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PROMIS Paediatric Lower Limbs Mobility Tool is Correlated with Accelerometer-Measured Physical Activity in Children with Hip Diseases

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ABSTRACT

Background: The reduction in mobility due to hip diseases in children is likely to affect their physical activity (PA) levels. Physical inactivity negatively influences quality of life and health. Our aim was to objectively measure PA in children with hip disease, and correlate it with the PROMIS Mobility Score (lower limbs).

Patients and Methods: 28 children (12 boys and 16 girls) with hip disease aged 8 to 17 years (mean 12±3 years) were studied between December 2018 and July 2019. Children completed the PROMIS Paediatric Item Bank v. 2.0 –Mobility Short Form 8a (lower limbs) and wore a hip worn accelerometer (ActiGraph) for 7 consecutive days. Sedentary time (ST), light PA, moderate to vigorous PA (MVPA) and vigorous PA were calculated from accelerometers data. Lower limbs mobility from the PROMIS questionnaire was classified as normal; mild; and moderate functions, based on the PROMIS cut scores on the physical function metric. A one-way ANCOVA was used to assess differences among mobility (normal; mild; moderate) and measured PA and relationships between these variables were assessed using bivariate Pearson Correlations.

Results: Children classified as normally functioning on the PROMIS had less ST (p=0.002); higher MVPA (p=0.002) and VPA (p=0.004) compared to those classified as mild or moderate function. A moderate correlation was evident between the overall PROMIS score and daily LPA ($\mathbf{r}=0.46$, $\mathbf{n}=28$, $\mathbf{p}=0.01$), moderate-to-vigorous PA ($\mathbf{r}=0.67$, $\mathbf{n}=28$, $\mathbf{p}=0.01$) and vigorous PA (VPA) ($\mathbf{r}=0.54$, $\mathbf{n}=28$, $\mathbf{p}=0.01$). No correlation was evident between the average daily ST and overall PROMIS score ($\mathbf{r}=-0.28$, $\mathbf{n}=28$, $\mathbf{p}=0.15$)

Conclusion: PROMIS lower limbs mobility tool is correlated to the PA level of children with hip disease and may provide a general overview of PA.

KEYWORDS: physical activity; children's hip disease; accelerometers; PROMIS; mobility

INTRODUCTION

Several conditions with varying clinical significance and severity can affect the hip joint in childhood, such as Perthes' Disease, Developmental Hip Dysplasia (DDH) and Slipped Capital Femoral Epiphysis (SCFE) (1, 2). Irrespective of the cause, the main symptoms of childhood hip disease are pain and limitations in hip joint mobility (1, 2). These symptoms negatively influence the quality of life of affected children through limitations in their usual activities, including walking, running and playing with their friends (3).

Patient Reported Outcome Measures (PROMs), that assess the physical, emotional and social impact of a disease, provide information on the daily activities and the related quality of life (4, 5). In the US, The National Institute of Health's Patient Reported Outcomes Measurement Information System (PROMIS) has been developed (6). The PROMIS questionnaires were designed to overcome limitations present in legacy outcome tools, and to standardise the tools used and the outcomes measured (6). The PROMIS tools were created using a robust programme of development to detect differences in outcomes of diseases in adults and children, such as pain or limitations. PROMIS tools have already been used to assess the impact of different symptoms (e.g. pain; physical function) on the quality of life of patients with paediatric conditions (e.g. Perthes' Disease; Cerebral Palsy) (7, 8). There are many versions of PROMIS, of which the PROMIS Paediatric Mobility specifically examines the functional impact of conditions on lower limbs in children.

Pain and reduced mobility of the hip joint likely limit physical activities in children with hip disease, supporting sedentary behaviour (e.g. sitting and lying). There are few studies within the literature that examine the impact of hip disease on physical activity, and most employ only a patient reported outcome questionnaire (e.g. PROMIS Mobility or EQ-5D-3L) (9, 10). Accelerometers have effectively been used in research to assess physical activity and sedentary behaviour in children (11, 12). However, no study has used accelerometers to assess if physical activity is reduced and sedentary behaviour is increased, in children with hip disease. Furthermore, no study has explored the correlation between reported mobility, using the PROMIS Mobility outcome tool, and the objectively

assessed level of physical activity and sedentary behaviour in children affected by hip disease. The aim of our study was to objectively measure PA in children with hip disease, and correlate it with the PROMIS Mobility Score (lower limbs).

METHODS

Participants: We sought to recruit 30 children with hip disease during routine clinical appointments between December 2018 and July 2019. We included children diagnosed with hip disease (e.g. Perthes' Disease; Slipped Capital Femoral Epiphysis, Developmental Dysplasia of the Hip) aged between 8 and 18 years old. Patients were approached during the clinical visit by the research team; the study was explained verbally and in writing before informed consent was obtained. The sample size sought was inflated by 10% to 34 children, to anticipate dropouts and poor-quality data. The study was approved from the Research Ethics Committee and adhered to the 1964 declaration of Helsinki and its later amendments. Patient notes were screened by the clinician in charge to assess for study eligibility. Exclusions were made for children with restricted activity that was not solely related to hip disease (e.g. children with neuromuscular diseases); those unable to adhere to the protocol (i.e. through learning difficulties or problems with communication); or those with an enforced period of inactivity (i.e. bed rest period; wheelchair; cast).

