Pain in Autistic Children

Bethany Donaghy

A thesis submitted in partial fulfilment of the requirements of Liverpool John Moores University for the degree of Doctor of Philosophy

May 2025

Contents

Abstract

Autistic children and young people experience pain at an alarmingly high rate. For example, Autistic children and young people are twice as likely to experience pain than their non-Autistic peers and represent 14% of paediatric chronic pain in tertiary pain management settings. Despite this high pain prevalence, understanding of Autistic children and young people's pain experiences remain sparse. Without this understanding, methods for addressing this health inequity cannot be implemented leaving a continually underserved population to be overlooked. To address this knowledge gap, the aim of this thesis was to examine factors which relate to Autistic children and young people's pain experiences and expression. To study these factors a range of methods were used, including four interrelated studies co-produced with Autistic children and young people, and their caregivers.

An initial systematic review of 87 psychophysical studies aided the synthesis of an ethical protocol for assessing pain thresholds in paediatric populations (see Chapter 2). Findings highlighted that established adult psychophysical protocols are feasible in use when assessing pain in paediatric populations. However ethical considerations pertaining to diagnostic groups, and the number of pain modalities used should be considered when adapting to a paediatric design.

In practice, the protocol developed from the systematic review was used to assess differences in mechanical, pressure, and cold pain thresholds, cold pain tolerance and subsequent pain intensity ratings between 9 Autistic and 20 non-Autistic children and young people (see Chapter 3). Following removal of data from one non-Autistic young person who consistently met ceiling values, findings suggested pain experiences did not differ between diagnostic groups, but observed individual differences within groups reinforced the need to consider the subjective nature of Autistic pain experiences.

To understand subjective pain experiences further and identify potential influential factors of pain, 10 dyadic interviews with Autistic children and young people, and their caregiver were conducted (see Chapter 4). Amongst other psychological and cognitive components, interpersonal factors like trust appeared to act as a gatekeeper to Autistic children and young people disclosing pain.

Further understanding of intent to disclose pain to caregivers, teachers, and healthcare providers was developed using an online survey including caregivers of 64 Autistic and 80 caregivers of non-Autistic children and young people (see Chapter 5). Autistic children and young people were consistently less likely to disclose pain to teachers and healthcare providers with communicative and social expectations identified as influential to their intent to disclose.

Whilst findings cannot explain the high pain prevalence amongst Autistic children and young people explicitly, a need to shift focus from a hyposensitive pain profile and towards understanding subjective pain experience was emphasised. Future research should develop this understanding, particularly considering how interpersonal factors like previous experiences of bystanders disbelieving pain impact an Autistic child or young person's behavioural intent to disclose pain. With this, guidance for teachers and healthcare providers in better supporting Autistic children and young people's pain should be developed to ensure this population receives timely pain appraisal and management in all environments. Hopefully, this development would contribute to decreasing the alarmingly high pain rates amongst this population.

Declaration

The author, Bethany Donaghy, declares that no portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

Acknowledgements

This thesis represents the subsequent end to my formal academic journey, and to say this was completed alone would be a detriment to all the support I have received over the years – here I want to take the time to thank everyone who has helped me in reaching this milestone.

To my Mum, Dad, Mia, and Grandma, thank you for all you have ever done for me. No opportunity was ever off the cards as you were there to help me achieve, whether that be supporting me emotionally, financially, or literally being my plus one to help me socially. Even when I felt a failure, you have picked me up every time and reminded me of my worth, helping to do anything and everything to get to this point. Although I don't say it much, I am very grateful for your continued support and this thesis is truly the collective reflection of how strong we are as a family – Team Donaghy as mum would say. Hopefully the Dunelm sign will be disappearing sooner than we think.

To my husband, Thomas, you met me at the start of my university journey, and I am thankful for some reason you stuck around to see it through to the end. You make it clear I can do anything I put my mind to and are the most selfless person I know doing everything in your power, so I achieve my dreams. Even following me across the world to sit in a hotel room whilst I attend an international conference just so that when I am struggling, you are there with me. You have been my rock throughout this whole PhD making sure I eat, sleep, regulate my emotions, and know how special I am. I am not always the best with words (ironic I know), but I appreciate all that you do. I am the luckiest person in the world to share this moment and my life with you. Now this is submitted, and you are finally reading, let's go to Disney World, or maybe I can compromise with Canada … the EPCOT pavilion.

To my primary supervisor, Dr David Moore, you have supported me and my academic development since the day I met you. To have had the opportunity to learn from your expertise over the last 7 years has shaped me into the academic I am today; we have come a long way since the internship interview. Thank you for believing in me when I often did not and listening to the 101 ways my mind likes to interpret a thought, grounding me to what I really need to be considering. You have never declined a need for me to talk things through, or to take opportunities that have allowed me to develop my skillset - I truly would

not have been able or wanted to do this without you. To my wider supervisory team: Professor Helen Poole, Dr Ben Rosser, and Dr Michelle Failla, your insight into how to complete a true breadth of methodologies has taught me so much. I always have gone away from our meetings grateful for your time and the opportunity to learn the best way to do research. Thank you for being a part of my supervisory team, and ensuring my work reflects my ability.

To my friends I have made during my time in academia, thank you for lifting me when I was down. You have taught me academia does not need to be about "*who is the best*", it is about working together to become the best academic versions of ourselves. Particularly in discussing friendship I would like to acknowledge Shaunna Devine - thank you for showing me what true friendship is, I feel very lucky that completing this PhD led me to meeting you. To my mentor, Angie, you helped me overcome many mental health difficulties in getting back to attending university. Thank you for always being there to listen and process what is going on in my life, I do not know if I would have even got to master's level let alone the end of a PhD without your support.

To all the Autistic children, non-Autistic children and caregivers who have taken the time to participate or advise; it is not easy to be vulnerable in your experiences, but you all took the time to do so in the aim of helping others - thank you for trusting me with these experiences. All knowledge acquired here has resulted from you and whatever becomes of this line of work is thanks to you. You are as big a part of this work as I am, and I hope you are proud of the way I have represented you.

Lastly, I want to thank the 7-year-old, undiagnosed Autistic girl in the picture below who first began this dream of becoming a Dr – a dentist to be exact. You pushed through many obstacles to get to where we are today and worked tirelessly towards your dream. Whilst we may not be the Dr we first intended; we are the Dr we should have always been, helping Autistic children just like you. I wish I could go back in time to tell you all that we have achieved, but for now in true Disney fashion: **Dreams Do Come True**

List of Tables

List of Figures

Research Publications

Ashworth, E., Bray, L., Hanlon, C., Stanway, H., Pavlopoulou, G., Moore, D. J., **Donaghy, B.**, Coen, E., & Firth, E. (2024). “Accumulating Harm and Waiting for Crisis”: Parents’ Perspectives of Accessing Child and Adolescent Mental Health Services for their Autistic Child Experiencing Mental Health Difficulties. *medRxiv*, 2024.2004.2009.24305538. https://doi.org/10.1101/2024.04.09.24305538

Boerner, K.E., Pawliuk, C., Heran, A., **Donaghy, B.,** Moore, D. J., Leong, K., Devan, H., & Oberlander, T. F. (n.d.) Systematic Review of Autistic Representation in the Treatment Literature for Pediatric Chronic Pain. *Journal of Pain.* (Accepted).

Donaghy, B. (Guest). (2024, March). Pain in Autistic Children and Young People. (No. 59) [Audio Podcast]. In *The Middletown Centre for Autism Podcast.* Retrieved from https://www.middletownautism.com/social-media/podcast-pain-in-autistic-children-and-young-people-with-bethany-donaghy-6-2024

Donaghy, B., & Failla, M. D. (Guests). (2022, October). Autism and Pain Sensitivity: Paying Attention to Pain in Autism. (No. 290) [Audio Podcast]. In *The Healing Pain Podcast.* Retrieved from https://integrativepainscienceinstitute.com/latest_podcast/autism-and-pain-sensitivity-paying-attention-to-pain-in-autism-with-bethany-donaghy-msc-bsc-and-michelle-failla-phd/In *The Healing Pain Podcast.*

Donaghy, B., & Linkiewich, D. (2024). Research Imitates Life: Researching Within Your Lived Experience. Patient Experience Journal, 11, 4-8. https://doi.org/10.35680/2372-0247.1916

Donaghy, B. & Moore, D.J. (Guests). (2024, March). Pain in Autistic Children and Young People. [Audio Podcast]. In *What a Pain.*

Donaghy, B., Moore, D. J., & Green, J. (2023). Co-Occurring Physical Health Challenges in Neurodivergent Children and Young People: A Topical Review and Recommendation. *Child Care in Practice*, 1-19. https://doi.org/10.1080/13575279.2022.2149471

Donaghy, B., Walker, S. C., & Moore, D. J. (2022). Social distancing with chronic pain during COVID-19: A cross-sectional correlational analysis. *PLoS One*, *17*(11), e0275680. https://doi.org/10.1371/journal.pone.0275680

Hanlon, C., Ashworth, E., Moore, D. J., **Donaghy, B**., & Saini, P. (2022). Autism should be considered in the assessment and delivery of mental health services for children and young people. *Disability & Society*, 1-6. https://doi.org/10.1080/09687599.2022.2099252

Jordan, A., Parchment, A., Gauntlett-Gilbert, J., Jones, A., **Donaghy, B.**, Wainwright, E., Connell, H., Walden, J., & Moore, D. J. (2024). Understanding the impacts of chronic pain on autistic adolescents and effective pain management: a reflexive thematic analysis adolescent–maternal dyadic study. *Journal of Pediatric Psychology*. https://doi.org/10.1093/jpepsy/jsae004

Oral Presentations

Invited Talks

Donaghy, B. (Moderator), Failla, M. D., & Moore, D. J. (2023). Pain and Autism: From the Clinic to the Lab and Back Again. Pain in Intellectual and Developmental Disabilites Special Interest Group (PIDDSIG), IASP.

Moore, D. J., & **Donaghy, B.** (2023). Pain in Autism. In: University of Bath - Centre for Pain Research (CPR) and Centre for Applied Autism Research (CAAR).

Moore, D. J., & **Donaghy, B.** (2023). Pain in Autism. Centre for Research in Autism and Education (CRAE) Webinar Series, University College London (UCL), UK.

Donaghy, B. (2023). Pain in Autistic Children and Young People. In University of Liverpool Pain Early Career Network Group (Ed.).

Donaghy, B. (2023). Pain in Autistic Children and Young People: Current Research and Future Directions. In: Sefton Child and Adolescent Mental Health Services (CAMHS).

Donaghy, B. (2022). Pain in Autistic Children and Young People. In Pain Peer Support Group (Ed.).

Donaghy, B. (2022). Pain in Autistic Children and Young People. In Participatory Approaches to Research Special Interest Group (Ed.).

Donaghy, B. (2022). PhD Journey: Pain in Autistic Children and Young People. In CAMHelions - Alder Hey Children's Hospital Trust (Ed.).

Donaghy, B., Ashworth, E., & Moore, D. J. (2023). Pain in Autistic Children and Young People: What We Know and Where We Need to Go. Beyond Learning Disabilities/Difficulties and Autism Workstream. Cheshire and Wirral Partnerships, NHS, UK.

Conference Talks

Donaghy, B. (2023, October 1-4). Pain in Autistic Children: Interviews with Child-Parent Dyads. In T. Oberlander (Chair), *Pain, Placebos and Autism: Looking at Multiple Dimensions of Pain in Autistic Children and Youth to Charting a Way for More Inclusive, Accessible Research.* [Symposium]. International Symposium on Paediatric Pain, Halifax, Canada.

Donaghy, B., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2022). Pain in Autistic Children: Interviews with Child-Parent Dyads. Institute for Health Research Children, Young People and Families Conference, Liverpool, UK.

Donaghy, B. (2021). Pain in Autistic Children: A PhD Outline. Pain Relief Foundation Conference, Liverpool, UK.

Conference Posters

Donaghy, B., Hanlon, C., Williams, S., Tomlinson, E., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2023). Co-creating a Psychophysical Protocol for Studying Pain in Autistic Children. International Symposium for Paediatric Pain, Halifax, Canada.

Donaghy, B., Hanlon, C., Williams, S., Tomlinson, E., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2023). Co-creating a Psychophysical Protocol for Studying Pain in Autistic Children. Liverpool Neuroscience Day, Liverpool, UK.

Donaghy, B., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2022). Pain in Autistic Children: Interviews with Child-Parent Dyads. International Association for the Study of Pain, Toronto, Ontario.

Donaghy, B., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2022). Pain in Autistic Children: Interviews with Child-Parent Dyads. LJMU Research Knowledge and Exchange Conference, Liverpool, UK.

Donaghy, B., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2022). Pain in Autistic Children: Interviews with Child-Parent Dyads. Neurodevelopmental Disorders Annual Seminars, Edinburgh, Scotland.

Donaghy, B., Poole, H., Rosser, B., Failla, M. D., & Moore, D. J. (2022). Pain in Autistic Children: Interviews with Child-Parent Dyads. Postgraduate Researcher Conference, Liverpool, UK.

Awards

Listed below are monetary awards received during the span of this PhD:

1) Pro-Vice Chancellor Scholarship, LJMU, UK.

Awarded scholarship on basis of research experience and project strength which provided waived student fees, a monthly stipend for 3 years, and annual bench fees.

2) Funding Grant, LJMU, UK. Total: Up to £3000

Grant collaborator and Autistic lead on Claire Hanlon's project: Improving support for neurodivergent students.

3) Conference Travel Fund for IASP, LJMU Doctoral Academy, UK. Total: £350.

Internal grant awarded to selected applicants – awarded to attend IASP conference, Toronto, Canada.

4) Trainee Travel Award for ISPP, Pain in Child Health (PICH), Canada. Total: $1500 CAD.

Funding award from PICH allocated to 5 good standing Trainee PICH members to support travel ISPP. Paid for international flights to attend and present (symposium and poster) at ISPP, Halifax, Canada.

5) Conference Travel Fund for ISSP, LJMU Doctoral Academy, UK. Total: £350.

Internal grant awarded to selected applicants -awarded to attend ISSP conference, Halifax, Canada.

6) Full registration fee for ISPP, IASP, USA. Total: $464 USD.

Funding award from IASP allocated to waive registration fee at ISPP for selected applicants.

7) Pain Education Day Fee for ISPP, PICH, Canada. Total: $100 CAD.

Funding award from PICH allocated to 5 good standing Trainee PICH members to attend Pain Education Day.

Listed below are non-monetary awards received during the span of this PhD:

1) Best Poster Presentation, LJMU Research Knowledge & Exchange Conference, Liverpool, UK.

Contributions

As the author of this thesis, I confirm that I, Bethany Donaghy, undertook the creation, design, data collection and write up of all studies presented as Chapters which have been reviewed by my supervisors: Dr David Moore, Professor Helen Poole, Dr Ben Rosser, and Dr Michelle Failla.

Within the systematic review outlined in Chapter 2, external collaborators Professor Tine Vervoort and Dr Emma Rheel assisted Dr David Moore, Professor Helen Poole, Dr Ben Rosser, Dr Michelle Failla and I in the conception of protocol design. Dr Emma Rheel and Dr David Moore provided additional assistance in the screening of journal articles however I (Bethany Donaghy) completed the data extraction and synthesis alone.

Studies outlined in Chapters 3 through 5 were designed between me (Bethany Donaghy), Dr David Moore, Professor Helen Poole, Dr Ben Rosser, and Dr Michelle Failla. Mia Donaghy assisted in editing and animating the recruitment video outlined in Chapter 4. However, all data collection, data analysis and data synthesis were completed by I (Bethany Donaghy) for the purpose of this thesis.

Abbreviations in Alphabetical Order

ADHD	Attention-Deficit Hyperactivity Disorder
ALL	Acute Lymphoblastic Leukaemia
ASD	Autism Spectrum Disorder
BPD	Borderline Personality Disorder
CAS	Coloured Analogue Scale
CD	Can't Determine
CDTh	Cold Detection Threshold
CFS	Chronic Fatigue Syndrome
cm	Centimetre
Cntrl	Control
CoVAS	Computerised-Visual Analogue Scale
COVID-19	Coronavirus Disease
CP	Cerebral Palsy
CPO	Chronic Pain and Obesity
CPM	Conditioned Pain Modulation
CPT	Cold Pressor Task
CPTh	Cold Pain Threshold
CPTol	Cold Pain Tolerance
CRPS	Complex Regional Pain Syndrome
CTTH	Chronic Tension-Type Headache
CU	Callous Unemotional
CYP	Children and Young People
DFNS	German Research Network on Neuropathic Pain
DM	Diabetes Mellitus
DMA	Dynamic Mechanical Allodynia
DS	Down Syndrome
DSM-5	Diagnostic Statistical Manual-5
EDS	Ehlers-Danlos Syndrome
ERA	Enthesitis-Related Arthritis

EU	European
f	Female
FAP/FAPD	Functional Abdominal Pain/Functional Abdominal Pain Disorder
FETTH	Frequent Episodic Tension-Type Headache
FLACC	Face-Legs-Activity-Cry-Consolability Scale
FoP	Fear of Pain
FOPQ-P	Fear of Pain Questionnaire-Parents
FPS-R	Face Pain Scale-Revised
g	Gram
GJH	Generalized Joint Hypermobility
GI	Gastrointestinal
HC	Healthy Control
HCP	Healthcare Provider
HMS	Hypermobility Syndrome
HPTh	Heat Pain Threshold
HPTol	Heat Pain Tolerance
HSD	Hypermobility Spectrum Disorders
HSO	Health Standards Organization
GPain	Growing Pains
IASP	International Association for the Study of Pain
IBS	Irritable Bowel Syndrome
ICD-11	International Classification of Diseases-11
ID	Intellectual Disability
IFL	Identity First Language
IPA	Interpretative Phenomenological Analysis
IQ	Intelligence Quotient
IU	Intolerance of Uncertainty
JCA	Juvenile Chronic Arthritis
JFM	Juvenile Fibromyalgia
JIA	Juvenile Idiopathic Arthritis

kg	Kilogram
kPa	Kilopascal
kPa/s	Kilopascal per Second
K-S	Kolmogorov Smirnov
LEP	Laser Evoked Potential
LJMU	Liverpool John Moores University
log_{10}	Log Base 10
m	Male
M	Mean
MCA	Mental Health Capacity Act
Mdn	Median
MDTh	Mechanical Detection Threshold
mm	Millimetre
mN	Micronewton
MPS	Mechanical Pain Sensitivity
MPSumm	Mechanical Pain Summation
MPTh	Mechanical Pain Threshold
mQST	Modified Quantitative Sensory Testing
M_{Age}	Mean Age
M_{Temp}	Mean Temperature
MSK	Musculoskeletal
mths	Months
NA	Not Applicable
NCCPC-R	Non-Communicating Children's Pain Checklist-Revised
NHLBI	National Heart, Lung, and Blood Institute (NIH)
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health and Care Research
NR	Not Reported
NRS	Numerical Rating Scale

NSSI	Non-Suicidal Self-Injury
O	Obesity
OSD	Osgood-Schlatter Disease
PCS-P	Pain Catastrophizing Scale-Parents
PFL	Person First Language
PFPS	Patellofemoral Pain Syndrome
PHS	Paradoxical Heat Sensitivity
PIS	Participant Information Sheet
PMS	Premenstrual Syndrome
PPI	Public and Patient Involvement
PPTh	Pressure Pain Threshold
PPTol	Pressure Pain Tolerance
PTh	Pain Threshold
QST	Quantitative Sensory Testing
Q.U.E.S.T	(Q)uestion the Child (U)se Pain Rating Tools (E)valuate Behaviour (S)ensitise Parents (T)ake Action
RAP	Recurrent Abdominal Pain
ROD	Rate of Decrease
ROI	Rate of Increase
s	Seconds
SCD	Sickle Cell Disease
SD	Standard Deviation
SEN	Special Educational Needs
SH	Self-Harm
SIB	Self-Injurious Behaviour
SPSS	Statistical Package for the Social Sciences
TD	Typically Developing

TDTh	Tactile Detection Threshold
TMD	Temporomandibular Disorder
TPS	Thermal Perceptual Sensitization
TSL	Thermal Sensory Linen
TSP	Temporal Summation of Pain
TTH	Tension-Type Headache
UK	United Kingdom
UKRI	UK Research and Innovation
UREC	University Research Ethics Committee
US	United States
VAS	Visual Analogue Scale
VDTh	Vibration Detection Threshold
VRS	Verbal Rating Scale
WDTh	Warm Detection Threshold
WHO	World Health Organization
WILDA	(W)ords to Describe Pain (I)ntensity (L)ocation (D)uration (A)ggravating and (A)lleviating Factors
WUR	Wind-Up Ratio
Yrs	Years
°C	Degree Celsius
°C/s	Degree Celsius per Second

Chapter 0.

Representing the Autistic Voice Through Research

0 Representing the Autistic Voice Through Research

0.1 Neurodiversity-Affirming Approaches to Research

Historically, a deficit-based model has been applied to Autism research, defining Autistic people by *"what Autistic individuals are missing rather than what they have great aptitude for"* (Anderson-Chavarria, 2022, p.1329). Countering these deficit-based views, Autistic advocates have rallied for a neurodiversity movement which appreciates the variation in how brains work and elevates neurodivergent rights for inclusion and autonomy (den Houting, 2018; Kapp, 2020). Consequentially, this movement encouraged a shift towards neurodiversity-affirming research, emphasising the importance of defining Autistic people by their strengths, and utilising the Autistic community's preferred language. For example, describing perceived "deficits" as differences or difficulties.

As an Autistic person, I appreciate the inclusive nature the neurodiversity movement presents by including Autistic people's preferences – particularly where research serves to support them. Given my appreciation, I have implemented a neurodiversity-affirming approach throughout this thesis with examples of how being provided in Chapter 3 through 5's methodology.

0.1.1 Language Use

The preferred language of the Autistic community has been utilised throughout this thesis when describing the interaction between Autism diagnosis and identity. Preferences were identified through my lived experience as an Autistic person, Bottema-Beutel et al.'s (2021) impactful paper exploring ableist Autism language, and grey literature including Autistic lived experiential statements on social media outlets like X. Based on the latter, identity-first language (IFL; i.e. Autistic person) was utilised as opposed to person-first language (PFL; i.e. person with Autism), as PFL is deemed to project Autism as an 'accessory' rather than being encompassed into an individual's identity. Further, despite "Autism Spectrum Disorder" or "ASD" being used for diagnostic purposes within the

Diagnostic Statistical Manual-5 (DSM-5; American Psychiatric Association (2023)), “Autism” is used throughout. The removal of “Spectrum” and “Disorder” limits the discussed deficit assumptions; a language shift the National Health Service (NHS) and academics in the Autism field are beginning to use (Bottema-Beutel et al., 2021). However, whilst IFL and the term “Autism” was predominantly used, if participants preferred PFL or described their diagnosis as “ASD”, I respected their choice and adapted my language towards their preference in these interactions. Moreover, use of “deficit” language has been changed to “difference”, particularly when referencing the DSM-5’s (American Psychiatric Association, 2023) diagnostic criteria.

Additionally, whilst outdated evidence supports suggestions of *“persistent deficits”* particularly when socialising, these are not at the onus of the Autistic person but rather society’s predominant non-Autistic perceptions of the *“correct”* way to socially engage. Views that project social deficits rather than reflect intentional and behavioural differences must be readdressed and redefined. The outcomes of this thesis intend to contribute towards these efforts.

However, it must be acknowledged that the preferred language utilised here are akin to the research period and reflect the current understanding of best language use. Inclusive language exists as a living process which adapts to the preferences of society and the Autistic community at the time. Any individual engaging with this research should actively understand what inclusive language looks when beginning their own research and adapt to the Autistic community’s preferences regardless of if they differ from those utilised here. Always be mindful when implementing an inclusive approach.

0.1.2 Co-Producing Research

The neurodiversity-affirming approach discussed in Chapter 0.1 emphasises that research should encompass “Nothing About Us, Without Us”, to best represent the voices of the people research serves to support (Hughes, 2016). To incorporate this statement, the NIHR INVOLVE (2018) outlined a need to co-produce research with members of the public

with lived experience (including patients), as they know what research should address, how studies can be accessibly designed and how to best disseminate findings for maximum impact. In agreement with the NIHR INVOLVE's (2018) recommendations, Public and Patient Involvement (PPI) panels were involved in co-producing study design, material assessment of Chapter 5, and in validating analysis of Chapters 3, and 4. Doing so was perceived to have improved research inclusivity, provided representative analysis, and above all put Autistic people at the forefront of research both about, and involving them.

To ensure PPI panels represented target populations, Autistic children and young people (CYP), Autistic adults and caregivers of Autistic CYP participated. Specifically, Chapters 3 and 5 included a PPI panel of one Autistic CYP aged 11-16 years, one maternal caregiver and one Autistic adult. The same individuals participated in Chapter 4's PPI panel with the addition of a second maternal caregiver, and a second Autistic CYP aged 11-16 years. Panels were recruited via social media and word-of-mouth, with clear guidance that meetings were online and that whilst monetary incentives were not available due to limited availability of funds, authorship would be provided on conference posters.

In conducting these panels, I sent the discussion structure, and all relevant materials or analyses to the PPI panel members one week prior to the arranged meeting for review. Throughout meetings, I signposted discussion points from the provided structure but allowed panel members to control the conversations whilst my lead supervisor noted key discussion points like areas for improvement. The following day, I disseminated these points to ensure members agreed with required changes - then one week later, amended materials or analyses were disseminated to affirm suggestions were correctly incorporated and represented.

0.2 Mental Capacity Act

Informed consent and relevant informed assent are fundamental for allowing participants to safely engage with research – these can be provided in written, verbal, or implied formats (UKRI, 2024). The principles outlined in Sections 1(1) (2) (3) (4) (5) and (6)

of the Mental Health Capacity Act (MCA) (2005) explain that all individuals aged 16 and over are deemed to possess capacity to provide informed consent. However as outlined in Section 2(1), and Sections 3(1) (a) (b) (c) and (d) of the MCA (2005), exceptions to these principles include any individuals who experience a difference in the functioning of their mind or brain. When these differences in functioning impact an individual's ability to understand and process study information, and to communicate their decision to participate by any means, an assessment of capacity must be completed to confirm their ability to consent.

As I interacted with all participating CYP aged 16 years during Chapter 3 and 4's studies, I had ample opportunity to ensure their consent was informed by answering questions and demonstrating procedures prior to study commencement. However, as Chapter 5 involved an anonymous online survey, LJMU University Research Ethics Committee (UREC) required consideration of Section 2(1), and Sections 3(1) (a) (b) (c) and (d) of the MCA (2005) to ensure consent was informed. This was consequential to anonymity preventing the ability to determine whether CYP aged 16 years had fully understood and processed the presented study information. The ability to mitigate this was further limited as requesting additional informed assent from the CYP, and consent from their caregiver aged over 18 years to affirm the latter would have violated Sections 1(1) (2) (3) (4) (5) and (6) of the MCA (2005) which states capacity must be assumed unless otherwise established. Thus, as capacity could not be established nor could informed assent for CYP aged 16 be lawfully obtained, the inclusion age of Chapter 5 was limited to 11-15 years.

0.3 Reflections as an Autistic Researcher

As an Autistic PhD researcher researching within their lived experience, additional benefits and challenges arose throughout the research process which I have reflected on. Whilst the biggest benefit of researching within my lived experience was the ability to embed my own and my community's expertise, this became emotionally exhausting. Particularly I reflected on my own experiences as an Autistic person experiencing healthcare which

provided great strength in attaining research applicability and impact, but additionally caused distress in highlighting how systems and people had let me down. If I were to have researched a topic separate from my Autistic identity, the need to manage such an emotional toll may have been mitigated but the skills my lived experience provides in helping address Autistic health inequities would never have been included. Additionally, I felt emotionally attached to Chapter 4 of this thesis and experienced immense self-inflicted pressure to “get it right” in correctly analysing and disseminating the Autistic CYP’s voices. I knew how important this piece was for highlighting their often-overlooked experiences and consequentially found it difficult to not overanalyse. Here, it feels like I was faced with a consistent risk vs. reward battle, one that can often be unappreciated by those who do not engage with lived experience researchers.

This emotional toll extended further when continually engaging with research portraying a deficit-based perspective of Autism. Reading multiple articles which stated what an Autistic person could not do and should not do caused me feelings of upset and anger. This became easier upon finding vital work produced by neurodiversity-affirming academics, however I continued to feel haste when seeing other academics who perpetuated these deficit views. Here, I suggest that when talking about any Autistic individual, narratives present a neurodiversity-affirming approach rather than continuing to support the deficit-based models – unintentionally or not.

From a research design perspective, learning how to balance the fine line of too little, or too much lived experience was a difficult task that became evident within Chapters 3, 4, and 5’s ethics application. These ethics applications were extensive given I wanted to cover all possibilities that could arise within the study, and ensure materials were thorough in providing relevant details. In covering these possibilities, I continually reflected on what I would want from a study as an Autistic participant and applied my own and my communities lived experience to support my suggested adaptations. However, it was difficult to determine at what point I should stop providing lived experience as there is always a point where the evidence provided is sufficient to emphasise a need, yet I did not want any Autistic individual to feel their experience was not reflected in my design decisions.

Although I found certain areas difficult when researching within my lived experience, the overall experience has been enjoyable. Truly I perceive all the benefits and challenges I faced along this PhD journey to be entirely worth it if this research serves one sole purpose; helping address Autistic health inequities.

Chapter 1.

Literature Review

1 Literature review

1.1 Autism

1.1.1 Defining Autism

Approximately 1% of the global paediatric population are diagnosed Autistic (Zeidan et al., 2022), comparative to approximately 3% of the English paediatric population (O'Nions et al., 2023). These figures may increase further when considering individuals who are currently waiting on the diagnostic pathway; are overlooked due to Autism stereotypes; or will receive a "*late-diagnosis*" throughout adulthood (O'Nions et al., 2023; Zeidan et al., 2022). As the current thesis includes diagnosed Autistic CYP, the diagnostic criterion for Autism is outlined below.

Both the DSM-5 (American Psychiatric Association, 2023) and the International Classification of Diseases-11 (ICD-11; World Health Organization (2022a)) diagnostically assess Autism against 4 key diagnostic classifications (see Table 1 for an outline of the discussed classifications):

1) Persistent differences in social communication and social interaction:

Three social diagnostic domains are presented in these criteria to suggest Autistic people have differing social abilities, particularly for *"Social-Emotional Reciprocity"*, *"Non-Verbal Communication",* and *"Developing and Maintaining Relationships at Developmental Level"* (American Psychiatric Association, 2023; World Health Organization, 2022a). Reported examples of how these may manifest for Autistic people include:

(1) limited awareness of, and attention to others social, emotional, and interpersonal cues (Downs & Smith, 2004; Macdonald et al., 1989),

(2) avoidance of social eye-contact often misinterpreted as rudeness or inattentiveness (American Psychiatric Association, 2023; Madipakkam et al., 2017),

(3) fewer friendships than non-Autistic peers (Calder et al., 2013; Wainscot et al., 2008).

However, neurodiversity-affirming literature counters the "*social deficit*" perspective of Autism which these diagnostic classifications outline. When each neurotype are compared, they may display differences in priorities for seeking, developing, and maintaining social relationships, however social relationships do exist (Black et al., 2022; Finke et al., 2019; O'Hagan & Hebron, 2017; Sosnowy et al., 2019). This may explain why Autistic people often report feeling a sense of belonging amongst other Autistic people, or those with shared interests (Crompton et al., 2020; Morrison et al., 2020; Rowley et al., 2012). Yet as non-Autistic people often possess higher societal power in driving understanding of socialisation, their social preferences and motivations are deemed the norm. Thus, when Autistic people's social preferences, communication style and motivations often divert from these normative views, they are deemed to be of a social deficit enforcing a pathologized perspective of their social nature rather than demonstrating a mere social difference.

2) Restricted ("focused"), repetitive patterns of behaviour, interests, or activities:

Behavioural aspects of Autism are discussed within the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) criterion, both requiring at least two symptomatic manifestations for diagnosis. These include *"Stereotyped or Repetitive Motor Movements, Speech, and Use of Objects"*, "*Excessive Adherence to Routine and Ritualised Patterns*", *"Focused Ares of Interest"* and *"Hyper- or Hypo-reactivity to Sensory Inputs".*

The use of *"Stimming",* otherwise known as self-stimulatory behaviour, is deemed synonymous with Autism. Exemplar behaviours involve engaging with repetitive vocalisations or behaviours such as hand-flapping or spinning, and self-injurious behaviours (SIB) such as scratching, picking, or biting (Canitano, 2006; Charlton et al., 2021; Kapp et al., 2019; National Autistic Society, 2020c). Whilst to bystanders the use of stimming may be unknown, it is regularly considered to self-regulate and serves as a cognitive mechanism (i.e. distraction and concentration), an act of emotional expression, and a means of managing sensory overload (Charlton et al., 2021; Kapp et al., 2019; Nwaordu & Charlton, 2023). An example of using stimming to manage sensory overload includes when an Autistic person is experiencing pain,

with evidence to suggest that stimming behaviours may act as a method for pain reduction by providing a distraction and promoting emotional relief (Kalingel-Levi et al., 2022). In contrast use of SIB to self-stimulate has misconstrued beliefs, with theories to suggest that SIB are not as painful for Autistic people, and that repeated engagement alters their pain sensitivity (Duerden et al., 2014; Edelson, 2021). However, evidence to support these assumptions are limited at best, and thus this assumption should not be taken as fact.

Often an experience of change, or an intolerance of uncertainty (IU), can increase levels of anxiety in some Autistic people (Jenkinson et al., 2020; Wigham et al., 2015). To help alleviate these feelings, make sense of the world and maintain familiarity, some Autistic people show a preference for routine and patterns. An example of this may be eating the same meals or going to bed at the same time each day (National Autistic Society, 2020a). However, the level which using these routines and patterns are helpful depends on the individual, and can further increase anxiety when routine deviates by matters outside of one's control (Dallman et al., 2023; Featherstone et al., 2023)

Focused areas of interest are very common amongst Autistic people. For example, Turner-Brown et al. (2011) demonstrated 89% of their study population possessed a focused area of interest at some point in their lives, 85% of which had persisted since early childhood. Trains are often an exemplar focused area of interest; however, exact topics differ between Autistic individuals (National Autistic Society, 2020b; Turner-Brown et al., 2011). Accessing interests can produce positive outcomes for Autistic people including promoting subjective wellbeing, providing intrinsic reward, assisting management of sensory overload, and facilitating socialisation by providing a conversational topic (Grove et al., 2018; Grove et al., 2016; Kapp et al., 2019; Patten Koenig & Hough Williams, 2017; Winter-Messiers et al., 2007).

Sensory inputs such as sight, sound, taste, touch, and smell are discussed at large in the context of Autism. Diagnostic criteria, literature about Autistic people and Autistic anecdotal reports consistently suggest Autistic people experience hypo- or hyperreactivity to sensory input (referred to from here as hypo- or hypersensitivity) (Corbett et al., 2016; Neil et al., 2016; Vaughan et al., 2020). Although a sensory input often overlooked is pain, despite the DSM-5 (American Psychiatric Association, 2023) diagnostic criterion specifying Autistic

people experience *"an indifference to pain"* and a *"high tolerance for pain"*. Current research considering pain experiences of Autistic people is emerging but remains limited. Yet an understanding of the underlying mechanisms of this perceived hypo- or hypersensitivity to pain has begun to emerge using lab-based studies which administer experimental pain through psychophysical assessments (Symons et al., 2022; Vaughan et al., 2020). However, lab-based evidence still does not provide clarification to support this perceived unidirectional response within the Autistic population, nor do they provide clear application to the experiences of pain within a real-world setting. Therefore, despite its presence within diagnostic criteria, the exact directional relevance of pain hypo- or hypersensitivity for the Autistic population remains poorly understood. This leaves but one overarching and alarming fact: we still do not understand the role of pain within Autism (Moore, 2014; Ortiz Rubio et al., 2023; Vaughan et al., 2019).

3) Presence of symptoms ("traits") in early childhood:

As a neurodevelopmental condition, Autism is not something a person can *"get", "acquire"* or *"grow out of"* - an individual is born Autistic with traits remaining present throughout the lifespan (Atherton et al., 2022; NHS, 2022; Parmeggiani et al., 2019). Unfortunately, Autistic traits in early childhood may be perceived as different things, often leading to misdiagnoses or missed opportunities for diagnosis (Fusar-Poli et al., 2022; Gould & Ashton-Smith, 2011; Wilson et al., 2023). Common examples of misdiagnosis prior to a late-diagnosis of Autism include personality, anxiety, and mood conditions - especially amongst Autistic women (Gesi et al., 2021; Kentrou et al., 2024). Although these conditions may constitute an initial misdiagnosis, this does not mean late-diagnosed Autistic individuals do not experience personality, anxiety, and mood conditions. In fact, evidence highlights late-diagnosed Autistic CYP experience more mental health conditions than early diagnosed Autistic CYP (French et al., 2023; Mandy et al., 2022). Additionally, whilst late-diagnosed Autistic adults may experience positive emotions when diagnosed, early diagnosed Autistic adults may experience a higher quality of life and well-being (Oredipe et al., 2022; Stagg & Belcher, 2019). To limit the potential impact of late-diagnosis amongst Autistic individuals, healthcare providers (HCP) should receive better training to understand the differing

presentation of Autistic traits in early childhood and facilitate more Autistic CYP receiving both an early-diagnosis and adequate support.

4) Limitation or impairment of everyday functioning:

Clinical significance for an Autism diagnosis is determined by the level which an individual's Autistic traits effect their ability to perform everyday functions including that of social, household, and occupational activities (American Psychiatric Association, 2023; World Health Organization, 2022a).

Despite receiving an Autism diagnosis using the same classifications, presentations of Autism can vary particularly in the context of co-occurring conditions and differing communication styles. One of the most likely conditions to co-occur with Autism is an intellectual disability (ID). Data collected by O'Nions et al. (2023) suggests as of 2018, ~9.34% of Autistic males and ~9.47% of Autistic females aged 10-19 years' experience a co-occurring ID. This means for these CYP, their intellectual functioning and adaptive behaviour (deemed as behaviours performed in everyday lives) is approximately 2+ standard deviations below the standardised mean of the population (World Health Organization, 2022b). The Autistic population have historically been classified into *"Levels of Functioning"* based on a co-occurring ID and their intelligence quotient (IQ), with this application of IQ still prevailing as a categorical purpose in the newly coined *"Profound Autism"* (Alvares et al., 2020; Bal et al., 2017; Lord et al., 2022; Wolff et al., 2022). Autistic people were previously deemed as *"High-functioning"* if they did not have a co-occurring ID and possessed an IQ ≥ 70, and *"Low-functioning"* if they did have a co-occurring ID and possessed an IQ < 70 (Alvares et al., 2020; Bottema-Beutel et al., 2021; Wolff et al., 2022). However, whilst some literature and The Lancet's terminology of *"Profound Autism"* continues to define Autistic people by IQ, neurodiversity-affirming movements have pushed for a predominant step away from this language (Bottema-Beutel et al., 2021; Bottini et al., 2023; Lord et al., 2022). Instead, there is a call to recognise the support an Autistic person requires as opposed to oversimplifying them to a functional label which may fluctuate dependent on, for example,

environmental circumstances and testing validity (Bottema-Beutel et al., 2021; Bottini et al., 2023; Kapp, 2023).

Moreover, differing communication styles are prevalent across the Autistic population, with up to 65% of Autistic CYP utilising predominantly verbal communication and up to 35% utilising predominantly non-verbal communication such as gestures, reaching and use of images (National Autistic Society, 2020d; Rose et al., 2016; Schaeffer et al., 2023; Tager-Flusberg & Kasari, 2013). However, the two communication styles are not mutually exclusive; an individual can be predominantly verbal yet become non-verbal in high stress environments such as hospitals (Cummins et al., 2020; Haydon et al., 2021; Muskat et al., 2015). Importantly, communication styles can also change across development, for example an individual may use non-verbal communication in very early childhood yet develop verbal communication styles as their age or engagement with aspects of, for example, speech and language therapy progresses (Broome et al., 2023; Tager-Flusberg & Kasari, 2013). Thus, just because two individuals may be diagnosed Autistic, their experiences of Autism may differ. However, not discussed here is how this diagnosis comes to fruition in an identity-based perspective or importantly how these impacts perceptions in healthcare management; the latter which will be outlined in Chapter 1.1.2 and 1.1.3.

Table 1

An Outline of The Key Autism Diagnostic Classifications

Diagnostic Classification	Domain
Persistent differences in social communication and social interaction	Social-Emotional Reciprocity Non-Verbal Communication Developing and Maintaining Relationships at Developmental Level
Restricted ("focused"), repetitive patterns of behaviour, interests, or activities	Stereotyped or Repetitive Motor Movements, Speech, and Use of Object Excessive Adherence to Routine and Ritualised Patterns Focused Ares of Interest Hyper- or Hypo-reactivity to Sensory Inputs
Presence of symptoms ("traits") in early childhood	-
Limitation or impairment of everyday functioning	Effect on ability to perform everyday functions including that of social, household, and occupational activities

Note. Formatted from the DSM-5 (American Psychiatric Association, 2023) and the ICD-11 (World Health Organization, 2022a) Autism diagnostic criteria. Additional consideration is provided for how these criteria co-occur with, for example, intellectual disability.

1.1.2 Diagnostic Procedure

To be clinically diagnosed as Autistic, an individual's experience of Autistic traits is assessed against one of the two aforementioned diagnostic criteria: DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a). Methods for these assessments are outlined by the National Institute for Health and Care Excellence (NICE) (2021a) which states all Autism diagnostic assessments following the paediatric pathway (individuals aged <19 years) must include:

- Details of parental/carer, and the CYP's concerns,
- Details of the CYP's home, scholastic, and social care life,
- A developmental history assessment including the CYP's current developmental profile,
- Observational and interactional assessment of communication skills and behaviour,
- A physical examination of, for example, signs of injury,
- Consideration of differential diagnosis or co-occurring conditions, including Attention-Deficit Hyperactivity Disorder (ADHD).

However, due to extensive wait times (at least a 13 week wait before initial assessment) and costly private assessment fees, many are choosing to self-diagnose as Autistic before receiving or even trying to access a clinical diagnosis (Lewis, 2016a, 2016b, 2017; McDonald, 2020; NHS Digital, 2023; Overton et al., 2023). Whilst self-diagnoses are useful, advice and support are often only accessible to those clinically recognised as Autistic through diagnosis. This illuminates how the medical arena's dominant symptomatic-based approach for diagnosis continues to prevail and remains a gatekeeper to accessing relevant support needs (Huang et al., 2020). Although a call for further funding to address this matter and recognise the importance of self-diagnosis and identification has been highlighted by The Westminister Commission on Autism (2021), recruited individuals within this thesis will predominantly be clinically diagnosed as Autistic. Whilst this does not entirely align with the neurodiversity-affirming approach outlined in Chapter 0.1, a focus on clinical diagnosis allows findings to be contextualised to the current need for an Autism diagnosis within healthcare

settings and create a foundation of evidence that can be extended to the wider Autistic community.

1.1.3 Healthcare Implications

Many Autistic people report difficulties with the current Autism diagnostic procedure. These include difficulties in accessing clinical appointments, negative emotions from previous misdiagnoses, and even having their eventual Autism diagnosis undermined particularly based on sex due to differences in Autism presentation like, for example, masking behaviours in females (Atherton et al., 2022; Bargiela et al., 2016; Fusar-Poli et al., 2022; Harmens et al., 2022; Hull et al., 2020; Milner et al., 2019). Whilst it may be assumed these negative healthcare experiences are limited to the difficult and lengthy diagnostic procedure, they unfortunately extend to the wider healthcare system. For example, Autistic people often describe feeling the presence and severity of their physical health symptoms are not believed, particularly symptoms of pain (Doherty et al., 2021; Doherty et al., 2022; Shaw et al., 2023). Reasoning for this remains unknown, although one plausible explanation could refer to Autism diagnostic criterion.

As the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) diagnostically suggest Autistic individuals are more hyposensitive to pain, HCPs may not perceive an Autistic individual's pain severity to be valid for their injury as their pain threshold is expected to be increasingly high. Consequentially, Autistic individual's pain may be undermined and misunderstood which not only limits the pain management offered but can exacerbate wider inequities Autistic individuals experience. For example, from a healthcare progression standpoint, pain oversights may increase the possibilities for Autistic people to disproportionately experience chronic pain consequential to mismanagement at an acute presentation (Friedrichsdorf et al., 2016; Lavand'homme, 2011). The implications this chronification can have on the individual's life include but are not limited to: (1) reductions in physical activity, (2) restrictions in abilities to socialise and (3) loss of occupation due to pain symptomatology (Brown et al., 2021; Dueñas et al., 2016; Mills et al., 2019). Thus, misunderstanding of pains mere presence could therefore be an

influential factor to the high pain prevalence amongst the Autistic population. However, without clearly understanding pain's true positionality to Autism criterion and the outcome of these professional interactions, these healthcare and pain related concerns will only continue to perpetuate. We must do better to provide understanding of all aspects of Autistic health and how we can best provide effective and inclusive methods to manage concerns.

1.2 Pain

1.2.1 Defining Pain

Fundamentally pain serves as a function of survival, providing protection from harm by eliciting "an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage" (Raja et al., 2020, p. 14). Whilst the sensory aspect of pain is deemed as a change in physical state, its emotional experience is associated with a change in affective state (Finan et al., 2022; Hanssen et al., 2017; Price, 2000; Wiech & Tracey, 2009). The affective change often creates a negative internal shift which can initiate innate motivational drives to seek relief and facilitate movement towards a positive affect (Navratilova & Porreca, 2014; Porreca & Navratilova, 2017). Additionally, these innate drives allow us to learn about environmental dangers, prevent sustained tissue damage, promote behavioural analgesia by retracting from the 'site', or 'stimulus' of pain, and even seek external methods of pain relief such as medical care (Moayedi & Davis, 2013). However, there are many factors to consider beyond this primary mechanistic understanding of pain. To understand the whole experience of pain, we must work in theoretical frameworks that consider modulators and moderators which can alter these pain experiences like psychological, cognitive, and social components.

1.2.2 Theories of Pain

There are multiple theories to explain how pain is perceived and experienced. Early theories like Descartes et al.'s (1662) Cartesian Dualistic Theory, focused solely on physiology attributing pain sensations because of physical injury, and mutually exclusive from

psychological injury (Trachsel et al., 2023). However, with developments of modern science, new theories recognising a multifaceted perspective of pain emerged as dominant considering more than just these physiological inputs. At the initial forefront was Melzack and Wall's (1965) Gate-Control Theory, which recognised similar nociceptive mechanisms as early theories but offered an additional construct: a "gate". The "gate" is thought to originate within the dorsal horn, allowing neural transmission to the brain when "open" for pain facilitation, and preventing neural transmission to the brain when "closed" for pain inhibition (Melzack & Wall, 1965). Assisting in controlling whether the "gate" opens or closes required the vital introduction of cognitive, and psychological components alongside the established physiological input (Melzack, 1996; Melzack & Wall, 1965).

Contextualised to the Gate-Control Theory, physiological input facilitates pain perception via the strength of neural stimulation, with larger injuries invoking stronger neural stimulation to "open" the gate and allow for greater pain percept's (Melzack & Wall, 1965). As for inhibiting pain, medication providing analgesic effects assist in "closing" the gate – however this is of less relevance to the current thesis (Blieden et al., 2014; Corder et al., 2018; Kirkpatrick et al., 2015; Pathan & Williams, 2012). Yet with the additional recognition of cognitive and psychological components, bidirectional relationships are discussed in further facilitating the "opening", or "closing" of the gate. For example, cognitive states can inhibit pain by "closing" the gate and decrease pain sensitivity by diverting attention away or distracting someone from their pain through the likes of attention focused tasks, and neural stimulation such as touch (Eccleston, 1995; Hoegh et al., 2019; Kammers et al., 2010; Khera & Rangasamy, 2021; Mancini et al., 2014; Sloan & Hollins, 2017). However existing as a bidirectional relationship, pain itself can inhibit attention through its demanding nature calling for individuals to take notice of its presence. This demand allows for the gate to remain "open", with evidence demonstrating pain's ability to interfere cognitively with individuals in pain displaying limited ability to maintain their attention towards focus-demanding tasks like the n-back (Moore et al., 2012; Villemure & Bushnell, 2002). Moreover, psychological factors which contribute to the "opening" and "closing" of this gate include past experiences of pain and their associated affective components (Linton & Shaw, 2011; Melzack & Wall, 1965). For example, research suggests previous pain experiences can facilitate current pain perception as

individuals who possess greater histories of pain and potentially negative emotions towards pain tend to exhibit lower pain tolerances (Gentsch & Kuehn, 2022; Rollman et al., 2004; Yoo et al., 2023). This suggests emotions which are a product of past pain or consequential to reliving pain, play a role in allowing this gate to "open" at a much lower level of neural stimulation than others. However, these psychological and cognitive components are not exhaustive – an array of additional psychological and cognitive considerations such as pain catastrophizing, fear of pain, memory and information processing will be discussed throughout the empirical research studies conducted within this thesis.

Whilst the physiological, psychological and cognitive driven aspects of pain perception, facilitation and inhibition are provided by the Gate-Control Theory, their explanations fail to incorporate the social nature of pain highlighted by Raja et al. (2020). Instead, the Biopsychosocial model addresses this gap with a heuristic approach to pain that recognises how our environment influences our internal state to create a rounded experience of pain as opposed to a mere neural somatosensation (Gatchel et al., 2007). Loeser's (1980) Biopsychosocial model of pain arguably achieves the latter, providing 4 dimensions which interact to define one's pain experience:

(1) Nociception - the use of nociceptive mechanisms to detect tissue damage,

(2) Pain - the perception created from processing a noxious stimulus,

(3) Suffering - the negative psychological and affective state induced by pain including fear and anxiety,

(4) Pain behaviour – an individual's response to a pain suffering. For example, displaying facial expressions like grimacing, or actively seeking relief through medication and resting.

Loeser (2006) described their Biopsychosocial model of pain as an "onion-skin pattern" reinforcing nociception as central to initiating pain mechanisms, followed by pain and suffering as additional internal processes that cannot externally be observed. The outermost layer provides an outward expression of pain through pain behaviour; a means for external observers to objectify the individuals internal pain experience (Loeser, 2006). Despite self-describing as a Biopsychosocial model, Loeser's (1980) model dilutes the observer

objectification of pain to recognising *"things a person says, does, or does not do"* (Loeser, 2000), reducing the complexity of dyadic social communication to a summative behavioural performance of *"is it there, or not?"*. This summation means distinct categories of communication such as verbal vs non-verbal cues are overlooked limiting bystander interpretations of how to respond. Collectively these limitations suggest Loeser's (1980) Biopsychosocial model provides a limited view of the role social components play in the experience of pain. Instead, Craig's (2015) Social Communication Model of Pain which explicitly incorporates the entirety of biological, psychological, behavioural and socio-communicative components is preferred.

Craig's (2015) Social Communication Model of Pain utilises a timeline to visualise the differing physical, intra- and inter-personal factors that influence pain perception. These include Antecedents, Pain Experiences, Pain Expression, Decoding Pain Expression, and Action; each recognise that social, economic, and physical ecological contexts influence how bystanders care for others in pain.

(1) *Antecedents* exist prior to a painful event and anticipate how an individual initially reacts to pain. These are determined by predisposition factors like genetics, fear of pain, catastrophizing and health (intrapersonal), and stress and expectancy cues (interpersonal).

(2) Replicable to previous models, *pain experiences* are defined by an array of sensory, emotional, cognitive, social, and motivational features which affect how pain is perceived. Influences on this perception recognise involvement of social components such as the meaning of pain for the individual (intrapersonal), and vicarious cues to indicate a need for distress (interpersonal).

(3) Once perceived, an individual *expresses their pain* spontaneously or deliberately utilising, for example, verbal and non-verbal cues. Decisions regarding how internal pain experiences are externally expressed and communicated involve modulation from an individual's social abilities (intrapersonal), or perceived consequences of their social environment (interpersonal).

The latter 3 components describe an individual's own suffering highlighting the personal processing of pain. The final 2 components of the model instead describe how bystanders can observe, process, and appraise another's pain.

(4) First, bystanders *decode these pain expressions* to produce their own reactions (e.g. empathy and distress, or pain appraisal). Determining factors of this encoding involve the bystanders own stress levels, ability to attend and clinical judgements (intrapersonal), and social relationships to the individual experiencing pain (interpersonal).

(5) This decoding allows for an assessment to identify appropriate *actions* and responses, whether that be to remain indifferent to pain or provide care. Decisions may depend on the bystander's previous experiences with pain (intrapersonal) and their understanding of current professional best practices to respond to pain (interpersonal).

In recognising the importance of social determinants, this model supports existing evidence that suggests social factors like access to social support and satisfaction with social participation can alter pain intensity (Donaghy et al., 2022; McClelland & McCubbin, 2008). Moreover, the inclusion of bystanders within Craig's (2015) model provides further strengths in understanding pain communication, demonstrating humans innate behavioural need to seek relief and the dual interpretation of pain by the self and others in facilitating the latter. Yet like previous models, Craig's (2015) ability to describe the role of social involvement for Autistic individuals may be limited by differences between Autistic and non-Autistic social intentions and expressions. For example, the intent of an Autistic individual to seek social interaction may differ from a non-Autistic individual. Here, the application of the model may be limited in use to bystanders who know the Autistic individual personally and are thus best able to socially perceive their pain; particularly a caregiver who is often key for understanding their CYP's needs (Ely et al., 2016; Kalingel-Levi et al., 2022). In this context it is likely the interpersonal caregiver-CYP relationship produces an innate understanding of outward pain expressions that are specific to the child and perhaps different from what someone such as an HCP may expect. Additionally, as experts in their CYP's pain, caregivers are likely to be best suited to disclose their CYP's previous experience of pain,

with the additional awareness of how to best seek the relevant clinical judgement (Smith-Young et al., 2022). However, consideration for the quality, and presence of a caregiver-CYP relationship is needed when placing caregivers as experts in their CYP's pain. For example, some CYP may experience negative relationships with their caregiver(s) for a multitude of reasons including, but not limited to, experiences of abuse. In these instances, CYP may be less inclined to outwardly express their pain around their caregiver, or even HCPs (Drouineau et al., 2017). Therefore, whilst an innate understanding between a caregiver and CYP can exist, consideration must be given to how the dynamics of these interpersonal relationships effect a CYP's pain expression.

In conclusion, Craig's (2015) Social Communication Model of Pain arguably presents the most rounded view of pain which clearly aligned with Raja et al.'s (2020) componential definition. However, given its application to the neurodivergent population may be less valid, understanding the social nature context of neurodivergent pain experience may provide a nuanced insight, rather than trying to mould a neurotypical perception around that of a neurodivergent perception.

1.2.3 Paediatric Pain

For many years, paediatric pain has been neglected, overlooked, and mismanaged. These unfortunate realities can be explained through the continued application of paediatric pain myths highlighted respectively by Loizzo et al.'s (2009) and Twycross' (1998) reviews. For example, for many years professionals believed there was no need to understand or manage paediatric pain as they assumed CYP (particularly infants and toddlers) do not feel pain; or if they did feel pain, they would not remember it later in life. Statistics alone counter these myths, demonstrating that paediatric pain most definitely exists. For example, evidence shows experiences of pain in early childhood can predict development of chronic pain in later life (Palermo, 2020). Additionally, developments within paediatric research have provided extensive understanding for the presentation, and assessments of paediatric pain; even informing the first global set of guidelines for paediatric management in 2023 (Health Standards Organization, 2023). With this recognised need for paediatric research, the 2021

Lancet Child & Adolescent Health Commission (Eccleston et al., 2021) also presented guidance in how research can be directed to improve the understanding of, and care for, paediatric pain through four transformative goals: (1) make pain matter, (2) make pain understood, (3) make pain visible, and (4) make pain better. Available paediatric research has inexplicably addressed goal 3 of the 2021 Lancet Child & Adolescent Health Commission (Eccleston et al., 2021), creating overwhelming guidance of how individuals should assess pain in CYP.

When assessing a CYP's pain, evidence reaches a consensus in emphasising the importance of listening to what a child says, observing what they do, and interpreting how their body reacts (Palermo et al., 2010; Stinson & Jibb, 2013; Stinson & McGrath, 2010; Webb & Sanders, 2020; Wong et al., 2012). For example, Baker and Wong (1987) coined the term "Q.U.E.S.T", presenting steps that each identify a component of a CYP's pain that must be continually assessed until the required method of pain relief is sought:

(*Q)uestion the Child* – Talk to the child about their pain; consider the language they are using and what this is telling you about their pain.

(U)se Pain Rating Tools – Use to help quantitatively understand what the child is feeling.

(E)valuate Behaviour – Consider differences in the child's behaviour (i.e. posture, appetite or sleeping patterns), expressions or reactions (i.e. rubbing site of pain) and their physiological responses (i.e. flushed or sweating).

(S)ensitise Parents – Ask parents about their child's medical history, current and previous experiences of pain and related behaviours and emotions to gain further clarification.

(T)ake Action – Once adequately assessing and reassessing, identify what should happen next to help manage the child's pain.

However, despite its vastness, this evidence base is predominantly based on neurotypical populations with a particular focus on chronic pain, leaving out a key intersect

of the paediatric population who are increasingly at risk of pain: the Autistic paediatric population. It could be argued that as most of our knowledge base comes from paediatric chronic pain, we should gain insight into Autistic paediatric pain given the two's co-occurring nature (Jones & Shivamurthy, 2022; Lipsker et al., 2018). However, this alone is not enough – research dedicated to the Autistic paediatric population must be conducted to create a fundamental and inclusive understanding. This is something even the World Health Organization (2020) and the 2021 Lancet Child & Adolescent Health Commission (Eccleston et al., 2021) have highlighted, calling for the funding of research studying pain in neurodivergent CYP to ensure we can address this health inequity. Thus, there is no debating the direction future paediatric pain research must follow - furthering our understanding of how Autistic CYP experience pain, and the methods of assessments we can utilise to acknowledge this.

1.2.4 Pain Communication

Methods to socially communicate, and assess pain consist of either verbalisation through pain description and non-verbalisations through pain behaviours and facial expressions (Craig, 1992). Considering the Autistic population may use different methods of verbal and non-verbal communication than the non-Autistic population, the role of pain communication needs clear consideration for neurodivergent applications. Here, how we initially understand pain, and an in-depth consideration of the communicative and assessment basis which allow others to understand our pain will be discussed. Evidence will be presented mostly in the context of paediatric pain as the focus of this thesis, however broader literature including that of adult populations will be considered.

1.2.4.1 Verbal Communication of Pain

When someone recognises an individual is in pain, it is common they ask questions to allow the individual to verbally express their pain (Baker & Wong, 1987; National institute for Health and Care Excellence (NICE), 2021b). These verbalisations of pain are referred to as self-reports and are deemed as gold standard for the assessment of pain to gain

qualitative insight into its subjective nature (Haefeli & Elfering, 2006; National institute for Health and Care Excellence (NICE), 2021b; Raja et al., 2020; The Royal Children's Hospital, 2022). Typically, these verbalisations are most utilised in pain management settings requiring an individual to states or indicate pain presence, location, duration and quality (Swann, 2021). For example, a person reporting a throbbing pain at the front of their head at a low intensity would likely be interpreted by an HCP to be experiencing a tension-type headache. However, verbal reports alone run the risk of not providing enough descriptive information for bystanders to understand someone's experiences of pain, particularly within clinical settings where pain assessment are indicative of pain management efficacy (Nehemkis & Charter, 1984; von Baeyer, 2006). Here Fink (2000) presents a pain assessment approach named WILDA to address the breadth of description needed to understand pain, and begin to disentangle the complex, and subjective nature of pain in an objective manner. The acronym WILDA encompasses the differing descriptions a healthcare provider may need to understand an individual's pain; Words to describe pain: Intensity, Location, Duration, Aggravating and Alleviating factors (Fink, 2000).

1.2.4.1.1 Pain Description.

To understand and create a picture of what an individual in pain is experiencing and allow identification of the best course of action, a bystander (i.e., caregiver, teacher, healthcare provider) may rely on descriptive characteristics. Particularly in clinical settings, the acronym WILDA reflects descriptive elements used in both real-world and predominant pain management environments.

Words to describe pain allow for identification of pain nature and quality (i.e., understanding nociceptive pathways). In the WILDA (Fink, 2000) approach a Verbal Rating Scale (VRS) is implemented where patients select a set of descriptive words to reflect their current pain quality. These words are the same as those often used in real-world context, providing subjective pain understanding through verbal adjectives such as aching, stabbing and throbbing. Differing words attribute to differing pain qualities, for example those describing their pain as burning would be suggested to experience neuropathic pain, and those describing their pain as aching suggested to experience somatic pain (Fink, 2000).

Similar approaches of using descriptive natures to understand pain quality have existed prior to this approach, most notably that McGill Pain Questionnaire which presents 78 adjectives for pain at affective and sensory levels (Melzack, 1975; Melzack, 1987).

Yet the use of VRS alone may limit terminology use as individuals may not fully agree with the descriptors they see, or even understand what these descriptors mean for them. Here an in-depth conversation may allow for a more rounded description where the likes of metaphorical terminology can provide an initial in-depth visual image of pain quality by saying the pain "is" something abstract which is then discussed to provide meaning (Stewart & Ryan, 2019). For example, Camerota et al. (2023) conducted a linguistic analysis of how individuals with Ehlers-Danlos Syndrome (EDS) described their pain, with metaphors like "*a bomb bursting inside*" being used to ascribe pain intensity. However, evidence suggests that these descriptions may extend to explain the associated affective and behavioural states when in pain. Thus, further context must be provided with these descriptors to understand which aspects of the pain experience a metaphor is referencing (Munday et al., 2022). Similarly, like many standard communicative expectations of pain, the use of metaphorical language amongst the Autistic population may not be utilised as often. Research suggests Autistic people's use of metaphorical comprehension differs from non-Autistic peers with reports of lower comprehensive accuracy (Kalandadze et al., 2019; Morsanyi et al., 2020). Conflicting evidence is present, however, to suggest that levels of comprehensive accuracy may differ on an individual basis. For example, Autistic people have displayed no difference, or even increased efficacy in generating general metaphorical descriptions when compared to non-Autistic people with similar language abilities (Kalandadze et al., 2018; Kasirer & Mashal, 2016). Thus, a reliance on a metaphorical understanding of pain alone may not be applicable for the Autistic population overall, and if used in identifying experiences of pain, it must be on an individualistic basis.

To assess pain intensity when utilising WILDA, quantity assessments such as a 0-10 Numerical Rating Scale (NRS) with 0 ascribing to no pain, and 10 being the worst pain imaginable are used. Measures of intensity are, if anything, utilised as often as descriptive words when understanding pain with the concept that a discrete value can be attributed to pain severity. These scales often present as NRS whereby numbers are descriptive of pain

intensity, or Face Pain Scales whereby facial emotional displays expressing pain intensity like the Wong-Baker Faces Pain Rating Scale (Baker & Wong, 1987; Wong & Baker, 1988) and Face Pain Scale-Revised (FPS-R) (Bieri et al., 1990; Hicks et al., 2001). However, measures of intensity are often criticised in its ambiguous nature (i.e., what defines a 1/10 intensity) leading to subjective biases in ratings that struggle to be standardized across populations (Hjermstad et al., 2011; Morley, 2016). This is particularly concerning within the Autistic population who anecdotally express difficulties in ascribing a “face” or “numerical” value to an internal sensation, meaning reflections of these measures may not say much about an individual’s pain compared to more descriptive and qualitative (Bogdanova et al., 2022; Ely et al., 2016; Eveleth, n.d.). Thus, these limitations of intensity ratings mean sole reliance on these self-reports reduces the complexity of pain subjectivity.

Location provides understanding of where pain is within the body; however, validity of this can be impacted by interoceptive processes. For example, evidence suggests interoceptive abilities within chronic pain populations are not accurate thus presenting a question of where alone may not provide the most qualitative or correct responses (Di Lernia et al., 2016; Grabli et al., 2022). The latter question may also not be of relevance in cases of referred pain where pain is perceived in a location distant from the site of injury, thus interoceptive abilities here may appear limited despite the differing localisation of pain-injury site (Jin et al., 2023; Murray, 2009).

Further questions about duration may provide insight into the chronicity of pain with additional understanding of aggravating and alleviating factors pain presenting again the multifaceted view of an individual’s pain (Fink, 2000; Raja et al., 2020). Individuals may be asked further aspects about how this pain is affecting their psychological and cognitive experiences to understand this bidirectional modulatory aspect of nociception; however, this modulatory perspective is not recognised explicitly in the current acronym (Fink, 2000).

When used in its entirety, WILDA can provide a full description of pain. However, even addressing collective parts allows for some level of understanding about an individual’s subjective pain experience (Fink, 2000). It may be particularly useful to take parts of WILDA and adapt an approach that’s inclusive of neurodivergent population as if a VRS for example was solely used, some Autistic individuals may have difficulties engaging with the

terminology used. Instead, WILDA may just highlight the key points we should consider when we ask an individual for a description of their pain. Perhaps we as bystanders, HCPs, and even as the PhD researcher of this thesis, must implement adaptations to reflect the individual to truly understand descriptive aspects of their pain as supported by the Health Standard Organization's (2023) guidelines.

1.2.4.2 Non-Verbal Communication of Pain

Often pain can cause individuals to experience difficulties in verbalising sensations for an array of reasons (i.e. age, development or being non-verbal), similarly these verbalisations may not be reflective of the full pain experience when considered alone (Herr et al., 2011; O'Rourke, 2004; Stanford et al., 2006; von Baeyer, 2006; von Baeyer et al., 2011). Therefore, non-verbal pain expressions can be utilised to gain an understanding of whether an individual is in pain, and even pain intensity (Craig, 2009; Craig, 2015). Below the role of pain behaviour, and facial expressions as a mean of communicating pain non-verbally will be discussed.

1.2.4.2.1 Pain Behaviour.

Behaviour is a means of communicating pain through actions (Visser & Davies, 2009). These behaviours serve a multitude of functions including protecting from pain, regulating emotions, and communicating pain severity (Akbari et al., 2020). Changes to existing behaviour and emergence of new behaviours can be indicative of the presence of new pain, or increases in pain intensity (Mathews, 2011). Exemplar emerging behaviours may include protecting or rubbing the site of pain, new social support behaviours such as crying to indicate a need for help, and changes in old behaviour such as choosing to not engage in enjoyable activities (Dueñas et al., 2016; Grabovac & Dorner, 2019; Martel et al., 2012). However, behavioural displays heavily depend on factors such as age, and developmental stage with clinicians and researchers demonstrating a need to base assessment use on such factors (Birnie et al., 2019; Mathews, 2011; Sansone et al., 2023). For example, early research from Gilbert-MacLeod et al. (2000) found behavioural

differences between CYP without developmental delays, and CYP with developmental delays when experiencing pain. Such differences include CYP without developmental delays crying more, showing intense distress, and displaying more social responses such as increased help-seeking behaviours and withdrawal comparative to CYP with developmental delays. This demonstrates how differing CYP display pain differently and highlights a need for an adaptable assessment - however this topic will be discussed further in the following evidence presented.

Observational assessments assess a set of standardised behaviours which can be monitored to provide clinical and day-to-day understanding of if a CYP is in pain. Face-Legs-Activity-Cry-Consolability scale (FLACC) is a common observational assessment used to identify pain in children, with behavioural movements such as kicking or drawing legs up, crying and squirming acting as behavioural markers for severe discomfort or pain (Merkel et al., 2002; Merkel et al., 1997). However, the breadth of behaviour in this assessment is very limited suggesting we may be overlooking important behaviours which could provide understanding of pain in specific populations (Crellin et al., 2015). The Non-Communicating Children's Pain Checklist-Revised (NCCPC-R) is an additional assessment often used in paediatric populations which in comparison to FLACC considers a larger number of behaviours (Breau, Finley, et al., 2002; Breau, McGrath, et al., 2002). These behaviours are split into 7 differing observational measures which an HCP may use to initially understand if an individual is in pain before interpreting the intensity. These behaviours are rated in terms of frequency in the past 2 hours with higher scores being indicative of higher pain intensities:

(1) Vocal. E.g., crying or screaming.

(2) Social. E.g., less interaction or seeking comfort.

(3) Facial. E.g., furrowed brow.

(4) Activity. E.g., less active movement.

(5) Body and limbs. E.g., flinching or protecting sites of pain.

(6) Physiological. E.g., shivering or holding breath.

(7) Eating/sleeping. E.g., eating less.

However, the validity of utilising these observational measures may be questioned in terms of their direct relation to pain, as opposed to something else. For example, seeking in

a social context may not always be indicative of pain, children may seek comfort for an array of reasons (i.e., to regulate emotions, or because they just like affective touch), vocalisations may be indicative of tiredness, eating and sleeping changes may be due to again anxieties or illness (Kiel et al., 2020; McMakin & Alfano, 2015). Here, contextual details may be needed to understand more about the reason for these expressions. Yet the most visually driven behavioural change highlighted by the NCCPC-R may be that of facial expressions which is discussed in vast more detail as a method of non-verbal pain communication.

1.2.4.2.2 Facial Expressions of Pain.

Facial expressions are indicative of an individual's pain with brow lower, eye squeeze, eye squint, cheek raise, nose wrinkle, upper lip raise, and facial grimaces recognised as facial coders of pain (Craig, 2009; Krahé et al., 2013; LeResche, 1982; Prkachin, 2009). Their use in defining an individual's pain appears applicable in understanding pain intensity and both affective and cognitive states with the likelihood, intensity and duration of a facial expression increasing with the perception of pain (Craig, 1992; Mieronkoski et al., 2020; Prkachin, 1992). However, the effectiveness of using these facial expressions to understand pain experiences are affected by a multitude of sociocultural factors. For example, for facial expressions to be identified as consequential to pain, social context of, and awareness of the cultural norms surrounding the pain-inducing environment or event is required (Dansie & Turk, 2013; Dildine et al., 2023).

Assessment tools for non-verbal pain cues often utilise understanding of pain through the face; a method particularly used in CYP with developmental differences where cognitive and vocabulary understanding are not at a threshold for valid self-report method use (Emerson & Bursch, 2020; Manocha & Taneja, 2016; O'Rourke, 2004). Factors which may drive these differences include developmental stages, age, and disability (Emerson & Bursch, 2020; Manocha & Taneja, 2016; O'Rourke, 2004). Recommended observational assessments where facial movements and expressions are coded to understand pain include the FLACC, but of more relevance here the Facial Action Coding System (FACS) (Brand & Al-Rais, 2019; Ekman & Friesen, 1976; Merkel et al., 1997; Rojo et al., 2015). Both recommended assessments rely on the bystander's own interpretation of these facial

movement and expressions, often making their judgement of whether a person is in pain in conjunction with the previously discussed verbal expressions where available. However, when these bystander beliefs of how pain is facially expressed are not congruent to that of the individual they are observing, they may wrongly assume pain is not present. Further, little is addressed here for how expressions habituate over time and their applicability for a neurodivergent population given they are designed from a global perspective as opposed to population-based perspective as highlighted by Noyek et al. (2023) in their most recent review. Moreover, if considering Milton's (2012) Double-Empathy Problem, bystanders conducting assessments of Autistic CYP may also need to be Autistic to ensure changes in facial expressions are more likely to be socially perceived as pain as opposed to a social response to something else. For example, Ebrahimpour et al. (2019) raise key points in their concept analysis that may allow a universal method of assessment as opposed to specifically focusing on chronic vs acute pain, per se, and opening differing methods of communication which have not been implemented prior.

Despite research having outlined well established assessments for paediatric pain, the continuation of understanding the communicative context of paediatric pain should allow for future adaptations or creation of assessments. Such assessments should aim to address these concerns and pose the question as to what relevancy these assessments and bystander reports hold in neurodivergent populations, particularly for those who are non-verbal where perhaps HCPs rely more heavily on these observational reports (Oberlander et al., 2010). For example, Ebrahimpour et al.'s (2019) work is highlighting the use of drawings to represent child's pain with discrete analyses allowing inference of discrete pain quality, symptoms, and location such as the colour red displaying discomfort with its position on the drawing indicating location of pain. This may provide wider use in paediatric populations who are non-verbal or prefer not to communicate verbally and perhaps address concerns previously discussed. However, this may still present validity concerns around if the interpretation of drawings is correctly represented as pain by a bystander. Thus, at current, validation of these assessments within neurodivergent populations are positioned as a vital need, not a mere want.

1.2.4.3 Acute Pain

When considering pain importance in neurotypical adults and CYP, two of the most common pain classifications often discussed are acute pain and chronic pain (Bonezzi et al., 2020). Acute pain adheres to the protective nature of pain; it is defined as a warning sign of disease or threat which responds to injury or illness and typically resolves within three months once its underlying cause is treated or healed (International Association for the Study of Pain, n.d.). Throughout this thesis acute pain will be of particular interest given its relevance in daily lives ranging from a stubbed toe to a migraine, however it is likely chronic pain may be mentioned given the prevalence of pain throughout the Autistic lifespan. Yet one challenge of understanding acute pain how to quantifiably measure its presence; in this thesis, pain thresholds and tolerance have been utilised as one measure to understand Autistic CYP's acute pain experiences.

1.2.4.3.1 Pain Threshold and Tolerance.

One option to understand pain mechanisms is through psychophysical assessments of pain thresholds and tolerances. Pain thresholds are defined as "the lowest intensity at which a given stimulus is perceived as painful" (Kanner, 2009). On the other hand, pain tolerances are defined as "the greatest level of pain that a subject is prepared to endure" (Kanner, 2009). Both involve the perception of pain; however, perceptual thresholds are the intensity which an individual first detects pain whilst perceptual tolerances are the intensity which an individual can no longer withstand pain.

Most common methods of assessing thresholds and tolerances in a quantitative and systematic procedure is through Quantitative Sensory Testing (QST). The standardised protocol used amongst adults was created by the German Research Network on Neuropathic Pain (DFNS) (Rolke et al., 2006) and is widely accepted as a valid method for understanding sensory thresholds amongst both clinical and research settings (Uddin & MacDermid, 2016). The protocol outlines a battery of 7 tests to assess 13 sensory parameters including thermal detection or pain thresholds with contact thermodes, and mechanical detection or pain thresholds with, for example, weighted pinprick mechanical

stimulators. However, adaptations of the protocol are regularly implemented within adult populations often including the omission or inclusion of certain batteries (i.e. utilising a cold-pressor task to assess cold-pain threshold and tolerance as opposed to a contact thermode (von Baeyer et al., 2005)). Despite the DFNS' standardised QST protocol, a standardised method of applying a QST protocol in a paediatric population remains limited, with the only real clear guidance being presented by Blankenburg et al. (2010). However, it must be noted more bodies of work are emerging to broach this topic. Further application of QST for neurodivergent CYP is sparse with recent work from Symons et al. (2022) providing some guidance, but still no clear standardised protocol that can be widely applied to a neurodivergent paediatric population is available. Without this protocol, a replicable and valid method to compare differences in pain thresholds and tolerances between neurotypical and neurodivergent populations cannot be conducted and thus the development in understanding neurodivergent pain experiences remains stunted.

Factors which affect differences in pain threshold and tolerance values have been studied at large with modulating factors previously discussed for pain inhibition and facilitation continuing to be prevalent. For example, experience of anxiety and depression associated to decreases in pain tolerance (Michaelides & Zis, 2019) and social presence of another person increasing pain threshold (Edwards et al., 2017). Evidence also suggests demographics account for variance in pain thresholds and tolerance with men generally displaying higher pain thresholds and tolerances than women and pain threshold generally increasing with age (Bartley & Fillingim, 2013; Bek et al., 2002; Fillingim et al., 2009; Lautenbacher et al., 2017; Schmitz et al., 2013; Woodrow et al., 1972). However, often for an experimenter to effectively identify a participant's pain threshold and tolerance value, a level of communication from the participant is required for the experimenter to understand their subjective experience. Implications of this communicative dependence are becoming clear within neurodivergent populations (including Autistic), where communication style may differ dependent of an array of factors (i.e. predominantly non-verbal, non-verbal in times of stress) (Barney et al., 2020). Features related to these communicative differences of pain in the Autistic community will be highlighted and explored further throughout this thesis.

1.3 Pain and Autism

When described through their defining characteristics, both Autism and pain have an overriding sensory element. For example, pain as an unpleasant sensory experience (Raja et al., 2020), and Autism's hyper- or hyposensitivity to sensory input (American Psychiatric Association, 2013). Therefore, as pain is a sensory experience itself, it might be expected that Autistic people would experience pain either more, or less intensely than neurotypical peers; which the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) suggests hyposensitivity in its Autism diagnostic criteria. Similarly, Autism is characterised through differences in emotional processing, cognitive function, and social-communication – all of which are critical for moderating the experience of pain, and pain expression (Prkachin & Craig, 1995; Raja et al., 2020; Turk & Flor, 1987; Williams & Craig, 2016). Thus, such differences in physiological, emotive, cognitive, behavioural and socio-communicative domains deemed critical for the modulation and moderation of pain may account for differences in Autistic pain experiences.

1.3.1 Researching Pain in Autism

Early evidence of Autistic pain experiences derived from anecdotal accounts assumed Autistic people did not experience pain given their lack of pain expression and behaviours (Clarke, 2015; Goldschmidt, 2016). Pertinent cases displaying a lack of pain in Autism include a girl playing in the snow without clothes, a boy who was not aware their hand was on a stove until they smelt burning and a boy who grabbed a hot frying pan but did not respond as expected (as presented in Moore's (2014) review). These accounts demonstrate a clear context where pain is expected, yet perception and behavioural displays of pain deviates from the norm. However, such cases cannot be reduced to account for the entire Autistic population's given the subjective nature of pain.

Use of anecdotal accounts as an explanation of pain insensitivity in Autistic people has since been discredited with systematic reviews of literature concluding that Autistic people may experience differing levels of pain sensitivity (Allely, 2013; Moore, 2014; Ortiz Rubio et al., 2023). Instead, a hyposensitivity approach to pain is utilised and reflected within

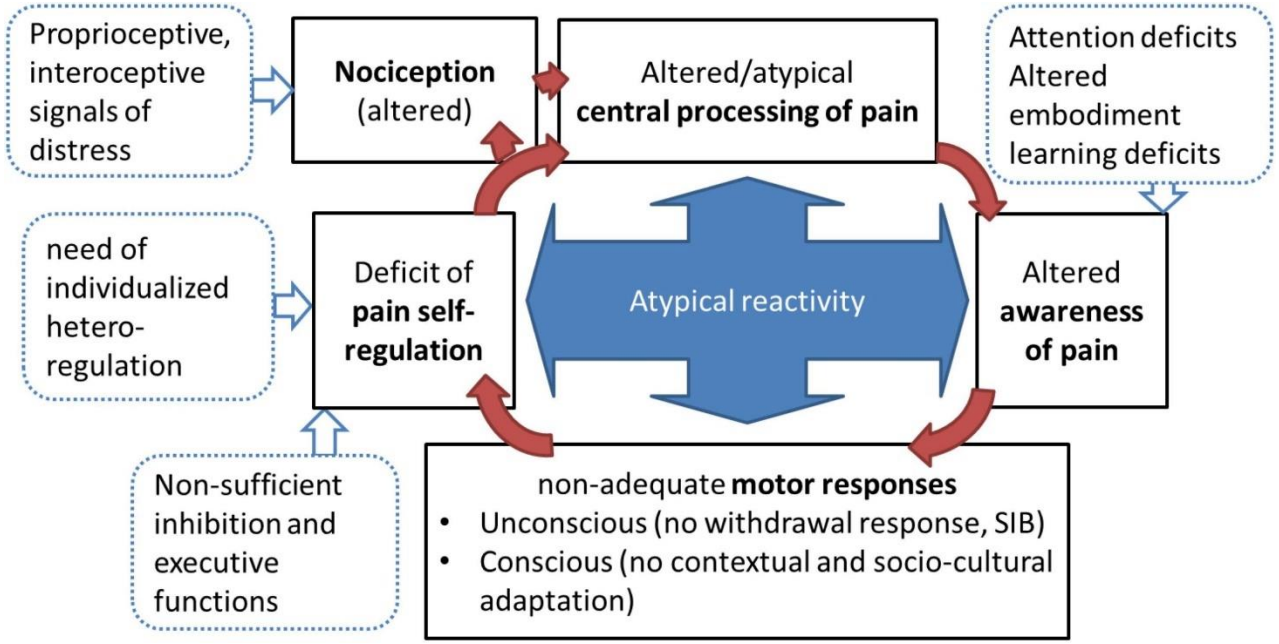
the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) with Autistic people perceived to display a decreased (hyposensitivity) sensitivity to pain. Evidence of Autistic pain hyposensitivity is present within the literature though not consistent. Differing experimental and survey-based studies highlight conflicting perspectives of pain hyposensitivity (i.e., higher pain thresholds) (Yasuda et al., 2016), hypersensitivity (i.e., lower pain thresholds) (Cascio et al., 2008; Hoffman et al., 2022) or no difference in response (Failla et al., 2020; Fründt et al., 2017; Vaughan et al., 2020) when compared to non-Autistic adults. However, additional psychological factors continue to modulate the direction of this sensitivity with higher levels of anxiety and fear contributing to higher pain intensity ratings even when threshold levels don't appear to differ (Failla et al., 2020; Failla et al., 2017; Garcia-Villamisar et al., 2019). Thus, studies considering pain in Autism must look past predominant physiological components and reconsider the interactions of the multifaceted components so regularly outlined (Raja et al., 2020).

More recently, Bogdanova et al. (2022) proposed a diagrammatic explanation described as an Integrated Model of the Pain Cycle in Autism to understand pain perception in Autism (see Figure 1). Bogdanova et al.'s (2022) model explores how differences in Autistic individuals pain perception may arise from differences in interactions between the perception, transmission, expression and modulation of pain. In particular, Bogdanova et al.'s (2022) model attributes observed differences in Autistic pain sensitivity to a change in neural relationships between altered self-awareness and interoception with pain processing and pain coping. Moreover, Bogdanova et al. (2022) explains in this model how methods for conscious pain appraisal and communication could be limited within Autism, particularly in early childhood where pain management can be dysregulated due to differences in pains physiological responses that in turn limits pain management and exacerbates potential increase in co-occurring mental health. Yet unlike Craig's (2015) Social Communication Model of Pain, how these factors interact to affect a different percept of pain in Autistic individuals are not clear and thus present limitations in the model's application. However, Bogdanova et al. (2022) themselves recognise further research is required to further develop this model; here clarity in the role of social factors may provide improvement given the emphasis of social communication broadly within Autism.

Whilst Bogdanova et al. (2022) proposed a model beginning to outline framework for a neural understanding of pain and its responses in Autistic people. The inclusion of pain appraisal, coping and communication begins to develop a multifaceted perspective of Autistic pain which initial models explicitly overlooked. Still, further developments are required to increase the model's explanatory use. Alas, our current understanding of pain in Autism remains limited to that of a basic consideration: Autistic people may have different experiences of pain.

Figure 1

The Integrated Model of the Pain Cycle in Autism



Note. A diagram of the "Integrated Model of the Pain Cycle in Autism" as proposed by Bogdanova et al. (2022). From "The Current View on the Paradox of Pain in Autism Spectrum Disorders," by O. V. Bogdanova et al., 2022, *Frontiers in Psychiatry, 13*, p. 13 (doi:10.3389). CC-BY 4.

1.3.2 Pain in Autistic Children

Autism and pain appear to regularly co-occur throughout childhood, with Autistic CYP being twice as likely to experience pain than non-Autistic peers (Whitney & Shapiro, 2019). Co-occurring pain statistics further support this with neurodivergent CYP (including Autistic) experiencing increased risk for and co-occurrences of both primary (i.e., chronic pain) and secondary pain conditions (i.e., Hypermobility-Syndrome Disorders (HSD), Ehlers-Danlos Syndrome (EDS) and Gastrointestinal (GI) conditions) (Donaghy et al., 2023; Jones & Shivamurthy, 2022; Margolis et al., 2019; Mazurek et al., 2014; Stallard et al., 2001; Trajkovski, 2018). Moreover, 14% of paediatric chronic pain cases in tertiary pain management settings involve Autistic CYP (defined as children aged 8-17), continuing to emphasise the clear prevalence of pain in the Autistic paediatric population (Lipsker et al., 2018). However, we still do not know why Autistic CYP frequently experience pain, but careful consideration for the pain mechanisms, and pain appraisal for Autistic CYP could be an important next step.

1.3.2.1 Pain Mechanisms in Autistic Children: Psychophysical Research

Given the high prevalence of pain conditions, it may be assumed Autistic CYP possess a hypersensitive pain profile rather than the diagnostically perceived hyposensitive pain profile. However, in measuring psychophysical pain thresholds of Autistic CYP, studies report both hypersensitivity (Fan et al., 2013; Riquelme et al., 2016) and hyposensitivity (Li et al., 2024). Similar inconsistent patterns in Autistic CYP's pain sensitivity profiles are presented within Posar and Viconti's (2018) review of Autistic paediatric pain, concluding the same reports of both hyper- and hyposensitive paediatric sensory profiles. These conflicts in sensitivity profile suggest not enough is known about the pain mechanisms of Autistic CYP to understand why pain is prevalent amongst this population, or how these experiences may differ from non-Autistic CYP for diagnostic hyposensitive pain profiles to be proposed. At this stage, additional psychophysical evidence is required to help understand the mechanistic direction of Autistic pain sensitivity and understand the clinical significance.

1.3.2.2 Appraising Autistic Children's Pain

It could be theorised that Autistic CYP experience more pain as their methods of pain communication differ from society's neurotypical understanding and thus limit the subsequent pain appraisal provided from bystanders. These can be discussed in both verbal, and non-verbal communication of pain.

Autistic CYP with or without intellectual disabilities are described within the paediatric literature to utilise similar verbal and descriptive self-reports of pain to non-Autistic CYP. For example, Autistic CYP are shown to describe pain, locate pain and answer questions regarding pain (Bandstra et al., 2012; Ely et al., 2016; Fitzpatrick et al., 2022). However, the level of description provided is suggested to differ, with Autistic CYP not focusing on pain duration, utilising simplistic language (i.e. hurt), and allowing parents to provide their input, interpretation and displays of trusted support (Benich et al., 2018; Ely et al., 2016). To provide additional insight into Autistic CYP's pain, non-verbal cues may be considered however paediatric literature suggests the direction of behavioural reactivity remain unclear. For example, some studies suggest there are no differences in frequency or reactivity (Dubois et al., 2017; Nader et al., 2004; Rattaz et al., 2013) and others suggest there are differences in the ability to express, or the duration of expression (Courtemanche et al., 2016; Daughters et al., 2007; Militerni et al., 2000; Tordjman et al., 2009).

Many of the described studies have reached their understanding through comparison to non-Autistic CYP. Whilst this provides insight into Autistic CYP's methods of pain communication, the design of using non-Autistic CYP's experiences as the comparative references immediately suggests Autistic CYP are differential to society's neurotypical preference. Additionally, a large emphasis has been placed on the communication methods of Autistic CYP, few studies have gained direct insight from Autistic CYP pertaining what their pain experiences are like to broadly understand important factors in shaping their experience, and how bystanders can correctly appraise their pain. For example, scaffolded approaches to facilitate Autistic paediatric pain communication may be absent from clinical practices, yet a caregiver may be required to articulate pain on an Autistic CYP behalf. Here understanding of Autistic CYP's broad pain experiences is vital to ensure their pain is valued in a neurodivergent space, as opposed to being viewed through a neurotypical lens.

1.3.2.3 What Are the Consequences of Overlooking Autistic Children's Pain?

Without comprehensive understanding of the mechanisms of Autistic CYP's pain, and how we can learn to appraise their pain, no valid evidence or guidelines can be utilised in the environments where they frequent to ensure pain is appraised and managed correctly. By failing to address the questions above adequately and in ample time, a population which are of higher risk in developing of pain will continue to experience a health inequity through no fault of their own and potentially face a chronic pain epidemic that could define the health of this population (Lipsker et al., 2018; Sharpe et al., 2019).

1.4 Brief Summary

Current Autistic literature remains inconsistent, with researchers and clinicians alike not understanding the Autistic experience of pain (see Theories of Pain in Autism and Pain in Autistic Children). Despite the breadth of research highlighting and distinguishing differences between Autistic and non-Autistic people (see Defining Autism), we continue to view Autistic pain through a neurotypical lens. By ascribing what we have learnt about pain experiences across neurotypes the subjective nature of pain, and the correct methods of pain assessment and management for Autistic people are overlooked; an ironic reality given the medical model's diagnostic criterion for Autism continues to highlight why these methods may not be as effective. It is this exact absence of Autistic based understanding within the pain arena which means a clear pain endemic is steadily progressing across all ages. We must understand how to tailor our knowledge to address the individual in front of us regardless of their positionality or intersectionality to pain, as currently we only know Autistic people are experiencing pain alarmingly more than non-Autistic people, yet we do not know:

(1) Why pain is more common within the Autistic community.

(2) If pain anecdotes and statistics correlate to pain hyper- or hyposensitivity.

(3) Which factors are important when addressing this health inequity.

The aim of this thesis is to examine factors which relate to pain experience and expression in Autistic CYP; a population who faces increased risk of chronic pain and lack of

effective pain management. To address this knowledge gap, a range of methods will be utilised to understand Autistic CYP's experiences of pain. First a systematic review of literature considering psychophysical parameters for testing experimental paediatric pain will be conducted to synthesise a psychophysical protocol for the following lab-based experimental study. This lab-based experimental study will utilise psychophysical methodology to measure mechanical pain threshold, pressure pain threshold, cold pain threshold, cold pain tolerance and associated pain intensity in both Autistic and non-Autistic CYP. Threshold, tolerance and intensity measures will be compared between the two groups to either support or oppose diagnostic criterion suggesting hyper- or hyposensitivity amongst the Autistic paediatric population. Next, separate interviews with Autistic CYP, followed by with their parent or guardian, will be conducted to gain a thorough understanding of the CYP's lived experiences of daily pain and the factors which may influence it. Finally, utilising the evidence-based landscape created through these methodologies, an online survey will capture an overarching caregiver perspective of their Autistic or non-Autistic child's pain from a larger and global participant pool. This survey will build particularly on interview themes in understanding difference in CYP's decisions to disclose pain to caregivers, teachers and HCPs. Throughout, methods will reflect the need for Autistic-based pain understanding, assessment, and management by ensuring the Autistic voice and lens is captured, included, and valued to make appropriate, and relevant healthcare adaptations. In doing so, findings will begin to provide an understanding of Autistic CYP's experiences of pain which can begin to dismantle the health inequity they face.

Chapter 2.

Psychophysical Methodologies to Measure Pain Responses in Children and Adolescents: A Systematic Review

2 Psychophysical Methodologies to Measure Pain Responses in Children and Adolescents: A Systematic Review

2.1 Introduction

For many years, a psychophysical approach to quantifiably measure pain has been applied in laboratory settings. Whilst considered a modern-day necessity in pain research, the approach is a product of Gustav Theodor Fechner's (1860) development of psychophysics and the self-titled formula: Fechner's law. Fechner's law proposes *"the magnitude of a sensation is proportional to the logarithm of the intensity of the stimulus causing it"* (Colman, 2015), emphasising the ability to quantifiably measure relationships between a noxious stimulus and reported pain response through psychophysical methods (Greenspan, 2009). Since Fechner, psychophysical methods have continued to develop allowing researchers to assess pain experimentally. In modern day, the DFNS' QST protocol (Rolke et al., 2006) provides the most standardised psychophysical methodology. This protocol outlines how to quantifiably measure differences in somatosensory functioning (i.e. pain and touch) between diagnostic groups and control reference values (Backonja et al., 2013). Although the DFNS' protocol consists of a battery of 7 tests to assess detection and thresholds across differing sensory modalities (Rolke et al., 2006), pain thresholds of relevance include:

(1) Thermal pain: cold pain thresholds (CPTh), heat pain thresholds (HPTh) and paradoxical heat sensations (PHS) measured using a contact thermode,

(2) Mechanical pain: mechanical pain thresholds (MPTh), mechanical pain sensitivity (MPS) and wind-up ratio (WUR) measured using weighted pinprick mechanical stimulators, and pressure pain threshold (PPTh) measured using a pressure gauge.

To experimentally measure a pain response using psychophysics, nociceptive mechanisms must be initiated to allow the encoding and processing of noxious stimuli for a report of pain perception (Dubin & Patapoutian, 2010; Institute of Medicine Committee on Pain & Chronic Illness, 1987; Wall et al., 1989). Initiation is ensued through application of noxious stimuli to the periphery (e.g. skin, muscle and joints) where primary afferent sensory

neurons transduce stimuli into electrical signals for transmission up specified central pathways for the encoding and processing of pain (Dubin & Patapoutian, 2010; Eilers & Schumacher, 2005; McEntire et al., 2016). The initial perception of pain is described as pain threshold and can be reported in differing ways, although in adult populations a method of self-report is suffice for threshold measures and subjective intensity for sensitivity measures (i.e. MPS and WUR) (Rolke et al., 2006). Contextualising thresholds to somatosensory functioning, references values in diagnostic-free adults are available for comparison to identify how pain mechanisms clinically differ between diagnostic groups (Rolke et al., 2006). However, whilst use of these psychophysical methodologies is well documented in adult populations (Vaughan et al., 2019), their use in assessing CYP's experimental pain remain an understudied field.

Pain psychophysical methods have been used in paediatric populations, yet no standardised protocol adapted for age is available. Decisions of pain methodological parameters are often at researcher discretion, with McGrath and Brown (2006) emphasising a need for developing standards to ensure precision of measures. Currently Blankenburg et al.'s (2010) modification of the DFNS' protocol for CYP is discussed as psychophysical guidance for assessment. Blankenburg et al. (2010) concluded the DFNS' protocol is feasible in assessing sensory thresholds in CYP aged 6-16 years, but wording of questions must be amended for comprehension. Additional studies support the concept of applying adult parameters to the paediatric population, only suggesting a requirement for identifying age-appropriate control reference values as opposed to utilising adult values (Hirschfeld et al., 2012; Magerl et al., 2010).

However, whilst DFNS parameters may be feasible, conclusions highlight wider ethical considerations that emphasise a requirement for adapting pain psychophysical methodology for CYP. For example, qualitative findings from Pate et al. (2019) suggest CYP's concept of pain is dependent on age, with younger CYP frequently responding "I don't know" when asked what pain is. Whilst Blankenburg et al. (2010) adapted questions to aid in understanding, included participants were not considered a clinical population with the explicit exclusion of CYP with chronic headache, and ADHD. Thus, a need for creating psychophysical scripts based on age, language comprehension and communicative

preference may be required to ensure pain threshold is correctly reported to increase data validity and minimise risk of tissue damage. Additionally suggested reliance on DFNS parameters alone cannot describe which psychophysical parameters are most precise in assessing paediatric pain thresholds as McGrath and Brown (2006) requested. Instead, reviewing when and how psychophysical parameters are used across broad paediatric literature may bridge this gap.

Discussions pertaining these parameters have recently emerged in the literature and have provided a partial evidence-based synthesis of psychophysics in CYP. For example, Tutelman et al. (2024) recently published a scoping review which began to provide paediatric insight for QST. Their summary of 301 studies described differing equipment and test-sites used in psychophysical testing and the feasibility of their use. Yet despite being referenced, only 8.31% studies used the DFNS which does not suggest Rolke et al. (2006) provides standardised methodological guidance for paediatric pain assessments, and there remains researcher discretion. Instead, consideration of additional psychophysical parameters may provide a better understanding of how to best assess paediatric pain thresholds. Birnie et al. (2012) provided an understanding of such psychophysical pain assessments, using methods separate from those described by the DFNS. In their systematic review, Birnie et al. (2012) described the use of the cold-pressor task (CPT) in assessing paediatric CPTh and cold pain tolerance (CPTol) – a well-documented method of pain assessment (von Baeyer et al., 2005). Birnie et al. (2012) considered the procedures and methodology used in CPT, highlighting pain threshold and tolerance as frequent outcomes, and continuing to outline clear methodological age-appropriate parameters regarding water temperatures and immersion time. For example, frequent use of 10°C ±1°C water temperature, however suggesting where tolerance is of interest water temperature should be below 10°C, particularly when children are over the age of 8 years (i.e. 5-7°C). To date, Birnie et al.'s (2012) review has been cited 118 times highlighting the benefit of synthesising in-depth methodological guidance.

Contextualised to the current thesis, Autistic CYP are twice as likely to experience pain than their non-Autistic peers yet understanding of why remains unclear (Whitney & Shapiro, 2019). Diagnostic criterion suggests differences in sensory responsivity amongst

Autistic CYP as an explanation, yet without understanding Autistic CYP's pain mechanisms these explanations remain theoretical. To examine if Autistic children differ to their non-Autistic peers on objective pain responses one option is to assess pain responses through psychophysics; an approach already used in Autistic adults by Vaughan et al. (2020). Whilst the latter reviews have provided initial recognition of what psychophysical methods are available for assessing CYP's pain and how their use effects findings, no study has summarised psychophysical parameters and ethical considerations for assessing pain in CYP with, and without a clinical diagnosis. Thus, before applying a psychophysical approach to measure Autistic CYP's pain, a systematic consideration of the ethical and methodological adaptations to facilitate safety, accessibility and inclusivity is required.

Given the lack of guidance on how to adapt psychophysical methods for paediatric populations, an understanding of the recommended parameters for measuring paediatric pain thresholds is required. Whilst existing reviews have explored psychophysical assessments (see Tutelman et al. (2024) and Birnie et al. (2012)), an evidence synthesis of both DFNS' and broad psychophysical parameters (i.e. the CPT) is not readily available. Without this synthesis, recommendations for the available and most appropriate psychophysical methodology remains overlooked. Moreover, the latter studies have not provided succinct insight into the ethical considerations needed for paediatric populations. Without this, samples representative of differing age, sex, and health status may not be included in psychophysical studies due to a lack of accessibility, or inclusivity (for example, Autistic CYP are often underrepresented in experimental research as highlighted in Nicolardi et al. (2023)). Therefore, to address this knowledge gap and outline a protocol for use in Chapter 3, the following research questions were identified as important in informing common methodological points: *What psychophysical methodology(s) are used when measuring child/adolescent pain thresholds?* Additional consideration of (1) *how methodological recommendations differ between age sex, and health status*, (2) *which methodology is most valid for measuring pain threshold*, and (3) *what ethical considerations are needed for paediatric populations*, will provide academics with a summarised understanding of how psychophysical methods can be adapted to safely measure CYP's pain.

2.2 Methodology

2.2.1 Protocol and Registration

An initial search on International Prospective Register of Systematic Reviews (PROSPERO) (https://www.crd.york.ac.uk/prospero/) confirmed similar reviews had not been registered. Review protocol was registered on PROSPERO on October 22nd, 2021 (ID: CRD42021281274) and a systematic search of existing literature focusing on psychophysics utilised in CYP was conducted. Protocol methodology followed Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Page et al., 2021) (http://www.prisma-statement.org/).

2.2.2 Search Strategy and Selection Process

An initial systematic literature search (1st) and an updated search (2nd) of article titles, abstracts and keywords were conducted in electronic databases PubMed, Web of Science and PsycINFO. Searches were time constrained (1st: database inception to 15-Jun-2021, 2nd: 16-Jun-2021 to 23-Oct-2023) and utilised a search strategy comprised of pain-related and population keywords (see Table 2).

Sourced references (including abstract and keywords) were exported to EndNote X9 for removal of duplicates and upon completion exported to Rayyan for eligibility screening of articles (Ouzzani et al., 2016). Article titles, abstracts and keywords were blindly assessed against inclusion criteria by the PhD researcher (B.D.) to assess eligibility, 10% of which were blindly assessed by a second member of the research team (Dr Emma Rheel, a postdoctoral researcher in physiotherapy and rehabilitation services (E.R.)) to audit decisions. The 10% of articles were selected by a random number generator (Randomness and Integrity Services Ltd., 2024), with the numerical value of the corresponding article determining its consideration. At full-text screening, 100% of articles were blindly assessed against inclusion criteria by B.D. and E.R.. Discrepancies in inclusion were discussed between both B.D. and E.R.; for articles where an amicable consensus was not reached a

full team discussion was arranged for a final decision (B.D., E.R.,T.V., and D.M.). Articles satisfying the following criteria were eligible for inclusion:

(1) Quantitative studies in humans aged 4-19 years old who were defined as healthy, clinically ill, or by diagnosis. An upper age limit of 19 years old was decided to adhere to the World Health Organization's (n.d.) specification of adolescence health (ages up to 19). Studies including participants aged over 19 were considered when grouped separately from participants aged 4-19 years (i.e. 17-19 years vs 20-22 years). Studies using animal models were excluded.

(2) Full-text available in English, French or Dutch. Abstracts, reviews, secondary analysis, longitudinal studies, and qualitative studies were excluded.

(3) Studies involving pain threshold measurement such as cold, heat or pressure pain. Cold pain tolerance was considered when additional pain thresholds were reported. Somatosensory articles obtaining threshold measurements other than pain (i.e. touch detection), and studies utilising methods of pain alleviation (i.e. anaesthesia) were excluded.

(4) Studies utilising at least one psychophysically sound pain assessment method, including calibrated needles, Von-Frey filaments, pressure algometers, thermodes, lasers (e.g. carbon dioxide and copper vapour), and cold-pressor task. However, CPT studies published before April-19-2012 were excluded due to likely inclusion in Birnie et al.'s (2012) review. Additional repeatable and robust psychophysical methods were considered when testing absolute rather than discriminative thresholds. Use of non-repeatable procedures (i.e. lumbar punctures and vaccinations) or methods without a standardised mode of pain induction (i.e. water load tests) were excluded.

Table 2

Database Search Strategy Keywords for Systematic Review

Topic	Keywords
Pain-Related	QST or “Quantitative Sensory Testing” or “experimental pain” or nociception or nociceptors or Aδ or A-delta or “c-fibre” or “c-fiber” or “thermal pain” or somatosensation or “pain thresholds” or “thermal detection” or “mechanical pain” or “dynamic mechanical allodynia” or “wind-up ratio” or “pressure pain” or electrocutaneous or “cold pressor” or “heat pain” or “pain sensitivity”
Population	Child** or pediatric or paediatric or adolescent or adolescence or youth or youngster or teen or infant

Note. This table provides the keywords used in each database search categorised by two topics: pain-related and population.

2.2.3 Data Extraction

Data from eligible articles was extracted into an Excel sheet independently by B.D. and shared amongst the team on BaseCamp 4 (37signals, 2024). Variables used for data extraction can be found in Table 3. Where data was missing referenced protocols were sought or authors were contacted for extraction. To ensure data was correctly extracted 10% of data extraction sheets were screened by a second member of the research team (D.M. or E.R.). A random number generator (Randomness and Integrity Services Ltd., 2024) was utilised to select the corresponding article number. Discrepancies in extraction were not recorded.

Table 3

Variables Used to Guide Data Extraction for Systematic Review

Variable Name	Data Extracted
Reference Information	Title, Author, Published Year and Country
Population Demographics	N Value, Diagnostic Information, Age (i.e. Mean, SD, and Range), Gender
Pain Induction	Methodology, Safety, Pain Induction Site (i.e. front arm), Written Description
Measurement Variables	Pain Measurement (i.e. VAS, NRS), Pain Report (i.e. self or observer), Mentioned Psychometric Measures
Design	Dependent Variable(s), Independent Variable(s), Co-Variates, Statistical Test(s)
Results	Summary of Findings
Ethics	Summary of Reported Ethics

Note. This table provides the differing datapoints extracted for each variable. Abbreviations used include SD = Standard Deviation; VAS = Visual Analogue Scale; NRS = Numerical Rating Scale.

2.2.4 Quality Assessment in Individual Studies

Two methods of assessing quality assessment concerned with methodical approach were conducted independently by B.D. and E.R. for 100% of the articles. Prior to beginning assessments, B.D. and E.R. independently decided whether studies were deemed as case-control, or cohort, with a meeting to discuss and resolve discrepancies. Independent assessments were conducted before discrepancies in ratings were discussed to reach a 100% agreement rate.

Final review studies with a clear distinction between 'case' group, and 'control' group were assessed using the NHLBI (2013) Quality Assessment of Case-Control Studies. Studies were scored against 12 different question criteria pertaining research question, study population, measures and analysis. A study received a “Yes” if the criteria were clearly met, “No” if the criteria were not clearly met, and either “Not Reported”, “Can’t Determine”, or “Not Applicable” if there was no clear distinction of criteria being met or not. Context of criteria 9 and 10 were adapted to reflect methodology for pain threshold, for example, having a clear established protocol. The number of “Yes” responses were totalled with a score of 0-4 indicating poor quality, 5-8 indicating fair quality, and 9-12 indicating good quality.

Final review studies with no clear distinction between a 'case' group, and 'control' group were assessed using the NHLBI (2013) Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies. Studies were scored against 14 different question criteria pertaining research question, study population, measures and analysis. A study received a “Yes” if the criteria were clearly met, “No” if the criteria were not clearly met, and either “Not Reported”, “Can’t Determine”, of “Not Applicable” if there was no clear distinction of criteria being met or not. The number of “Yes” responses were totalled with a score of 0-4 indicating poor quality, 5-10 indicating fair quality, and 11-14 indicating good quality.

2.2.5 Data Synthesis

Methodological summaries were synthesised using extracted data variables (see Table 3.). Summaries were grouped by a collective threshold measure (i.e. thermal

threshold), and sub-grouped by psychophysical equipment to their describe use in discrete pain threshold measurements (i.e. cold pain threshold).

2.3 Results

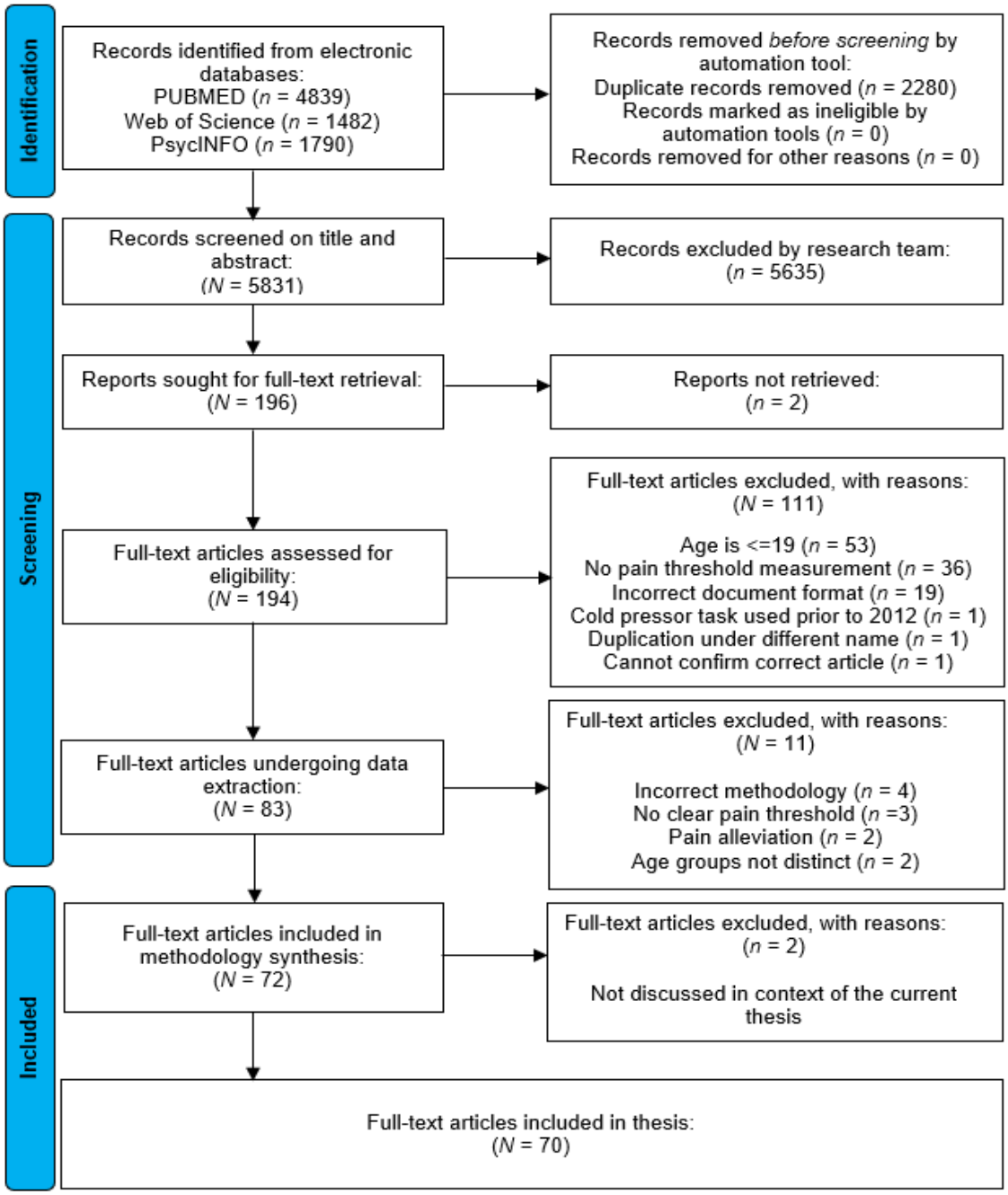
2.3.1 Study Selection

Following removal of duplicates, B.D. independently screened the titles and abstracts of 6614 articles against inclusion and exclusion criteria. A detailed flow chart of each search can be found in Figure 2 and Figure 3. Provided below is a detailed response of discrepancy values within study selection.

Of the 662 articles independently assessed for title and abstract eligibility; a 91% agreement rate was yielded. A total of 58 discrepancies (1st search: *n* = 56, 2nd search: *n* = 2) were discussed between the two resulting in a 100% mutual agreement without the need for full team discussion. Of the 254 articles independently screened for full-text eligibility, an 88% agreement rate was yielded. A total of 30 discrepancies (1st search: *n* = 16, 2nd search: *n* = 13) were again discussed by the two. A final consensus could not be met for 9 articles (1st search: *n* = 5, 2nd search: *n* = 4) which was discussed by the full team (B.D., E.R.,T.V., and D.M.). Following, 101 articles progressed to data extraction (1st search: *n* = 83, 2nd search: *n* = 18), where further exclusions were made upon analysing text in more detail (1st search: *n* = 11, 2nd search: *n* = 3). A total of 87 articles were therefore synthesised within this systematic review.

Figure 2

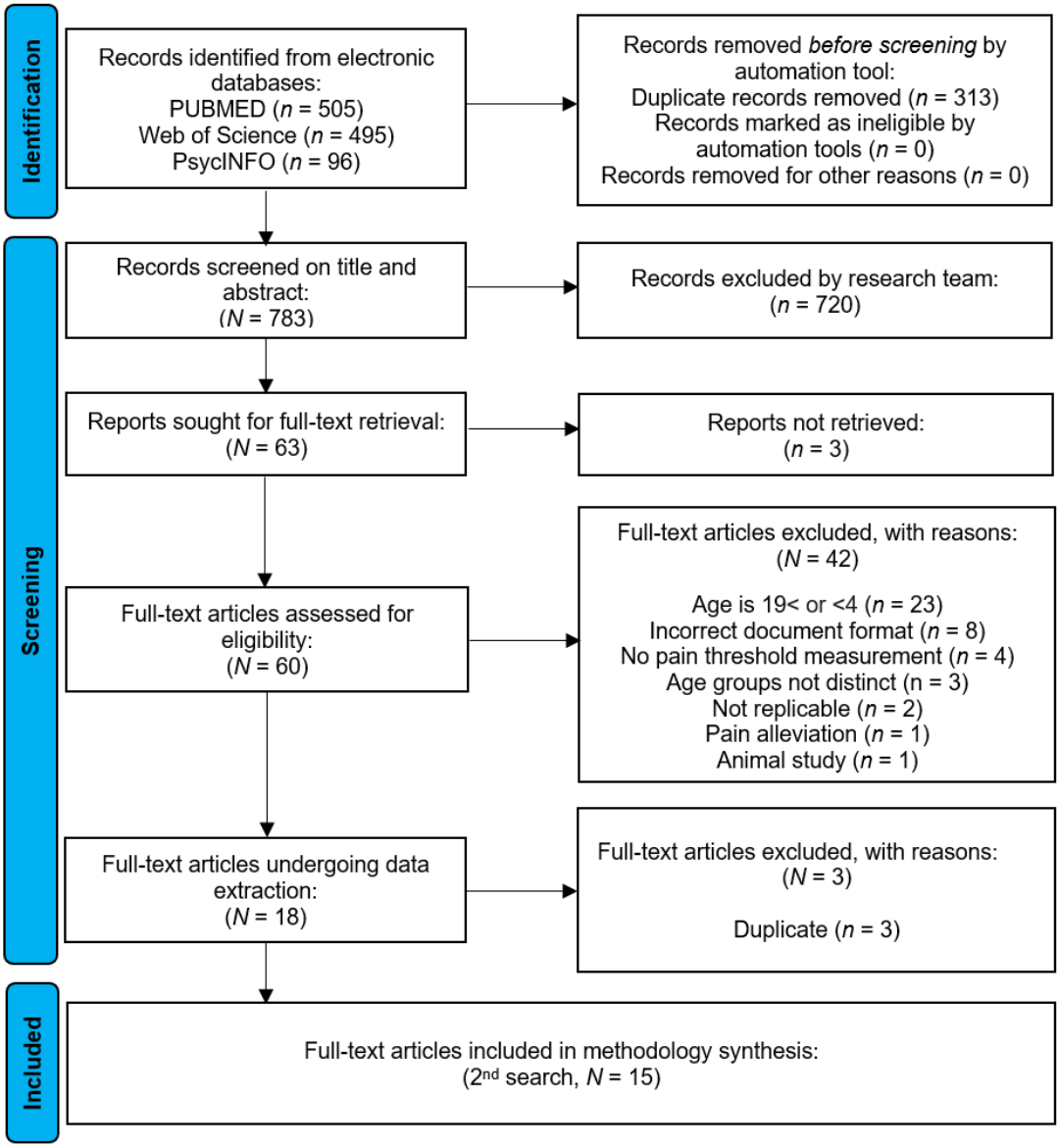
PRISMA Flow Diagram for the 1st Systematic Literature Search in Systematic Review



Note. A flow diagram amended from Page et al. (2021) to display the screening stages of the 1st systematic literature search. Adapted from "The PRISMA 2020 statement: an updated guideline for reporting systematic reviews," by M. J. Page et al., 2021, *BMJ, 372*(71). CC-BY 4.0.

Figure 3

PRISMA Flow Diagram for the 2nd Systematic Literature Search in Systematic Review



Note. A flow diagram amended from Page et al. (2021) to display the screening stages of the 2nd systematic literature search. Adapted from "The PRISMA 2020 statement: an updated guideline for reporting systematic reviews," by M. J. Page et al., 2021, *BMJ, 372*(71). CC-BY 4.0.

2.3.2 Quality Assessments

Results of quality assessments can be found in Table 4 for case-control studies, and Table 5 for cohort studies. Across all studies an 86% agreement rate was yielded between BD and ER. Discussions of discrepancies resulted in mutual agreement for 86 studies, with only one criterion for one article requiring full team discussion before reaching agreement.

The overall assessment across all studies yielded an 80.46% “Fair” rating, with both case-control and cohort or cross-sectional studies being rated mostly “Fair” (80.60% and 80% respectively). Only 10.34% of all studies were rated of “Good” quality”, however more cohort or cross-sectional studies received this rating (15%) than case-control studies (8.96%). A rating of “Poor” was only provided to 9.20% of studies, however case-control studies received more “Poor” ratings (10.45%) than cohort or cross-sectional studies (5%). Case-control studies typically omitted information regarding how cases and/or controls were selected, if controls were concurrent, and whether assessors were blind to the case/control status of the participant. Similarly, cohort or cross-sectional studies typically omitted information describing if assessors were blinded to participant diagnostic status. Given the prevalence of fair ratings, any bias in the reported methodologies is less likely to invalidate the interpretation in this review (NHLBI, 2013).

Table 4

NHLBI Quality Assessments for Included Case-Control Studies in Systematic Review

Reference (*n* = 67)	Quality Assessment	Q.1	Q.2	Q.3	Q.4	Q.5	Q.6	Q.7	Q.8	Q.9	Q.10	Q.11	Q.12
Abad et al. (2002)	Fair	Y	Y	N	NR	Y	Y	NA	NR	N	Y	NR	N
Alfvén (1993)	Fair	Y	Y	NR	Y	N	N	N	Y	Y	Y	Y	Y
Anttila et al. (2002)	Fair	Y	Y	Y	Y	N	Y	Y	NR	N	Y	Y	N
Balta and Arslan (2021)	Fair	Y	Y	N	Y	N	N	NR	NR	Y	Y	Y	N
Blankenburg et al. (2010)	Fair	Y	Y	N	Y	Y	Y	NR	NR	Y	N	NR	Y
Blankenburg et al. (2011)	Fair	Y	Y	Y	Y	Y	Y	NA	NR	Y	N	NR	Y
Blankenburg et al. (2012)	Fair	Y	Y	N	N	N	Y	NR	NR	Y	Y	NR	Y
Buskila et al. (2003)	Fair	Y	Y	N	N	N	Y	NR	CD	N	Y	NR	Y
Campi et al. (2020)	Fair	Y	Y	N	Y	Y	Y	NA	NR	Y	Y	NR	Y
Chae et al. (2007)	Fair	Y	Y	N	Y	N	Y	NR	NR	N	Y	NR	N
Chaves et al. (2013)	Fair	Y	Y	Y	Y	Y	Y	NA	NR	Y	Y	NR	N
Chen et al. (2000)	Poor	Y	N	N	Y	N	N	NR	NR	Y	N	N	Y
Cheng et al. (2012)	Fair	Y	Y	N	Y	Y	Y	NR	NR	N	Y	NR	N
Cornelissen et al. (2014)	Fair	Y	Y	Y	N	Y	Y	NR	N	Y	N	NR	Y
de Araújo Vitor et al. (2021)	Fair	Y	Y	Y	N	N	N	NR	CD	Y	Y	NR	N
de Tommaso et al. (2016)[a]	Fair	Y	Y	N	N	Y	Y	NR	CD	Y	N	Y	N
Duarte et al. (2000)	Fair	Y	Y	Y	Y	Y	Y	NR	N	N	Y	NR	N
Duerden et al. (2015)	Fair	Y	Y	N	N	N	Y	NR	NR	Y	Y	NR	Y
Engebretsen et al. (2018)	Fair	Y	Y	N	Y	N	Y	NA	NR	N	Y	N	Y
Fernández-de-Las-Peñas et al. (2010)	Good	Y	Y	Y	Y	Y	Y	NA	NR	N	Y	Y	Y
Fernández-de-Las-Peñas et al. (2011)	Good	Y	Y	N	Y	Y	Y	NR	CD	Y	Y	Y	Y
Fernández-Mayoralas et al. (2010)	Good	Y	Y	Y	Y	Y	Y	NA	NR	N	Y	Y	Y
Ferracini et al. (2014)	Fair	Y	Y	N	Y	Y	Y	NR	CD	Y	Y	N	Y
Goffaux et al. (2008)	Fair	Y	Y	N	N	Y	Y	NR	NR	N	Y	NR	Y
Gulewitsch et al. (2019)	Fair	Y	Y	N	Y	Y	Y	NR	NR	N	Y	NR	N
Habechian et al. (2018)	Fair	Y	Y	N	Y	Y	N	NR	NR	Y	Y	NR	N
Hainsworth et al. (2020)	Fair	Y	Y	Y	Y	Y	Y	N	NR	Y	Y	NR	N
Han et al. (2012)	Fair	Y	Y	N	Y	N	Y	NR	Y	N	Y	NR	N
Hashkes et al. (2004)	Fair	Y	Y	N	Y	N	Y	NR	NR	N	Y	N	Y
Hogeweg, Kuis, Huygen, et al. (1995)	Fair	Y	Y	N	N	N	N	NR	Y	Y	Y	Y	Y
Hogeweg, Kuis, Oostendorp, et al. (1995)	Fair	Y	N	Y	N	N	Y	NR	CD	Y	Y	Y	N
Hogeweg et al. (1996)	Fair	Y	Y	N	Y	N	Y	CD	NR	Y	Y	N	Y
Jedel et al. (2007)[b]	Fair	Y	Y	N	Y	N	N	NR	CD	Y	Y	NR	N
King et al. (2017)	Fair	Y	Y	Y	Y	Y	Y	NR	NR	Y	N	NR	Y
Lieber et al. (2018)	Poor	Y	N	N	N	N	N	NR	N	Y	N	NR	N
Ludäscher et al. (2015)	Fair	Y	Y	N	Y	Y	Y	NR	NR	Y	Y	NR	Y
Lyng et al. (2023)	Fair	Y	Y	N	Y	Y	Y	N	NR	Y	N	N	N
Mensink et al. (2022)	Poor	Y	Y	Y	N	N	N	N	NR	Y	N	NR	N
Metsahonkala et al. (2006)	Fair	Y	Y	N	Y	Y	N	Y	CD	N	Y	Y	Y
Morris et al. (2022)	Fair	Y	Y	N	Y	Y	Y	N	N	Y	Y	CD	Y
O'Leary et al. (2014)	Fair	Y	Y	Y	Y	Y	N	N	CD	Y	Y	NR	N
Pas et al. (2019)	Good	Y	Y	Y	Y	Y	Y	NR	CD	Y	Y	Y	N
Rathleff et al. (2013)	Good	Y	Y	Y	Y	Y	Y	Y	NR	Y	Y	NR	Y
Riquelme et al. (2016)	Fair	Y	Y	N	Y	Y	Y	NR	NR	Y	N	CD	Y
Riquelme et al. (2023)	Fair	Y	Y	N	Y	Y	Y	N	N	Y	Y	N	Y
Sá and Silva (2017)	Good	Y	Y	Y	Y	Y	Y	N	CD	Y	Y	N	Y
Sacramento et al. (2017)	Fair	Y	Y	Y	Y	Y	Y	N	CD	Y	Y	N	N
Saxena et al. (2015)	Poor	Y	N	N	NR	Y	Y	CD	CD	N	Y	NR	N
Scheper et al. (2017)	Fair	Y	Y	Y	N	Y	Y	NA	CD	Y	N	Y	Y
Schmitz et al. (2013)	Fair	Y	Y	N	Y	N	Y	N	CD	Y	Y	N	Y
Sethna et al. (2007)	Poor	Y	Y	N	N	N	N	N	N	Y	N	N	Y
Sherry and Sapp (2003)	Fair	Y	Y	N	N	Y	Y	Y	N	N	Y	NR	N
Soee et al. (2013)	Fair	Y	Y	N	N	Y	Y	N	NR	Y	N	N	Y
Stabell et al. (2014)	Fair	Y	Y	N	Y	Y	Y	N	CD	N	Y	NR	Y

Truffyn et al. (2021)	Fair	Y	Y	N	Y	Y	Y	NR	CD	N	N	NR	Y
Valentino et al. (2020)	Fair	Y	Y	Y	N	Y	Y	NR	CD	Y	Y	NR	Y
Valkenburg et al. (2015)	Poor	Y	N	N	N	N	N	NR	NR	Y	Y	N	Y
van der Venne et al. (2021)	Fair	Y	Y	N	N	Y	Y	NR	CD	N	CD	NR	Y
Vervoort et al. (2008)	Fair	Y	Y	N	Y	Y	Y	NR	NA	N	N	NR	Y
Weidenbacker et al. (1963)	Poor	Y	N	N	N	N	N	CD	CD	N	Y	CD	N
Weiss et al. (2014)	Fair	Y	Y	N	Y	Y	N	N	NR	N	Y	NR	N
Winger et al. (2014)	Fair	Y	Y	Y	Y	Y	N	NR	CD	Y	Y	N	N
Wollgarten-Hadamek et al. (2009)	Fair	Y	Y	N	Y	Y	Y	N	CD	Y	Y	NR	Y
Wollgarten-Hadamekl et al. (2011)	Fair	Y	Y	N	Y	Y	Y	N	CD	Y	Y	NR	N
Zohsel et al. (2006)	Fair	Y	Y	Y	Y	N	Y	NR	CD	Y	Y	NR	Y
Zohsel et al. (2008a)	Fair	Y	Y	N	Y	N	Y	NR	CD	Y	Y	NR	N
Zohsel et al. (2008b)	Fair	Y	Y	N	Y	Y	Y	NR	CD	Y	Y	NR	Y

Note. This table outlines the quality assessment decisions for each question on the NHLBI quality assessments for included case-control studies. Abbreviations used include Y = Yes; N = No; NR = Not Reported; NA = Not Applicable; and CD = Can't Determine. Q1 refers to research question, Q2-9 refers to the study population, Q10-11 refers to the measures, and Q12 refers to the statistical analysis.

[a] Discussed in broader systematic review, but not within this chapter due to sole use of laser-evoked potential.

[b] Discussed in broader systematic review, but not within this chapter due to sole use of electrocutaneous pain.

Table 5

NHLBI Quality Assessments for Included Cohort and Cross-Sectional Studies in Systematic Review

Reference (*n* = 20)	Quality Assessment	Q.1	Q.2	Q.3	Q.4	Q.5	Q.6	Q.7	Q.8	Q.9	Q.10	Q.11	Q.12	Q.13	Q.14
Buchanan and Midgley (1987)	Fair	N	N	NR	N	NR	Y	CD	Y	N	Y	Y	N	NA	Y
Cummins et al. (2021)	Fair	Y	Y	NR	Y	Y	Y	Y	Y	Y	N	Y	N	NA	Y
Derman et al. (2004)	Fair	Y	N	CD	Y	N	Y	N	Y	Y	N	N	Y	NA	N
Goubert et al. (2009)	Fair	Y	Y	Y	Y	Y	Y	N	NA	Y	N	Y	NA	NA	N
Hoehn et al. (2022)	Fair	Y	Y	Y	Y	Y	Y	Y	NA	NA	NA	Y	N	Y	Y
Jacob et al. (2015)	Fair	Y	Y	Y	Y	N	NA	N	Y	Y	N	Y	NR	NA	N
Jovellar-Isiegas et al. (2022)	Fair	Y	Y	NR	Y	Y	Y	Y	N	Y	N	Y	N	NA	Y
Kersch et al. (2020)	Fair	Y	Y	NR	Y	N	Y	N	Y	N	NA	Y	NR	NA	N
Kjeldgaard Pedersen et al. (2023)	Good	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	N
Leone et al. (2021)	Good	Y	Y	Y	Y	N	Y	Y	Y	Y	N	Y	N	Y	Y
Li et al. (2023)	Fair	Y	Y	NR	Y	N	Y	Y	Y	Y	N	Y	N	NA	Y
Marche et al. (2015)	Fair	Y	N	NR	N	N	Y	N	Y	Y	N	Y	NR	NA	N
Meier et al. (2001)	Poor	N	N	NR	Y	N	N	Y	N	N	Y	N	NA	NA	N
Nikolajsen et al. (2011)	Fair	Y	Y	Y	Y	N	Y	N	Y	CD	Y	Y	NR	NA	Y
Ocay et al. (2022)	Fair	Y	Y	Y	Y	N	Y	Y	Y	Y	N	Y	N	Y	N
Rheel et al. (2021)	Good	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	N	Y	Y
Richards et al. (2021)	Fair	Y	Y	Y	Y	Y	Y	Y	N	Y	N	Y	N	Y	N
Sieberg, Lunde, Shafrir, et al. (2023)	Fair	Y	Y	NR	Y	N	Y	Y	Y	Y	N	Y	N	NA	N
Sieberg, Lunde, Wong, et al. (2023)	Fair	Y	Y	NR	Y	N	Y	Y	NR	Y	N	Y	N	Y	N
van den Bosch et al. (2017)	Fair	Y	Y	Y	Y	N	Y	Y	Y	Y	N	Y	NR	N	Y

Note. This table outlines the quality assessment decisions for each question on the NHLBI Quality assessments for included cohort and cross-sectional studies. Abbreviations used include Y = Yes; N = No; NR = Not Reported; NA = Not Applicable; and CD = Can't Determine. Q1 refers to research question, Q2-5 refers to the study population, Q6-13 refers to the measures, and Q14 refers to the statistical analysis.

2.3.3 Study Characteristics

Of the 87 included studies, publication dates ranged between the years 1963 and 2023; 15 of which were published prior to Rolke et al. (2006). Excluding control values used by Cornelissen et al. (2014), Lieber et al. (2018), and Sethna et al. (2007) due to their use of existing reference values, approximately 11,026 CYP participated in at least one pain threshold assessment (~6115 females and ~4892 males). Inclusive age ranged between 4 and 19 years, with eight studies including an adult group: four assessing their pain thresholds (50%).

A total of 32 studies used more than one pain assessment specified in inclusion criteria, remaining studies were interested in PPTh (*n* = 47, including two studies described as MPTh), HPTh (*n* = 6), laser-evoked pain (LEP) (*n* = 1) and electrocutaneous pain (*n* = 1). Studies involving LEP (*n* = 2; 50% as a singular measure) and electrocutaneous pain (*n* = 1) are not included in the context of this thesis due to the low yield limiting synthesis in use. Overall studies compared pain threshold between diagnostic and comparator groups (66.67%), or assessed thresholds in control populations (14.94%), in specified diagnostic groups (10.34%), in athletic groups (2.30%), whilst observed by parents (3.45% - value does not include one observational study between diagnostic and comparator) and compared to an adult population (2.30%). Common diagnostic group diagnoses were pain-related (*n* = 44), neurodevelopmental (*n* = 9), neuropathic (*n* = 5), mental health (*n* = 5), orthopaedic (*n* = 2) and other (*n* = 2) (see Appendix 1 for categorisation).

2.3.3.1 General Ethical Considerations in Study Protocol

Ethical considerations in study design were generally discussed, yet only one study piloted their protocol in their target population prior to study commencement. Many studies required experienced or trained researchers for testing (*n* = 19), Li et al. (2023) was most intensive requiring 20 hours of training, a proficiency assessment, a QST mock and a bi-yearly training record review once testing begun. Before testing began, nine studies were selective of testing days to prevent fatigue and harm; six pain-related studies (i.e. a headache free day in four studies) and three pain-free studies (i.e. between 12am and 2pm

in female CYP aged 17-19 years for circadian rhythm). When conducting assessments, a calm testing environment was promoted (*n* = 12) with additional equipment and procedural familiarisation (*n* = 7). Standardised instructions were additionally provided (*n* = 11), however Valkenburg et al. (2015) created a comic book and plush animal with a contact thermode tail to promote inclusive and accessible understanding in CYP with down syndrome.

Despite the age range of CYP, only 15 studies discussed parental presence. There were conflicts in allowing parental presence (*n* = 7), preventing parental presence (*n* = 6), or providing CYP with autonomy in deciding their parental presence (*n* = 2). However, of the three studies involving Autistic CYP, two studies specified the need for adult presence to ensure safety in case of non-communication (Riquelme et al., 2016; Riquelme et al., 2023). Only 14 studies provided CYP with a reward for participating, two of which additionally reimbursed parents. Hoehn et al. (2022) promoted rewarding participants further, by providing incentive to individuals who referred participants and those who expressed participatory interest. Whether this assisted their recruitment remains unclear.

2.3.4 What Psychophysical Methodology(s) Are Used to Measure Children and Young People's Pain Thresholds?

2.3.4.1 Thermal Threshold

Within the included studies, thermal thresholds were assessed in 43.68% (*n* = 38). Of these 38 studies, HPTh (86.84%), CPTh (57.89%), CPTol (15.79%), and PHS (13.16%) were measured. Below psychophysical methodologies are discussed in more detail, except for PHS as this measure fails to provide thresholds.

2.3.4.1.1 Heat Pain Threshold.

2.3.4.1.1.1 Contact Thermode for Heat Pain Threshold

Of the 33 studies assessing HPTh (see Table 6), 32 used a contact thermode to induce heat pain, and one study a versatile cold/hot plate. However, use of the latter may

reflect ease in additional methodological blood withdrawal (van der Venne et al., 2021). Eight of these studies (24.24%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

There were ~4756 CYP aged between 6 and 19 years who participated in a study involving a HPTh assessment (~2667 females, ~2089 males). These values exclude control values utilised by Cornelissen et al. (2014), Lieber et al. (2018), and Sethna et al. (2007) as these represented reference values from a differing study. Whilst one study included an adult population (Sieberg, Lunde, Shafrir, et al., 2023), methods did not appear to adapt for a paediatric population. Most studies were interested in comparing HPTh between a diagnostic and control group (*n* = 22), or within a diagnostic group (*n* = 4). Pain-related diagnoses were most frequent (*n* = 14), with additional studies considering neuropathic diagnoses (*n* = 5), mental health diagnoses (*n* = 4), and neurodevelopmental diagnoses (*n* = 3). The remaining focused on control populations (*n* = 7). Methodological parameters typically referenced Rolke et al. (2006) (*n* = 9) and Blankenburg et al. (2010) (*n* = 6) as a standardised protocol followed.

Where a contact thermode size was reported (*n* = 31), 70.97% of studies reported use of a single thermode size of 9cm^2 (including semantic descriptions: 9cm^3, 30x30mm and 3x3cm). Four studies adapted thermode size to 16x16mm or 1.6cm^2 when testing over small sites including thenar eminence and/or trigeminal nerve (Cornelissen et al., 2014; Hainsworth et al., 2020; Wollgarten-Hadamek et al., 2009; Zohsel et al., 2006). Additional sizes included 1.6x3.6cm (Abad et al., 2002), 3.2cm^2 (Cummins et al., 2021) and 572.5mm^2 (Rheel et al., 2021). Yet despite 13 studies testing multiple sites of differing sizes, only O'Leary et al. (2014) and Zohsel et al. (2008b) used two differing thermodes; both a 9cm^2 (30x30mm) at volar forearm or abdomen, respectively, and 2.56cm^2 (16x16mm) at thenar eminence.

HPTh was measured at either 1 (*n* = 21; 28.57% bilateral and 4.76% quadrilateral), 2 (*n* = 11; 9.09% bilateral) or 3 (*n* = 1; 100% bilateral) sites. Where one site is reported, this value includes one study involving a control group site measured unilaterally, and a pain diagnostic group measured bilaterally (Mensink et al., 2022). Where two sites are reported,

this value includes two studies involving a control group being measured in one control site and a pain-related diagnostic group in two sites (one pain, and one control site (Ocay et al., 2022), or one bilateral pain site, and one control site (Truffyn et al., 2021)).

A full list of testing location can be found in the methodology section of Table 6, however broadly the hand (*n* = 16; 62.50% thenar eminence), and the forearm (*n* = 13, 30.77% inner dominant forearm and 23.08% inner non-dominant forearm) were tested most. For nine studies, testing sites were dependent on participants diagnostic experience (seven pain-related, one mental health, and one neuropathy). Diagnostic experience included adapting for pain diagnosis whereby control site was maintained between a diagnostic and control group, but pain site adapted to diagnosis (*n* = 7; 28.57% control data accessed from reference values). For example, Mensink et al. (2022) defined the right knee as a control site in their control group, however in the juvenile idiopathic arthritis (JIA) group pain site was defined as the knee “affected” by arthritis, and control site as the contralateral knee. As an additional example, Truffyn et al. (2021) defined the dominant volar forearm as a control site and an additional pain vs. contralateral pain site in a complex regional pain syndrome-1 (CRPS-1) group. Diagnostic experience additionally included ethical consideration of preventing harm (*n* = 2). For example, in a group of CYP with a mental health diagnosis Cummins et al. (2021) originally specified use of volar forearm but adapted to dorsal forearm or upper arm if there were scarring. Moreover, Lieber et al. (2018) altered location to prevent harm in a group with a neuropathic diagnosis, shifting from the dorsum of the feet to the dorsum of the hand dependent of clinical symptoms. Seven studies described blinding participants from viewing the computer monitor displaying values during testing (21.21%).

In the 31 studies reporting a baseline and maximum temperature, a 32°C baseline and 50°C maximum across age, gender, and diagnoses was most common (*n* = 27). Where maximum temperature differed from 50°C, diagnostic comparison or methodological difference was described. For example, only Morris et al. (2022), and Gulewitsch et al. (2019) included functional abdominal pain populations, both using a 32°C baseline but higher maximum at non-dominant forearm (51°C and 52.5°C respectively). This may suggest CYP with functional abdominal pain require higher temperatures when assessing HPTh or reflect Morris et al.’s (2022) CPM use; neither pattern can be affirmed. Additionally,

Jacob et al. (2015) increased maximum temperature to 52.5°C at the thenar eminence in sickle cell disease (SCD) populations, although this may be research discretion as O'Leary et al. (2014) assessed the same site and population but maintained maximum of 50°C. Rheel et al. (2021) increased maximum temperature over trials assessing HPTh from a 32°C baseline, with an initial 42°C target and increasing each trial target by 1°C if pain not rated moderate, whilst adhering to a 54°C maximum; however this reflected a need for moderate HPTh in an additional heat pain task. In contrast, a pattern in reported rates of increase was not observed as varied rates were reported (*n* = 33): 1°C/s (*n* = 20; 60.61%), 1.5°C/s (*n* = 8; 24.24%), 0.3°C/s (*n* = 1; 3.03%), 0.5°C/s (*n* = 1; 3.03%), 0.7°C/s (*n* = 1; 3.03%), 1°C over 13.3 seconds (*n* = 1; 3.03%), or 70°C/s over 300 milliseconds (*n* = 1; 3.03%).

Of the studies providing information on how HPTh was reported (*n* = 31), 100% required a method of self-report. However, Rheel et al. (2021) included verbalisation to understand if pain was mild, moderate, or severe and Ocay et al. (2022) a verbalisation of "stop". Remaining studies utilised a button once heat turned painful (*n* = 29; 93.55%). With button use, participants were instructed to press *(n* = 28; 96.55%) or release the button as Valkenburg et al. (2015) suggested in their study involving CYP with down-syndrome this method was easier for CYP to understand (*n* = 1; 3.45%). Truffyn et al. (2021) also provided ethical consideration for button use, training CYP with CRPS-1 and controls how to press the button. To guide on when the button should be pressed, four studies provided descriptive clarity using language to indicate pain quality such as "aching", "stinging" or "burning" (*n* = 3), or an age-appropriate description to participants providing context (*n* = 1). For example, Cornelissen et al. (2014) a likened heat pain to holding of a cup of hot coffee or hot chocolate for CYP aged between 9.6 and 15.5 years. Studies providing this clarity typically included a diagnostic group (neuropathy, *n* = 1 and pain-related, *n* = 1).

Typically, three HPTh trial values were recorded at each site (*n* = 19; 10.53% recorded the 1st value as a rehearsal and removed from analysis). However, four HPTh trial values (*n* = 2; 100% recorded 1st value as a rehearsal and removed from analysis), and five HPTh trial values (*n* = 8; 25% recorded 1st value as a rehearsal and removed from analysis) were also common. Additional numbers of trial values can be found in Table 6. Trial values were typically expressed as a mean across experimental trials (*n* = 29; 87.88%), a mean

change from baseline across three trials (*n* = 1; 3.03%), or temperature when pain (including moderate) was first reported (*n* = 3; 9.09%). Patterns in trial decision or threshold calculation were not clear across age, gender, or diagnoses.

Based on the synthesised evidence, evidence suggests that when using a contact thermode to measure HPTh, researchers record 3 trial values at the relevant testing site using a 9cm^2 thermode, and a 32°C baseline with a maximum of 50°C. Heat stimulation should stop as soon as the participant reports a sensation of pain, or the maximum value of 50°C is met.

2.3.4.1.2 Cold Pain Threshold.

2.3.4.1.2.1 Contact Thermode for Cold Pain Threshold.

Of the 22 studies assessing CPTh, 18 studies used a contact thermode and one study a thermal plate to induce cold pain (see Table 6). Eight of these studies (44.44%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

Excluding control values utilised by Cornelissen et al. (2014), Lieber et al. (2018), and Sethna et al. (2007) as these represented participants from a differing study, ~1417 CYP with an age range between 6 and 19 years participated in a study involving a CPTh assessment with a contact thermode (~815 females, ~602 males). Whilst one study included an adult population (Sieberg, Lunde, Shafrir, et al., 2023), methods did not appear to adapt for a paediatric population. Most studies compared CPTh between a diagnostic and control group (*n* = 13), others compared within a control group (*n* = 5) or a within diagnostic group (*n* = 1). Diagnoses included were pain-related (*n* = 4), neuropathic (*n* = 4), neurodevelopmental (*n* = 3), or mental health (*n* = 3). Methodological parameters typically referenced Rolke et al. (2006) (*n* = 7) and Blankenburg et al. (2010) (*n* = 4) as a standardised protocol followed.

Where contact thermode size was reported (*n* = 17), 82.35% of studies reported use of a single thermode size of 9cm^2 (including semantic descriptions: 9cm^3, 30x30mm and 3x3cm), 5.88% 16x16mm when testing over small sites including thenar eminence and joints or 5.88% utilising 3.2 cm^2. However, only O'Leary et al. (2014) used two differing thermodes

dependent on testing site size: 9cm^2 at volar forearm and 2.56cm^2 at thenar eminence. Thermode size was not reported in one study, nor was thermal plate size.

CPTh was measured at either one (*n* = 14; 50% bilateral and 7.14% quadrilateral) two (*n* = 4; 25% bilateral) or three (*n* = 1; 100% bilateral) sites. Where one site is reported, this value includes one study involving a control group site measured unilaterally, and a pain diagnostic group measured bilaterally (Mensink et al., 2022). Where two sites are reported, this value includes two studies involving a control group being measured in one control site and a pain-related diagnostic group in two sites (one bilateral pain site, and one control site (Truffyn et al., 2021)).

A full list of testing location can be found in the methodology section of Table 6, however broadly the hand (*n* = 11; 54.55% thenar eminence), the foot (*n* = 5; 66.67% dorsal), and the forearm (*n* = 4, 50% inner dominant) were used most. For six studies, testing sites were dependent on participants diagnostic experience (four pain-related, one mental health and one neuropathy). As reported in Chapter 2.3.4.1.1.1 diagnostic experience included locations that were affected by diagnosis such as in pain vs not (*n* = 4; 50% control data accessed from reference values) and ethical consideration of preventing harm (*n* = 2). Five studies described blinding participants from viewing the computer monitor displaying values during testing (26.32%).

All studies reported a minimum temperature of 0°C, with 18 studies stating a 32°C baseline and Riquelme et al. (2023) a baseline at participants thermal detection threshold. There were varied reported rates of decrease (*n* = 19): 1°C/s (*n* = 12; 63.16%) and 1.5°C/s (*n* = 7; 36.84%). Riquelme et al. (2023) additionally assessed a second CPTh measured in seconds, applying a constant temperature at 0°C with a ceiling of 180s, although discussed number of measures above and CPTh reports below remained consistent.

All studies provided information on how CPTh was reported. Use of a button once cold turned painful was most frequent (*n* = 18; 94.74%), with differences in participants being informed to press the button *(n* = 17) or release the button as Valkenburg et al. (2015) previously described (*n* = 1). Truffyn et al. (2021) ethically considered button use again, and four studies provided descriptive clarity on when to press the button, using same HPTh

language to indicate pain quality (*n* = 3), or age-appropriate, contextualised description to participants (*n* = 1). For example, Cornelissen et al. (2014) a likened cold pain to being like holding ice or a popsicle on your skin for CYP aged between 9.6 and 15.5 years. Studies providing this clarity typically included a diagnostic group (neuropathy, *n* = 1 and pain-related, *n* = 1). Riquelme et al. (2023) was the only study to ask Autistic CYP to remove their hand when the cold sensation became painful and allowed parents of Autistic CYP to observe signs of distress and stop procedure if pain were not communicated.

Typically, most studies reported that three CPTh trial values were recorded at each site (*n* = 17), however use of five CPTh trial values was also reported (*n* = 2; 100% recorded 1st value as a rehearsal and removed from analysis). CPTh values were expressed as a mean across trial trials (94.74%), or a mean change from baseline across three trial values (5.26%). Patterns in trial decision or threshold calculation were not clear across age, gender, or diagnoses.

Based on the synthesised evidence, evidence suggests that when using a contact thermode to measure CPTh, researchers record 3 trial values at the relevant testing site using a 9cm^2 thermode, and a 32°C baseline with a minimum of 0°C. Cold stimulation should stop as soon as the participant reports a sensation of pain, or the minimum value of 0°C is met.

Table 6

Included Studies in Systematic Review Measuring Heat Pain Thresholds or Cold Pain Thresholds with a Contact Thermode

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	HPTh Methodology	CPTh Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Abad et al. (2002)	1. Type-1 DM (*n* = 35, *M* = 13.6 (1.4), 18f, 17m) 2. Age-matched HC (*n* = 35, *M* = 12.4 (2.6),16f, 19m)	HPTh	CDTh WDTh	Contact Thermode (1.6 x 3.6cm)	Investigator applied thermode to dorsal surface of the right hand and foot Baseline: 40°C Max: - ROI: 0.7°C/s 7 measures Interstimulus interval: 10s	Not applicable	Pressed button at first sensation of pain	Mean of 7 measures	1. Assessment in a quiet room at 24°C 2. 2 stimuli used for habituation
2	Blankenburg et al. (2010)	1. HC (*N* = 176, 88f, 88m) Range: 6-16.12 yrs Age bands: 6-8 yrs (24f, 24m) 9-12.2 yrs (32f, 32m) 13-16.12 yrs (32f, 32m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Contact Thermode ($9cm^2$)	Thermode applied to both facial cheeks, dorsum of hands and feet in randomised order Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures MLI	Thermode applied to both facial cheeks, dorsum of hands and feet in randomised order Baseline: 32°C Min: 0°C ROD: 1°C/s 3 measures	Pressed button when felt “aching”, “stinging”, or “burning”.	Mean of 3 measures	1. 1 of 2 assessors who received 4-week training in DFNS centre for QST research 2. Demonstration of pain induction at practice area above test area 3. Participant decided whether parents stayed in room or outside 4. 4-hr sessions with short breaks if concentration declined 5. Monetary reimbursement
3	Blankenburg et al. (2011)	1. HC (*N* = 173) Age bands: 7 yrs (42f, 44m) 14 yrs (43f, 44m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Contact Thermode ($9cm^2$)	Thermode applied to both dorsum of hands in randomised order Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures MLI	Thermode applied to both dorsum of hands in randomised order Baseline: 32°C Min: 0°C ROD: 1°C/s 3 measures	Pressed button when felt “aching”, “stinging”, or “burning”.	Mean of 3 measures	1. Described to children presence and absence of pain as a likened to difference between sharp and blunt 2. Subjects decided whether their parents stayed in the testing room or outside 3. Monetary reimbursement
4	Blankenburg et al. (2012)	1. Type-1 DM (*n* = 45, *M* = 13.2 (2.8), 23f, 22m) 2. HC (*n* = 45, *M* = 13.2 (2.6), 23f, 22m age-gender matched)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Contact Thermode ($9cm^2$)	Thermode applied to both feet in randomised order Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures MLI	Thermode applied to both feet in randomised order Baseline: 32°C Min: 0°C ROD: 1°C/s 3 measures	Pressed button when felt “aching”, “stinging”, or “burning”.	Mean of 3 measures	1.. Monetary reimbursement
5	Cornelissen et al. (2014)	1. JIA (*n* = 60, *Mdn* = 13.0 (9.6 – 15.5), 44f, 16m)	MPTh PPTh CPTh HPTh	MDTh VDTh CDTh WDTh	Contact Thermode ($1.6cm^2$)	Whilst on a clinic bed, two sites for JIA: 1a) If active joint*: joint with most significant inflammation, swelling or tenderness	Whilst on a clinic bed, two sites for JIA: 1a) If active joint*: joint with most significant inflammation,	Pressed button when felt “feels so hot you don’t want it on your skin	Mean change from baseline across 3 trials	1. QST script 2. Participants provided time to adapt to room and equipment 3. Participant blinded from monitors

		2a. US HC (n = 92, *Mdn* = 13.0 (10.0 – 13.8), 50% f)* 2b. EU HC (n = 151, *Mdn* = 11.0 (9.0 – 14.0), 50% f)** *Obtained from Meier et al. (2001) **Obtained from Blankenburg et al. (2010)					1b) If no active joint, inactive joint*: joint with most inflammation in past 2) Control site: contralateral thenar eminence** Baseline: 32°C Max: 50°C ROI: 1.5°C/s Return to Baseline: 10°C/s 3 measures *Joint test site either distal or proximal interphalangeal joint, wrist, knee or ankle **Control data was pre-existing, comparing JIA thresholds to control thenar eminence	swelling or tenderness 1b) If no active joint, inactive joint*: joint with most inflammation in past 2) Control site: contralateral thenar eminence** Baseline: 32°C Min: 0°C ROD: 1.5°C/s Return to baseline: 10°C/s 3 measures *Joint test site either distal or proximal interphalangeal joint, wrist, knee or ankle **Control data was pre-existing, comparing JIA thresholds to control thenar eminence	anymore, like holding a cup of hot coffee or hot chocolate" (HPTh) Pressed button when "feels so cold you don't want it on your skin anymore, like holding ice or a popsicle" (CPTh)		4. Participants asked to keep eyes closed 5. Parent/guardian present during testing
6	Cummins et al. (2021)	1. Community cntrl (*n* = 14, *M* = 16.4 (0.7), 12f, 2m) 2. No SH in last year) Residential (*n* = 17, *M* = 16.5 (1.0), 1f, 16m) 3. SH 1-4 Episodes (in last year) Residential (*n* = 12, *M* = 16.2 (1.4), 4f, 8m) 4. SH 5+ Episodes (in last year) Residential (*n* = 21, *M* = 16.3 (1), 17f, 4m) Range: 13-17 yrs	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Contact Thermode ($3.2cm^2$)	Whilst seated thermode applied to volar forearm (in some cases dorsal forearm or upper arm if scarification) Baseline 32°C Max: 50°C ROI: 1°C/s Baseline return rate: 8s 3 measures Interstimulus interval: 6s	Whilst seated thermode applied to volar forearm (in some cases dorsal forearm or upper arm if scarification) Baseline: 32°C Min: 0°C ROD: 1°C/s Baseline return rate: 8s 3 measures Interstimulus interval: 6s	Pressed stop button at first pain sensation	Arithmetic mean of 3 measures	1. Testing site avoided naive skin or scarification 2. Monetary reimbursement (withheld until consent to avoid inducement)
7	Duerden et al. (2015)	1. Autistic (*n* = 20, *M* = 14.6 (1.9), 5f, 15m) 2. TD (*n* = 55, *M* = 15.7 (1.1), 28f, 27m)	HPTh CPTh	WDTh CDTh	Contact Thermode ($9cm^2$)	Whilst participant sat comfortable in a chair, one female investigator applied to thermode dominant volar forearm 10cm proximal to the skin crease at the wrist Baseline: 32°C Max: 50°C ROI: 1°C/s Return to baseline: 10°C/s 3 measures	Whilst participant sat comfortable in a chair, one female investigator applied to thermode dominant volar forearm 10cm proximal to the skin crease at the wrist Baseline: 32°C Min: 0°C ROD: 1°C/s Return to baseline:	Pressed button with contralateral index finger when felt heat pain (HPTh) Pressed button with contralateral index finger when felt	Mean of 3 measures	1. Time for participants to adapt to room temperature and be familiarised with equipment and one of the test temperatures stimuli 2.Trials conducted without parental presence 3. Participants provided with standardised instructions 4. Screen positioned so participant was blind to values

						Interstimulus interval: 20s	10°C/s 3 measures Interstimulus interval: 20s	cold pain (CPTh)		5. Participants asked to keep eyes open throughout 6. Participant able to remove thermode at any time 7. Limited to 3 measures to minimise adaptation or sensitisation
8	Engebretsen et al. (2018)	1. Cntrl (*n* = 997, 481f, 516m) Most adolescents 15–17 yrs	HPTh PPTh CPTol	HPTol PPTol	Contact Thermode (30x30mm)	Thermode applied to ulnar side of forearm Baseline: 32°C Max: 50°C ROC: 1°C/s Return to baseline: 8 C/s 3 measures	*See cold pressor task table	Pressed button when sensation changed from warm to pain	Mean of last 2 measures	NR
9	Goffaux et al. (2008)	1. Born full term (38 weeks gestational age) (*n* = 13, *M* = 9.3 (1.3), 6f, 7m), Range: 7–11 yrs 2. Low-pain born preterm (32 weeks gestational age) (*n* = 6, *M* = 9.2 (1.5), 3f, 3m), Range: 8-11 yrs 3. High-pain born preterm (32 weeks gestational age) (*n* = 7, *M* = 9.4 (1.1), 4f, 3m), Range: 8-11 yrs	HPTh	TPS HPSuprathreshold (including CPM)	Contact Thermode (3x3 cm)	Thermode applied to left calf and left forearm Baseline: 32°C Max: - ROI:1°C/s	Not applicable	When pain was reported (unspecified how)	Baseline recording	NR
10	Gulewitsch et al. (2019)	1. Children with FAP (*n* = 20, *M* = 11.91 (1.96), 12f, 8m) 2. HC (*n* = 22, *M* = 11.91 (1.77), 13f, 9m) Range: 8.5–16 yrs	HPTh	None	Contact Thermode (30x30mm)	**Contact thermode applied to non-dominant forearm at: Baseline: 32°C Max: 52.5°C ROI: 1°C/s 5 measures Interstimulus interval: 8s **Premeasurement and postmeasurement trial to a cyberball paradigm, same methodology in each	Not applicable	Pressed button when heat became unpleasant, and pain experienced	Unclear individual threshold, but presented as a group mean	1. Informed not to bear up against the pain 2. Informed thermode could not produce temperature that would burn skin 3. Two cinema coupons as compensation
11	Hainsworth et al. (2020)	1. Chronic pain (*n* = 12, *M* = 15.3 (1.6), 7f, 5m) Range: 13.0–17.5 yrs 2. CPO (*n* = 19, *M* = 16.0 (1.2), 10f, 9m) Range: 13.8–17.8 yrs 3. O (*n* = 14, *M* = 15.1 (1.4), 10f,	HPTh MPTh	HPS MPSen	Contact Thermode (16x16mm)	Thermode applied to thenar eminence of non-dominant hand and then lateral dorsum of the foot Baseline: 32°C Max temp: 50°C ROI: 1°C/s 5 measures Interstimulus interval: ~12s	Not applicable	Pressed button when first felt pain	Mean of 5 measures	1. Primary author (extensive psychophysics training in psychophysics) conducted study in paediatric hospital 2. Testing in quiet room, acclimatising to temperature (i.e. taking off footwear on foot site) 10–15 minutes prior to testing. 3. Tested alone 4. Test site and computer screen

		4m) Range: 13.1–17.3 yrs								blinded from participant 5. No cues to suggest start of stimulus
12	Jacob et al. (2015)	1. Children with SCD (*n* = 48, *M* = 13.7 (2.0), 22f, 26m) Range: 10-17 yrs	CPTh HPTh	MDTh MPS WDTh CDTh	Contact Thermode (9cm^2)	Thermode applied to left or right thenar eminence of the hand at random. Baseline: 32°C Max: 52.5°C ROI: 1.5°C/s Return to baseline: 10°C/s 3 measures	Thermode applied to left or right thenar eminence of the hand at random. Baseline: 32°C Min: 0°C ROD: 1.5°C/s Return to baseline: 10°C/s 3 measures	Pressed button when felt heat pain (HPTh) Pressed button when felt cold pain (CPTh)	Mean of 3 measures	1. Monetary reimbursement
13	Leone et al. (2021)	1. Patient diagnosed with NSSI (*n* = 30, *M* = 15.7 (11-8), 27f, 3m) 2. HC (*n* = 20, *M* = 16.8 (11-18), 18f, 2m)	CPTh HPTh	CDTh WDTh Laser CPM	Contact Thermode (30x30mm)	Thermode applied to right-hand dorsum Baseline: 32°C Max: 50°C ROI: 1°C/s Return to baseline: ~5°C/s 3 measures	Thermode applied to right-hand dorsum Baseline: 32°C Min: 0°C ROD: 1°C/s Return to baseline: ~5°C/s 3 measures	Pressed button when felt pain	Mean of 3 measures	1. Participants mentally count laser stimulations perceived for vigilance and attention
14	Li et al. (2023)	1. Post MSK surgery (*n* = 100, *M* = 14.5 (2.0), 80f, 20m) Range: 10-18 yrs 2. Post MSK injury (*n* = 177, *M* = 14.4 (1.9), 84f, 93m) Range: 11-17 yrs	PPTh HPTh CPTol	TSP CPM	Contact Thermode (30x30mm)	Thermode applied to dominant inner forearm. Baseline: 32°C Max: 50°C ROI: 1.5°C/s Return to baseline: 1°C/s. 4 measures Interstimulus interval: 10s	*See cold pressor task table	Pressed trigger when heat stimulation became painful.	Mean of last 3 measures	1. Each of the 15 researchers completed standardised training of 8-hour workshop on QST, 20-hours of training and independent practice over 5 weeks, an initial proficiency assessment and a final mock QST testing demonstration to confirm that the procedures were mastered. 2. Each researcher needed to either conduct 2 QST testing per month or one testing and observe an additional testing per month. 3. Each researcher QST proficiency were evaluated every 6 months with a training record 4. Children provided scripted instructions, expectations and taught how to use the pain-rating scale 5. Breaks provided where children waited in quiet private area without electronic or family access 6. HPTh practice trial 7. Participant not aware of CPTol ceiling time
15	Lieber et al. (2018)	1. Survivors of paediatric ALL (*n* = 46, *M* = 9.8 (3.1), 18f, 28m), Range: 12.5 yrs 2. HC (*n* = 46, age-gender matched)	CPTh HPTh MPTh PPTh	MDTh VDTh CDTh WDTh PHS TSL MPS DMA WUR	Contact Thermode (9cm^2)	Thermode applied bilaterally using dorsum of both feet, or if clinical symptoms an affected body site in randomised order Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures	Thermode applied bilaterally using dorsum of both feet, or if clinical symptoms an affected body site in randomised order Baseline: 32°C Min: 0°C	Pressed button when pain experienced	Mean of 3 measures	1. Test site dependent on symptoms 2. QST was done in a quiet, darkened room to receive full attention 3. Short breaks if necessary

		*Obtained from Blankenburg et al. (2010) and Blankenburg et al. (2011)					ROD: 1°C/s 3 measures			
16	Ludäscher et al. (2015)	1. BPD (*n* = 20, *M* = 16.4 (1.7), 20f) 2. HC (*n* = 20, *M* = 15.1 (1.4), 20f)	CPTh HPTh	Vibration Sensitivity CDTh WDTh	Contact Thermode (9cm^3)	Thermode applied to both hands, always starting with right Baseline: 32°C Max: 50°C ROI: 1.5°C/s 3 measures	Thermode applied to both hands, always starting with right Baseline: 32°C Min: 0°C ROD: 1.5°C/s 3 measures	Pressed button when perceived thermal sensation as painful	Mean of 3 measures	1. Assessments performed in quiet and comfortably warm room
17	Meier et al. (2001)	1. Ctrl (*N* = 101, *M* = 11.5 (3), 53f, 48m) Range: 6 – 17yrs	CPTh HPTh	CDTh WDTh VDTh	Contact Thermode (9cm^2)	Investigator applied thermode to left or right thenar eminence and lateral dorsum of the foot in randomised order. Baseline: 32°C Max: 50°C ROI: 1.5°C/s Return to baseline: 10°C/s 3 measures	Investigator applied thermode to left or right thenar eminence and lateral dorsum of the foot in randomised order. Baseline: 32°C Min: 0°C ROD: 1.5°C/s Return to Baseline: 10°C/s 3 measures	Pressed button when perceived thermal sensation as painful	Mean of 3 measures	1. Tested in quiet room with testing sites exposed to ambient temperature 10-15 min 2. Provided standard instructions and a familiarisation of sensory tests 3. Participants informed would experience brief pain but could terminate whenever 4. Participant could not see computer for values 5. Participant not given cues for start of stimulus
18	Mensink et al. (2022)	1. Patients with JIA (*n* = 16, *M* = 14.8 (2.1), 12 f, 4 m) Range: 9-18 yrs 2. HC (*n* = 16, matched by age (within 1 year) and age)	CPTh HPTh MPTh PPTh	CDTh WDTh TSL MDTh WUR VDTh MP/DM Allodynia	Contact Thermode (3x3cm^2)	Thermode applied to affected knee and the unaffected control knee in JIA, and right knee in healthy control. Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures	Thermode applied to affected knee and the unaffected control knee in JIA, and right knee in healthy control. Baseline: 32°C Min: 0°C ROD: 1°C/s 3 measures	Pressed button when change from neutral to painful sensation	Mean of 3 measures	1. QST performed by trained personnel 2. Practice trial performed before each test to ensure understanding
19	Morris et al. (2022)	1. Youth with FAP (*N* = 183, *M* = 14.6 (1.9), 119 f, 64 m)	HPTh	HPTol CPM	Contact Thermode (30x30mm)	Thermode applied to non-dominant ventral forearm. Baseline: 32°C Max: 51°C ROI: 0.5°C/s 4 measures Interstimulus interval: 25s	Not applicable	Pressed button when stimulus first perceived as painful	Mean of last 3 measures	1. Double blind study
20	Ocay et al. (2022)	1. Chronic MSK Pain patients (*n* = 302, *M* = 14.93 (1.95), 247f, 55m) Range: 10-18 yrs 2. Age-matched Cntrl (*n* = 80, *M* = 14.99 (1.96), 32f, 48m) Range: 10-18 yrs	PPTh HPTh	MDTh WUR DMA VDTh MPSumm WDTh CPM	Contact Thermode (9cm^2)	Thermode applied to left volar forearm in control and left volar forearm as control area followed by most painful anatomical region (affected area) in MSK Baseline: 32°C Max: 50°C ROI: 0.3°C/s 3 measures	Not applicable	Verbalisation of “stop” when pain experienced	Mean of 3 measures	1. Adapted protocol to reduce complexity, time and fit with clinical constraints

21	O'Leary et al. (2014)	1. SCD (*n* = 27, *M* = 14.8 (2.37), 15f, 12m) 2. Cntrl (*n* = 28, *M* = 14.4 (1.96), 10f, 18m)	HPTh CPTh	HDTh CDTh TPS	Contact Thermode at Volar Forearm (9.0 cm^2) Contact Thermode at Thenar Eminence (2.56 cm^2)	Whilst participant seated, same investigator held thermode to volar surface of dominant forearm (two thirds of the distance between the medial epicondyle and the ulnar process) and thenar eminence of non-dominant hand: Baseline: 32°C Max: 50°C ROI: 1°C/s Returned to baseline: 10°C/s 3 measures Interstimulus interval: 20s	Whilst participant seated, same investigator held thermode to volar surface of dominant forearm (two thirds of the distance between the medial epicondyle and the ulnar process) and thenar eminence of non-dominant hand: Baseline: 32°C Min: 0°C ROD: 1°C/s Returned to baseline: 10°C/s 3 measures Interstimulus interval: 20s	Pressed button when felt heat pain (HPTh) Pressed button when felt cold pain (CPTh)	Mean of 3 measures	1. Standardised verbal instruction in quiet, temperature-controlled room 2. Time for familiarisation with equipment and test trial 3. Testing without parental presence 4. Thermode not fixed to allow participant to withdraw 5. Participant could not see monitor 6. Participant asked to close eyes
22	Rheel et al. (2021)	1. Children experimental group (*n* = 44, *M* = 12.02 (1.87), 20f, 24m)* 2. Children control group (*n* = 45, *M* = 11.71 (1.71), 19f, 26m)* *Two groups of parents not involved in threshold	HPTh	None	Contact Thermode (572.5mm^2 area)	**Contact thermode applied to inside of participants non-dominant forearm, with heat applied in a staircase method completed twice. 1st baseline: 32°C 1st increased to target: 42°C for 300ms If 1st target temp perceived mild, continued until moderate pain. Starting from baseline: 32°C Increased to target: 1°C higher than subsequent target for 300ms Max: 54°C Accelerated velocity: 70°C/s **Served purpose as both outcome measure, and experimental heat pain task	Not applicable	Verbalisation when pain perceived whether it was mild, moderate, or severe	Highest temperature indicated by participant to be at least moderate pain	1. Heat pain procedure described, and equipment shown 2. Monetary reimbursement
23	Riquelme et al. (2023)	1. Autistic (*n* = 38, *M* = 10.94 (4.15), 14f, 24m) 2. TD (*n* = 34, *M* = 9.68 (2.75), 20f, 14m)	CPTh (°C and s at 0°C) PPTh	TDTh WDTh CDTh	Thermal Plate	Not applicable	Participant made skin contact between thermal plate and thenar eminence of hand palms bilaterally 1) CPTh as a °C Baseline: Non-painful baseline (TDT) Min: 0°C Mean ROD: 1°C/s 3 measures	Retracting hand when cold sensation became painful	Mean of 3 measures of bilateral sites (in °C and in s from 0°C)	1. Familiarised with procedure using different stimuli in the body locations other than hand palm (i.e. hand dorsum, arm) 2. Examiner experienced in assessing thresholds 3. Ensure children correctly understood and dealt with any distress.

							2) CPTh as a time in s Constant rate: 0°C Ceiling time: 180s 3 measures			4. In case non-communication of pain in Autistic children, parent observed procedure to report signs of distress that would stop procedure.
24	Sethna et al. (2007)	1. CRPS (*n* = 42, *M* = 13.2 (2.6), 40f, 2m) 2. HC (*n* = 101, Range: 7-17)* *Obtained from Meier et al. (2001)	CPTh HPTh	CDTh WDTh VDTh Mechanical Dynamic Allodynia Static Dynamic Allodynia Allodynia to Punctate Temporal Summation	Contact Thermode ($9cm^2$)	Whilst participant was seated investigator applied thermode to affected and unaffected limb (foot) Baseline: 32°C Max: 50°C ROI: 1.5°C/s Return to baseline: 10°C/s 3 measures	Whilst participant was seated investigator applied thermode to affected and unaffected limb (foot) Baseline: 32°C Min: 0°C ROD: 1.5°C/s Return to baseline: 10°C/s 3 measures	Pressed button when perceived thermal sensation as painful	Mean of 3 measures	1. Single researcher conducted sensory testing in quiet room whilst participant seated or reclined 2. Skin sites exposed to ambient temperature for 10-15 min before testing. 3. Participant could not see computer for values 4. Participant not given cues for start of stimulus 5. Given a dry run before testing
25	Sieberg, Lunde, Shafrir, et al. (2023)	1. Female pain free (*N* = 118, *M* = 23 (7), 12-19 yrs *n* = 58) 2. Male pain free (*N* = 63, *M* = 30 (9), 12-19 yrs *n* = 10) *Thresholds reported by decade age 12-19 yr	MPTh PPTh HPTh CPTh	DMA MDTh TSP WDTh CDTh	Contact Thermode	Thermode applied to 4 sites: centre of the upper left and right quadrants and lower left and right quadrants of the abdomen. Baseline: 32°C Max: 50°C ROI: 1°C 3 measures	Thermode applied to 4 sites: centre of the upper left and right quadrants and lower left and right quadrants of the abdomen. Baseline: 32°C Min: 0°C ROD: 1°C 3 measures	Pressed button first sensation of pain	Mean of 3 measures	1. Researchers could not test until proficiency demonstrated 2. Continuous protocol with natural breaks 3. Detail script to read verbatim for participants every study visit. 4. Monetary reimbursement
26	Stabell et al. (2014)	Students (*n* = 961, *M* = 16.1 (0.4), 469f, 492m)* *Includes 861 cntrl and 77 IBS participants Range: 15–17 yrs	HPTh PPTh CPTol	PPTol HPTol	Contact Thermode (30x30mm)	Thermode applied to volar surface of right forearm. Baseline: 32°C Max: 50°C ROI: 1°C/s Return to baseline: 8°C/s 3 measures	*See cold pressor task table	Pressed button when sensation changed from warmth to pain	Mean of last 2 measures	NR
27	Truffyn et al. (2021)	1. Lower limb CRPS-1 (*n* = 34, *M* = 12.03 (2.4), 28f, 6m) 2. HC (*n* = 56, *M* = 15.7 (1.1), 28f, 28m)	HPTh CPTh	WDTh CDTh	Contact Thermode (3x3cm)	Whilst resting arm on padded surface, thermode applied using support stand at constant pressure to 10cm above wrist on dominant volar forearm. CRPS-1 group additionally tested pain site and contralateral pain site. Baseline: 32°C Max: 50°C ROI: 1°C/s 3 measures Interstimulus interval: 20s	Whilst resting arm on padded surface, thermode applied using support stand at constant pressure to 10cm above wrist on dominant volar forearm. CRPS-1 group additionally tested pain site and contralateral pain site. Baseline: 32°C Min: 0°C ROD: 1°C/s 3 measures Interstimulus interval: 20s	Pressed button when sensation changed to a 'hurt feeling'	Mean of 3 measures	1. Participants told could withdraw arm any time 2. Participants trained to press button

28	Valkenburg et al. (2015)	1. Children with DS (*n* = 42, $M_{yr:mth}$ = 12:10 (3:0), 21f, 21m) 2. Siblings (*n* = 24, $M_{yr:mth}$ = 15:0 (4:10), 8f, 16m)	CPTh HPTh	MDTh CDTh WDTh	Contact Thermode (30x30mm)	Thermode applied to thenar eminence of the non-dominant hand. Baseline: 32°C Max: 50°C ROI: 1.5°C/s 5 measures (1st acted as rehearsal)	Thermode applied to thenar eminence of the non-dominant hand. Baseline: 32°C Min: 0°C ROD: 1.5°C/s 5 measures (1st acted as rehearsal)	Release button when stimulus became so hot was painful (HPTh) Release button when stimulus became so cold was painful (CPTh)	Mean of last 4 measures	1. Participant released button instead of pressed button when pain perceive as considered easier to understand. 2. Designed comic book to prepare participant for testing; available online so parent and child could read at home. 3. Comic book main character was plush animal, designed with thermode of TSA as tail. This was brought to participant. 4. Before testing began comic was reread and plush animal introduced. 5. Parents were present during study but asked to minimise interference.
29	van den Bosch et al. (2017)	1. 8-9 yrs (*n* = 14, *Mdn* = 9.0 (8.7 – 9.4), 8f, 6m) 2. 10-11 yrs (*n* = 31, *Mdn* = 11.1 (10.6 – 11.3), 18f, 13m) 3. 12-13 yrs (*n* = 12, *Mdn* = 12.5 (12.5 – 13.0), 8f, 4m) 4. 14-17 yrs (*n* = 12, *Mdn* = 16.5 (14.7 – 17.6), 7f, 5m)	CPTh HPTh	CDTh WDTh	Contact Thermode (30x30mm)	Researcher applied thermode to thenar eminence of non-dominant hand . Baseline: 32°C Max: 50°C ROI: 1.5°C/s Returned baseline: 10°C/s 5 measures (1st acted as rehearsal) Interstimulus interval: 10s	Researcher applied thermode to thenar eminence of non-dominant hand . Baseline: 32°C Min: 0°C ROD: 1.5°C/s Returned baseline: 10°C/s 5 measures (1st acted as rehearsal) Interstimulus interval: 10s	Pressed button when thermode felt painful	Mean of last 4 measures	1. Same researcher conducted all measures 2. Emphasised to participant and parent testing would not harm hand 3. Parents asked to not interact with participant during testing
30	van der Venne et al. (2021)	1. HC (*n* = 35, *M* = 14.9 (1.29), 35f) 2. NSSI (*n* = 94, *M* = 14.9 (1.44), 94f)	HPTh	HPTol	Versatile cold/hot plate	Non-dominant hand placed flat on plate once baseline temperature reached, 3-min adaptation phase before temperature raised Baseline: 32°C. Max: 50°C ROI: 1°C/13.3 s	Not applicable	NR; °C taken at first pain sensation for threshold. Then rated on a 0-100 NRS once pain threshold was met	Temperature at the first pain sensation (°C)	1. Monetary reimbursement
31	Wollgarten-Hadamek et al. (2009)	1. Moderate burn injuries (*n* = 24, *M* = 11.9 (2.0), 11f, 13m) Range: 9–16 yrs 2. Severe burn injuries (*n* = 24, *M* = 12.0 (2.0), 12f, 12m) Range: 9–15 yrs 3. Control (*n* = 24, *M* = 11.2 (1.9), 12f, 12m) Range: 9–15 yrs	HPTh MPTh	WDTh TPS MDTh MPSen	Contact Thermode (16x16mm)	Whilst participant was seated, thermode applied to non-dominant thenar and trigeminal nerve Baseline: 32°C Max: 50°C ROI: 1°C/s 5 measures Interstimulus interval: 12s	Not applicable	Pressed button when felt heat pain	Mean of 5 measures	1. Sites chosen as those not affected by burn injury 2. All equipment shown to reduce fear 3. Both participant and parent could withdraw at any point 4. Experimental trials had an initial familiarisation trial not included in analysis 5. Participant tested alone with parent outside

										6. Computer with thermode values out of site
32	Wollgarten-Hadamekl et al. (2011)	1. Moderate burn injuries (*n* = 12, *M* = 12.3 (1.9), 8f, 4m) Range: 10–16 yrs 2. Severe burn injuries (*n* = 10, *M* = 12.7 (1.8), 6f, 4m) Range: 10–16 yrs 3. Cntrl (*n* = 20, *M* = 13.3 (1.9), 10f, 10m) Range: 10–16 yrs	HPTh PPTh	HPTol PPTol Ischemic PTh Ischemic Ptol	Contact Thermode (30x30mm)	Whilst seated, thermode applied to participant's non-dominant volar forearm. Baseline: 32°C Max: 50°C ROI: 1°C/s 3 probe trails 5 measures Interstimulus interval: 10s	Not applicable	Pressed button when felt heat pain	Mean of 5 measures	1. Participant familiarised with lab and equipment 2. Participant could not see value on computer 3. Monetary compensation
33	Zohsel et al. (2006)	1. With migraine (*n* = 25, *M* = 11.0 (1.8), 11f, 14m) 2. Cntrl (*n* = 28, *M* = 11.0 (1.8), 16f, 12m) Range: 9–15yrs	HPTh MPTh	TPS MPSen	Contact Thermode (16x16mm)	Thermode applied to skin at: 1) Trigeminal nerve was chosen (area of the upper cheek) (Pain-relevant site; either predominantly affected side in migraineurs group, or random in control/bilateral migraineur pain) 2) Thenar eminence of the non-dominant hand (Distal site) Baseline: 32°C Max: 50°C ROI: 1°C/s 5 measures Interstimulus interval: 12s	Not applicable	Pressed button when felt pain	Mean of 5 measures	1. Participant and parent could withdraw any time 2. All equipment shown and explained to participant and parent to reduce excitement and fear 3. Monetary reimbursement
34	Zohsel et al. (2008b)	1. RAP (*n* = 20, *M* = 10.7 (1.7), 11f, 9m) Range: 8–14 yrs 2. Cntrl (*n* = 23, *M* = 11.0 (1.5), 10f, 13m) Range: 9–14 yrs	HPTh MPTh	TPS MPSen	Contact Thermode at thenar (16x16mm) Contact Thermode at abdomen (30x30mm)	Thermode applied to skin at: 1) M. abdominus near the umbilicus (Pain-relevant site; either predominantly affected side in RAP group, or random in control) 2) Thenar eminence of the non-dominant hand (Distal site) Baseline: 32°C Max: 50°C ROI: 1°C/s. 5 measures Interstimulus: 12s	Not applicable	Pressed button when felt pain	Mean of 5 measures	1. Participant and parent could withdraw any time 2. All equipment shown and explained to participant and parent to reduce excitement and fear 3. Parent waited in adjacent room during testing 4. Monetary reimbursement

Note. This table provides the data extracted from each study measuring heat pain threshold or cold pain thresholds with a contact thermode. Abbreviations ordered alphabetically ALL = Acute Lymphoblastic Leukaemia; BPD = Borderline Personality Disorder; CDTh = Cold Detection Threshold; Cntrl = Control; CPM = Conditioned Pain Modulation; CPO = Chronic Pain and Co-Occurring Overweight/Obesity; CPTh = Cold Pain Threshold; CPTol = Cold Pain Tolerance; CRPS = Complex Regional Pain Syndrome; DFNS = Quantitative Sensory Testing in the German Research Network on Neuropathic Pain; DM = Diabetes Mellitus; DMA = Dynamic Mechanical Allodynia; DS = Down Syndrome; EU = European; f = Female; FAP = Functional Abdominal Pain; HC = Healthy Control; HPTh = Heat Pain Threshold; HPTol = Heat Pain Tolerance; IBS = Irritable Bowel Syndrome; JIA = Juvenile Idiopathic Arthritis; m = Male; M = Mean; Mdn = Median; MDTh = Mechanical

Detection Threshold; MPS = Mechanical Pain Sensitivity; MPSen = Mechanical Perceptual Sensitisation; MPSumm = Mechanical Pain Summation; MPTh = Mechanical Pain Threshold; mths = Months; MSK = Musculoskeletal Pain; n = Sub-Group Sample Size; N = Total Sample Size; NR = Not Reported; NRS = Numerical Rating Scale; NSSI = Non-Suicidal Self-Injury; O = Obesity; PHS = Paradoxical Heat Sensation; PTh = Pain Threshold; PPTh = Pressure Pain Threshold; PTol = Pain Tolerance; PPTol = Pressure Pain Tolerance; QST = Quantitative Sensory Testing; RAP = Recurrent Abdominal Pain; ROD = Rate of Decrease; ROI = Rate of Increase; s = Seconds; SCD = Sickle Cell Disease; SH = Self-Harm; TD = Typically Developing; TDTh = Tactile Detection Threshold; TPS = Thermal Perceptual Sensitisation; TSL = Thermal Sensory Linen; TSP = Temporal Summation of Pain; US = United States; VDTh = Vibration Detection Threshold; WDTh = Warm Detection Threshold; WUR = Wind-Up Ratio; and Yrs = Years

2.3.4.1.2.2 Cold Pressor Task for Cold Pain Threshold.

Of the 22 studies assessing CPTh since the year 2012, three used a CPT (66.67% with continuous water circulation) (see Table 7). None of these studies (0%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter). A total of ~831 CYP aged between 7.30 and 17 years participated (~402 females, ~409 males) and were not defined by diagnosis, although Saxena et al. (2015) only assessed males and included an adult population.