Experimental design: Following informed consent, children completed the PROMIS paediatric item bank v. 2.0 –Mobility Short Form 8a (lower limbs). Children were then fitted with a hip-worn accelerometer (ActiGraph) and instructed to wear the accelerometer for 7 days during waking hours.

Measurements:

a) PROMIS Questionnaire. Participants completed the PROMIS paediatric item bank v. 2.0 – Mobility Short Form 8a (lower limbs) (6, 13). The questionnaire has 8 items to investigate the general impact that patient's lower limbs mobility had on their daily activity tasks (such as playing with

friends; or walking up the stairs), and was scored from 1 (*not able to do it*) to 5 (*with no trouble*). As per PROMIS guidelines (16), raw score was calculated as the sum of each item score. Scale score was calculated from raw score using the PROMIS conversion table (13).

b) Physical Activity. Physical activity was monitored using a tri-axial accelerometer (Actigraph wGT3x-BT). Participants wore the accelerometer on their right hip for 7 consecutive days. The hip on which the accelerometer was worn did not depend on the side of the disease, as this was simply a standardised location to record general physical activity. Participants were allowed to remove the device for sleeping and water-based activities. Participants recorded in a diary the times the device was worn each day. Accelerometer non-wear time was defined as 90 consecutive minutes of zero counts.min⁻¹ (14). Data were deemed acceptable for analysis if there was \geq 7 hours of wear time per day (15), for a minimum of 4 days, including one weekend day (16). The ActiLife software, version 6.2 (ActiGraph, Pensacola, Florida) was used to download the data and to perform scoring and wear-time validation analysis. Raw acceleration data were converted to 60s-epoch activity count data (counts·min⁻¹). Physical activity intensity was determined using the following cut points (17): light (\geq 150 counts.min-1), moderate (\geq 500 counts.min-1), and vigorous (\geq 4000 counts.min-1). Physical activity data were exported and handled in Excel (Microsoft), and total time (minutes) spent in light, moderate and vigorous Physical activity was calculated (Figure 1).

Statistical Analysis:

Data were checked for outliers and Shapiro-Wilk Test was employed to ensure residuals were normally distributed (p>0.05). A Bivariate Pearson Correlation Test (two-tailed) was employed to examine linear correlations between the average daily physical activity in minutes (sedentary time -ST; light – LPA; moderate-vigorous – MVPA; vigorous – VPA) and PROMIS questionnaire Scale Score. A one-way ANCOVA was employed to examine differences in physical activity levels (ST; LPA; MVPA; VPA) among groups divided by PROMIS Scale Score (moderate; mild; normal). Analyses were adjusted for the effects of age, BMI, height, weight and accelerometer wear time. Statistically significant group differences were followed up with Bonferroni pairwise comparisons. The statistical analysis was performed using IBM SPSS Statistics for Windows, version 26.0 (IBM Corp., Armonk, N.Y., USA).

RESULTS

28 children returned the activity monitors with data acceptable for analysis using the predefined criteria. This included 12 boys and 16 girls. The mean age of participants was 12 years (SD \pm 3 years). The diseases included in the analysis were Perthes' Disease (16), SCFE (3) and DDH (9, of whom 1 had severe AVN). 24 children had undergone prior surgery. A breakdown of participants is available in Appendix A.

Activity Monitors. Average daily ST was higher (73% of wear time) than average daily Total Activity (TA) time (27% of wear time). Of the average daily TA, the highest amount of time was spent in MVPA (18%), with remaining activity time spent in LPA (8%), and only 1% of the activity time spent in VPA (Figure 1).

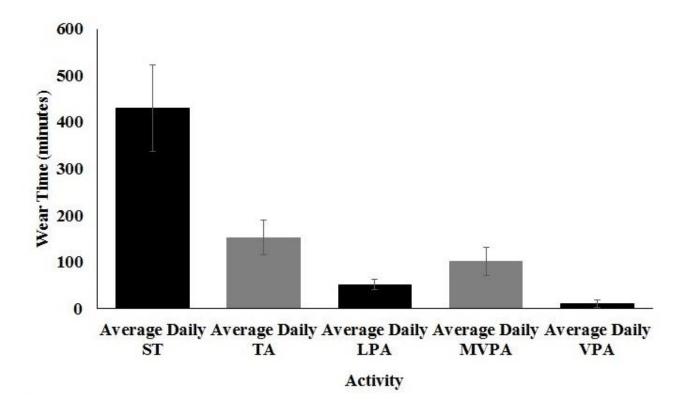


Figure 1: Children's PA data collected using accelerometers. ST=Sedentary Time; TA=Total Activity; LPA=Light Physical Activity; MVPA=Moderate-to-Vigorous Physical Activity; VPA=Vigorous Physical Activity.