Prior to testing, two studies instructed participants to immerse their non-dominant arm into warm water for 2 minutes for similar group skin temperature. Marche et al. (2015) immersed arm into 34.8°C at elbow level, whilst Schmitz et al. (2013) immersed arm into 36°C (±1°C) 5cm above the wrist with upwards palm. When testing CPTh, all participants immersed a hand into cold water with Marche et al. (2015) and Schmitz et al. (2013) using instructions above, and Saxena et al. (2015) immersing non-dominant hand 2cm above the wrist with fingers spread and palm down. Each study used different cold-water temperatures and ceiling times including 10.8°C at informed 4-minute ceiling (Marche et al., 2015), 6°C (±1°C) at uninformed 3-minute ceiling (Schmitz et al., 2013), and 0-1°C at not reported ceiling (Saxena et al., 2015). Saxena et al. (2015) may have used colder water temperature as more participants were aged over 18 (*n* = 63), than under 18 (*n* = 17). Participants self-reported CPTh but did not remove their hand to allow continued CPTol measurement, however only Marche et al. (2015) specified verbal report. Time elapsed between immersion and self-report of pain was defined as the CPTh in a single measure (*n* = 3).

Two studies reported methodological considerations, Marche et al. (2015) allowed participants to rest their arm on an insulated cover to minimise temperature fluctuations, and Schmitz et al. (2013) tested participants alone with researcher stood behind the participant to avoid eye contact. Whether this improved result validity cannot be determined.

Based on the synthesised evidence, evidence suggests that when using a cold pressor task to measure CPTh, researchers should ask participants to immerse their hand into cold water and self-report when a sensation of pain is felt. However, as testing

parameters are unclear, researchers should defer to similar protocols or reviews for guidance on temperature, and ceiling time.

2.3.4.1.2.3 Cold Pressor Task for Cold Pain Tolerance.

A total of six studies since 2012 considered CPTol using a CPT (see Table 7), including 3 CPTh studies outlined in Chapter 2.4.1.2.2, with ~3066 participants aged between 7.30 and 17 years (~1516 females, ~1530 males). The additional three studies omitted CPTh in older participants (between 11 and 17 years) to instead measure HPTh and PPTh (Engebretsen et al., 2018; Li et al., 2023; Stabell et al., 2014). Only two studies compared within a pain-diagnostic group (musculoskeletal and IBS), remaining four included control populations. None of these studies (0%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

Methodological description of CPTol for Marche et al. (2015), Saxena et al. (2015), and Schmitz et al. (2013) are consistent with Chapter 2.4.1.2.2 (except for reporting CPTol). Warm water bath was not utilised in additional three studies. Instead, participants immersed hand up to wrist level (non-dominant hand (*n* = 1), open dominant hand (*n* = 1), and left hand (*n* = 1)) into 3°C water (*n* = 2), or 8°C (*n* = 1), colder water in older control populations, and warmer water in diagnostic populations.

Participants only removed their hand when they could no longer maintain the water temperature (*n* = 6), with five studies including ceiling time of 4 minutes (40%), 3 minutes (20%), and 1 minute 45 seconds (40%). Shorter ceiling time was reported for Stabell et al. (2014) and Engebretsen et al. (2018) where an older control population were utilised (range 15-17 years), however studies included three tolerance measures so shortened ceiling may be for safety. CPTol was measured once (*n* = 6) and recorded as time hand was removed (83.33%), or difference between CPTh and total immersion time (16.67%).

Based on the synthesised evidence, evidence suggests that when using a cold pressor task to measure CPTol, researchers should ask participants to immerse their hand into cold water for a singular trial and remove their hand when they can no longer maintain

the water temperature. However, as testing parameters are unclear, researchers should defer to similar protocols or reviews for guidance on temperature, and ceiling time.

Table 7

Included Studies in Systematic Review Measuring Cold Pain Threshold or Cold Pain Tolerance with a Cold Pressor Task

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	CPTh Methodology	CPTol Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Engebretsen et al. (2018)	1. Cntrl (*N* = 997, 481f, 516m) Most adolescents 15–17 yrs	HPTh PPTh CPTol	HPTol PPTol	13-L plexi-glass container and circulating water bath	Not applicable	Non-dominant hand and wrist placed in in 3°C water and maintained for as long as participants could. Max: 105s	Removal of hand from water when could no longer maintain	Time hand kept in water	NR
2	Li et al. (2023)	1. Post MSK surgery (*n* = 100, *M* = 14.5 (2.0), 80f, 20m) Range: 10-18 yrs 2. Post MSK injury (*n* = 177, M = 14.4 (1.9), 84f, 93m) Range: 11-17 yrs	PPTh HPTh CPTol	TSP CPM	Cooling unit with a finger guard and an immersion circulator (Cole Parmer Ltd., Vernon Hills, IL)	Not applicable	Dominant hand whilst open and relax immersed up to the wrist in 8˚C cold water bath Max: 4 min	Hand removed when could no longer tolerate cold	Hand immersion time	1. Each of the 15 researchers completed standardised training of 8-hour workshop on QST, 20-hours of training and independent practice over 5 weeks, an initial proficiency assessment and a final mock QST testing demonstration to confirm that the procedures were mastered. 2. Each researcher needed to either conduct 2 QST testing per month or one testing and observe an additional testing per month. 3. Each researcher QST proficiency were evaluated every 6 months with a training record 4. Children provided scripted instructions, expectations and taught how to use the pain-rating scale 5. Breaks provided where children waited in quiet private area without electronic or family access 6. HPTh practice trial 7. Participant not aware of CPTol ceiling time
3	Marche et al. (2015)	1. HC (*N* = 86; demographics only available for 66 children with 56% male, *M* = 9.85 (2.02), 29f, 37m) Range: 7.30-15.42 yrs	CPTh CPTol	None	Modified JetSpray Machine (inc. built-in thermoregulator, thermostat with external temperature control, cooler, and water pump to cool and circulate over 18L of water)	First participant submerged non-dominant arm up to elbow, in warm water (M_{temp} = 34.8°C) for 2min for group skin temp prior to CPT immersions. Next, participant submerged non-dominant arm up to elbow in cold circulating water	First participant submerged non-dominant arm up to elbow, in warm water (M_{temp} = 34.8°C) for 2min for group skin temp prior to CPT immersions. Next, participant submerged non-dominant arm up to elbow in cold circulating water	Verbalisation when arm first started to hurt (not to remove hand) (CPTh) Removed arm when hurt too much to keep in tank (CPTol)	Time pain reported (CPTh) Time arm removed from water (CPTol)	1. Armrest an insulation covers to minimise temperature fluctuations. 2. Participant given reward

						(M_{temp} = 10.8°C). Immersed arm for as long as could or until 4min ceiling. Time measured on stopwatch	(M_{temp} = 10.8°C). Immersed arm for as long as could or until 4min ceiling. Time measured on stopwatch			
4	Saxena et al. (2015)	*N* = 79 1. Children Range: 8-13 yrs (*n* = 17, 17m) 2. Young Adults Range: 18-25 yrs (*n* = 21, 21m) 3. Middle Aged Adults Range: 35-45 yrs (*n* = 22, 22m) 4. Old Adults Range: 55-70 yrs (*n* = 19, 19m)	CPTh CPTol	None	Cold water bath	Non-dominant hand immersed into cold water bath (0-1°C). Water needed to cover up to 2cm above wrist whilst palm down and fingers spread out. CPTh and total time of immersion recorded (s) on two separate stop watches.	Non-dominant hand immersed into cold water bath (0-1°C). Water needed to cover up to 2cm above wrist whilst palm down and fingers spread out. CPTh and total time of immersion recorded (s) on two separate stop watches.	Self-report when felt pain (not to remove hand) (CPTh) Removal of hand from water when chose to not bear pain anymore (CPTol)	Time in s pain reported (CPTh) Calculation of subtracting CPTh from total time of immersion (CPTol)	NR
5	Schmitz et al. (2013)	*N* = 728, *M* = 12.6 (1.9), 373f, 355m Range: 9-17 yrs Grade 4 (*n* = 110) Grade 5 (*n* = 107) Grade 6 (*n* = 146) Grade 7 (*n* = 112) Grade 8 (*n* = 111) Grade 9 (*n* = 142)	CPTh CPTol	None	Thermostatically controlled refrigeration unit with continuous water circulation	First participant submerged non-dominant forearm with water up to 5cm above wrist, with palm facing upwards in 36 °C (±1 °C) warm water for 2 minutes. Once hand was dry, participants submerged identical forearm in 6 °C (±1 °C) cold water, resting arm on armrest, and to keep there as long as possible (even if uncomfortable) with 3min ceiling* *Additional pain endurance measure calculated	First participant submerged non-dominant forearm with water up to 5cm above wrist, with palm facing upwards in 36°C (±1°C) warm water for 2 minutes. Once hand was dry, participants submerged identical forearm in 6°C (±1°C) cold water, resting arm on armrest, and to keep there as long as possible (even if uncomfortable) with 3min ceiling* *Additional pain endurance measure calculated	Self-report when pain in hand experienced (not to remove hand) (CPTh) Removal of hand when pain became intolerable (CPTol)	Time in s from forearm immersion to pain report (CPTh) Total immersion duration in seconds (CPTol)	1. Participants tested individually (in line with von Baayer et al. (2005) 2. Nine trained, female experimenters (sex constant factor in experiment) 3. Participants not informed of elapsed, or ceiling time 4. Experimenter stood behind participant to avoid eye contact
6	Stabell et al. (2014)	Students (*N* = 961, *M* = 16,1 (0.4), 469f, 492m)* *Includes 861 cntrl and 77 IBS participants Range: 15-17 yrs	HPTh PPTh CPTol	HPTol PPTol	Cold water bath (13L external acrylic glass container with a flow rate of 22L/min	Not applicable	Left hand hand wrist immersed into 3°C cold water as long as able to. Max: 105s Time to withdrawal of the hand was recorded	Removal of hand when no longer able to withhold pain	Time hand removed from water	NR

Note. This table provides the data extracted from each study measuring cold pain threshold or cold pain tolerance with a cold pressor task. Abbreviations ordered alphabetically Cntrl = Control; CPM = Conditioned Pain Modulation; CPTh = Cold Pain Threshold; CPTol = Cold Pain Tolerance; DFNS = Quantitative Sensory Testing in the German Research Network on Neuropathic Pain; f = Female; HC = Healthy Control; HPTh = Heat Pain Threshold; HPTol = Heat Pain Tolerance; IBS = Irritable Bowel Syndrome; m = Male; M = Mean; MSK = Musculoskeletal Pain; M_{Temp} = Mean Temperature; n = Sub-Group Sample Size; N = Total Sample Size; NR = Not Reported; PPTh = Pressure Pain Threshold; PPTol = Pressure Pain Tolerance; QST = Quantitative Sensory Testing; s = Seconds; TSP = Temporal Summation of Pain; and Yrs = Years.

2.3.4.2 Mechanical Threshold

Mechanical thresholds were assessed in 75.86% of studies (*n* = 66). Of these 66 studies, MPS (9.09%), MPTh (19.70%), PPTh (93.94%) and WUR (10.61%) were measured. Below psychophysical methodologies are discussed in more detail, except for WUR and MPS as these measures fail to provide thresholds.

2.3.4.2.1 Mechanical Pain Threshold.

2.3.4.2.1.1 Punctate Probes for Mechanical Pain Threshold.

Of the 13 studies assessing MPTh, 10 utilised a punctate probe (see Table 8). Of these studies, six (60%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

Excluding control values used by (Lieber et al., 2018) as these represented participants from a differing study, ~794 CYP aged between 6 and 18 years participated in a study involving a MPTh assessment with punctate probes (~405 females, ~389 males). Studies predominantly compared MPTh between a diagnostic and control group (*n* = 7), with additional studies comparing within controls (*n* = 2), and within diagnoses (*n* = 1). Diagnoses included were predominantly pain related (*n* = 5), however neuropathy (*n* = 2) and mental health (*n* = 1) were considered. Methodological parameters typically referenced Rolke et al. (2006) (*n* = 6) and Blankenburg et al. (2010) (*n* = 4) as a standardised protocol followed.

The same punctate probes were used across studies; 100% used standard equipment of 7 weighted mechanical pinpricks/punctate filaments with a 0.2mm diameter contact area (forces: 8, 16, 32, 64, 128, 256, and 512mN). Stimuli were applied to 1 (*n* = 5; 80% bilateral), 2 (*n* = 4; 0% bilateral), and 3 (*n* = 1; 100% bilateral). Where one site is reported, this value includes one study involving a pain-related diagnostic group being measured in 1 site bilaterally, and a control group where site was measured unilaterally (Mensink et al., 2022). A full list of testing location can be found in the methodology section of Table 8, however broadly the hand (*n* = 6; 66.67% thenar eminence), and the foot (*n* = 4; 75% dorsal) were used most. For five studies, testing sites were dependent on participants diagnostic experience (three pain-related, one mental health and one neuropathy). As

reported in Chapter 2.3.4.1.1.1, diagnostic experience included locations that were affected by diagnosis such as in pain vs not (*n* = 3) and ethical consideration of preventing harm (*n* = 2).

Studies involving fixed force intensities between 8 and 512mN followed the same protocol, applying 5 series of ascending (prick) and descending (blunt) stimuli as a 2 second contact time to each site (*n* = 10) however only 30% reported blindfolding participants. Participants self-reported in each ascending or descending series the first percept of sharpness and bluntness, respectively (*n* = 8), or verbalised “yes” or “no” for if a sensation was painful (*n* = 2). Each of these studies expressed MPTh as a geometric mean across the 10 sharp and blunt stimuli.

Based on the synthesised evidence, evidence suggests that when using punctate probes to measure MPTh, researchers utilise 7 weighted pinprick mechanical stimulators (8-512mN) and apply a series of 5 ascending (prick) and descending (blunt) stimuli to the relevant testing site. Participants should be asked to self-report at the first percept of sharpness or bluntness.

Table 8

Included Studies in Systematic Review Measuring Mechanical Pain Threshold with a Punctate Probes

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Blankenburg et al. (2010)	1. HC (*N* = 176, 88f, 88m) Range: 6-16.12 yrs Age bands: 6-8 yrs (24f, 24m) 9-12.2 yrs (32f, 32m) 13-16.12 yrs (32f, 32m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Pinprick stimuli applied to both facial cheeks, dorsum of hands and feet in randomised order Contact time: 2s Five series of ascending (prick) and descending (blunt) stimuli.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean across stimuli.	1. 1 of 2 assessors who received 4-week training in DFNS centre for QST research 2. Demonstration of pain induction at practice area above test area 3. Participant decided whether parents stayed in room or outside 4. 4-hr sessions with short breaks if concentration declined 5. Monetary reimbursement
2	Blankenburg et al. (2011)	1. HC (*N* = 173) Age bands: 7 yrs (42f, 44m) 14 yrs (43f, 44m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Pinprick stimuli applied to both dorsum of hands in randomised order Contact time: 2s Five series of ascending (prick) and descending (blunt) stimuli. MLE	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean across stimuli	1. Described to children presence and absence of pain as a likened to difference between sharp and blunt 2. Subjects decided whether their parents stayed in the testing room or outside 3. Monetary reimbursement
3	Blankenburg et al. (2012)	1. Type-1 DM (*n* = 45, *M* = 13.2 (2.8), 23f, 22m) 2. HC (*n* = 45, *M* = 13.2 (2.6), 23f, 22m age-gender matched)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Pinprick stimuli applied to both feet in randomised order* Contact time: 2s Five series of ascending (prick) and descending (blunt) stimuli.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean across stimuli	1. Monetary reimbursement
4	Cummins et al. (2021)	1. Community cntrl (*n* = 14, *M* = 16.4 (0.7), 12f, 2m) 2. No SH (in last year) Residential (*n* = 17, *M* = 16.5 (1.0), 1f, 16m) 3. SH 1-4 Episodes (in last year) Residential (*n* = 12, *M* = 16.2 (1.4), 4f, 8m) 4. SH 5+ Episodes (in last year) Residential (*n* = 21, *M* = 16.3 (1), 17f, 4m) Range: 13-17 yrs	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Whilst participant was seating, pinprick applied to volar forearm (in some cases dorsal forearm or upper arm if scarification) in 'up-down' pattern. Five supra- and five subthreshold responses.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean of values	1. Testing site avoided naive skin or scarification 2. Monetary reimbursement (withheld until consent to avoid inducement)

5	Hainsworth et al. (2020)	1. Chronic pain (*n* = 12, *M* = 15.3 (1.6), 7f, 5m) Range: 13.0-17.5 yrs 2. CPO (*n* = 19, *M* = 16.0 (1.2), 10f, 9m) Range: 13.8-17.8 yrs 3. O (*n* = 14, *M* = 15.1 (1.4), 10f, 4m) Range: 13.1-17.3 yrs	HPTh MPTh	HPS MPSen	7 Standardised Punctate Filaments at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Punctate filament applied thenar eminence of non-dominant hand and then lateral dorsum of the foot. Five series of ascending (prick) and descending (blunt) stimuli.	Prick: Verbalisation of "yes" for pain threshold Blunt: Verbalisation of "no" for non-painful stimulus	Geometric mean of the 10 values above and below threshold	1. Primary author (extensive psychophysics training in psychophysics) conducted study in paediatric hospital 2. Testing in quiet room, acclimatising to temperature (i.e. taking off footwear on foot site) 10–15 minutes prior to testing. 3. Tested alone 4. Test site and computer screen blinded from participant 5. No cues to suggest start of stimulus
6	Lieber et al. (2018)	1. Survivors of paediatric ALL (*n* = 46, *M* = 9.8 (3.1), 18f, 28m), Range: 12.5 yrs 2. HC (*n* = 46, age-gender matched)* *Obtained from Blankenburg et al. (2010) and Blankenburg et al. (2011)	CPTh HPTh MPTh PPTh	MDTh VDTh CDTh WDTh PHS TSL MPS DMA WUR	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Pinprick stimuli applied bilaterally using dorsum of both feet, or if clinical symptoms on an affected body site in randomised order Contact time: 2s Five series of ascending (prick) and descending (blunt) stimuli.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean across stimuli	1. Test site dependent on symptoms 2. QST was done in a quiet, darkened room to receive full attention 3. Short breaks if necessary
7	Mensink et al. (2022)	1. Patients with JIA (*n* = 16, *M* = 14.8 (2.1), 12f, 4m) Range: 9-18 yrs 2. HC (*n* = 16, matched by age (within 1 year) and age)	CPTh HPTh MPTh PPTh	CDTh WDTh TSL MDTh WUR VDTh MP/DM Allodynia	7 Weighted Pinpricks Mechanical Stimulators at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Pinprick applied to affected knee and the unaffected control knee in JIA, and right knee in healthy control. Contact time: 2s Five series of ascending (prick) and descending (blunt) stimuli.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean across stimuli	1. QST performed by trained personnel 2. Practice trial performed before each test to ensure understanding
8	Wollgarten-Hadamek et al. (2009)	1. Moderate burn injuries (*n* = 24, *M* = 11.9 (2.0), 11f, 13m) Range: 9–16 yrs 2. Severe burn injuries (*n* = 24, *M* = 12.0 (2.0), 12f, 12m) Range: 9–15 yrs 3. Cntrl (*n* = 24, *M* = 11.2 (1.9), 12f, 12m) Range: 9–15 yrs	HPTh MPTh	WDTh TPS MDTh MPSen	7 Pinpricks Punctate Probes at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Whilst blindfolded, pinpricks applied to the non-dominant thenar and trigeminal nerve in ascending and descended series starting with lowest intensity. Repeated 5 times per series.	Prick: First reported percept of sharpness in series (ascending) Blunt: First reported percept of blunt touch in series (descending)	Geometric mean of five sub- and five supra-threshold intensities	1. Sites chosen as those not affected by burn injury 2. All equipment shown to reduce fear 3. Both participant and parent could withdraw at any point 4. Experimental trials had an initial familiarisation trial not included in analysis 5. Participant tested alone with parent outside 6. Computer with thermode values out of site
9	Zohsel et al. (2006)	1. With migraine (*n* = 25, *M* = 11.0 (1.8), 11f, 14m) 2. Cntrl (*n*= 28, *M* = 11.0 (1.8), 16f, 12m) Range: 9-15yrs	HPTh MPTh	TPS MPSen	7 Pinpricks Punctate Probes at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Whilst blindfolded, starting with lowest intensity pinpricks applied in ascending and descending series to: 1) Trigeminal nerve was chosen (area of the upper cheek) (Pain-relevant site; either predominantly affected side in migraineurs group, or random in control/bilateral migraineur pain) 2) Thenar eminence of the non-dominant hand (Distal site) Repeated 5 times per series.	Self-report verbalisation of "yes" or "no" if painful sensation	Geometric mean of 10 values above and below threshold	1. Participant and parent could withdraw any time 2. All equipment shown and explained to participant and parent to reduce excitement and fear 3. Monetary reimbursement

10	Zohsel et al. (2008b)	1. RAP (*n* = 20, *M* = 10.7 (1.7), 11f, 9m) Range: 8-14 yrs 2. Cntrl (*n* = 23, *M* = 11.0 (1.5), 10f, 13m) Range: 9-14 yrs	HPTh MPTh	TPS MPSen	7 Pinpricks Punctate Probes at Fixed Intensity Forces 8, 16, 32, 64, 128, 256, and 512 mN 0.2mm diameter contact area	Whilst blindfolded, starting with lowest intensity pinpricks applied in ascending and descending series to: 1) M. abdominus near the umbilicus (Pain-relevant site; either predominantly affected side in RAP group, or random in control) 2) Thenar eminence of the non-dominant hand (Distal site) Repeated 5 times per series.	Self-report verbalisation of "yes" or "no" if painful sensation	Geometric mean of 10 values above and below threshold	1. Participant and parent could withdraw any time 2. All equipment shown and explained to participant and parent to reduce excitement and fear 3. Parent waited in adjacent room during testing 4. Monetary reimbursement

Note. This table provides the data extracted from each study measuring mechanical pain threshold with punctate probes. Abbreviations ordered alphabetically ALL = Acute Lymphoblastic Leukaemia; CDTh = Cold Detection Threshold; Cntrl = Control; CPTh = Cold Pain Threshold; DFNS = Quantitative Sensory Testing in the German Research Network on Neuropathic Pain; DM = Diabetes Mellitus; DMA = Dynamic Mechanical Allodynia; f = Female; HC = Healthy Control; HPTh = Heat Pain Threshold; JIA = Juvenile Idiopathic Arthritis; m = Male; M = Mean; MDTh = Mechanical Detection Threshold; mN = Millinewtons; MPS = Mechanical Pain Sensitivity; MPSen = Mechanical Perceptual Sensitisation; MPTh = Mechanical Pain Threshold; n = Sub-Group Sample Size; N = Total Sample Size; PHS = Paradoxical Heat Sensation; PPTh = Pressure Pain Threshold; QST = Quantitative Sensory Testing; RAP = Recurrent Abdominal Pain; s = Seconds; SH = Self-Harm; TPS = Thermal Perceptual Sensitisation; TSL = Thermal Sensory Linen; VDTh = Vibration Detection Threshold; WDTh = Warm Detection Threshold; WUR = Wind-Up Ratio; and Yrs = Years.

2.3.4.2.1.2 Von-Frey Hairs for Mechanical Pain Threshold.

Of the 13 studies assessing MPTh, three used Von-Frey hairs with an observed pattern of equipment use coinciding with a MDTh measure (see Table 9). Two of these studies (66.66%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

Excluding control values used by Cornelissen et al. (2014) as these represented participants from a differing study, ~160 CYP between the ages of 9.6 and 19 years participated in a study involving a MPTh assessment with Von-Frey Hairs (~127 female, ~33 male). MPTh's were compared within a control group (*n* = 1), between a pain-related diagnostic and control group (*n* = 1), or within a pain-related diagnostic group (*n* = 1). Meier et al. (2001) (*n* = 1) and Rolke et al. (2006) (*n* = 2) were discussed as a standardised protocol. One study utilising Rolke et al. (2006) involved a predominant adult population (Sieberg, Lunde, Shafrir, et al., 2023), but protocol adaptation was not made for CYP (only CYP <19 years are accounted for in the above *n* value).

Studies used Von-Frey Hairs between 0.008 and 300g (*n* = 2), or 0.25g and 512g (*n* = 1). Decisions in application adhered to diagnostic group, with two studies assessing in a pain population applying to two sites (0% bilateral). In these two studies, testing site continued to be determined by pain location (i.e. active or inactive joint, and surgical incision), with a comparative control site in the hand (thenar eminence, *n* = 1; dorsum, *n* = 1), or non-dominant forearm (*n* = 1). In contrast, one study assessing in a pain-free population applied to one site, selecting the abdomen as the testing site (*n* = 1; 100% quadrilateral abdomen).

When testing, Sieberg, Lunde, Shafrir, et al. (2023) and Sieberg, Lunde, Wong, et al. (2023) used the Von-Frey hair identified as MDTh as a minimum, increasing weight at each site until pain was self-reported (Sieberg, Lunde, Shafrir, et al., 2023) or described as 2 out of 10 on a NRS (Sieberg, Lunde, Wong, et al., 2023). This was repeated three times and expressed in analysis as a mean. Cornelissen et al. (2014) on the other hand did not report a minimum and increased in weight at each site until a self-verbalised "sharp" sensation. This was repeated twice and expressed in analysis as a mean.

Based on the synthesised evidence, evidence suggests that researchers consider the coinciding measure of MDTh when deciding whether Von-Frey hairs as appropriate for measuring MPTh. If appropriate, researchers should increase the weight of the hair until participants self-report a percept of pain.

Table 9

Included Studies in Systematic Review Measuring Mechanical Pain Threshold with a Von-Frey Hair Filaments

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Cornelissen et al. (2014)	1. JIA (*n* = 60, *Mdn* = 13.0 (9.6 – 15.5), 44f, 16m) 2a. US HC (n = 92, *Mdn* = 13.0 (10.0 – 13.8), 50% f)* 2b. EU HC (n = 151, *Mdn* = 11.0 (9.0 – 14.0), 50% f)** *Obtained from Meier et al. (2001) **Obtained from Blankenburg et al. (2010)	MPTh PPTh CPTh HPTh	MDTh VDTh CDTh WDTh	Von-Frey Hairs between 0.008g - 300g	Whilst on a clinic bed, Von Frey applied perpendicularly to skin at slightly different areas to avoid habituation at two sites for JIA: 1a) If had active joint*: affected joint with most significant inflammation, swelling or tenderness 1b) If no active joint, inactive joint*: joint with most inflammation in past 2) Control site: contralateral thenar eminence** 2 measures *Joint test site either distal or proximal interphalangeal joint, wrist, knee or ankle **Control data was pre-existing, comparing JIA thresholds to control thenar eminence	Verbalisation of "sharp" if sharp sensation	Mean of 2 measures	1. QST script provided 2. Participants provided time to adapt to room and equipment 3. Participant blinded from monitors 4. Participants asked to keep eyes closed 5. Parent/guardian present during testing
2	Sieberg, Lunde, Shafrir, et al. (2023)	1. Female pain free (*n* = 118, *M* = 23 (7); 12-19yrs, *n* = 58*) 2. Male pain free (*n* = 63, *M* = 30 (9); 12-19yrs, *n* = 10*) *Thresholds reported by decade age 12-19 yrs	MPTh PPTh HPTh CPTh	DMA MDTh TSP WDTh CDTh	Von-Frey Hairs between 0.008g - 300g	Von Frey filament applied to 4 sites: centre of the upper left and right quadrants and lower left and right quadrants of the abdomen. Filament weight ascending in strength. Min: Filament determined MDTh 3 measures	First self-report of pain.	Mean of 3 measures	1. Researchers could not test until proficiency demonstrated 2. Continuous protocol with natural breaks 3. Detail script to read verbatim for participants every study visit. 4. Monetary reimbursement
3	Sieberg, Lunde, Wong, et al. (2023)	1. Male with chronic post-surgical pain (*n* = 7, *M* = 15.0 (1.3)) Range: 14-17 yrs 2. Female with chronic post-surgical pain (*n* = 25, *M* = 13.6 (1.7)) Range: 10-16 yrs	PPTh MPTh	MDTh	Von-Frey Filaments between 0.25g and 512g 0.5mm diameter	*Filament applied to skin on dorsum of the left or right hand (random), or non-dominant forearm (control), and randomly to 3cm of left or right of surgical incision on lumbar spine (test). Min: Filament size recorded as MDTh *Completed first surgery visit and 4-6 months later	When stimulus weight that became painful; described as 2 / 10 on a NRS	Mean of 3 measures	1. Adapted protocol to be quicker and reduce burden in younger participants

Note. This table provides the data extracted from each study measuring mechanical pain threshold with von-Frey hairs. Abbreviations ordered alphabetically CDTh = Cold Detection Threshold; CPTh = Cold Pain Threshold; DMA = Dynamic Mechanical Allodynia; EU = European; f = Female; HC = Healthy Control; HPTh = Heat Pain Threshold; JIA = Juvenile Idiopathic Arthritis; m = Male; M = Mean; Mdn = Median; MDTh = Mechanical Detection Threshold; MPTh = Mechanical Pain Threshold; n = Sub-Group Sample Size; N = Total Sample Size; PPTh = Pressure Pain Threshold; QST = Quantitative Sensory Testing; TSP = Temporal Summation of Pain; US = United States; VDTh = Vibration Detection Threshold; WDTh = Warm Detection Threshold; and Yrs = Years.

2.3.4.2.2 Pressure Pain Threshold.

2.3.4.2.2.1 Deep Pressure Algometry for Pressure Pain Threshold.

Of the 62 studies testing PPTh, 58 included a form or deep pressure algometry (see Table 10). Eight of these studies (13.79%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter).

Excluding control values used by Cornelissen et al. (2014), Lieber et al. (2018) as these represented participants from a differing study, ~8494 CYP aged between 4 and 19 years participated in a study involving a PPTh assessment with deep pressure algometry (~4746 females and ~3749 males). Whilst three studies included an adult population (Sacramento et al., 2017; Scheper et al., 2017; Sieberg, Lunde, Shafrir, et al., 2023), methods did not appear to adapt for a paediatric population. Most studies compared PPTh between a diagnostic and control group (*n* = 37), with additional studies comparing within control groups (*n* = 12), within diagnostic groups (*n* = 7) and within athletic groups (*n* = 1). Where diagnoses were involved, the majority were pain-related (*n* = 33; 87.88% between group), neuropathic (*n* = 2; 100% between group), neurodivergent (*n* = 3; 100% between group), mental health (*n* = 2; 100% between group), orthopaedic disorder (*n* = 2; 0% between group) or other (*n* = 2). Methodological parameters typically referenced Fischer (1986) (*n* = 8), Rolke et al. (2006) (*n* = 8) and Blankenburg et al. (2010) (*n* = 4) as a standardised protocol. However, where Fischer (1986) is cited, 75% of studies were published prior to Rolke et al.'s (2006) protocol.

An algometer (including dolorimeter or pressure gauge) was most often used to induce pressure pain (*n* = 58), with one of these studies using an additional 13cm manually inflating blood pressure cuff as 2nd method to induce pressure. There was little consistency in the 38 studies describing size of the probe applied to the skin, including a 1cm^2 tip (*n* = 29; including semantics of 1cm, 1.1cm, and 10mm), 0.5cm^2 (*n* = 3), 1.5cm^2 (*n* = 2; including semantics of 1.52cm^2), 1.4cm^2 (*n* = 1) 3.5cm^2 (*n* = 1), 0.74cm^2 (*n* = 1), and 0.79cm^2 (*n* = 1).

On average, PPTh was tested at more sites than previous discussed thresholds measures ranging between one and 25 sites, however for studies where additional pain measures of interests were used number of sites were between one and three. Full list of

testing locations can be found in Table 10, however common sites include muscles in the hand (*n* = 28), back (*n* = 25), leg (*n* = 14), face (*n* = 14), and knee (*n* = 11). When comparing between a diagnostic and control group (*n* = 36) only 19.44% of studies discussed reported altering testing site for CYP (*n* = 7). Of these studies, six considered adapted pain-site to diagnosis (100% pain-diagnosis), and one ethical consideration of preventing harm (100% neuropathy).

Lack of consistency was observed when reporting both maximum and ramp rate values. Where both were available (*n* = 29), the most consistently described was a maximum of 10kg/cm^2 (or 1000kPa or 100N) at a ramp rate of 0.5kg/cm^2 (or 50kPa or 5N) (*n* = 7; 28.57% followed Blankenburg et al. (2010)), or a maximum of 11kg/cm^2 at a ramp rate of 1kg/cm^2 (*n* = 7; 85.71% followed Fischer (1986)). Additional maximum and ramps rate values reported in more than one study included a maximum of 10kg/cm^2 (or 1000kPa or 100N) and a ramp rate of 0.3kg/cm^2 (or 30kPa or 3N) (*n* = 3) and a maximum of 20kg/cm^2 and a ramp rate of 0.5 kg/cm^2 (*n* = 3). Ramp rate was often reported as a singular parameter (*n* = 22), common values included 30 k/Pa (*n* = 8), 1kg/cm^2 (or 1kg/s or 100N) (*n* = 7), 0.5kg/cm^2 (or 0.5kg/s or 5N) (*n* = 3), 20k/Pa (*n* = 3), and 10k/Pa (*n* = 1). Additionally used maximum and ramp rate values can be found in Table 10.

CYP were usually asked to self-report verbally PPTh (*n* = 27), including using “stop” (*n* = 12, including 1 study allowing hand raise too), “now” (*n* = 8), “yes” (*n* = 3), or a word of participants choice in a study with CYP as young as 5 (*n* = 1). Additionally where available, self-report included the CYP’s own signal (*n* = 1), or pressing a button when pain was felt (*n* = 14); two of these studies gave clarity with instructions, for example “You need to push the button on this controller when the pressure becomes painful to you” (*n* = 1), and “When you feel the sensation changes from pressure to the slightest pain, press the button immediately” (*n* = 1). A final method of self-report was provided by Ferracini et al. (2014) who asked CYP diagnosed with migraine, or without migraine to ring a bell when pressure became painful, providing instructions to report the exact moment sensation changed. PPTh algometry was the only method to include a study with researcher observations but did not specify what factors were considered. A further 12 studies explained CYP were asked to report when they

felt pain, but did not specify how. Two studies did not report whether PPTh was self or observer report.

Of the 47 studies that reported the number of measures they took at each site, typically three PPTh trial values were recorded (*n* = 37; 16.22% recorded the 1st value as a rehearsal and removed from analysis). Other studies reported two PPTh trial values (*n* = 8), and four PPTh trial values (*n* = 2; 100% recorded the 1st value as a rehearsal and removed from analysis). Reported PPTh was expressed in analysis as a mean across experimental trials (*n* = 48; 8.33% specified geometric); minimum quantity of pressure necessary for sensation of pain at each site (*n* = 9); and the reading value when an observer determined a perception of pain (*n* = 1).

Based on the synthesised evidence, evidence suggests that when using deep pressure algometry to measure PPTh, researchers record 3 trial values at the relevant testing site using a $1cm^2$ tip of an algometer. Forces delivered are at researcher discretion between a $10kg/cm^2$ maximum at a $0.5kg/cm^2$ ramp rate, or an $11kg/cm^2$ maximum at a $1kg/cm^2$ ramp rate. However, force should stop as soon as the participant reports a sensation of pain, or the relevant maximum value is met.

Table 10

Included Studies in Systematic Review Measuring Pressure Pain Threshold Measured with Deep Pressure Algometry

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Alfvén (1993)	Total (*N* = 140*, aged ~11yrs, 71f, 69m) 1. No Abdominal Pain, No Headache, No Chest Pains (*n* = 50) 2. RAP During Past 3 Months (*n* = 49) *Includes other groups not reported	PPTh	None	Algometer (0.74cm^2 tip)	Physician blind to group applied algometer to a quadricep muscle as a reference, and 5 left side muscles as test site: Temporal, Trapezius, Subclavius, Lateral insertion of the greater pectoral and Rectus abdominus near the umbilicus (identified as increased tenderness and tension amongst children with RAP) Ramp: 30 kpa/s Interpressure time: 10s 3 measures	Pressed button at first change of pressure to pain	Mean of last 2 measures	1. Age suitable to answer questions and reached puberty. 2. Before testing children acquainted and instructed with equipment 3. The test done in a calm, non-disturbing environment in school.
2	Anttila et al. (2002)	1. Migraine (*n* = 59, 32f, 27m) 2. Episodic TTH (*n* = 65, 21f, 44m) 3. No headache (*n* = 59, 37f, 22m) Group M_{Age}: 13.4 yrs	PPTh	None	Fischer Dolorimeter (1cm^2 tip)	Trained physiotherapist blind to group applied dolorimeter to 5 bilateral points (frontal muscle, temporal muscle, suboccipital muscle insertion, upper trapezius muscle, levator scapulae muscle). Max: 11kg/cm^2 Ramp: 1kg/cm^2/s	NR	Mean dolorimeter score across sites	1. Excluded supraspinatus muscles and anterior aspects of C5-7 because measurement is difficult in children
3	Balta and Arslan (2021)	1. Idiopathic Chest Pain (*n* = 72, *Mdn* = 15.00 (13.25 - 16.00), 38f, 34m) 2. HC (*n* = 54, *Mdn* = 15.00 (10.00 - 15.00), 28f, 26m)	PPTh	None	Dolorimeter	Algologist blinded to group applied algometer vertically and bilaterally to extrathoracic region (the supraspinatus origin, trapezius middle fiber midpoint, deltoid midpoint, and proximal medial tibial point) and the thoracic region (where the midclavicular line intersected with thoracic 2, 4, and 10 dermatomes). Max: 22kg/cm^2 Ramp: ~1kg/cm^2/s	Verbalisation of "stop" or raising hand when slight discomfort felt	Arithmetic mean of bilateral measures	NR
4	Blankenburg et al. (2010)	1. HC (*N* = 176, 88f, 88m) Range: 6-16.12 yrs Age bands: 6-8 yrs (24f, 24m) 9-12.2 yrs (32f, 32m) 13-16.12 yrs (32f, 32m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Pressure Gauge Device (1cm^2 probe)	Pressure gauge applied to both facial cheeks masseter, both dorsum of hands thenar muscle and ball of the feet in randomised order. Max: 10kg/cm^2 Ramp: ~0.5kg/cm^2/s 3 measures	Verbalisation of "now" when sensation becomes painful	Geometric mean of 3 measures	1. 1 of 2 assessors who received 4-week training in DFNS centre for QST research 2. Demonstration of pain induction at practice area above test area 3. Participant decided whether parents stayed in room or outside 4. 4-hr sessions with short breaks if concentration declined 5. Monetary reimbursement
5	Blankenburg et al. (2011)	1. HC (*N* = 173) Age bands: 7 yrs (42f, 44m) 14 yrs (43f, 44m)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA	Algometer (1cm^2 probe)	Algometer applied to both dorsum of hands thenar muscle. Max: 20kg/cm^2 Ramp: ~0.5kg/cm^2/s 3 measures	Verbalisation of "now" when sensation becomes painful	Geometric mean of 3 measures	1. Described to children presence and absence of pain as a likened to difference between sharp and blunt 2. Subjects decided whether their parents stayed in the testing room or outside

				WUR VDTh					3. Monetary reimbursement
6	Blankenburg et al. (2012)	1. Type-1 DM (*n* = 45, *M* = 13.2 (2.8), 23f, 22m) 2. HC (*n* = 45, *M* = 13.2 (2.6), 23f, 22m age-gender matched)	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Algometer	Algometer applied to both ball of the feet in randomised order Max: 10kg/cm^2 Ramp: ~0.5kg/cm^2/s 3 measures	Verbalisation of "now" when sensation becomes painful	Geometric mean of 3 measures	1. Monetary reimbursement
7	Buchanan and Midgley (1987)	1. HC (*N* = 190, *M* = 18.3 (17 - 9), 95f, 95m)	PPTh	None	Dolorimeter (incorporating a string gauge)	Dolorimeter applied to both dominant and non-dominant medial aspect of calcaneum, medial aspect of upper tibia, dorsal surface between thumb and forefinger, lateral aspect of midpoint of forearm and lower forehead. Measure repeated within 5 minutes In 40 participants repetition included exaggerating pain in expected response before tested only on the dorsal surface between right thumb and forefinger In 40 participants repetition included blindfolding whilst tested only on the dorsal surface between right thumb and forefinger	Intra- and inter-observer value, not specified on what communication.	Reading value	1. Avoid the influence of circadian variation in females, performed at same time of day i.e. between 12 a.m. and 2 p.m
8	Buskila et al. (2003)	1. Born preterm (*n* = 60, *M* = 14.1 (1.7), 36f, 24m) 2. Born full term (*n* = 60, *M* = 13.9 (1.7), 33f, 27m) Range: 11-18 yrs	PPTh	None	Dolorimeter (1.4 cm diameter footplate)	Dolorimeter applied vertically by one experienced observer to 9 tenderpoint sites - 5 sites on the right and 2 sites on both sides. Location stabilised by examiners non-dominant hand: 1) Right trapezius, midpoint of the upper fold. 2) Left trapezius, midpoint of the upper fold. 3) Right occiput below occipital prominence. 4) Right cervical spine, anterior aspect of intertransverse space at C5-7 5) Right second costochondral junction, just lateral to junction, on upper surface. 6) Right medial knees, medial fat pad of the knees overlying medial collateral ligament. 7) Left medial knees, medial fat pad of the knees overlying medial collateral ligament. 8) Right lateral elbow, 2 cm distal to lateral epicondyle; 9) Right greater trochanter, 2 cm posterior to greater trochanter. 4 control point sites used: 1) Forehead (middle) 2) Forearm (right distal third) 3) Lateral knee (right) 4) Shaft of the third metatarsal (right) Max: 9kg Ramp: 1kg/s	Verbalisation of "yes" when sensation changed from pressure to definite pain	Mean dolorimeter score	1. Measures conducted by one experienced observer 2. Site held by examiner to prevent painful shifting 3. Control site familiarisation to discourage anticipation of exaggerated response
9	Campi et al. (2020)	1. Without signs of painful TMD (*n* = 578, *M* = 12.70 (0.76), 317f, 261m) 2. With signs of painful TMD (*n* = 112, *M* = 12.80 (0.80), 72f, 40m)	PPTh	None	Algometer	One researcher applied algometer perpendicularly and bilaterally to temporal muscles, masseter muscles, lateral pole of the temporomandibular joints, trapezius muscles, and anterior tibial muscles.	Pressed button at first onset of pain	Mean of 3 measures	1. The researcher who was responsible for this examination underwent 15 hours of training to ensure an accurate assessment

		Range: 12-14 yrs				Ramp: 0.5kg/cm^2/s 3 measures Interstimulus interval: 5 min			
10	Chae et al. (2007)	1. Female adolescents (*N* = 46, *M* = 13.3 (0.1)) a. Low premenstrual syndrome b. High premenstrual syndrome	PPTh	None	Algometer (1cm diameter)	Whilst participant sat comfortable on an adjustable chair examiner blinded to group applied algometer perpendicularly to 8 point (6 acupuncture, 2 non-acupuncture) bilaterally whilst child sat comfortably in an adjustable chair: *Acupuncture points:* (A) PC6: 2 cm above the wrist crease between the tendons of the palmaris longus and flexor carpi radialis (B) TE5: 2 cm above TE4 between the radius and the ulna on the TE4 (on the transverse crease of the dorsum of the wrist in a depression on the ulnar side of the extensor digitorum communis tendon); TE10: 1 cm superior to the olecranon process in a depression with the elbow flexed (C) LI4: in the middle of the second metacarpal bone on the radial side (D) SP6: 3 cm directly above the tip of the medial malleolus on the posterior border of the tibia (E) GB39: 3 cm above the tip of the external malleolus in a depression between the posterior border of the fibula and the tendons of the peroneus longus and brevis (F) LR3: on the dorsum of the foot in a depression distal to the junctions of the first and second metatarsal bones. *Non-acupuncture points:* (A) 2 cm proximal to PC6. (D) 2 cm anterior to SP6 Applied at constant rate: 30 kPa/s 2 measures Interstimulus interval: ~5 minutes	When pressure became painful (not stated how)	Mean of 2 measures	1. Initial training session to familiarise participants with procedure and how to make threshold judgements 2. Conducted in a quiet room.
11	Chaves et al. (2013)	1. No Pain (GWP) (*n* = 62, *M* = 9.05 (1.29), 22f, 40m) 2. Joint Pain (GJ) (*n* = 10, *M* = 9.10 (1.20), 8f, 3m) (*does not add to 10 but as reported)* 3. Joint and Muscle Pain (GJMM) (*n* = 12, *M* = 9.67 (1.23), 7f, 5m) 4. Muscle Pain (GMM) (*n* = 5, *M* = 9.00 (1.58), 2f, 3m) 5. Pain during Mastication (GMAST) (*n* = 11, *M* = 9.45 (1.13), 7f, 4m) Range: 7-12 yrs Pain in context of temporomandibular joint and orofacial muscle pain	PPTh	None	Algometer (1cm^2 tip)	One of two examiners blind to group applied algometer perpendicular in a randomized order of sites. Bilateral sites included: MO, MB and MI: masseter origin, belly and insertion; TA, TM and TP: Anterior, middle and posterior portions of the temporalis muscle; TMJ: Temporomandibular joint, lateral pole, TR: Thenar region Max: 10kg/cm^2 Ramp: 0.5kg/cm^2/s 3 measures Interstimulus interval: 5 min (to re-evaluate each structure)	When pressure became painful (not stated how)	Mean of 3 measures	1. Algometry performed by two examiners trained for 15 hours 2. Familiarisation to thenar region of both examiner and child's right hand 3. Procedure was first explained in detail to the children, instructed about the difference in the perception of pressure and perception of the beginning of pain. 4. Rubber disk adapted to metal tip of the to avoid surface damage

12	Chen et al. (2000)	1. Cntrl (*N* = 44*, *M* = 13.1 (2.0), 41f, 3m) Range: 8-19 yrs **n* = 25 in control and experimental, *n* = 19 in just experimental	PPTh	None	Algometer (1cm diameter)	Whilst seated in a comfortable and relaxed position, algometer applied perpendicularly to myofascial trigger point (identified from palpitating middle finger extensor portion of the extensor digitorum communis muscle) Max: 11 kg/cm^2 Ramp: 1 kg/cm^2/s 3 measures Interstimulus interval: 10-20s	Verbalisation at a noticeable pain or discomfort	Mean of 3 measures	1. Procedure was explained clearly to the subject
13	Cheng et al. (2012)	1. Young offenders with low CU traits (*n* = 15, *M* = 16.7 (1.00), 15m) 2. Young offenders with high CU traits (*n* = 13, *M* = 16.9 (0.85), 13m) 3. TD (*n* = 15, *M* = 17.5 (1.77), 15m)	PPTh	None	Algometer (1.52 cm^2 probe)	Algometer applied perpendicularly to dorsal side of the proximal phalanx of the index finger on both hands. 3 measures	Verbalisation of "Yes" when feeling pain or discomfort	Mean of 3 measures	NR
14	Cornelissen et al. (2014)	1. JIA (*n* = 60, *Mdn* = 13.0 (9.6 – 15.5), 44f, 16m) 2a. US HC (n = 92, *Mdn* = 13.0 (10.0 – 13.8), 50% f)* 2b. EU HC (n = 151, *Mdn* = 11.0 (9.0 – 14.0), 50% f)** *Obtained from Meier et al. (2001) **Obtained from Blankenburg et al. (2010)	MPTh PPTh CPTh HPTh	MDTh VDTh CDTh WDTh	Algometer (1.1cm^2 area)	Whilst on a clinic bed, algometer applied perpendicular to two sites for JIA: 1a) If had active joint*: affected joint with most significant inflammation, swelling or tenderness 1b) If no active joint, inactive joint*: joint with most inflammation in past 2) Control site: contralateral thenar eminence** Max: 10kg/cm^2 Ramp: ~0.5kg/cm^2/s 3 measures MLI *Joint test site either distal or proximal interphalangeal joint, wrist, knee or ankle **Control data was pre-existing, comparing JIA thresholds to control thenar eminence	Verbalisation of "now" when pressure becomes uncomfortable	Mean of 3 measures	1. QST script provided 2. Participants provided time to adapt to room and equipment 3. Participant blinded from monitors 4. Participants asked to keep eyes closed 5. Parent/guardian present during testing
15	Cummins et al. (2021)	1. Community cntrl (*n* = 14, *M* = 16.4 (0.7), 12f, 2m) 2. No SH (in last year) Residential (*n* = 17, *M* = 16.5 (1.0), 1f, 16m) 3. SH 1-4 Episodes (in last year) Residential (*n* = 12, *M* = 16.2 (1.4), 4f, 8m) 4. SH 5+ Episodes (in last year) Residential (*n* = 21, *M* = 16.3 (1), 17f, 4m) Range: 13-17 yrs	CPTh HPTh MPTh PPTh	CDTh WDTh PHS TSL MDTh MPS DMA WUR VDTh	Algometer	Algometer applied to muscle at the base of the thumb. Max: 20 kg/cm^2 Ramp: 0.5kg/s 3 measures	Verbalisation of "now" when pressure first became painful	Arithmetic mean of 3 measures	1. Testing site avoided naive skin or scarification 2. Monetary reimbursement (withheld until consent to avoid inducement)
16	Derman et al. (2004)	1. Patients with Dysmenorrhea (*N* = 27, *M* = 15.6 (1.4), 27f) Range: 13-19 yrs	PPTh	PPTol	Algometer	Researcher blind to pain rating (not PPTh related) applied algometer perpendicularly to both sides of the trapezius muscle, palmar muscle and anterior proximal thigh. 3 measures	NR	Mean of 3 measures	1. Algometer calibrated before each subject
17	de Araújo Vitor et al. (2021)	1. With ID (*n* = 25, *M* = 11.5 (3.5), 15f, 10m) 2. Without ID (*n* = 25, *M* = 11.9 (3.7), 11f, 14m)	PPTh	None	Algometer (3.5cm diameter)	One single trained examiner applied algometer perpendicular to the extra oral surface of the temporal and masseter muscles, beginning on right side in following sequence: front, middle and back of the temporal muscle, followed by the	Activated algometer locking device when sensation changed from	Minimum quantity of pressure necessary for the first	1. Children told to relax facial muscle prior to measurement 2. All performed by single

						masseter muscle, (superior, middle, and inferior). Initial force: 0.5 kgf/cm^2/s Interstimulus time: 1 minute 12 evaluations per participant	pleasant to discomfort	sensation of pain	examiner trained in palpation of temporal and masseter muscles
18	Duarte et al. (2000)	1. Without RAP (*n* = 100, *Mdn* = 9.0, 55f, 45m) 2. With RAP (*n* = 100, *Mdn* = 9.2, 55f, 45m) Range: 5.0-15.8 yrs	PPTh	None	Algometer (1cm^2 tip)	One investigator applied algometer 90 degrees to body part, site order randomised. Sites as followed: 1) Hypochondriac regions, located on the hemi clavicular lines below the last costal margin 2) Epigastric region, located 4 cm below the xiphoid process on the alba line 3) Lateral regions, located at the intersection of the hemi clavicular line with the horizontal line, passing over the umbilicus 4) Umbilical region, located 2 cm from the umbilical scar on the right side of the body 5) Inguinal regions, located at the intersections of the hemi clavicular lines with the line joining the tubercles of the iliac crest 6) Pubic region, located 4 cm above the pubic tubercle on the alba lines 7) Tibial regions, located 5 cm from the tibial tubercles in the medial part of the tibias 8) Trapezial regions, located on the midpoints of the upper margin of the trapezius muscles 9) Deltoid regions, located 4 cm below the acromion in the lateral region of the deltoid muscles 10) Supraspinous regions, located immediately above the midpoint of the scapular spine above the supraspinous muscles Child lay supine decubitus position with examiner next to bed when threshold measures, other than trapezial and supraspinous regions when would be seated. Max: 10kg/cm^2 Ramp: 0.5kg/cm^2	Verbalisation at beginning of pain. Verbalisation chosen by child.	Minimum quantity of pressure necessary for the first sensation of pain	1. Algometer checked at 15-day intervals using weights of 0.7 and 4.2 kg. Calibrated at start and end of day with 10.0-kg weight. 2. Measurement during medical examination between 8 a.m. and 4 p.m., after a fast of at least 1 hour. 3. When the patient had daily pain attacks, a 24-hour period from previous before measuring PPTs. 4. The investigator and the examining room were the same for all patients.
19	Engebretsen et al. (2018)	1. Cntrl (*N* = 997, 481f, 516m) Most adolescents 15-17 yrs	HPTh PPTh CPTol	HPTol PPTol	Algometer (1cm^2 probe)	Algometer applied to cuticle of the ring fingernail of the non-dominant hand and on the non-dominant trapezius muscle, midway between the neck and shoulder joint. Max: 1000kPa Ramp: 30kPa/s 3 measures	Pressed button when sensation changed from pressure to pain	Mean of last 2 measures	NR
20	Fernández-de-Las-Peñas et al. (2010)	1. FETTH (*n* = 25, *M* = 8.9 (1.8), 19f, 6m) 2. HC (*n* = 50, *M* = 8.8 (1.7), 38f, 12m) Range: 5-11 yrs	PPTh	None	Algometer	Assessor blind to condition applied to bilateral temporalis muscle, upper trapezius muscle, second metacarpal, and the tibialis anterior muscle in a randomised order. Ramp: 30kPa/s 3 measures Interstimulus interval: 30s	Pressed switch at first change of pressure to pain	Mean of 3 measures	1. Children attended a preliminary session for familiarisation with the pressure test procedures. 2. Children were examined on days that were headache-free.
21	Fernández-de-Las-Peñas et al. (2011)	1. CTTH (*n* = 70, *M* = 9 (2), 51f, 19m) 2. HC (*n* = 70, *M* = 9 (1.8), 51f, 19m) Range: 6-11 yrs	PPTh	None	Algometer	Assessor blind to condition applied to bilateral temporalis muscle, upper trapezius muscle, second metacarpal, and the tibialis anterior muscle in a randomised order. Ramp: 30 kPa/s 3 measures Interstimulus interval: 30s	Pressed switch when sensation changed from pressure to pain	Mean of 3 measures	1. Children attended a preliminary session for familiarisation with the pressure test procedures. 2. Children were examined on days that were headache-free.

22	Fernández-Mayoralas et al. (2010)	1. FETTH (*n* = 30, *M* = 8.6 (1.7), 23f, 7m) 2. HC (*n* = 50, *M* = 8.5 (2.1), 36f, 14m)	PPTh	None	Algometer	Assessor blind to condition applied to bilaterally over supra-orbital (V1), infra-orbital (V2), mental (V3), median (C5), ulnar (C7), and radial (C6) nerves in a randomised order. (Identified by manual palpation). Ramp: 30 kPa/s 3 measures Interstimulus interval: 30s	Pressed switch when sensation changed from pressure to pain	Mean of 3 measures	1. Children attended a preliminary session for familiarisation with the pressure test procedures. 2. Children were examined on days that were headache-free.
23	Ferracini et al. (2014)	1. Diagnosis of migraine without aura (*n* = 50, *M* = 9.9 (1.6), 34f, 16m) 2. No history of headache for at least 6 mths (*n* = 50, *M* = 9.0 (1.4), 34f, 16m)	PPTh	None	Algometer (1cm^2 tip)	Assessor not blind to condition applied algometer perpendicularly to 18 sites divided bilaterally in a random order whilst standing, and child sat (sites identified by anatomical reference points and manual palptation): 1) insertion of the suboccipital muscles (IOM) 2) anterior aspect from the fifth to the seventh cervical (lower cervical) vertebra (AC5-C7) 3) supraspinal muscle (SS) 4) rostral margin of the trapeze (RMT) 5 extracephalic points: 5A) second costochondral joint (second intercostal space) (SIS) 5B) lateral epicondyle of the elbow (LEE) 5C) superolateral quadrant of the gluteal region below the iliac spine (SLQG) 5D) muscle insertions in the femoral trochanter major (FTM) 5E) medial condyle of the femur or medial margin of the knee (MMK), in the fat pad close to the joint line Max: 5kg/cm^2 Ramp: 0.5kg/cm^2/s 2 measures Interstimulus interval: 5 min	Rung table bell with a metal base (G-471®) when stimulus stopped being pressure and became painful.	Mean of 2 measures	1. Researcher demonstrated the test on self then placed in the child's hand for familiarisation. 2. Perception of pressure and of pain explained by pressing rubber disk to ventral region of arm 3. Research previously trained specifically with the aid of an algometry 4. Children instructed not to bear up with the pain an communicate the exact moment of change 5. Sites defined by American College of Rheumatology 7. Parental presence
24	Goubert et al. (2009)	1. Child (*n* = 53, *M* = 11.74 (1.73), 29f, 24m) Range: 9.17-15 yrs 2. Caregiver (*n* = 53, *M* = 40.51 (3.76), 43f, 10m) Range: 35-52 yrs** **Observer, used in other ratings but not for threshold and not tested on	PPTh	None	Algometer (10mm diameter tip)	Algometer applied perpendicularly to two pre-defined tender points of non-dominant side of child's body in counterbalanced order: 1) Suboccipital muscle insertion or occiput region 2) Anterior aspect of the interspaces between the transverse processes of C5–C7 or low cervical region** 6 pressures given below, on and above site. **Additionally measured 0.70 kg/cm2 above and below threshold. Used in pain test.	Verbalisation of "stop" when sensation of pressure changed to pain	Minimum quantity of pressure necessary for the first sensation of pain at each site	1. Child and caregiver received monetary reimbursement
25	Habechian et al. (2018)	1. Non practitioners (*n* = 30, *M* = 11.50 (1.94), 14f, 16m) 2. Amateur swimmers (*n* = 30, *M* = 11.56 (1.81), 18f, 12m) 3. Competitive swimmers (*n* = 30, *M* = 12.63 (2.02), 17f, 13m)	PPTh	None	Algometer (1cm^2 tip attached to string gauge)	Algometer applied over the UT (midpoint between the C7 vertebra and the posterolateral acromion), infraspinatus (muscle belly below the midpoint of the scapular spine), supraspinatus (muscle belly above the midpoint of the scapular spine), middle deltoid (muscle belly close to the inferolateral insertion), and tibialis anterior (halfway between the most superior attachment to the tibia and its tendon in the upper one-third of the muscle belly) in a randomised order. Rate: 1 kgf/s 3 measures Interstimulus interval: 30s	Pressed hand-controlled switch when sensation changed from pressure to pain	Mean of 3 measures	1. Familiarisation trial 2. Tested on non-swimming day (~24 hours without practice)

26	Han et al. (2012)	1. Boys (*n* = 258, *M* = 7.8 (2.1)) 2. Girls (*n* = 247, *M* = 8.0 (2.1)) Range: 4-11 yrs	PPTh	None	Algometer	One well trained examiner applied algometer whilst child sat comfortably and relaxed to three different sites in the brachioradialis muscle (random order): 1) the lateral epicondyle at elbow (site A, assumed to be the attachment trigger point site) 2) the mid-point of the muscle belly (site B, assumed to be the myofascial trigger point site) 3) the muscle-tendon junction as a control site (site C). Ramp: 1kg/s 3 measures Interstimulus interval: ~1 min	Verbalisation of "Yes" when begin to feel pain or discomfort	Mean of 3 measures	1. One well-trained examiner conducted all measurements 2. Procedure clearly explained to child
27	Hashkes et al. (2004)	1. GPain (*n* = 44), 17f (*M* = 8.1 (2.4)); 27m (*M* = 8.3 (2.5)) 2. HC (*n* = 46), 18f (*M* = 8.7 (1.8)); 28m (*M* = 8.5 (1.4)) Range: 4-12yrs	PPTh	None	Fischer Dolorimeter	Same experienced physician who as unblinded to group applied algometer to attain 25 measurements across the following sites: 1) 18 predefined points of FM (not stated) 2) 3 control points (distal right arm, forehead, and left thumb) 3) The mid-anterior tibia below the tibial tuberosity in both legs, where patients usually report pain during attacks and the lower back for possible referred pain Max: 10kg Ramp: 1 kg/s	When pressure became painful (not stated how)	Mean dolorimeter score	1. Same experience physician across all measurements
28	Hoehn et al. (2022)	1. Physically HC (*N* = 54, *M* = 9.05 (1.84), 20f, 34m) Range: 6-12 yrs	PPTh	CPM	Algometer (0.5 cm^2 diameter probe)* *Metronome assist to provide steady ramp rater on thumbnail	**Experimenter applied algometer perpendicular to child's right thumbnail. Max: 60N Ramp: 2N/s 2 measures **Same methodology used for measuring Experimental trial 1 (baseline, nonpainful conditioning stimulus, and painful conditioning stimulus), and Experimental trial 2 (baseline, painful conditioning stimulus, and nonpainful conditioning stimulus)	Verbalisation of "stop" when first felt painful	Mean of 2 measures	1. Monetary reimbursement and small toy for child, monetary reimbursement for parent 2. Raffle draw for interested families 3. Monetary reward for referring participants 4. Child comprehension of instructions assessed and repeated when needed. 5. Experimenter explained and demonstrated PPTh procedure on own thumb 6. 10 undergraduate researchers as experimenters, underwent rigorous training 7. Included practice PPTh before CPM experimental trial to familiarise procedure and have similar hand surface temp across participants. CPM set to 35C in this phase 8. Participant feedback questionnaire after testing
29	Hogeweg, Kuis, Huygen, et al. (1995)	1. HC (*n* = 69), 36f (*M* = 11.5 (3.1)); 33m (*M* = 11.5 (2.2)) 2. JCA (*n* = 57), 39f (*M* = 11.8 (3.2)); 18m (*M* = 12.2 (2.6))	PPTh	None	Algometer	An assessor blind to group status applied algometer to left and right of: 1) Elbow, wrist, knee and ankle at the level of their articular capsules. 2) The spinal processes of C6, TI, T3, T6, T10, LI, L3, L5	When pressure became painful (not stated how)	Mean of 3 measures	1. Participant unable to see results

						When elbow and wrist measured, participant sat. When knee and ankle measured, participant supine. When spinal processes measured, measured paravertebrally in spinal region. Max: 11kg/cm^2 Ramp: 1kg/cm^2/s 3 measures Interstimulus interval: 30s			
30	Hogeweg, Kuis, Oostendorp, et al. (1995)	1. JCA (*N* = 33) 1a. JCA inflamed knee (*n* = 16, 8f *M* = 11.6 (3.2), 8m *M* = 11.6 (2.1)) 1b. JCA inflamed ankle (*n* = 17, 14f *M* = 12.1 (2.9), 3m *M* = 11.3 (1.5)) 2. HC (*n* = 69, 36f *M* = 11.5 (3.1), 33m *M* = 11.5 (2.2)) Range: 6-17 yrs	PPTh	None	Algometer (1cm diameter tip)	An assessor blind to group status applied algometer to left and right of: 1) The spinous processes of C6, TI, T3, T6, TI0, LI, L3, L5 2) Knee at the medial side on the level of the articular capsule 3) Ankle at the lateral side at the level of the articular capsule When knee and ankle measured, participant supine. When spinal region measured, participant in prone position. Max: 11kg/cm^2 Ramp: 1kg/cm^2/s 3 measures Interstimulus interval: 30s	When pressure became painful (not stated how)	Mean of 3 measures	NR
31	Hogeweg et al. (1996)	1. 6-11 yrs (*n* = 38, *M* = 9.4 (1.3), 20f, 18m) 2. 12-17 yrs (*n* = 31, *M* = 14.0 (1.6), 16f, 15m)	MPTh (PPTh)	None	Algometer (1cm diameter tip)	Examiner blind to measurement applied algometer perpendicular to 4 peripheral joints, and paraspinal sites with segmental innervation of peripheral joints on both sides of body: 1) Elbow 2) Wrist 3) Knee 4) Ankle 5) Back (vertebrae C-6, T-1, T-3, T-6, T-10, L-1, L-3, and L-5) Max: 11 kg/cm^2 Ramp: 1kg/cm^2/s 3 measures Interstimulus interval: 30s Elbow and wrist examined with participant sitting Knees and ankles examined when in supine position (examiner sat right side of participant) Back examined in prone position (examiner sat left side of participant)	Verbalise "stop" first moment pressure became uncomfortable	Mean of 3 measures	1. Participant unable to see results 2. Explanation and demonstration of algometer on back of subject's hand for familiarisation 3. Examined in quiet room
32	Jovellar-Isiegas et al. (2022)	1. TD (*n* = 24, *M* = 9.60 (2.30), 10f, 14m) 2. Unilateral CP (*n* = 23, *M* = 9.27 (2.27), 14f, 9m)	PPTh	Tactile registration Single-point localization Two-point discrimination Double simultaneous Graphesthesia Sterognosis of familiar object Stereognosis of forms Texture perception Proprioception	Algometer	Whilst participants were blinded, sat up right with feet on floor, arm on table and elbow flexed 90degrees, algometer was applied perpendicularly to two sites bilaterally on the palm of both hands, starting with less affected then more affected hand: 1) Deltoid zone (middle fibres) 2) Dorsal zone of hand (space between the 2nd and 3rd metacarpal). Algometer stabilised between the researcher's index and middle fingers. Ramp: 1kg/cm^2/s	Make a signal when pain felt	Mean of 3 measures	1. 2 trials on less-affected hand before scoring - 1 eyes open, 1 closed to ensure understanding of instructions 2. Tested on two non-consecutive days to prevent fatigue (somatosensory always second day) 3. Tests performed in the same order and instructions 4. Noise-and-distraction-free room for testing

				Functional sensibility		3 measures Interstimulus interval: 1 min			
33	Kersch et al. (2020)	1. Children with chronic pain (*N* = 98, *M* = 13.10 (2.43), 65f, 33m) Range: 7-18 yrs	Deep PPTh (1a) Deep PPTh (2a)	Pain intensity to static light touch Pain intensity to dynamic light touch Pain intensity punctate pressure Pain intensity to repetitive punctate pressure Pain intensity to cool stimuli (1b) Repetitive PPTh TPS (2b) Repetitive PPTh TPS	(1a) Algometer (2a) Manually inflating 13cm blood pressure cuff	(1a) Algometer applied opposite to pain site, ipsilateral and contralateral thenar eminence or ball of big toe and dorsal forearm or tibialis anterior (control sites) and again to site adjacent to but not directly over main pan site. Max: 7.5 kg/cm^2 (1b) Cuff applied to subsample of 51 participants. A single stimulation by inflating cuff at test sites (mid-upper arm or widest part of the lower leg) to determine deep PPTh. Limb nearest primary pain (pain site) and both contralateral and most remote limbs (two control sites). Max: 300 mmHg	(1a) Verbalisation of "Stop" when sensation just began to hurt (1b) Verbalisation of "Stop" when sensation just began to hurt	Minimum quantity of pressure necessary for the first sensation of pain at each site	1. Procedure explained and demonstrated before testing 2. Testing paused or stopped if signs of distress
34	King et al. (2017)	1. JFM (*n* = 34, *M* = 15.42 (1.41), 34f) 2. HC (*n* = 31, *M* = 14.57 (1.28), 31f)	PPTh	None	Algometer	Algometer applied in a randomised and stratified order bilaterally to: 1) palm of the hand (i.e., thenar eminence of the thumb) 2) above the supraorbital ridge of the forehead (i.e., arch of the eyebrow) Max: 4kg/cm^2 Ramp: 1kg/cm^2/s 3 measures Interstimulus interval: 30s	First sensation of pain (not clear how)	Mean of 3 measures from left and right sides averaged into a single score for each site	1. Monetary reimbursement 2. Interval to reduce sensitisation
35	Kjeldgaard Pedersen et al. (2023)	1. Orthopaedic disorder (*N* = 72, *Mdn* = 10.2 (6-14), 33f, 39m)	PPTh	None	Algometer	**Investigator applied algometer to thenar of hand, holding algometer perpendicular to first metacarpal bone. Ramp: 20kPa/s 2 measures **Part of four interventions, measure same each time	Verbalisation of "stop" when pressure begins to feel like pain	Mean of 2 measures	1. Standardised verbal information provided at start of study 2. One investigator completed all measurements
36	Li et al. (2023)	1. Post MSK surgery (*n* = 100, *M* = 14.5 (2.0), 80f, 20m) Range: 10-18 yrs 2. Post MSK injury (*n* = 177, 14.4 (1.9), 84f, 93m) Range: 11-17 yrs	PPTh HPTh CPTol	TSP CPM	Algometer (1cm^2 probe)	Whilst sat at a table with legs uncrossed and planted on floor, and arm on table, algometer applied to the dorsal forearm with the participant's arm palm down, 2 cm distal to the elbow crease at the maximal prominence of the extensor digitorum muscle body and then at the trapezius (C7 spinous process and the spine of the scapula), applied to biggest bulge of the interceding muscle body (3 fingers lateral of the spinous process and 2 fingers above scapular spine) Max: 1000kPa Ramp: 35kPa/s 4 measures Interstimulus interval: 10s	Pressed trigger when pressure stimulus became painful, "You need to push the button on this controller when the pressure becomes painful to you."	Mean of last 3 measures	1. Each of the 15 researchers completed standardised training of 8-hour workshop on QST, 20-hours of training and independent practice over 5 weeks, an initial proficiency assessment and a final mock QST testing demonstration to confirm that the procedures were mastered. 2. Each researcher needed to either conduct 2 QST testing per month or one testing and observe an additional testing per month. 3. Each researcher QST proficiency were evaluated every 6 months with a training record 4. Children provided scripted instructions, expectations and taught how to use the pain-rating

									scale 5. Breaks provided where children waited in quiet private area without electronic or family access 6. HPTh/PPTh practice trial 7. Participant not aware of CPTol ceiling time
37	Lieber et al. (2018)	1. Survivors of paediatric ALL (*n* = 46, *M* = 9.8 (3.1), 18f, 28m) Range: 12.5 yrs 2. HC (*n* = 46, age-gender matched)* *Obtained from Blankenburg et al. (2010) and Blankenburg et al. (2011)	CPTh HPTh MPTh PPTh	MDTh VDTh CDTh WDTh PHS TSL MPS DMA WUR	Algometer ($1cm^2$ probe)	Algometer applied bilaterally to dorsum of both feet, or if clinical symptoms on an affected body site (randomised order). Max: $20kg/cm^2$ Ramp: $0.5kg/cm^2/s$ 3 measures MLI	Verbalisation of "now" when pain experienced	Mean of 3 measures	1. Each of the 15 researchers completed standardised training of 8-hour workshop on QST, 20-hours of training and independent practice over 5 weeks, an initial proficiency assessment and a final mock QST testing demonstration to confirm that the procedures were mastered. 2. Each researcher needed to either conduct 2 QST testing per month or one testing and observe an additional testing per month. 3. Each researcher QST proficiency were evaluated every 6 months with a training record 4. Children provided scripted instructions, expectations and taught how to use the pain-rating scale 5. Breaks provided where children waited in quiet private area without electronic or family access 6. HPTh/PPTh practice trial 7. Participant not aware of CPTol ceiling time
38	Lyng et al. (2023)	1. OSD (*n* = 27, *M* = 13.0 (1.5), 12f, 15m) 2. Cntrl (*n* = 22, *M* = 13.4 (1.4), 11f, 11m)	PPTh	None	Algometer ($1cm^2$ tip)	*Whilst sat comfortably on a plinth with legs over the side, a trained assessor applied algometer perpendicularly bilaterally to: 1) Test limb at the mid-portion patellar tendon (OSD = painful knee, control = randomly assigned) 2) Muscle belly of the tibial anterior 3) Rectus femoris (at a site measured as 15 cm proximal from the basis of patella) Ramp: 30kPa/s *Performed before and after isometric wall squat	Pressed button on handheld device when pressure sensation changed to pain	Minimum quantity of pressure necessary for the first sensation of pain	1. Conducted by trained assessor with extensive testing experience and delivering information to adolescents 2. Experimental procedures piloted in young adolescents with and without OSD prior to testing to ensure study comprehension and comfort 3. Pain threshold instruction was standardised to age
39	Mensink et al. (2022)	1. Patients with JIA (*n* = 16, *M* = 14.8 (2.1), 12f, 4m) Range: 9-18 yrs 2. HC (*n* = 16, matched by age (within 1 year) and age)	CPTh HPTh MPTh PPTh	CDTh WDTh TSL MDTh WUR VDTh MP/DM Allodynia	Pressure Gauge Device ($1cm^2$ probe)	Algometer applied to affected knee and the unaffected control knee in JIA, and right knee in healthy control. Max: 10 kg/cm^2 Ramp: $0.5kg/cm^2/s$ 3 measures	Verbalisation of "now" when sensation of pressure becomes painful	Geometric mean of 3 measures	1. DFNS protocol with modifications for children and adolescents 2. QST performed by trained personnel 3. Practice trial performed before each test to ensure understanding

40	Metsahonkala et al. (2006)	1. With migraine (*n* = 48, 27f, 21m) 2. TTH (*n* = 61, 19f, 42m) 3. No headache (*n* = 59, 37f, 22m) Group *M*: 13.4	PPTh	None	Fischer Dolorimeter	Physiotherapist blinded to group applied dolorimeter to five cranial and neck-shoulder points (frontal muscle, temporal muscle, suboccipital muscle insertion, upper trapezius, and levator scapulae muscle) and three extracephalic points (elbow, knee and medial gluteal muscle). Max: 11kg/cm^2 Ramp: 1kg/cm^2/s	When pressure reported as painful (not clear how)	Mean scores across peri cranial and extracephalic	NR
41	Nikolajsen et al. (2011)	1. Orthopaedic disorder (*N* = 50, 23f (*M* = 8.5 (2.3)); 27m (*M* = 8.0 (2.4)) Range: 4-12 yrs	PPTh	None	Algometer (1cm^2 probe)	Algometer was applied to: (1) the lateral aspect of the calf on the affected side above the gastrocnemius muscle and approximately where the upper 1/3 of the lower leg meets the lower 2/3 of the lower leg (if both legs were affected, the leg ipsilateral to the dominant hand was examined) (2) the thenar of the dominant hand. Ramp: 20kPa/s 3 measures Trial based on who conducted assessment (both raters trained in pressure algometry): 1) Rater 1 examination (twice) 2) Rater 1 and 2 examination (one each) 3) Rater 1 examination (once) 15 to 30 minute between examinations	Verbalisation of ‘‘stop’’ when sensation of pressure changed to pain	Mean of last 2 measures	1. Algometry performed same day in quiet and non-stressful conditions 2. Participants carefully introduced to procedure, youngest children observed a demonstration on a teddy bear. 3. Raters trained in pressure algometry
42	Ocay et al. (2022)	1. Chronic MSK Pain patients (*n* = 302, *M* = 14.93 (1.95), 247f, 55m) Range: 10-18 yrs 2. Age-matched Cntrl (*n* = 80, *M* = 14.99 (1.96), 32f, 48m) Range: 10-18 yrs	PPTh HPTh	MDTh WUR DMA VDTh MPSumm WDTh CPM	Algometer	Algometer applied perpendicular to left volar forearm under underlying bone or muscle in cntrl, and left volar forearm as control area followed by most painful anatomical region (affected area) in MSK Ramp: 5N/s 3 measures	Verbalisation of "stop" when pain experienced	Mean of 3 measures	1. Adapted from Rolke et al. (2006) Blankenburg et al. (2010) and Ferland et al. (2018) to reduce complexity, time and fit with clinical constraints
43	Pas et al. (2019)	1. FAPD (*n* = 39, *Mdn* = 9 (3), 25f, 14m) 2. HC (*n* = 36, *Mdn* = 9 (2), 21f, 15m) *Parents also however threshold not measured	PPTh	CPM	Algometer (1cm^2 area tip)	Researcher blinded to group applied algometer perpendicularly in a random order to: 1) Symptomatic region: umbilical region 2) Remote test: trapezial region 3) Remote test: tibial region Ramp: 1 kg/cm^2/s 3 measures Interstimulus interval: 30s	Verbalisation of “stop” when sensation changed from pressure to pain	Mean of last 2 measures	1. Participants washed hands and drank fruit juice to avoid vasovagal reactions on CPM 2. All measures conducted by two trained researchers 3. Participant not to bear up pain, and communicate at exact moment
44	Rathleff et al. (2013)	1. HC (*n* = 22, *M* = 17.1 (.09), 22f) 2. PFPS (*n* = 57, *M* = 17.3 (1.1), 57f) Range: 15-19 yrs	PPTh	None	Algometer (1cm^2 probe area)	Algometer applied perpendicular to the skin at: 1) Knee at 4 bony landmarks (3 cm medial to the midpoint of the medial edge of the patella, 2 cm proximal to the superior edge of the patella, 3 cm lateral to the midpoint of the lateral edge of the patella, and at the centre of the patella). 2) Muscle belly of the tibialis anterior (5 cm distal to the tibial tuberosity) During measures participant reclined with knee flexed to approximately 20°, with a small pillow beneath knee. Ramp: 30 kPa/s 2 measures	When the sensation changed from sensation of pressure to pain (not clear how)	Mean of 2 measures	NR

45	Richards et al. (2021)	1. Elite youth football players (*N* = 34, *M* = 17 (1), 34m) Range: 16-18 yrs	PPTh	None	Algometer (1cm^2 probe)	*Whilst participant lay supine on a medical plinth with non-dominant knee bent to 90 degrees and exposed, with foot plantar side down on medical plinth, algometer applied perpendicular to tibial and sural nerves in random order. Ramp: 30kPa/s 3 measures Interstimulus interval: 30s *Initially inter-rated (R1, R2,R3) on all measures and then two weeks later remeasured for intra-rater (R1)	Verbalisation when sensation alters from non-unpleasant pressure to unpleasant pain	Mean of 3 measures	1. Standardised verbal instructions for PPTh and printed on a poster above medical plinth for reference 2. Familiarisation to procedure applying algometer to point on participant wrist 3. Tested same time and prior to training / 3 days post-competitive match to prevent fatigue 4. All raters undergone one-hour training session of neural points, protocol and recording 5. Rater practice on asymptomatic participants not involved in the study prior to testing
46	Sá and Silva (2017)	1. With chronic idiopathic neck pain (*n* = 40, 21f, 19m) 2. Asymptomatic (*n* = 40, 21f, 19m) Group M_{Age} = 17.2 ± 0.56	PPTh	None	Algometer (0.5 cm diameter probe)	Physiotherapist not blind to group applied algometer to: 1) Right and left upper trapezius (at the mid distance between the posterior angle of the acromion and C7) 2) Right and left articular pillar between C1 and C2 (approximately 1 cm lateral and above the spinous process of C2) 3) Right and left articular pillar of C5/C6 (approximately 1 cm lateral to the mid distance between the spinous processes of C5 and C6, which were identified by palpation) 4) Over the right tibialis anterior (lateral to the medial malleolus). Participants in prone with head aligned to be relaxed in all measures except tibialis anterior where participant was supine. Max: 60N Ramp: 3N/s 3 measures Interstimulus interval: 30s	Verbalisation "stop" when the sensation changed from pressure to pain	Mean of 3 measures	1. Participant familiarised with procedure by demonstration in their hand 2. Measures taken in school 3. Physiotherapist with 10+ years clinical practice
47	Sacramento et al. (2017)	1. Adult (*n* = 35, *M* = 23.4 (3.4), 21f, 14m) Range: 22.3-24.6 yrs 2. Children (*n* = 35, *M* = 9.1 (1.7), 20f, 15m) Range: 8.6-9.7 yrs	PPTh	None	Algometer (1cm^2 tip)	Physical therapist applied algometer bilaterally at 6 points in random order: 1) Upper trapezius (halfway between the neck and shoulder over the muscle fibres) 2) Infraspinatus (muscle belly below the spine of the scapula) 3) Supraspinatus (middle point over the fossa of the scapula) (4) Deltoid (muscle belly close to the deltoid tuberosity) 5) Articular pillar of C5-6 zygapophyseal joint 6) tibialis anterior (halfway between the superior most attachment to the tibia and its tendon in the upper third of the muscle belly). Whilst assessed, participant was seated in a chair with erect spine, feet on floor, and coxofemoral and tibiotarsal joints at 90 degrees. Upper limbs flexed and slightly abducted, hands relaxed on thighs, and head and cervical region relaxed, neutral position and forward facing.	Pressed switch when sensation of pressure became painful	Mean of 3 measures	1. Physical therapist familiarised participants by explaining and demonstrating on thenar muscles of hand to show how algometer eels and increased pressure that may or may not be painful 2. Region and muscles chosen due to high pain and mechanical stress in these regions of children

						Ramp: 1kg/cm^2/s 3 measures Interstimulus interval: 30s			
48	Scheper et al. (2017)	1. HMS/EDS-HT (*N* = 74) 1a. Child <18 (*n* = 43, *M* = 13.6 (2.8), 22f, 21m) 1b. Adult >=18 (*n* = 31, *M* = 37.4 (12.6), 31f) 2. GJH (*n* = 74, *M* = 20.0 (3.9), 64f, 10m) 3. Cntrl (*n* = 77, *M* = 25.8 (10.3), 61f, 17m) (does not total but as reported)	PPTh	None	Algometer (0.79 cm^2 area tip)	Algometer applied by assessors blind to hand dominance, group and symptoms bilaterally to 6 sites: 1) Middle of the deltoid muscle at the latitude of the axilla 2) 1st dorsal interosseus muscle, in the middle of the skin web between thumb and index finger 3) Extensor carpi radialis longus muscle at the proximal third of the forearm 4) Paraspinal muscles 5 cm lateral to the spinous process of L3 and T8 5) Tibialis anterior muscle, midway and lateral to the tibia 6) Middle of the gastrocnemius muscle at the proximal third of the calf Ramp: 1kg/s 3 measures Interstimulus interval: 10s	When uncomfortable pressure level reached, just short of pain (not clear how)	Mean of last 2 measures	NR
49	Sherry and Sapp (2003)	1. Reporting enthesalgia (*n* = 68, *M* = 11.9 (2.2), 45f, 23m) 2. Not reporting enthesalgia (*n* = 166, *M* = 11.8 (2.2), 95f, 71m)	PPTh	None	Pressure Gauge Device (1cm^2 probe)	Algometer applied to 7 entheses, and 5 control sites: Entheses: 1) Plantar fascia insertion on all metatarsal heads 2) Plantar fascia insertion on the calcaneus 3) Achilles tendon insertion 4) Tibial tuberosity 5) Inferior pole of the patella 6) Greater trochanter 7) Lower sacroiliac (SI) joint (at the inferior insertion of the dorsal iliosacral ligament) Control: 1) Thumbnail of the non-dominant hand 2) Superior posterior iliac spine 3) Mid-tibia 4) Calcaneus 5) Proximal great toe phalange Max: 11kg/cm^2 Ramp: 1kg/cm^2/s	When felt pain (not clear how)	Minimum quantity of pressure necessary for the first sensation of pain	NR
50	Sieberg, Lunde, Shafrir, et al. (2023)	1. Female pain free (*n* = 118, *M* = 23 (7); 12-19 yrs *n* = 58*) 2. Male pain free (*n* = 63, *M* = 30 (9); 12-19 yrs *n* = 10*) *Thresholds reported by decade age 12-19 yrs	MPTh PPTh HPTh CPTh	DMA MDTh TSP WDTh CDTh	Algometer	Algometer applied to 4 sites: centre of the upper left and right quadrants and lower left and right quadrants of the abdomen. Max:100N 3 measures Interstimulus: 30s	Self-report pain (not clear how)	Mean of 3 measures	1. Modified from Rolke et al. (2006) 2. Researchers could not test until proficiency demonstrated 3. Continuous protocol with natural breaks 4. Detail script to read verbatim for participants every study visit. 5. Monetary reimbursement
51	Sieberg, Lunde, Wong, et al. (2023)	1. Male with chronic post-surgical pain (*n* = 7, *M* = 15.0 (1.3)) Range: 14-17 yrs 2. Female with chronic post-surgical pain (*n* = 25, *M* = 13.6 (1.7)) Range: 10-16 yrs	PPTh MPTh	MDTh	Algometer	*Algometer applied to dominant forearm or randomly to left/right dorsum of the hand (control), and randomly to 3cm of left or right of surgical incision on lumbar spine (test). Max: 100N Ramp: 3N/s 3 measures	Self-report pain (not clear how)	Mean of 3 measures	1. Adapted protocol to be quicker and reduce burden in younger participants

						*Completed first surgery visit and 4-6 months later			
52	Soee et al. (2013)	1. FETTH (*n* = 23, *M* = 10.5 (2.5), 13f, 10m) 2. CTTH (*n* = 36, *M* = 12.6 (2.2), 28f, 8m) 3. HC (*n* = 57, *M* = 10.7 (2.3), 36f, 21m)	PPTh	SupraPPT	Algometer ($1cm^2$ tip)	Algometer applied perpendicularly to non-dominant sites: 1) Dorsum of the second finger's interphalanx 2) Anterior temporal region where palpation revealed the belly of the muscle during contraction 3) M. trapezius, the point halfway between C7 and acromion Ramp: 10kPa/s 3 measures Interstimulus interval: 2 min	Pressed button when pressure became painful - "When you feel the sensation changes from pressure to the slightest pain, press the button immediately".	Mean of 3 measures	1. Only non-dominant side of body tested as testing at upper limit of concentration, and to avoid differences in tissue composition and pain sensitivity according to hand dominance 2. Familiarisation demonstration at each site before testing
53	Stabell et al. (2014)	Students (*N* = 961, *M* = 16.1 (0.4), 469f, 492m)* *Includes 861 cntrl and 77 IBS participants Range: 15-17 yrs	HPTh PPTh CPTol	HPTol PPTol	Algometer ($1cm^2$ tip)	Pressure was applied to the cuticle of the ring fingernail of the right hand and on the midline of the right trapezius muscle and in shoulder height with a handheld algometer Max: 1000kPa Ramp: 30kPa/s 3 measures	Pressed button when non-painful pressure changed to pain	Mean of last 2 measures	NR
54	Valentino et al. (2020)	1. JIA (*n* = 51, *M* = 12 (3), 41f, 10m) 2) HC (*n* = 52, *M* = 12 (3), 29f, 23m)	PPTh	None	Algometer ($1cm^2$ tip)	Whilst participant sat in dental chair, relaxed with teeth apart, the algometer was applied by a single examiner in a randomised order perpendicular to: 1) Left and right anterior temporalis (AT) (determined by palpation) 2) Left and right masseter (MM) muscles (determined by palpation) 3) Left and right TMJ (over the lateral TMJ condyle pole) 4) Left and right thenar eminence (TH) (point that connects the longitudinal axis of the thumb and index finger) Ramp: 20kPa/s 4 measures Interstimulus interval between site: 5s Interstimulus interval between measurements: 2s	Pressed button as soon as the pressure sensation became painful	Mean of last 3 measures	1. First measurement excluded as usually highest
55	Vervoort et al. (2008)	1. Child (*n* = 84, *M* = 11.82 (1.70), 44f, 40m) Range: 9-15 yrs 2. Caregiver (*n* = 84, *M* = 40.39 (4.29), 71f, 13m) Range: 33-55yrs* *Observer, used in other ratings but not for threshold and not tested on	PPTh	None	Algometer ($1cm^2$ tip)	Algometer applied by examiner in a counterbalanced order to 2 tender points on child's non-dominant side: 1) Suboccipital muscle insertion region (i.e. region in the neck) 2) Anterior aspect of the interspaces between the transverse processes of C5–C7 (i.e. region on the shoulder)	Verbalisation of "stop" when started to feel pain	Minimum quantity of pressure necessary for the first sensation of pain at each site	1. Two examined described pain procedure and shown algometer 2. Difference between pain threshold with pain tolerance explained; PPTh does not mean 'not being able to stand it anymore' 3. Chosen tender points so experimenter could stay behind the child, and not interfere with experimental conditions
56	Weiss et al. (2014)	1. ERA (*n* = 30, *Mdn* = 13 (11 - 15), 12f, 18m) 2. Cntrl (*n* = 30, *Mdn* = 12 (7-15), 14f, 16m)	PPTh	None	Dolorimeter ($1.5cm^2$ tip)	Paediatric rheumatologist applied dolorimeter bilaterally to test sites: 1) Common extensor tendon on lateral humerus epicondyle 2) Common flexor tendon on medial humerus epicondyle 3) Quadriceps at superior patella 4) Patellar ligament at inferior patella 5) Achilles tendon 6) Plantar fascia at the calcaneus.	Verbalisation of pain	Minimum quantity of pressure necessary for the first sensation of pain at each site	1. Children blinded from pressure amount 2. Second paediatric rheumatologist examined 8 random children

						And control sites: 1) Non-dominant thumbnail 2) Bilateral mid-trapezius muscle 3) Bilateral 2 cm distal to the lateral epicondyle 4) Bilateral 1 cm posterior to greater trochanter Max: 4.5kg Ramp: ~ 0.5kg/s			
57	Winger et al. (2014)	1. Patients with CFS (*n* = 120, *M* = 15.4 (1.6), 86f, 34m) 2. HC (*n* = 39, *M* = 15.2 (1.6), 28f, 11m)	PPTh	None	Algometer (0.5cm^2 tip)	Assessed in the same order for all participants, a researcher not blinded to group applied algometer bilaterally to 3 predefined sites: 1) fingernail of the third finger 2) skin superficial to the trapezius (ascending part) 3) supraspinatus muscles 2 measures Interstimulus interval: 10 min	Verbalisation of "stop" when at pain threshold	Mean of 2 measures	NR
58	Wollgarten-Hadamekl et al. (2011)	1. Moderate burn injuries (*n* = 12, *M* = 12.3 (1.9), 8f, 4m) Range: 10–16 yrs 2. Severe burn injuries (*n* = 10, *M* = 12.7 (1.8), 6f, 4m) Range: 10–16 yrs 3. Cntrl (*n* = 20, *M* = 13.3 (1.9), 10 f, 10 m) Range: 10–16 yrs	HPTh PPTh	HPTol PPTol Ischemic PTh Ischemic PTol	Algometer (1cm^2 tip)	Whilst seated with eyes closed, algometer applied to participants non-dominant thenar Max: 10kg/cm^2 Ramp: 0.5kg/cm^2/s 1 probe trial at dominant thenar 3 measures	Verbalisation of "now" when pain experienced	Mean of 3 measures	1. Participant familiarised with lab and equipment 2. Participant could not see value on computer 3. Monetary compensation

Note. This table provides the data extracted from each study measuring pressure pain thresholds with deep pressure algometry. Abbreviations ordered alphabetically CDTh = Cold Detection Threshold; Cntrl = Control; CP = Cerebral Palsy; CPTh = Cold Pain Threshold; CFS = Chronic Fatigue Syndrome; CTTH = Chronic Tension-Type Headache; CU = Callous Unemotional; DFNS = Quantitative Sensory Testing in the German Research Network on Neuropathic Pain; DM = Diabetes Mellitus; DMA = Dynamic Mechanical Allodynia; ERA = Enthesitis-Related Arthritis; f = Female; FAPD = Functional Abdominal Pain Disorder; FETTH = Frequent Episodic Tension-Type Headache; GJH = Generalised Joint Hypermobility; GP = Growing Pains; HC = Healthy Control; HMS/EDS-HT = Hypermobility Syndrome; HPTh = Heat Pain Threshold; HPTol = Heat Pain Tolerance; IBS = Irritable Bowel Syndrome; ID = Intellectual Disability; JCA = Juvenile Chronic Arthritis; JFM = Juvenile Fibromyalgia; JIA = Juvenile Idiopathic Arthritis; kPa = Kilopascal; m = Male; M = Mean; M_{Age} = Mean Age; Mdn = Median; MDTh = Mechanical Detection Threshold; MPS = Mechanical Pain Sensitivity; MPSumm = Mechanical Pain Summation; MPTh = Mechanical Pain Threshold; mths = Months; MSK = Musculoskeletal Pain; n = Sub-Group Sample Size; N = Total Sample Size; NR = Not Reported; OSD = Osgood–Schlatter Disease; PHS = Paradoxical Heat Sensation; PFPS = Patellofemoral Pain Syndrome; PPTh = Pressure Pain Threshold; PPTol = Pressure Pain Tolerance; QST = Quantitative Sensory Testing; ROI = Rate of Increase; RAP = Recurrent Abdominal Pain; s = Seconds; SH = Self-Harm; TD = Typically Developing; TSL = Thermal Sensory Linen; TTH = Tension-Type Headache; TMD = Temporomandibular Disorder; TSP = Temporal Summation of Pain; VDTh = Vibration Detection Threshold; WDTh = Warm Detection Threshold; WUR = Wind-Up Ratio; and Yrs = Years.

2.3.4.2.2.2 Other Equipment for Pressure Pain Threshold.

Of the 62 studies assessing PPTh (two described MPTh), four studies used equipment other than an algometer (see Table 11). None of these studies (0%) were involved in a wider battery (defined as including 4+ thresholds of interest to this chapter). A total of ~219 CYP aged between 4 and 15 years participated in a study involving a PPTh assessment with other equipment use (~71 female and ~148 male). There were three studies comparing PPTh between a neurodevelopmental diagnostic and control group (66.66% Autism, 33.33% Cerebral Palsy) or pain-related diagnostic and control group (100% migraine). No study reported to follow a standardised protocol such as Rolke et al. (2006) or Blankenburg et al. (2010) as expected given the differing equipment use.

Studies involving an Autistic group used a dynamometer with a $1cm^2$ probe at a maximum force of 13kgf and a ramp rate of 5N/s (or 50kPa or $0.5kgcm^2$), however consistency in description reflects both studies being published by the same author. The study involving a Cerebral Palsy group used an esthesiometer with a $1mm^2$ tip applying varied weight (20-500g) at a 15s interstimulus interval, and a study involving migraine used a pneumatically accelerated plastic cylinder (6mm diameter) at a 3.9m/s velocity. All studies involved an initial familiarisation task, Autistic studies demonstrated at the testing site (1 study omitted thenar eminence), Cerebral Palsy demonstrated at a differing site (the arm) and migraine did not specify. Testing sites differed between studies. Studies involving Autistic CYP tested bilaterally, with one study involving the lips, cheeks, thenar eminence, thumb pads, index finger pads, and hands dorsi (*n* = 1), whilst the other only included the thenar eminence (*n* = 1). However, where one site was chosen additional thermal detection and pain thresholds were considered. In contrast, studies involving Cerebral Palsy tested on the plantar surface of the foot, including the big toe, first metatarsal, fifth metatarsal, mid arch and heel (bilaterality unclear) and migraine tested at the pad of the distal phalanx of left index finger. Only CYP with Cerebral Palsy were blinded from viewing the testing procedure, this was not specified in Autistic CYP. All studies conducted three measures at each testing location,

Verbal and non-verbal cues were used to self-report PPTh (*n* = 3), with Autism studies using the word “pain” or a non-verbal hand raise (*n* = 2) and Cerebral Palsy studies

using the word “now” or a pre-arranged signal (*n* = 1). Report of PPTh was not stated in the migraine study. Only studies involving CYP with Cerebral Palsy provided explanation in how to report threshold describing this to be when they would like the pain to go away Whilst studies involving Autistic CYP allowed parental (*n* =1) or teacher presence (*n* = 1) to observe signs of distress if pain was non-communicated and subsequently stop the procedure. Where migraine groups were included, consideration to prevent harm were evident with testing occurring approximately 27 days after last attack. All studies used a mean value of the three measures to express PPTh.

Based on the synthesised evidence, whilst other equipment could be used to measure PPTh when a wider battery is not of interest, evidence suggests that researchers defer to deep pressure algometry as a standardised approach.

Table 11

Included Studies in Systematic Review Measuring Pressure Pain Threshold with Equipment Other Than Deep Pressure Algometry

Table no.	Reference	Participant Information	Additional Pain Induction	Other Inductions	Equipment	Methodology	Threshold Measure	Data Processing	Ethical Considerations
1	Riquelme et al. (2016)	1. Autistic (*n* = 27, *M* = 6.3 (3.32), 7f, 20m) 2. TD (*n* = 30, *M* = 6.5 (3.37), 15f, 15m)	PPTh	MDTh Sterognosis Propriceptive skills Finer Motor Skills	Dynamometer (1cm^2 probe)	Dynamometer applied in a pseudorandom order to 12 bilateral sites: 1) Lips 2) Cheeks 3) Thenar eminences 4) Thumb pads 5) Index finger pads 6) Hand dorsi. Max: 13kgf Ramp: 5N/s 3 measures	Verbalisation of "pain", or raise hand when pressure became painful	Mean of 3 measures	1. Avoiding anxiety, children familiarised with procedure using nonpainful stimuli in the same body locations. 2. Ensure children correctly understood and dealt with any distress. 3. In case non-communication of pain in Autistic children, teacher observed procedure to report signs of distress that would stop procedure.
2	Riquelme et al. (2023)	1. Autistic (*n* = 38, *M* = 10.94 (4.15), 14f, 24m) 2. TD (*n* = 34, *M* = 9.68 (2.75), 20f, 14m)	CPTh (°C and s at 0°C) PPTh	TDTh WDTh CDTh	Dynamometer (1cm^2 probe)	Examiner unblinded to group applied dynamometer bilaterally to thenar eminence of hand palms. Max: 13kgf Ramp: 5N/s 3 measures	Verbalisation of "pain", or raise hand when pressure became painful	Mean of 3 measures	1. Familiarised with procedure using different stimuli in the body locations other than hand palm (i.e. hand dorsum, arm) 2. Examiner experienced in assessing thresholds 3. Ensure children correctly understood and dealt with any distress. 4. In case non-communication of pain in Autistic children, parent observed procedure to report signs of distress that would stop procedure.
3	Weidenbacker et al. (1963)	1. Cntrl (*n* = 30, *M* = 9.83) Range: 4.75–14.08 yrs 2. CP (*n* = 30, *M* = 9.58) Range: 4.08–14.83 yrs *Implied male population from use of he	PPTh	None	Esthesiometer (1mm^2 tip)	Algometer applied to skin at 5 areas of plantar surface of the foot: 1) Big toe (BT) 2) First metatarsal (1 MT) 3) Fifth metatarsal (5 MT) 4) Mid-arch (MA) 5) Heel (H). Range: 20 - 500g 3 measures Interstimulus interval: 15s Measurement at site: 15s Re-stimulation used from mark in skin	Verbalisation of "Now" or other prearranged signal	Mean of 3 measures	1. To explain how to report threshold, child told in a friendly way to report when, "you would like it to go away." 2. Identical and relaxed testing area to minimise fear and distraction 3. Familiarisation on the arm 4. Board in place to cover feet avoiding blindfold use to reduce child apprehension
4	Zohsel et al. (2008a)	1. Migraine (*n* = 15, *M* = 12 (1.5), 7f, 8m) Range: 10-14 yrs 2. Cntrl (*n* = 15, *M* = 12.3 (1.5), 8f, 7m) Range: 10-15 yrs	MPTh (PPTh)	None	Small plastic cylinder pneumatically accelerated in a guiding barrel (length: 12 mm, weight: 0.5 g, diameter: 6 mm)	Whilst seated comfortably stimuli administered to pad of distal phalanx of left index finger* Ramp: 3.9m/s 3 measures *Determined prior to an oddball paradigm	NR	Mean of 3 measures	1. Prior to the experiment, participants received several practice trials to ensure that the children had understood the task. 2. Migraine group tested approximately 26 days after last pain episode. 3. Parent waiting in adjacent room 4. Procedure and equipment shown to parent and child 5. Experiment administered in an electrically shielded room

	6. White noise headphones to cancel out noise

Note. This table provides the data extracted from each study measuring pressure pain thresholds with equipment other than deep pressure algometry. Abbreviations ordered alphabetically CDTh = Cold Detection Threshold; Cntrl = Control; CP = Cerebral Palsy; CPTh = Cold Pain Threshold; kgf = Kilogram Force; m = Male; M = Mean; n = Sub-Group Sample Size; N = Total Sample Size; PPTh = Pressure Pain Threshold; TD = Typically Developing; TDTh = Tactile Detection Threshold; WDTh = Warm Detection Threshold; and Yrs = Years.

2.4 Discussion

Given the lack of guidance for conducting pain psychophysics in paediatric populations, this systematic review aimed to summarise and critically appraise psychophysical methodologies used across age, sex and diagnosis. Evidence synthesis constituted from 87 studies, including a total of 11,026 participating CYP between 4 and 19 years, and a higher yield of female (~6115) to male (~4892) participants. In this review, synthesised data outlined each studies reported demographics, equipment use, and methodological parameters such as minimum, maximum and ramp values, testing location, pain threshold report and definition. Generally, studies provided clarity in outlining the methodological parameters used, often referring to Rolke et al.'s (2006) and Blankenburg et al.'s (2010) protocols for guidance. Yet whilst these referenced protocols suggest adult parameters are feasible and replicable in use amongst paediatric populations aged 4 to 19 years, methodological differences were still observed dependent upon the sole, or limited use of pain modalities and their broad ethical considerations.

In reviewing thermal thresholds, parameters for assessing HPTh, and CPTh were mostly standardised to Rolke et al. (2006) and Blankenburg et al. (2010). For example, applying a 9cm^2 contact thermode to the hand at a 32°C baseline, with a minimum of 0°C for CPTh, and maximum of 50°C for HPTh. This suggests when assessing these thermal thresholds, researchers defer to these methodological parameters as guidance. However, whilst CPTh methodology was consistent, differences in approach were evident when CPTol was an additional measure. A pattern in equipment-based decisions was observed with the suggested use of a 9cm^2 contact thermode when CPTh was assessed as a singular cold pain measure diverting to a form of CPT when both CPTh and CPTol were assessed. This pattern places emphasis on how researchers should adapt their equipment-based decisions dependent on the number of cold pain assessments they intend to use, utilising a contact thermode when assessing CPTh alone, and a CPT when assessing both CPTh and CPTol.

Similarly, despite both CPTh and CPTol using the hand as a testing site, temperature parameters were conflicted when CPTol was an additional measure. For example, whilst the previously described temperatures were implemented when assessing

CPTh alone, there was little consistency for assessing CPTol with suggestions ranging from 0 to 10.8°C. Additionally, there was little evidence for the ethical implications of how the chosen temperatures may impact a paediatric population. For example, studies like Saxena et al. (2015) failed to differentiate temperatures between age groups, instead applying a 0-1°C temperature across an 8–70-year age range. Whilst Birnie et al. (2012) did suggest these temperatures were feasible for assessing CPTol in paediatric populations, their review highlights few studies utilise a temperature as low as, for example, 1°C. Given this lack of consideration, researchers should consider Birnie et al.'s (2012) suggestion of a higher water temperature ranging between 5-10°C when CYP are involved and adapt their methodology for ethical purposes when multiple age groups are compared.

Studies considering broad mechanical thresholds (MPTh and PPTh) also displayed methodological inconsistencies, particularly in evidencing the use of standardised methodological parameters. Undeniably, PPTh was the most common psychophysical method of pain assessment amongst CYP of any diagnostic group, yet the most inconsistently reported. Although most PPTh studies used an algometer with a $1cm^2$ tip and applied this to the CYP's hand or back over three trials, only 50% of studies provided the maximum and ramp rate values. However, when these were reported, common parameters adhered to Blankenburg et al. (2010) ($10kg/cm^2$ maximum, $0.5kg/cm^2$ ramp rate) and Fischer (1986) ($11kg/cm^2$ maximum, $1kg/cm^2$ ramp rate). It could be argued that either would be appropriate methodology, however with most studies utilising Fischer (1986) being published prior to 2006, researchers defer to Blankenburg et al. (2010) as a modern protocol. In contrast, continued use of these protocols may not always be feasible as PPThs were most often tested as a sole pain measure, not as a battery like these standardised protocols suggest. In response, researchers should aim to create a standardised protocol for assessing PPTh as a sole measure, accounting for how this approach could be modified alongside other modalities as required.

In contrast, the assessment of MPTh provided more consistent recommendation for the use of weighted pinprick mechanical stimulators as Rolke et al. (2006) and Blankenburg et al. (2010) suggest. Use of these standardised protocols were further evident in the application of a series of 5 ascending and descending stimuli until a prick, or blunt sensation

were perceived. However, some studies outlined additional use of Von-Frey hairs; here equipment use may rely on the additional modalities used, with those following a QST protocol utilising pinpricks, and those considering MDTh as an additional modality Von-Frey hairs. Additionally, consideration of location for MPTh and MPS were not clear, although the thenar eminence of the non-dominant hand can typically be considered as a control site for MPTh in pain-diagnostic groups. In making these testing site decisions, consideration of diagnostic group may provide guidance in applicable use.

The involvement of a diagnostic group appeared to account for changes in psychophysical methodology. The diagnoses of a population were used to understand which testing site should be used to ensure participants were prevented from harm or tested in a diagnostically valid site. For example, in populations with a neuropathy or mental health diagnosis with a history of self-harm, researchers should determine an appropriate location that prevents participants experiencing unwarranted pain as Cummins et al. (2021) and Lieber et al. (2018) demonstrated. Furthermore, in populations with a pain-related diagnosis, the immediate, and contralateral location most affected by this diagnosis should be assessed. This suggests researchers should consider how even brief pain experiences can cause potential tissue damage in sensitive areas and modify their testing protocol for prevention. However, no study considered the impact of testing site amongst populations with a neurodevelopmental diagnosis which may be important. For example, in an Autistic population shifting testing location to account for increased sensitivity to sensory input could allow an Autistic CYP to complete a study as opposed to becoming distressed from overstimulation and withdrawing. Therefore, researchers should apply knowledge for adapting testing location to any diagnostic population to improve the feasibility for completing a psychophysical study in a safe, and ethical manner. Additionally, diagnoses of a population influenced decisions for parental presence. Although studies were broadly conflicted in these decisions studies involving Autistic CYP promoted parental presence, ensuring additional safety precautions by recognising their role in advocating for Autistic CYP's differing communication styles instead of relying on self-report alone (Riquelme et al., 2016; Riquelme et al., 2023). More studies should follow this example and consider modifying their protocol to facilitate an environment that can ensure the inclusion of CYP

who may use varying methods of pain communication. Such an approach in an experimental setting may facilitate in allowing Autistic CYP to engage with full QST batteries; an observation this review failed to provide despite their adult predecessors having experienced these (Vaughan et al., 2020).

Although sex was not considered when designing psychophysical studies, overall, more female participants participated in psychophysical studies than male participants suggesting female pain may be a greater research interest. Yet little ethical guidance was provided on how psychophysical procedure may be adapted for sex. For example, with a higher proportion of female participants, researchers may need to consider the impact of the menstrual cycle in the timing of when individuals participate. This is of importance as although conflicted, there is evidence to suggest an increase in pain perception during the perimenstrual, and menstrual period (Fatima et al., 2014). Therefore, if an individual participates during this period of their menstrual cycle, their experiences of pain may not be representative of their day-to-day experiences. Here, researchers should consider broader sex factors that could influence the representativeness of their psychophysical studies.

In contrast ethical considerations were provided in the context of paediatric developmental stage. Some studies emphasised a need for promoting pain comprehension to ensure CYP safety, and validity in self-reports. Methods to promote these included explanations of when to report pain (i.e. training CYP when to press a button (Truffyn et al., 2021) or releasing of button for age comprehension (Valkenburg et al., 2015)), or age-appropriate explanations of what pain is (i.e. Blankenburg et al. (2010) described CPTh as feeling stinging). Researchers should take both age and developmental stage into consideration in their study design to ensure material inclusivity and fundamentally prevent harm. However, further consideration should be provided to make these materials developmentally engaging for populations like Autistic CYP who are often not observed in pain research due to, for example a fear of pain (Karos et al., 2018). Here studies should consider modifying materials for their participants use which Valkenburg et al.'s (2015) use of comic books and a plush teddy for inclusivity of both an age and developmental perspective may provide guidance. However, researchers should include their population of

interest in the creation of these engagement-based approaches to ensure the applicability of their modified materials.

Although this review provides important insight, methodological limitations may hinder its application in experimental settings. Whilst a second reviewer was consulted to aid in inclusion and exclusion-based decisions, they only reviewed 10% of articles at each methodological stage. Without double screening of all articles the lead reviewer's own interpretative bias may have allowed studies that would broadly be considered as inclusive, to be excluded. Thus, studies which may have developed understanding particularly in less conclusive methods like CPTh testing location in the absence of CPTol could have been overlooked limiting the application of this review. Additionally, interpretation of ethical considerations was often at reviewer discretion, as consideration of typically reported ethics such as informed consent are presumed and thus not an information source in the current review. Instead, methodologies were analysed to select plausible ethical considerations; yet again the reviewer's selection bias may have caused ethical adaptations others would deem important to not be reported.

It could be argued a further limitation arose in the decision to exclude articles utilising a CPT published before April-19-2012 due to likely inclusion within Birnie et al.'s (2012) review, as available and eligible evidence was likely missed. However, whilst a relevant argument, with this evidence already being succinctly summarised previously, the available evidence was still utilised in supporting experimental recommendations in the current review as opposed to merely replicating existing work. Therefore, whilst eligible articles that may have contributed to the conclusion for CPT use in CPTh and CPTol may have been excluded initially, the continual reference to Birnie et al.'s (2012) work still provides insight into the broader available evidence, even if summarised by differing authors.

An additional limitation outlines the bias of available paediatric pain literature focusing on paediatric populations with a pain-related diagnosis. Consequentially, the findings of this review may be limited in application, only providing clarity in designing studies where pain-related diagnoses are included. Additionally, most studies focused on

PPTh, providing understanding of how mechanical pain can be assessed but limiting guidance for additional pain modalities such as thermal thresholds. To counter these limitations, future psychophysical research should include broad diagnostic populations, particularly mental health, neuropathy, or neurodevelopment which are widely underserved in paediatric pain. These psychophysical assessments should additionally include more modalities than PPTh alone to allow an extensive understanding of the differing sensory mechanisms – an especially beneficial approach for the Autistic population who reportedly experience sensory sensitivities as a diagnostic feature.

In conclusion, pain psychophysical assessments are feasible in use amongst paediatric populations when trying to understand differences in pain thresholds and tolerance. When considering methodological parameters, standardised guidance can be provided by Rolke et al. (2006) protocol, Blankenburg et al. (2010) protocol, and Birnie et al.'s (2012) systematic review. Still, researchers should show consideration for a paediatric population's own experience and comprehension of pain as a construct to ensure measures are precise, and fundamentally safe. Here, a requirement to create a standardised methodological protocol that is modified to facilitate in attaining an evidence base for populations that are often not represented in QST studies is emphasised. However, when applying such a protocol, researchers should consider the needs of their population to ensure results are reliable, and ethical.

Chapter 3.

Autistic and Non-Autistic Children and Young People's Pain Psychophysical Assessments

3 Autistic and Non-Autistic Children and Young People's Pain Psychophysical Assessments

3.1 Introduction

As discussed in Chapter 1.1.1, the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) diagnostically consider Autistic individuals to be hyposensitive to pain compared to non-Autistic peers. Psychophysical methods like those outlined in Chapter 2 have been used to assess whether pain hyposensitivity is plausible as a diagnostic feature within Autistic adults (Vaughan et al., 2020). However, these empirical findings have challenged the perceived unidirectional approach of pain sensitivity and emphasised inconsistent patterns in Autistic adults. To develop this understanding in a paediatric context, the continued use of pain psychophysics would allow identification of if, and how sensitivity in sensory modalities differ between Autistic and non-Autistic CYP. This would provide fundamental understanding of whether pain mechanisms may contribute to diagnostic overrepresentation or if consideration of the broad pain experience is required.

When considering pain sensitivity across a multitude of somatosensory domains (i.e. MPTh, PPTh, HPTh and CPTh), evidence predominantly suggests pain thresholds of Autistic adults do not significantly differ from non-Autistic adults (Bird et al., 2010; Failla et al., 2020; Failla et al., 2017; Fründt et al., 2017; Thaler et al., 2017; Vaughan et al., 2020). Thus, current adult evidence does not support diagnostic perceptions of Autistic pain hyposensitivity. Instead within-group differences in pain responses are observed, with both Vaughan et al. (2020) and Fründt et al. (2017) demonstrating subgroups of Autistic adults show clinically significant hypo- or hypersensitivity on a standard QST battery. For example, Vaughan et al. (2020) found that a greater number of Autistic adults than non-Autistic adults individually displayed clinically relevant data points (i.e. MPTh hyposensitivity). However, it must be acknowledged that the greater number of data points included the broader QST battery assessments such as touch detection, and measures of thermal thresholds using contact thermodes (i.e. HPTh and CPTh). Therefore, any clinically relevant data points for these assessments are not relevant to the purpose of the current thesis (pain thresholds), or

the apparatus used to measure CPTh (CPT). However, this still suggests subjective pain experiences that are typically considered and evaluated amongst neurotypical populations are present amongst Autistic adults. Yet these may be presumptuously overlooked due to diagnostic misconceptions or differences in observed pain expressions.

On the other hand, unidirectional differences in hypo- and hypersensitivity have been observed amongst thermal pain. For example, despite not showing significant differences in thermal thresholds, Yasuda et al. (2016) suggested Autistic adults possess a hyposensitive pain profile based on providing lower subjective pain intensity ratings. In contrast, Cascio et al. (2008) found significant thermal hypersensitivity, with both Hoffman et al. (2023) and Failla et al. (2020) reporting significant higher pain intensity ratings at lower heat pain thresholds. However, these differences may be indicative of a broader sensory facet related to Autism such as intolerance to temperature, reflecting a thermal sensitive phenotype rather than a diagnostically defined pain sensitivity (Casterman et al., 2024). Despite the overarching consensus the thermal threshold differences emphasising the complexity of pain within Autistic adults, and the presumptuous perception of pain hyposensitivity amongst this population should not be summarised to a subjective experience.

Compared to Autistic adults, availability of evidence for if Autistic CYP's pain thresholds differ from non-Autistic CYP is sparse, with Nicolardi et al. (2023) commenting that only one of the five QST studies included in their systematic review involved an Autistic adolescent group. Even within this sparsity, similar psychophysical conflicts identified in Autistic adults emerge. For example, Duerden et al. (2015) identified that despite displaying higher thermal thresholds, these did not significantly differ from non-Autistic CYP, whilst Li et al. (2024) suggested Autistic CYP experience MPTh hyposensitivity through higher MPTh's. However, the most current and pertinent profile amongst Autistic CYP is pain hypersensitivity, with studies suggesting Autistic CYP possess significantly lower PPTh's than non-Autistic peers (Chen et al., 2017; Fan et al., 2013; Riquelme et al., 2016). Whilst this could suggest Autistic CYP are hypersensitive to pain, particularly PPTh, the limited availability of data reduces the strength of this argument. Instead, a continued consideration of multiple pain modalities would contribute to broadening understanding of individual pain

experience, and the sensory mechanisms that contribute to these diagnostic perceptions of pain.

In line with available evidence, testing of MPTh and PPTh in Autistic CYP would provide clarification for conflicts in mechanical thresholds, with inclusion of an additional thermal threshold corresponding to Duerden et al.'s (2015) approach. However, whilst Duerden et al. (2015) assessed both HPTh and CPTh, evidence suggests Autistic adults have differing experiences of pain at suprathreshold levels (Failla et al., 2020; Hoffman et al., 2023). Yet whether these findings are replicated in the paediatric population remains unclear. Here CPTol should be assessed as a thermal suprathreshold alongside CPTh, as Chapter 2 highlights these assessments are frequently used together.

Moreover, findings suggest Autistic adults self-rate pain higher than non-Autistic adults which may suggest pain appraisal pathways differ amongst the Autistic population (Failla et al., 2020; Hoffman et al., 2023). For example, with the high co-occurrence of anxiety within the Autistic population, pain assessments may be influenced by a heightened emotional state which in turn heightens the intensity of the pain experience (Failla et al., 2020). However, whether these appraisal pathways differ again between Autistic and non-Autistic CYP remains unknown, as predominant pain sensitivity evaluations have been provided through bystander-proxy reports during medical procedures (Allely, 2013). Thus, measuring self-reported pain intensity ratings would provide thoughtful insight into Autistic CYP's self-appraisal of pain, and highlight whether differences in pain are fundamentally mechanistic, or potentially consequential to appraisal.

Chapter 2 clarified paediatric pain thresholds and tolerance can be feasibly measured experimentally using psychophysical methods, providing clear understanding of QST parameters that allow assessment validity. However, evidence synthesis highlighted a need for adapting protocols to the diagnostic population to facilitate engagement. Recent literature has further emphasised this need through a modified QST (mQST) which adapts typical QST protocol for populations who often rely on non-verbal communication for pain to be perceived (Barney et al., 2020). Some psychophysical research has begun to consider such modifications amongst Autistic CYP, including allowing parental presence to help

monitor non-verbal pain communication, and report pain on the CYP's behalf when required (Riquelme et al., 2016; Riquelme et al., 2023). However, these mQST's are not a standardised necessity to facilitate the participation of populations who may find QST research difficult, which may explain the current limited availability of Autistic pain threshold and tolerance data (Karos et al., 2018). Considering this, the current study will apply psychophysical modifications developed alongside Autistic individuals to promote participatory engagement - a nuanced approach in the current pain landscape.

Fundamentally understanding of Autistic CYP's pain is limited, emphasising a need to provide further and concise evidence by assessing pain thresholds and tolerances. Using a between subjects' design, this study used QST parameters outlined in Chapter 2 to assess differences in MPTh, PPTh, CPTh, CPTol and subsequent pain intensity measures between Autistic and non-Autistic CYP. Additional modifications were applied in line with literature and PPI advice to increase Autistic accessibility and inclusivity. However, such modifications did not contribute to biased results in favour of Autistic CYP as evidence suggests neurodivergent adaptations equally support non-Autistic CYP (Alderson, 2018). Findings aim to broaden understanding as to whether a diagnostic criterion for pain hyposensitivity reflects Autistic CYP's pain experiences. However, hypotheses were generated from available literature which predominantly suggests PPTh hypersensitivity in Autistic CYP compared to non-Autistic CYP, and higher self-ratings of pain in Autistic adults than non-Autistic adults. Therefore, it is hypothesised:

H_1: Autistic CYP will experience significantly lower pain thresholds and tolerance than non-Autistic CYP

H_2: Autistic CYP will rate their pain significantly higher than non-Autistic CYP

3.2 Method

3.2.1 Design

A case-control design was used to measure differences in pain thresholds, tolerance, and intensities between Autistic and non-Autistic CYP. Four psychophysical pain

assessments were used to measure MPTh, PPTh, CPTh and CPTol, with additional self-reported measures of MPTh, PPTh and CPTol pain intensity.

3.2.1.1 Design Validation and Accessibility

A PPI panel (see Chapter 0.1.2) was consulted to assess applicability, validity and inclusivity of study design and materials. All recommendations for improvement were implemented prior to study commencement. For example, panel members explained age-appropriate context should be provided on study flyers as if they saw an advert which inflicted pain, they would not want to take part, but if they understood this pain as an uncomfortable feeling their anxiety would reduce. The panel members provided thoughtful insight in defining each QST measure within the study flyer, such as a likening cold pain felt during the cold-pressor to cold pain when holding an iced slushy. Additionally, the PPI panel suggested adding context to pain intensity numerical values as the originally proposed scale included a standard NRS. Upon further discussion of interview findings (see Chapter 4.3.2.1) and PPI member's lived experience, this need for context was emphasised and deemed important in aiding pain description and comprehension. Here a PPI member referenced a pain intensity scale which provided such context that they, and other Autistic people found useful; this was amended for study use (see Chapter 3.2.3.3.4).

In the lab, CYP and their caregiver were encouraged to familiarise themselves with the environment. When comfortable, the PhD researcher provided age and diagnostic appropriate PISs to both the CYP and their caregiver, before explaining verbatim the study procedure and allowing discussion of what participation would entail. The researcher then demonstrated study procedure on themselves and if comfortable, the CYP and caregivers who satisfied safety-based inclusion criteria. There were no time constraints to this familiarisation process.

3.2.2 Participants

From January 2023 to February 2024 a snowball sample of 29 CYP with low support needs were recruited for participation. Diagnosed Autistic participants were grouped as Autistic CYP (n = 9, M_{age} = 12.55 , SD = 1.74, 6 Male) and participants not diagnosed as, or suspected to be Autistic grouped as non-Autistic CYP (n = 20, M_{age} = 12.50, SD = 1.19, 11 Male).

3.2.2.1 Inclusion Criteria

All participants were aged 11-16 years to ensure study comprehension, a similar adolescent development and of an age where assessment of interventions is more pliable (Kelly, 2022; Visser-Bochane et al., 2020; World Health Organization, n.d.).

To prevent harm, participants were not eligible for inclusion if they had circulatory problems (including Raynaud Syndrome), sensory neuropathy (including diabetes), skin complaints (including eczema), or a severe neuropsychiatric condition (including severe depression). Interpretation of neuropsychiatric condition severity was of caregiver discretion given the high co-occurrence amongst Autistic CYP (Mingins et al., 2020; Stewart et al., 2022). Additionally, to prevent skewed data, participants must not have been experiencing current pain which would affect their daily behaviours and must not have consumed pain medication in the 24 hours prior to participation, or at an average of 3+ times a week.

3.2.3 Materials

3.2.3.1 Recruitment Materials

3.2.3.1.1 Social Media Poster.

Primary caregiver recruitment was targeted using social media posters (see Appendix 2). Posters provided inclusion criteria, brief methodology and contact details for the caregiver to express interest on their CYP's behalf.

3.2.3.1.2 YouTube Videos.

3.2.3.1.2.1 Video Information Sheet.

Two subtitled videos hosted through private YouTube link were created to support PISs and reduce anxiety from interactive uncertainty (Gowen et al., 2019). During each video, the PhD researcher orally introduced themselves, provided a study explanation and specified inclusion for Autistic CYP (see Appendix 3.1), or non-Autistic CYP (see Appendix 3.2). Videos were sent to caregivers upon expression of interest.

3.2.3.1.2.2 YouTube Video – Study Walkthrough for All CYP.

To prevent feelings of anxiety regarding study uncertainty (Gowen et al., 2019; Neilson & Bond, 2023), a first-person walkthrough of the study was filmed and posted via private YouTube link. The video began at the initial meeting on LJMU property, displayed psychophysics equipment, demonstrated study procedure and ended by leaving LJMU property (see Appendix 3.3).

3.2.3.2 Questionnaire

Demographic information (i.e. age and identifying gender) was obtained via a paper questionnaire. Instructions of how to answer questions were provided in a visual and verbal format to be inclusive of differing processing styles (see Appendix 4).

3.2.3.3 Quantitative Sensory Testing Battery

The QST battery and relevant values reported are consistent with systematic findings for psychophysical pain assessment outlined in Chapter 2.4.

3.2.3.3.1 Mechanical Pain Threshold.

MPThs were measured using 7 pinprick mechanical stimulators, each weighted at a fixed intensity force between: 8, 16, 32, 64, 128, 256 and 512mN (MRC Systems, 2010a).

Using a method of limits (Rolke et al., 2006), the 0.25mm diameter tip of each pinprick was applied perpendicularly to the left volar forearm with a 2-second contact period.

The applied fixed intensity force systematically ascended until participants self-reported an initial sharp pain percept through verbalisation or by displaying a “NOW” sign which stopped the ascending application. The fixed intensity force of the pinprick perceived as painful was recorded as trial prick value (mN). If participants did not report sharp pain at 512mN, the trial prick value was reported as a ceiling of 1024mN. Upon reporting sharp pain or reaching ceiling, participants rated their perceived pain intensity using a pain intensity scale (see Chapter 3.2.3.3.4). Stimuli was then reapplied systematically descending from the fixed intensity force perceived as sharp (if ceiling reached, from 512mN) until participants self-reported no percept of sharp pain through verbalisation or by displaying a “NOW” sign (recorded in mN as trial blunt value). This process was repeated five times.

3.2.3.3.2 Pressure Pain Threshold.

PPThs were measured using a SenseBox Algometer (Somedic SenseLab AB, n.d.) controlled by purpose-built software which allows delivered force to be visualised, monitored and controlled. The 1cm² probe of the algometer was applied perpendicularly to the thenar eminence of the participant’s non-dominant hand. Based on participant preference, the hand was stabilised by the PhD researcher’s non-dominant hand, a vacuum cushion, or a stable wooden table.

Using a method of levels, the applied force continually increased from 0kPa at a steady ramp rate of 50kPa/s. Participants self-reported the first moment the applied pressure became painful by pressing a button held in their adjacent hand to stop the software or displaying a “NOW” sign for algometer removal. If ramp rate increased to 1000kPa before pain was reported, the algometer was removed for safety purposes, and participants were considered to have “ceilinged out” of the trial. The on-screen value when pressure became painful, or a ceiling value of 1000kPa was recorded as the trial value (kPa). Upon reporting pain or reaching ceiling, participants rated their perceived pain intensity using a pain

intensity scale (see Chapter 3.2.3.3.4). This process was repeated three times at 30s intervals over a slightly shifted stimulation site to minimise carryover effects.

3.2.3.3.3 Cold Pain Threshold and Tolerance.

A custom CPT (Dancer Design) was used to measure CPTh and CPTol. The CPT consisted of two interconnected tanks: a water-filled stimulus tank maintained at a constant predefined temperature (7°C ±0.10°C) for participant use, and an ice water-filled reservoir tank (0°C) which facilitates the maintenance of the stimulus tank temperature. As outlined in Chapter 2.4, use of the reported methodological temperatures and durations are established as safe, and reliable for paediatric populations (Birnie et al., 2012).

Whilst standing, participants immersed their non-dominant hand into the stimulus tank water, ensuring water reached wrist level, digits were spread apart, and their hand did not touch the base or sides of the tank. The PhD researcher continually monitored participants during hand immersion and utilised a stopwatch to measure immersion time.

Participants were instructed to self-report their first percept of pain through verbalisation or display of the “NOW” sign; the time at this report was recorded as the CPTh (s). Upon the self-report, participants were instructed to maintain their hand immersion until they could no longer tolerate their pain, at which point they could remove their hand without informing the PhD researcher. If a participant had not reported pain threshold, and/or tolerance by 3 minutes they were instructed to remove their hand from the tank for safety purposes and considered to have “ceilinged out” of the trial. The time at hand removal was recorded as CPTol (s), or a ceiling value of 180s. Upon removing their hand, participants rated their perceived pain intensity using a pain intensity scale (see Chapter 3.2.3.3.4) Only one trial was conducted for both CPTh and CPTol.

3.2.3.3.4 Pain Intensity Scale.

Pain intensity scales were amended using a scale identified by the study’s PPI panel (Anonymous, n.d.) (see Appendix 5.1). The amended scale (see Appendix 5.2) comprised an

11-point NRS and VRS of differing pain intensities (0 describe as no pain, 10 described as unable to move). Both 0-10 NRS and VRS were deemed applicable given their successful use amongst CYP aged 6-17 for rating pain (Ruskin et al., 2014; Tsze et al., 2018). Use of qualitative anchors were amended alongside NRS and VRS points to provide examples of how pain intensity may interfere with daily tasks or school. Use of colours in the amended scale remained to aid in contextualisation of NRS and VRS (i.e. red worst pain) (Pope et al., 2012) and increase scale accessibility in CYP aged 11-16 (i.e. Colour Analogue Scales (CAS) (McGrath et al., 1996). All text was formatted in font Comic Sans MS due to its preference in CYP and facilitation in readability for those who are dyslexic (Bernard et al., 2002; Khan et al., 2018).

3.2.3.3.5 Pain Threshold "NOW" and Pain Tolerance "STOP" Sign.

To promote valid pain measures and prevent harm from unrecognised pain, two signs were created for use in signalling threshold, tolerance, or stimuli withdrawal amongst CYP with a preference of, or need for non-verbal communication (Shaw et al., 2022).

A "NOW" sign was created to communicate pain threshold (i.e. "I am starting to feel pain now"). The sign was blue to prevent negative emotive connotations (i.e. perceived as most intense pain) (Pope et al., 2012). Additionally, a "STOP" sign was created to communicate pain tolerance or a need for stimuli withdrawal. The sign was red to build on schemas of real world stop signs and urgency (Pravossoudovitch et al., 2014). Although availability was made clear to participants, their use was not required.

3.2.4 Procedure

3.2.4.1 Recruitment

Following ethical approval by LJMU UREC (ref: 22/PSY/077), CYP recruitment involved accessing caregivers on social media (posting recruitment poster on academic profiles, and caregiver-led groups), public engagement (presentations to CYP and caregiver

groups), and word-of-mouth (caregivers sharing the study amongst their scholastic groups and friends).

Caregivers expressed participatory interest on behalf of their CYP over email; 24 hours later, age and diagnostic appropriate PISs including contact information for support outlets and YouTube videos were sent for CYP to engage with. Caregivers were encouraged to discuss potential benefits and limitations of participating with CYP and email the PhD researcher any questions. If CYP agreed their interest, a date and time convenient for both CYP and a caregiver was arranged for participation. CYP were offered an opportunity to attend the research site prior to this allotted day for familiarisation however no one agreed.

3.2.4.2 Lab-Based Experiment

On the day of testing, study procedure was explained, discussed, and demonstrated to both CYP and their caregiver. Once understood, the researcher allowed CYP and caregivers to discuss participatory intent. If CYP expressed willingness for participation written informed consent (16+ years), or informed assent (11-15 years) with caregiver informed consent (18+ years) was obtained. However, if there were reason to suspect participatory intent was consequential to pressure by either the CYP or caregiver, the CYP's participation was declined; this did not however occur with any participants.

Upon obtaining consent participants were instructed to sit at a desk whilst their caregiver sat behind at the back of the room. Caregiver presence was deemed critical for aiding their CYP's comfortability in engaging with the study, and holding ability to withdraw their CYP's participation if they were to recognise nuanced signs of distress the PhD researcher could not (Riquelme et al., 2016; Riquelme et al., 2023). Both CYP and caregivers were reinformed of the CYP's, caregiver's and the PhD researcher's ability to stop a stimulus or withdraw from the study at any point through verbalisation or display of the "STOP" sign. An example of this case included if the CYP expressed distress or if the PhD researcher observed signs of the CYP's distress.

3.2.4.2.1 Questionnaire.

When comfortable, a demographic questionnaire was handed to the CYP for completion. They were reminded the questionnaire was not a test; they could ask their caregiver or the PhD researcher for question clarification as required, and that irrelevant (i.e. if began menstruating) or uncomfortable questions could be left blank.

3.2.4.2.2 Quantitative Sensory Testing Battery.

Upon completing the questionnaire, the outlined QST battery began in the described order (see Chapter 2.4). Throughout CYP were reminded that in this study, pain was described as an unpleasant or "not nice" feeling. Before each psychophysical assessment began as explained in Chapter 3.2.3.3, CYP were reminded they could stop at any point and asked if they were comfortable and happy to continue before the stimulus was applied. If a participant felt uncomfortable completing one threshold measure, that measure was not performed and CYP were asked their preference for study continuation, or withdrawal before the protocol continued. Upon psychophysical completion or withdrawal, CYP and their accompanied caregiver were debriefed on study purpose, data use and provided contact details of support outlets before CYP received a £20 shopping voucher as a thank you for their time.

3.2.5 Analysis

3.2.5.1 Data Processing

Consistent with DFNS (Rolke et al., 2006), raw scores for MPTh (see Chapter 3.2.3.3.1) and PPTh trials (see Chapter 3.2.3.3.2) were logarithmically transformed (log_{10}) prior to mean calculations. For analysis, MPTh was calculated as a geometric mean of participant prick and blunt trial values; and MPTh pain intensity, PPTh, and PPTh pain intensity as an arithmetic mean of their respective participant trial values. Mean values could not be calculated for CPTh, CPTol, and CPTol pain intensity thus raw scores were sufficed for use.

Missing values are reported in two participants. CPTh was not recorded in one Autistic CYP as the CYP removed their hand immediately at CPTol and thus did to not report their CPTh. Additionally, PPTh in one non-Autistic CYP was not recorded due to inability to have algometer pushed into their thenar eminence.

Moreover, during data processing, one non-Autistic CYP's data was removed due to consistently reaching ceiling values across threshold and tolerance measures and disclosing evidence of an existing extremely high pain tolerance. Removal was deemed appropriate as this was the only observed pattern of hyposensitivity in a single participant and deemed a subjective anomaly. Therefore, a total of 19 non-Autistic participants were used in analysis.

3.2.5.2 QST Data Normalisation

Originally data was normalised to Blankenburg et al.'s (2010) QST reference values which are categorised by gender and age (6-8, 9-12 and 13-16 years). However, as the outlined QST battery was adapted for inclusivity of participants, normalisation of Blankenburg et al.'s (2010) reference values where these adaptations were not implemented was considered not fit for analytic purpose in this thesis. Instead, data from the non-Autistic group were normalised and compared to the Autistic group.

Control group reference values were created for MPTh, PPTh, CPTh and CPTol. To do so, non-Autistic CYP were grouped by gender and calculated as the respective log-transformed or raw mean and SD value across each threshold or tolerance (see Table 12). Age was not considered in reference values as the limited inclusive range did not provide variative insight.

For data normalisation, Autistic CYP's data were Z-transformed respective to control group reference values by subtracting the mean reference values of the gender-matched, control group from each individual Autistic CYP's QST value and dividing by the respective control group SD value. Each Autistic CYP's threshold and tolerance data were the represented as a Z-score for analysis. The following equation expression provides clarity of this calculation:

$$Z - score = \frac{(Value_{Autistic\ Participant} - Mean_{Control\ Group})}{SD_{Control\ Group}}$$

In this normative space, a positive Z-score indicated Autistic CYP had a higher pain threshold than non-Autistic CYP, a negative Z-score that Autistic CYP had a lower pain threshold than non-Autistic CYP, and a Z-score of 0 that Autistic CYP's thresholds do not differ from non-Autistic CYP. Sensory profiles are deemed clinically significant if they exist outside of the 95% confidence interval, with Z-scores above +1.96 reflecting a hyposensitive profile, and Z-scores below -1.96 a hypersensitive profile. Z-scores are presented in analysis as a mean across Autistic CYP, and discrete of Autistic CYP; use will be clarified in presentation of values. For clarity in understanding differences between groups, untransformed mean values (SD) are reported in Table 13.

Table 12

Data Reference Values Represented as Non-Autistic CYP's Mean (SD) Values for QST Parameters in Psychophysical Assessment

Pain Measure	Male (*n* = 11)	Female (*n* = 8)
MPTh* (mN)	2.71 (0.24)	2.46 (0.09)
PPTh* (kPa)	2.45 (0.14)	2.42 (0.15)
CPTh (s)	16.26 (10.84)	14.73 (9.50)
CPTol (s)	43.06 (36.31)	29.77 (13.19)

Note. This table outlines the data references values calculated for each QST parameter from non-Autistic CYP's data. Abbreviations ordered alphabetically kPa = Kilopascal; mN = Millinewton; s = Seconds.

* Represented from $\log_{10}$ transformed data.

3.2.5.3 Statistical Analysis

All data satisfied assumptions for parametric testing: Kolmogorov-Smirnov (K-S) tests indicated normal distribution across all data, $p > .05$, with skew values of an acceptable range (±1.96). Pain intensity measures were compared between samples with Levene's Test used to ascertain homogeneity of variance amongst data. Homogeneity was assumed for PPTh and CPTol pain intensities ($p > .05$), but not MPTh ($F(26, 10.837) = 4.63$, $p = .041$). However, due to small *n* value across groups, this was not deemed to violate homogeneity, and thus parametric assumptions were satisfied.

One-sample T-tests compared if Autistic CYP's mean pain thresholds and tolerances significantly differ from non-Autistic normative values, and independent T-tests compared differences in mean pain intensity between Autistic and non-Autistic groups. Throughout each test $p > .05$ deemed non-significant results (Coolican, 2004). For the original analytic examination of threshold and tolerance results using MPTh and PPTh data normalised to Blankenburg et al. (2010) and a discussion of their context, see Appendix 6.

3.3 Results

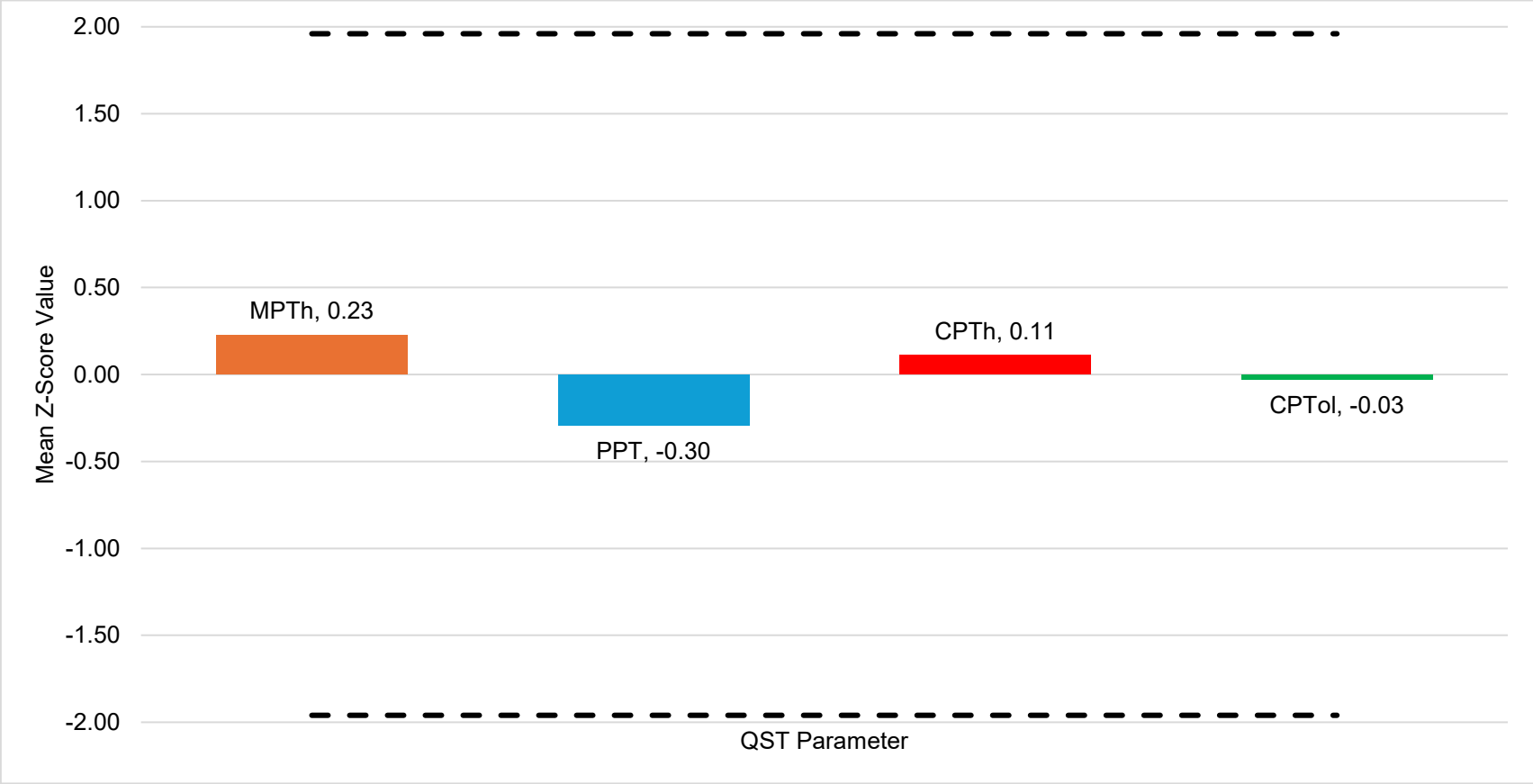
3.3.1 Grouped Autistic Participant Z-Scores Across QST

Figure 4 displays QST Z-scores for Autistic CYP, mean pain intensity values are displayed in Table 13. Compared to normative values, Autistic CYP displayed slight hyposensitivity to MPTh ($M = 0.23$, $SD = 2.00$) and CPTh ($M = 0.11$, $SD = 0.81$). However, thresholds did not significantly differ from normative values failing to support a profile of MPTh or CPTh hyposensitivity amongst Autistic CYP ($t(8) = 0.34$, $p = .741$ and $t(7) = 0.39$, $p = .708$, respectively). Similarly, marginal pain hypersensitivity in Autistic CYP was observed in PPTh ($M = -0.30$, $SD = 1.43$) and CPTol ($M = -0.03$, $SD = 0.62$), suggesting a lower force or temperature is required to detect pressure pain, and to perceive cold pain as intolerable. However again, pain thresholds and tolerance did not significantly differ from normative values failing to support a profile of PPTh or CPTol hypersensitivity ($t(8) = -0.62$, $p = .552$ and $t(8) = -0.15$, $p = .887$, respectively). Given no significant results were determined, H_0 is

accepted with the assumption that in this sample, Autistic CYP's pain thresholds do not significantly differ from non-Autistic normative values.

Figure 4

Autistic CYP's Grouped Mean Z-Score Across QST Parameters in Psychophysical Assessment



Note. Figure 4 provides a visual description of the Autistic CYP groups, grouped mean Z-score of each QST parameter. 95% confidence interval of clinical significance set to ±1.96. Bars to express group mean Z-score values for Autistic CYP. SD not reported due to small *N* value. Figure 4. provides further insight into individual data distribution.

Table 13

Group Untransformed Raw Mean (SD) Values for Pain Measures in Psychophysical Assessment

Pain Measure	Autistic CYP (*n* = 9)	Non-Autistic CYP (*n* = 19)
MPTh* (mN)	429.30 (192.11)	436.17 (212.58)
PPTh* (kPa)	275.11 (130.35)	286.44 (91.46)**
CPTh (s)	16.65 (7.98)**	15.54 (9.97)
CPTol (s)	35.57 (17.58)	36.77 (27.98)
MPTh Pain Intensity	2.60 (1.78)	2.16 (1.07)
PPTh Pain Intensity	3.63 (1.25)	3.08 (1.25)
CPTol Pain Intensity	6.11 (1.76)	4.82 (1.47)

Note. This table outlines the mean scores for each group. Rows 1 through 4 are untransformed raw mean group values for each QST parameter, and rows 5 through 8 untransformed raw mean values for each groups pain intensity measures. Abbreviations ordered alphabetically kPa = Kilopascal; mN = Millinewton; s = Seconds.

* MPTh and PPTh are not represented from $\log_{10}$ transformed data.

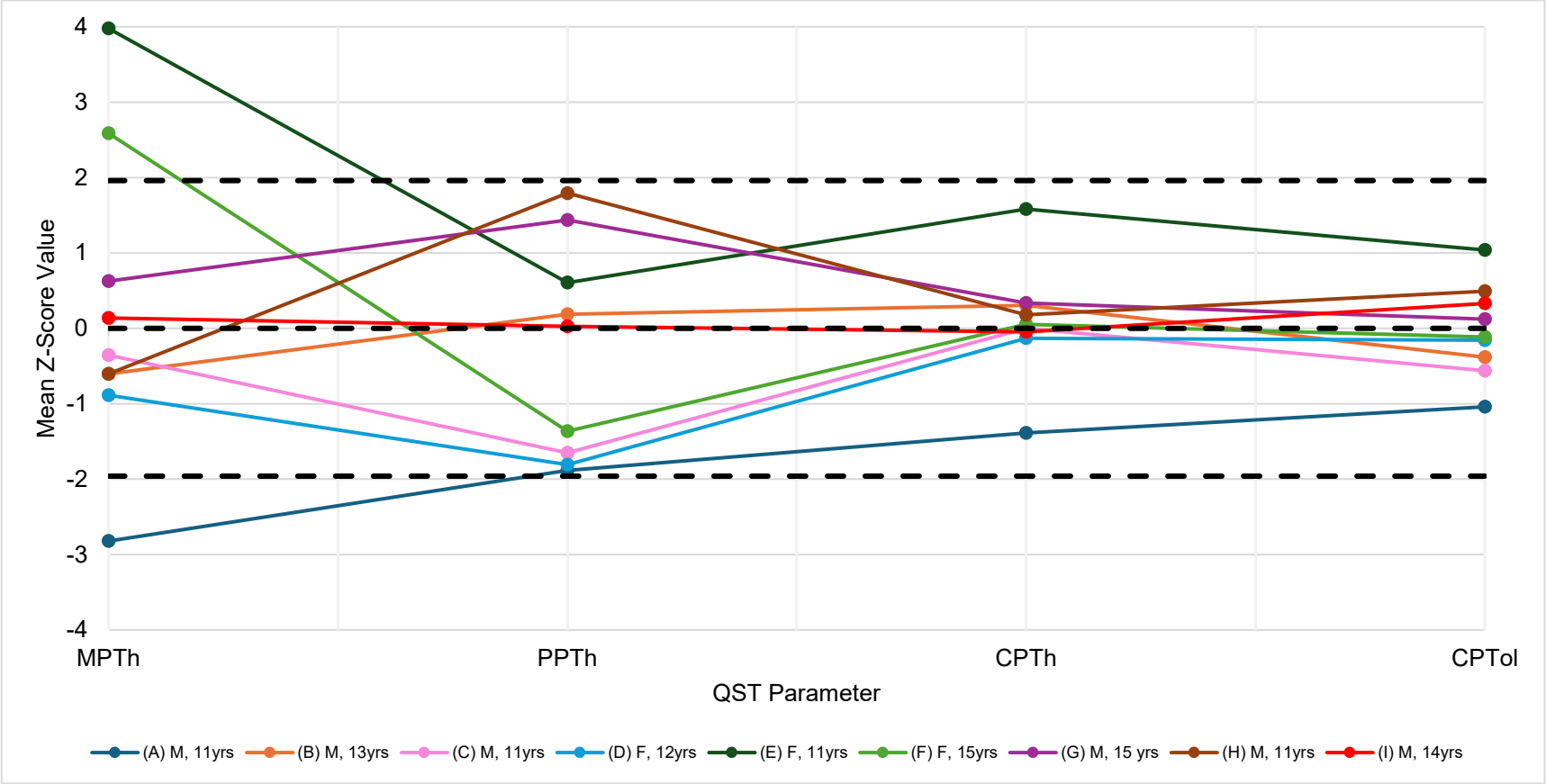
** Data excluded listwise were participant thresholds not available.

3.3.2 Discrete Autistic Participant Z-Scores Across QST

Whilst data is still descriptive, as displayed in Figure 5, some sensitivity patterns are present that might be interesting to examine in future research. Across the Autistic sample (*n* = 9), there were no clinically relevant data points. However, for MPTh for there were three sensory profiles of clinical significance highlighting the subjective nature of pain. These included two hyposensitive females (E and F), and one hypersensitive male (A). Moreover, males tend to display varied sensory profiles in their individual threshold and tolerance patterns whereas female participants D and E displayed a uniformed pattern of higher MPTh's and CPTh's than PPTh's and CPTol's. Similar patterns were observed amongst some Autistic CYP displaying hypersensitive phasic profile through a PPTh trend towards clinical significance (A, C, D, and F). Additionally, both CPTh and CPTol across participants appeared most consistently like the non-Autistic group, except for CPTh in participants A and E who trended towards hyper- and hyposensitivity respectively. However, participants A and E reflect distinct individual differences in sensitivity profiles across all QST parameters, thus emphasising pain subjectivity across Autistic CYP. Overall, a similar pattern outlined in Chapter 1.3.1 emerges with no consistent sensory profile across Autistic CYP, instead pain can be deemed a subjective experience.

Figure 5

Autistic CYP's Individual Mean Z-Score Trends Across QST Parameters in Psychophysical Assessment



Note. Figure 5 provides a visual of each Autistic CYP's individual Z-score on each QST parameter. description 95% confidence interval of clinical significance set to ±1.96. CPTh was not provided for Participant C, however for charting purposes Z-score was set to 0. SD not reported due to small *N* value.

3.3.3 Grouped Autistic Participant Pain Intensity Ratings Across QST

Mean pain intensity values are displayed in Table 13. Autistic CYP consistently rated MPTh and PPTh at a marginally higher intensity than non-Autistic CYP, however differences were non-significant ($t(26) = 0.82$, $p = .418$) and $t(25) = 0.97$, $p = .343$, respectively). A large effect consistent with Cohen's d values (Cohen, 2013) ($d = 0.83$) was observed with Autistic CYP rating cold pain more intensely ($M = 6.11$, $SD = 1.76$) than their non-Autistic peers ($M = 4.82$, $SD = 1.47$). Although differences in CPTol intensity failed to meet criteria for significance ($t(26) = 2.05$, $p = .051$), given the exploratory nature and relatively small sample size this may suggest Autistic CYP experience suprathreshold stimuli more intensely than their non-Autistic peers and future research exploring pain tolerance could be of interest. Again, no significant results were determined therefore H_0 is accepted with the assumption that in this sample Autistic CYP did not significantly rate pain more intensely than non-Autistic CYP.

3.4 Discussion

Pain hyposensitivity is delineated as a potential diagnostic feature of Autism, yet previous studies involving Autistic adults suggests no systemic differences in pain thresholds across psychophysical parameters. This study aimed to further understanding of Autistic pain sensitivity profiles from a paediatric perspective, by examining if Autistic CYP's MPTh, PPTh, CPTh, CPTol and subsequent pain intensity measures differed from non-Autistic CYP. No differences in pain threshold or tolerance were observed at group level between Autistic and non-Autistic CYP, both experienced pain thresholds, tolerance and intensity similarly at a mechanistic level. On the other hand, individual differences were apparent amongst Autistic CYP, with observed threshold and tolerance variance including hyposensitivity, hypersensitivity, or no difference comparative to non-Autistic CYP. Thus, whilst Autistic CYP pain experiences do not significantly differ from non-Autistic CYP at group level, individual differences in pain experiences should not be overlooked.

Autistic CYP do not fundamentally differ from their non-Autistic peers in how noxious stimuli are coded as painful demonstrating Autistic CYP have the neural architecture to feel pain. For example, much like in the data available for Autistic adults, Autistic CYP displayed no differences in pain thresholds or tolerance compared to their non-Autistic peers (Bird et al., 2010; Failla et al., 2020; Failla et al., 2017; Fründt et al., 2017; Thaler et al., 2017; Vaughan et al., 2020). Thus, providing no evidence for a diagnostically perceived pain hyposensitivity. Additionally, similarities with the Autistic adult data suggests developmentally at a sensory mechanistic level, Autistic individuals' experiences of pain may not differ across the lifespan. This could provide insight in understanding Autistic CYP's pain mechanisms through application of adult findings as theoretical guidance where paediatric studies are not as widely available. However, such application should not be considered as a solution to the lack of available knowledge.

Additionally, findings mirror variability observed by Vaughan et al. (2020) and Fründt et al. (2017) in Autistic adult's pain thresholds. At group level, these findings failed to support previous evidence that Autistic CYP possess a hypersensitive PPTh profile (Chen et al., 2017; Fan et al., 2013; Riquelme et al., 2016). However, when analysed as individuals,

subjectivity in these experiences is observed, particularly through variance amongst broadly across mechanical pain sensitivity profiles (MPTh and PPTh). In contrast, thermal thresholds and tolerance appeared most similar across Autistic CYP, with similar Z-scores positioned around 0 supporting thermal threshold may be similar between neurotypes. Collectively, these findings challenge the developmental similarity discussed above, that whilst Autistic adults may possess a thermal sensitive phenotype these are not observed amongst Autistic CYP (Casterman et al., 2024). Developmentally, pain mechanisms involving C-fibers such as in CPTh may become increasingly reactive as age progresses and thus may be of particular interest for future QST studies. Similarly, whilst these findings mirrored those of Vaughan et al. (2020), their observed MPTh subjectivity did not display as many outlying datapoints as these. Again, this may suggest from a developmental perspective mechanical pain sensitivity does not persist throughout adulthood. However, unlike thermal sensitivity where a mechanistic perspective may be beneficial, consideration of influential socio-affective factors such as social interaction and anxieties may provide additional relevance for mechanical thresholds. Below the presented argument suggests socio-affective factors may influence subjective pain hypersensitivity amongst Autistic CYP. However, these influential relationships could be less relevant for explaining Autistic CYP with hyposensitive profiles other than they may not have possessed an existing sensory responsiveness, phobia, or aversion to touch. Whilst it could be suggested these socio-affective factors reduce an Autistic CYP's ability to direct their attention towards a social interaction, meaning they do not need to address pain in a social context, this inference leaves further questions which future research should consider broadening understanding.

Across painful experiences in medical settings social interaction appears to be influential of pain disclosure, as Autistic CYP often experience distrust in HCPs when their pain is dismissed (Mason et al., 2019; Shaw et al., 2023). Although the PhD researcher here is not an HCP and the inclusion of a PPI panel helped to distinguish the laboratory environment from a hospital environment, it is plausible these past negative experiences of pain drive an altered top-down regulation in their appraisal of pain like Bogdonova's (2022) Model (see Chapter 1.3.1). For example, within medical settings there is typically a hierarchy of authority which the HCP holds control in how the Autistic CYP's pain is managed.

Similarly, an authoritative interaction may have been experienced during MPTh and PPTh assessments as the PhD researcher controlled the application or removal of noxious stimuli as instructed by the participant. Given the need for the PhD researcher to remove noxious stimuli, Autistic participants were required to socially disclose pain and trust the PhD researcher would act accordingly. Here these past negative pain experiences may have exacerbated a feeling of pain invalidation when social interaction is required, which may have caused Autistic CYP to report pain earlier to take back control before a negative affect was experienced. Additional inclusion of environmental and social context in influencing top-down pain appraisal is discussed in Chapter 4.4.

Moreover, Autistic CYP's affective state towards the psychophysical assessments may be influenced by pain specific anxieties like fear of pain (FoP) which are understood to increase pain sensitivity amongst both neurotypical and Autistic adults (Cimpean & David, 2019; Failla et al., 2020; Michaelides & Zis, 2019). Whilst not relevant to all Autistic CYP, needle phobias are common amongst Autistic CYP (Leyfer et al., 2006; McMurtry et al., 2015). Given the similarity between needles and pinprick equipment (MRC Systems, 2010b), some Autistic CYP may have been more fearful of this parameter before force was applied despite being informed the pinprick would not break the skin. Whilst this explanation may not provide insight into differences in pain sensitivity directly, these findings may highlight fear as a confounding variable which could be utilised in future mQST protocols to implement method of fear reduction. However, although affective state is discussed as potentially influential here, no measure of affective state was utilised within the current study. To have considered these influential factors at this stage would have complicated an understanding of how Autistic CYP experience pain, before even creating a foundation of knowledge to suggest if Autistic CYP's pain at its most mechanistic level differs from non-Autistic CYP.

Additionally, the required physical (hand support), and social interaction with the PhD researcher may have promoted pain hypersensitivity. Evidence suggest Autistic individuals who experience generally high hypersensitivity experience increased states of anxiety in response to sensory stimuli (i.e. touch) and avoid interpersonal touch due to its social nature (Green & Ben-Sasson, 2010; Henderson, 2022; Ujiie & Takahashi, 2022). Therefore, it could be theorised Autistic CYP with PPTh hypersensitivity may have

experienced high levels of anxiety from both noxious, and touch stimuli which promoted a trend towards clinical significance. On the other hand, low PPTh's could reflect touch avoidance which Autistic CYP reported feeling pain earlier to stop a social interaction as suggested by Henderson (2022). Or the level of distress caused by this social interaction promoted anxiety which effected pain intensity, much like Failla et al. (2020) suggested in Autistic adults. Here, a measure of Autistic CYP's sensory sensitivity, state anxiety, FoP and attitudes towards touch may provide understanding as to how these factors can impact direction in sensitivity profiles.

Psychophysical studies involving Autistic CYP typically recruit 20-40 participants (Chen et al., 2017; Duerden et al., 2015; Fan et al., 2013; Riquelme et al., 2016). However, whilst the non-Autistic sample broached this (total recruited, *n* = 20, data inclusion, *n* = 19), the Autistic sample did not (*n* = 9) which suggests sample size may be a large study limitation. For example, given the low sample of Autistic CYP, the ability to generalise these findings to the broader Autistic population are reduced. This reduction arguably is even more relevant when considering the participating Autistic CYP may have been considered as having low support needs (although this cannot be presumed as fact given this question was not asked). Here, the low Autistic sample size, and representation of a low support needs population may limit the ability to confidently argue Autistic CYP experience pain on a similar sensory and intensity level as non-Autistic CYP. Rather, this subgroup of Autistic CYP experience pain on a similar sensory and intensity level as non-Autistic CYP. As ever, further research could be required to strengthen this argument through a higher sample size.

There are a multitude of additional reasons to consider in not attaining a large Autistic sample, most prevalent here is ability to recruit. Both Autistic individuals, and CYP are considered "hard-to-reach" groups with gatekeeper engagement acting in this case as a barrier to accessing participants (Gowen et al., 2019; Poppe & Abela, 2023). Although gatekeepers are paramount amongst paediatric populations, additional attempts to build rapport with schools and Autism-based group to access gatekeepers were required. Even with the PhD researcher's best efforts to build rapport and engage with existing working relationships, many gatekeepers would not share the study even with efforts to promote understanding like providing a video of the study. This suggests when trying to engage

gatekeepers creating recruitment information that is catered and accessible to them for study understanding may be just as vital as it is for Autistic CYP.

These findings demonstrate Autistic CYP can tolerate pain in laboratory settings when adaptations are amenable and considerate of Autistic CYP's accessibility needs and research expectations. Here, researchers should modify this protocol for future research use in the aims of furthering knowledge of Autistic CYP's pain which Chapter 2 emphasised is severely lacking. However, despite providing study modifications (see Chapter 3.2) which a PPI panel suggested would assist in CYP recruitment, these did not assist in reaching target sample. In hindsight a survey or brief interview at the end of the study could have been utilised to understand what influenced decisions to participate, what modifications were useful, and how the CYP would improve these modifications for future use. However, even still, findings would be biased towards those willing to participate and would not have provided insight into those who decline. Here an additional survey that can be appended with recruitment materials to identify what deters participation may improve future participant retention.

Moreover, researchers should consider experience of CPTol in CYP from both an Autistic, and non-Autistic perspective as the current findings highlighted two interesting patterns within, and between each population. The first interesting pattern emerges when considering reference values for non-Autistic CYP (see Table 12), as non-Autistic males displayed a higher CPTol than their non-Autistic female peers. As the only measure in the current study of tolerance, this could imply sex differences have a larger influence on pain tolerance than pain threshold. However, because menstruation was the only sex difference recorded in the current study, there is little evidence to support this suggestion. Instead, future research should consider the role of differing sex factors in influencing pain tolerance; an insight that could contribute to how psychophysical studies can be adapted for participant's sex as discussed in Chapter 2.4. The second interesting pattern emerges when considering Autistic CYP's CPTol pain intensity rating (see Table 13), as although not significant, Autistic CYP rated CPTol pain more intensely than their non-Autistic peers. Here, pain tolerance could have contributed to a heightened emotional state in a population known to experience co-occurring anxiety at baseline, in turn heightening their pain intensity (Failla

et al., 2020). However as already discussed, affective state was not considered in the current study, therefore these suggestions remain theoretical. Future studies should explore differences in CPTol pain intensity between Autistic and non-Autistic populations to aid in understanding the pain appraisal pathways of the Autistic population.

Although differences in CPTol intensity failed to meet criteria for significance ($t(26) = 2.05$, $p = .051$), given the exploratory nature and relatively small sample size this may suggest Autistic CYP experience suprathreshold stimuli more intensely than their non-Autistic peers and future research exploring pain tolerance could be of interest

To conclude, findings suggest Autistic CYP experience pain on a similar sensory and intensity level as non-Autistic CYP. However, whilst subjectivity in pain experiences were clear, no measures were implemented to understand factors that drive such subjectivities. Chapter 4 will qualitatively consider factors that may shape subjective pain experiences to begin to understand why pain is overrepresented amongst the Autistic population despite sensory experiences being similar across these paediatric neurotypes.

Chapter 4.

An Autistic Experience of Pain: Qualitative Understanding of Autistic Children and Young People, and their Maternal Caregiver's Perspectives

4 An Autistic Experience of Pain: Qualitative Understanding of Autistic Children and Young People and their Maternal Caregiver's Perspectives

4.1 Introduction

Chapter 3 provided an objective investigation of differences in pain thresholds, tolerance, and intensity measures between Autistic and non-Autistic CYP. No significant differences were found to support beliefs that Autistic CYP experience pain hypo- or hypersensitivity, which lends the question: *if pain is not experienced differently on a sensory level, then why is pain overrepresented within Autistic CYP?* As outlined in Chapter 1.2.2 pain is a multifaceted experience, which affective states, cognitive evaluation, and social context are consistently evidenced as important factors in shaping non-Autistic adults and CYP's pain (Craig, 2015). As the role of these factors is less understood amongst Autistic CYP, consideration of how these psychological and social factors shape Autistic CYP's pain may provide experiential understanding for deciphering this pain overrepresentation.

Experiences of anxiety and depression are shown to increase pain sensitivity amongst non-Autistic individuals (Beesdo et al., 2010; Means-Christensen et al., 2008). Similarly, this moderating relationship is observed amongst Autistic adults and CYP, with qualitative and survey-based evidence reporting heightened negative affective states increase Autistic individuals pain sensitivity (Kalingel-Levi et al., 2022; Mazurek et al., 2013; Williams et al., 2015). As Autistic CYP experience frequent and intense co-occurring negative affective states (i.e. anxiety and depression), the facilitatory nature of affect may be strengthened to exacerbate a hypersensitive profile (Mingins et al., 2020; Stewart et al., 2022). However, Hazen et al. (2014) suggests this causal relationship is less clear: *does anxiety promote pain hypersensitivity as suggested above, or does pain hypersensitivity promote anxiety?* Pain hypersensitivity certainly can promote negative affect, including a predisposition to pain-related anxiety and fear (Carpenter et al., 2019; Failla et al., 2020; Syu et al., 2020; Tsao et al., 2006). However, how social context influences behavioural avoidance of these anxious states may contribute to pain overrepresentation more so than anxiety itself (Babalola et al., 2024). For example, Autistic individuals who report difficulties in processing hospital environments and feel misunderstood by HCPs are less likely to

attend preventative-based hospital appointments to mitigate health-related anxieties (Babalola et al., 2024; Doherty et al., 2020; Nicolaidis et al., 2013; Strömberg et al., 2022). Recent studies have suggested adaptations to overcome healthcare barriers and manage health-related anxieties to reduce this worsened health burden (Doherty et al., 2022; Haydon et al., 2021). Still, additional knowledge of how social factors influence affect in the context of pain may provide unique insight into Autistic CYP's experiences.

Interruption, interference and identity are three important cognitive processes for pain evaluation (Morley, 2008). Pain's interruptive has capacity to capture attention and promote analgesic behaviour, however this focus consequently interferes with an ability to engage with, and perform other tasks (Van Damme & Moore, 2012). Arguably to facilitate pain's interruptive capacity, CYP need to interoceptively attend to their internal cues and recognise a sensation of pain (Garfinkel et al., 2015; Murphy et al., 2019). Yet review-based evidence presented by Dubois et al. (2016) leans towards Autistic CYP experiencing hyposensitivity in pain interoception, meaning Autistic CYP may be less susceptible to recognising pain and experiencing its interruptive effects (Bogdanova et al., 2022; Trevisan et al., 2021). Without these attentional shifts, Autistic CYP may not appraise the threatening nature of a stimuli omitting a need to seek relief, theoretically ascribing pain interruption as a facilitator to Autistic pain progression. On the other hand, Autistic interoceptive abilities are subjective (Garfinkel et al., 2016); some Autistic CYP experience opposing difficulties in shifting their attention away from pain which exacerbates the implications of pain interference (Duerden et al., 2015; Lipsker et al., 2021). The consequences of pain interference are most apparent and pronounced in scholastic settings where CYP spend most of their time (Logan et al., 2008). However as engaging with education can already be cognitively demanding for Autistic CYP, less energy may be available to manage such interfering effects (Donaghy et al., 2023; Horgan et al., 2023). Autistic CYP recognise the latter difficulties in their pain experiences, with disparities between peers contributing to their sense of self. For example, qualitative evidence from Jordan et al. (2024) shows Autistic CYP experiencing chronic pain recognise their pain experience differs from non-Autistic peers, and report this reduces their ability to engage with pain treatment. Although these interactions between interference and identity may not contribute to a sensitivity profile,

perceived effects of reduced engagement with treatment may contribute to pain overrepresentation. Additionally, Jordan et al. (2024) further suggests social appraisal of Autistic pain behaviours reinforce this sense of self with interpretative incongruence promoting perceived treatment barriers, which additionally emphasises how social factors continue to influence access to care is important.

Bystander's observational interpretation of CYP's pain behaviours (i.e. crying) can socially appraise pain severity and provide understanding of the adequate method of relief (Akbari et al., 2020; Craig, 2015). However behavioural displays deemed as gold standard in pain identification (i.e. FLACC and NCCPC-R) are based on neurotypical understanding, failing to acknowledge neurodivergent CYP's differential displays – particularly how behavioural expression may hold different communicative purpose (Noyek et al., 2023). Allely's (2013) literature review suggests Autistic CYP experience a need for higher pain intensities before reactivity is shown, alluding Autistic behavioural expression and use of behavioural measures may be most useful in understanding subjectively high pain intensity. de Knegt et al.'s (2013) systematic review supports this perspective ascribing increasing pain intensity as most influential in predicting CYP with an ID's (including Autistic) behavioural displays of pain. If true, methods to improve bystander behavioural interpretations and accurate use of pain measures may intersect pain overrepresentation allowing knowledge that Autistic pain behaviours indicate an immediate need for relief, rather than a portrayal of severity progression. However, upon further reflection of Chapter 3.3.3 findings, Autistic CYP may rate pain at higher intensities than non-Autistic CYP. Therefore, if behavioural expressions are indicative of such intensity, Autistic CYP should consistently show heightened reactivity for bystander interpretation, yet evidence does not reflect this (Courtemanche et al., 2016; Daughters et al., 2007; Militerni et al., 2000; Tordjman et al., 2009). Instead, additional social context may influence behavioural expression which must be understood to aid in improving observational interpretation of Autistic CYP's pain.

Caregivers are vital in managing CYP's pain (see Wong and Baker (1988)), yet caregiver involvement in decoding Autistic CYP's pain behaviours and actioning relief may be increased. To lessen bystander difficulties in decoding pain, caregivers often scaffold

Autistic CYP's pain management by informing others of what their pain displays mean, communicating their current and past pain experiences, and advocating for access to relief (Ely et al., 2016; Jordan et al., 2024; Lindly et al., 2017). Yet Noyek's (2023) review further highlights discrepancies in self- vs. observational reports of pain, with caregivers both overreporting, and underreporting pain intensity. Whilst this could discredit the validity of caregiver perspective in understanding Autistic CYP's pain, caregivers typically observe their authentic pain behaviours in settings (i.e. home) where external social pressures and anxieties to mask are mitigated (Chapman et al., 2022). Thus, caregivers of Autistic CYP are arguably best placed in interpreting pain, given their knowledge of nuanced patterns of pain expression, and what they represent (Jordan et al., 2024; Kalingel-Levi et al., 2022). Additionally, caregiver's role in social action may be emphasised by their active awareness of when and what relief their Autistic CYP requires. For example, Autistic CYP use typical methods of pain relief, i.e. medication and distraction, yet caregivers provide routine in accessing relief and facilitate acquiring items aligned with sensory needs or focused areas of interest (Dobson et al., 2023; Dubois et al., 2017; Jordan et al., 2024). Thus, their perspectives of how to socially facilitate relief may allow understanding of often overlooked methods and provide adequate and person-centred pain management that could reduce this pain overrepresentation. Whilst caregivers undeniably hold great social relevance here, gaining knowledge of how Autistic CYP's pain is understood in differing social contexts (i.e. school and hospital) is paramount. Doing so will identify and address barriers to pain expression and management which this introduction has suggested may promote overrepresentation of pain.

Recent literature has explored the relevance of cognition, affect and social context in Autistic CYP's broad experiences of pain. For example, qualitative evidence from Dobson et al. (2023) and Jordan et al. (2024) suggests whilst unique in nature, these psychological and social factors drive Autistic pain experiences. Particularly their findings highlight anxiety and caregiver relationships hold large importance in moderating pain intensity and management. However given their focus on chronic pain and medical procedure, their application may be limited to a medical arena. Thus, the current study aimed to qualitatively understand how emotional, cognitive, behavioural, and social factors influence Autistic

CYP's experiences of pain, with reference to their own previous experiences of pain. In-depth knowledge was obtained through separate qualitative interviews with Autistic CYP and their primary caregiver providing a self-reported and observational experiential view. With the exploratory nature of this study no hypotheses were determined, instead objectives to explore the following were outlined:

1) The importance of emotional, cognitive, behavioural and relationship factors in Autistic CYP experiences of pain.
2) Comparisons and contrasts in experiential perspectives between Autistic CYP and their caregiver.

4.2 Method

4.2.1 Design

Using cross-sectional semi-structured interviews, an inductive qualitative research design was implemented to provide in-depth understanding of Autistic CYP's pain experiences from both their own and their caregiver's perspective. In doing so, Smith et al.'s (1996) Interpretative Phenomenological Analysis (IPA) was adhered to as an analytic approach (see Chapter 4.2.8 for reasoning).

4.2.1.1 Design Validation

A PPI panel (see Chapter 0.1.2) was consulted to assess applicability, validity and inclusivity of study design and materials. All recommendations for improvement were implemented prior to study commencement (i.e., non-ambiguous questions, poster colour scheme providing readability).

4.2.2 Participants

From December 2021 to July 2022 a snowball sample of eleven child-caregiver dyads were recruited via word of mouth, and social media. As one Autistic CYP withdrew at

interview, ten female dyads were included in analysis (Autistic CYP: *M* = 12.50, *SD* = 1.58; Caregiver: *M* = 41.80, *SD* = 6.96).

4.2.2.1 Inclusion Criteria

Autistic CYP eligible for participation were: (1) formally diagnosed Autistic by an HCP and (2) aged 11-16 years for similar comprehension and understanding of questions involving 'why' (Visser-Bochane et al., 2020). Caregivers eligible for participation were: (1) one caregiver of a participating Autistic CYP and (2) aged 18+ years to provide Autistic CYP's consent.

4.2.3 Materials

4.2.3.1 Recruitment Materials

4.2.3.1.1 Social Media Poster.

Caregiver recruitment was targeted using social media posters (see Appendix 7). Posters provided inclusion criteria, methodology and contact details to express dyadic participatory interest.

4.2.3.1.2 YouTube Video.

A video hosted through a private YouTube link was created to support PIS and reduce anxiety from interactive uncertainty (Gowen et al., 2019). During the video, the PhD researcher orally introduced themselves and provided study explanation with subtitles and animations (see Appendix 8).

4.2.3.2 Interview Materials

4.2.3.2.1 Questionnaire.

A bespoke questionnaire was hosted by Qualtrics (2021) and consisted of 2 participant-focused blocks: Autistic CYP block (see Appendix 9.1), and caregiver block (see

Appendix 9.2). The purpose of the questionnaire was to collect key demographics (i.e., age and identifying gender) and information for interview utilisation (i.e., Autistic CYP identifying a recent pain scenario for interview context, and dyads providing interview adaptations like cameras off). Identifying information for interview utilisation was crucial for improving research inclusivity, accessibility, and engagement by minimising distressing feelings from, for example, interacting with someone new (Gowen et al., 2019; Neilson & Bond, 2023).

4.2.3.2.2 Interview Schedule.

An interview schedule (see Appendix 10.1 for CYP and 10.2 for caregiver's full interview schedule) was created using a cognitive-behavioural approach to incorporate pain-related thoughts, feelings, and behaviours. The schedule outlined 15 questions for Autistic CYP, and 16 for their caregiver. These questions addressed psychological and social factors that affect pain experiences (see Table 14) in the context of the participant's provided pain scenario. If a scenario was not provided or no longer relatable, 5 likely pain scenarios were identified as common experiences where a CYP may experience pain (see Appendix 11) (McGrath et al., 1994). Given no CYP required their use, their use within the interview schedule is not reported.

Table 14

Topic of Interest and Key Points for Both CYP and Caregiver Interview Questions

Topic of Interest	Key Points to Address
Pain Importance	Pain frequency
	Interoceptive identification
Pain Description	Pain description
	Recognising pain quality, location, and duration
Pain Behaviours	Helpful pain behaviours
	Unhelpful pain behaviours
	Pain disclosure
Cognitive Pain States	Thoughts about pain
	Attention differences (in pain vs. not in pain)
	Memory differences (in pain vs. not in pain)
	Communicative processing differences (in pain vs. not in pain)
Emotional Pain States	Emotional recognition
	Emotional expression

Note. This table outlines the topics discussed in each interview, and the subsequent key points that were addressed.

4.2.4 Procedure

Following ethical approval (LJMU UREC ref: 21/PSY/034), caregivers were recruited through word-of-mouth and social media (i.e. academic X profile, and Autism-focused, or Autistic-led Facebook groups). Caregivers assessed dyadic eligibility before contacting the PhD researcher via email to express interest; 24 hours later, age-appropriate PISs including contact information for support outlets, and accompanying YouTube video were sent. Dyads were encouraged to discuss potential participatory benefits and limitations and asking the PhD researcher questions before recontacting to complete initial assent and/or consent forms (see Chapter 3.2.4.2). If there were reason to suspect participatory intent was not dyadically mutual, neither were eligible for participation.

Once assent and/or consent was obtained, the questionnaire link was emailed to caregivers 24-hours later. Ensuring continued willingness to participate, dyads were informed questionnaire completion satisfied assent and/or consent for this portion of the study and exiting the questionnaire constituted data withdrawal. Dyads completed their relevant questionnaire block each within 30 minutes – during which, caregivers were encouraged to support Autistic CYP in comprehending and answering questions when requested. One week after completion, semi-structured online interviews were conducted and recorded audibly via Microsoft Teams. Upon obtaining verbal assent and/or consent, dyads were interviewed consecutively, Autistic CYP were consistently interviewed first with the option for caregiver presence. Nine Autistic CYP chose to interview with their caregiver present, and one Autistic CYP aged 16 years chose to interview alone. During the interview requested adaptations were implemented and participants were reminded of their ability to take breaks from, stop, or withdraw. On average, Autistic CYP's interviews lasted 33 minutes and 20 seconds, and caregiver's 42 minutes and 20 seconds.

Upon interview completion or withdrawal, participants were debriefed by the PhD researcher on study purpose, data use and provided contact details of support outlets before each receiving a £20 shopping voucher as a thank you for their time.

4.2.5 Descriptive Analysis

Questionnaire data was exported from Qualtrics (2021) and imported into Microsoft Excel for data cleaning. Once cleaned, data was imported into SPSS v27.0 (IBM Corp, 2020) for descriptive analysis.

4.2.6 Qualitative Analysis

4.2.6.1 Data Processing

Using Microsoft Word, the PhD researcher created verbatim transcripts for each interview from their audio recordings. Personal data was removed from transcripts for anonymity with quality-assurance provided by the primary supervisor.

4.2.6.2 Interpretative Phenomenological Analysis

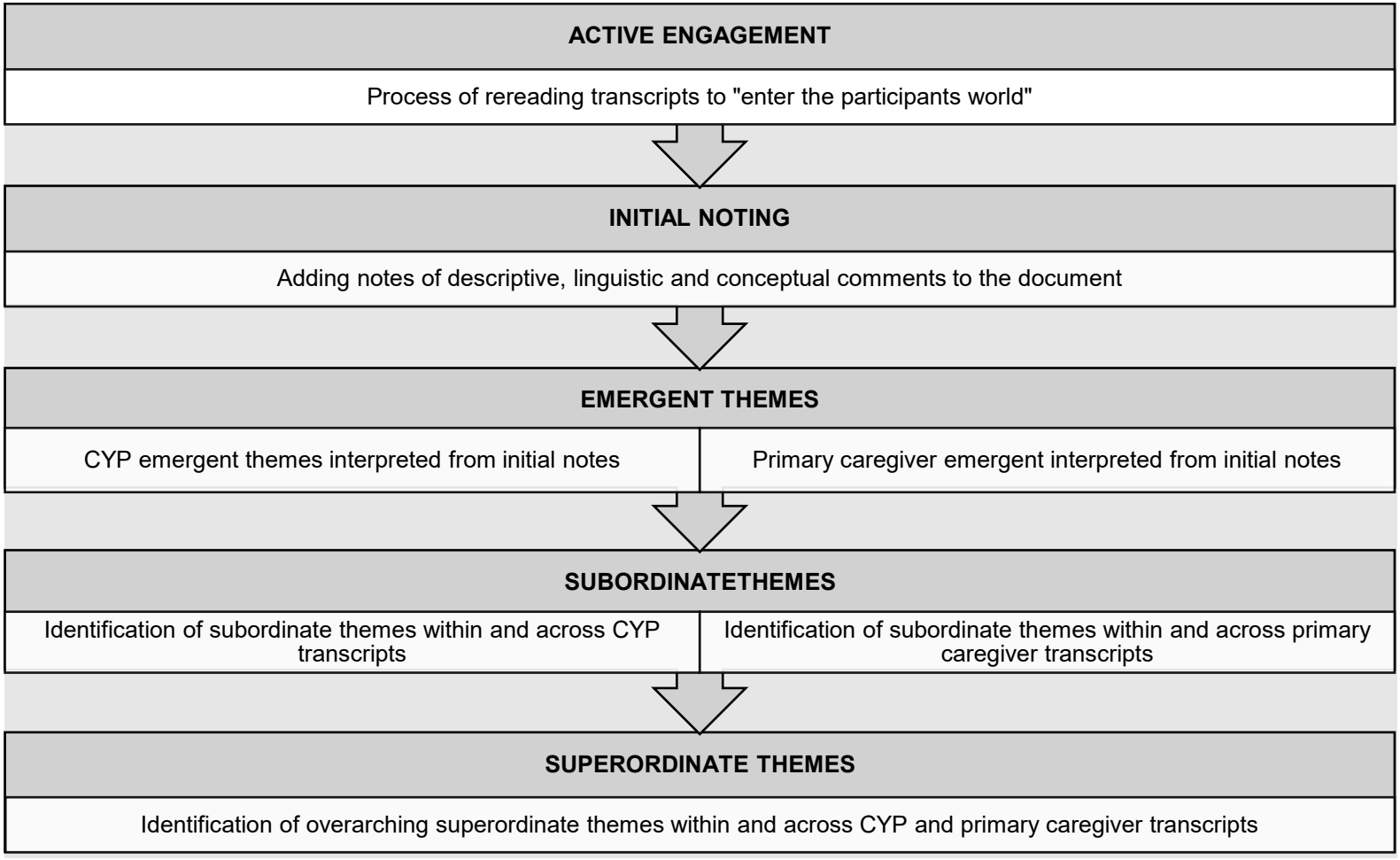
Transcripts were analysed using IPA (Smith, 1996): a form of qualitative analysis used in Autistic CYP, pain and wider health research to provide an inductive perspective of experiential meanings and bodily feelings (Rasmussen & Pagsberg, 2019; Smith & Osborn, 2015; Willig & Stainton Rogers, 2017).

Smith et al.'s (2009, p.79-107) analytic outline was followed (see Figure 6). First the PhD researcher actively engaged with and reread physical copies of transcripts to "enter the participants world" whilst simultaneously noting descriptive, linguistic, and conceptual comments (see Appendix 12). The PhD researcher interpreted what these comments contributed to pain experiences to create emergent themes (see Appendix 13). Using emergent themes, subordinate themes within and across transcripts (see Appendix 14). As dyads participated, two transcripts corresponding to either the CYP or their caregiver were present. In the latter analytic approach, all Autistic CYP's transcripts were analysed first followed by all caregiver transcripts. Initial subordinate themes were identified across each group separately and converged into superordinate themes to identify similarities and differences in experiential perspectives. Although it was feasible to compare and contrast within dyads, this order was implemented to place Autistic CYP's voices at the forefront of interpretations and understand their experience as a subset of the Autistic population, with

caregiver insights providing additional experiential insight. Developed themes are presented within the results as interrelated superordinate themes and discussion of polarising perspectives within dyads.

Figure 6

Outline of IPA Steps Followed for Interview Analysis



Note. Figure 6 provides a visual description of the methodical IPA steps taken in Chapter 4's analysis.

4.2.7 Research Quality

Smith et al. (2009, p.180-183) outlined how IPA meets Yardley's (2000) 4 criteria for assessing quality in qualitative research. All criteria were considered and followed with approaches discussed below.

4.2.7.1 Sensitivity of Context

Prior to conducting interviews, the PhD researcher had theoretical awareness of qualitative approaches before identifying inclusive adaptations. Derived from academic literature, grey literature and lived experience, inclusive approaches emphasised importance of Autism language and surrounding stigma from a sociocultural context. In response, language use was adapted to participant preference (i.e. IFL) and appreciation for the interactional nature with methods to put participants at ease when discussing sensitive topics (i.e. YouTube video, interview adaptations).

4.2.7.2 Commitment and Rigour

Demonstrating commitment, the PhD researcher thoroughly engaged with each transcript to correctly interpret their words and identify underlying meaning. The PhD researcher's supervisor, Professor Helen Poole (an expert in qualitative research), helped provide rigour by auditing 4 annotated transcripts to identify potential analytic biases and assess overall interpretation. Rigour was ensured through supervisory discussions of superordinate themes and their formation. The PPI panel further assessed validity by discussing if themes aligned with their own experiences and expectations of the wider Autistic community – which they agreed.

4.2.7.3 Transparency and Coherence

Study design involved frequent supervisory meetings to evaluate study materials and justification; decisions were noted and embedded within chapter methods to highlight methodological transparency. Particularly coherence was considered when writing qualitative

results; supervisors suggested the PhD researcher recorded theme explanations to create flow in explaining how they encompass pain experiences, whilst still being mindful of IPA's hermeneutic phenomenology in representing an individual's voice.

4.2.7.4 Impact and Importance

As an Autistic person, the PhD researcher provided unique ability in applying a neurodivergent analytical perspective to a neurodivergent issue. Additionally, the PhD researcher and lead supervisor understood research impact and importance from their Autistic community outreach with PPI panels, which provided further understanding of how research can be applied to improve, for example, medical knowledge of Autistic CYP's pain experiences.

4.2.8 Reflexivity

As an Autistic female, the PhD researcher reflected on how their data interpretations may be biased by their personal experiences before choosing IPA (Smith, 1996) as the most appropriate analytic approach. This decision was based on IPA's use of descriptive interpretation which lent well to the emotional nature embedded throughout pain.

Throughout data collection the PhD researcher jotted interpretative thoughts, interview reflections, and noted behavioural factors that may impact data (i.e., participant often talking to someone off camera). An exemplar reflection which has been highlighted in Chapter 0.3 includes the PhD researcher's empathic nature towards Autistic CYP experiences based on how they mirrored their own and a want to express that Autistic CYP feelings were valid and "understood". However, the PhD researcher consistently refrained from commenting to prevent biased responses but felt guilt as in doing so they may have prevented a CYP feeling "normal" in their experiences.

Additional recognised influences in data interpretation includes supervisory input from H.P. (a Professor of Applied Health Psychology with over 20 years-experience in pain research), B.R. (a Clinical Psychologist with a background in providing Autism assessments), M.F. (a neurodivergent Assistant Professor with 9 years-experience in Autism

and pain research), and D.M. (a neurodivergent Associate Professor with 15 years-experience in Autism and pain research; diagnosed dyslexic and self-identified ADHD). Here a mixture of neurodivergent vs. neurotypical perspectives and clinical vs. research perspectives were provided in discussing the findings of this study. Whilst B.D. provided the initial analytic viewpoint, contributions from the wider supervisory team influenced the reported results to provide context in an array of settings.

4.3 Results

4.3.1 Descriptive Analysis

Co-occurring conditions were common amongst dyads, with both Autistic CYP (*n* = 5) and their caregivers (now described as maternal caregivers) (*n* = 7) reporting physical (see Table 15), mental health or neurodivergent conditions (see Table 16). Most maternal caregivers reported disclosing their CYP's Autism diagnosis at medical appointments (*n* = 7). All participants identified a pain scenario for interview context (see Table 17), and some provided adaptative requests (see Table 18).

Table 15

Frequency of Co-Occurring Physical Conditions in Autistic CYP and Their Maternal Caregiver

Condition Name	Autistic CYP	Maternal Caregiver
EDS	1[a]	2[a]
Scoliosis	1[b]	-
Sciatica	1[b]	-
Migraine	1[b]	-
Back Pain	1[b]	-
Ear Pain	1[b]	-
Joint Pain	2[b]	1[a]
Dislocations/Subluxations	-	1[a]
Hyperthyroidism	-	1[a]
IBS	-	1[a]
Interstitial Cystitis	-	1[a]
Lumbar Spondylosis	-	1[a]
ME/CFS	-	1[a]
Scheuermann's Disease	-	1[a]
Unspecified Allergies	-	1[a]

Notes. This table displays the frequency of co-occurring physical conditions disclosed within each participating group. Ordered by Frequency of Physical Condition(s) in Autistic CYP. Values do not distinguish diagnoses from experiential. Values do not equate to number of participants given

the ability for each participant to experience multiple physical conditions. Abbreviations ordered alphabetically EDS = Ehlers-Danlos Syndrome; IBS = Irritable Bowel Syndrome; ME/CFS = Myalgic Encephalomyelitis/Chronic Fatigue Syndrome.

[a] Specified as a chronic diagnosis.

[b] Specified as recurring pain.

Table 16

Frequency of Co-Occurring Mental Health or Neurodivergent Conditions in Autistic CYP and Their Maternal Caregiver

Condition Name	Autistic CYP	Maternal Caregiver
Autism	10	7[a]
Anxiety	3	3
Depression	2	2[b]
ADHD	2[c]	2[d]
Dyslexia	-	1
OCD	-	1

Notes. This table displays the frequency of co-occurring mental health or neurodivergent conditions disclosed within each participating group. Ordered by Frequency of Mental Health or Neurodivergent Condition(s) in Autistic CYP. Values do not distinguish from clinical and self-reported diagnosis. Autism not included given inclusion criteria. Abbreviations ordered alphabetically ADHD = Attention-Deficit Hyperactivity Disorder; OCD = Obsessive Compulsive Disorder.

[a] Includes 6 participants with a suspected diagnosis.

[b] Includes 1 participant who specified low mood.

[c] Includes 1 participant identified as on the waiting list for diagnosis.

[d] Includes 1 participant identified as on the waiting list for diagnosis.

Table 17

Frequency of Pain Scenarios Identified for Interview Context by Autistic CYP

Pain Description Provided	Frequency
Migraine/Headache	4
Sports Injury	2
Menstrual Pain	2
Abdominal Migraine	1
Bacterial Infection	1
Stubbed Toe	1
Leg Pain	1
Spinal Injury	1

Note. This table outlines the frequency of pain scenario topics identified by Autistic CYP. Ordered by “Frequency of Pain Description Provided”. Total frequency exceeds *n* = 10 as some Autistic CYP provided more than 1 scenario.

Table 18

Interview Adjustments as Expressed by Participating CYP and Their Maternal Caregiver

Expressed by CYP on Behalf of	**Expressed by Maternal Caregiver on Behalf of**	
Self (*n* = 4)	**CYP (*n* = 6)[a]**	**Self (*n* = 2)**
No Cameras (*n* = 3)	No Cameras (*n* = 2)	No Cameras (*n* = 1)
Use of Fidget Toy (*n* = 1)	Use of Breaks (*n* = 2)	Recorded Footage Use (*n* = 1)
Use of Chat Function (*n* = 1)[b]	Use of Audio (*n* = 1)	
	May Look away (*n* = 1)	
	Topics to Avoid (*n* = 1)	
	Option to Type (*n* = 1)	

Notes. This table describes the interview adjustments requested by each participating group. Total adjustments exceed *n* value due to more than one adjustment request

[a] Adjustments expressed by maternal caregiver on behalf of CYP total exceeds n value as select maternal caregiver expressed more than one adjustment.

[b] Determined during interview.

4.3.2 Qualitative Analysis

Four superordinate themes were constructed to encapsulate Autistic CYP's pain experiences (see Table 19). An experiential narrative was provided to outline how pain is recognised ("*Making Sense of a Feeling"),* experienced (*"How Pain is Experienced"*), expressed or disclosed (*"Role of Relationships"*), and subsequently relieved (*"Playing an Active Role in Pain Management"*). Superordinate themes are summarised below with anonymised quotations from Autistic CYP's (e.g. A1), and maternal caregiver's (e.g. M2) to exemplify analytic points.

Table 19

Superordinate Themes and Their Subsumed Subordinate Themes for CYP and Maternal Caregivers

Superordinate Theme	Subordinate Theme
Making Sense of a Feeling	Using the External to Understand the Internal
	Recognising the Changes in Feeling
	Putting Words to the Feeling
How Pain is Experienced	The Emotional Threshold of Pain
	Self-Awareness of Cognitive Differences
	Knowing How and When to “Get on With It”
The Role of Relationships	The Importance of the Primary Caregiver
	Building the Foundation for Pain Disclosure
	Creating Barriers for Pain Disclosure
Playing an Active Role in Pain Management	Knowledgeable of One’s Own Needs in Pain
	Paying Attention to the Importance of Cognition
	What Relief is Available?
	How to Choose the Most Effective Method of Relief

Note. This table displays the superordinate and subsequent subordinate themes identified across transcripts.

4.3.2.1 Making Sense of a Feeling

Autistic CYP reported using internal and environmental cues to interoceptively attend to their pain. Internal cues were driven by a change in internal state, however their use in pain recognition was limited only to pain of higher intensities as A10 explains, *"I usually notice it [pain] when it gets really bad…"*. By contrast, environmental cues provided understanding of a potential cause where a perceived search for an interoceptive cause allows pain recognition. Understanding from environmental cues can be a product of interactions between the body and space, "*the nurses were touching me in the hospital and then I could feel it [pain] then"* (A1), or visual cues, "*I can like cut myself or scratch myself and I won't even notice and then when I actually look at it, that's when I feel the pain"* (A8).

Maternal caregivers described how they socially facilitate their CYP's pain recognition through communicative prompts. Upon observing a change in their CYP's demeanour or behaviour, i.e. their "*reaction to other people around"* (M9), maternal caregivers ask direct questions which allow Autistic CYP to process whether changes are consequential to unrecognised pain. Closed questions were described as most useful in gaining pain recognition as demonstrated by M10 whose child would be *"more actively able to answer"* as opposed to when an individual asks "*right well, tell me what's going on?".* Additionally, some questions required comparative experiential context as exemplified by M5 who describes they need to use previous experiential context to identify a type of pain sensation, "*'Is it like when you felt sick on the coach or is it like last week when your temperature was really high, and you felt really grotty?".* Communication prompts appeared to have retrospective use also which may prove useful in medical settings, as maternal caregivers sometimes reframed researcher questions to help Autistic CYP discuss a historical pain experience in increased detail. Such an interaction is demonstrated between A11 and M11:

A11: *"... I just don't know what to say because I don't know.",*

M11: *"Can you think of what pain is like for you, is it annoying? Is it something you, like, is it something that's horrible? Is it something you makes you feel alive or?",*

A11: *“Makes me like quite sensory overwhelmed erm like quite fidgety you know like if I feel pain I get really fidgety and stuff and like it’s all sensory overwhelming and I can't really like cope and stuff and like, yeah I don’t know.”*

Once pain has been recognised, Autistic CYP use words to describe what their pain means to others. These descriptions do not appear to provide extensive insight to pain nature and quality with both maternal caregivers and Autistic CYP explaining CYP’s descriptive vocabulary as brief and blunt. Descriptions typically extend to *“hurting”* or *“sore”* rather than providing abstract and metaphorical understanding. Yet upon providing direct questions Autistic CYP’s descriptive language did allow some understanding of pain quality although still limited to “*stabbing*”. Instead, use of comparison as a description appeared more useful. Maternal caregivers explained their Autistic CYP contextualise pain intensity to a “*pain anchor”,* comparing their current pain experience to a previous pain experience in the aims of providing context to their pain intensity. For example, A10 used a previous experience of breaking their arm to suggest their headaches are of a higher intensity, “*definitely like worse than breaking an arm”*. Additionally, A4 provided a personalised pain intensity scale through comparison of multiple pain experiences, “*banged head-first onto a concrete floor that would be very severe. But like I don't know, just, I don't- falling over onto my knee, that's fine.*”.

Autistic CYP also provided descriptive comparison through similes, explaining their pain as “like” something as opposed to typical metaphorical language which would explain their pain “is” something. For example, A3 used a simile to describe their headache as *“someone’s like hammering my head”* rather than typical metaphorical language like *“my head is banging”*. Use of pain anchors overlapped with these similes to provide personal rather than theoretical experiential context. For example, A6 anchors their simile to a sport they play using an experiential consequence that evokes a feeling pain to reference their own pain, “*It hurt really bad, whenever I stood I thought I would fall over and my legs felt too weak to support me they felt all shaky like if I did jiu jitsu too long.”*. Here a bystander can easily picture what this CYP is feeling through a created image in their mind’s eye. On the other hand, whilst use of similes was common Autistic CYP occasionally used metaphorical

language. Yet these descriptions were less detailed and appeared to be socially modelled, like A10 who described their pain to feel "*as if someone was pressing on my tummy all the time"* which is synonymous with HCPs questions when trying to physically understand location of pain. A10's pain scenario was in fact related to an experience in hospital, meaning their provided metaphor in this context may mirror HCPs language as opposed to their true pain description.

4.3.2.2 How Pain is Experienced

Pain is a multifaceted experience; much like in the non-Autistic population, pain causes differences cognitive abilities, affective states, and behaviours which Autistic CYP and maternal caregivers recognised and could discuss.

Autistic CYP possessed high self-awareness in how pain changes their cognitive abilities. Autistic CYP and maternal caregivers reported differences in communicating when in pain, such as being a likened to talking "*gibberish*" or not making sense; a point that is interesting given the communicative involvement required to recognise pain. M8 describes this could be consequential to the interrupting nature of pain, with an ability to focus on anything other than pain being difficult, *"I think the pain that is like it just trump's everything in those moment I think she can't function properly in pain."*. Additionally, cognitive differences were prevalent in in scholastic settings with Autistic CYP describing a reduced ability to attend to, and process information which A5 attributed as a cause for worsened academic performance:

> *"I normally have to concentrate really hard on something like that so if I'd like erm if I- if I'd hurt, hit my ankle earlier that day and it was still kind of hurting I'd probably wouldn't do as well as I normally would if I wasn't in pain [when doing homework], or I'd find it difficult to like pay attention if a teacher was explaining it."*

Interestingly, differences in memory were only explicitly identified by maternal caregivers who's Autistic CYP either had or were seeking an ADHD diagnosis. These caregivers could not determine whether differences were due to pain, or ADHD co-

occurrence, *"I mean, she struggles with them things anyways because of the ADHD so that is a daily thing …"* (M8). Despite maternal caregivers of Autistic CYP without ADHD typically suggesting pain did not impact their child's memory, "*I don't think it would have that much of an impact on remembering things for him"* (M4), anecdotes from Autistic CYP appear to suggest some facets of memory are difficult. A5 describes experiencing a headache in class, during which a familiar topic was discussed but despite having prior knowledge they *"couldn't get it and I just couldn't remember so I had to like look it up online later when I got home."*. This suggests the cognitive difficulties in consolidating and recalling memory-based information is a concern as opposed to the creation of an initial memory itself which again could worsen academic performance long-term.

Autistic CYP reported feeling increased negative emotions such as *"sadness"* and *"anger"* when in pain. However, cause of these negative affective states was varied with Autistic CYP and their maternal caregivers explaining feelings can be consequential to:

1) Pain severity, as described by A11 who reports their feelings of increased anger, sadness, and fatigue is consequential to how much their pain hurts, *"I feel really like maybe angry, you know, and like quite sad and tired … Like because it hurts so much, I'm angry that it hurts so much."*.

2) A limited capacity to cope with external factors, particularly with the social demands of others as A8 becomes increasingly frustrated by other people's behaviours, *"When I feel physical pain, and someone does something little a lot of things would trigger me and I'll get very frustrated at people."*.

3) And the behavioural limitations consequential to pain, such as limiting ability to engage with enjoyable activities, *"I think impacts upon her in terms of she'll want to bake or she'll want to go do something with her plants which is another thing she likes, but she doesn't feel like it because she's got a headache."* (M9).

Interestingly negative affective states caused an additional type of pain coined as *"emotional pain"*. As described by A4 emotional pain is caused by external matters, "*like say my teacher takes away erm my sketchbook then that, then that would make me feel mental pain"*, whilst physical pain is an internal sensation, "*like I don't know, tummy bug and erm and other things inside there. But like also like scratches and bumps."*. Typically, emotional

pain appeared to be more intense than physical pain, "*like I feel like people saying stuff affects me more than people actually hurting me."* (A8), and difficult to process when an exteroceptive understanding of a cause is absent: *"I think physical pain she can usually identify where the source of pain is coming from and sort of maybe what it is so it's like she can identify sciatic pain or back pain or a headache but if it's like emotional type pain and distress, she struggles to communicate the feelings around why that might be or what's causing it."* (M10).

Whether emotions were expressed was socially dependent on who an Autistic CYP was with and where they were. For example, A10 described their ability to express negative emotions was limited to the home as they felt safe in their authentic emotional display, and could mitigate negative social responses to their chosen method of expression, *"Erm so if I'm like with my family and something, I might shout at them while- even though I don't mean to like, just get a bit frustrated with anything really.".* Maternal caregivers further supported this interpretation with M8 highlighting incongruence between their Autistic CYP's emotional expressions of pain at home compared to at school. However, reason to rationalise incongruence was limited suggesting M8 may have developed a schematic understanding of their CYP's emotional expression which rarely deviates in their presence:

> *"… I've always been told by your school teachers since you were tiny that when you really hurt yourself in the playground or something and you've been to the school nurse or the teachers have had to like look after you, you've never ever cried when you've hurt yourself, which is the polar opposite of when she hurts herself at home and I don't know why that is so I don't know if you wanna think about that.",*

Autistic CYP displayed conscious awareness of how to *"get on"* with their pain to mitigate discussed negative affect from missing out or appearing different, *"say like I don't know, I've stubbed my toe in a game of tag I would just ignore it and keep playing tag because I care more about the game of tag"* (A4). Changes in these behavioural abilities to push through pain were indicative of pain severity, with Autistic CYP being less likely to change their behaviour by masking pain at higher severities. M1 explains how their child is increasingly unable to mask in school when their pain is at its worst, needing to go to sleep

on the table as opposed to conform to classroom expectations, *" she'd just go to sleep on a desk or whatever"*.

4.3.2.3 The Role of Relationships

Primary caregivers like mothers and fathers are key in validating Autistic CYP's pain; even when not prompting the processing of pain, they facilitate and can be the first and sometimes only person Autistic CYP disclose pain to. Additionally, primary caregivers act as advocates by disclosing pain on Autistic CYP's behalf when they feel unable to. For example, Autistic CYP felt more comfortable with their maternal caregivers disclosing pain to HCPs on their behalf than disclosing themselves, "*I usually tell her all of the information and, and then I'm like tell it at the doctors."* (A2). However, to aid in accurate advocation, maternal caregivers report rehearsing what they will say, in doing so they ask Autistic CYP "*what types of things we, we need to talk to the doctor about"* (A2), to ensure they are merely repeating their CYP's self-report rather than providing their own interpretation.

Whilst maternal caregivers perceived their advocation as predominantly useful, they recognised a lack of longevity and felt concern in how Autistic CYP could disclose pain in their absence, *"I guess the next stage as he kind of goes into the teenage years as he may not wish erm not to have me erm being his advocate and how he could kind of like get around that but still be able to erm effectively communicate to medical professionals"* (M4). There was discussion of how maternal caregivers were encouraging Autistic CYP to disclose their pain during healthcare appointments, however additional methods to facilitate pain disclosure are required. Specifically, Autistic CYP and maternal caregivers highlighted a need to build a trusting and familiar relationship to create comfort, "*if it was someone that he wasn't comfortable with then he probably wouldn't someone that's familiar and someone that he trusts, he has a big thing on trust."* (M3). This need was highlighted by Autistic CYP disclosing pain to a family member or a close friend that they trusted, and knew would help them access relief; more often than not, grandparents were the person an Autistic CYP would disclose pain to, " *I'd probably, probably go to my nan 'cause she's made me hot*

water bottles in the past and also she has, like, a massive cupboard of medicine in her house …" (A8).

Primary caregivers aim to improve the quality of healthcare Autistic CYP receive through disclosure of their Autism diagnosis; although Autistic CYP did not report this to yield benefit, maternal caregivers seem to think otherwise. However diagnostic disclosure creates an emotional toll for the maternal caregiver as negative perceptions of their intention are thrust upon them by the HCP. Described perceptions typically alluded to an attempt to conceal something or appear to want to direct the conversation. Yet maternal caregivers continue disclosing, putting their CYP's needs above their own feelings to ensure their Autistic CYP receive the care they deserve. M6 provides thoughtful insight of how diagnostic disclosure provides an emotional toll but recognises this is the only way to ensure their treatment is taken seriously:

> *"I always disclose it as soon as we walk in because doctors are brutal if you don't, I shouldn't have to disclose but the difference in care is night and day, so I always disclose … I had a child with a broken arm who had basically gone non-verbal, he was refusing to answer any of their questions, and they were taking it as trauma - let's get him away from mum so that we can talk to him on his own. And he was just feeling more traumatized, you're trying to take my mum away from me and trying to get ahead of the game and say look, he's Autistic, please don't stress him out any more than we have to but also please don't report me to social services again for an injury that you know just because my son can't speak when he comes in, he's not hiding anything for you, he literally can't speak, he just can't and it's not because we're playing a game and we don't want to talk to you it's because he genuinely can't do it."*

Unfortunately, Autistic CYP had a general mistrust of HCPs due to previous experiences of disclosing pain where they had not been believed or received support in managing pain. M1 explained their Autistic CYP's pain was dismissed in an emergency setting, "*I took her to the Walk-In Centre for them to look thinking it was a fracture or something, and they were sort of quite dismissive and sort of 'well no, this is just this oh*

you'll be fine". In response, their CYP felt upset feeling as though her pain was not validated, internalising this interaction to mean *'Is it nothing, am I just meant to feel nothing?'*. Consequentially Autistic CYP reported they were reluctant, or did not want to show vulnerability in disclosing pain to HCPs to avoid continual invalidation so instead chose to not say anything, *"[In context of not wanting to talk to HCP] like they lie … I don't believe them anymore but there's no point in arguing … because they don't listen."* (A9). This is a concerning pattern of behaviour that could manifest throughout adulthood and potentially facilitate the progression of acute to chronic pain.

Feelings of distrust were not reserved to HCPs, Autistic CYP reported refraining from informing their teacher of pain if they could not trust them. For example, if a teacher had not, or a CYP believed they would not help relieve or validate pain, the likelihood of disclosure decreased - almost like a *"What is the Point?"* approach, *"she probably wouldn't tell the teacher that it was because she was in pain because she'd be worried, they didn't believe her."* (M11). This approach was exacerbated through fear that pain would get Autistic CYP into trouble as teachers were not sensitive to how pain impacted their cognitive ability. Here A4 expressed all they would like is an appreciation of their pain experiences, and recognition that changes in their cognitive abilities are at the onus of their pain, not themselves as a human:

> *"… they'd normally tell me off for it instead of sympathising because they're only noticing that I'm not doing enough work, they're not noticing that I'm hurt, like they don't really understand why I wouldn't be able to do it because of that. Erm, I wish that they could understand that it would, it slows me down even if I'm feeling pain, I just like them to like not tell me off for it I don't mind if they don't really do much."*

An additional need for familiarity in scholastic settings was required for Autistic CYP to disclose pain, "*like a trusted adult, someone that knows her specifically, knows her medical history."* (M2). Here, Autistic CYP specifically reference a need for someone they were 'close' to, spent a lot of their time with to facilitate trust, *"Erm, probably with someone on the SEN team my friend he's got a one to one, so considering [my friend] and I sit next to*

each other in almost all of our classes and I can just tell her and she'd probably do something." (A3).

However, if an Autistic CYP did disclose pain in the absence of a familiar and trusting scholastic relationship, maternal caregivers highlighted this as indicative of high pain intensity, "*if it was a bad thing, I think I don't think he'd hesitate 'cause I think he'd realise 'I need to do something about this'."* (M4). In this case, maternal caregivers explain teachers should refrain from providing sweeping judgement of their CYP's intent to disclose pain, as this behaviour is indicative of help-seeking thus a need for relief is urgent. M5 provides an example of this in their statement:

> *"… when he's had headaches and he's actually said it to the teachers, she's gone 'oh well we all get headaches' and you know, and I've said no for him to say that, that means he's pushed to like this is really bad and I feel my head is going to explode, not I've got a little bit of a headache and I can't be bothered listening you."*

To overcome these disclosure barriers, HCPs were suggested to employ advocacy-based approach in their medical assessments to facilitate pain disclosure by relieving pressure from Autistic CYP. This involves mirroring maternal caregivers scaffolding behaviours, such as asking CYP direct questions free from typical text-book jargon to help in recognising and disclosing pain, "*I just think that they would say more dependent on the person asking them the questions and how comfortable they felt with the person."* (M1), and *"Doctors will go well how does the pain feel? And it's such a stupid question."* (M5). Similarly, maternal caregivers suggest HCPs take time to understand Autistic CYP's previous medical history, utilising these experiences as an anchor in the current disclosure context.

Moreover, teachers were recommended to take time to learn from primary caregivers the discrete changes in Autistic CYP's demeanour to aid in pain recognition. M3 highlights how this knowledge of something as little as a change in facial expression is highly beneficial for initiating relief, even when a CYP has not recognised a sensation of pain themselves:

"Yeah, it's his teacher in the infant always used to say he used to go like green she said she could tell be the colour of his face whether he had a migraine or whether it was gonna be a bad one of not because when he was very little and he wasn't in the in like when he was unable to kind of notice himself she just used to kind of watch the colour that his face was if it went a bit of a green yellow colour, she knew there was one coming."

4.3.2.4 Playing an Active Role in Pain Management

Autistic CYP can be self-sufficient in relieving pain, even playing an active role in knowing what relief they need and how to seek this (i.e. can relief by accessed alone or with the help of their primary caregiver). Here the use of A5's first-person language (*"I*") demonstrates their ownership in actively managing their relief, displaying knowledge of what relief they need, *"Normally I just rest it [ankle pain]. I'd probably, I'd probably have to just rest it on like, on like a chair or on my bed if that makes sense … it depends what type of pain it was but sometimes I might take paracetamol like for example if I had a sore throat or like a stomach-ache, erm I'd take paracetamol or calpol.".*

Typically medicine, distraction with TV, and self-soothing by using sensory seeking behaviour were described as common sources for pain relief, yet certain methods for relief were reserved for certain types of pain at differing intensity levels. Here Autistic CYP explained how they used their knowledge of their pain patterns and previous behavioural prompts, i.e. vomiting, to choose an adequate form of relief. A3 describes how the intensity of their pain helps them access relief due to their schematic understanding of their migraine experiences:

"*It like slowly comes. It just starts as like a little bit of throbbing in my head and then about a minute or so later then if we don't get medicine or something in that minute time, then that's when I feel like I wanna rip my head off … medicine's definitely the first one we'll go for … but then if it gets worse it's probably the stress ball and a warm bath."*

Additionally M2 exemplifies the use of medical input such as physiotherapy and pain analgesic's to manage their child's muscular and joint pain, whilst migraines can be managed with at-home solutions like 4-head cooling stick and calpol, *"So with the joints her muscles, it is just a physiotherapy PRN, pain meds as and when needed, warm bath… with the migraines we do have the you know 4-head thing erm and calpol"* (M2).

Traditional analgesic methods involved communication with an adult to attain required medication. Autistic CYP expressed continued autonomy in seeking their primary caregiver when medication was needed and describing which medication they needed. For example, A2 possessed an active role in managing their pain ("*I"*), however their mum was required to access calpol, *"usually when I feel it's- feeling it starts erm I go to mum to ask for calpol.".* In this case, caregivers acted as a gatekeeper to analgesic's rather than a provider, as CYP described routine knowledge of when they needed to use medication, "*Like every four hours or whatever it is the paracetamol and then ibuprofen before I go to bed."* (A1).

In comparison to relieve pain without the need of a gatekeeper, Autistic CYP used methods of distraction like watching TV to divert their attention away pain, *"something that she can maybe just focus on other than the pain…"* (M2). The cognitive demand a method of distraction required influenced its effect as a passive pain relief. For example, at higher pain intensities more interactive methods of distraction like reading a book or playing a game were difficult to engage with, and thus TV was an easier method of distraction, *"it's harder with a book because I have to focus on it and imagine it but with the TV it just happens and I don't have to do anything"* (A6).

Minimising the cognitive demand of TV, Autistic CYP watched a familiar or special interest programme (i.e., *"The Simpsons"* or *"Brooklyn 99"*) ensuring predictability of the programme mitigated the requirement of information processing whilst still being enjoyable enough to divert attention from pain, "*I'm just watching it for the sake of it I'm not actually you know paying attention to it that much, just watching it just listening to what they're saying innit."* (A1). Moreover, TV could not have a high amount of sensory input like loud noises and flashing lights, as explained by A3 whom will avoid Marvel films as the required sensory

processing worsens their pain opting for a calmer show typically directed for a younger audience:

> *"I think it depends what program it is. I think it's just like it's a bit louder and it's just a lot. Yeah and there's loads of like flashing lights, especially with Marvel 'cause you've got like all the different powers. Generally like "Sam and Cat" or "The Loud House", that's fine."*

Sensory seeking behaviour was often used alone, and involved tactile objects like soft blankets, clothing tags and stress balls to self-soothe and distract, *"like these tags, they're smooth- they help me kind of calm down like they help me like focus on them and not the migraine."* (A2). This method is not unexpected given the commonality between using sensory objects to regulate emotions and Autism as a diagnosis. The tactile object sought was dependent on the sensation, and often indicated Autistic CYP's pain severity as maternal caregivers reported they saw their Autistic CYP reaching for a specific teddy bear for comfort, when they were in a lot of or experienced a specific type of pain, *"I'd know that if she's just sat there with this teddy that there's something very wrong with her that, you know, we need to address it…"*. This behaviour was unconscious in nature for Autistic CYP as they did not reaffirm this pattern.

Unsurprisingly, the efficacy of relief was dependent of the Autistic CYP's location. When at home, pain disclosure was easier and there was access to a wider range of relief options, whereas in school options were limited with disclosure-based barriers preventing access to relief. For example, A9 describes experiencing a headache at school yet only accessed relief when at home, *"When I got home erm well it got a little bit better before and it got quite a bit better, but that was like I only just had it [ibuprofen] really so. And also when I put on the cool patch it got quite lot better. Ibuprofen probably helped the best.".* However, A9's maternal caregiver, M9, explained they in fact had ibuprofen at school but this was never accessed due to barriers in requesting relief, *" she's got medicine in school but she's never, ever gone and had medicine in school because she doesn't tell them when she feels like she needs it.".* This appeared to cause concern amongst maternal caregivers as despite

facilitating physical access to relief outside of the home, when they were not there to scaffold in communicatively acquiring, for example ibuprofen, relief was not used.

4.4 Discussion

Knowledge gaps in understanding how psychological and social factors shape Autistic CYP's pain experiences were addressed using a qualitative approach. Findings suggest that cognition influences Autistic CYP's ability to recognise pain, whilst emotion holds greater relevance in how pain is experienced. However, social factors like interpersonal relationships and behavioural appraisal both influence effects of these psychological factors and the ability for pain expression, pain disclosure and access to relief. Here, Autistic CYP's social expectations of how others respond to their pain may act as a fundamental gatekeeper to their ability to access pain management and have a greater role to play in explaining pain overrepresentation. This is discussed in further detail below.

Despite evidence suggesting Autistic CYP display hyporeactive interoceptive abilities (Dubois et al., 2016), current findings highlight Autistic CYP can interoceptively attend to internal cues for pain recognition. Schauder et al. (2015) originally suggested Autistic CYP disproportionately attend to interoceptive internal cues ascribing this as a core symptom of Autism, yet our findings highlight such interoceptive attention is limited to higher pain severity. Instead, findings reflect an Autistic interoceptive pattern more salient with Bogdonova's (2022) model (see Chapter 1.3.1), which explains interoceptive differences constitute an altered top-down regulation which diminishes the ability for conscious pain appraisal. Current findings support Bogdonova's (2022) model that conscious pain appraisal may be difficult in Autistic CYP; however, we suggest Autistic CYP may utilise top-down regulation to promote pain processing through a reliance on social-context. Here, findings could suggest pain interruption in Autistic CYP is consequential to exteroceptive attention towards social environment and relationships for social appraisal, rather than an internal sensation for self-appraisal. For example, environmental context and communicative prompts incorporating past pain experiences facilitate top-down processing and pain appraisal by providing Autistic CYP with a causal expectation of pain. Whilst this may not

explain pain overrepresentation, it presents a method of assisting Autistic CYP in recognising pain which places social involvement as important to interoceptive understanding.

Mirroring expectations of Craig's (2015) Social Communication Model of Pain, Autistic CYP recognised cognitive and emotional differences due to their pain, increasingly so when in a scholastic environment. Whilst cognitive differences may be easier to identify at school as CYP spend extensive time there, Horgan et al. (2023) suggests loud noise and social demands create a less inviting scholastic environment for Autistic people which in this context is likely to influence a worsening of pain cognitions. Observed cognitive differences mirror Donaghy et al.'s (2023) and Horgan et al.'s (2023) reviews, finding pain interfered with Autistic CYP's ability to attend to and process information with consequential worsened academic performance. A change in affective state was additionally experienced with Autistic CYP describing feeling anger and sadness when in pain, however these affective states were not deemed as a pain facilitator as research suggests (Kalingel-Levi et al., 2022; Mazurek et al., 2013; Williams et al., 2015). Instead, emotions were a consequence of higher pain intensities, a lack of ability to engage with enjoyable activities, and a more salient Autistic diagnostic focus of processing sensory environment as described in the context of hospitals environments. Unlike non-Autistic populations (Fisher et al., 2016), cognitive differences and highly negative affective states did not appear to promote pain-avoidance behaviours as Autistic CYP continued through their pain in the aims of not socially differing from other CYP and missing out on activities. Whilst Autistic CYP did not explicitly position pain in their identity, the need to appear socially congruent to others may suggest Autistic CYP's pain experiences promoted a perceived sense of difference similar to Jordan et al.'s (2024) findings.

On the other hand, masking of pain appeared to also mitigate social consequences that create a state of vulnerability and distress, perhaps demonstrating a unique emotional pain-avoidance model for disclosure. This notion of whom an Autistic CYP expresses their pain to may be more driven by emotional state than pain itself, causing its own visceral pain reaction when social consequences are negative, better coined by participants as *"emotional pain"*. Whether an Autistic CYP emotively, behaviourally, or verbally expressed their pain

was dependent on social environment and quality of an interpersonal relationship, which only those with trusted and familiar relationships would outwardly observe their pain. In the absence of these interpersonal qualities, Autistic CYP's schematic understanding of who will cause emotional distress by invalidating, misinterpreting, and mismanaging their pain, and who will help was important for pain expression. However, ability to continue masking decreased as pain intensities increased, with caregivers highlighting this change as an observable method in understanding Autistic CYP's pain at its worst. This strengthens Allely's (2013) and de Knegt's (2013) perspectives of behavioural expression supporting that differences in pain behaviour reflect a higher pain intensity amongst Autistic CYP. These findings provide additional understanding that behavioural pain expression may not reflect a conscious intent to express pain, but rather an inability to cope as cognitive energy is no longer available to mask pain (see "Spoon Theory" by Miserandino (2003)). At this point a need of relief is a necessity, not a matter of debate

Consistent with Ely et al.'s (2016) and Kalingel-Levi et al.'s (2022) findings, caregivers were described as the first, and often only, individual Autistic CYP would express and disclose their pain to. On the other hand, untrustworthy HCPs and teachers were omitted from observing expression and disclosure due to historical experiences of them disbelieving and misinterpreting an Autistic CYP's pain; an experience a breadth of systematic reviews highlights as a healthcare barrier for many of the Autistic population (see Haydon et al. (2021); Mason et al. (2019) and Walsh et al. (2023)). It is likely these misinterpretations result from a lack of training in how Autistic CYP communicate pain, as HCPs often explain they feel lack knowledge about Autism, and the differing ways an Autistic person communicates pain (McCormack et al., 2020; Zerbo et al., 2015). However, whether teachers predominantly fail to appraise pain is less understood from available literature. These findings suggest Autistic CYP's pain communication were once again not congruent to teachers perceived neurotypical understanding and thus teachers did not appraise pain, reinforcing the same observed emotional pain-avoidance model that HCPs contributed to. These barriers to disclosure additionally prevented Autistic CYP from accessing pain relief, as often teachers and HCPs acted as a gatekeeper to effective analgesic relief in the absence of a caregiver. Consequentially Autistic CYP were unable to

verbally or expressively request this need for analgesia when a teacher or HCP was not trusted to help, contributing to a potential worsening in their pain. This may suggest why Autistic CYP appear to have taken an active role in managing their pain through methods of distraction and sensory seeking, as doing so mitigates required expression to others which has not been successful and created a highly negative emotive state.

Barriers outlined in these findings including lack of trust, disbelief of symptoms and previous negative experiences are synonymous with wider researched Autistic barriers to healthcare (Haydon et al., 2021). A continuation of Autistic CYP being in social environments that do not overcome barriers to promote feelings of safety in their pain appraisal may cause lifelong trauma with Autistic CYP feeling their healthcare experiences are not important and presents a burden to them accessing treatment (Doherty et al., 2021; Langhinrichsen-Rohling et al., 2021). It is paramount that understanding how to overcome these barriers is addressed in early childhood, as Palermo (2020) highlights vulnerabilities in CYP's health behaviours may impact their outcomes as an adult. Thus, intersecting the cycle of Autistic CYP not expressing or disclosing pain may prevent their pain from being continually untreated, and potentially progressing in chronicity throughout adulthood (Mallen et al., 2006). Previous studies have suggested relevant adaptations for Autistic individuals that typically lean towards an improvement in training and sensory adaptive environments (Doherty et al., 2021; Rios-Vega et al., 2024). Whilst relevant, these findings provide further adaptation knowledge catered to an Autistic paediatric population. For example, von Baeyer et al. (2004) suggests the benefits of asking CYP "Wh-" questions to provide open responses. Our findings counter this, as when asking Autistic CYP about their pain direct and closed questions appeared to yield an ease of response. Additionally, HCPs and teachers should adopt a person-centred approach in deciphering Autistic CYP's pain. For example, Autistic CYP described the importance of comparing their current pain to a previous pain to indicate intensity and provided description through simile. This finding was not limited to these participants, with Han et al.'s (2024) analysis of Autistic CYP's case-notes and clinical observations recognising the same use of personal descriptions. Given its real-world evidence in clinical settings, HCPs who take the time to understand Autistic CYP's past experiences will gain a greater understanding of their current pain, and refrain from

typical methods of pain assessments (see Chapter 1.2.4) to gauge a rating of intensity. This need for a person-centred approach extends further to utilising a change in behaviour to understand pain at its highest intensity rather than as an indicator of pains presence; this could be particularly useful in non-verbal Autistic CYP. This use of behaviour has already been recognised by Liu et al. (2020) who proposed a need for modified observational assessments for pain reflecting findings here. Whilst not an immediate cause and effect in Autistic CYP's pain communication, utilising these adaptions may allow pain to be more adequately managed and perhaps prevent effects of pain overrepresentation in childhood, and into adulthood. Yet to better tailor these adaptive approaches in facilitating young people, a further understanding of what behaviours have contributed to Autistic CYP's schematic understanding of who will and who will not appraise their pain is required.

Although this research provided a forum for Autistic CYP's voices about their pain to be shared, there are limitations to the application of these data. For example, participants identified a pain scenario for interview context which was deemed vital in assisting Autistic CYP's engagement. However, during these scenarios Autistic CYP presumably engaged with prompts to recognise pain, leaving limited understanding of difficulties in pain recognition when prompts are inaccessible. Findings therefore may overgeneralise use of prompts in real-world context and not appreciate the magnitude of interoceptive difficulties. Moreover, acknowledging Milton's (2012) Double-Empathy Problem, the PhD researcher's Autistic insight may have biased analytic interpretations by subconsciously ascribing their own lived experience to explain Autistic CYP's experiences. However, this is not critical to analytic validity given methods use to ensure data rigour, and IPA's recognition of the researcher's lived experience in directing interpretation.

More than ever, research is needed to further understand Autistic CYP's pain experiences. Given the relevance of social factors, psychophysical methodology outlined in Chapter 3.2.4.2.2 should be adapted where possible to understand how these differing social factors influence Autistic CYP's psychophysical pain intensity and ability to disclose pain. The review-based framework provided by Krahé (see Krahé et al. (2013) and Krahé and Fotopoulou (2018)) could be used to in inform further social adaptations that may be pertinent (i.e. parental presence vs absence and caregiver-child attachment styles). The

current PhD had aimed to conduct this study, but due to COVID-19 restrictions was unable to.

Overall, Chapter 4 findings suggest that much like in non-Autistic populations, Autistic CYP's pain experiences are actively shaped by psychological and social factors. Psychological factors like cognition and emotion appear to influence the broader experience of pain like pain recognition. Yet the relevance of social factors such as behavioural appraisal and interpersonal relationships may exude greater impact on an Autistic CYP's ability to express, disclose and relieve pain. This becomes apparent amongst differing relationships, with changes in outward pain expressions being observed for caregivers, teachers, and HCPs. Ultimately, the ability to express pain appears to act as a gatekeeper to Autistic CYP's pain relief, with a need to understand how to facilitate pain disclosure amongst Autistic CYP to prevent their pain being mismanaged and progressing in chronicity being paramount. Chapter 5 will begin to broach this understanding through exploring factors that may influence Autistic CYP's decisions to disclose pain with relevance to bystander pain appraisal. With the suggested importance of social relationships in Autistic CYP's decisions to express and disclose, relationships deemed significant here will be considered in the role of bystander: caregivers, teachers, and HCPs.

Chapter 5.

“They Often Feel Like If They Speak Up, They’ll Be a Burden”: Exploring Autistic Children and Young People’s Pain Disclosure from a Caregiver Perspective

5 “They Often Feel Like If They Speak Up, They’ll Be a Burden”: Exploring Autistic Children and Younge People’s Pain Disclosure from a Caregiver Perspective

5.1 Introduction

A qualitative exploration of Autistic CYP’s pain experiences in Chapter 4 provided insight which factors influence Autistic CYP’s pain. As expected, psychological factors like cognition and emotion were discussed. However, a large emphasis was placed upon how social factors like trust, disbelief of symptoms and previous negative experience influence an Autistic CYP’s ability to socially communicate pain through outward expression and interpersonal disclosure. Here, a focused understanding of why Autistic CYP do not disclose pain in certain relationships will complement Chapter 4’s findings and facilitate the development of methods to better support Autistic CYP in pain.

Within Chapter 1.3.2, available literature outlined the communicative processes Autistic CYP use to self-report pain, indicating verbalisations are a common method for pain disclosure in verbal Autistic CYP (Bandstra et al., 2012; Ely et al., 2016; Fitzpatrick et al., 2022). Many bystanders rely on this verbalisation to gain insight into an Autistic CYP’s pain including the location, duration and quality, as bystander interpretation of non-verbal behavioural or expressive cues can often be incongruent to the individuals internal state (Courtemanche et al., 2016; Daughters et al., 2007; Militerni et al., 2000; Tordjman et al., 2009). However, as identified in Chapter 4.3.2.3, a lack of trust, external disbelief of symptoms, and negative social experiences can limit an Autistic CYP’s behavioural intent to verbalise pain. If Autistic CYP continue to experience these negative interpersonal interactions, they may continue to avoid disclosing pain to HCPs and teachers. This is of particular concern as methods to socially communicate pain develop in childhood (Emerson & Bursch, 2020), and whilst emotional distress may be mitigated when Autistic CYP do not disclose pain in certain interpersonal relationships, harmful consequences are likely. For example, a lack of adequate care from not disclosing pain may contribute to the worsening of symptoms and promote the development of chronic pain in adulthood (Palermo et al., 2010). An in-depth understanding of what influences these disclosure decisions is therefore

required to understand how Autistic CYP can be better supported in expressing pain, and bystanders in appraising their pain.

Experiences of mistrust and disbelief are not specific to Autistic CYP in pain; non-Autistic CYP share these negative interactions. For example, Carter (2002) found that CYP in search of a chronic pain diagnosis often had their pain diminished and disbelieved by HCPs. These experiences are broadly reported to produce lasting hostility within the interpersonal relationship where pain was disbelieved like teachers, friends, and even family members (Defenderfer et al., 2018). Additionally, Wakefield et al. (2021) suggests CYP with chronic pain may continue to hide their symptoms to avoid the emotional distress that accompanies disbelief. However, these feelings of distress are much higher when a caregiver (reported by 38%), or HCP (reported by 17%) dismiss their pain which could suggest these two relationships are of importance for disclosure decisions, much like in Autistic CYP.

Whilst these shared experiences may limit the relevancy of applying a social lens to understanding differences in Autistic CYP's experiences of pain disclosure, broader healthcare experiences may contribute to reinforcing the behavioural effects these factors have. For example, in mental health literature, caregivers of Autistic CYP report that when their CYP trusts their therapist they are more likely to be truthful in their experiences, highlighting that interpersonal factors are important across an array of healthcare systems (Jackson et al., 2020). Additionally, these experiences of mistrust and disbelief in non-Autistic CYP are provided in the context of chronic pain, where the effects of disbelief may be strengthened when pain experiences hold a greater relevance to their identity (Jordan et al., 2018). Instead, the broad experiences of disclosing pain amongst interpersonal relationships should include any CYP in pain and compare between Autistic and non-Autistic CYP's experiences to identify if similar themes are observed, or stark differences.

While Chapter 4 highlighted important social factors for pain disclosure, summarising the complexities of behavioural intent to the findings of a singular study may undermine Autistic CYP's lived experience, and limit the development of directed methods for support. Additionally, how these social factors differ between interpersonal relationships

like caregivers, teachers and HCPs as identified in Chapter 4 are not provided in detail to suggest how these methods of support can be adapted. To develop this understanding, the aim of this mixed-methods study was to utilise a caregiver perspective to explore decisions for, and compare differences between Autistic, and non-Autistic CYP's pain disclosure amongst three interpersonal relationships: caregivers, teachers and HCPs.

5.2 Methods

5.2.1 Design

A mixed-methods online survey was used to understand if experiences in pain disclosure differed between Autistic and non-Autistic CYP from a caregiver perspective. Quantitative and qualitative data approaches were used concurrently (Schoonenboom & Johnson, 2017); however, findings were qualitatively driven to further develop relationship-based themes discussed in Chapter 4.

Originally psychological predictors, and experiential data were collected from both participating CYP (Autistic and non-Autistic), and their caregiver's perspective - placing CYP's voices at the forefront of this research. However, only caregiver materials and responses are reported, as difficulties in recruiting CYP meant their involvement was stopped during data collection. For design transparency, all participants in the intended design completed materials listed in Chapter 5.2.3 in the context of themselves, with CYP completing an additional unreported Child Pain Anxiety Symptom Scale (Pagé et al., 2010).

5.2.1.1 Design Validation

A PPI panel (see Chapter 0.1.2) was consulted to assess applicability, validity and inclusivity of study design and materials with awareness of how previous study findings influenced this methodological design. All recommendations for improvement were implemented prior to study commencement. For example, given the use of open-ended questions (see Chapter 5.2.3), the PPI panel provided extensive insight into question

relevancy and wording. This facilitated participant understanding of questions and allowed comparable answers between groups for analytic comparison.

5.2.2 Participants

From May 2023 to May 2024, a snowball sample of caregivers were recruited for participation. Initial recruitment was predominantly indirect using social media and gatekeeper outreach. For example, accessing the Autistica Network of participants. However, due to low sample size in the non-Autistic group, use of Prolific (2024) allowed direct sampling from an online pool of participants in April 2024. The inclusive sample totalled 144 caregivers (QuestionPro, *n* = 63; Prolific, *n* = 81) across 18 countries; 3 most common being the UK (62.50%), South Africa (11.11%), and Poland (6.25%).

5.2.2.1 Inclusion Criteria

Participants were eligible for inclusion if they were the caregiver of a CYP aged 11-15 years who was clinically diagnosed as Autistic (Autistic group), or who was not clinically diagnosed or suspected to be Autistic (non-Autistic group). As reported in Chapter 0.2, the inconsistent age range of 11-15 years is a result of consideration for Section 2(1), and Sections 3(1) (a) (b) (c) and (d) of the MCA (2005).

5.2.3 Materials

5.2.3.1 Recruitment

5.2.3.1.1 Social Media Poster.

Social media posters were used to recruit caregivers (see Appendix 15). Posters provided inclusion criteria, methodology, contact details for any questions, and a QR code to redirect interested participants to the survey host website, QuestionPro (2024).

5.2.3.2 Questionnaires

A series of validated, and survey-created questionnaires were used to collect data within three domains of the pain experience: pain description, pain disclosure, and psychological predictors.

5.2.3.2.1 Pain Description.

5.2.3.2.1.1 Pain Description Open Text.

As language use for describing pain was discussed during previous qualitative interviews (see Chapter 4.3.2.1), two questions were created to provide understanding of how caregivers perceive their CYP to describe pain. These questions were consistent with Chapter 4's interview study, asking caregivers to use the open text function to:

1) Describe a time their CYP had been in pain, ***"****Please tell us in the box below about a time your child has experienced pain in the last 12 weeks (British Pain Society, 2021)?"*
2) Describe how their CYP described this pain to an adult, *"In the box below please state how your child would describe the pain they experienced as if they were telling an adult?"*

5.2.3.2.1.2 Common Pain Frequency.

Caregivers were asked, *"How often in the last 6 months has your child you felt X",* which X was replaced with seven common childhood pains: headache, migraine, stomach-ache, dental pain, menstrual pain, muscle pain, and sore throat (McGrath et al., 1994). Distinctions were provided between headache *(pain or discomfort in one or more areas of the head or face)* and migraine *(type of headache that recurs (keeps coming back), the pain is often throbbing and can happen on one or both sides of the head) were outlined to limit overreporting).* Caregivers rated pain frequency using a 6-point Likert scale (1 – Everyday to 6 – Never) with an additional open text option called "Other" to specify their own frequency.

Menstrual pain provided a 7th option of “Not Relevant”. A higher score indicated a CYP experienced the described pain more frequently.

5.2.3.2.1.3 Common Pain Intensity.

Where a value other than “6 – Never” was selected for pain frequency, caregivers were asked: *“Please rate how painful your child’s worst X in the past 6 months was”.* Ratings were provided using the same pain intensity scale outlined in Chapter 3.2.3.3.4, with a higher score indicating their CYP experienced increasingly intense pain.

5.2.3.2.2 Pain Disclosure.

5.2.3.2.2.1 Disclosing to a Caregiver.

To understand if CYP disclosed pain to their caregiver, a binary question (yes or no) was presented to caregivers: *“If your child was with you and began to feel in pain, would they tell you that they are in pain?”*

Where CYP would disclose pain, caregivers indicated who their CYP would tell first between a maternal, paternal, or both caregivers. Caregivers specified “other” if these options were not inclusive, i.e. single caregiver, multiple caregivers or caregivers of the same identifying gender. Where CYP would not disclose pain, caregivers were presented with free text to identify reasoning for this answer.

5.2.3.2.2.2 Disclosing to a Teacher.

To understand if CYP disclosed pain to their teacher, a binary question (yes or no) was presented to caregivers: *“If your child was at school and began to feel in pain, would they tell a member of staff (i.e. class teacher, SEN) that they are in pain?”*

Where CYP would disclose pain, caregivers indicated which scholastic staff CYP would tell first, choosing between a class teacher, SEN tutor, or both. Caregivers specified “other” if these options were not inclusive. To aid in understanding staff availability (i.e. if a

CYP was preferring to disclose to a teacher over an SEN, or they only had access to an SEN), caregivers indicated if an SEN was available. Where CYP would not disclose pain, caregivers were presented with free text to identify reasoning for this answer.

5.2.3.2.2.3 Disclosing to a General Practitioner.

To understand if CYP disclosed pain to their general practitioner (GP), a binary question (yes or no) was presented to caregivers asking: *"If your child was at you doctors (GP) and was in pain, would they tell their doctor (GP) that they are in pain?"*. Where CYP would not disclose, caregivers were presented with free text to identify reasoning for this answer.

An additional question as which communicative method CYP used most to interact with their GP was asked to further understand how healthcare experiences may influence these decisions. Forced responses included: face to face, online chat, telephone, and other with free text for clarification.

5.2.3.2.3 Psychological Predictors.

5.2.3.2.3.1 Pain Catastrophizing Scale-Parents (PCS-P).

The 13-item, validated questionnaire was used to assess how caregivers think and feel when their CYP is in pain (Goubert et al., 2006). Using a 5-point Likert scale (0 – Not at all to 4 – Extremely), caregivers rated how frequently they engage with 13 statements referencing thoughts and feelings about their CYP's pain. Scores were calculated as a total PCS-P ranging from 0-52, and as three subscales of catastrophizing: Rumination, Worry, and Helplessness. A higher score indicated the caregiver was more likely to catastrophize their CYP's pain.

5.2.3.2.3.2 Fear of Pain Questionnaire-Parents (FOPQ-P).

The 21-item, validated questionnaire was used to assess caregivers' pain-related fears and avoidance behaviours in response to their CYP's pain (Simons et al., 2015).

Caregivers were presented with 21 statements explaining how they may think or behave in response to their CYP's pain and rated on a 5-point Likert scale how much the statement reflected themself (0 – Strongly Disagree to 4 – Strongly Agree). Scores were calculated as a total FOPQ-P ranging from 0-84, and 4 subscales: Fear of Pain, Avoidance, Fear of School, and Fear of Movement. A higher score indicated a caregiver engages with higher levels of pain-related fears and avoidance of behaviours in response to their CYP's pain.

5.2.4 Procedure

Upon receiving ethical approval (LJMU UREC ref: 23/PSY/026), participants were recruited indirectly through social-media (i.e. academic X profile, Autism-focused, or Autistic-led Facebook groups, and parent Facebook groups), and outreach (i.e. Autistic participant pool, local Autism groups, and North-West schools). Direct recruitment through Prolific (2024) was used to increase sample size, with a final participant cohort being paid to participate.

Participants self-identified if they satisfied inclusion criteria, with eligible participants accessing the online survey host website, QuestionPro (2024), through QR code or an accompanying link. Here, relevant PISs including contact information for support outlets were provided which emphasised participation was anonymous, voluntary, questions could be left blank, and required breaks were encouraged. Participants were additionally informed whilst they could withdraw by exiting the browser during participation, completed data could not be withdrew due to anonymity preventing identification. Informed consent was acquired by participants ticking a box to signify their awareness of the study's purpose and understanding that consent was implied by completing the questionnaire.

Participants provided demographical information before completing questionnaires in the following domain order:

1) Pain Description
2) Pain Disclosure
3) Psychological Predictors

Upon survey completion or withdrawal, participants were provided a debrief sheet outlining study purpose, data use and contact details of support outlets. Participants recruited through social media and outreach who wanted to be entered into a prize draw to win 1 of 5 £20 Love2Shop vouchers were redirected to a separate study hosted via QuestionPro (2024) to enter their email. The redirected survey stored the personal information separately from survey responses to maintain anonymity. Participants recruited through Prolific (2024) were not eligible for the prize draw and instead compensated £3 upon evidencing completion through a separate Prolific (2024) link.

5.2.5 Data Analysis

To explore pain disclosure, quantitative and qualitative data were analysed separately and then integrated to provide understanding of perceived disclosure decisions. This approach has previously been used to analyse healthcare experiences from the perspective of Autistic CYP's caregivers (Ashworth et al., 2024).

QuestionPro (2024) data were imported into Microsoft Excel for data cleaning whereby the PhD researcher looked to identify anomalies in data to ensure validity of response. Originally, a sample of 161 participants were exported however if caregivers disclosed their CYP were suspected to be, but not clinically diagnosed as Autistic, their response was excluded (n = 17). Datapoints were also highlighted where caregivers disclosed their CYP was home-educated in free-text for analytic purposes. Once cleaned the qualitative data of 144 participants were analysed in Microsoft Excel, and quantitative data imported for analysis in SPSS v27.0 (IBM Corp, 2020).

Originally, additional analysis comparing data between participating CYP (Autistic and non-Autistic), and their caregiver's perspective was proposed. However, as CYP's involvement was stopped due to difficulties in recruitment, some data points were less valid in use. This includes pain description data, which are not reported as supporting CYP perspectives were not available to validate accuracy in caregiver reports. Additionally, pain descriptions did not provide meaningful data because of the free response option. Data was

predominantly brief providing little qualitative understanding, for example, saying a CYP describes their pain as “it hurts”.

5.2.5.1 Quantitative Analysis

To describe the participant sample, descriptive statistics were calculated using demographic information (i.e. verbal communication). Where data was missing during quantitative analysis, the participant was excluded from the relevant analysis and reported in text.

A Chi-Square Test of Independence were used to compare whether diagnostic group and reported common childhood pain frequency, or pain disclosure decisions were significantly associated. However, where assumptions for Chi-Square Test of Independence were violated (i.e. expected cell frequency <5), a Fishers-Exact Test for 2x2 contingency tables, and a Fishers-Freeman-Halton Exact Test larger contingency tables were reported (Coolican, 2024; Lydersen et al., 2007). For either test a $p > .05$ was deemed a non-significant result (Coolican, 2004). Where significant associations were observed, adjusted standardised residuals were calculated for each contingency table cell; value’s higher than 2 were considered to drive the overall significance (Bewick et al., 2004). These statistics were contextualised utilising qualitative analysis as listed in Chapter 5.2.5.2.

Statistical testing was further utilised in identifying differences in pain intensity ratings and pain psychological factors between diagnostic groups; $p > .05$ was deemed a non-significant result (Coolican, 2004). Dental pain ratings, and both total and subscale data of the FOPQ-P, and PCS-P satisfied parametric assumptions for Levene’s Test for homogeneity of variance ($p > .05$), with observed skew values within an acceptable range (±1.96). K-S tests further indicated normal distribution of data for PCS Total score, and subscales Fear of Pain for the FOPQ-P, and Rumination for the PCS. However, data violated this parametric assumption for headache, migraine, stomach-ache, menstrual pain, muscle pain, and sore throat pain intensity ratings, FOPQ-P Total score, and remaining PCS and FOPQ-P subscales. ($p < .05$). Therefore, independent t-tests were used in analysing dental pain intensity ratings, PCS Total score, and subscales Fear of Pain for the FOPQ-P,

and Rumination for the PCS, whilst Mann-Whitney U tests were used in analysing remaining pain intensity ratings, FOPQ-P Total score, and remaining PCS and FOPQ-P subscales.

5.2.5.2 Qualitative Analysis

Open-ended question responses were extracted and categorised by Autistic vs. non-Autistic in Microsoft Excel for qualitative content analysis (Hsieh & Shannon, 2005). Responses were independently coded to identify reasoning for not disclosing pain or diagnosis before being categorised into meaningful clusters to provide an overarching theme. Clusters were then integrated with quantitative disclosure findings to provide context to disclosure reasonings. These clusters are presented after quantitative data are described, for example, where X% of CYP do not disclose pain, these themes were identified to suggest why.

5.3 Results

As a total sample (*n* = 144), caregivers identified their gender as either male (*n* = 40), female (*n* = 102), non-binary (*n* = 1), or not disclosed (*n* = 1), and ranged in age between 22 and 68 years (M_{age} = 42.26, *SD* = 6.43). At time of participation, 71.50% did not suspect they were Autistic (*n* = 103), 13.20% suspected they were Autistic but were not on a diagnostic pathway (*n* = 19), 5.60% suspected they were Autistic and were on diagnostic pathway (*n* = 8), 9% were diagnosed Autistic (*n* = 13), and 0.70% identified as other (*n* = 1). Subgroup demographics for caregivers of Autistic and non-Autistic CYP can be found in Table 20. In response to their CYP's pain, caregivers of Autistic CYP were less likely to catastrophize (*M* = 18.42, *SD* = 1.50), and display pain-related fears or avoidance of behaviours (*M* = 28.17, *SD* = 1.19) than non-Autistic caregivers (*M* = 19.61, *SD* = 1.36, and *M* = 32.13, *SD* = 1.97, respectively). However, there were no significant differences between groups across any psychological total, or subscale score; PCS total score, *t*(141) = -0.58, *p* = .561, and POFPQ total score, *U* = 2856, *p* = .183. Therefore, qualitative reports are not perceived to be influenced by these psychological factors or dissimilar in comparison.

Excluding missing data for age due to technical error (n = 10), caregiver responses were contextualised to CYP aged between 11 and 15 years (M_{age} = 12.80, SD = 1.26). Excluding missing data for identifying gender due to technical error (n = 11), CYP were described to have identified as male (n = 68), female (n = 60), non-binary (n = 3), gender-fluid (n = 1), or not disclosed (n = 1). Subgroup demographics for Autistic and non-Autistic CYP can be found in Table 21.

Pain frequency ratings can be found in Table 22, with Autistic CYP experiencing all described common childhood pains more frequently than non-Autistic CYP except for sore-throat. A significant association was observed between diagnosis and frequency of headaches (X^2(5, N = 144) = 29.15, p < .001), migraines (Fisher-Freeman-Halton Exact Test, p = .034), stomach-aches (X^2(5, N = 144) = 14.03, p = .015) and muscle pain (X^2(5, N = 144) = 21.17, p < .001). This suggests a CYP's diagnosis may have influenced how frequently they experienced these childhood pains. However, no significant associations were observed between diagnosis and frequency of dental pain (Fisher-Freeman-Halton Exact Test, p = .365), menstrual pain (Fisher-Freeman-Halton Exact Test, p = .054), and sore throat (Fisher-Freeman-Halton Exact Test, p = .851) which suggests frequency of these childhood pains do not significantly differ between diagnostic groups.

Pain intensity ratings can be found in Table 23; values do not include those described as "other" or "never". Autistic CYP were perceived to experience headaches (U = 730.00, p < .001), migraines (U = 62.50, p = .03) and muscle pain (p = .016) at a significantly higher pain intensity. However marginally higher stomach-ache pain ratings in the Autistic CYP did not significantly differ from non-Autistic CYP (U = 1001.50, p = .439). Similarly, no group differences in pain intensity ratings for dental pain (t(29) = -0.45, p = .654), menstrual pain (U = 218.50, p = .215) and sore-throat (U = 463.00, p = .704) were observed. This suggests Autistic CYP may find some common childhood pains more intense that non-Autistic CYP, however this is dependent on pain type.

Table 20

Caregiver Demographic Information Grouped by Whether Participants Were the Caregiver for an Autistic or Non-Autistic CYP

Demographic	Caregiver of Autistic CYP (*n* = 64)	Caregiver of Non-Autistic CYP (*n* = 80)
Age (years)		
M	42.84	41.80
SD	7.23	5.71
Identifying Gender (*n*)		
Male	9	31
Female	55	47
Non-Binary	0	1
Not Disclosed	0	1
Autism Diagnosis (*n)*		
Not Autistic	29	74
Suspect Autistic, Not Diagnostic Pathway	15	4
Suspect Autistic, On Diagnostic Pathway	7	1
Diagnosed Autistic	12	1
Other	1	-

Note. This table describes the demographic information provided by each participating caregiver group.

Table 21

CYP Demographic Information Grouped by Whether Participants Were an Autistic or Non-Autistic CYP

Demographic	Autistic CYP (*n* = 64)	Non-Autistic CYP (*n* = 80)
Age[a] (years)		
M	13.11	12.58
SD	1.36	1.15
Age at Autism Diagnosis (years)		
M	9.56	-
SD	3.58	-
Identifying Gender[b] (*n*)		
Male	28	40
Female	25	35
Non-Binary	2	1
Gender-Fluid	1	0
Not Disclosed	0	1

Notes. This table describes the demographic information provided by each participating caregiver group on behalf of their CYP.

[a] Data missing for 8 Autistic and 2 non-Autistic CYP.

[b] Data missing for 8 Autistic and 3 non-Autistic CYP.

Table 22

The Frequency Each Group of CYP Were Reported by their Caregiver to Experience Common Childhood Pains

Pain Frequency	Headache		Migraine		Stomach-Ache		Dental Pain		Menstrual Pain		Muscle Pain		Sore Throat	
	Autistic CYP	Non-Autistic CYP	Autistic CYP	Non-Autistic CYP	Autistic CYP	Non-Autistic CYP	Autistic	Non-Autistic CYP	Autistic CYP[a]	Non-Autistic CYP[b]	Autistic	Non-Autistic CYP	Autistic	Non-Autistic CYP
Everyday	0%	1.25%	0%	0%	6.25%	1.25%	0%	0%	0%	0%	12.50%	0%	0%	0%
More Than Once a Week, But not Everyday	26.56%	0%	6.25%	1.25%	17.19%	3.75%	6.25%	1.25%	0%	0%	18.75%	5%	3.13%	3.75%
Once a Week	15.63%	10%	6.25%	1.25%	17.19%	10%	0%	0%	5.41%	0%	18.75%	16.25%	3.13%	1.25%
Once a Month	31.25%	52.50%	23.44%	13.75%	29.69%	46.25%	20.31%	16.25%	59.46%	46%	25%	40%	37.50%	38.75%
Never	12.50%	23.75%	56.25%	78.75%	18.75%	22.50%	67.19%	76.25%	24.32%	48%	20.31%	27.50%	29.69%	35%
Other	14.10%	12.50%	7.81%	5%	10.94%	16.25%	6.25%	6.25%	10.81%	6%	4.69%	11.25%	26.56%	21.25%

Notes. This table outlines how frequent CYP experience each of the provided common childhood pains as reported by their caregiver. Unless specified, *n* values for each subgroup are as follows: Autistic CYP (*n* = 64) and non-Autistic CYP (*n* = 80)

[a] Number of Autistic CYP who menstruate (*n* = 37)

[b] Number of non-Autistic CYP who menstruate (*n* = 50)

Table 23

The Mean Pain Intensity Each Group of CYP Were Reported by their Caregiver to Rate Common Childhood Pains

Pain Type	Autistic CYP	Non-Autistic CYP
Headache		
n	47	51
M	6.01*	4.57*
SD	1.88	2.00
Migraine		
n	23	13
M	7.24**	5.31**
SD	1.72	1.64
Stomach-Ache		
n	45	49
M	5.41	5.02
SD	1.91	1.81
Dental Pain		
n	17	14
M	4.24	4.62
SD	2.31	2.39

Menstrual Pain		
n	24	23
M	5.39	4.67
SD	1.88	1.92
Muscle Pain		
n	48	49
M	4.97***	3.89***
SD	2.22	1.66
Sore Throat		
n	28	35
M	4.50	4.59
SD	2.02	1.58

Notes. This table outlines the mean pain intensity CYP experience for each of the provided common childhood pains as reported by their caregiver. Values provided for "Other" were not included. Unless specified, *n* values for each subgroup are as follows: Autistic CYP (*n* = 64) and non-Autistic CYP (*n* = 80)

* $p < .001$. ** $p = .03$. *** $p = .016$.

5.3.1 Pain Disclosure Approach

Below quantitative and qualitative data are synthesised to describe if caregivers predict their CYP would disclose pain to a caregiver, teacher, or HCP. In discussing method of pain disclosure, it can be assumed both Autistic CYP (*n* = 41, excluding missing data, *n* = 21, and no responses, *n* = 2), and non-Autistic CYP (*n* = 74, excluding missing data, *n* = 4, and no responses, *n* = 2) are comparable with a predominant use of verbal communication. However, factors which influence use of verbal communication differed between groups in social vs emotional context, with Autistic CYP highlighted as more susceptible to changing their communication style, an apparent theme throughout the discussed interpersonal relationships. For example, emotional factors appeared to influence non-Autistic CYP's patterns of verbalisation, with 2.70% of verbal non-Autistic CYP (*n* = 2) described to only verbalise when in a specific emotional state (i.e. not feeling shy or stressed). In comparison social factors appeared to influence Autistic CYP's patterns of verbalisation, with 31.71% of verbal Autistic CYP (*n* = 13) described to only verbalise when they felt comfortable with the interpersonal relationship - three of whom would become selectively mute if not. Adjustments in aiding Autistic CYP's ability to communicate were described (*n* = 5), these included: messaging applications (*n* = 3), Augmentative and Alternative Communication (AAC) (*n* = 1); one participant did not specify.

Overall, CYP disclosed pain more to a caregiver (93.06%; *n* = 134), than an HCP (86.81%; *n* = 125), or scholastic staff (71.94%; *n* = 100). However, in considering individual relationships differences in disclosure patterns emerge. These between group differences are discussed in detail below.

5.3.2 Caregiver Disclosure

A Fisher's Exact Test was used to determine whether there was a significant association between diagnostic group and likeliness to disclose pain to their caregiver (see Table 24). No significant differences were observed from the results of the Fisher's Exact Test, *p* = .109, suggesting this group of Autistic CYP (89.06%; *n* = 57) are as likely as their non-Autistic peers (96.25%; *n* = 77) to disclose pain to a caregiver.

For the CYP who were reported to disclose pain to a caregiver, observed differences in preference for disclosing pain to a specified caregiver are apparent; data is missing from 2 Autistic CYP whom omitted answering (*n* = 55; see Table 25). Autistic CYP were reportedly more likely to disclose their pain to a maternal caregiver (69.09%; *n* = 38) than their non-Autistic peers (49.35%; *n* = 38). In comparison, non-Autistic CYP were more likely to not show a preference in whom they disclosed their pain to between two maternal and/or paternal caregivers (38.96%; *n* = 30), than Autistic peers (21.81%; *n* = 12). These findings suggest Autistic CYP may value a certain caregiver relationship in their seeking behaviour than non-Autistic CYP; Autistic CYP showing preference towards a maternal caregiver, and non-Autistic CYP displaying neutrality in caregiver disclosure decisions.

For the few Autistic (10.93%; *n* = 7) and non-Autistic CYP (3.75%; *n* = 3) who were described to not disclose pain to a caregiver, one interrelated theme suggesting why was identified: Communicative Incongruence.

5.3.2.1 Communicative Incongruence

In the context of disclosing to a caregiver, communicative incongruence refers to an Autistic CYP's ability to interpret bodily sensations of pain. Caregivers reference differences in interoceptive ability, reporting that their Autistic CYP's pain *"just doesn't occur to him"* (A46), or that their likelihood to *"routinely ignore her body"* (A64) further limits pain recognition. Here, a lack of pain disclosure continues to reflect an incongruence in Autistic CYP's interpretation of their bodily sensation as opposed to consciously deciding to not disclose pain as explained by A30:

> *"I think it is due to problems she has in understanding what her body is telling her - for example, what is pain, what is discomfort, what is excruciating. It is all confused and just distressing, so she literally is unable to explain what is wrong."*

However, other caregivers suggest Autistic CYP do interpret their bodily sensations of pain, instead waiting until a higher severity to disclose their pain and access support as explained by A47, providing incongruence as to the role of interoception:

“He seems to accept whatever happens to him as normal and therefore accepts pain as part of life unless he really can't cope with it.”

Similarly, non-Autistic CYP were reported to display some level of communicative incongruence. For example, one caregiver describes that their CYP’s decision to disclose pain is dependent on the pain type; although this determines which caregiver the CYP will disclose to as opposed to if they will disclose, “*she tells mom first about menstrual pain*” (NA13). However as only 3 non-Autistic CYP were reported to not disclose pain, further supporting evidence as to why cannot be provided.

Table 24

Contingency Table Displaying Values of CYP's Pain Disclosure to a Caregiver Within and Between Each Diagnostic Group

Disclosure Decision[a]		**Autistic CYP (*n* = 64)**	**Non-Autistic CYP (*n* = 80)**
Would Disclose to a Caregiver	*n*	57	77
	%	89.06%	96.25%
Would Not Disclose to a Caregiver	*n*	7	3
	%	10.94%	3.75%

Notes. This table outlines how many CYP would disclose their pain to a caregiver, and how many would not.

[a] Missing data for 2 Autistic CYP.

Table 25

Number of CYP per Diagnostic Group Who Would Disclose Their Pain to a Maternal and/or Paternal Caregiver(s)

Disclosed To		Autistic CYP (*n* = 55)	Non-Autistic CYP (*n* = 77)
Maternal Caregiver	*n*	38	38
	%	69.09%	49.35%
Paternal Caregiver	*n*	5	9
	%	9.09%	11.69%
Both Caregivers	*n*	12	30
	%	21.81%	38.96%

Note. This table describes which caregiver CYP would disclose their pain to, if they were perceived to disclose their pain to a caregiver in Table 24.

5.3.3 Teacher Disclosure

A 2x2 contingency Chi-Square Test of Independence was used to determine whether there was a significant difference between diagnostic group and likeliness to disclose pain to a teacher (see Table 26). Data for home-educated CYP were removed (n = 5), leaving a total of 60 Autistic CYP and 79 non-Autistic CYP in the analysis.

A significant association between diagnosis and likeliness to disclose pain to a teacher's was observed, $X^2(1, N = 139) = 18.11$, $p < .001$. Whilst at group level all CYP were more likely to disclose their pain in a scholastic setting than not, interpretation of adjusted residual values (4.3) suggested non-Autistic CYP were significantly more likely to disclose pain (86.08%; n = 68) than their Autistic peers (53.33%; n = 32). Therefore, Autistic CYP may experience more barriers in their ability to disclose pain scholastically than their non-Autistic peers.

A significant association between diagnosis and who a CYP would disclose their pain to was observed using a Fishers-Freeman-Halton-Exact Test, p <.001. Of the available scholastic staff members, both Autistic (43.75%; n = 14), and non-Autistic (94.12%; n = 64) CYP were most likely to disclose their pain to a class teacher (see Table 27). Yet interpretation of adjusted residuals suggested Autistic CYP were more likely to tell both a class teacher and SEN (4.1), whilst non-Autistic CYP were more likely to tell a class teacher (5.7). However, a significant association between diagnosis and access to an SEN was observed, $X^2(1, N = 98) = 29.23$, $p < .001$, which could suggest decisions to disclose to specific scholastic members of staff reflects a difference in SEN access (see Table 28). Interpretations of adjusted residual values further support this suggestion (5.4), with Autistic CYP being significantly more likely to have access to an SEN (66.67%; n = 26) than non-Autistic CYP (13.56%; n = 8). Thus, this observed pattern in which scholastic member of staff may only highlight that non-Autistic CYP have no option other than to tell a class teacher, whilst Autistic CYP are able to differentiate between either.

For Autistic (46.67%; n = 28) and non-Autistic CYP (13.92%; n = 11) who were described to not disclose pain in a scholastic setting, two themes suggesting why were identified: A Difference in Negative Experiences and Communicative Expectations.

5.3.3.1 A Difference in Negative Experiences

For both groups, expectations of pain dismissal in a social setting limit intent to disclose pain. Although experiences of being dismissed were reported more often for Autistic CYP, both groups' expectations were consequential to previous negative experiences of disclosing pain which have led them to believe their teacher will not appraise their pain:

"Do not care" (NA52),

"They're dismissive, gas light him, and are mostly unavailable" (A6),

"Does not think they would believe her" (A23),

"Don't take her seriously" (A31),

"Would think they won't care, or listen" (A48).

Additional factors reportedly contribute to creating negative scholastic experiences. Non-Autistic CYP were reported to not disclose pain for both social and emotive reasons such as they *"do not like to draw attention"* (NA50) to themselves or feeling *"timid"* (NA26) in disclosing. However, Autistic CYP were described to be predominantly exposed to a negative social experience. For example, pain disclosure provides unwanted social attention from interrupting a lesson to make a *"fuss"* (A20 and A58). The desire to not be socially perceived by others, and have bystander's focus upon a personal, and subjective experience fundamentally prevents pain disclosure:

"When we've asked the usual responses we get are "I didn't want to interrupt" or "I didn't want to draw attention to myself" (A36),

"Would not want to draw attention to themselves." (A48).

Autistic CYP were also reported to experience social barriers to communicating verbally, instead choosing not to disclose their pain as they *"do not feel confident"*, *"get nervous"* or are *"scared"* when needing to speak up in class. In the classroom, interpersonal relationships are not limited to the CYP and teacher; classmates act as bystanders and

provide additional social expectations of how pain will be socially perceived which facilitate an environment where disclosure is prevented.

5.3.3.2 Communicative Expectations

Expectations that an individual in pain would initiate the disclosure process means Autistic CYP's pain can be overlooked in scholastic settings. Here, caregivers explain Autistic CYP do not want to *"start"* (A2) or *"initiate"* (A3) the conversation and thus will not be forthcoming in their pain experience. Like Chapter 5.3.3.1, social factors influence this ability with factors such as fear, *"scared to talk to other people"* (A50), and trust, *"selective with who he communicates with and what he communicates about"* (A49), impacting the likelihood for pain communication. Yet for some Autistic CYP the means of verbally expressing pain are not applicable as they are *"often mute"* (A1), have *"difficulty with verbal communication"* (A19), or interoceptive recognition *"not occurred"* (A28). Thus, if the onus is placed solely on the Autistic CYP to facilitate pain disclosure, their pain could potentially be mismanaged in scholastic settings. Instead, Autistic CYP will directly contact the interpersonal relationship where they feel ease in being forthcoming: their caregiver: *"She would ring me as won't tell school"* (A24).

Whilst these communicative expectations were not reported for non-Autistic CYP, both groups prevented the expression of pain to bystanders. Masking behaviours are often reported in Autistic CYP, with extensions here to the context of pain being hidden *"until they are home"* (A21). Interestingly, non-Autistic CYP were reported to display similar behaviours and wait until they were in a safe environment to express their pain, a likely consequence to the experiences of pain dismissal:

> *"They would wait until they get home"* (NA34),
>
> *"He'd wait till he was at home"* (NA76).

Table 26

Contingency Table Displaying Values of CYP's Pain Disclosure to a Teacher Within and Between Each Diagnostic Group

Disclosure Decision[a]		**Autistic CYP (*n* = 60)**	**Non-Autistic CYP (*n* = 79)**
Would Disclose to a Teacher	*n*	32	68*
	%	53.33%	86.08%
Would Not Disclose to a Teacher	*n*	28*	11*
	%	46.67%	13.92%

Notes. This table outlines how many CYP would disclose their pain to a teacher, and how many would not.

[a] Missing data for 5 home-educated CYP.

* Adjusted standardised residual $p < .001$.

Table 27

Number of CYP per Diagnostic Group Who Would Disclose Their Pain to a Teacher and/or SEN, or a Different Member of Staff

Disclosed To		Autistic CYP (*n* = 32)	Non-Autistic CYP (*n* = 68)
Class Teacher	*n*	14	64*
	%	43.75%	94.12%
SEN	*n*	5	0
	%	15.63%	0%
Both Class Teacher and SEN	*n*	9*	1
	%	28.13%	1.47%
Other	*n*	4[a]	3[b]
	%	12.50%	4.41%

Notes. This table describes which scholastic member of staff CYP would disclose their pain to, if they were perceived to disclose their pain to a scholastic member of staff in Table 26.

[a] Disclosed as a separate teaching assistant (*n* = 2), or the school office (*n* = 2).

[b] Disclosed as the scholastic medical team (*n* = 2), or friends (*n* = 2).

* *p* <.001

Table 28

Number of CYP Reported to have Access to an SEN Tutor

Access to a SEN?[ab]		Autistic CYP[c] (*n* = 39)	Non-Autistic CYP[d] (*n* = 59)
Yes	*n*	26*	8
	%	66.67%	13.56%
No	*n*	13	51*
	%	33.33%	86.44%

Notes. The table outlines how many CYP have access to an SEN.

[a] Missing data for 20 CYP due to technical error.

[b] Missing data for 3 CYP due to no response.

[c] Excluding 1 don't know responses.

[d] Excluding 17 don't know responses.

* $p < .001$.

5.3.4 General Practitioner Disclosure

A 2x2 contingency Chi-Square Test of Independence was used to determine whether there was a significant difference between Autistic and non-Autistic CYP's likeliness to disclose pain to an HCP (see Table 29). A significant difference between diagnosis and likeliness to disclose pain to an HCP was observed, $X^2(1, N = 144) = 27.36$, $p < .001$. Interpretation of adjusted residual values (5.2) suggested non-Autistic CYP were significantly more likely to disclose pain to an HCP (100%; $n = 80$), than Autistic CYP (70.31%; $n = 45$). Therefore, Autistic CYP may experience more barriers in their ability to disclose pain medically than their non-Autistic peers. This difference was not significantly impacted by differences in method of interacting with an HCP as observed using a Fisher-Freeman-Halton Exact Test, $p = .099$, with face-to-face remaining the most common engagement method for both groups (see Table 30).

Despite 78.38% of CYP whose caregiver do disclose their Autism diagnosis ($n = 37$) also deciding to disclose pain to an HCP ($n = 29$), providing HCPs with Autism diagnostic awareness did not significantly impact CYP's pain disclosure (see Table 31) as observed using a Fisher-Freeman-Halton Exact Test, $p = .114$. Still, disclosing an Autism diagnosis was described to be important for improving the healthcare experience as discussed as a theme below: Disclosing an Autism Diagnosis (see Chapter 5.3.4.1). Therefore, despite not observing a significant interaction in decisions for pain disclosure, Autism diagnostic disclosure may facilitate a positive environment which allow the majority of Autistic CYP (70.31%; $n = 45$) to disclose pain. For Autistic CYP whom would not disclose pain (29.61%; $n = 19$), two additional themes suggesting why were identified: To Be Socially Perceived (Chapter 5.3.4.2) and Communicative Incongruence (Chapter 5.3.4.3).

5.3.4.1 Disclosing an Autism Diagnosis

Caregivers reported that providing this information created diagnostic recognition of Autism, whereby they could affirm HCPs knew about the entirety of their CYP's health, sensory and interoceptive differences that co-occur:

"To enable proper diagnosis and medical history" (A37)

“So that the doctor can know my child's health history” (A57)

“In case whatever is ailing my child is related to his autism and also so they are aware of his sensory issues.” (A7)

Additionally diagnostic disclosure ensured HCPs were able to interpret differences in CYP’s communication styles, with caregivers emphasising that without this information misinterpretation could cause Autistic CYP to not receive the correct level of pain management. These communication styles include both verbal, *“Because she has sensory difficulties and often is not able to explain how she is feeling physically and how or where she is experiencing pain”* (A31), and behavioural differences, *“To help them understand her behaviour at the app”* (A38). However, caregivers expected this disclosure to be met with a change in HCPs therapeutic approach, shifting towards a neurodiversity-affirming healthcare as demonstrated by A9 who highlights the expectation:

“My son's communication and interactions are noticeably different. It helps health care professionals understand his behaviour in appointments and adjust accordingly.”.

This point is further reinforced by A62 who clearly states the level of information they provide, and outline what adaptations for a neurodiversity-affirming approach are expected to facilitate the HCP interaction:

“I share the diagnosis when I make the appointment and give a brief overview, as autism can take many different forms. I make sure that it is known that our son is verbal but has problems with unclear, imprecise and long statements. I also let him know that touching should be announced and that our son may not be able to tolerate it.”.

5.3.4.2 To Be Socially Perceived

As a method of protection from emotions associated with a bystander’s social perception, some Autistic CYP were described to refrain from disclosing pain to a HCP to prevent feelings of discomfort arising from the specific social interaction. For example,

caregivers suggested feelings of nervousness, *"wait for me to speak to the doctor as nervous"* (A28) , or embarrassment, *"clam up when talking to Drs/feel embarrassed"* (A17).

The cause of this discomfort appears to be consequential to a need for a relationship to disclose pain, with GPs being referred to as a *"stranger".* This suggests if CYP were to see their GP more frequently, they may feel more at ease to disclose pain: *"She would behave differently with a stranger, and she rarely needs to go to the GP so they aren't familiar."* (A10). However similar reasoning as those outlined in Chapter 4.3.2.3 are apparent as previous social experiences acted as an emotional predictor for Autistic CYP's current healthcare decisions. For example, caregivers report their CYP *"don't trust the GP to make it better"* (A29), with previous experience of pain dismissals contributing to this reasoning as *"previously doctors didn't believe level of pain."* (A21). Some Autistic CYP appear to rely on their caregiver to advocate pain on their behalf. Mitigating the need to self-disclose pain, many CYP want their caregiver to "*talk-*" or "*speak for them"* however these behaviours are consequential to the previously described discomfort HCP-interactions have promoted:

> *"They would wait for me to speak to the doctor as nervous"* (A28)
>
> *"They would want me to do this for them"* (A42)
>
> *"Often they want me to talk for them as they are not comfortable with the doctors"* (A48)

Although, these reports do not always reference previous healthcare experiences, general past experiences are still described as a prevention from disclosure. This suggests a societal change towards creating inclusive social environments which promote positive experiences of pain disclosure, rather than reinforcing a learnt negative social consequence are paramount:

> *"Our child has a large fear of being "bothersome." They often feel like if they speak up people will become annoyed or that they'll be a burden. Even when it's a person specifically there to help them."* (A36)

5.3.4.3 Communicative Incongruence

Many Autistic CYP were described to experience communicative incongruence like those outlined in Chapter 5.3.2.1. For example, difficulties in interoception prevent pain recognition for pain disclosure as explained by both A30 "*she would be unable to understand her pain*", and A9 "*he has difficulties with interoception and so reduced awareness of his body*". This suggests their CYP would not interoceptively process their pain, which without this recognition would prevent the ability in disclosing to an HCP; however, whether their caregiver recognises to allow them attending the GP is unclear.

Further communicative incongruence was observed between the social dynamic of caregiver-CYP, whereby difficulties in providing the gold standard verbalisation of pain prevented Autistic CYP from being able to disclose. For example, caregivers described their Autistic CYP experienced difficulties in different facets of verbal communication, including the ability to find the language, "*they would try but would struggle to find language to express their pain in a way the Dr would understand*" (A6), requirement of directed questions "*only if asked specifically*" (A32), and limited expression "*finds it hard to express also doesn't show pain in expected way*" (A21). Despite most caregivers explaining they disclose their CYP's Autism diagnosis to mitigate these difficulties expecting HCPs to adapt to a neurodiversity-affirming approach, this insight suggests HCPs often do not place this approach in practice, or perhaps do not understand how to.

Table 29

Contingency Table Displaying Values of CYP's Pain Disclosure to an HCP Within and Between Each Diagnostic Group

Disclosure Decision		Autistic CYP (*n* = 64)	Non-Autistic CYP (*n* = 80)
Would Disclose to an HCP	*n*	45	80*
	%	70.31%	100%
Would Not Disclose to an HCP	*n*	19*	0
	%	29.69%	0%

Notes. This table outlines how many CYP would disclose their pain to an HCP, and how many would not.

* $p < .001$.

Table 30

CYP's Most Frequent Method of Communication with HCPs

Disclosure Decision		Autistic CYP (*n* = 64)	Non-Autistic CYP (*n* = 80)
Face-to-Face	*n*	43	63
	%	67.19%	78.75%
Telephone	*n*	19	12
	%	29.69%	15%
Online Chat	*n*	2	5
	%	3.13%	6.25%

Notes. This table outlines which method of communication CYP most frequently use.

Table 31

All Caregiver Decisions in Disclosing their CYP's Autism Diagnosis to an HCP

Autism Disclosure Decision		Will Disclose Pain to HCP	Will Not Disclose Pain to HCP
Always Disclose to an HCP	*n*	29	8
	%	45.31%	12.50%
Sometimes Disclose to an HCP	*n*	13	8
	%	20.31%	12.50%
Never Disclose to an HCP	*n*	0	1
	%	0%	1.56%
Other	*n*	2	0
	%	3.13%	0%
Don't Know	*n*	1	2
	%	1.56%	3.13%

Note. This table describes how often a caregiver discloses their CYP's Autism diagnosis, dependent on their pain disclosure response in Table 29.

5.4 Discussion

In Chapter 4, mistrust, disbelief of symptoms and negative experiences were identified as barriers to Autistic CYP disclosing pain in certain interpersonal relationships. To develop this understanding further, this study utilised the perspectives of caregivers to explore decisions and differences for Autistic and non-Autistic CYP's pain disclosure in specified interpersonal relationships. Interestingly across all relationships, caregivers perceived that both Autistic and non-Autistic CYP were more likely to disclose their pain than not suggesting Autistic CYP are seeking support in accessing pain relief. However, in considering specific interpersonal relationships, caregivers identified clear experiential differences to suggest these decisions were dependent on the interpersonal relationship. Similar themes pertaining relationships with caregivers, teachers and HCPs as those in Chapter 4 were identified, with factors like distrust being evident amongst Autistic and non-Autistic CYP. Additional consideration of how interoception, and communicative incongruence impact decisions were identified by caregivers to Autistic CYP, but not non-Autistic CYP. How these patterns in disclosure differ across relationships and their associated reasoning are discussed further below.

Of all the specified relationships, caregivers suggested Autistic CYP were more likely to disclose pain to their caregiver. Caregivers reported differences in interoception can affect their Autistic CYP's ability to disclose pain, emphasising that a lack disclosure is consequential to difficulties in attending to internal sensations rather than a conscious intent. This explanation reflects available literature which highlights interoception as a common difficulty amongst Autistic individuals, with reports of not recognising cues of thirst, hunger, or in this example, pain (Dubois et al., 2016). However, consistent with Chapter 4.3.2.1, caregivers suggested some Autistic CYP recognise their internal sensations of pain but only disclosed to seek support when experiencing a higher pain severity. This continued discordance in understanding the role of interoception in disclosing pain creates a complex debate of Autistic CYP's interoceptive abilities. However, with the provided clarity that interpersonal factors like distrust or disbelief do not influence pain disclosure to caregivers, a focus on the role of intraindividual factors like a CYP's anxiety levels may provide required insight.

As expected, caregivers highlighted Autistic CYP continued to face barriers in disclosing pain to HCPs and teachers with mistrust and dismissal of symptoms remaining a key factor, yet a prominence of communicative incongruence emerged as an addition. An emphasis for Autistic CYP to amend their communication style to appease the expectations of teachers and HCPs created difficulty for disclosure. For example, HCPs reportedly relied on CYP to continually verbalise their pain and teachers reportedly relied on CYP to initiate these conversations. Existing literature and these discussed interactions with caregivers highlight Autistic CYP can verbalise their pain, however the lack of trust and experience of dismissal likely influences their ability to be forthcoming with this information (Bandstra et al., 2012; Ely et al., 2016; Fitzpatrick et al., 2022). Instead, a collaborative effort should be made between Autistic CYP and their teacher or HCP to understand how changes in behaviour can be used for disclosure rather than placing the onus on Autistic CYP to provide verbalisations and experience consequential distress. Yet as bystander interpretations of Autistic CYP's non-verbal behavioural or expressive cues can be incongruent to their internal state (Noyek et al., 2023), a general need for educating professionals on neurodivergent pain communication is vital.

Barriers to disclosing pain were most prevalent in scholastic settings, with 46.67% of Autistic CYP being reported by their caregiver to refrain from disclosing. When disclosing pain at school, Chapter 4 contextualised findings primarily to interactions between the CYP and their teacher. However, to prevent peers socially perceiving their experience of pain, Autistic CYP were reported to not disclose their pain in class settings. The contextualisation to class is important, here isolated one-to-one interactions between teacher and CYP rarely exist, instead multiple interpersonal relationships interact with the presence of additional pupils. For these Autistic CYP, the concept of being negatively evaluated by peers could be distressing due to the adolescent age group, or potential co-occurring social anxiety (Caouette & Guyer, 2014; Somerville, 2013). Although social anxiety was not measured as a self-report or by-proxy, with 50% of the Autistic population experiencing social anxiety could be an influential factor (Maddox & White, 2015; Spain et al., 2018). Therefore, whilst the listed interpersonal factors may limit disclosure to teachers, the additional interpretation and dismissal of public disclosure to peers could exacerbate any distress and decrease

likelihood of disclosure further. Thus, continuing to consider scholastic interpersonal factors in duality may undermine the social norms and developmental perspectives at play.

Interestingly, despite only 13.92% of non-Autistic CYP being reported by their caregiver to refrain from pain disclosure, negative social experiences were identified as a barrier. To mitigate social attention directed towards them and emotional distress from scholastic disbelief, non-Autistic CYP were suggested to mask their pain until they were home like Autistic CYP in Chapter 4.3.2.3. Although literature suggests their symptoms of pain are typically disbelieved in healthcare settings (Carter, 2002; Defenderfer et al., 2018; Newton et al., 2013; Wakefield et al., 2021), consistency in all non-Autistic CYP disclosing pain to HCPs suggests this interpersonal factor is more influential to scholastic experiences. As a means of improving these scholastic experiences, a toolkit like that of The School Toolkit for EDS and JHS (2021) may be advantageous. The School Toolkit for EDS and JHS (2021) provides guidance of how schools can better understand, and support CYP with JHS or EDS including in their experiences of pain. Adapting this approach specifically for CYP to disclose pain by equipping teachers with an understanding of how to facilitate an environment that promotes pain disclosure, and how peers can better support one another may mitigate the demands of interpersonal relationships. Yet, more research would be required to create a valid, and practical set of guidance from both an Autistic and non-Autistic CYP's perspective.

Caregivers additionally provided information regarding the frequency and intensity of common childhood pains. Autistic CYP were perceived by their caregiver to experience stomach-aches, headaches, migraines, and muscle pain more frequently, and to rate the latter three pains more intensely than their non-Autistic peers. This pattern is unsurprising given existing evidence has established that these pain types frequently co-occur within the Autistic population (Chakraborty et al., 2021; Pan et al., 2021; Whitney & Shapiro, 2019). However, as only these common childhood pains were perceived to be more frequent and intense for the Autistic population, it could be inferred Autistic CYP may appear more frequently in specific pain clinics, rather than all. Two clinical approaches could be implemented from this inference: 1) screen all CYP whom appear at headache, gastrointestinal or muscular pain clinics for an Autism diagnosis to facilitate adaptations to

the treatment approach they receive as Donaghy et al. (2023) suggests, and 2) create a clinical pathway which refers Autistic CYP who present with these symptoms to a neurodiversity-affirming clinic. However, further research is required to identify which approach would be most beneficial to Autistic CYP, and cost-effective for the healthcare services. In contrast, menstrual pain was not perceived to be more frequent for Autistic CYP despite evidence the Autistic population may experience dysmenorrhea (Ingudomnukul et al., 2007; Pohl et al., 2014). However future research should aim to develop this insight further as this finding may be related to the infrequency of the populations cycle (Harley et al., 2024), or an overshadowing of symptoms that co-occur with the menstrual cycle like headaches (Chaudhary, 2021).

However, these findings do not represent a universal experience in disclosing pain as the validity in these disclosure findings may be skewed through use of caregiver proxy-report. For example, in asking if a CYP always discloses pain to the caregiver, data pertaining when a CYP "does not disclose pain" instead may infer that the caregiver has observed their CYP is in pain, and recognised their CYP has not told them. Additionally, given the time intensive nature of this survey participating caregivers may have been more likely to have a good familial relationship at home whereby they felt more comfortable in answering questions about their CYP. In turn this could have increased the frequency of CYP who were perceived to disclose pain to their caregiver as this good familial relationship was present. Thus, this does not capture instances where a CYP has been in pain without their caregiver knowing, or CYP who tend to divert from disclosing to a caregiver. Similarly, the validity of these proxy-reports may be limited in the context of HCPs and teachers. Caregivers of CYP may have exaggerated how often their CYP discloses pain to an HCP to omit negative perceptions that they do not provide their CYP with autonomy (Akre & Suris, 2014; Paron, 2024); particularly for caregivers of Autistic CYP as discussed within Chapter 4. Yet whilst caregiver perceptions of HCP interactions may have been more representative due to their likely presence at appointments, their perception on teacher interactions may be presumptuous and not reflect their CYP's lived experience. Therefore, whilst the observed patterns in disclosure may reflect the CYP's real-world experiences, a bias in caregiver perception may inflate or reduce the strength of these disclosure decisions.

Several limitations could be reported for Chapter 5, however most pertinent are those pertaining COVID-19 lockdowns. Throughout the duration of this thesis, COVID-19 has impacted the design and practicality of all studies, however its effects are exacerbated here. The format of this study changed multiple times consequential to the social landscape COVID-19 drove. In the limited ability to explore interpersonal factors in a laboratory setting whereby social context and the relationship of interest could have been manipulated to assess impacts on pain disclosure, social distancing mandates prevented this design. Whilst I and my supervisory team perceived an online survey as necessary to adhere to the gold standard research approach at the time, its flaws limited optimality for assessing an actively social construct in a passive manner are acknowledged. However, whilst not optimal in design, the ability to access such anonymised data may have provided an accurate insight into disclosure patterns, with caregivers potentially feeling less nervous to disclose their own perceptions of their CYP's behaviour without judgement. Future research should aim to incorporate online studies in parallel with experimental designs to understand how perceived behaviour, and observed behaviour compare.

Additionally, participant recruitment was extremely difficult. Despite best efforts to recruit including but not limited to contacting all age-appropriate schools within the area, accessing Autistica's Network of participants, and strategically posting the survey following public engagement with Autistic CYP and caregivers - recruitment was slow. To mitigate the impact of recruitment rate on the overall data, a team decision to include only caregivers was made. Whilst caregivers did not display differences in psychological components that may have decreased their comparability, arguably this decision may limit the applicability of findings, as throughout this thesis self-reported pain experiences have been provided yet in this instance proxy-reports are used for comparison. Future research should aim to gain this disclosure insight from CYP's lived experience to strengthen these findings and highlight if disparities are presence in self- vs. proxy-reports. However, a survey conducted by Harrop et al. (2021) exploring how COVID impacted early career researchers' studies involving Autistic participants suggested approximately 65% of researchers experienced recruitment issues. Whilst this implies a universal experience in recruitment difficulties amongst Autistic research, this does not mitigate the effects on data validity.

In summary, Chapter 5 develops understanding of why Autistic CYP disclose pain in certain interpersonal relationships through a caregiver perspective. Whilst Autistic CYP were considered by their caregiver to be as likely to disclose their pain to caregiver, teacher, and HCP relationships, differences in their decisions to disclose pain were observed. Mistrust and dismissal were reported by caregivers in both groups as barriers, but a focus on communicative incongruence between Autistic CYP and their teachers and HCPs appeared to strengthen decisions to not disclose. Additionally, a focus on interactions between Autistic CYP and teachers failed to appreciate the social evaluation of peers present within a classroom that reinforce decisions to not disclose. However, negative experiences of pain disclosure in the classroom were not reserved for Autistic CYP as a need to mitigate social perception was perceive by caregivers to prevent non-Autistic CYP from disclosing too. Here a continued focus of how HCPs interact with pain across all paediatric literature may be overlooking a key experience where the progression of pain is being facilitated – schooling. Although training on how HCPs should best manage Autistic CYP's pain, a requirement for scholastic understanding is required to the benefit of both Autistic, and non-Autistic CYP.

Chapter 6.

General Discussion

6 General Discussion

6.1 General Discussion

To address a knowledge gap in understanding Autistic CYP's pain, a range of methods were used to examine for low level differences in stimulus processing, as well as to explore the experiential state of pain for Autistic CYP. Within Chapter 1.4, three gaps in the Autistic paediatric literature were outlined to provide guidance for how the findings reported in this thesis could develop an understanding of Autistic CYP's pain. These included:

(1) Why pain is more common within the Autistic community,

(2) If pain anecdotes and statistics correlate to perceived pain hyper- or hyposensitivity,

(3) Which factors are important when addressing this health inequity.

Findings provide a preliminary explanation for the latter two points, with their application inferring a possible explanation for why pain is more common within the Autistic community. Below the relevance of Chapter 2 through 5's findings are discussed and positioned to create a foundation of knowledge for academics, and non-academics alike. Particularly, academics can continue to progress through Autistic co-production, and non-academics like teachers and clinicians can develop their own understanding and better support the Autistic community in their work.

6.1.1 Do pain anecdotes and statistics correlate to perceived pain hyper- or hyposensitivity?

Theoretical questioning arose in how the DSM-5's (American Psychiatric Association, 2023) assumption that Autistic CYP are hyposensitive to, or don't feel pain prevails despite medical statistics, the Autistic community, academics, and pain professionals alike advocating the opposite. Throughout this thesis, this incongruence in opinion is discussed with the aims of producing evidence that can provide a realistic understanding of Autistic CYP's pain experience. The psychophysical protocol and

subsequent quantitative evidence outlined in Chapters 2, and 3 provide insight into Autistic CYP's psychophysical experiences of pain which counters the DSM-5's (American Psychiatric Association, 2023) assumption of Autistic pain hyposensitivity.

As a quantity that bystanders cannot observe, understanding of pain can be provided by measuring pain mechanisms. Psychophysical assessment of pain threshold and tolerance are the most frequent method to quantify the relationship between a response and a stimulus magnitude (Greenspan, 2009). However, with no standardised approach for paediatric psychophysical assessments available, a systematic review of existing literature that psychophysically assessed paediatric pain was conducted to develop an ethical protocol. Previous literature predominantly applied adult parameters (Rolke et al., 2006) to a paediatric population (Blankenburg et al., 2010) as a feasible method of psychophysical assessment. However, adaptations for the inclusion of psychophysical assessments in diagnostic populations from a full battery assessment to select pain modalities were observed (see Chapter 2.3.4.3). Additionally, methodology should be adapted for safety and accessibility of CYP however ethical considerations were limited for adapting methodology, and difficult to infer for an Autistic population as only three studies were included in evidence synthesis (Duerden et al., 2015; Riquelme et al., 2016; Riquelme et al., 2023). Whilst the latter studies included equipment familiarisation prior to study commencement, and provided an explanation of instructions to aid comprehension were provided, conflicts emerged in decisions for parental presence. As a safety precaution, a recommendation for parental presence was appropriate for psychophysical studies involving Autistic CYP consistent with Riquelme et al. (2016) and Riquelme et al. (2023). Psychophysical assessments predominantly rely on verbal self-report, with evidence to suggest Autistic CYP provide similar reports to non-Autistic CYP (Benich et al., 2018; Ely et al., 2016). However, further evidence suggests a shift to non-verbal communication can be observed when distressed; an emotion that is often consequential to pain (Cummins et al., 2020; Haydon et al., 2021; Muskat et al., 2015). Thus, as a safety precaution to monitor non-verbal pain communication and report pain on the CYP's behalf when required, parental presence for Autistic CYP should be considered.

In creating protocol, ethical considerations were applied to commonly used measures of MPTh, PPTh, CPTh and CPTol in assessing Autistic pain experiences (see Chapter 3.1). In exploring differences in pain threshold, tolerance, and intensity between Autistic and non-Autistic CYP, Chapter 3's findings provide three key arguments in challenging the DSM-5's (American Psychiatric Association, 2023) current perception of pain in the Autistic population:

(1) Like in Autistic adults (Bird et al., 2010; Failla et al., 2020; Failla et al., 2017; Fründt et al., 2017; Thaler et al., 2017; Vaughan et al., 2020), sensory mechanisms involved in eliciting a pain response for Autistic CYP do not significantly differ from non-Autistic CYP,

(2) Individuals experiences of pain are subjective, with Autistic CYP displaying differences in hyper-, hypo- or no clinical sensitivity to differing pain modalities; another finding which replicated existing adult literature (Fründt et al., 2017; Vaughan et al., 2020),

(3) Autistic CYP appear to possess the neural archetype to feel pain.

Overall, no evidence was provided to support anecdotal claims of Autistic pain hyper- or hyposensitivity. Although some individuals displayed hyposensitivity to support the DSM-5's (American Psychiatric Association, 2023) claims, the collective of these experiences only highlights the subjective nature of pain which Raja et al.'s (2020) definition suggests.

By continuing to suggest through the likes of the DSM-5 (American Psychiatric Association, 2023) and ICD-11 (World Health Organization, 2022a) that Autistic individuals as a diagnostic group experience pain hyposensitivity, Autistic CYP's pain may be misinterpreted by bystanders by applying an incorrect diagnostic perception to explain an experience they often do not understand. For example, HCPs report accessing their understanding of Autism through broad personal experiences including self-directed reading, and formal learning experiences including lectures (Snow et al., 2022). As a diagnostic manual, the DSM 5 (American Psychiatric Association, 2023) may be a material accessed to develop knowledge, however if the criterion reported do not reflect the lived experiences of

Autistic CYP misconceptions may negatively shape diagnostic approach. Thus, by continuing to promote this explanation as a synonymous feature to Autistic people's sensory experience, harm will prevail in preventing an understanding of the basic sensory mechanisms involved in Autistic pain.

The DSM-5 (American Psychiatric Association, 2023) should update their diagnostic criterion to include recognition for subjective pain experiences amongst the Autistic population. Using an approach like how advances in paediatric pain literature contributed to dispelling the harmful myth that no CYP experience pain described in Chapter 1.2.3 (see Loizzo et al. (2009) and Twycross (1998)), efforts should be placed in dispelling the same myth in Autistic CYP by developing the literature to strengthen arguments for change. However, focus should shift from contributing evidence to show Autistic CYP experience pain as existing adult and paediatric literature psychophysically are ample in providing this diagnostic understanding - even if contradictory in direction of sensitivity (Bird et al., 2010; Cascio et al., 2008; Chen et al., 2017; Duerden et al., 2015; Failla et al., 2020; Failla et al., 2017; Fan et al., 2013; Fründt et al., 2017; Hoffman et al., 2023; Li et al., 2024; Riquelme et al., 2016; Riquelme et al., 2023; Thaler et al., 2017; Vaughan et al., 2020). Instead, research should focus on deciphering the differential factors that contribute to the Autistic pain experience, and how the DSM 5's (American Psychiatric Association, 2023) perceptions of pain hyposensitivity create systemic barriers for healthcare that contribute to worsening Autistic CYP's pain experiences.

6.1.2 Which factors are important when addressing the health inequity?

Pain continues to be prevalent amongst Autistic CYP, but with Chapter 3 providing no evidence for differences in experience at a sensory level, further understanding of moderating inter- and intra- social or personal factors was needed. Given the exploratory nature in identifying unspecified factors, a qualitative approach was utilised to attain an in-depth understanding of Autistic CYP's broad pain experience, rather than quantitatively measuring responses to an isolated pain.

Chapter 4 provided an initial framework for how differential psychological, cognitive and social factors shape Autistic CYP's pain. In interviewing Autistic CYP and their caregivers about daily experiences of pain, expectations of Craig's (2015) Social Communication Model of Pain were mirrored with Autistic CYP recognising interactions between their pain and cognition or affective state. For example, Autistic CYP recognised how pain affects their cognition with an exacerbation in scholastic settings, and how they mask their changing affective state dependent of who they are with. However, of greatest importance to pain experiences were the social factors which influences Autistic CYP's comfortability in expressing or disclosing pain. This insight suggested an understanding of how differing social contexts influences pain is vital to addressing Autistic CYP's pain inequity.

Like existing research identifying healthcare barriers for Autistic individuals have suggested (Doherty et al., 2021; Haydon et al., 2021; Langhinrichsen-Rohling et al., 2021; Mason et al., 2019; Walsh et al., 2023), interpersonal factors of distrust, symptom disbelief and previous negative experiences influenced Autistic CYP's ability to access pain management through verbal disclosure. These factors were only discussed in the context of HCPs and teachers, with Autistic CYP reporting their efforts to seek aid in these interactions were met with a lack of pain appraisal or assistance. Consequentially, a *"What's the Point?"* view was continually reinforced influencing their lack of intent to continue to disclose pain in the future in these relationships. However, the role of negative healthcare experiences is not nuanced, the impacts these experiences have in accessing healthcare and health outcomes are well-documented. For example, evidence suggests individuals reporting negative healthcare experiences had poorer health outcomes, lower levels of institutional trust, and not accessing healthcare support in the future (Eriksen et al., 2023; Schwei et al., 2017). Given the extent which interpersonal factors impact Autistic CYP's healthcare behaviours – social factors may be the largest contributor to the worsening of pain outcomes in Autistic CYP.

In furthering knowledge of how interpersonal factors act as a continued barrier to pain disclosure, an online survey described in Chapter 5 was used to explore additional interpersonal factors that affect Autistic CYP's decisions to disclose pain. In continuation

from Chapter 4, dismissal of Autistic CYP's pain continued to influence behavioural intent to disclose pain to HCPs and teachers. However, when interacting with HCPs, a communicative incongruence continually emerged whereby HCPs applied their understanding of neurotypical disclosure to their interaction with a neurodivergent population. For example, expecting a verbalisation of the CYP's pain without adapting to a neurodivergent approach. Understandably, HCPs are taught a standard approach for assessing pain, whereby verbal reports are deemed the gold standard for gaining qualitative insight to pain and thus may not be comfortable in differing approaches to assessment (Haefeli & Elfering, 2006; National institute for Health and Care Excellence (NICE), 2021b; Raja et al., 2020; The Royal Children's Hospital, 2022). Potentially this lack of understanding in approach may be detrimental to Autistic CYP's outcomes. However much like available paediatric literature suggests, Autistic CYP described their ability to describe pain, locate pain and answer questions regarding pain (Bandstra et al., 2012; Ely et al., 2016; Fitzpatrick et al., 2022). Thus, Autistic CYP may require a scaffolded approach being required to disclose, or even recognise the presence of pain, which a reliance on verbal self-report alone may not provide. Instead, a step-by-step process in assessing paediatric pain like Baker and Wong's (1987) "Q.U.E.S.T", and Fink's (2000) "WILDA" provides may be more beneficial for Autistic CYP. These highlight the importance of asking CYP questions about their pain to gain qualitative insight, rather than awaiting a forthcoming disclosure, and would likely provide the scaffolded approach so many Autistic CYP and caregivers emphasised. Whilst the onus is often placed on CYP to suggest the methods of pain communication develop in childhood can predict development of chronic pain in adulthood (Emerson & Bursch, 2020; Palermo et al., 2010), here a lack of HCP understanding of Autistic CYP's pain expression may be of greater concern.

In recommending adaptations for approach, HCPs and teachers must be supported in how to recognise, appraise and support Autistic CYP's pain. In response, the development of neurodiversity-affirming training for approaching pain assessment is vital. Such training would be especially beneficial to HCPs as evidence suggests HCPs recognise their lack of knowledge in managing neurodivergent experiences of pain (see Boshoff et al.'s (2021) systematic review for evidence synthesis). Similarly, the National Autistic Society

(2021) report only 39% of mainstream teachers have received more than half-a-days Autism, which suggests generally teachers' knowledge of Autistic pain experiences could be limited (Derguy et al., 2023). Training considering the neurodiversity-affirming movement is regularly developed to assist individuals in public facing roles understand Autistic experiences from a diagnostic, and healthcare perspective. For example, National Autism Trainer Programme (Evans et al., 2022) and The Oliver McGowan Mandatory Training on Learning Disability and Autism (Health Education England, 2022). However, a focused approach to pain assessment would be a beneficial addition. Examples of what training should include are regularly suggested in Chapters 4 and 5. For instance, caregivers identified that if their Autistic CYP were to be asked about their pain in a specific manner, they would find ease in disclosing their experience. Additionally, Autistic CYP described their contextualisation of current pain experiences to previous pain experiences to anchor their pain intensity; a descriptive approach that has previously been identified in Autistic CYP's use (Han et al., 2024). Methods of how HCPs and teachers can adapt their questioning and interpret provided descriptive reports are evident in Chapter 4 and 5's findings, yet as ever, further research providing qualitative understanding is needed to allow the creation of such training. However, to ensure training is representative of lived experience and possesses real world impact in its applicability, training should be co-produced with Autistic CYP to position their voice in how approaches should be adapted. Additionally, the findings presented throughout this thesis reflect the experiences of Autistic CYP with low support needs and do not reflect an array of Autistic experiences. Therefore, training should be developed from a multitude of Autistic experiences, including Autistic CYP with ID, Autistic CYP with high support needs and Autistic CYP who utilise predominant non-verbal communication.

6.1.3 Where do we go from here?

The findings of this thesis cannot explicitly explain why pain is more common within the Autistic community. However, how the implications of the discussed diagnostic misconceptions and the negative experiences pertaining Autistic CYP's health have shaped these pain outcomes may provide insight. If society continues to apply a stereotypical

understanding of Autistic pain experiences and ascribe a neurotypical understanding of pain communication for Autistic pain management, adequate treatment cannot be provided to lessen the likelihood of pain persisting. The provided suggestions for improving Autistic CYP's pain experiences may provide some support in addressing the pain health inequity (i.e. updating the DSM-5's (American Psychiatric Association, 2023) diagnostic criteria and developing training). However broader understanding, and clinical applications are required to both understand the commonality and address the pain health inequity.

This thesis provides insight into how Autistic CYP experience pain, but more so emphasises the inequities that are engrained into their daily lives. For example, Autistic CYP experiencing pain but having to mask their affective state in school to avoid social consequences or having to learn how to "get on" with their pain as self-management is easier than having to disclose. Conducting research to identify where Autistic inequities lie is useful for knowing which aspects of society need to be transformed to encompass a neurodiversity-affirming approach. However the reality is, the Autistic population experience societal stigma that contributes to many health, and social inequities throughout their lives (Trundle et al., 2023; Turnock et al., 2022). These experiential inequities do not exist in isolation; they interact to shape our expectations of future outcomes. For example, some Autistic individuals report a need to mask their authentic self to socially fit in (Chapman et al., 2022). This experience evidently translates to Autistic CYP masking their pain in school to not appear different from their peers as these findings highlighted. Another example includes Autistic individuals having expectations that they will not receive support as their Autism diagnosis and co-occurring health conditions make them complicated (Camm-Crosbie et al., 2018). This experience mirrors these findings that Autistic individuals feel as though there is no point in telling HCPs about their pain, as they won't receive the relevant support. The broader experiences the Autistic population have in how their Autism is perceived by society influences all aspects of their life; thus, wider consideration of how social perceptions impact Autistic individuals' ability to express, and experience pain is paramount.

Moreover, it is not enough to change diagnostic criteria or provide training for HCPs and teachers to engage with alone. The unfortunate reality is only those aware of the

implications of, and interested in Autism and pain are likely to engage with training (Harackiewicz et al., 2016), and the learning of pain indifference is embedded into the healthcare system. Broader clinical adaptations to the systemic structure must be implemented to better support Autistic CYP.

An important first step would be to create neurodiversity-affirming pain services specifically for Autistic CYP. These pain services should incorporate the existing sensory-based evidence (Doherty et al., 2021; Rios-Vega et al., 2024), in addition to the communicative approaches discussed in this thesis to support Autistic CYP in having their healthcare needs met. For example, HCPs taking time to build a trusting relationship with an Autistic CYP through a conversational approach where they expose their own vulnerable and personable side, before expecting the Autistic CYP to be forthcoming with their own experiences. This could include discussing their own interests or difficulties with the healthcare system. Yet to strengthen this approach, more research should be conducted to understand what an Autistic CYP requires from their HCP to feel both safe, and comfortable. However, whilst academic studies discussing the broad healthcare experiences of the Autistic CYP are available, a disparity in the availability of clinical research involving Autistic CYP is apparent (Boerner et al., n.d.). Without this clinical research, an understanding of how Autistic CYP respond to existing, and subsequently adapted pain-related interventions cannot be determined. Extending the methods for engaging Autistic CYP suggested within this thesis to clinical research would be an important next step in ensuring these healthcare experiences are equitable. Additionally, as these experiences rely on interpersonal relationships, individuals in healthcare or scholastic settings should work to address these systemic issues, beginning with a self-analysis as to how they may contribute to promoting inequities, before discussing with peers how they can amend them (Goldbach et al., 2015).

Implementing a collective of these approaches to address these systemic issues in conjunction with developing knowledge for the impact of how social factors impact Autistic CYP's pain specifically would provide both a direct, and active approach in progressing this field. However, it must be acknowledged that these adaptations will take time to both develop and collect evidence that these suggestions are both a feasible, and cost-effective approach for healthcare systems to use. In the interim, it is important to still view Autistic

individuals, as individuals, meaning anyone who interacts with an Autistic CYP should take the time to understand that individual and their needs, rather than rely on the discussed neurotypical approach as a gold standard. By taking an individualistic approach for a group often considered heterogenous by diagnosis (Jeste & Geschwind, 2014), tailored adaptations can be identified to create an environment where Autistic CYP are supported and facilitate their ability to engage with a variety of services effectively, whether that be in education or healthcare as discussed in Chapters 4 and 5, or in research as discussed in Chapters 2 and 3.

6.2 Thesis Limitations

Within each individual Chapter, methodological and finding-based limitations are outlined. However, in designing studies to complement one another and encompass a consistent rationale, the limitations of the thesis itself precede the validity and applicability of these findings. Limitations can be categorised into three differing points: (1) a lack of intersectionality, (2) differences in eligibility criteria, and (3) methods of communicating pain research.

6.2.1 A Lack of Intersectionality

Despite promoting a neurodiversity-affirming approach, the findings of this thesis fail to represent intersectional experiences of pain. Inclusion criteria focused on the need for caregiver involvement, and a clinical diagnosis of Autism to aid in the creation of a dyadic evidence base that could be contextualised across the Autistic community (see Chapter 1.1.2). However, this decision and the implicit trauma this provides arguably presented the largest limitation to the application of findings.

As summarised by Smooth (2013, p. 11), intersectionality is: *"the assertion that social identity categories such as race, gender, class, sexuality, and ability are*

interconnected and operate simultaneously to produce experiences of both privilege and marginalization". Autism as an intersect of identity is recognised throughout this thesis from a neurodivergent perspective (i.e. use of IFL), and how societal perceptions of this identity can create experiences of marginalisation for Autistic CYP. For example, Autistic CYP's pain experiences regularly includes a description of pain dismissal from teachers and HCPs who lack understanding in how Autistic CYP communicate pain. However, the findings of this thesis are mostly contextualised to the experiences of White, verbal, females with predominantly required low support needs; a population whose healthcare experiences may be positioned in privilege comparative to Autistic CYP of differing ethnicities, gender, and social backgrounds. Consequentially, findings may not be valid in understanding pain in broad Autistic intersects such as ethnicities, LBTQIA+ and care-experience (Mallipeddi & VanDaalen, 2022).

An inclusion of predominantly White participants could reflect the recruitment catchment area: North-West England as the Office for National Statistics (2022) suggests 81.2% of the population identify as White British, and 4.4% as White other. However, this overrepresentation may also reflect the exclusion of self-diagnosis, which limited the participatory opportunities for Black, Latine and Asian CYP due to a lower likelihood of receiving a clinical diagnosis (as summarised in Aylward et al.'s (2021) literature review). Moreover, evidence suggests neurodivergent individuals are more likely than neurotypical individuals to identify as gender-diverse, or transgender (Brunissen et al., 2021; George & Stokes, 2017; Kourti & MacLeod, 2018; Warrier et al., 2020). Yet once again the recruited population were not representative of these lived experiences as most Autistic CYP defined their gender identity as binary: male, or female. This lack of representation may reflect more than a limited access to a diagnosis, as gender-diverse CYP report negative healthcare experiences including discrimination from a service provider when accessing treatment (Goulding et al., 2023). These experiences may have acted as an additional barrier to participation which the study design did not account for, with discussing pain experiences causing increased feelings of distress (Ramos, 2021). Similarly, emphasis on the need for caregiver involvement also may have limited the ability for CYP whose parental

responsibility is held by their local authority to participate, i.e. care experienced children and young people - a high proportion of which could be diagnosed Autistic (Annis, 2016).

The neurodiversity-affirming approach implemented in this thesis aimed to be inclusive in nature. However, failing to adapt for the implicit, and explicit systemic barriers that contribute to accessing a clinical diagnosis, or even discussing healthcare experiences may have limited real-world validity of findings. Without facilitating an environment to engage with a variety of Autistic intersectional identities, this research may contribute to a biased evidence base that continually fails to recognise the differing experiences of negative pain experiences in all social domains. As Smooth (2013) suggested, categories of social identity are interconnected and operate simultaneously, thus when designing methodology to be inclusive of neurotype, intersects that are prevalent amongst the population must and should be considered to accurately represent the whole community, rather than subgroups. Future research should adapt their recruitment methods to be inclusive of, and accessible to how individuals with intersecting identities engage with research, and be mindful of how their lived experiences of, for example, stigma impact this. However, researchers should recognise that such adaptations cannot be created in isolation; researchers should collaborate with those with lived experience to understand how these methods should be developed, instead of relying on existing literature alone.

6.2.2 Differences in Eligibility Criteria

Like criteria-based discussions in Chapter 6.2.1, differences in eligibility criteria between Chapters may have contributed to recruitment; particularly the specified need to not possess, or lack of recognition for a chronic pain diagnosis.

Recruitment figures outlined in Chapter 3.2.2 portray that the identification of Autistic CYP whom wanted to complete a psychophysical protocol was a difficult process – however the exclusion criteria may have only contributed to these difficulties. Stringent exclusion criteria were provided to prevent any CYP with chronic pain from completing the psychophysical protocol for safety purposes. Yet with chronic pain disproportionately

effecting Autistic CYP compared to non-Autistic CYP (Lipsker et al., 2018; Whitney & Shapiro, 2019), these criteria could have significantly reduced the eligible participant pool.

In contrast, criteria pertaining pain diagnoses were not provided in the eligibility criteria of Chapters 4 and 5 to allow for a broad understanding of daily pain experiences. Although this decision may have facilitated in gaining a broad insight, Autistic CYP and their caregivers may have interpreted recruitment materials describing a study about pain to only be eligible to those with chronic, or recurring pain. Thus, the experiences of Autistic CYP without chronic or recurring pain in their daily lives could be underrepresented throughout these Chapters. Upon reflection, in designing this research clarity in how these criteria were explained should have been provided, like how the differing pain experiences were described to CYP in Chapter 3. Future research should provide further information and clarification for their inclusion criteria pertaining pain, particularly when multiple studies intersect to limit the likelihood of such misinterpretations.

6.2.3 Methods of Communicating Pain Research

Generally, recruitment for pain research can be difficult with exemplar barriers to participant engagement including location, mistrust, fear, and a lack of communication (Anastasi et al., 2023; Kennedy et al., 2022). However, for an Autistic population with previous negative pain experiences, the concept of voluntarily evoking pain may be distressing. Despite adhering to the guidance of a PPI panel (see Chapter 0.1.2), engaging with the Autism community and providing Autistic participants with study details, intended data use, and multimedia sources for study information; study recruitment remained difficult, and data availability was limited (Gowen et al., 2019).

Whilst efforts to increase recruitment through accessibility and inclusivity were implemented; clearly more could have been done. For example, Janevic et al. (2022) contextualised the unique barriers underserved populations experience when participating in pain research, outlining how health literacy should be a consideration in inclusivity. Whilst

PISs and supporting videos were created to aid in CYP's understanding of what pain research involves, this does not equate to confirming their comprehension. Instead, implementing active recruitment methods may have been beneficial. For example, creating workshops in a familiar, or online environment to engage potential participants in the research experience whilst diminishing their fear of unexpected pain to encourage participation. Additionally, CYP's participation involved a caregiver in some capacity who would have had their own perceptions of how this research would impact their CYP. Acting as the initial gatekeeper to CYP, more focus should have been provided to how caregivers would interpret the introductory information provided on study flyers. Perhaps the intended outcomes of this research should have been amplified through similar workshop approaches to demonstrate the real-world impact their CYP's participation could provide. Future research should consider these recommendations when communicating pain to CYP and their caregiver, and if implemented, assess the feasibility of approach to support future paediatric pain researchers in their own recruitment.

6.3 Future Research

Whilst these findings provide an important first step in understanding the pain experiences of Autistic CYP, further research is required to continue this developing field. In positioning how these findings can inform of future research, both methodological and theory-based framework are highlighted.

Although the participating Autistic CYP in this thesis provided retrospective and experimentally induced insight into their pain experiences, validity in how these findings represent real-world experiences remains limited. For example, emotional descriptions of a pain that occurred last week, or pain intensity rating of an experimentally controlled pain are not a true representation of how an Autistic CYP might react when pain spontaneously presents. To provide this real-world understanding and identify how pain impacts an Autistic CYP in their daily life, future research should utilise "obser-views" – a method of combining observations with immediate and reflective interviews (Kragelund, 2013; Kragelund et al., 2015). With the unpredictability of acute pain, researchers should begin by observing Autistic

CYP with chronic pain to increase the likelihood for the presence of pain. Observations should adhere to the three interpersonal relationships discussed in Chapters 4 and 5: caregivers at home, teachers at school, and HCPs at medical appointments – a likely event for a population overrepresented in pain management settings (Lipsker et al., 2018). Throughout, researchers should collect data on the CYP's daily pain intensity; observe if, when, who and how the Autistic CYP discloses pain; and converse to reflect on the CYP's daily behaviours. For example, "Your pain today was as bad as when you broke your leg today (9/10), how did you manage your pain today?". Doing so would provide identify additional facilitators or barriers to pain disclosure, and could outline representative guidance of how caregivers, HCPs and teachers can appraise Autistic CYP's pain and support them in disclosing.

Whilst these observations of would be beneficial, the apprehension ethics committees often express towards both paediatric, and neurodivergent research could mean developing understanding experimentally should be considered. Future research should develop an understanding of how Autistic CYP's previous experiences of distress consequential to pain dismissal experimentally predicts their behavioural intent to disclose pain. For example, throughout Chapters 4 and 5, previous negative experiences of pain prevented Autistic CYP from disclosing in certain interpersonal relationships consequential to the perceived threat of their pain being dismissed. In broad pain literature, research suggests social threats influence the ability for non-Autistic individuals to verbally or facially express pain, observing increased inhibition of pain disclosure in the presence of a stranger (Krahé et al., 2013; Vervoort et al., 2008; Vlaeyen et al., 2009). However, these expressive patterns are conflicted in CYP with increased pain anxiety, for example the pain expressions of CYP who highly-catastrophize pain are more pronounced regardless of the interpersonal relationship present (Vervoort et al., 2011; Vervoort et al., 2008). Although the latter findings are contextualised to non-Autistic CYP, the implications could be applied to understand social patterns of pain expression. For example, Autistic CYP and caregivers in Chapter 4 described that when pain severity worsens, the CYP's intent to disclose pain increases regardless of the interpersonal relationship present. Here, an understanding of how anxiety caused by higher severity of experimental pain modulates an Autistic CYP's intent to

disclose pain between trusted, and untrusted interpersonal relationships may be important to understand the importance of physical threat (pain), vs. social threat (interpersonal variables) in pain disclosure.

Moreover, future research interested in understanding Autistic CYP's experimental pain responses should continue to challenge available protocols through co-production with Autistic individuals. Co-production is a common method to engage when designing qualitative research like the interviews, however less common for experimental designs like the psychophysical assessment. Here, co-production was highlighted as important for designing the psychophysical protocol used in Chapter 3 as whilst standardised approaches like Rolke et al. (2006) and Blankenburg et al. (2010) are available, they fail to consider neurodivergent experiences. Co-producing this protocol with Autistic individuals bridged this gap to facilitate design inclusivity, accessibility, participant engagement and prevent withdrawals from distress. It could be argued that adapting these established and acclaimed protocols provides too much data variability and encourages criticism from other researchers. However, the retention rate of Chapter 3 only emphasises how co-production can strengthen the knowledgebase surrounding Autistic pain experiences and identify additional adaptations to those listed in Chapter 3.2 to continue developing pain research protocols that are more inclusive, accessible, and overall - neurodiversity-affirming.

6.4 Key Recommendations

1. Within Autistic diagnostic criteria, the statement "insensitive to pain" does not reflect experiences. This criterion has the potential to create misunderstanding and medical harm for Autistic CYP. Update Autism diagnostic criterion to reflect pain as a subjective experience, whereby Autistic CYP predominantly show no difference in pain thresholds.
2. Place future research efforts into understanding the systemic barriers that contribute to the worsening of Autistic CYP's pain. From this, develop training for HCPs and teachers to support them in better appraising, managing, and treating Autistic CYP's pain.

3. Recognise the importance of a neurodiversity-affirming approach to healthcare by adapting communicative methods for understanding pain to the individual Autistic CYP's needs.

6.5 Conclusion

Historically, Autistic CYP have been diagnostically and anecdotally perceived to not experience pain; a perception that the DSM-5 (American Psychiatric Association, 2023) contributes to in providing pain hyposensitivity as a diagnostic feature of Autism. Yet statistics counter these perceptions and suggest Autistic CYP experience pain at alarmingly high rates, emphasising a clear discordance in our understanding of Autistic pain experiences. To address this knowledge gap, the aim of this thesis was to examine factors which relate to Autistic CYP's pain experiences and expression. A range of methods were utilised to gain thorough insight into the differing facets of pain experiences, including psychophysical pain responses, influential factors of pain and pain disclosure behaviours. Unequivocally, the findings of this thesis demonstrated that Autistic CYP have the neural archetype to feel pain, with no evidence to support that Autistic CYP's pain perceptions differ from a neurotypical population. However, interpersonal factors such as a lack of trust from previous pain dismissal acted as a gatekeeper to Autistic CYP socially expressing or disclosing their pain. For example, Autistic CYP would refrain from disclosing their pain to HCPs and teachers to avoid consequential distress however this would limit their ability to access pain management. Without understanding of how to facilitate Autistic CYP in disclosing pain, and support HCPs and teachers in recognising Autistic CYP in pain, ample opportunity presents for the currently high pain prevalence rate to increase further. Future research should identify how Autistic CYP's distress translates into the medical trauma that prevents them from disclosing pain and develop methods of reducing this emotional response. Additionally, further training is required for HCPs and teachers in understanding how Autistic CYP express pain. Currently researchers are regularly educating HCPs on what neurodivergence is, and the broad healthcare experiences of Autistic CYP. However, a focused understanding for both HCPs and teachers of how Autistic CYP communicate pain,

and how this presentation may differ from neurotypical CYP is paramount in ensuring pain is correctly appraised, and managed.

Chapter 7.

References

7 References

37signals. (2024). *Basecamp 4*. In (Version 4) [Project Management]. 37signals. https://basecamp.com/

Abad, F., Díaz-Gómez, N. M., Rodríguez, I., Pérez, R., & Delgado, J. A. (2002). Subclinical pain and thermal sensory dysfunction in children and adolescents with Type 1 diabetes mellitus. *Diabetic Medicine*, *19*(10), 827-831. https://doi.org/10.1046/j.1464-5491.2002.00793.x

Akbari, F., Dehghani, M., Mohammadi, S., Goubert, L., Sanderman, R., & Hagedoorn, M. t. (2020). Why do Patients Engage in Pain Behaviors? A Qualitative Study Examining the Perspective of Patients and Partners. *The Clinical Journal of Pain*, *36*(10). https://doi.org/10.1097/AJP.0000000000000868

Akre, C., & Suris, J.-C. (2014). From controlling to letting go: what are the psychosocial needs of parents of adolescents with a chronic illness? *Health Education Research*, *29*(5), 764-772. https://doi.org/10.1093/her/cyu040

Alderson, P. (2018). How the rights of all school students and teachers are affected by SEND (special educational needs or disability) services: teaching, psychology, policy. *London Review of Education*, *16*, 175-190. https://doi.org/10.18546/LRE.16.2.01

Alfvén, G. (1993). The pressure pain threshold (PPT) of certain muscles in children suffering from recurrent abdominal pain of non-organic origin. An algometric study. *Acta Paediatrica*, *82*(5), 481-483. https://doi.org/10.1111/j.1651-2227.1993.tb12727.x

Allely, C. S. (2013). Pain sensitivity and observer perception of pain in individuals with autistic spectrum disorder. *The Scientific World Journal*, *2013*, 916178-916178. https://doi.org/10.1155/2013/916178

Alvares, G. A., Bebbington, K., Cleary, D., Evans, K., Glasson, E. J., Maybery, M. T., Pillar, S., Uljarević, M., Varcin, K., Wray, J., & Whitehouse, A. J. (2020). The misnomer of 'high functioning autism': Intelligence is an imprecise predictor of functional abilities at diagnosis. *Autism*, *24*(1), 221-232. https://doi.org/10.1177/1362361319852831

American Psychiatric Association. (2023). Neurodevelopmental Disorders. In *Diagnostic and Statistical Manual of Mental Disorders: Fifth Edition Text Revision (DSM-5-TR)* (Fifth ed.). https://doi.org/10.1176/appi.books.9780890425787.x01_Neurodevelopmental_Disorders

Anastasi, J. K., Capili, B., Norton, M., McMahon, D. J., & Marder, K. (2023). Recruitment and retention of clinical trial participants: understanding motivations of patients with chronic pain and other populations. *Frontiers in Pain Research*, *4*, 1330937. https://doi.org/10.3389/fpain.2023.1330937

Anonymous. (n.d.). 0-10 Scale of Pain Severity.

Anttila, P., Metsähonkala, L., Mikkelsson, M., Aromaa, M., Kautiainen, H., Salminen, J., Viander, S., Jäppilä, E., & Sillanpää, M. (2002). Muscle Tenderness in Pericranial and Neck-Shoulder Region in Children with Headache. A Controlled Study. *Cephalalgia*, *22*(5), 340-344. https://doi.org/10.1046/j.1468-2982.2002.00352.x

Ashworth, E., Bray, L., Hanlon, C., Stanway, H., Pavlopoulou, G., Moore, D., Donaghy, B., Coen, E., & Firth, E. (2024). "Accumulating harm and waiting for crisis": Parents' perspectives of accessing Child and Adolescent Mental Health Services for their autistic child experiencing mental health difficulties. *medRxiv*, 2024.2004.2009.24305538. https://doi.org/10.1101/2024.04.09.24305538

Atherton, G., Edisbury, E., Piovesan, A., & Cross, L. (2022). Autism Through the Ages: A Mixed Methods Approach to Understanding How Age and Age of Diagnosis Affect Quality of Life. *Journal of Autism and Developmental Disorders*, *52*(8), 3639-3654. https://doi.org/10.1007/s10803-021-05235-x

Aylward, B. S., Gal-Szabo, D. E., & Taraman, S. (2021). Racial, Ethnic, and Sociodemographic Disparities in Diagnosis of Children with Autism Spectrum Disorder. *Journal of Developmental & Behavioral Pediatrics*, *42*(8), 682-689. https://doi.org/10.1097/dbp.0000000000000996

Babalola, T., Sanguedolce, G., Dipper, L., & Botting, N. (2024). Barriers and Facilitators of Healthcare Access for Autistic Children in the UK: a Systematic Review. *Review Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s40489-023-00420-3

Backonja, M. M., Attal, N., Baron, R., Bouhassira, D., Drangholt, M., Dyck, P. J., Edwards, R. R., Freeman, R., Gracely, R., Haanpaa, M. H., Hansson, P., Hatem, S. M., Krumova, E. K., Jensen, T. S., Maier, C., Mick, G., Rice, A. S., Rolke, R., Treede, R.-D., Serra, J., Toelle, T., Tugnoli, V., Walk, D., Walalce, M. S., Ware, M., Yarnitsky, D., & Ziegler, D. (2013). Value of quantitative sensory testing in neurological and pain disorders: NeuPSIG consensus. *PAIN*, *154*(9). https://doi.org/10.1016/j.pain.2013.05.047

Baker, C. M., & Wong, D. L. (1987). QUEST: a process of pain assessment in children. *Orthopaedic Nursing*, *6*(1), 11-21.

Bal, V. H., Farmer, C., & Thurm, A. (2017). Describing Function in ASD: Using the DSM-5 and Other Methods to Improve Precision. *Journal of Autism and Developmental Disorders*, *47*(9), 2938-2941. https://doi.org/10.1007/s10803-017-3204-3

Balta, S., & Arslan, D. (2021). Pain pressure thresholds in children and adolescents with idiopathic chest pain. *Cardiology in the Young*, 1-5. https://doi.org/10.1017/s1047951121001840

Bandstra, N. F., Johnson, S. A., Filliter, J. H., & Chambers, C. T. (2012). Self-reported and Parent-reported Pain for Common Painful Events in High-functioning Children and Adolescents With Autism Spectrum Disorder. *The Clinical Journal of Pain*, *28*(8), 715-721. https://doi.org/10.1097/AJP.0b013e318243ecf6

Bargiela, S., Steward, R., & Mandy, W. (2016). The Experiences of Late-diagnosed Women with Autism Spectrum Conditions: An Investigation of the Female Autism Phenotype. *Journal of Autism and Developmental Disorders*, *46*(10), 3281-3294. https://doi.org/10.1007/s10803-016-2872-8

Barney, C. C., Andersen, R. D., Defrin, R., Genik, L. M., McGuire, B. E., & Symons, F. J. (2020). Challenges in pain assessment and management among individuals with intellectual and developmental disabilities. *Pain reports*, *5*(4), e821. https://doi.org/10.1097/pr9.0000000000000822

Bartley, E. J., & Fillingim, R. B. (2013). Sex differences in pain: a brief review of clinical and experimental findings. *British Journal of Anaesthesia*, *111*(1), 52-58. https://doi.org/10.1093/bja/aet127

Beesdo, K., Jacobi, F., Hoyer, J., Low, N. C. P., Höfler, M., & Wittchen, H.-U. (2010). Pain associated with specific anxiety and depressive disorders in a nationally representative population sample. *Social Psychiatry and Psychiatric Epidemiology*, *45*(1), 89-104. https://doi.org/10.1007/s00127-009-0045-1

Bek, N., Uygur, F., Bayar, B., & Armutlu, K. (2002). Analysis of age and gender related differences in pressure pain threshold and pressure pain tolerance levels. *The Pain Clinic*, *14*(4), 309-314. https://doi.org/10.1163/15685690260494843

Benich, S., Thakur, S., Schubart, J. R., & Carr, M. M. (2018). Parental Perception of the Perioperative Experience for Children With Autism. *AORN Journal*, *108*(1), 34-43. https://doi.org/10.1002/aorn.12274

Bernard, M., Chaparro, B., Mills, M., & Halcomb, C. (2002). Examining children's reading performance and preference for different computer-displayed text. *Behaviour & IT*, *21*, 87-96. https://doi.org/10.1080/01449290210146737

Bewick, V., Cheek, L., & Ball, J. (2004). Statistics review 8: Qualitative data - Tests of association. *Critical care (London, England)*, *8*, 46-53. https://doi.org/10.1186/cc2428

Bieri, D., Reeve, R. A., Champion, D. G., Addicoat, L., & Ziegler, J. B. (1990). The Faces Pain Scale for the self-assessment of the severity of pain experienced by children: development, initial validation, and preliminary investigation for ratio scale properties. *PAIN*, *41*(2), 139-150. https://doi.org/10.1016/0304-3959(90)90018-9

Bird, G., Silani, G., Brindley, R., White, S., Frith, U., & Singer, T. (2010). Empathic brain responses in insula are modulated by levels of alexithymia but not autism. *Brain*, *133*(5), 1515-1525. https://doi.org/10.1093/brain/awq060

Birnie, K. A., Hundert, A. S., Lalloo, C., Nguyen, C., & Stinson, J. N. (2019). Recommendations for selection of self-report pain intensity measures in children and adolescents: a systematic review and quality assessment of measurement properties. *PAIN*, *160*(1). https://doi.org/10.1097/j.pain.0000000000001377

Birnie, K. A., Petter, M., Boerner, K. E., Noel, M., & Chambers, C. T. (2012). Contemporary Use of the Cold Pressor Task in Pediatric Pain Research: A Systematic Review of Methods. *The Journal of Pain*, *13*(9), 817-826. https://doi.org/10.1016/j.jpain.2012.06.005

Black, M. H., Kuzminski, R., Wang, J., Ang, J., Lee, C., Hafidzuddin, S., & McGarry, S. (2022). Experiences of Friendships for Individuals on the Autism Spectrum: A Scoping Review. *Review Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s40489-022-00332-8

Blankenburg, M., Boekens, H., Hechler, T., Maier, C., Krumova, E., Scherens, A., Magerl, W., Aksu, F., & Zernikow, B. (2010). Reference values for quantitative sensory testing in children and adolescents: Developmental and gender differences of somatosensory perception. *PAIN*, *149*(1), 76-88. https://doi.org/10.1016/j.pain.2010.01.011

Blankenburg, M., Kraemer, N., Hirschfeld, G., Krumova, E. K., Maier, C., Hechler, T., Aksu, F., Magerl, W., Reinehr, T., Wiesel, T., & Zernikow, B. (2012). Childhood diabetic neuropathy: functional impairment and non-invasive screening assessment. *Diabetic Medicine*, *29*(11), 1425-1432. https://doi.org/10.1111/j.1464-5491.2012.03685.x

Blankenburg, M., Meyer, D., Hirschfeld, G., Kraemer, N., Hechler, T., Aksu, F., Krumova, E. K., Magerl, W., Maier, C., & Zernikow, B. (2011). Developmental and sex differences in somatosensory perception--a systematic comparison of 7- versus 14-year-olds using quantitative sensory testing. *PAIN*, *152*(11), 2625-2631. https://doi.org/10.1016/j.pain.2011.08.007

Blieden, M., Paramore, L. C., Shah, D., & Ben-Joseph, R. (2014). A perspective on the epidemiology of acetaminophen exposure and toxicity in the United States. *Expert review of clinical pharmacology*, *7*(3), 341-348.

Boerner, K.E., Pawliuk, C., Heran, A., Donaghy, B., Moore, D. J., Leong, K., Devan, H., & Oberlander, T. F. (n.d.) Systematic Review of Autistic Representation in the Treatment Literature for Pediatric Chronic Pain. *Journal of Pain.* (Accepted).

Bogdanova, O. V., Bogdanov, V. B., Pizano, A., Bouvard, M., Cazalets, J. R., Mellen, N., & Amestoy, A. (2022). The Current View on the Paradox of Pain in Autism Spectrum Disorders. *Frontiers in Psychiatry*, *13*, 910824. https://doi.org/10.3389/fpsyt.2022.910824

Bonezzi, C., Fornasari, D., Cricelli, C., Magni, A., & Ventriglia, G. (2020). Not All Pain is Created Equal: Basic Definitions and Diagnostic Work-Up. *Pain and Therapy*, *9*(Suppl 1), 1-15. https://doi.org/10.1007/s40122-020-00217-w

Boshoff, K., Bowen-Salter, H., Gibbs, D., Phillips, R. L., Porter, L., & Wiles, L. (2021). A meta-synthesis of how parents of children with autism describe their experience of

accessing and using routine healthcare services for their children. *Health & Social Care in the Community*, *29*(6), 1668-1682. https://doi.org/10.1111/hsc.13369
Bottema-Beutel, K., Kapp, S. K., Lester, J. N., Sasson, N. J., & Hand, B. N. (2021). Avoiding ableist language: Suggestions for autism researchers. *Autism in Adulthood*. https://doi.org/10.1089/aut.2020.0014
Bottini, S. B., Morton, H. E., Buchanan, K. A., & Gould, K. (2023). Moving from Disorder to Difference: A Systematic Review of Recent Language Use in Autism Research. *Autism in Adulthood*. https://doi.org/10.1089/aut.2023.0030
Brand, K., & Al-Rais, A. (2019). Pain assessment in children. *Anaesthesia & Intensive Care Medicine*, *20*(6), 314-317. https://doi.org/10.1016/j.mpaic.2019.03.003
Breau, L. M., Finley, G. A., McGrath, P. J., & Camfield, C. S. (2002). Validation of the Non-communicating Children's Pain Checklist–Postoperative Version. *Anesthesiology*, *96*(3), 528-535. https://doi.org/10.1097/00000542-200203000-00004
Breau, L. M., McGrath, P. J., Camfield, C. S., & Finley, G. A. (2002). Psychometric properties of the non-communicating children's pain checklist-revised. *PAIN*, *99*(1), 349-357. https://doi.org/10.1016/S0304-3959(02)00179-3
British Pain Society. (2021). *USEFUL DEFINITIONS AND GLOSSARY*. Retrieved 15th July from https://www.britishpainsociety.org/people-with-pain/useful-definitions-and-glossary/#pain
Broome, K., McCabe, P., Docking, K., Doble, M., & Carrigg, B. (2023). Speech Development Across Subgroups of Autistic Children: A Longitudinal Study. *Journal of Autism and Developmental Disorders*, *53*(7), 2570-2586. https://doi.org/10.1007/s10803-022-05561-8
Brown, D., Schenk, S., Genent, D., Zernikow, B., & Wager, J. (2021). A scoping review of chronic pain in emerging adults. *Pain reports*, *6*(1). https://doi.org/10.1097/PR9.0000000000000920
Brunissen, L., Rapoport, E., Chawarska, K., & Adesman, A. (2021). Sex Differences in Gender-Diverse Expressions and Identities among Youth with Autism Spectrum Disorder. *Autism Research*, *14*(1), 143-155. https://doi.org/10.1002/aur.2441
Buchanan, H. M., & Midgley, J. A. (1987). Evaluation of pain threshold using a simple pressure algometer. *Clinical Rheumatology*, *6*(4), 510-517. https://doi.org/10.1007/bf02330587
Buskila, D., Neumann, L., Zmora, E., Feldman, M., Bolotin, A., & Press, J. (2003). Pain Sensitivity in Prematurely Born Adolescents. *Archives of Pediatrics & Adolescent Medicine*, *157*(11), 1079-1082. https://doi.org/10.1001/archpedi.157.11.1079
Calder, L., Hill, V., & Pellicano, E. (2013). ‘Sometimes I want to play by myself’: Understanding what friendship means to children with autism in mainstream primary schools. *Autism*, *17*(3), 296-316. https://doi.org/10.1177/1362361312467866
Camerota, F., Mariani, R., Cordiano, G., Di Trani, M., Lodato, V., Ferraris, A., Pasquini, M., & Celletti, C. (2023). The Language of Pain in the Hypermobile Ehlers–Danlos Syndrome: Metaphors as a Key to Understanding the Experience of Pain and as a Rehabilitation Tool. *Brain sciences*, *13*(7), 1042. https://doi.org/10.3390/brainsci13071042
Camm-Crosbie, L., Bradley, L., Shaw, R., Baron-Cohen, S., & Cassidy, S. (2018). ‘People like me don’t get support’: Autistic adults’ experiences of support and treatment for mental health difficulties, self-injury and suicidality. *Autism*, *23*(6), 1431-1441. https://doi.org/10.1177/1362361318816053
Campi, L. B., Visscher, C. M., Ongaro, P. C. J., do Vale Braido, G. V., Fernandes, G., & Gonçalves, D. A. G. (2020). Widespread Pain and Central Sensitization in Adolescents with Signs of Painful Temporomandibular Disorders. *Journal of Oral & Facial Pain and Headache*, *34*(1), 83–91. https://doi.org/10.11607/ofph.2288

Canitano, R. (2006). Self injurious behavior in autism: clinical aspects and treatment with risperidone. *Journal of Neural Transmission*, *113*(3), 425-431. https://doi.org/10.1007/s00702-005-0337-x

Caouette, J. D., & Guyer, A. E. (2014). Gaining insight into adolescent vulnerability for social anxiety from developmental cognitive neuroscience. *Developmental Cognitive Neuroscience*, *8*, 65-76. https://doi.org/10.1016/j.dcn.2013.10.003

Carpenter, K. L. H., Baranek, G. T., Copeland, W. E., Compton, S., Zucker, N., Dawson, G., & Egger, H. L. (2019). Sensory Over-Responsivity: An Early Risk Factor for Anxiety and Behavioral Challenges in Young Children. *Journal of Abnormal Child Psychology*, *47*(6), 1075-1088. https://doi.org/10.1007/s10802-018-0502-y

Carter, B. (2002). Chronic Pain in Childhood and the Medical Encounter: Professional Ventriloquism and Hidden Voices. *Qualitative Health Research*, *12*(1), 28-41. https://doi.org/10.1177/104973230201200103

Cascio, C., McGlone, F., Folger, S., Tannan, V., Baranek, G., Pelphrey, K. A., & Essick, G. (2008). Tactile Perception in Adults with Autism: a Multidimensional Psychophysical Study. *Journal of Autism and Developmental Disorders*, *38*(1), 127-137. https://doi.org/10.1007/s10803-007-0370-8

Casterman, N., Rossignol, M., Colomar, A., & Cassioli, F. (2024). Thermoception in Autism Spectrum Disorder: A Concise Systematic Review. *Review Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s40489-024-00445-2

Chae, Y., Kim, H. Y., Lee, H. J., Park, H. J., Hahm, D. H., An, K., & Lee, H. (2007). The alteration of pain sensitivity at disease-specific acupuncture points in premenstrual syndrome. *The Journal of Physiological Sciences*, *57*(2), 115-119. https://doi.org/10.2170/physiolsci.RP012706

Chakraborty, P., Carpenter, K. L. H., Major, S., Deaver, M., Vermeer, S., Herold, B., Franz, L., Howard, J., & Dawson, G. (2021). Gastrointestinal problems are associated with increased repetitive behaviors but not social communication difficulties in young children with autism spectrum disorders. *Autism*, *25*(2), 405-415. https://doi.org/10.1177/1362361320959503

Chapman, L., Rose, K., Hull, L., & Mandy, W. (2022). “I want to fit in… but I don’t want to change myself fundamentally”: A qualitative exploration of the relationship between masking and mental health for autistic teenagers. *Research in Autism Spectrum Disorders*, *99*, 102069. https://doi.org/10.1016/j.rasd.2022.102069

Charlton, R. A., Entecott, T., Belova, E., & Nwaordu, G. (2021). “It feels like holding back something you need to say”: Autistic and Non-Autistic Adults accounts of sensory experiences and stimming. *Research in Autism Spectrum Disorders*, *89*, 101864. https://doi.org/10.1016/j.rasd.2021.101864

Chaudhary, A. (2021). Migraine Associated with Menstruation An Overlooked Trigger. *JNMA J Nepal Med Assoc*, *59*(238), 611-613. https://doi.org/10.31729/jnma.6332

Chaves, T. C., Nagamine, H. M., de Sousa, L. M., de Oliveira, A. S., Regalo, S. C., & Grossi, D. B. (2013). Differences in pain perception in children reporting joint and orofacial muscle pain. *Journal of Clinical Pediatric Dentistry*, *37*(3), 321-327. https://doi.org/10.17796/jcpd.37.3.013017332p40j65u

Chen, C., Hung, A.-Y., Fan, Y.-T., Tan, S., Hong, H., & Cheng, Y. (2017). Linkage between pain sensitivity and empathic response in adolescents with autism spectrum conditions and conduct disorder symptoms. *Autism Research*, *10*(2), 267-275. https://doi.org/10.1002/aur.1653

Chen, S.-M., Chen, J.-T., Kuan, T.-S., Hong, J., & Hong, C.-Z. (2000). Decrease in Pressure Pain Thresholds of Latent Myofascial Trigger Points in the Middle Finger Extensors Immediately After Continuous Piano Practice. *Journal of Musculoskeletal Pain*, *8*(3), 83-92. https://doi.org/10.1300/J094v08n03_07

Cheng, Y., Hung, A. Y., & Decety, J. (2012). Dissociation between affective sharing and emotion understanding in juvenile psychopaths. *Development and Psychopathology*, *24*(2), 623-636. https://doi.org/10.1017/s095457941200020x

Cimpean, A., & David, D. (2019). The mechanisms of pain tolerance and pain-related anxiety in acute pain. *Health Psychology Open*, *6*(2), 2055102919865161. https://doi.org/10.1177/2055102919865161

Clarke, C. (2015). Autism Spectrum Disorder and Amplified Pain. *Case Reports in Psychiatry*, *2015*, 930874. https://doi.org/10.1155/2015/930874

Cohen, J. (2013). *Statistical power analysis for the behavioral sciences*. Routledge.

Colman, A. M. (2015). Fechner's law. In: Oxford University Press.

Coolican, H. (2004). *Research Methods and Statistics in Psychology* (4 ed.). Hodder & Stoughton.

Coolican, H. (2024). *Research Methods and Statistics in Psychology*. Taylor & Francis Group. http://ebookcentral.proquest.com/lib/ljmu/detail.action?docID=31005775

Corbett, B. A., Muscatello, R. A., & Blain, S. D. (2016). Impact of sensory sensitivity on physiological stress response and novel peer interaction in children with and without autism spectrum disorder. *Frontiers in Neuroscience*, *10*, 278.

Corder, G., Castro, D. C., Bruchas, M. R., & Scherrer, G. (2018). Endogenous and Exogenous Opioids in Pain. *Annual Review of Neuroscience*, *41*, 453-473. https://doi.org/10.1146/annurev-neuro-080317-061522

Cornelissen, L., Donado, C., Kim, J., Chiel, L., Zurakowski, D., Logan, D. E., Meier, P., Sethna, N. F., Blankenburg, M., Zernikow, B., Sundel, R. P., & Berde, C. B. (2014). Pain hypersensitivity in juvenile idiopathic arthritis: a quantitative sensory testing study. *Pediatric Rheumatology*, *12*(1), 39. https://doi.org/10.1186/1546-0096-12-39

Courtemanche, A. B., Black, W. R., & Reese, R. M. (2016). The Relationship Between Pain, Self-Injury, and Other Problem Behaviors in Young Children With Autism and Other Developmental Disabilities. *Am J Intellect Dev Disabil*, *121*(3), 194-203. https://doi.org/10.1352/1944-7558-121.3.194

Craig, K. D. (1992). The facial expression of pain Better than a thousand words? *APS Journal*, *1*(3), 153-162. https://doi.org/10.1016/1058-9139(92)90001-S

Craig, K. D. (2009). The social communication model of pain. *Canadian Psychology/Psychologie canadienne*, *50*(1), 22. https://doi.org/10.1037/a0014772

Craig, K. D. (2015). Social communication model of pain. *PAIN*, *156*(7), 1198-1199. https://doi.org/10.1097/j.pain.0000000000000185

Crellin, D. J., Harrison, D., Santamaria, N., & Babl, F. E. (2015). Systematic review of the Face, Legs, Activity, Cry and Consolability scale for assessing pain in infants and children: is it reliable, valid, and feasible for use? *PAIN*, *156*(11). https://doi.org/10.1097/j.pain.0000000000000305

Crompton, C. J., Hallett, S., Ropar, D., Flynn, E., & Fletcher-Watson, S. (2020). 'I never realised everybody felt as happy as I do when I am around autistic people': A thematic analysis of autistic adults' relationships with autistic and neurotypical friends and family. *Autism*, *24*(6), 1438-1448. https://doi.org/10.1177/1362361320908976

Cummins, C., Pellicano, E., & Crane, L. (2020). Autistic adults' views of their communication skills and needs. *International Journal of Language & Communication Disorders*, *55*(5), 678-689. https://doi.org/10.1111/1460-6984.12552

Cummins, T. M., English, O., Minnis, H., Stahl, D., O'Connor, R. C., Bannister, K., McMahon, S. B., & Ougrin, D. (2021). Assessment of Somatosensory Function and Self-harm in Adolescents. *JAMA Netw Open*, *4*(7), e2116853. https://doi.org/10.1001/jamanetworkopen.2021.16853

Dallman, A. R., Wilson, K., & Friesner, E. (2023). Social Agency and Neurodivergent Routines: Supports and Barriers to the Mental Health of Autistic Adults During the

COVID-19 Pandemic. *The American Journal of Occupational Therapy*, *77*(2). https://doi.org/10.5014/ajot.2023.050049

Dansie, E. J., & Turk, D. C. (2013). Assessment of patients with chronic pain. *BJA: British Journal of Anaesthesia*, *111*(1), 19-25. https://doi.org/10.1093/bja/aet124

Daughters, H., Palermo, T., & Koh, J. (2007). (720): Procedural pain and distress in children with autism- A pilot study. *The Journal of Pain*, *8*(4, Supplement), S31. https://doi.org/10.1016/j.jpain.2007.02.124

de Araújo Vitor, C. A., de Oliveira Lira Ortega, A., Ferreira, A., da Silva, C. A. L., Gallottini, M. H. C., & Santos, M. (2021). Pressure pain threshold of masticatory muscles in children and adolescents with and without intellectual disability: a pilot study. *European Archives of Paediatric Dentistry*, *22*(4), 587-593. https://doi.org/10.1007/s40368-020-00593-z

de Knegt, N. C., Pieper, M. J. C., Lobbezoo, F., Schuengel, C., Evenhuis, H. M., Passchier, J., & Scherder, E. J. A. (2013). Behavioral Pain Indicators in People With Intellectual Disabilities: A Systematic Review. *The Journal of Pain*, *14*(9), 885-896. https://doi.org/10.1016/j.jpain.2013.04.016

de Tommaso, M., Sciruicchio, V., Ricci, K., Montemurno, A., Gentile, F., Vecchio, E., Barbaro, M. G., Simeoni, M., Goffredo, M., & Livrea, P. (2016). Laser-evoked potential habituation and central sensitization symptoms in childhood migraine. *Cephalalgia*, *36*(5), 463-473. https://doi.org/10.1177/0333102415597527

Defenderfer, E. K., Bauer, K., Igler, E., Uihlein, J. A., & Davies, W. H. (2018). The Experience of Pain Dismissal in Adolescence. *The Clinical Journal of Pain*, *34*(2). https://doi.org/10.1097/AJP.0000000000000530

den Houting, J. (2018). Neurodiversity: An insider's perspective. *Autism*, *23*(2), 271-273. https://doi.org/10.1177/1362361318820762

Mental Health Capacity Act, § 1(1) (2) (3) (4) (5) (6), 2(1), 3(1) (a) (b) (c) (d) (2005). https://www.legislation.gov.uk/ukpga/2005/9/contents

Derguy, C., Aubé, B., Rohmer, O., & Loyal, D. (2023). Evaluation of teachers' knowledge and attitudes regarding the school inclusion of autistic children: Validation of a brief assessment toolkit. *British Journal of Educational Psychology*, *93*(2), 467-481. https://doi.org/10.1111/bjep.12569

Derman, O., Kanbur, N. Ö., Baltaci, G., Akbayrak, T., & Tokur, T. (2004). The pain intensity level in adolescents with primary dysmenorrhea. *The Pain Clinic*, *16*(3), 349-352. https://doi.org/10.1163/1568569041798353

Descartes, R., Clerselier, C., de La Forge, L., & Schuyl, F. (1662). *L'homme... et un traitté de la formation du fœtus. Du mesme autheur*. Charles Angot.

Di Lernia, D., Serino, S., & Riva, G. (2016). Pain in the body. Altered interoception in chronic pain conditions: A systematic review. *Neuroscience & Biobehavioral Reviews*, *71*, 328-341. https://doi.org/10.1016/j.neubiorev.2016.09.015

Dildine, T. C., Amir, C. M., Parsons, J., & Atlas, L. Y. (2023). How Pain-Related Facial Expressions Are Evaluated in Relation to Gender, Race, and Emotion. *Affective Science*. https://doi.org/10.1007/s42761-023-00181-6

Dobson, O., Symons, F. J., & McMurtry, C. M. (2023). Making needle procedures comfortable for autistic children: Caregiver perspectives. *Research in Autism Spectrum Disorders*, *107*, 102208. https://doi.org/10.1016/j.rasd.2023.102208

Doherty, A. J., Atherton, H., Boland, P., Hastings, R., Hives, L., Hood, K., James-Jenkinson, L., Leavey, R., Randell, E., Reed, J., Taggart, L., Wilson, N., & Chauhan, U. (2020). Barriers and facilitators to primary health care for people with intellectual disabilities and/or autism: an integrative review. *BJGP Open*, *4*(3), bjgpopen20X101030. https://doi.org/10.3399/bjgpopen20X101030

Doherty, M., Haydon, C., & Davidson, I. A. (2021). Recognising autism in healthcare. *British Journal of Hospital Medicine*, *82*(12), 1-7. https://doi.org/10.12968/hmed.2021.0313

Doherty, M., Neilson, S., O'Sullivan, J., Carravallah, L., Johnson, M., Cullen, W., & Shaw, S., C. K. (2022). Barriers to healthcare and self-reported adverse outcomes for autistic adults: a cross-sectional study. *BMJ Open*, *12*(2), e056904. https://doi.org/10.1136/bmjopen-2021-056904

Donaghy, B., Moore, D., & Green, J. (2023). Co-Occurring Physical Health Challenges in Neurodivergent Children and Young People: A Topical Review and Recommendation. *Child Care in Practice*, 1-19. https://doi.org/10.1080/13575279.2022.2149471

Donaghy, B., Walker, S. C., & Moore, D. J. (2022). Social distancing with chronic pain during COVID-19: A cross-sectional correlational analysis. *PLoS One*, *17*(11), e0275680. https://doi.org/10.1371/journal.pone.0275680

Downs, A., & Smith, T. (2004). Emotional Understanding, Cooperation, and Social Behavior in High-Functioning Children with Autism. *Journal of Autism and Developmental Disorders*, *34*(6), 625-635. https://doi.org/10.1007/s10803-004-5284-0

Drouineau, M.-H., Guenego, E., Sebille-Rivain, V., Vrignaud, B., Balençon, M., Blanchais, T., Levieux, K., Vabres, N., Picherot, G., & Guen, C. G.-l. (2017). Do abused young children feel less pain? *Child Abuse & Neglect*, *65*, 248-254. https://doi.org/https://doi.org/10.1016/j.chiabu.2017.02.002

Duarte, M. A., Goulart, E. M. A., & Penna, F. J. (2000). Pressure Pain Threshold in Children With Recurrent Abdominal Pain. *Journal of Pediatric Gastroenterology and Nutrition*, *31*, 280-285. https://doi.org/10.1097/00005176-200009000-00015

Dubin, A. E., & Patapoutian, A. (2010). Nociceptors: the sensors of the pain pathway. *The Journal of Clinical Investigation*, *120*(11), 3760-3772. https://doi.org/10.1172/JCI42843

Dubois, A., Michelon, C., Rattaz, C., Zabalia, M., & Baghdadli, A. (2017). Daily living pain assessment in children with autism: Exploratory study. *Research in Developmental Disabilities*, *62*, 238-246. https://doi.org/10.1016/j.ridd.2017.01.003

Dubois, D., Ameis, S. H., Lai, M.-C., Casanova, M. F., & Desarkar, P. (2016). Interoception in Autism Spectrum Disorder: A review. *International Journal of Developmental Neuroscience*, *52*, 104-111. https://doi.org/10.1016/j.ijdevneu.2016.05.001

Dueñas, M., Ojeda, B., Salazar, A., Mico, J. A., & Failde, I. (2016). A review of chronic pain impact on patients, their social environment and the health care system. *Journal of Pain Research*, *9*, 457-467. https://doi.org/10.2147/jpr.S105892

Duerden, E. G., Card, D., Roberts, S. W., Mak-Fan, K. M., Chakravarty, M. M., Lerch, J. P., & Taylor, M. J. (2014). Self-injurious behaviours are associated with alterations in the somatosensory system in children with autism spectrum disorder. *Brain Structure and Function*, *219*(4), 1251-1261. https://doi.org/10.1007/s00429-013-0562-2

Duerden, E. G., Taylor, M. J., Lee, M., McGrath, P. A., Davis, K. D., & Roberts, S. W. (2015). Decreased Sensitivity to Thermal Stimuli in Adolescents With Autism Spectrum Disorder: Relation to Symptomatology and Cognitive Ability. *The Journal of Pain*, *16*(5), 463-471. https://doi.org/10.1016/j.jpain.2015.02.001

Ebrahimpour, F., Pashaeypoor, S., Salisu, W. J., Cheraghi, M. A., & Sadat Hosseini, A. S. (2019). Children's description of pain through drawings and dialogs: A concept analysis. *Nursing Open*, *6*(2), 301-312. https://doi.org/10.1002/nop2.211

Eccleston, C. (1995). The attentional control of pain: methodological and theoretical concerns. *PAIN*, *63*(1), 3-10. https://doi.org/10.1016/0304-3959(95)00093-8

Eccleston, C., Fisher, E., Howard, R. F., Slater, R., Forgeron, P., Palermo, T. M., Birnie, K. A., Anderson, B. J., Chambers, C. T., Crombez, G., Ljungman, G., Jordan, I., Jordan, Z., Roberts, C., Schechter, N., Sieberg, C. B., Tibboel, D., Walker, S. M., Wilkinson, D., & Wood, C. (2021). Delivering transformative action in paediatric pain:

a Lancet Child & Adolescent Health Commission. *The Lancet Child & Adolescent Health*, *5*(1), 47-87. https://doi.org/10.1016/S2352-4642(20)30277-7
Edelson, S. M. (2021). Comparison of Autistic Individuals Who Engage in Self-Injurious Behavior, Aggression, and Both Behaviors. *Pediatric Reports*, *13*(4), 558-565. 10.3390/pediatric13040066
Edwards, R., Eccleston, C., & Keogh, E. (2017). Observer influences on pain: an experimental series examining same-sex and opposite-sex friends, strangers, and romantic partners. *PAIN*, *158*(5). https://doi.org/10.1097/j.pain.0000000000000840
Eilers, H., & Schumacher, M. A. (2005). Mechanosensitivity of Primary Afferent Nociceptors in the Pain Pathway. In A. Kamkin & I. Kiseleva (Eds.), *Mechanosensitivity in Cells and Tissues*. Academia.
Ekman, P., & Friesen, W. V. (1976). Measuring facial movement. *Environmental psychology and nonverbal behavior*, *1*, 56-75. https://doi.org/10.1007/BF01115465
Ely, E., Chen-Lim, M. L., Carpenter, K. M., II, Wallhauser, E., & Friedlaender, E. (2016). Pain Assessment of Children with Autism Spectrum Disorders. *Journal of Developmental & Behavioral Pediatrics*, *37*(1). https://doi.org/10.1097/DBP.0000000000000240
Emerson, N. D., & Bursch, B. (2020). Communicating with Youth about Pain: Developmental Considerations. *Children (Basel)*, *7*(10). https://doi.org/10.3390/children7100184
Engebretsen, S., Frigessi, A., Engø-Monsen, K., Furberg, A. S., Stubhaug, A., de Blasio, B. F., & Nielsen, C. S. (2018). The peer effect on pain tolerance. *Scandinavian Journal of Pain*, *18*(3), 467-477. https://doi.org/10.1515/sjpain-2018-0060
Eriksen, A. A., Fegran, L., Fredwall, T. E., & Larsen, I. B. (2023). Patients' negative experiences with health care settings brought to light by formal complaints: A qualitative metasynthesis. *Journal of clinical nursing*, *32*(17-18), 5816-5835. https://doi.org/10.1111/jocn.16704
Evans, C., Pavlopoulou, G., Moyse, R., Mills, R., Atkins, C., Towells, J., Nedelcu, I., & Owen, R. (2022). *National Autism Trainer Programme*. https://www.hee.nhs.uk/our-work/autism/national-autism-trainer-programme
Eveleth, R. (n.d.). Beyond the Smiley-Face Pain Scale. https://www.theatlantic.com/health/archive/2015/01/beyond-the-smiley-face-pain-scale/384049/
Failla, M. D., Gerdes, M. B., Williams, Z. J., Moore, D. J., & Cascio, C. J. (2020). Increased pain sensitivity and pain-related anxiety in individuals with autism. *Pain reports*, *5*(6), e861. https://doi.org/10.1097/pr9.0000000000000861
Failla, M. D., Moana-Filho, E. J., Essick, G. K., Baranek, G. T., Rogers, B. P., & Cascio, C. J. (2017). Initially intact neural responses to pain in autism are diminished during sustained pain. *Autism*, *22*(6), 669-683. https://doi.org/10.1177/1362361317696043
Fan, Y.-T., Chen, C., Chen, S.-C., Decety, J., & Cheng, Y. (2013). Empathic arousal and social understanding in individuals with autism: evidence from fMRI and ERP measurements. *Social Cognitive and Affective Neuroscience*, *9*(8), 1203-1213. https://doi.org/10.1093/scan/nst101
Fatima, N., Babu, P. R., Sisinty, V. S., & Tarakji, B. (2014). Pain Perception and Anxiety Levels during Menstrual Cycle Associated with Periodontal Therapy. *International Journal of Dentistry*, *2014*(1), 472926. https://doi.org/https://doi.org/10.1155/2014/472926
Featherstone, C., Sharpe, R., Axford, N., Asthana, S., & Husk, K. (2023). Autistic adults' experiences of managing wellbeing and implications for social prescribing. *Disability & Society*, 1-29. https://doi.org/10.1080/09687599.2023.2263628
Fechner, G. T. (1860). *Elemente der psychophysik* (Vol. 2). Breitkopf u. Härtel.
Ferland, C. E., Villemure, C., Michon, P.-E., Gandhi, W., Ma, M.-L., Chouchou, F., Parent, A. J., Bushnell, M. C., Lavigne, G., Rainville, P., Ware, M. A., Jackson, P. L., Schweinhardt, P., & Marchand, S. (2018). Multicenter assessment of quantitative

sensory testing (QST) for the detection of neuropathic-like pain responses using the topical capsaicin model. *Canadian Journal of Pain*, *2*(1), 266-279. https://doi.org/10.1080/24740527.2018.1525682

Fernández-de-Las-Peñas, C., Ambite-Quesada, S., Rivas-Martínez, I., Ortega-Santiago, R., de-la-Llave-Rincón, A. I., Fernández-Mayoralas, D. M., & Pareja, J. A. (2011). Genetic Contribution of Catechol-O-Methyltransferase Polymorphism (Val158Met) in Children With Chronic Tension-Type Headache. *Pediatric Research*, *70*(4), 395-399. https://doi.org/10.1203/PDR.0b013e318229448a

Fernández-de-Las-Peñas, C., Fernández-Mayoralas, D. M., Ortega-Santiago, R., Ambite-Quesada, S., Gil-Crujera, A., & Fernández-Jaén, A. (2010). Bilateral, wide-spread, mechanical pain sensitivity in children with frequent episodic tension-type headache suggesting impairment in central nociceptive processing. *Cephalalgia*, *30*(9), 1049-1055. https://doi.org/10.1177/0333102410362806

Fernández-Mayoralas, D. M., Fernández-de-las-Peñas, C., Ortega-Santiago, R., Ambite-Quesada, S., Jiménez-García, R., & Fernández-Jaén, A. (2010). Generalized mechanical nerve pain hypersensitivity in children with episodic tension-type headache. *Pediatrics*, *126*(1), e187-194. https://doi.org/10.1542/peds.2010-0012

Ferracini, G. N., Stuginsk-Barbosa, J., Dach, F., & Speciali, J. G. (2014). A comparison pressure pain threshold in pericranial and extracephalic regions in children with migraine. *Pain Medicine*, *15*(4), 702-709. https://doi.org/10.1111/pme.12353

Fillingim, R. B., King, C. D., Ribeiro-Dasilva, M. C., Rahim-Williams, B., & Riley, J. L. (2009). Sex, Gender, and Pain: A Review of Recent Clinical and Experimental Findings. *The Journal of Pain*, *10*(5), 447-485. https://doi.org/10.1016/j.jpain.2008.12.001

Finan, P. H., Hunt, C. A., Mun, C. J., Lerman, S. F., Tennen, H., Smith, M. T., & Haythornthwaite, J. A. (2022). Association of affective state with the assimilation of daily pain expectancy and pain experience. *PAIN*, *163*(11). https://doi.org/10.1097/j.pain.0000000000002624

Fink, R. (2000). Pain assessment: the cornerstone to optimal pain management. *Baylor University Medical Center Proceedings*, *13*(3), 236-239. https://doi.org/10.1080/08998280.2000.11927681

Finke, E. H., McCarthy, J. H., & Sarver, N. A. (2019). Self-perception of friendship style: Young adults with and without autism spectrum disorder. *Autism & Developmental Language Impairments*, *4*, 2396941519855390. https://doi.org/10.1177/2396941519855390

Fischer, A. A. (1986). Pressure threshold meter: Its use for quantification of tender spots. *Archives of Physical Medicine and Rehabilitation*, *67*(11), 836-838. https://doi.org/10.5555/uri:pii:0003999386901802

Fisher, E., Keogh, E., & Eccleston, C. (2016). Adolescents' approach-avoidance behaviour in the context of pain. *PAIN*, *157*(2), 370-376. https://doi.org/10.1097/j.pain.0000000000000383

Fitzpatrick, R., McGuire, B., & Lydon, H. (2022). Improving pain-related communication in children with autism spectrum disorder and intellectual disability. *Paediatric and Neonatal Pain*, *4*. https://doi.org/10.1002/pne2.12076

French, B., Daley, D., Groom, M., & Cassidy, S. (2023). Risks Associated With Undiagnosed ADHD and/or Autism: A Mixed-Method Systematic Review. *Journal of Attention Disorders*, *27*(12), 1393-1410. https://doi.org/10.1177/10870547231176862

Friedrichsdorf, S. J., Giordano, J., Desai Dakoji, K., Warmuth, A., Daughtry, C., & Schulz, C. A. (2016). Chronic Pain in Children and Adolescents: Diagnosis and Treatment of Primary Pain Disorders in Head, Abdomen, Muscles and Joints. *Children (Basel)*, *3*(4). https://doi.org/10.3390/children3040042

Fründt, O., Grashorn, W., Schöttle, D., Peiker, I., David, N., Engel, A. K., Forkmann, K., Wrobel, N., Münchau, A., & Bingel, U. (2017). Quantitative Sensory Testing in adults

with Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders*, *47*(4), 1183-1192. https://doi.org/10.1007/s10803-017-3041-4

Fusar-Poli, L., Brondino, N., Politi, P., & Aguglia, E. (2022). Missed diagnoses and misdiagnoses of adults with autism spectrum disorder. *European Archives of Psychiatry and Clinical Neuroscience*, *272*(2), 187-198. https://doi.org/10.1007/s00406-020-01189-w

Garcia-Villamisar, D., Moore, D., & Garcia-Martínez, M. (2019). Internalizing Symptoms Mediate the Relation Between Acute Pain and Autism in Adults. *Journal of Autism and Developmental Disorders*, *49*(1), 270-278. https://doi.org/10.1007/s10803-018-3765-9

Garfinkel, S. N., Seth, A. K., Barrett, A. B., Suzuki, K., & Critchley, H. D. (2015). Knowing your own heart: distinguishing interoceptive accuracy from interoceptive awareness. *Biological Psychology*, *104*, 65-74. https://doi.org/10.1016/j.biopsycho.2014.11.004

Garfinkel, S. N., Tiley, C., O'Keeffe, S., Harrison, N. A., Seth, A. K., & Critchley, H. D. (2016). Discrepancies between dimensions of interoception in autism: Implications for emotion and anxiety. *Biological Psychology*, *114*, 117-126. https://doi.org/10.1016/j.biopsycho.2015.12.003

Gatchel, R. J., Peng, Y. B., Peters, M. L., Fuchs, P. N., & Turk, D. C. (2007). The biopsychosocial approach to chronic pain: scientific advances and future directions. *Psychological Bulletin*, *133*(4), 581-624. https://doi.org/10.1037/0033-2909.133.4.581

Gentsch, A., & Kuehn, E. (2022). Clinical Manifestations of Body Memories: The Impact of Past Bodily Experiences on Mental Health. *Brain sciences*, *12*(5). https://doi.org/10.3390/brainsci12050594

George, R., & Stokes, M. A. (2017). Gender identity and sexual orientation in autism spectrum disorder. *Autism*, *22*(8), 970-982. https://doi.org/10.1177/1362361317714587

Gesi, C., Migliarese, G., Torriero, S., Capellazzi, M., Omboni, A. C., Cerveri, G., & Mencacci, C. (2021). Gender Differences in Misdiagnosis and Delayed Diagnosis among Adults with Autism Spectrum Disorder with No Language or Intellectual Disability. *Brain sciences*, *11*(7). https://doi.org/10.3390/brainsci11070912

Gilbert-MacLeod, C. A., Craig, K. D., Rocha, E. M., & Mathias, M. D. (2000). Everyday Pain Responses in Children With and Without Developmental Delays. *Journal of Pediatric Psychology*, *25*(5), 301-308. https://doi.org/10.1093/jpepsy/25.5.301

Goffaux, P., Lafrenaye, S., Morin, M., Patural, H., Demers, G., & Marchand, S. (2008). Preterm births: can neonatal pain alter the development of endogenous gating systems? *European Journal of pain*, *12*(7), 945-951. https://doi.org/10.1016/j.ejpain.2008.01.003

Goldbach, J. T., Amaro, H., Vega, W., & Walter, M. D. (2015). The grand challenge of promoting equality by addressing social stigma.

Goldschmidt, J. (2016). What Happened to Paul? Manifestation of Abnormal Pain Response for Individuals With Autism Spectrum Disorder. *Qualitative Health Research*, *27*(8), 1133-1145. https://doi.org/10.1177/1049732316644415

Goubert, L., Eccleston, C., Vervoort, T., Jordan, A., & Crombez, G. (2006). Parental catastrophizing about their child's pain. The parent version of the Pain Catastrophizing Scale (PCS-P): A preliminary validation. *PAIN*, *123*(3), 254-263. https://doi.org/10.1016/j.pain.2006.02.035

Goubert, L., Vervoort, T., Cano, A., & Crombez, G. (2009). Catastrophizing about their children's pain is related to higher parent-child congruency in pain ratings: an experimental investigation. *Eur J Pain*, *13*(2), 196-201. https://doi.org/10.1016/j.ejpain.2008.03.009

Gould, J., & Ashton-Smith, J. (2011). Missed diagnosis or misdiagnosis? Girls and women on the autism spectrum. *Good Autism Practice (GAP)*, *12*(1), 34-41.

Goulding, R., Goodwin, J., O'Donovan, A., & Saab, M. M. (2023). Transgender and gender diverse youths' experiences of healthcare: A systematic review of qualitative studies. *Journal of Child Health Care*, *0*(0), 13674935231222054. https://doi.org/10.1177/13674935231222054

Gowen, E., Taylor, R., Bleazard, T., Greenstein, A., Baimbridge, P., & Poole, D. (2019). Guidelines for conducting research studies with the autism community. *Autism Policy Pract*, *2*(1 A new beginning), 29-45.

Grabli, F. E., Quesque, F., Borg, C., Witthöft, M., Michael, G. A., Lucas, C., Pasquier, F., Lebouvier, T., & Bertoux, M. (2022). Interoception and social cognition in chronic low back pain: a common inference disturbance? An exploratory study. *Pain Management*, *12*(4), 471-485. https://doi.org/10.2217/pmt-2021-0090

Grabovac, I., & Dorner, T. E. (2019). Association between low back pain and various everyday performances. *Wiener klinische Wochenschrift*, *131*(21), 541-549. https://doi.org/10.1007/s00508-019-01542-7

Green, S. A., & Ben-Sasson, A. (2010). Anxiety Disorders and Sensory Over-Responsivity in Children with Autism Spectrum Disorders: Is There a Causal Relationship? *Journal of Autism and Developmental Disorders*, *40*(12), 1495-1504. https://doi.org/10.1007/s10803-010-1007-x

Greenspan, J. D. (2009). Pain Psychophysics. In M. D. Binder, N. Hirokawa, & U. Windhorst (Eds.), *Encyclopedia of Neuroscience* (pp. 3077-3080). Springer Berlin Heidelberg. https://doi.org/10.1007/978-3-540-29678-2_4365

Grove, R., Hoekstra, R. A., Wierda, M., & Begeer, S. (2018). Special interests and subjective wellbeing in autistic adults. *Autism Research*, *11*(5), 766-775. https://doi.org/https://doi.org/10.1002/aur.1931

Grove, R., Roth, I., & Hoekstra, R. A. (2016). The motivation for special interests in individuals with autism and controls: Development and validation of the special interest motivation scale. *Autism Research*, *9*(6), 677-688. https://doi.org/https://doi.org/10.1002/aur.1560

Gulewitsch, M. D., Jusyte, A., Weimer, K., & Schönenberg, M. (2019). Does Social Exclusion Alter Sensory and Pain Thresholds in Children and Adolescents with Functional Abdominal Pain? - Results from a Preliminary Study. *Pain Medicine*, *20*(8), 1472-1478. https://doi.org/10.1093/pm/pny266

Habechian, F. A. P., Lozana, A. L., Cools, A. M., & Camargo, P. R. (2018). Swimming Practice and Scapular Kinematics, Scapulothoracic Muscle Activity, and the Pressure-Pain Threshold in Young Swimmers. *Journal of Athletic Training*, *53*(11), 1056-1062. https://doi.org/10.4085/1062-6050-100-17

Haefeli, M., & Elfering, A. (2006). Pain assessment. *European spine journal*, *15*, S17-S24. https://doi.org/10.1007/s00586-005-1044-x

Hainsworth, K. R., Simpson, P. M., Ali, O., Varadarajan, J., Rusy, L., & Weisman, S. J. (2020). Quantitative Sensory Testing in Adolescents with Co-occurring Chronic Pain and Obesity: A Pilot Study. *Children (Basel)*, *7*(6). https://doi.org/10.3390/children7060055

Han, G. T., Heavner, H. S., Rains, T. R., Hoang, A. H., & Stone, A. L. (2024). Chronic Pain in Autistic Youth: Clinical Prevalence and Reflections on Tailoring Evidence-Based Interventions from an Interdisciplinary Treatment Team. *Children*, *11*(3), 312. https://doi.org/10.3390/children11030312

Han, T.-I., Hong, C.-Z., Kuo, F.-C., Hsieh, Y.-L., Chou, L.-W., & Kao, M.-J. (2012). Mechanical pain sensitivity of deep tissues in children - possible development of myofascial trigger points in children. *BMC Musculoskeletal Disorders*, *13*(1), 13. https://doi.org/10.1186/1471-2474-13-13

Hanlon, C., Ashworth, E., Moore, D., Donaghy, B., & Saini, P. (2022). Autism should be considered in the assessment and delivery of mental health services for children and young people. *Disability & Society*, 1-6. https://doi.org/10.1080/09687599.2022.2099252

Hanssen, M. M., Peters, M. L., Boselie, J. J., & Meulders, A. (2017). Can positive affect attenuate (persistent) pain? State of the art and clinical implications. *Current Rheumatology Reports*, *19*(12), 80. https://doi.org/10.1007/s11926-017-0703-3

Harackiewicz, J. M., Smith, J. L., & Priniski, S. J. (2016). Interest Matters: The Importance of Promoting Interest in Education. *Policy Insights from the Behavioral and Brain Sciences*, *3*(2), 220-227. https://doi.org/10.1177/2372732216655542

Harley, K. G., Watson, A., Robertson, S., Vitzthum, V. J., & Shea, A. (2024). Menstrual Cycle Characteristics of U. S. Adolescents According to Gynecologic Age and Age at Menarche. *Journal of Pediatric and Adolescent Gynecology*, *37*(4), 419-425. https://doi.org/10.1016/j.jpag.2024.03.005

Harmens, M., Sedgewick, F., & Hobson, H. (2022). Autistic women's diagnostic experiences: Interactions with identity and impacts on well-being. *Womens Health (Lond)*, *18*, 17455057221137477. https://doi.org/10.1177/17455057221137477

Harrop, C., Bal, V., Carpenter, K., & Halladay, A. (2021). A lost generation? The impact of the COVID-19 pandemic on early career ASD researchers. *Autism Research*, *14*(6), 1078-1087. https://doi.org/10.1002/aur.2503

Hashkes, P. J., Friedland, O., Jaber, L., Cohen, H. A., Wolach, B., & Uziel, Y. (2004). Decreased pain threshold in children with growing pains. *The Journal of rheumatology*, *31*(3), 610-613.

Haydon, C., Doherty, M., & Davidson, I. A. (2021). Autism: making reasonable adjustments in healthcare. *British Journal of Hospital Medicine*, *82*(12), 1-11. https://doi.org/10.12968/hmed.2021.0314

Hazen, E. P., Stornelli, J. L., O'Rourke, J. A., Koesterer, K., & McDougle, C. J. (2014). Sensory Symptoms in Autism Spectrum Disorders. *Harvard Review of Psychiatry*, *22*(2). https://doi.org/10.1097/01.HRP.0000445143.08773.58

Health Education England. (2022). *The Oliver McGowan Mandatory Training on Learning Disability and Autism*. https://www.hee.nhs.uk/our-work/learning-disability/current-projects/oliver-mcgowan-mandatory-training-learning-disability-autism

Health Standards Organization. (2023). *Pediatric Pain Management*. Health Standards Organization Retrieved from https://healthstandards.org/standard/pediatric-pain-management-can-hso-13200-2023-e/

Henderson, E. F. (2022). Autism, Autonomy, and Touch Avoidance. *Disability Studies Quarterly*, *42*(1). https://doi.org/10.18061/dsq.v42i1.7714

Herr, K., Coyne, P. J., McCaffery, M., Manworren, R., & Merkel, S. (2011). Pain Assessment in the Patient Unable to Self-Report: Position Statement with Clinical Practice Recommendations. *Pain Management Nursing*, *12*(4), 230-250. https://doi.org/https://doi.org/10.1016/j.pmn.2011.10.002

Hicks, C. L., von Baeyer, C. L., Spafford, P. A., van Korlaar, I., & Goodenough, B. (2001). The Faces Pain Scale-Revised: toward a common metric in pediatric pain measurement. *PAIN*, *93*(2), 173-183. https://doi.org/10.1016/s0304-3959(01)00314-1

Hirschfeld, G., Zernikow, B., Kraemer, N., Hechler, T., Aksu, F., Krumova, E., Maier, C., Magerl, W., & Blankenburg, M. (2012). Development of somatosensory perception in children: a longitudinal QST-study. *Neuropediatrics*, *43*(1), 10-16. https://doi.org/10.1055/s-0032-1307450

Hjermstad, M. J., Fayers, P. M., Haugen, D. F., Caraceni, A., Hanks, G. W., Loge, J. H., Fainsinger, R., Aass, N., & Kaasa, S. (2011). Studies Comparing Numerical Rating Scales, Verbal Rating Scales, and Visual Analogue Scales for Assessment of Pain

Intensity in Adults: A Systematic Literature Review. *Journal of Pain and Symptom Management*, *41*(6), 1073-1093. https://doi.org/10.1016/j.jpainsymman.2010.08.016
Hoegh, M., Seminowicz, D. A., & Graven-Nielsen, T. (2019). Delayed effects of attention on pain sensitivity and conditioned pain modulation. *European Journal of pain*, *23*(10), 1850-1862. https://doi.org/https://doi.org/10.1002/ejp.1458
Hoehn, J. L., Dahlquist, L. M., & Zeroth, J. A. (2022). Conditioned Pain Modulation in Children: The Effects of Painful and Nonpainful Conditioning Stimuli. *The Journal of Pain*, *23*(7), 1208-1219. https://doi.org/10.1016/j.jpain.2022.02.004
Hoffman, T., Bar-Shalita, T., Granovsky, Y., Gal, E., Kalingel-Levi, M., Dori, Y., Buxbaum, C., Yarovinsky, N., & Weissman-Fogel, I. (2022). Indifference or hypersensitivity? Solving the riddle of the pain profile in individuals with autism. *PAIN*, 10.1097/j.pain.0000000000002767. https://doi.org/10.1097/j.pain.0000000000002767
Hoffman, T., Bar-Shalita, T., Granovsky, Y., Gal, E., Kalingel-Levi, M., Dori, Y., Buxbaum, C., Yarovinsky, N., & Weissman-Fogel, I. (2023). Indifference or hypersensitivity? Solving the riddle of the pain profile in individuals with autism. *PAIN*, *164*(4), 791-803. https://doi.org/10.1097/j.pain.0000000000002767
Hogeweg, J. A., Kuis, W., Huygen, A. C., de Jong-de vos van Steenwijk, C., Bernards, A. T., Oostendorp, R. A., & Helders, P. J. (1995). The pain threshold in juvenile chronic arthritis. *Rheumatology*, *34*(1), 61-67. https://doi.org/10.1093/rheumatology/34.1.61
Hogeweg, J. A., Kuis, W., Oostendorp, R. A., & Helders, P. J. (1996). The influence of site of stimulation, age, and gender on pain threshold in healthy children. *Physical Therapy*, *76*(12), 1331-1339. https://doi.org/10.1093/ptj/76.12.1331
Hogeweg, J. A., Kuis, W., Oostendorp, R. A. B., & Helders, P. J. M. (1995). General and segmental reduced pain thresholds in juvenile chronic arthritis. *PAIN*, *62*(1), 11-17. https://doi.org/10.1016/0304-3959(94)00207-u
Horgan, F., Kenny, N., & Flynn, P. (2023). A systematic review of the experiences of autistic young people enrolled in mainstream second-level (post-primary) schools. *Autism*, *27*(2), 526-538. https://doi.org/10.1177/13623613221105089
Hsieh, H.-F., & Shannon, S. (2005). Three Approaches to Qualitative Content Analysis. *Qualitative Health Research*, *15*, 1277-1288. https://doi.org/10.1177/1049732305276687
Huang, Y., Arnold, S. R., Foley, K.-R., & Trollor, J. N. (2020). Diagnosis of autism in adulthood: A scoping review. *Autism*, *24*(6), 1311-1327. https://doi.org/10.1177/1362361320903128
Hughes, J. (2016). Increasing neurodiversity in disability and social justice advocacy groups [whitepaper]. *Autistic Self Advocacy Network*.
Hull, L., Petrides, K. V., & Mandy, W. (2020). The Female Autism Phenotype and Camouflaging: a Narrative Review. *Review Journal of Autism and Developmental Disorders*, *7*(4), 306-317. https://doi.org/10.1007/s40489-020-00197-9
IBM Corp. (2020). *IBM SPSS Statistics for Windows*. In (Version 27.0) IBM Corp.
Ingudomnukul, E., Baron-Cohen, S., Wheelwright, S., & Knickmeyer, R. (2007). Elevated rates of testosterone-related disorders in women with autism spectrum conditions. *Hormones and Behavior*, *51*(5), 597-604. https://doi.org/10.1016/j.yhbeh.2007.02.001
Institute of Medicine Committee on Pain, D., & Chronic Illness, B. (1987). In M. Osterweis, A. Kleinman, & D. Mechanic (Eds.), *Pain and Disability: Clinical, Behavioral, and Public Policy Perspectives*. National Academies Press (US). https://doi.org/10.17226/991
International Association for the Study of Pain. (n.d.). *Acute Pain*. https://www.iasp-pain.org/resources/topics/acute-pain/

Jackson, L., Keville, S., & Ludlow, A. K. (2020). Mothers' Experiences of Accessing Mental Health Care for their Child with an Autism Spectrum Disorder. *Journal of Child and Family Studies*, *29*(2), 534-545. https://doi.org/10.1007/s10826-019-01662-8

Jacob, E., Chan, V. W., Hodge, C., Zeltzer, L., Zurakowski, D., & Sethna, N. F. (2015). Sensory and Thermal Quantitative Testing in Children With Sickle Cell Disease. *Journal of Pediatric Hematology/Oncology*, *37*(3), 185-189. https://doi.org/10.1097/mph.0000000000000214

Janevic, M. R., Mathur, V. A., Booker, S. Q., Morais, C., Meints, S. M., Yeager, K. A., & Meghani, S. H. (2022). Making Pain Research More Inclusive: Why and How. *The Journal of Pain*, *23*(5), 707-728. https://doi.org/10.1016/j.jpain.2021.10.004

Jedel, E., Carlsson, J., & Stener-Victorin, E. (2007). Health-related quality of life in child patients with temporomandibular disorder pain. *European Journal of pain*, *11*(5), 557-563. https://doi.org/10.1016/j.ejpain.2006.07.007

Jenkinson, R., Milne, E., & Thompson, A. (2020). The relationship between intolerance of uncertainty and anxiety in autism: A systematic literature review and meta-analysis. *Autism*, *24*(8), 1933-1944. https://doi.org/10.1177/1362361320932437

Jeste, S. S., & Geschwind, D. H. (2014). Disentangling the heterogeneity of autism spectrum disorder through genetic findings. *Nat Rev Neurol*, *10*(2), 74-81. https://doi.org/10.1038/nrneurol.2013.278

Jin, Q., Chang, Y., Lu, C., Chen, L., & Wang, Y. (2023). Referred pain: characteristics, possible mechanisms, and clinical management. *Frontiers in Neurology*, *14*, 1104817. https://doi.org/10.3389/fneur.2023.1104817

Jones, G., & Shivamurthy, V. (2022). OA02 Paediatric chronic pain and Autism Spectrum Disorder: is there a link? *Rheumatology Advances in Practice*, *6*(Supplement_1). https://doi.org/10.1093/rap/rkac066.002

Jordan, A., Noel, M., Caes, L., Connell, H., & Gauntlett-Gilbert, J. (2018). A developmental arrest? Interruption and identity in adolescent chronic pain. *Pain reports*, *3*(7). https://doi.org/10.1097/PR9.0000000000000678

Jordan, A., Parchment, A., Gauntlett-Gilbert, J., Jones, A., Donaghy, B., Wainwright, E., Connell, H., Walden, J., & Moore, D. J. (2024). Understanding the impacts of chronic pain on autistic adolescents and effective pain management: a reflexive thematic analysis adolescent–maternal dyadic study. *Journal of Pediatric Psychology*. https://doi.org/10.1093/jpepsy/jsae004

Jovellar-Isiegas, P., García, C. C., Jaén-Carrillo, D., Palomo-Carrión, R., Alonso, C. P., & Roche-Seruendo, L. E. (2022). Somatosensation and motor performance in the less-affected and more-affected hand of unilateral cerebral palsy children: a cross-sectional study. *Disability and Rehabilitation*. https://doi.org/10.1080/09638288.2022.2127938

Kalandadze, T., Bambini, V., & Næss, K.-A. B. (2019). A systematic review and meta-analysis of studies on metaphor comprehension in individuals with autism spectrum disorder: Do task properties matter? *Applied Psycholinguistics*, *40*(6), 1421-1454. https://doi.org/10.1017/S0142716419000328

Kalandadze, T., Norbury, C., Nærland, T., & Næss, K. B. (2018). Figurative language comprehension in individuals with autism spectrum disorder: A meta-analytic review. *Autism*, *22*(2), 99-117. https://doi.org/10.1177/1362361316668652

Kalingel-Levi, M., Schreuer, N., Granovsky, Y., Bar-Shalita, T., Weissman-Fogel, I., Hoffman, T., & Gal, E. (2022). "When I'm in Pain, Everything Is Overwhelming": Implications of Pain in Adults With Autism on Their Daily Living and Participation. *Frontiers in Psychology*, *13*, Article 911756. https://doi.org/10.3389/fpsyg.2022.911756

Kammers, M. P. M., de Vignemont, F., & Haggard, P. (2010). Cooling the Thermal Grill Illusion through Self-Touch. *Current Biology*, *20*(20), 1819-1822. https://doi.org/10.1016/j.cub.2010.08.038

Kanner, R. (2009). Chapter 1 - Definitions. In C. E. Argoff & G. McCleane (Eds.), *Pain Management Secrets (Third Edition)* (pp. 9-14). Mosby. https://doi.org/10.1016/B978-0-323-04019-8.00001-9

Kapp, S. K. (2020). Introduction to the Neurodiversity Movement. In *Autistic community and the neurodiversity movement: Stories from the frontline* (pp. 330). Springer Nature. https://doi.org/10.1007/978-981-13-8437-0

Kapp, S. K. (2023). Profound Concerns about “Profound Autism”: Dangers of Severity Scales and Functioning Labels for Support Needs. *Education Sciences*, *13*(2), 106. https://doi.org/10.3390/educsci13020106

Kapp, S. K., Steward, R., Crane, L., Elliott, D., Elphick, C., Pellicano, E., & Russell, G. (2019). 'People should be allowed to do what they like': Autistic adults' views and experiences of stimming. *Autism*, *23*(7), 1782-1792. https://doi.org/10.1177/1362361319829628

Karos, K., Alleva, J. M., & Peters, M. L. (2018). Pain, Please: An Investigation of Sampling Bias in Pain Research. *The Journal of Pain*, *19*(7), 787-796. https://doi.org/10.1016/j.jpain.2018.02.011

Kasirer, A., & Mashal, N. (2016). Comprehension and generation of metaphors by children with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *32*, 53-63. https://doi.org/10.1016/j.rasd.2016.08.003

Kelly, Y. (2022). Adolescent Health and Wellbeing in the UK. In A. Heinen, R. Samuel, C. Vögele, & H. Willems (Eds.), *Wohlbefinden und Gesundheit im Jugendalter: Theoretische Perspektiven, empirische Befunde und Praxisansätze* (pp. 647-664). Springer Fachmedien Wiesbaden. https://doi.org/10.1007/978-3-658-35744-3_29

Kennedy, N., Nelson, S., Jerome, R. N., Edwards, T. L., Stroud, M., Wilkins, C. H., & Harris, P. A. (2022). Recruitment and retention for chronic pain clinical trials: a narrative review. *Pain reports*, *7*(4). https://doi.org/10.1097/PR9.0000000000001007

Kentrou, V., Livingston, L. A., Grove, R., Hoekstra, R. A., & Begeer, S. (2024). Perceived misdiagnosis of psychiatric conditions in autistic adults. *EClinicalMedicine*, *71*. https://doi.org/10.1016/j.eclinm.2024.102586

Kersch, A., Perera, P., Mercado, M., Gorrie, A., Sainsbury, D., McGrath, T., Aouad, P., Sarraf, S., Jaaniste, T., & Champion, D. (2020). Somatosensory Testing in Pediatric Patients with Chronic Pain: An Exploration of Clinical Utility. *Children*, *7*(12), 275. https://doi.org/10.3390/children7120275

Khan, R. U., Oon, Y. B., Haq, M. I. U., & Hajarah, S. (2018). Proposed user interface design criteria for children with dyslexia. *International Journal of Engineering & Technology*, *7*(4), 5253-5257. https://doi.org/10.14419/ijet.v7i4.25496

Khera, T., & Rangasamy, V. (2021). Cognition and Pain: A Review [Review]. *Frontiers in Psychology*, *12*. https://doi.org/10.3389/fpsyg.2021.673962

Kiel, E. J., Price, N. N., & Premo, J. E. (2020). Maternal comforting behavior, toddlers' dysregulated fear, and toddlers' emotion regulatory behaviors. *Emotion*, *20*(5), 793-803. https://doi.org/10.1037/emo0000600

King, C. D., Jastrowski Mano, K. E., Barnett, K. A., Pfeiffer, M., Ting, T. V., & Kashikar-Zuck, S. (2017). Pressure Pain Threshold and Anxiety in Adolescent Females With and Without Juvenile Fibromyalgia: A Pilot Study. *The Clinical Journal of Pain*, *33*(7), 620-626. https://doi.org/10.1097/ajp.0000000000000444

Kirkpatrick, D. R., McEntire, D. M., Hambsch, Z. J., Kerfeld, M. J., Smith, T. A., Reisbig, M. D., Youngblood, C. F., & Agrawal, D. K. (2015). Therapeutic Basis of Clinical Pain Modulation. *Clinical and Translational Science*, *8*(6), 848-856. https://doi.org/https://doi.org/10.1111/cts.12282

Kjeldgaard Pedersen, L., Fisker, L. Y. V., Rölfing, J. D., Ahlburg, P., Veien, M., Vase, L., & Møller-Madsen, B. (2023). Virtual reality increases pressure pain threshold and lowers anxiety in children compared with control and non-immersive control-A randomized, crossover trial. *European Journal of pain*, *27*(7), 805-815. https://doi.org/10.1002/ejp.2108

Kourti, M., & MacLeod, A. (2018). “I Don't Feel Like a Gender, I Feel Like Myself”: Autistic Individuals Raised as Girls Exploring Gender Identity. *Autism in Adulthood*, *1*(1), 52-59. https://doi.org/10.1089/aut.2018.0001

Kragelund, L. (2013). The obser-view: a method of generating data and learning. *Nurse Researcher*, *20*(5), 6-10. https://doi.org/10.7748/nr2013.05.20.5.6.e296

Kragelund, L., Moser, A., & van Zadelhoff, E. (2015). Using the Obser-View in Qualitative Research: Benefits and Challenges. *International Journal of Qualitative Methods*, *14*(5), 1609406915621426. https://doi.org/10.1177/1609406915621426

Krahé, C., & Fotopoulou, A. (2018). Psychological and neurobiological processes in coping with pain: The role of social interactions. In *The Routledge International Handbook of Psychobiology* (pp. 73-92). Routledge.

Krahé, C., Springer, A., Weinman, J., & Fotopoulou, A. (2013). The Social Modulation of Pain: Others as Predictive Signals of Salience – a Systematic Review [Review]. *Frontiers in Human Neuroscience*, *7*. https://doi.org/10.3389/fnhum.2013.00386

Langhinrichsen-Rohling, J., Lewis, C. L., McCabe, S., Lathan, E. C., Agnew, G. A., Selwyn, C. N., & Gigler, M. E. (2021). They’ve been BITTEN: reports of institutional and provider betrayal and links with Ehlers-Danlos Syndrome patients’ current symptoms, unmet needs and healthcare expectations. *Therapeutic Advances in Rare Disease*, *2*, 26330040211022033. https://doi.org/10.1177/26330040211022033

Lautenbacher, S., Peters, J. H., Heesen, M., Scheel, J., & Kunz, M. (2017). Age changes in pain perception: A systematic-review and meta-analysis of age effects on pain and tolerance thresholds. *Neuroscience & Biobehavioral Reviews*, *75*, 104-113. https://doi.org/10.1016/j.neubiorev.2017.01.039

Lavand’homme, P. (2011). The progression from acute to chronic pain. *Current Opinion in Anesthesiology*, *24*(5). https://doi.org/10.1097/ACO.0b013e32834a4f74

Leone, C., Galosi, S., Mollica, C., Fortunato, M., Possidente, C., Milone, V., Misuraca, S., Berillo, L., Truini, A., Cruccu, G., Ferrara, M., & Terrinoni, A. (2021). Dissecting pain processing in adolescents with Non-Suicidal Self Injury: Could suicide risk lurk among the electrodes? *European Journal of pain*, *25*(8), 1815-1828. https://doi.org/10.1002/ejp.1793

LeResche, L. (1982). Facial expression in pain: A study of candid photographs. *Journal of Nonverbal Behavior*, *7*(1), 46-56. https://doi.org/10.1007/BF01001777

Lewis, L. F. (2016a). Exploring the Experience of Self-Diagnosis of Autism Spectrum Disorder in Adults. *Archives of Psychiatric Nursing*, *30*(5), 575-580. https://doi.org/10.1016/j.apnu.2016.03.009

Lewis, L. F. (2016b). Realizing a diagnosis of autism spectrum disorder as an adult. *International Journal of Mental Health Nursing*, *25*(4), 346-354. https://doi.org/10.1111/inm.12200

Lewis, L. F. (2017). A Mixed Methods Study of Barriers to Formal Diagnosis of Autism Spectrum Disorder in Adults. *Journal of Autism and Developmental Disorders*, *47*(8), 2410-2424. https://doi.org/10.1007/s10803-017-3168-3

Leyfer, O. T., Folstein, S. E., Bacalman, S., Davis, N. O., Dinh, E., Morgan, J., Tager-Flusberg, H., & Lainhart, J. E. (2006). Comorbid Psychiatric Disorders in Children with Autism: Interview Development and Rates of Disorders. *Journal of Autism and Developmental Disorders*, *36*(7), 849-861. https://doi.org/10.1007/s10803-006-0123-0

Li, R., Holley, A. L., Palermo, T. M., Ohls, O., Edwards, R. R., & Rabbitts, J. A. (2023). Feasibility and reliability of a quantitative sensory testing protocol in youth with acute musculoskeletal pain postsurgery or postinjury. *PAIN*, *164*(7), 1627-1638. https://doi.org/10.1097/j.pain.0000000000002865

Li, T., Xiong, Q., Shi, R., & Yi, L. (2024). Autistic and Non-autistic Children's Pain Perception is Modulated by Their First-Hand Pain Sensitivity and Theory of Mind. *Research on Child and Adolescent Psychopathology*. https://doi.org/10.1007/s10802-024-01176-y

Lieber, S., Blankenburg, M., Apel, K., Hirschfeld, G., Hernáiz Driever, P., & Reindl, T. (2018). Small-fiber neuropathy and pain sensitization in survivors of pediatric acute lymphoblastic leukemia. *European Journal of Paediatric Neurology*, *22*(3), 457-469. https://doi.org/10.1016/j.ejpn.2017.12.019

Lindly, O., Thorburn, S., Heisler, K., Reyes, N., & Zuckerman, K. (2017). Parent disclosure of complementary health approaches used for children with autism spectrum disorder: Barriers and facilitators. *Complementary Therapies in Medicine*, *35*, 47-52. https://doi.org/10.1016/j.ctim.2017.09.003

Linton, S. J., & Shaw, W. S. (2011). Impact of Psychological Factors in the Experience of Pain. *Physical Therapy*, *91*(5), 700-711. https://doi.org/10.2522/ptj.20100330

Lipsker, C. W., Bölte, S., Hirvikoski, T., Lekander, M., Holmström, L., & Wicksell, R. K. (2018). Prevalence of autism traits and attention-deficit hyperactivity disorder symptoms in a clinical sample of children and adolescents with chronic pain. *Journal of Pain Research*, *11*, 2827-2836. https://doi.org/10.2147/jpr.S177534

Lipsker, C. W., Hirvikoski, T., Balter, L. J. T., Bölte, S., Lekander, M., Holmström, L., & Wicksell, R. K. (2021). Autistic Traits and Attention-Deficit Hyperactivity Disorder Symptoms Associated With Greater Pain Interference and Depression, and Reduced Health-Related Quality of Life in Children With Chronic Pain. *Frontiers in Neuroscience*, *15*, 716887. https://doi.org/10.3389/fnins.2021.716887

Liu, J., Chen, L. L., Shen, S., Mao, J., Lopes, M., Liu, S., & Kong, X. (2020). Challenges in the Diagnosis and Management of Pain in Individuals with Autism Spectrum Disorder. *Review Journal of Autism and Developmental Disorders*, *7*(4), 352-363. https://doi.org/10.1007/s40489-020-00199-7

Loeser, J. D. (1980). Perspectives on Pain. In P. Turner, C. Padgham, & A. Hedges (Eds.), *Clinical Pharmacology & Therapeutics: Proceedings of Plenary Lectures Symposia and Therapeutic Sessions of the First World Conference on Clinical Pharmacology & Therapeutics London, UK, 3–9 August 1980* (pp. 313-316). Palgrave Macmillan UK. https://doi.org/10.1007/978-1-349-05952-2_34

Loeser, J. D. (2000). Pain and Suffering. *The Clinical Journal of Pain*, *16*(2), S2-S6. https://doi.org/10.1097/00002508-200006001-00002

Loeser, J. D. (2006). Chapter 2 Pain as a disease. In F. Cervero & T. S. Jensen (Eds.), *Handbook of Clinical Neurology* (Vol. 81, pp. 11-20). Elsevier. https://doi.org/10.1016/S0072-9752(06)80006-0

Logan, D. E., Simons, L. E., Stein, M. J., & Chastain, L. (2008). School Impairment in Adolescents With Chronic Pain. *The Journal of Pain*, *9*(5), 407-416. https://doi.org/10.1016/j.jpain.2007.12.003

Loizzo, A., Loizzo, S., & Capasso, A. (2009). Neurobiology of pain in children: an overview. *The Open Biochemistry Journal*, *3*, 18-25. https://doi.org/10.2174/1874091x00903010018

Lord, C., Charman, T., Havdahl, A., Carbone, P., Anagnostou, E., Boyd, B., Carr, T., De Vries, P. J., Dissanayake, C., & Divan, G. (2022). The Lancet Commission on the future of care and clinical research in autism. *The Lancet*, *399*(10321), 271-334. https://doi.org/10.1016/S0140-6736(21)01541-5

Ludäscher, P., von Kalckreuth, C., Parzer, P., Kaess, M., Resch, F., Bohus, M., Schmahl, C., & Brunner, R. (2015). Pain perception in female adolescents with borderline

personality disorder. *European Child & Adolescent Psychiatry*, *24*(3), 351-357. https://doi.org/10.1007/s00787-014-0585-0

Lydersen, S., Pradhan, V., Senchaudhuri, P., & Laake, P. (2007). Choice of test for association in small sample unordered r × c tables. *Statistics in Medicine*, *26*(23), 4328-4343. https://doi.org/doi.org/10.1002/sim.2839

Lyng, K. D., Sorensen, L. B., Olesen, J. L., Rathleff, M. S., & Holden, S. (2023). Do adolescents with Osgood-Schlatter display nociplastic pain manifestations compared to controls: A cross-sectional study*. *Journal of Science and Medicine in Sport*, *26*(7), 358-364. https://doi.org/10.1016/j.jsams.2023.05.005

Macdonald, H., Rutter, M., Howlin, P., Rios, P., Conteur, A. L., Evered, C., & Folstein, S. (1989). Recognition and Expression of Emotional Cues by Autistic and Normal Adults. *Journal of Child Psychology and Psychiatry*, *30*(6), 865-877. https://doi.org/10.1111/j.1469-7610.1989.tb00288.x

Maddox, B. B., & White, S. W. (2015). Comorbid Social Anxiety Disorder in Adults with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *45*(12), 3949-3960. https://doi.org/10.1007/s10803-015-2531-5

Madipakkam, A. R., Rothkirch, M., Dziobek, I., & Sterzer, P. (2017). Unconscious avoidance of eye contact in autism spectrum disorder. *Sci Rep*, *7*(1), 13378. https://doi.org/10.1038/s41598-017-13945-5

Magerl, W., Enax-Krumova, E., Baron, R., Toelle, T., Treede, R.-D., & Maier, C. (2010). Reference data for quantitative sensory testing (QST): Refined stratification for age and a novel method for statistical comparison of group data. *PAIN*, *151*, 598-605. https://doi.org/10.1016/j.pain.2010.07.026

Mallen, C. D., Peat, G., Thomas, E., & Croft, P. R. (2006). Is chronic pain in adulthood related to childhood factors? A population-based case-control study of young adults. *The Journal of rheumatology*, *33*(11), 2286-2290. https://www.jrheum.org/content/jrheum/33/11/2286.full.pdf

Mancini, F., Nash, T., Iannetti, G. D., & Haggard, P. (2014). Pain relief by touch: a quantitative approach. *PAIN*, *155*(3), 635-642. https://doi.org/10.1016/j.pain.2013.12.024

Mandy, W., Midouhas, E., Hosozawa, M., Cable, N., Sacker, A., & Flouri, E. (2022). Mental health and social difficulties of late-diagnosed autistic children, across childhood and adolescence. *Journal of Child Psychology and Psychiatry*, *63*(11), 1405-1414. https://doi.org/10.1111/jcpp.13587

Manocha, S., & Taneja, N. (2016). Assessment of paediatric pain: a critical review. *Journal of Basic and Clinical Physiology and Pharmacology*, *27*(4), 323-331. https://doi.org/10.1515/jbcpp-2015-0041

Marche, T. A., Briere, J. L., & von Baeyer, C. L. (2015). Children's Forgetting of Pain-Related Memories. *Journal of Pediatric Psychology*, *41*(2), 220-231. https://doi.org/10.1093/jpepsy/jsv111

Margolis, K. G., Buie, T. M., Turner, J. B., Silberman, A. E., Feldman, J. F., Murray, K. F., McSwiggan-Hardin, M., Levy, J., Bauman, M. L., Veenstra-VanderWeele, J., Whitaker, A. H., & Winter, H. S. (2019). Development of a Brief Parent-Report Screen for Common Gastrointestinal Disorders in Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *49*(1), 349-362. https://doi.org/10.1007/s10803-018-3767-7

Martel, M.-O., Trost, Z., & Sullivan, M. J. (2012). The Expression of Pain Behaviors in High Catastrophizers: The Influence of Automatic and Controlled Processes. *The Journal of Pain*, *13*(8), 808-815. https://doi.org/10.1016/j.jpain.2012.05.015

Mason, D., Ingham, B., Urbanowicz, A., Michael, C., Birtles, H., Woodbury-Smith, M., Brown, T., James, I., Scarlett, C., Nicolaidis, C., & Parr, J. R. (2019). A Systematic Review of What Barriers and Facilitators Prevent and Enable Physical Healthcare

Services Access for Autistic Adults. *Journal of Autism and Developmental Disorders*, *49*(8), 3387-3400. https://doi.org/10.1007/s10803-019-04049-2

Mathews, L. (2011). Pain in children: neglected, unaddressed and mismanaged. *Indian Journal of Palliative Care*, *17*(Suppl), S70-73. https://doi.org/10.4103/0973-1075.76247

Mazurek, M. O., Keefer, A., Shui, A., & Vasa, R. A. (2014). One-year course and predictors of abdominal pain in children with autism spectrum disorders: The role of anxiety and sensory over-responsivity. *Research in Autism Spectrum Disorders*, *8*(11), 1508-1515. https://doi.org/10.1016/j.rasd.2014.07.018

Mazurek, M. O., Vasa, R. A., Kalb, L. G., Kanne, S. M., Rosenberg, D., Keefer, A., Murray, D. S., Freedman, B., & Lowery, L. A. (2013). Anxiety, Sensory Over-Responsivity, and Gastrointestinal Problems in Children with Autism Spectrum Disorders. *Journal of Abnormal Child Psychology*, *41*(1), 165-176. https://doi.org/10.1007/s10802-012-9668-x

McClelland, L. E., & McCubbin, J. A. (2008). Social influence and pain response in women and men. *Journal of Behavioral Medicine*, *31*(5), 413-420. https://doi.org/10.1007/s10865-008-9163-6

McCormack, G., Dillon, A. C., Healy, O., Walsh, C., & Lydon, S. (2020). Primary Care Physicians' Knowledge of Autism and Evidence-Based Interventions for Autism: A Systematic Review. *Review Journal of Autism and Developmental Disorders*, *7*(3), 226-241. https://doi.org/10.1007/s40489-019-00189-4

McDonald, T. A. M. (2020). Autism Identity and the "Lost Generation": Structural Validation of the Autism Spectrum Identity Scale (ASIS) and Comparison of Diagnosed and Self-Diagnosed Adults on the Autism Spectrum. *Autism Adulthood*, *2*(1), 13-23. https://doi.org/10.1089/aut.2019.0069

McEntire, D. M., Kirkpatrick, D. R., Dueck, N. P., Kerfeld, M. J., Smith, T. A., Nelson, T. J., Reisbig, M. D., & Agrawal, D. K. (2016). Pain transduction: a pharmacologic perspective. *Expert review of clinical pharmacology*, *9*(8), 1069-1080. https://doi.org/10.1080/17512433.2016.1183481

McGrath, P., Finley, A., Ritchie, J., Dowden, S., & Med. (1994). Pain, Pain Go Away: Helping Children with Pain. https://www.rch.org.au/uploadedFiles/Main/Content/anaes/Pain_go_away.pdf

McGrath, P. A., & Brown, S. C. (2006). Quantitative Sensory Testing in children: practical considerations for research and clinical practice. *PAIN*, *123*(1-2), 1-2. https://doi.org/10.1016/j.pain.2006.03.024

McGrath, P. A., Seifert, C. E., Speechley, K. N., Booth, J. C., Stitt, L., & Gibson, M. C. (1996). A new analogue scale for assessing children's pain: an initial validation study. *PAIN*, *64*(3), 435-443. https://doi.org/10.1016/0304-3959(95)00171-9

McMakin, D. L., & Alfano, C. A. (2015). Sleep and anxiety in late childhood and early adolescence. *Current Opinion in Psychiatry*, *28*(6), 483-489. https://doi.org/10.1097/yco.0000000000000204

McMurtry, C. M., Pillai Riddell, R., Taddio, A., Racine, N., Asmundson, G. J. G., Noel, M., Chambers, C. T., Shah, V., HelpinKids, & Adults, T. (2015). Far From "Just a Poke": Common Painful Needle Procedures and the Development of Needle Fear. *The Clinical Journal of Pain*, *31*. https://doi.org/10.1097/AJP.0000000000000272

Means-Christensen, A. J., Roy-Byrne, P. P., Sherbourne, C. D., Craske, M. G., & Stein, M. B. (2008). Relationships among pain, anxiety, and depression in primary care. *Depression and Anxiety*, *25*(7), 593-600. https://doi.org/https://doi.org/10.1002/da.20342

Meier, P. M., Berde, C. B., DiCanzio, J., Zurakowski, D., & Sethna, N. F. (2001). Quantitative assessment of cutaneous thermal and vibration sensation and thermal pain

detection thresholds in healthy children and adolescents. *Muscle Nerve*, *24*(10), 1339-1345. https://doi.org/10.1002/mus.1153
Melzack, R. (1975). The McGill Pain Questionnaire: major properties and scoring methods. *PAIN*, *1*(3), 277-299. https://doi.org/10.1016/0304-3959(75)90044-5
Melzack, R. (1987). The short-form McGill pain questionnaire. *PAIN*, *30*(2), 191-197. https://doi.org/10.1016/0304-3959(87)91074-8
Melzack, R. (1996). Gate control theory: On the evolution of pain concepts. *Pain Forum*, *5*(2), 128-138. https://doi.org/10.1016/S1082-3174(96)80050-X
Melzack, R., & Wall, P. D. (1965). Pain mechanisms: a new theory. *Science*, *150*(3699), 971-979. https://doi.org/10.1126/science.150.3699.971
Mensink, M. O., Eijkelkamp, N., Veldhuijzen, D. S., & Wulffraat, N. M. (2022). Feasibility of quantitative sensory testing in juvenile idiopathic arthritis. *Pediatric Rheumatology*, *20*(1), 63. https://doi.org/10.1186/s12969-022-00715-5
Merkel, S., Voepel-Lewis, T., & Malviya, S. (2002). Pain Assessment in Infants and Young Children: The FLACC Scale: A behavioral tool to measure pain in young children. *AJN The American Journal of Nursing*, *102*(10). https://doi.org/10.1097/00000446-200210000-00024
Merkel, S. I., Voepel-Lewis, T., Shayevitz, J. R., & Malviya, S. (1997). The FLACC: a behavioral scale for scoring postoperative pain in young children. *Pediatric Nursing*, *23*(3), 293-297. https://pubmed.ncbi.nlm.nih.gov/9220806/
Metsahonkala, L., Anttila, P., Laimi, K., Aromaa, M., Helenius, H., Mikkelsson, M., Jäppilä, E., Viander, S., Sillanpää, M., & Salminen, J. (2006). Extracephalic tenderness and pressure pain threshold in children with headache. *European Journal of pain*, *10*(7), 581-585. https://doi.org/10.1016/j.ejpain.2005.08.005
Michaelides, A., & Zis, P. (2019). Depression, anxiety and acute pain: links and management challenges. *Postgraduate Medicine*, *131*(7), 438-444. https://doi.org/10.1080/00325481.2019.1663705
Mieronkoski, R., Syrjälä, E., Jiang, M., Rahmani, A., Pahikkala, T., Liljeberg, P., & Salanterä, S. (2020). Developing a pain intensity prediction model using facial expression: A feasibility study with electromyography. *PLoS One*, *15*(7), e0235545. https://doi.org/10.1371/journal.pone.0235545
Militerni, R., Bravaccio, C., Falco, C., Puglisi-Allegra, S., Pascucci, T., & Fico, C. (2000). Pain reactivity in children with autistic disorder. *The Journal of Headache and Pain*, *1*(1), 53-56. https://doi.org/10.1007/s101940050011
Mills, S. E. E., Nicolson, K. P., & Smith, B. H. (2019). Chronic pain: a review of its epidemiology and associated factors in population-based studies. *British Journal of Anaesthesia*, *123*(2), e273-e283. https://doi.org/10.1016/j.bja.2019.03.023
Milner, V., McIntosh, H., Colvert, E., & Happé, F. (2019). A Qualitative Exploration of the Female Experience of Autism Spectrum Disorder (ASD). *Journal of Autism and Developmental Disorders*, *49*(6), 2389-2402. https://doi.org/10.1007/s10803-019-03906-4
Milton, D. E. M. (2012). On the ontological status of autism: the 'double empathy problem'. *Disability & Society*, *27*(6), 883-887. https://doi.org/10.1080/09687599.2012.710008
Mingins, J. E., Tarver, J., Waite, J., Jones, C., & Surtees, A. D. R. (2020). Anxiety and intellectual functioning in autistic children: A systematic review and meta-analysis. *Autism*, *25*(1), 18-32. https://doi.org/10.1177/1362361320953253
Miserandino, C. (2003). The Spoon Theory. *ButYouDontLookSick.com*. https://butyoudontlooksick.com/articles/written-by-christine/the-spoon-theory/
Moayedi, M., & Davis, K. D. (2013). Theories of pain: from specificity to gate control. *Journal of Neurophysiology*, *109*(1), 5-12. https://doi.org/10.1152/jn.00457.2012
Moore, D. J. (2014). Acute pain experience in individuals with autism spectrum disorders: A review. *Autism*, *19*(4), 387-399. https://doi.org/10.1177/1362361314527839

Moore, D. J., Keogh, E., & Eccleston, C. (2012). The interruptive effect of pain on attention. *Quarterly Journal of Experimental Psychology*, *65*(3), 565-586. https://doi.org/10.1080/17470218.2011.626865

Morley, S. (2008). Psychology of pain. *BJA: British Journal of Anaesthesia*, *101*(1), 25-31. https://doi.org/10.1093/bja/aen123

Morley, S. (2016). Bias and reliability in pain ratings. *PAIN*, *157*(5). https://doi.org/10.1097/j.pain.0000000000000448

Morris, M. C., Bruehl, S., Stone, A. L., Garber, J., Smith, C., Palermo, T. M., & Walker, L. S. (2022). Place and Pain: Association Between Neighborhood SES and Quantitative Sensory Testing Responses in Youth With Functional Abdominal Pain. *Journal of Pediatric Psychology*, *47*(4), 446-455. https://doi.org/10.1093/jpepsy/jsab113

Morrison, K. E., DeBrabander, K. M., Jones, D. R., Faso, D. J., Ackerman, R. A., & Sasson, N. J. (2020). Outcomes of real-world social interaction for autistic adults paired with autistic compared to typically developing partners. *Autism*, *24*(5), 1067-1080. https://doi.org/10.1177/1362361319892701

Morsanyi, K., Stamenković, D., & Holyoak, K. J. (2020). Metaphor processing in autism: A systematic review and meta-analysis. *Developmental Review*, *57*, 100925. https://doi.org/10.1016/j.dr.2020.100925

MRC Systems. (2010a). *PinPrick Stimulator Set*. In https://www.mrc-systems.de/en/products/pinprick#:~:text=MRC's%20PinPrick%20stimulators%20are%20used,nociceptor%20activation%20of%20the%20skin

MRC Systems. (2010b). PinPrick Stimulator Set User Manual. In (Vol. 3, pp. 2). https://www.mrc-systems.de/en/products/pinprick#produktbeschreibung: MRC Systems.

Munday, I., Kneebone, I., Rogers, K., & Newton-John, T. (2022). The Language of Pain: Is There a Relationship Between Metaphor Use and Adjustment to Chronic Pain? *Pain Medicine*, *23*(12), 2073-2084. https://doi.org/10.1093/pm/pnaa467

Murphy, J., Catmur, C., & Bird, G. (2019). Classifying individual differences in interoception: Implications for the measurement of interoceptive awareness. *Psychonomic Bulletin & Review*, *26*(5), 1467-1471. https://doi.org/10.3758/s13423-019-01632-7

Murray, G. M. (2009). Guest Editorial: referred pain. *Journal of Applied Oral Science*, *17*(6), i. https://doi.org/10.1590/s1678-77572009000600001

Muskat, B., Burnham Riosa, P., Nicholas, D. B., Roberts, W., Stoddart, K. P., & Zwaigenbaum, L. (2015). Autism comes to the hospital: The experiences of patients with autism spectrum disorder, their parents and health-care providers at two Canadian paediatric hospitals. *Autism*, *19*(4), 482-490. https://doi.org/10.1177/1362361314531341

Nader, R., Oberlander, T. F., Chambers, C. T., & Craig, K. D. (2004). Expression of pain in children with autism. *The Clinical Journal of Pain*, *20*(2), 88-97. https://doi.org/10.1097/00002508-200403000-00005

National Autistic Society. (2020a). Dealing with change - a guide for all audiences. https://www.autism.org.uk/advice-and-guidance/topics/behaviour/dealing-with-change/all-audiences#H2_1

National Autistic Society. (2020b). *Obsessions and repetitive behaviour*. https://www.autism.org.uk/advice-and-guidance/topics/behaviour/obsessions/all-audiences#:~:text=Intense%20interests,-%E2%80%9CMy%20mind%20was&text=Many%20autistic%20people%20have%20intense,dinosaurs%20or%20particular%20cartoon%20characters.

National Autistic Society. (2020c). *Stimming*. https://www.autism.org.uk/advice-and-guidance/topics/behaviour/stimming

National Autistic Society. (2020d). *Understanding and developing communication*. https://www.autism.org.uk/advice-and-guidance/topics/communication/understanding-and-developing-communication
National Autistic Society. (2021). *Education*. https://www.autism.org.uk/what-we-do/campaign/education#:~:text=Seven%20in%20ten%20autistic%20children,14%25%20for%20secondary%20school%20teachers.
National Institute for Health and Care Excellence (NICE). (2021a). *Autism spectrum disorder in under 19s: recognition, referral and diagnosis (CG128)*. (CG128). https://www.nice.org.uk/guidance/cg128: National Institute for Health and Care Excellence (NICE)
National institute for Health and Care Excellence (NICE). (2021b). *Patient experience in adult NHS services: improving the experience of care for people using adult NHS services*. Retrieved from https://www.nice.org.uk/guidance/cg138
Navratilova, E., & Porreca, F. (2014). Reward and motivation in pain and pain relief. *Nature neuroscience*, *17*, 1304-1312. https://doi.org/10.1038/nn.3811
Nehemkis, A. M., & Charter, R. A. (1984). The limits of verbal pain descriptors. *Perceptual and Motor Skills*, *59*(1), 251-254. https://doi.org/10.2466/pms.1984.59.1.251
Neil, L., Olsson, N. C., & Pellicano, E. (2016). The Relationship Between Intolerance of Uncertainty, Sensory Sensitivities, and Anxiety in Autistic and Typically Developing Children. *Journal of Autism and Developmental Disorders*, *46*(6), 1962-1973. https://doi.org/10.1007/s10803-016-2721-9
Neilson, C., & Bond, C. (2023). The experience of anxiety for autistic children and young people: A thematic synthesis review. *Research in Autism Spectrum Disorders*, *109*, 102274. https://doi.org/10.1016/j.rasd.2023.102274
Newton, B. J., Southall, J. L., Raphael, J. H., Ashford, R. L., & LeMarchand, K. (2013). A Narrative Review of the Impact of Disbelief in Chronic Pain. *Pain Management Nursing*, *14*(3), 161-171. https://doi.org/10.1016/j.pmn.2010.09.001
NHLBI. (2013). *Study Quality Assessment Tools*. https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools
NHS. (2022). *What is autism?* NHS.co.uk. https://www.nhs.uk/conditions/autism/what-is-autism/
NHS Digital. (2023). *Autism Statistics, January to December 2022*. Retrieved from https://digital.nhs.uk/data-and-information/publications/statistical/autism-statistics/january-to-december-2022#
Nicolaidis, C., Raymaker, D., McDonald, K., Dern, S., Boisclair, W. C., Ashkenazy, E., & Baggs, A. (2013). Comparison of Healthcare Experiences in Autistic and Non-Autistic Adults: A Cross-Sectional Online Survey Facilitated by an Academic-Community Partnership. *Journal of General Internal Medicine*, *28*(6), 761-769. https://doi.org/10.1007/s11606-012-2262-7
Nicolardi, V., Fanizza, I., Accogli, G., Macchitella, L., Scoditti, S., & Trabacca, A. (2023). Pain assessment in autism: updating the ethical and methodological challenges through a state-of-the-art review. *Neurological Sciences*, *44*(11), 3853-3861. https://doi.org/10.1007/s10072-023-06942-2
NIHR INVOLVE. (2018). Guidance on co-producing a research project. In: National Institute for Health and Care Research (NIHR).
Nikolajsen, L., Kristensen, A., Pedersen, L., Rahbek, O., Jensen, T., & Møller-Madsen, B. (2011). Intra- and interrater agreement of pressure pain thresholds in children with orthopedic disorders. *Journal of children's orthopaedics*, *5*, 173-178. https://doi.org/10.1007/s11832-011-0336-4
Noyek, S., Jessa, J. S., Faulkner, V., Boerner, K. E., Dewan, T., Doyle, D., Genik, L., Grainger-Schatz, S., McMorris, C., McMurtry, C. M., Nania, C. G., Oberlander, T., Lorenzetti, D., Turner, K., & Birnie, K. A. (2023). A systematic review of self and

observer assessment of pain and related functioning in youth with brain-based developmental disabilities. *PAIN*, 10.1097/j.pain.0000000000003066. https://doi.org/10.1097/j.pain.0000000000003066

Nwaordu, G., & Charlton, R. A. (2023). Repetitive Behaviours in Autistic and Non-Autistic Adults: Associations with Sensory Sensitivity and Impact on Self-Efficacy. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-023-06133-0

O'Leary, J. D., Crawford, M. W., Odame, I., Shorten, G. D., & McGrath, P. A. (2014). Thermal pain and sensory processing in children with sickle cell disease. *The Clinical Journal of Pain*, *30*(3), 244-250. https://doi.org/10.1097/AJP.0b013e318292a38e

O'Nions, E., Petersen, I., Buckman, J. E. J., Charlton, R., Cooper, C., Corbett, A., Happé, F., Manthorpe, J., Richards, M., Saunders, R., Zanker, C., Mandy, W., & Stott, J. (2023). Autism in England: assessing underdiagnosis in a population-based cohort study of prospectively collected primary care data. *The Lancet Regional Health – Europe*, *29*. https://doi.org/10.1016/j.lanepe.2023.100626

O'Rourke, D. (2004). The Measurement of Pain in Infants, Children, and Adolescents: From Policy to Practice. *Physical Therapy*, *84*(6), 560-570. https://doi.org/10.1093/ptj/84.6.560

O'Hagan, S., & Hebron, J. (2017). Perceptions of friendship among adolescents with autism spectrum conditions in a mainstream high school resource provision. *European Journal of Special Needs Education*, *32*(3), 314-328. https://doi.org/10.1080/08856257.2016.1223441

Oberlander, T. F., Burkitt, C. C., & Symons, F. J. (2010). Pain in Individuals with Intellectual Disabilities. In M. E. Lynch, K. D. Craig, & P. H. Peng (Eds.), *Clinical Pain Management : A Practical Guide* (pp. 328-329). John Wiley & Sons, Incorporated. http://ebookcentral.proquest.com/lib/ljmu/detail.action?docID=624674

Ocay, D. D., Larche, C. L., Betinjane, N., Jolicoeur, A., Beaulieu, M. J., Saran, N., Ouellet, J. A., Ingelmo, P. M., & Ferland, C. E. (2022). Phenotyping Chronic Musculoskeletal Pain in Male and Female Adolescents: Psychosocial Profiles, Somatosensory Profiles and Pain Modulatory Profiles. *Journal of Pain Research*, *15*, 591-612. https://doi.org/10.2147/jpr.S352607

Office for National Statistics. (2022). *Regional ethnic diversity*. GOV.UK: GOV.UK Retrieved from https://www.ethnicity-facts-figures.service.gov.uk/uk-population-by-ethnicity/national-and-regional-populations/regional-ethnic-diversity/latest/

Oredipe, T., Kofner, B., Riccio, A., Cage, E., Vincent, J., Kapp, S. K., Dwyer, P., & Gillespie-Lynch, K. (2022). Does learning you are autistic at a younger age lead to better adult outcomes? A participatory exploration of the perspectives of autistic university students. *Autism*, *27*(1), 200-212. https://doi.org/10.1177/13623613221086700

Ortiz Rubio, A., de María Romero Ayuso, D. N., Torres Sánchez, I., Cabrera Martos, I., Rodríguez Torres, J., López López, L., & Valenza, M. C. (2023). Pain Experiences of People Diagnosed With Autism Spectrum Disorder: A Systematic Review of Case–Control Studies. *The American Journal of Occupational Therapy*, *77*(2). https://doi.org/10.5014/ajot.2023.050050

Ouzzani, M., Hammady, H., Fedorowicz, Z., & Elmagarmid, A. (2016). Rayyan—a web and mobile app for systematic reviews. *Systematic Reviews*, *5*(1), 210. https://doi.org/10.1186/s13643-016-0384-4

Overton, G. L., Marsà-Sambola, F., Martin, R., & Cavenagh, P. (2023). Understanding the Self-identification of Autism in Adults: a Scoping Review. *Review Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s40489-023-00361-x

Pagé, M. G., Fuss, S., Martin, A. L., Escobar, E. M. R., & Katz, J. (2010). Development and Preliminary Validation of the Child Pain Anxiety Symptoms Scale in a Community

Sample. *Journal of Pediatric Psychology*, *35*(10), 1071-1082. https://doi.org/10.1093/jpepsy/jsq034

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., Shamseer, L., Tetzlaff, J. M., Akl, E. A., Brennan, S. E., Chou, R., Glanville, J., Grimshaw, J. M., Hróbjartsson, A., Lalu, M. M., Li, T., Loder, E. W., Mayo-Wilson, E., McDonald, S., McGuinness, L. A., Stewart, L. A., Thomas, J., Tricco, A. C., Welch, V. A., Whiting, P., & Moher, D. (2021). The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ*, *372*(71). https://doi.org/10.1136/bmj.n71

Palermo, T. M. (2020). Pain prevention and management must begin in childhood: the key role of psychological interventions. *PAIN*, *161*, S114-S121. https://doi.org/10.1097/j.pain.0000000000001862

Palermo, T. M., Koh, J. L., & Zeltzer, L. K. (2010). Pain in Children. In M. E. Lynch, K. D. Craig, & P. W. H. Peng (Eds.), *Clinical Pain Management : A Practical Guide* (pp. 320-321). John Wiley & Sons, Incorporated. http://ebookcentral.proquest.com/lib/ljmu/detail.action?docID=624674

Pan, P.-Y., Bölte, S., Kaur, P., Jamil, S., & Jonsson, U. (2021). Neurological disorders in autism: A systematic review and meta-analysis. *Autism*, *25*(3), 812-830. https://doi.org/10.1177/1362361320951370

Parmeggiani, A., Corinaldesi, A., & Posar, A. (2019). Early features of autism spectrum disorder: a cross-sectional study. *Italian Journal of Pediatrics*, *45*(1), 144. https://doi.org/10.1186/s13052-019-0733-8

Paron, K. (2024). Exploring Child-Patient Autonomy: Findings from an Ethnographic Study of Clinic Visits by Children. *Child Indicators Research*, *17*(1), 99-121. https://doi.org/10.1007/s12187-023-10077-3

Pas, R., Rheel, E., Van Oosterwijck, S., Leysen, L., Van De Vijver, E., Nijs, J., Ickmans, K., & Meeus, M. (2019). Endogenous pain modulation in children with functional abdominal pain disorders. *PAIN*, *160*(8), 1883-1890. https://doi.org/10.1097/j.pain.0000000000001566

Pate, J. W., Noblet, T., Hush, J. M., Hancock, M. J., Sandells, R., Pounder, M., & Pacey, V. (2019). Exploring the concept of pain of Australian children with and without pain: qualitative study. *BMJ Open*, *9*(10), e033199. https://doi.org/10.1136/bmjopen-2019-033199

Pathan, H., & Williams, J. (2012). Basic opioid pharmacology: an update. *British Journal of Pain*, *6*(1), 11-16. https://doi.org/10.1177/2049463712438493

Patten Koenig, K., & Hough Williams, L. (2017). Characterization and Utilization of Preferred Interests: A Survey of Adults on the Autism Spectrum. *Occupational Therapy in Mental Health*, *33*(2), 129-140. https://doi.org/10.1080/0164212X.2016.1248877

Pohl, A., Cassidy, S., Auyeung, B., & Baron-Cohen, S. (2014). Uncovering steroidopathy in women with autism: a latent class analysis. *Molecular Autism*, *5*, 27. https://doi.org/10.1186/2040-2392-5-27

Pope, D. J., Butler, H., & Qualter, P. (2012). Emotional Understanding and Color-Emotion Associations in Children Aged 7-8 Years. *Child Development Research*, *2012*, 975670. https://doi.org/10.1155/2012/975670

Poppe, K., & Abela, A. (2023). Engaging with hard-to-reach children and parents using a creative methodology. *Qualitative Research*, 14687941231202378. https://doi.org/10.1177/14687941231202378

Porreca, F., & Navratilova, E. (2017). Reward, motivation, and emotion of pain and its relief. *PAIN*, *158*, S43-S49. https://doi.org/10.1097/j.pain.0000000000000798

Posar, A., & Visconti, P. (2018). Sensory abnormalities in children with autism spectrum disorder. *Jornal de Pediatria*, *94*(4), 342-350. https://doi.org/10.1016/j.jped.2017.08.008

Pravossoudovitch, K., Cury, F., Young, S. G., & Elliot, A. J. (2014). Is red the colour of danger? Testing an implicit red–danger association. *Ergonomics*, *57*(4), 503-510. https://doi.org/10.1080/00140139.2014.889220

Price, D. D. (2000). Psychological and neural mechanisms of the affective dimension of pain. *Science*, *288*(5472), 1769-1772. https://doi.org/10.1126/science.288.5472.1769

Prkachin, K. M. (1992). The consistency of facial expressions of pain: a comparison across modalities. *PAIN*, *51*(3), 297-306. https://doi.org/10.1016/0304-3959(92)90213-U

Prkachin, K. M. (2009). Assessing pain by facial expression: facial expression as nexus. *Pain Research and Management*, *14*(1), 53-58. https://doi.org/10.1155/2009/542964

Prkachin, K. M., & Craig, K. D. (1995). Expressing pain: The communication and interpretation of facial pain signals. *Journal of Nonverbal Behavior*, *19*(4), 191-205. https://doi.org/10.1007/BF02173080

Prolific. (2024). Version April 2024) Prolific. https://www.prolific.com/

Qualtrics. (2021). *Qualtrics Software*. In (Version December 2021) Qualtrics. https://www.qualtrics.com/

QuestionPro. (2024). *QuestionPro Survey Software*. In https://www.questionpro.com/

Raja, S. N., Carr, D. B., Cohen, M., Finnerup, N. B., Flor, H., Gibson, S., Keefe, F. J., Mogil, J. S., Ringkamp, M., Sluka, K. A., Song, X. J., Stevens, B., Sullivan, M. D., Tutelman, P. R., Ushida, T., & Vader, K. (2020). The revised International Association for the Study of Pain definition of pain: concepts, challenges, and compromises. *PAIN*, *161*(9), 1976-1982. https://doi.org/10.1097/j.pain.0000000000001939

Ramos, N. (2021). Medical Trauma in LGBTQIA Youth: Adapting Trauma-Informed Affirming Clinical Practices. *Pediatric Annals*, *50*(9), e379-e383. https://doi.org/10.3928/19382359-20210818-02

Randomness and Integrity Services Ltd. (2024). *RANDOM.ORG*. Randomness and Integrity Services Ltd. https://www.random.org/

Raponi, M. (n.d.). Playground School Children Playing. In. Pixabay.

Rasmussen, P. S., & Pagsberg, A. K. (2019). Customizing Methodological Approaches in Qualitative Research on Vulnerable Children with Autism Spectrum Disorders. *Societies*, *9*(4), 75. https://doi.org/10.3390/soc9040075

Rathleff, M. S., Roos, E. M., Olesen, J. L., Rasmussen, S., & Arendt-Nielsen, L. (2013). Lower Mechanical Pressure Pain Thresholds in Female Adolescents With Patellofemoral Pain Syndrome. *Journal of Orthopaedic & Sports Physical Therapy*, *43*(6), 414-421. https://doi.org/10.2519/jospt.2013.4383

Rattaz, C., Dubois, A., Michelon, C., Viellard, M., Poinso, F., & Baghdadli, A. (2013). How do children with autism spectrum disorders express pain? A comparison with developmentally delayed and typically developing children. *PAIN*, *154*(10), 2007-2013. https://doi.org/10.1016/j.pain.2013.06.011

Rheel, E., Ickmans, K., Wauters, A., Van Ryckeghem, D. M. L., Malfliet, A., & Vervoort, T. (2021). The effect of a pain educational video intervention upon child pain-related outcomes: A randomized controlled study. *European Journal of pain*, *25*(10), 2094-2111. https://doi.org/10.1002/ejp.1822

Richards, D., Jones, S., Jeffery, J., Lowe, M., Godwin, M., & Willett, M. (2021). Intra-Rater and Inter-Rater Reliability of Pressure Pain Algometry of the Sural and Tibial Nerves in Asymptomatic Elite Youth Footballers. *Sports (Basel)*, *9*(9). https://doi.org/10.3390/sports9090132

Rios-Vega, L., Carroll, A., Dumont, R., Treadwell-Deering, D., Fields, M., & Schaaf, R. (2024). Designing sensory adaptive environments to enhance participation in healthcare for autistic children. *Discover Psychology*, *4*(1), 22. https://doi.org/10.1007/s44202-024-00118-5

Riquelme, I., Hatem, S. M., & Montoya, P. (2016). Abnormal Pressure Pain, Touch Sensitivity, Proprioception, and Manual Dexterity in Children with Autism Spectrum Disorders. *Neural Plasticity*, *2016*, 1723401. https://doi.org/10.1155/2016/1723401

Riquelme, I., Hatem, S. M., Sabater-Garriz, A., & Montoya, P. (2023). A multidimensional investigation of the relationship between skin-mediated somatosensory signals, emotion regulation and behavior problems in autistic children. *Frontiers in Neuroscience*, *17*, Article 1227173. https://doi.org/10.3389/fnins.2023.1227173

Rojo, R., Prados-Frutos, J. C., & López-Valverde, A. (2015). Pain assessment using the Facial Action Coding System. A systematic review. *Medicina Clínica (English Edition)*, *145*(8), 350-355. https://doi.org/10.1016/j.medcle.2014.08.002

Rolke, R., Baron, R., Maier, C., Tölle, T. R., Treede, D. R., Beyer, A., Binder, A., Birbaumer, N., Birklein, F., Bötefür, I. C., Braune, S., Flor, H., Huge, V., Klug, R., Landwehrmeyer, G. B., Magerl, W., Maihöfner, C., Rolko, C., Schaub, C., Scherens, A., Sprenger, T., Valet, M., & Wasserka, B. (2006). Quantitative sensory testing in the German Research Network on Neuropathic Pain (DFNS): standardized protocol and reference values. *PAIN*, *123*(3), 231-243. https://doi.org/10.1016/j.pain.2006.01.041

Rollman, G. B., Abdel-Shaheed, J., Gillespie, J. M., & Jones, K. S. (2004). Does past pain influence current pain: biological and psychosocial models of sex differences. *European Journal of pain*, *8*(5), 427-433. https://doi.org/10.1016/j.ejpain.2004.03.002

Rose, V., Trembath, D., Keen, D., & Paynter, J. (2016). The proportion of minimally verbal children with autism spectrum disorder in a community-based early intervention programme. *Journal of Intellectual Disability Research*, *60*(5), 464-477. https://doi.org/10.1111/jir.12284

Rowley, E., Chandler, S., Baird, G., Simonoff, E., Pickles, A., Loucas, T., & Charman, T. (2012). The experience of friendship, victimization and bullying in children with an autism spectrum disorder: Associations with child characteristics and school placement. *Research in Autism Spectrum Disorders*, *6*(3), 1126-1134. https://doi.org/10.1016/j.rasd.2012.03.004

Ruskin, D., Lalloo, C., Amaria, K., Stinson, J. N., Kewley, E., Campbell, F., Brown, S. C., Jeavons, M., & McGrath, P. A. (2014). Assessing pain intensity in children with chronic pain: convergent and discriminant validity of the 0 to 10 numerical rating scale in clinical practice. *Pain Research and Management*, *19*(3), 141-148. https://doi.org/10.1155/2014/856513

Sá, S., & Silva, A. G. (2017). Repositioning error, pressure pain threshold, catastrophizing and anxiety in adolescents with chronic idiopathic neck pain. *Musculoskeletal Science and Practice*, *30*, 18-24. https://doi.org/10.1016/j.msksp.2017.04.011

Sacramento, L. S., Camargo, P. R., Siqueira-Júnior, A. L., Ferreira, J. P., Salvini, T. F., & Alburquerque-Sendín, F. (2017). Presence of Latent Myofascial Trigger Points and Determination of Pressure Pain Thresholds of the Shoulder Girdle in Healthy Children and Young Adults: A Cross-sectional Study. *Journal of Manipulative and Physiological Therapeutics*, *40*(1), 31-40. https://doi.org/10.1016/j.jmpt.2016.10.007

Sansone, L., Gentile, C., Grasso, E. A., Di Ludovico, A., La Bella, S., Chiarelli, F., & Breda, L. (2023). Pain Evaluation and Treatment in Children: A Practical Approach. *Children (Basel)*, *10*(7). https://doi.org/10.3390/children10071212

Saxena, I., Kumar, M., Barath, A. S., Verma, A., Garg, S., & Kumar, M. (2015). Effect of Age on Response to Experimental Pain in Normal Indian Males. *JOURNAL OF CLINICAL AND DIAGNOSTIC RESEARCH*, *9*(9), Cc05-08. https://doi.org/10.7860/jcdr/2015/15385.6516

Schaeffer, J., Abd El-Raziq, M., Castroviejo, E., Durrleman, S., Ferré, S., Grama, I., Hendriks, P., Kissine, M., Manenti, M., Marinis, T., Meir, N., Novogrodsky, R.,

Perovic, A., Panzeri, F., Silleresi, S., Sukenik, N., Vicente, A., Zebib, R., Prévost, P., & Tuller, L. (2023). Language in autism: domains, profiles and co-occurring conditions. *Journal of Neural Transmission*, *130*(3), 433-457. https://doi.org/10.1007/s00702-023-02592-y

Schauder, K. B., Mash, L. E., Bryant, L. K., & Cascio, C. J. (2015). Interoceptive ability and body awareness in autism spectrum disorder. *Journal of Experimental Child Psychology*, *131*, 193-200. https://doi.org/10.1016/j.jecp.2014.11.002

Scheper, M. C., Pacey, V., Rombaut, L., Adams, R. D., Tofts, L., Calders, P., Nicholson, L. L., & Engelbert, R. H. (2017). Generalized Hyperalgesia in Children and Adults Diagnosed With Hypermobility Syndrome and Ehlers-Danlos Syndrome Hypermobility Type: A Discriminative Analysis. *Arthritis Care & Research*, *69*(3), 421-429. https://doi.org/10.1002/acr.22998

Schmitz, A. K., Vierhaus, M., & Lohaus, A. (2013). Pain tolerance in children and adolescents: sex differences and psychosocial influences on pain threshold and endurance. *European Journal of pain*, *17*(1), 124-131. https://doi.org/10.1002/j.1532-2149.2012.00169.x

Schoonenboom, J., & Johnson, R. B. (2017). How to Construct a Mixed Methods Research Design. *Kolner Z Soz Sozpsychol*, *69*(Suppl 2), 107-131. https://doi.org/10.1007/s11577-017-0454-1

Schwei, R. J., Johnson, T. P., Matthews, A. K., & Jacobs, E. A. (2017). Perceptions of negative health-care experiences and self-reported health behavior change in three racial and ethnic groups. *Ethnicity & Health*, *22*(2), 156-168. https://doi.org/10.1080/13557858.2016.1244621

Sethna, N. F., Meier, P. M., Zurakowski, D., & Berde, C. B. (2007). Cutaneous sensory abnormalities in children and adolescents with complex regional pain syndromes. *PAIN*, *131*(1-2), 153-161. https://doi.org/10.1016/j.pain.2006.12.028

Sharpe, R. A., Curry, W., Brown, R., & Shankar, R. (2019). A public health approach to reducing health inequalities among adults with autism. *British Journal of General Practice*, *69*(688), 534-535. https://doi.org/10.3399/bjgp19X706133

Shaw, S. C., Davis, L.-J., & Doherty, M. (2022). Considering autistic patients in the era of telemedicine: the need for an adaptable, equitable, and compassionate approach. *BJGP Open*, *6*(1), BJGPO.2021.0174. https://doi.org/10.3399/bjgpo.2021.0174

Shaw, S. C. K., Carravallah, L., Johnson, M., O'Sullivan, J., Chown, N., Neilson, S., & Doherty, M. (2023). Barriers to healthcare and a 'triple empathy problem' may lead to adverse outcomes for autistic adults: A qualitative study. *Autism*, 13623613231205629. https://doi.org/10.1177/13623613231205629

Sherry, D. D., & Sapp, L. R. (2003). Enthesalgia in childhood: site-specific tenderness in healthy subjects and in patients with seronegative enthesopathic arthropathy. *The Journal of rheumatology*, *30*(6), 1335-1340. https://pubmed.ncbi.nlm.nih.gov/12784411/

Sieberg, C. B., Lunde, C. E., Shafrir, A. L., Meints, S. M., Madraswalla, M., Huntley, D., Olsen, H., Wong, C., DiVasta, A. D., Missmer, S. A., & Sethna, N. (2023). Quantitative somatosensory testing of the abdomen: establishing initial reference values across developmental age and biological sex. *PAIN*. https://doi.org/10.1097/j.pain.0000000000003001

Sieberg, C. B., Lunde, C. E., Wong, C., Manganella, J., Starkweather, A. R., Sethna, N., & Perry-Eaddy, M. A. (2023). Pilot Investigation of Somatosensory Functioning and Pain Catastrophizing in Pediatric Spinal Fusion Surgery. *Pain Management Nursing*, *24*(1), 27-34. https://doi.org/10.1016/j.pmn.2022.11.001

Simons, L. E., Smith, A., Kaczynski, K., & Basch, M. (2015). Living in fear of your child's pain: the Parent Fear of Pain Questionnaire. *PAIN*, *156*(4), 694-702. https://doi.org/10.1097/j.pain.0000000000000100

Skitterphoto. (n.d.). Girls Bicycle Helmets. In. Pixabay.
Sloan, P., & Hollins, M. (2017). Attention and pain: are auditory distractors special? *Experimental Brain Research*, *235*(5), 1593-1602. https://doi.org/10.1007/s00221-017-4903-x
Smith-Young, J., Chafe, R., Audas, R., & Gustafson, D. L. (2022). "I Know How to Advocate": Parents' Experiences in Advocating for Children and Youth Diagnosed With Autism Spectrum Disorder. *Health Services Insights*, *15*, 11786329221078803. https://doi.org/10.1177/11786329221078803
Smith, J. A. (1996). Beyond the divide between cognition and discourse: Using interpretative phenomenological analysis in health psychology. *Psychology & Health*, *11*(2), 261-271. https://doi.org/10.1080/08870449608400256
Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. SAGE Publications. https://books.google.co.uk/books?id=UTxdBAAAQBAJ
Smith, J. A., & Osborn, M. (2015). Interpretative phenomenological analysis as a useful methodology for research on the lived experience of pain. *British Journal of Pain*, *9*(1), 41-42. https://doi.org/10.1177/2049463714541642
Smooth, W. G. (2013). Intersectionality from Theoretical Framework to Policy Intervention. In A. R. Wilson (Ed.), *Situating Intersectionality: Politics, Policy, and Power* (pp. 11). Palgrave Macmillan US. https://doi.org/10.1057/9781137025135_2
Snow, S. L., Smith, I. M., Latimer, M., Stirling Cameron, E., Fox, J., & Chorney, J. (2022). A balancing act: An interpretive description of healthcare providers' and families' perspective on the surgical experiences of children with autism spectrum disorder. *Autism*, *26*(4), 839-848. https://doi.org/10.1177/13623613211034057
Soee, A. B., Skov, L., Kreiner, S., Tornoe, B., & Thomsen, L. L. (2013). Pain sensitivity and pericranial tenderness in children with tension-type headache: a controlled study. *Journal of Pain Research*, *6*, 425-434. https://doi.org/10.2147/jpr.S42869
Somedic SenseLab AB. (n.d.). *Algometer*. In http://somedic.com/en/algometer.html
Somerville, L. H. (2013). The Teenage Brain: Sensitivity to Social Evaluation. *Current Directions in Psychological Science*, *22*(2), 121-127. https://doi.org/10.1177/0963721413476512
Sosnowy, C., Silverman, C., Shattuck, P., & Garfield, T. (2019). Setbacks and Successes: How Young Adults on the Autism Spectrum Seek Friendship. *Autism in Adulthood*, *1*(1), 44-51. https://doi.org/10.1089/aut.2018.0009
Spain, D., Sin, J., Linder, K. B., McMahon, J., & Happé, F. (2018). Social anxiety in autism spectrum disorder: A systematic review. *Research in Autism Spectrum Disorders*, *52*, 51-68. https://doi.org/10.1016/j.rasd.2018.04.007
Stabell, N., Stubhaug, A., Flægstad, T., Mayer, E., Naliboff, B. D., & Nielsen, C. S. (2014). Widespread hyperalgesia in adolescents with symptoms of irritable bowel syndrome: results from a large population-based study. *The Journal of Pain*, *15*(9), 898-906. https://doi.org/10.1016/j.jpain.2014.05.007
Stagg, S. D., & Belcher, H. (2019). Living with autism without knowing: receiving a diagnosis in later life. *Health Psychology and Behavioral Medicine*, *7*(1), 348-361. https://doi.org/10.1080/21642850.2019.1684920
Stallard, P., Williams, L., Lenton, S., & Velleman, R. (2001). Pain in cognitively impaired, non-communicating children. *Archives of Disease in Childhood*, *85*(6), 460-462. https://doi.org/10.1136/adc.85.6.460
Stanford, E. A., Chambers, C. T., & Craig, K. D. (2006). The role of developmental factors in predicting young children's use of a self-report scale for pain. *PAIN*, *120*(1), 16-23. https://doi.org/10.1016/j.pain.2005.10.004
Stewart, M., & Ryan, S.-J. (2019). Do Metaphors Have Therapeutic Value for People in Pain? A Systematic Review. *Pain and Rehabilitation - the Journal of Physiotherapy*

Pain Association, *2020*(48), 10-23. https://www.ingentaconnect.com/content/ppa/pr/2019/00002020/00000048/art00003

Stewart, T. M., Martin, K., Fazi, M., Oldridge, J., Piper, A., & Rhodes, S. M. (2022). A systematic review of the rates of depression in autistic children and adolescents without intellectual disability. *Psychology and Psychotherapy: Theory, Research and Practice*, *95*(1), 313-344. https://doi.org/10.1111/papt.12366

Stinson, J., & Jibb, L. (2013). Pain Assessment. In A. Twycross, S. Dowden, & J. Stinson (Eds.), *Managing Pain in Children : A Clinical Guide for Nurses and Healthcare Professionals*. John Wiley & Sons, Incorporated. http://ebookcentral.proquest.com/lib/ljmu/detail.action?docID=7103754

Stinson, J. N., & McGrath, P. J. (2010). Measurement and Assessment of Pain in Pediatric Patients. In M. E. Lynch, K. D. Craig, & P. H. Peng (Eds.), *Clinical Pain Management : A Practical Guide* (pp. 64-65). John Wiley & Sons, Incorporated. http://ebookcentral.proquest.com/lib/ljmu/detail.action?docID=624674

Strömberg, M., Liman, L., Bang, P., & Igelström, K. (2022). Experiences of Sensory Overload and Communication Barriers by Autistic Adults in Health Care Settings. *Autism Adulthood*, *4*(1), 66-75. https://doi.org/10.1089/aut.2020.0074

Swann, M. (2021). Recognising the importance of language in effective pain assessment. *Nursing Standard*, *36*(2), 43-50. https://doi.org/10.7748/ns.2021.e11563

Symons, F. J., Burkitt, C. C., Wilcox, G., McAdams, B., Wendelschafer-Crabb, G., & Kennedy, W. R. (2022). Modifying quantitative sensory testing to investigate behavioral reactivity in a pediatric global developmental delay sample: Relation to peripheral innervation and chronic pain outcomes. *Developmental Psychobiology*, *64*(8), e22329. https://doi.org/10.1002/dev.22329

Syu, Y.-C., Huang, P.-C., Wang, T.-Y., Chang, Y.-C., & Lin, L.-Y. (2020). Relationship Among Sensory Over-Responsivity, Problem Behaviors, and Anxiety in Emerging Adults with Autism Spectrum Disorder. *Neuropsychiatric Disease and Treatment*, *16*(null), 2181-2190. https://doi.org/10.2147/NDT.S270308

Tager-Flusberg, H., & Kasari, C. (2013). Minimally verbal school-aged children with autism spectrum disorder: the neglected end of the spectrum. *Autism Research*, *6*(6), 468-478. https://doi.org/10.1002/aur.1329

Thaler, H., Skewes, J. C., Gebauer, L., Christensen, P., Prkachin, K. M., & Jegindø Elmholdt, E.-M. (2017). Typical pain experience but underestimation of others' pain: Emotion perception in self and others in autism spectrum disorder. *Autism*, *22*(6), 751-762. https://doi.org/10.1177/1362361317701269

The Royal Children's Hospital. (2022). *Pain assessment and measurement*. https://www.rch.org.au/rchcpg/hospital_clinical_guideline_index/Pain_assessment_and_measurement/

The School Toolkit for EDS and JHS. (2021). *The School Toolkit*. https://theschooltoolkit.org/

The Westminister Commission on Autism. (2021). *SUPPORT SURROUNDING DIAGNOSIS: An Inquiry into Pre- and Post-Support for the Autism Diagnosis Pathway* https://www.bath.ac.uk/publications/resources-for-researchers-and-the-autism-community/attachments/support-surrounding-diagnosis.pdf

Tordjman, S., Anderson, G. M., Botbol, M., Brailly-Tabard, S., Perez-Diaz, F., Graignic, R., Carlier, M., Schmit, G., Rolland, A. C., Bonnot, O., Trabado, S., Roubertoux, P., & Bronsard, G. (2009). Pain reactivity and plasma beta-endorphin in children and adolescents with autistic disorder. *PLoS One*, *4*(8), e5289. https://doi.org/10.1371/journal.pone.0005289

Trachsel, L. A., Munakomi, S., & Cascella, M. (2023). Pain Theory. In *StatPearls*. StatPearls Publishing LLC.

Trajkovski, V. (2018). Health Condition in Persons with Autism Spectrum Disorders. *Journal for ReAttach Therapy and Developmental Diversities*, *1*(2), 112 - 124. https://jrtdd.com/index.php/journal/article/view/14

Trevisan, D. A., Parker, T., & McPartland, J. C. (2021). First-Hand Accounts of Interoceptive Difficulties in Autistic Adults. *Journal of Autism and Developmental Disorders*, *51*(10), 3483-3491. https://doi.org/10.1007/s10803-020-04811-x

Truffyn, E. E., Moayedi, M., Brown, S. C., Ruskin, D., & Duerden, E. G. (2021). Sensory Function and Psychological Factors in Children With Complex Regional Pain Syndrome Type 1. *Journal of Child Neurology*, *36*(10), 823-830. https://doi.org/10.1177/08830738211007685

Trundle, G., Jones, K. A., Ropar, D., & Egan, V. (2023). Prevalence of Victimisation in Autistic Individuals: A Systematic Review and Meta-Analysis. *Trauma Violence Abuse*, *24*(4), 2282-2296. https://doi.org/10.1177/15248380221093689

Tsao, J. C. I., Lu, Q., Kim, S. C., & Zeltzer, L. K. (2006). Relationships among Anxious Symptomatology, Anxiety Sensitivity and Laboratory Pain Responsivity in Children. *Cognitive Behaviour Therapy*, *35*(4), 207-215. https://doi.org/10.1080/16506070600898272

Tsze, D. S., von Baeyer, C. L., Pahalyants, V., & Dayan, P. S. (2018). Validity and Reliability of the Verbal Numerical Rating Scale for Children Aged 4 to 17 Years With Acute Pain. *Annals of Emergency Medicine*, *71*(6), 691-702.e693. https://doi.org/10.1016/j.annemergmed.2017.09.009

Turk, D. C., & Flor, H. (1987). Pain > pain behaviors: The utility and limitations of the pain behavior construct. *PAIN*, *31*(3), 277-295. https://doi.org/10.1016/0304-3959(87)90158-8

Turner-Brown, L. M., Lam, K. S. L., Holtzclaw, T. N., Dichter, G. S., & Bodfish, J. W. (2011). Phenomenology and measurement of circumscribed interests in autism spectrum disorders. *Autism*, *15*(4), 437-456. https://doi.org/10.1177/1362361310386507

Turnock, A., Langley, K., & Jones, C. R. G. (2022). Understanding Stigma in Autism: A Narrative Review and Theoretical Model. *Autism Adulthood*, *4*(1), 76-91. https://doi.org/10.1089/aut.2021.0005

Tutelman, P. R., MacKenzie, N. E., Chambers, C. T., Coffman, S., Cornelissen, L., Cormier, B., Higgins, K. S., Phinney, J., Blankenburg, M., & Walker, S. (2024). Quantitative sensory testing for assessment of somatosensory function in children and adolescents: a scoping review. *Pain reports*, *9*(3), e1151. https://doi.org/10.1097/pr9.0000000000001151

Twycross, A. (1998). Perceptions about children's pain experience. *The Professional Nurse*, *13*(12), 822-826. https://pubmed.ncbi.nlm.nih.gov/10095674/

Uddin, Z., & MacDermid, J. C. (2016). Quantitative Sensory Testing in Chronic Musculoskeletal Pain. *Pain Medicine*, *17*(9), 1694-1703. https://doi.org/10.1093/pm/pnv105

Ujiie, Y., & Takahashi, K. (2022). Associations between self-reported social touch avoidance, hypersensitivity, and autistic traits: Results from questionnaire research among typically developing adults. *Personality and Individual Differences*, *184*, 111186. https://doi.org/10.1016/j.paid.2021.111186

UKRI. (2024). *Consent*. https://www.ukri.org/councils/esrc/guidance-for-applicants/research-ethics-guidance/consent/#:~:text=Research%20should%20not%20normally%20proceed,alternative%20approaches%20to%20record%20consent.

Unknown. (n.d.-a). Child Soccer Playing. In. Pixabay.

Unknown. (n.d.-b). Medication Child Hurts Got Sick. In. Pixabay.

Valentino, R., Rongo, R., Alessio, M., Alstergren, P., Bucci, R., Leone, G., D'Antò, V., & Michelotti, A. (2020). Pressure pain threshold over masticatory muscles and

temporomandibular joint in patients with juvenile idiopathic arthritis. *Journal of Oral Rehabilitation*, *47*(8), 944-950. https://doi.org/10.1111/joor.13024

Valkenburg, A. J., Tibboel, D., & van Dijk, M. (2015). Pain sensitivity of children with Down syndrome and their siblings: quantitative sensory testing versus parental reports. *Developmental Medicine & Child Neurology*, *57*(11), 1049-1055. https://doi.org/10.1111/dmcn.12823

Van Damme, S., & Moore, D. J. (2012). From the clinic to the lab (and back)-a call for laboratory research to optimize cognitive behavioural treatment of pain. *Translational Behavioral Medicine*, *2*(1), 102-105. https://doi.org/10.1007/s13142-011-0083-6

van den Bosch, G. E., van Dijk, M., Tibboel, D., & Valkenburg, A. J. (2017). Thermal quantitative sensory testing in healthy Dutch children and adolescents standardized test paradigm and Dutch reference values. *BMC Pediatrics*, *17*(1), 77. https://doi.org/10.1186/s12887-017-0827-7

van der Venne, P., Balint, A., Drews, E., Parzer, P., Resch, F., Koenig, J., & Kaess, M. (2021). Pain sensitivity and plasma beta-endorphin in adolescent non-suicidal self-injury. *Journal of Affective Disorders*, *278*, 199-208. https://doi.org/10.1016/j.jad.2020.09.036

Vaughan, S., Failla, M. D., Poole, H. M., Forshaw, M. J., McGlone, F., Cascio, C. J., & Moore, D. J. (2019). Pain Processing in Psychiatric Conditions: A Systematic Review. *Review of General Psychology*, *23*(3), 336-358. https://doi.org/10.1177/1089268019842771

Vaughan, S., McGlone, F., Poole, H., & Moore, D. J. (2020). A Quantitative Sensory Testing Approach to Pain in Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders*, *50*(5), 1607-1620. https://doi.org/10.1007/s10803-019-03918-0

Vervoort, T., Caes, L., Trost, Z., Sullivan, M., Vangronsveld, K., & Goubert, L. (2011). Social modulation of facial pain display in high-catastrophizing children: An observational study in schoolchildren and their parents. *PAIN*, *152*(7), 1591-1599. https://doi.org/10.1016/j.pain.2011.02.048

Vervoort, T., Goubert, L., Eccleston, C., Verhoeven, K., De Clercq, A., Buysse, A., & Crombez, G. (2008). The effects of parental presence upon the facial expression of pain: the moderating role of child pain catastrophizing. *PAIN*, *138*(2), 277-285. https://doi.org/10.1016/j.pain.2007.12.013

Villemure, C., & Bushnell, M. C. (2002). Cognitive modulation of pain: how do attention and emotion influence pain processing? *PAIN*, *95*(3), 195-199. https://doi.org/10.1016/S0304-3959(02)00007-6

Visser-Bochane, M. I., Reijneveld, S. A., Krijnen, W. P., van der Schans, C. P., & Luinge, M. R. (2020). Identifying Milestones in Language Development for Young Children Ages 1 to 6 Years. *Academic Pediatrics*, *20*(3), 421-429. https://doi.org/10.1016/j.acap.2019.07.003

Visser, E. J., & Davies, S. (2009). What is pain? II: Pain expression and behaviour, evolutionary concepts, models and philosophies. *Australasian Anaesthesia*(2009), 35-43. https://doi.org/10.3316/informit.485890435725678

Vlaeyen, J. W. S., Hanssen, M., Goubert, L., Vervoort, T., Peters, M., van Breukelen, G., Sullivan, M. J. L., & Morley, S. (2009). Threat of pain influences social context effects on verbal pain report and facial expression. *Behaviour Research and Therapy*, *47*(9), 774-782. https://doi.org/10.1016/j.brat.2009.05.008

von Baeyer, C. L. (2006). Children's Self-Reports of Pain Intensity: Scale Selection, Limitations and Interpretation. *Pain Research and Management*, *11*, 197616. https://doi.org/10.1155/2006/197616

von Baeyer, C. L., Marche, T. A., Rocha, E. M., & Salmon, K. (2004). Children's memory for pain: overview and implications for practice. *The Journal of Pain*, *5*(5), 241-249. https://doi.org/10.1016/j.jpain.2004.05.001

von Baeyer, C. L., Piira, T., Chambers, C. T., Trapanotto, M., & Zeltzer, L. K. (2005). Guidelines for the cold pressor task as an experimental pain stimulus for use with children. *The Journal of Pain*, *6*(4), 218-227. https://doi.org/10.1016/j.jpain.2005.01.349

von Baeyer, C. L., Uman, L. S., Chambers, C. T., & Gouthro, A. (2011). Can we screen young children for their ability to provide accurate self-reports of pain? *PAIN*, *152*(6), 1327-1333. https://doi.org/10.1016/j.pain.2011.02.013

Wainscot, J. J., Naylor, P., Sutcliffe, P., Tantam, D., & Williams, J. V. (2008). Relationships with peers and use of the school environment of mainstream secondary school pupils with Asperger syndrome (high-functioning autism): A case-control study. *International Journal of Psychology & Psychological Therapy*, *8*, 25-38. https://psycnet.apa.org/record/2008-02887-003

Wakefield, E. O., Puhl, R. M., Litt, M. D., & Zempsky, W. T. (2021). “If It Ever Really Hurts, I Try Not to Let Them Know:” The Use of Concealment as a Coping Strategy Among Adolescents With Chronic Pain. *Frontiers in Psychology*, *12*. https://doi.org/10.3389/fpsyg.2021.666275

Wall, P. D., Melzack, R., & Bonica, J. J. (1989). *Textbook of pain*. Churchill Livingstone Edinburgh.

Walsh, C., O’Connor, P., Walsh, E., & Lydon, S. (2023). A Systematic Review of Interventions to Improve Healthcare Experiences and Access in Autism. *Review Journal of Autism and Developmental Disorders*, *10*(2), 185-202. https://doi.org/10.1007/s40489-021-00279-2

Warrier, V., Greenberg, D. M., Weir, E., Buckingham, C., Smith, P., Lai, M.-C., Allison, C., & Baron-Cohen, S. (2020). Elevated rates of autism, other neurodevelopmental and psychiatric diagnoses, and autistic traits in transgender and gender-diverse individuals. *Nature Communications*, *11*(1), 3959. https://doi.org/10.1038/s41467-020-17794-1

Webb, T. L., & Sanders, J. E. (2020). Pediatric Pain. In D. H. Cisewski & S. M. Motov (Eds.), *Pain Management Guide* (1st ed.). EMRA.

Weidenbacker, R., Sandry, M., & Moed, G. (1963). Sensory Discrimination of Children with Cerebral Palsy: Pressure/Pain Thresholds on the Foot. *Perceptual and Motor Skills*, *17*(2), 603-610. https://doi.org/10.2466/pms.1963.17.2.603

Weiss, P. F., Chauvin, N. A., Klink, A. J., Localio, R., Feudtner, C., Jaramillo, D., Colbert, R. A., Sherry, D. D., & Keren, R. (2014). Detection of Enthesitis in Children With Enthesitis-Related Arthritis: Dolorimetry Compared to Ultrasonography. *Arthritis & Rheumatology*, *66*(1), 218-227. https://doi.org/10.1002/art.38197

Whitney, D. G., & Shapiro, D. N. (2019). National Prevalence of Pain Among Children and Adolescents With Autism Spectrum Disorders. *JAMA Pediatrics*, *173*(12), 1203-1205. https://doi.org/10.1001/jamapediatrics.2019.3826

Wiech, K., & Tracey, I. (2009). The influence of negative emotions on pain: Behavioral effects and neural mechanisms. *NeuroImage*, *47*(3), 987-994. https://doi.org/10.1016/j.neuroimage.2009.05.059

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2015). The Interplay Between Sensory Processing Abnormalities, Intolerance of Uncertainty, Anxiety and Restricted and Repetitive Behaviours in Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *45*(4), 943-952. https://doi.org/10.1007/s10803-014-2248-x

Williams, A. C. C., & Craig, K. D. (2016). Updating the definition of pain. *PAIN*, *157*(11), 2420-2423. https://doi.org/10.1097/j.pain.0000000000000613

Williams, S., Leader, G., Mannion, A., & Chen, J. (2015). An investigation of anxiety in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *10*, 30-40. https://doi.org/10.1016/j.rasd.2014.10.017

Willig, C., & Stainton Rogers, W. (2017). *The SAGE Handbook of Qualitative Research in Psychology* (2 ed.). SAGE Publications Ltd.

Wilson, R. B., Thompson, A. R., Rowse, G., & Freeth, M. (2023). The experience of seeking, receiving, and reflecting upon a diagnosis of autism in the UK: A meta-synthesis of qualitative studies conducted with autistic individuals. *Research in Autism Spectrum Disorders*, *103*, 102135. https://doi.org/10.1016/j.rasd.2023.102135

Winger, A., Kvarstein, G., Wyller, V. B., Sulheim, D., Fagermoen, E., Småstuen, M. C., & Helseth, S. (2014). Pain and pressure pain thresholds in adolescents with chronic fatigue syndrome and healthy controls: a cross-sectional study. *BMJ Open*, *4*(10), e005920. https://doi.org/10.1136/bmjopen-2014-005920

Winter-Messiers, M. A., Herr, C. M., Wood, C. E., Brooks, A. P., Gates, M. A. M., Houston, T. L., & Tingstad, K. I. (2007). How Far Can Brian Ride the Daylight 4449 Express?: A Strength-Based Model of Asperger Syndrome Based on Special Interest Areas. *Focus on Autism and Other Developmental Disabilities*, *22*(2), 67-79. https://doi.org/10.1177/10883576070220020701

Wolff, N., Stroth, S., Kamp-Becker, I., Roepke, S., & Roessner, V. (2022). Autism Spectrum Disorder and IQ – A Complex Interplay. *Frontiers in Psychiatry*, *13*. https://doi.org/10.3389/fpsyt.2022.856084

Wollgarten-Hadamek, I., Hohmeister, J., Demirakça, S., Zohsel, K., Flor, H., & Hermann, C. (2009). Do burn injuries during infancy affect pain and sensory sensitivity in later childhood? *PAIN*, *141*(1-2), 165-172. https://doi.org/10.1016/j.pain.2008.11.008

Wollgarten-Hadamekl, I., Hohmeisterl, J., Zohsell, K., Florl, H., & Hermannl, C. (2011). Do school-aged children with burn injuries during infancy show stress-induced activation of pain inhibitory mechanisms? *European Journal of pain*, *15*(4), 423.e421-423.e410. https://doi.org/10.1016/j.ejpain.2010.09.001

Wong, C., Lau, E., Palozzi, L., & Campbell, F. (2012). Pain management in children: Part 1 - Pain assessment tools and a brief review of nonpharmacological and pharmacological treatment options. *Canadian Pharmacists Journal*, *145*(5), 222-225. https://doi.org/10.3821/145.5.cpj222

Wong, D. L., & Baker, C. M. (1988). Pain in children: comparison of assessment scales. *Pediatric Nursing*, *14*(1), 9-17. https://pubmed.ncbi.nlm.nih.gov/3344163/

Woodrow, K. M., Friedman, G. D., Siegelaub, A. B., & Collen, M. F. (1972). Pain Tolerance: Differences According to Age, Sex and Race. *Psychosomatic Medicine*, *34*(6), 548-556. https://doi.org/10.1097/00006842-197211000-00007

World Health Organization. (2020). *Guidelines on the management of chronic pain in children*. https://www.who.int/publications/i/item/9789240017870

World Health Organization. (2022a). Autism Spectrum Disorder. In *ICD-11: International classification of diseases* (11th ed.). https://icd.who.int/browse/2024-01/mms/en#437815624

World Health Organization. (2022b). Disorders of Intellectual Development. In *ICD-11: International classification of diseases* (11th ed.). https://icd.who.int/browse11/l-m/en#/http%253A%252F%252Fid.who.int%252Ficd%252Fentity%252F605267007

World Health Organization. (n.d.). *Adolescent health*. World Health Organization. https://www.who.int/health-topics/adolescent-health#tab=tab_1

Yardley, L. (2000). Dilemmas in qualitative health research. *Psychology & Health*, *15*(2), 215-228. https://doi.org/10.1080/08870440008400302

Yasuda, Y., Hashimoto, R., Nakae, A., Kang, H., Ohi, K., Yamamori, H., Fujimoto, M., Hagihira, S., & Takeda, M. (2016). Sensory cognitive abnormalities of pain in autism

spectrum disorder: a case–control study. *Annals of General Psychiatry*, *15*(1), 8. https://doi.org/10.1186/s12991-016-0095-1

Yoo, H., Cho, Y., & Cho, S. (2023). Does past/current pain change pain experience? Comparing self-reports and pupillary responses [Original Research]. *Frontiers in Psychology*, *14*. https://doi.org/10.3389/fpsyg.2023.1094903

Zeidan, J., Fombonne, E., Scorah, J., Ibrahim, A., Durkin, M. S., Saxena, S., Yusuf, A., Shih, A., & Elsabbagh, M. (2022). Global prevalence of autism: A systematic review update. *Autism Research*. https://doi.org/10.1002/aur.2696

Zerbo, O., Massolo, M. L., Qian, Y., & Croen, L. A. (2015). A Study of Physician Knowledge and Experience with Autism in Adults in a Large Integrated Healthcare System. *Journal of Autism and Developmental Disorders*, *45*(12), 4002-4014. https://doi.org/10.1007/s10803-015-2579-2

Zohsel, K., Hohmeister, J., Flor, H., & Hermann, C. (2008a). Altered pain processing in children with migraine: an evoked potential study. *European Journal of pain*, *12*(8), 1090-1101. https://doi.org/10.1016/j.ejpain.2008.02.001

Zohsel, K., Hohmeister, J., Flor, H., & Hermann, C. (2008b). Somatic pain sensitivity in children with recurrent abdominal pain. *The American Journal of Gastroenterology*, *103*(6), 1517-1523. https://doi.org/10.1111/j.1572-0241.2008.01911.x

Zohsel, K., Hohmeister, J., Oelkers-Ax, R., Flor, H., & Hermann, C. (2006). Quantitative sensory testing in children with migraine: preliminary evidence for enhanced sensitivity to painful stimuli especially in girls. *PAIN*, *123*(1-2), 10-18. https://doi.org/10.1016/j.pain.2005.12.015

Chapter 8.

Appendices

8 Appendices

8.1 Appendix 1. Table 32. Diagnoses Categorised by Diagnostic Group Across Included Studies in Systematic Review

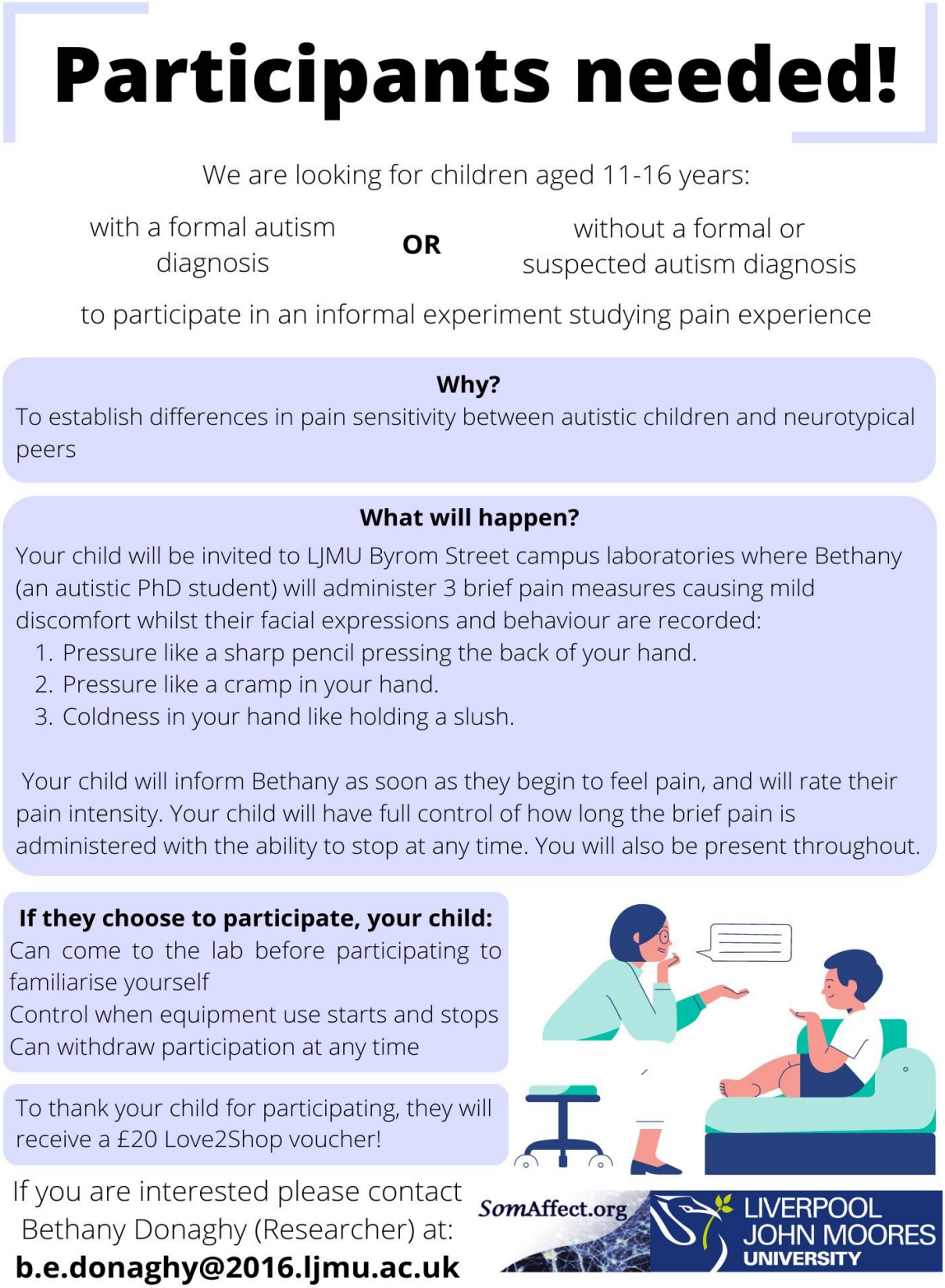
Diagnostic Group	Diagnosis	Number of studies
Pain-Related	Migraine/Headache	10
	Functional Abdominal Pain/Functional Abdominal Pain Disorder	3
	Juvenile Idiopathic Arthritis	3
	RAP	3
	Burn Injury	2
	Chronic Pain	2
	Complex Regional Pain Syndrome	2
	Enthesitis-Related Arthritis	2
	Juvenile Chronic Arthritis	2
	Musculoskeletal Pain	2
	Temporomandibular Disorder	2
	Post-Surgery	1
	Dysmenorrhea	1
	Growing Pain	1

	Irritable Bowel Syndrome	1
	Joint Pain	1
	Juvenile Fibromyalgia	1
	Osgood-Schlatter Disease	1
	Patellofemoral Pain Syndrome	1
	Hypermobility Syndrome/Ehlers Danlos Syndrome	1
	Idiopathic Neck Pain	1
	Chronic Idiopathic Chest Pain	1
Neurodevelopmental	Autism	3
	Cerebral Palsy	2
	Pre/Full-Term	2
	Down-Syndrome	1
	Intellectual Disability	1
Neuropathic	Diabetes Mellitus	2
	Sickle Cell Disease	2
	Acute Lymphoblastic Leukaemia	1
Mental Health	Non-Suicidal Self-Injury	2
	Borderline Personality Disorder	1

	Callous Unemotional Traits	1
	Self-Harm	1
Orthopaedic Disorder	Orthopaedic Disorder	2
Other	Chronic Fatigue Syndrome	1
	Pre-Menstrual Syndrome	1

Note. This table describes the frequency of diagnostic groups included in studies within Chapter 2's systematic review. Diagnostic group terminology determined by PhD researcher and affirmed by lead supervisor.

8.2 Appendix 2. Figure 7. Social Media Poster for Recruiting Participants for Pain Psychophysics Study



Note. This figure displays the social media poster used to recruit participants to a psychophysical study in Chapter 3.

8.3 **Appendix 3.1.** YouTube Video to Support Pain Psychophysics Study Participant Information Sheet for Autistic Children and Young People

To access the video link, please contact Dr David Moore (D.J.Moore@ljmu.ac.uk).

8.4 **Appendix 3.2.** YouTube Video to Support Pain Psychophysics Study Participant Information Sheet for Non-Autistic Children and Young People

To access the video link, please contact Dr David Moore (D.J.Moore@ljmu.ac.uk).

8.5 **Appendix 3.3.** YouTube Video to Display Study Environment and Full Demonstration of the Pain Psychophysics Study

To access the video link, please contact Dr David Moore (D.J.Moore@ljmu.ac.uk).

8.6 **Appendix 4.** Pain Psychophysics Study Demographic Questionnaire for All Children and Young People

Psychophysics Demographic Questionnaire

Below are a set of questions so I can learn more about you!

If you need help or if there is anything you do not know, your parent/guardian can help you!

For questions you want to answer, please tick the box next to your answer.

Here is an **example**:

Are you a human?

☑ Yes

☐ No

For questions you do not want to answer, please leave boxes blank.

Here is an **example**:

Are you an alien?

☐ Yes

☐ No

For questions with this line, please write your answer:

You will see a big smiley face when your questions end. Please let Bethany know, or ask your parent/guardian to let Bethany know when you see this face:

Insert Smiley Face

If that is all ok – please turn over and we will begin!

New page

Start of questions!

1) What is your identifying gender?

☐ Male

☐ Female

☐ Other

_____

☐ Prefer not to say

2) What is your age in years?

_____

3) What is your ethnicity?

☐ White

☐ Mixed or Multiple Ethnic Groups

☐ Asian or Asian British

☐ Black, African, Caribbean, or Black British

☐ Other Ethnic Group

_____

☐ Don't Know

☐ Prefer Not to Say

4) Do you have an Autism Spectrum Condition diagnosis?

☐ Yes

☐ No

5) What was your age in years when you received your Autism Spectrum Condition Diagnosis?

_____

6) Where did you receive your diagnosis?

☐ NHS assessment

☐ Private assessment

☐ Not sure

7) Do you have any co-occurring mental health conditions? E.g. anxiety. If yes, please specify.

☐ Yes

_____

☐ No

8) Do you have any siblings? Please select all that apply.

☐ Yes – My siblings and I share the same mum and dad

☐ Yes – My siblings only share the same Mum as I

☐ Yes – My siblings only share the same Dad as I

☐ Yes – My siblings do not share the same Mum and Dad as I

☐ No

9) Who do you currently live with?

_____

10) What is your current education level?

☐ Primary education (e.g., Primary school)

☐ Secondary education (e.g., High School)

☐ Further education (e.g., College, Sixth form)

☐ Apprenticeship

☐ None

☐ Prefer not to say

11) If you are currently attending secondar or further education, is your high school/college/sixth form:

☐ Mainstream

☐ Specialist

12) If relevant, have you begun menstruating (your period)?

☐ Yes

☐ No

☐ Prefer not to say

13) Are you currently receiving any regular medication?

☐ Yes

☐ No

☐ Don't know

☐ Prefer not to say

Thank you very much for answering these questions.

Please let Bethany know, or ask your parent/guardian to let Bethany know you have completed the questionnaire.

Insert Smiley Face

8.7 Appendix 5.1. Figure 8. Pain Intensity Scale Identified by Psychophysics PPI Panel for Guidance in Creating a Pain Intensity Scale for Pain Psychophysics Study

Note. Figure 8 displays the pain intensity scale provided by Chapter 3's PPI panel for guidance (Anonymous, n.d.).

8.8 Appendix 5.2. Amended Pain Intensity Scale Used for Pain Psychophysics Study

(Tick)	Severity		Description of Experience
	10	Unable to Move	I can't move due to my pain. I need someone to take me to the emergency room to get help for my pain
	9	Severe	My pain is all that I can think about. I can barely talk or move because of the pain.
	8	Intense	My pain is so severe that it is hard to think of anything else. Talking and listening are difficult.
	7	Unmanageable	I am in pain during other tasks (i.e. school work). It keeps me from repeating these tasks because of my pain.
	6	Distressing	I am thinking about my pain whilst doing other tasks (i.e. school work). I need to stop these tasks because of my pain.
	5	Distracting	I am thinking about my pain most of the whilst doing other tasks (i.e. school work). I might need to stop these tasks.
	4	Moderate	I am aware of my pain during other tasks (i.e. school work) but I can continue.
	3	Uncomfortable	My pain bothers me but I can ignore it most of the time.
	2	Mild	I have a low level of pain. I am aware of my pain only when I pay attention to it.
	1	Minimal	My pain is hardly noticeable.
	0	No pain	I have no pain.

8.9 **Appendix 6.** Pain Psychophysics Original Statistical Analysis Normative to Blankenburg (2010)

Data were assessed against parametric assumptions. Levene's Test was used to ascertain homogeneity of variance amongst data, which was assumed across both Autistic and non-Autistic threshold measures ($p > .05$). K-S tests and skew values (±1.96) indicated normal distribution across PPTh Autistic and non-Autistic, and MPTh Autistic data, $p > .05$, but not MPTh non-Autistic, $D(18) = 0.27$, $p = .001$. However, due to small n value across groups, this was not deemed to violate parametric assumptions and thus independent t-tests were used to compare z-score MPTh and PPTh differences between Autistic and non-Autistic CYP. Throughout each test $p > .05$ will deem non-significant results (Coolican, 2004).

On average, Autistic CYP ($M = 3.41$, $SD = 0.98$) and non-Autistic CYP ($M = 3.35$, $SD = 0.96$) both shown higher MPTh than normative values, However, thresholds did not significantly differ between groups failing to support a profile of MPTh hyposensitivity amongst Autistic CYP ($t(26) = 0.44$, $p = .884$). Moreover, Autistic CYP ($M = -2.35$, $SD = 1.56$) and non-Autistic CYP ($M = -1.81$, $SD = .96$) both showed lower PPTh than normative values. Additionally, Autistic CYP PPTh's suggested a much lower force was required to perceive pain than their comparative group and a profile of hypersensitivity could be considered. However, differences between groups were not significant failing to support a PPTh hypersensitive amongst Autistic CYP ($t(25) = -1.12$, $p = .273$).

8.10 Appendix 7. Figure 9. Social Media Poster for Recruiting Participants for Interview Study

Note. Figure 9 displays the social media poster used to recruit participants to an interview study in Chapter 4.

8.11 **Appendix 8.** YouTube Video to Support Interview Study Participant Information Sheet for Autistic Children and Young People

To access the video link, please contact Dr David Moore (D.J.Moore@ljmu.ac.uk).

8.12 Appendix 9.1. Table 33. Questions Asked to Autistic CYP in an Online Questionnaire

What We Must Know	What May Want to Know (If Comfortable Talking About)
Identifying gender	Education level
Age	School type (i.e. mainstream or specialist)
Autism diagnosis?	Pre-term birth?
Age at Autism diagnosis?	If female, have they began menstruating?
Formal pain diagnosis?	Currently receiving any medication?
Recurring pain problem?	
Co-occurring (i.e. anxiety, depression, etc.)	
Is the co-occurrence formal?	
Any siblings? Biological, half or adopted.	
Current familial structure (who lives in the home)?	
Ethnicity?	
Can you identify a recent time you have experienced pain lasting less than 12 weeks (British Pain Society, 2021) that we can talk about during our interview? Examples may include: Stomach-ache, Headache, Grazed knee, Twisted ankle	
If yes, can you give a brief description? For example, I had to stay off school last week because I had a stomach-ache.	
Is there anything ese you want to tell us?	
Is there anything we should know to be more comfortable in interview? E.g. presence of certain things?	

Note. This table describes the questions asked to Autistic CYP in Chapter 4's online questionnaire.

8.13 Appendix 9.2. Table 34. Questions Asked to Caregivers in an Online Questionnaire

What We Must Know	What May Want to Know (If Comfortable Talking About)
Identifying gender	Education Level
Age	Occupation
Ethnicity	Does parent experience chronic pain?
	Autism diagnosis?
	Does anyone else in the family have an Autism diagnosis?
	Is anyone in the family suspected to be Autistic?
	Co-occurring (i.e. anxiety, depression, etc.)
	Is the co-occurrence formal?
Is there anything should know to be more comfortable in interview? E.g. presence of certain things?	

Note. This table describes the questions asked to caregivers in Chapter 4's online questionnaire

8.14 Appendix 10.1. Table 35. Interview Schedule Used for Interviewing Autistic CYP

Topic of Interest	Interview Question	Prompts
Pain importance	What does pain feel like for you? In your own words.	
	How often do you feel pain?	Is there a pain you feel the most? i.e. stomach ache Is the pain regular e.g. is it there all the time? Is it only occasionally?
	Tell me about things you find difficult when you are in pain.	I.e. going to school, socialising Yes: Does this always happen? No: So you can still do everything you do when you are not in pain?
	Do you know when your pain starts or ends?	Interoception; do you know when you are in pain, can you identify this?
Informal chat about pain scenario given in pre-survey. If none/can't remember, introduction of possible pain scenario; scenario that resonated most to be used throughout. Images shown as a visual prompt. I am going to ask you a few questions now, does that sound ok?		
Pain scenario	Identifying information on scenario If struggling, ask if maybe *parent/guardian* can help?	What happened? How long ago was the *scenario* (yesterday, last week, month, year)? Who were you with when you *scenario*? (i.e. mum/dad, friend, teacher)

		Did they see you hurt yourself?
So, thinking about this pain we have just been talking about…		
Pain description	Can you describe the pain you felt to me? *Show picture for help*	Where do you feel in pain? Does the pain only stay there, or does it spread to other parts of your body? How does the pain feel? (Hot, itchy, stings etc.) Do you feel the pain straight away, for example as soon as you fell off your bike did your knee begin to hurt?
Pain behaviour	What do you do when you feel pain?	Do you tell anyone? – Who, why, how (if mum can ask to show), does this help (how)? Is there an object/thing that you use? i.e. special interest previously identified in conversation, a fidget toy, a weighted blanket, an iPad/tablet to watch something – What, why, does this help (how)? Do you take medicine? Or watch a tv programme? Sleep? Socialise? Is this what you did when you *scenario*

	Is there anything that doesn't help the pain go away or not be as bad? i.e. walking on a sore foot, won't go to a loud place.	Why won't you do that? Anything that makes the pain worse?
Cognitive pain states	Tell me what goes through your mind when you are in pain?	For example, some people may only be able to think that their head hurts?
	Some people find it more difficult to pay attention to things like the TV when in pain, does this happen to you and can you talk about it?	Why do you think that is?
	Some people find it more difficult to remember things like doing their homework when in pain, does this happen to you and can you tell me about it?	Why do you think that is?
	Some people find it more difficult to understand things like people speaking verbally to them, does this happen to you and can you tell me about it?	Why do you think that is?
Emotional pain states	Tell me about the emotions you feel when you are in pain.	i.e. Do you feel happy, sad, angry? Are any of the emotions you feel shown on this picture? Do you think you show these emotions?

	Tell me how you may show these emotions?	i.e. You might cry when you are sad, do you ever cry when you are in pain?
Closing	You have done so great today and given us lots of great information so we will finish in a second; but before we do is there anything else you want to tell me/us/giraffe?	

Note. This table provides the interviews schedule used to interview Autistic CYP in Chapter 4.

8.15 Appendix 10.2. Table 36. Interview Schedule Used for Interviewing Caregiver's

Topic of Interest	Interview Question	Prompts
Pain importance	Can you tell me about how often your child experiences pain?	What type of pain does your child experience most often? How long does this pain last?
	Tell me about anything you may use or have in place to help your child cope with/alleviate pain?	Is there a certain teddy that is used for comfort? Is there a certain pattern of behaviours you follow? Does your child seek more or less of their special interest when in pain?
	Does your child experience pain related fears?	What are they? Do you know the rationale/event behind these fears?
Pain description	Do you feel you are usually aware when your child is experiencing pain at the time of the pain?	Does your child let you know in the moment, or after the pain has passed? Do you know your child is in pain before they say?
	Tell me how your child communicates and describes their pain to you?	**Verbal:** type of language they would use, would they say directly to you? Would they write or draw it down? Would they act out using toys? **Implicit:**

		would they point to site of pain, would they cry, would their behaviour change, and you had to recognise this?
	Would your child communicate their pain to someone other than you?	If yes can you describe to me who they would communicate their pain to? Why do you think your child would communicate their pain to that/those individual(s)? Is there anyone you think you child would not communicate their pain to? Why do you think your child would not communicate their pain to that/those individual(s)? Would their communication differ, if yes how? Do you find you often explain how your pain feels pain for others? Do you need to prep before Dr appointments how to describe pain?
	Can you tell me about anything that may change how your child communicates their pain?	i.e. pain severity, external stressors, location, type of pain

	Do you think your child is able to communicate their pain effectively?	If yes, what makes their communication effective? If no, why is their communication not effective?
Pain behaviour	Tell me about any behaviours your child performs when in pain?	How is this behaviour useful to them? How is this behaviour useful to you (i.e. recognising pian severity)?
	Tell me about any behaviours your child does not perform when in pain?	Does your child show for example a higher level of avoidance behaviour?
Emotional pain states	Tell me about the emotions your child experiences and shows when they are in pain?	Happy, Sad, Anger, Neutral? How do you know they are showing this emotion?: Does your child show any emotional behaviours when in pain? I.e. does the child cry, does the child have a meltdown? Are there any emotions your child shows specific to the pain they are in? I.e. cry more when fall over, but more angry when have a stomach-ache
	How does this emotion differ to when your child is not in pain?	What types of things would cause a change in their emotions?

Cognitive pain states	Tell me about anything your child finds difficult to do when in pain?	For example, explaining themselves or socialising? If yes in what way? Is this dependent on pain severity? Does your child's ability to pay attention to things such as the tv differ – how, what is attention usually like? Does your child's ability to remember things such as completing homework differ - how, what is memory usually like? Does your child's ability to understand things such as verbal communication differ – how is their ability to understand communication usually?
	Is there anything your child finds easy to do when in pain?	For example, explaining themselves or socialising? If yes in what way? Is this dependent on pain severity?
Additional*	*if parent perhaps has a neurotypical child: Would you say your child's response to pain differs from their siblings/other individuals in your child's life?	Other: family/friends/children/schoolmates If, yes how?
Closing	Is there anything else we have not discussed you would like to tell us about?	

Note. This table provides the interviews schedule used to interview caregivers in Chapter 4.

8.16 Appendix 11. Table 37. Contextual Pain Scenarios Provided to Autistic CYP Who Did Not Identify or Want to Use a Disclosed Pain Scenario During Interviews

Scenario	Possible Approach
Falling over on playground and scraping *insert*	What do you do at lunchtime at school? *Child: I play with friends/I am with friends/Nothing [next scenario]* Do you play any games at lunchtime with your friends/What do you play? *Child: tag/hide and seek/football/nothing [next scenario]* Have you ever fell over whilst playing, maybe in the playground? *Child: Yes* What happened? *Child: Nothing, I got back up [next scenario]/I hurt my *knee/elbow/hand/wrist/leg** How did you hurt it? *Child: …* **Image 1**. Playground School Children Playing

	 *Note.* Image from Raponi (n.d.).
Falling when learning to ride a bike and hurting *insert*	Have you ever learned to ride a bike? *Child: Yes/No [next scenario]* I remember when I was learning to ride a bike/when I used to ride my bike and I fell off a few times, did this happen to you? *Child: Yes/No [next scenario, or parent might intervene and say yes you did; if so carry on]* Did you hurt yourself when you were fell off your bike? *Child: I hurt my *leg/hand/arm*/I was bleeding* How did you hurt it?

	Child: … **Image 2.** Girls Bicycle Helmets  *Note.* Image from Skitterphoto (n.d.).
Sports injury	Do you play any sports? *Child: Yes/No* Which sport do you play? *Child: Football/Rugby/Etc* Have you ever been injured/hurt yourself whilst playing?

Child: Yes/No [next scenario, or parent might intervene and say yes you did; if so carry on]

Do you think we can talk about this?

Child: Yes/No [next scenario]

Image 3.

Child Soccer Playing

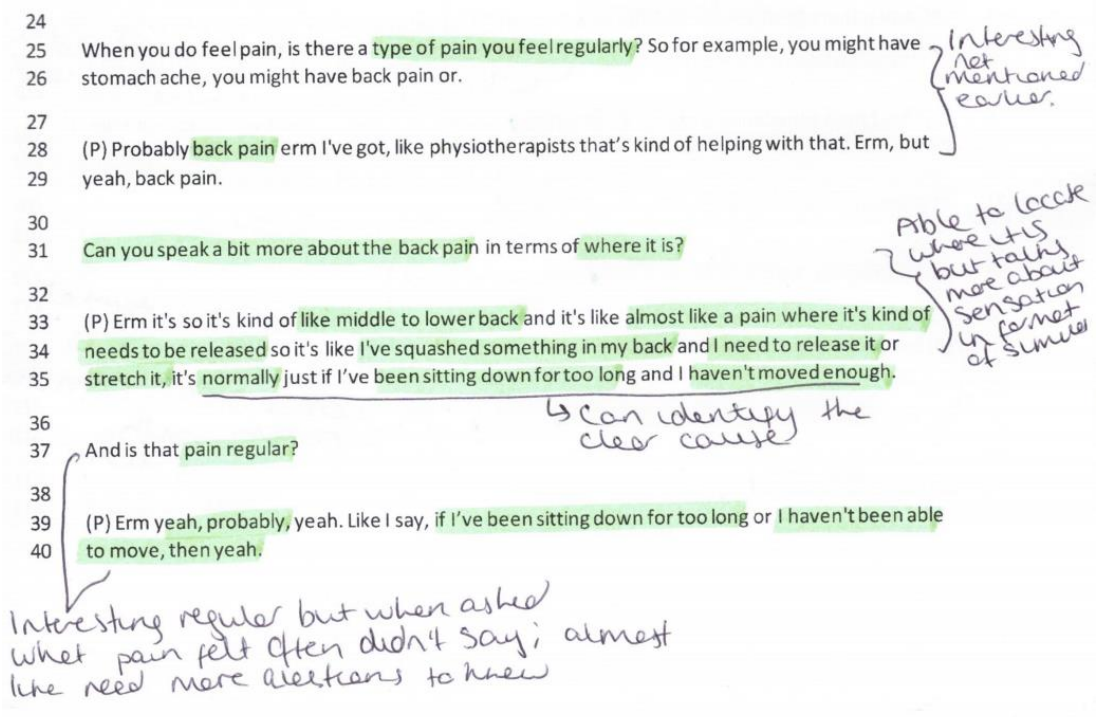
Note. Image from Unknown (n.d.-a).

Being ill with stomach-ache and having associated *stomach pain*	Can you remember a time you were poorly? *Child: Yes/No [next scenario]* Did you have a sore stomach, or stomach-ache when you were poorly? *Child: Yes/No [next scenario]* Ok, so did this hurt? *Child: …* **Used image removed as deleted by source**
Having a headache and associated *head pain*	Can you remember a time you were poorly? *Child: Yes/No [next scenario]* Did you have sore head, or headache, when you were poorly? *Child: Yes/No [next scenario]* Ok, so did this hurt? *Child: …* **Image 4.** Medication Child Hurts Got Sick

Note. Image from Unknown (n.d.-b).

Note. This table provides the exemplar pain scenarios prepared for interview use in Chapter 4.

8.17 Appendix 12. Figure 10. Example of a subsect of PhD researchers noting of descriptive, linguistic, and conceptual comments on a CYP's transcript



Note. Figure 10 displays an example of initial noting for a subsect of analysis for an Autistic CYP's transcript in Chapter 4.

8.18 Appendix 13. Table 38. Example of a subsect of interpreting exploratory comments from an Autistic CYP's transcript to create emergent themes

Emerging Themes	Original Transcript	Exploratory Comments
Patterns of pain	… would you be able to think back to it if we have a conversation about it? *Uh, they're kind of all the same, but probably yeah.* OK, that's fine. So your last headache, can you say what happened? *So I was just playing with. (2 second pause) two of my friends and I think my cousin, er. Then it was tea time, so I, I went down for tea and I just have this horrible headache and.*	Again, this patterned pain behaviour; interesting that the headaches are what are spoke about, perhaps a these as the most memorable.
Interoceptive ability	*Mostly when I've got a terrible headache, it means I can't eat.* What is it about the headache that makes it so you can't eat? *I just feel like so full for some reason.*	Interoceptive processing whereby notices an internal feeling of fullness despite having not eaten, could this be as headache is making participant feel sick? However still interpreted as a fullness.

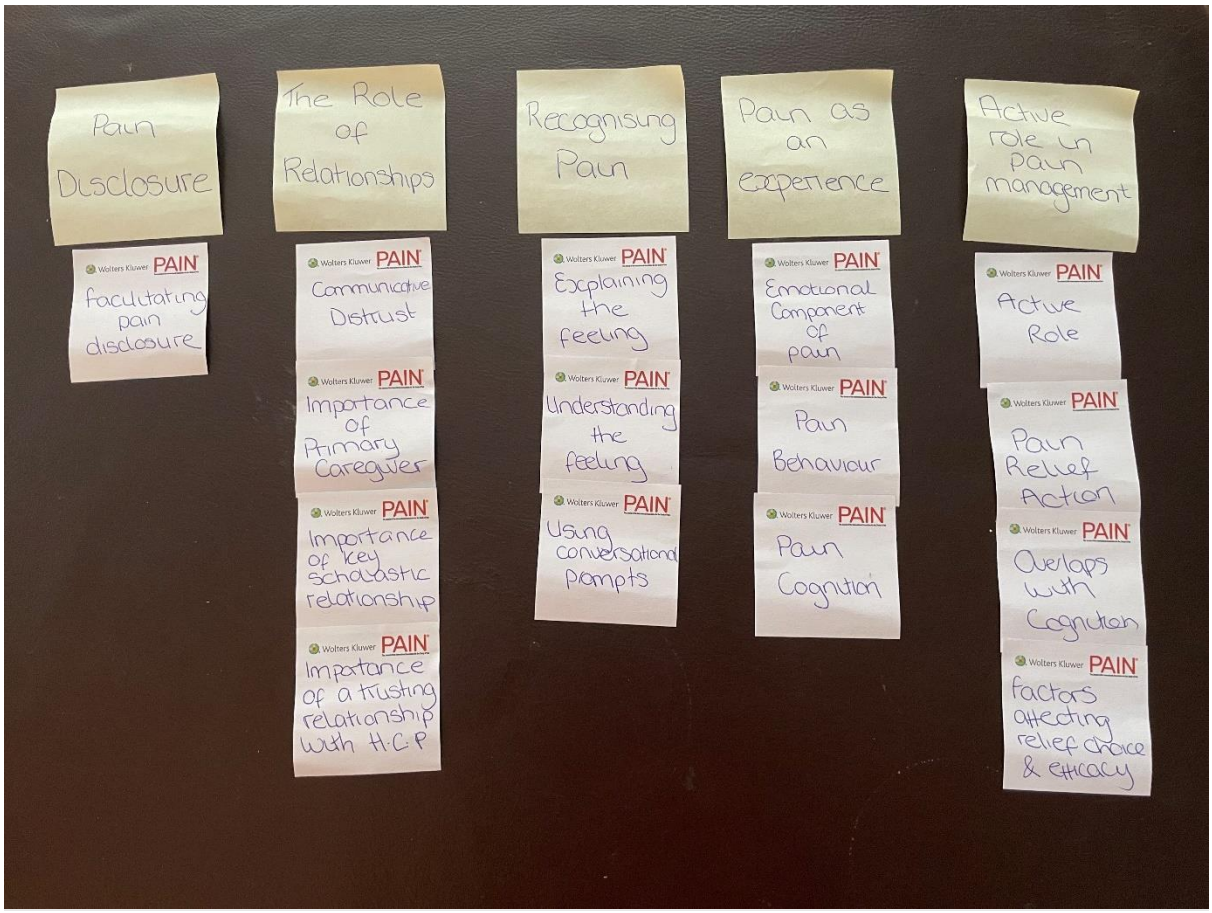
Mum-Child relationship (disclosure)	who was you with then? *Erm mum, brother and sister. And then that dad was away.* Ok and dad was away. Right, OK, er, so did you tell anyone you had a headache? *Er I, I can't remember.* *(Mum speaks don't know what is said)* *Yeah, I normally do when I come down 'cause as mum said, I'm not. She normally just puts it in the oven until I'm ready.* **…** So it's usually mum then that you tell when you have a headache. *Yeah.*	Mum was informed that participant was in pain; perhaps told as they are the one who provides a caring context (i.e. putting tea in the oven until feels better). However, appears it is usually mum who is told.
Parental scaffolding	*In context of telling anyone in pain* *(Mum speaks don't know what is said)*	Mum interjects to help participant answer when they can't remember.

Pain cause identified (pattern of pain)	*If I've got a horrible headache and I keep eating then I just throw up and that's not fun.*	Clear cause and effect which helps to explain behaviour, and why such behaviour must be sustained.
Pain location ability	**…** are you able to say where you felt the pain? *It's normally just the same place right at the front of my head.* And does the pain only stay there? *Yeah.*	Able to locate and communicate pain location without prompts, and how this continual location remains.

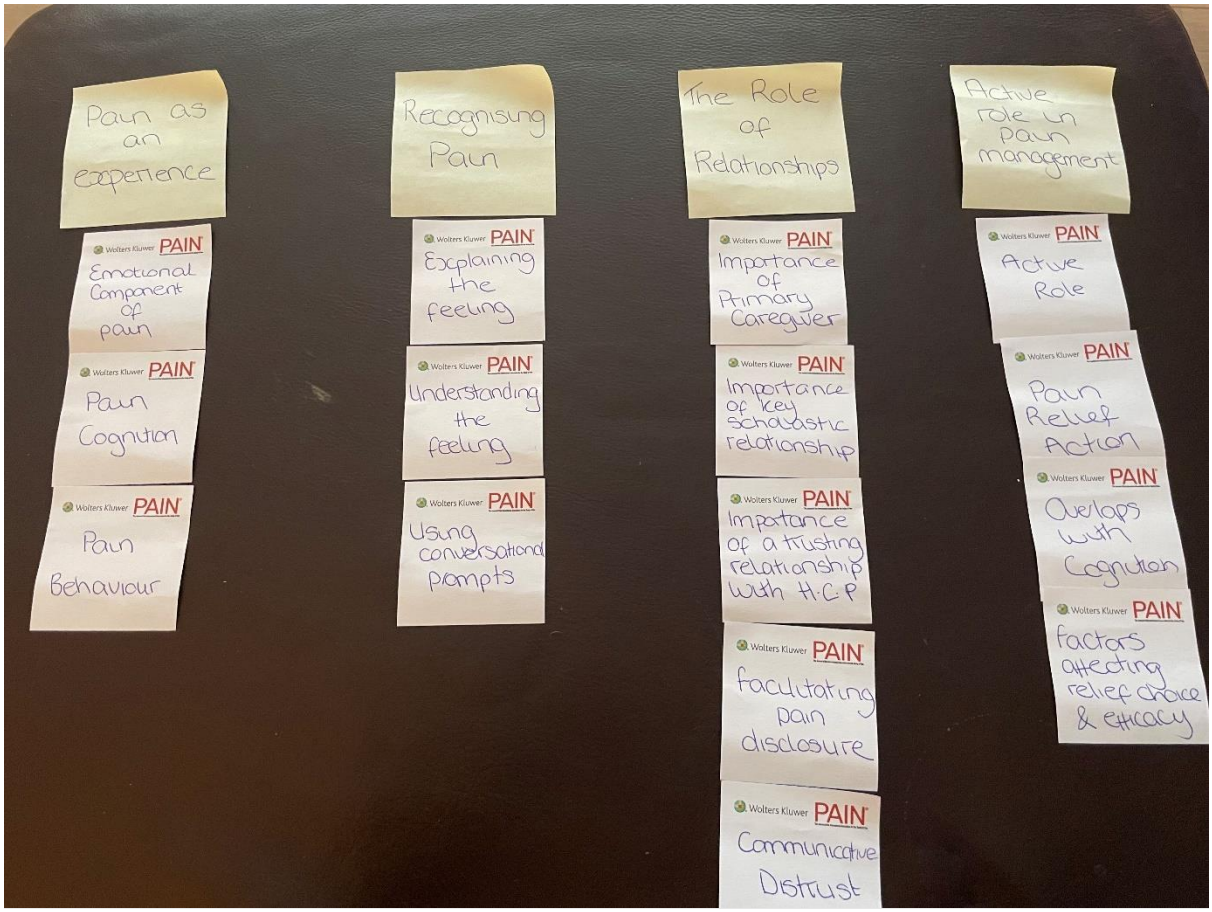
Note. This table provides an example of interpreting exploratory comments in an Autistic CYP's transcript in Chapter 4.

8.19 Appendix 14. Figure 11. Theme Naming Process for IPA

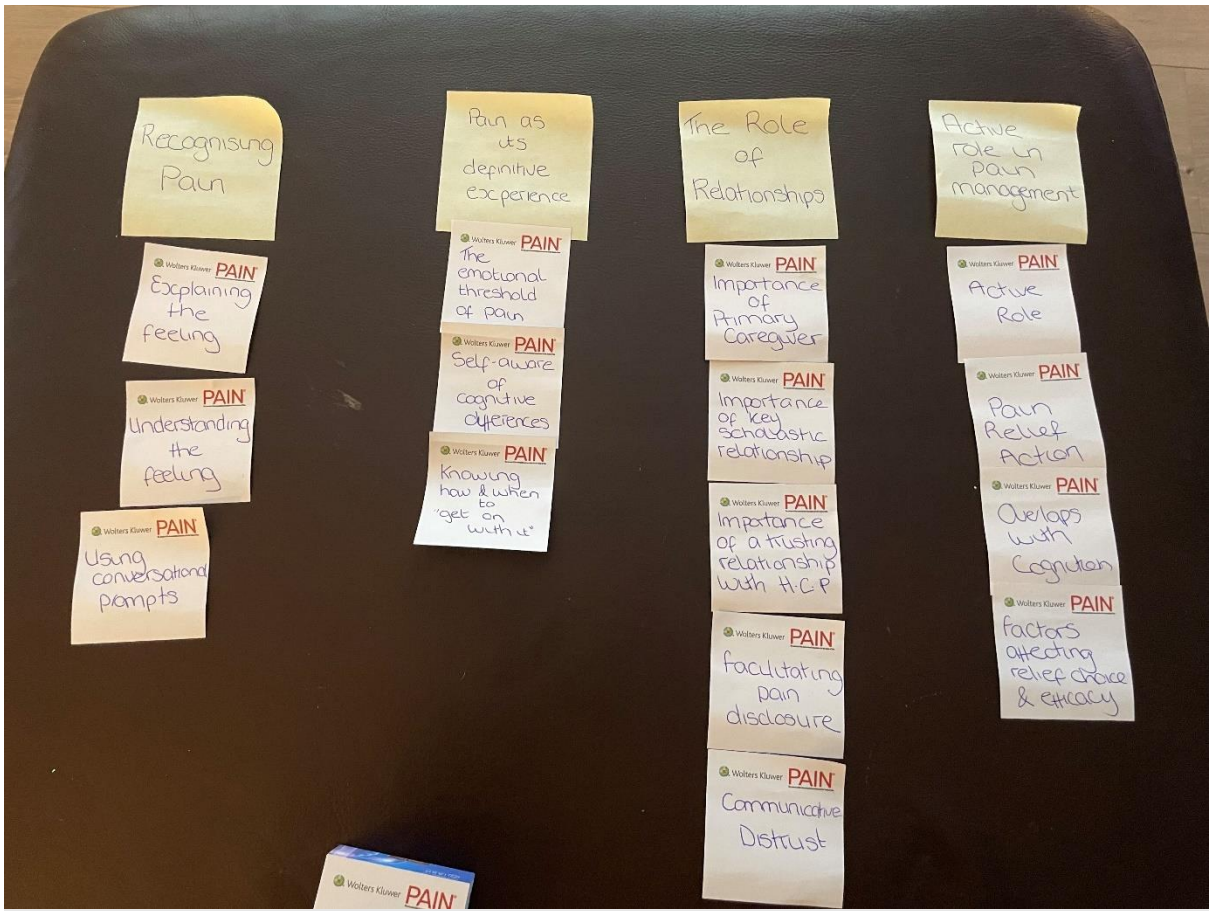
Step 1: Original themes as discussed between B.D., D.M. and H.P.



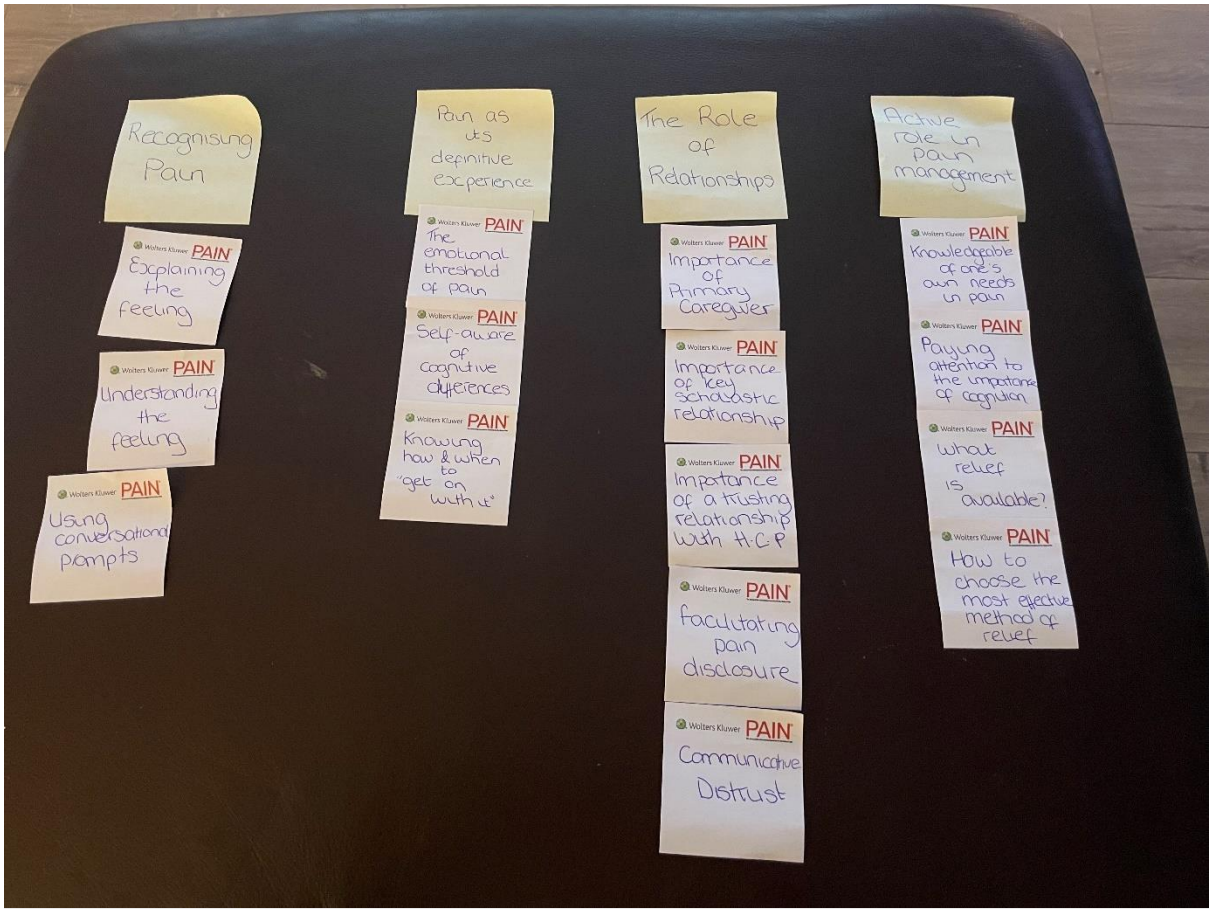
Step 2: (1) Pain disclosure as a superordinate theme collapsed into The Role of Relationships. (2) Order of superordinate themes in explaining the “story” reorganised.



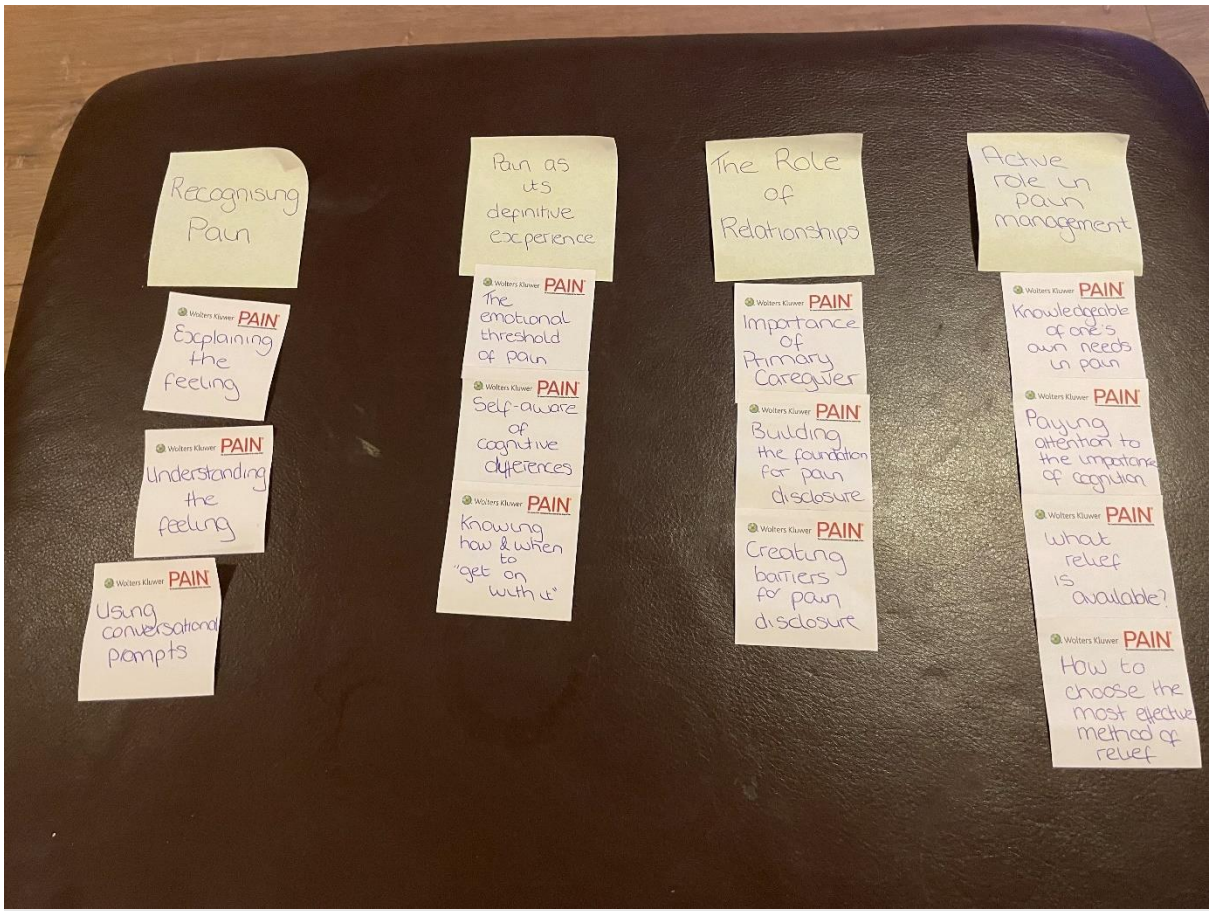
Step 3: (1) Order of superordinate themes in explaining the “story” reorganised. (2) “Pain as an experience” superordinate theme renamed. (3) Subordinate themes within “Pain as its definitive experience” renamed to be more statement based and explanatory. Later changed to “How Pain is Experienced” in thesis corrections.



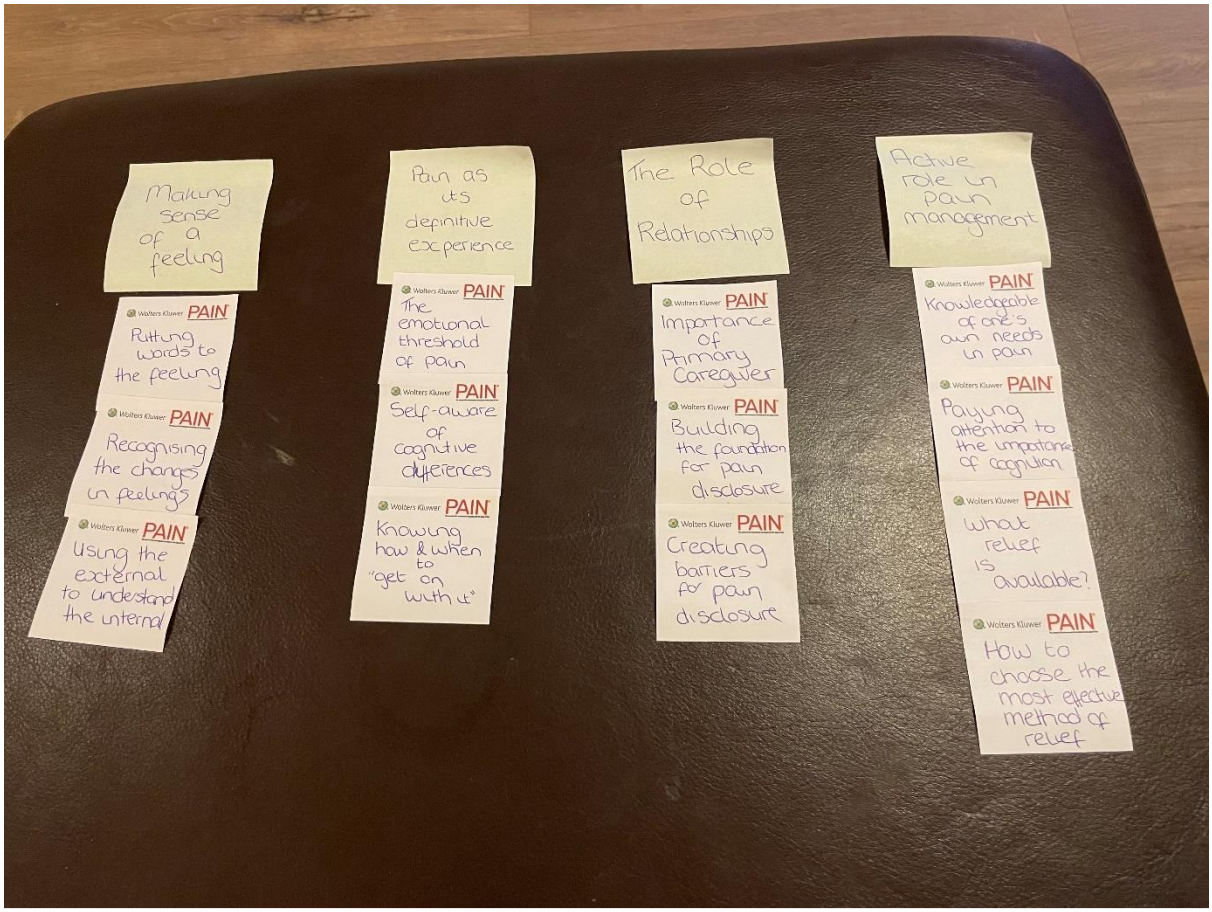
Step 4: Subordinate themes within “Active role in pain management” renamed to be more statement based and explanatory and ordered into a process.



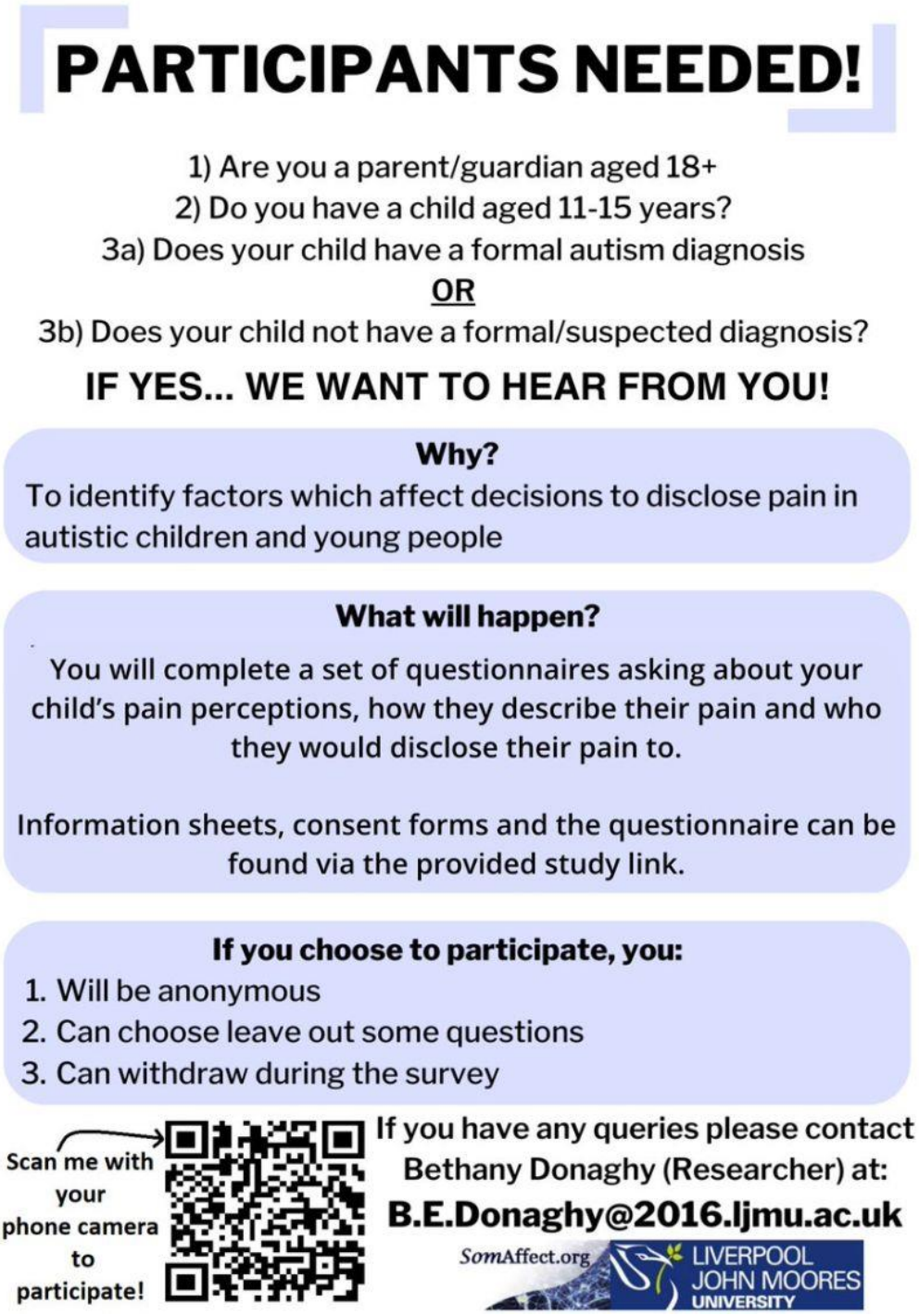
Step 5: (1) Subordinate themes within “Role of Relationships” renamed to be more statement based. (2) Importance of key scholastic relationships and HCP collapsed and now encompassed by their own traits (i.e. trust, communication) with mention in explanations to how these traits effect disclosure in the latter relationships.



Step 6: (1) “Recognising pain” superordinate theme them renamed. (2) Subordinate themes within “Making sense of a feeling” renamed to be more statement based and explanatory.



8.20 Appendix 15. Figure 12. Social Media Poster for Recruiting Participants for Online Survey Study



Note. Figure 12 displays the social media poster used to recruit participants to an online survey study in Chapter 5.