PROMIS Questionnaire. The overall score of each PROMIS questionnaire was converted in the Scale Score and the results were classified as 'moderate limitation', 'mild limitation' and 'normal function', based on the PROMIS cut scores on physical function metric (Figure 2) (18). "Normal function" was identified in 30% of the children (Scale Score \geq 50), whilst 44% reported "mild limitations" (Scale Score 40 to 48) and 26% reported "moderate limitations" (Scale Score 30 to 40) (Figure 3).

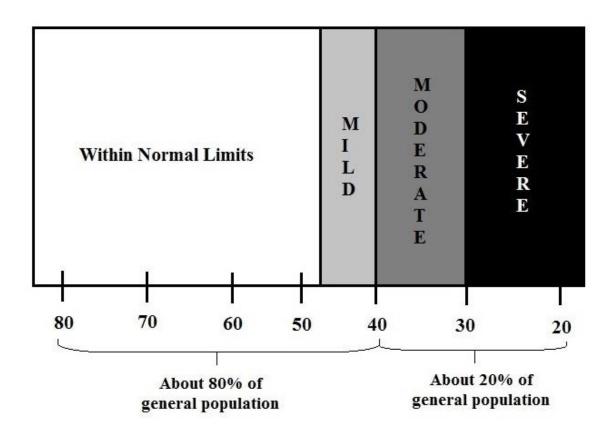


Figure 2: PROMIS cut scores on physical function metric (Cella et al., 2010).

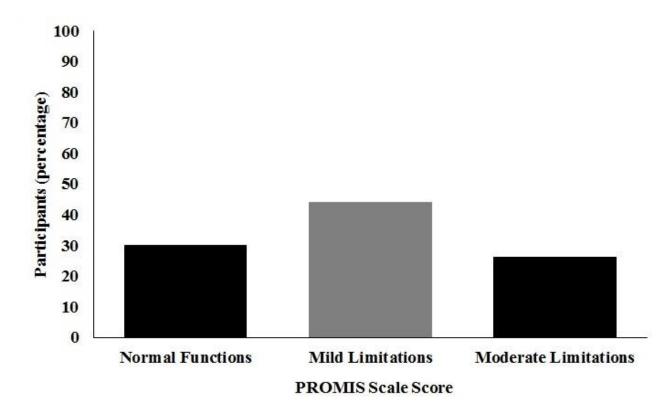


Figure 3: PROMIS scale score results.

Correlation between PA and PROMIS Scale Score. There was evidence of a moderate to strong correlation between daily MVPA and the overall Scale Score of the PROMIS questionnaire (r=0.67, n=28, p=0.01) (Figure 4a). A moderate correlation was evident between the daily LPA and the overall Scale Score (r=0.46, n=28, p=0.01) (Figure 4a). A moderate correlation was evident between the daily VPA and the overall Scale Score (r=0.46, n=28, p=0.01) (Figure 4a). A moderate correlation was evident between the daily VPA and the overall Scale Score (r=0.54, n=28, p=0.01) (Figure 4a). No correlation was detected between the average daily ST and the overall Scale Score (r=-0.28, n=28, p=0.15) (Figure 4b).

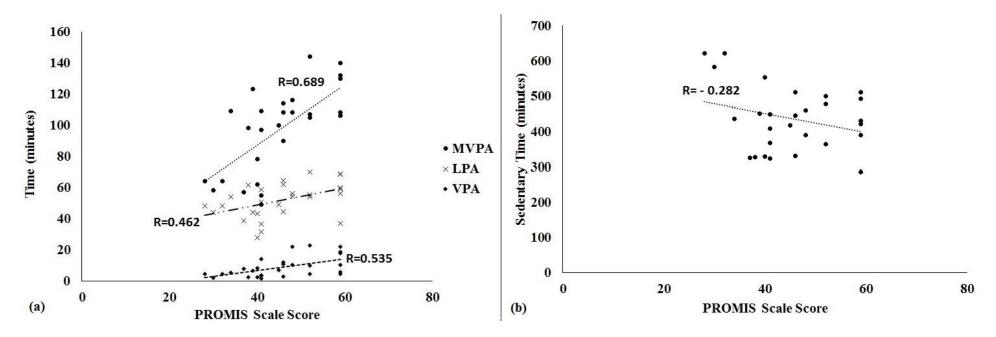


Figure 4: (a) Correlation between PA time and Scale Score. (b) Correlation between ST and Scale Score. LPA=Light Physical Activity; MVPA=Moderate-to-Vigorous Physical Activity; VPA=Vigorous Physical Activity.

Differences in PA level among PROMIS Scale Score Groups. There was a significant difference in Sedentary Time (p=0.002); MVPA (p=0.002) and VPA (p=0.004) among Scale Score sub-groups. ST was lower in the normal function group (*mean 407.7±9.0 minutes*) compared to the moderate (*mean difference -54.3±13.8 minutes*, p=0.00) and the mild (*mean difference -36.6±12.3 minutes*, p=0.02) groups; but no statistically significant differences were evident between the moderate and the mild limitation group (*mean difference 17.6±13.0 minutes*, p=0.58). MVPA was higher in the normal function group (*mean 126.0±7.8 minutes*) compared to the mild (*mean difference 33.4±10.6 minutes*, p=0.01) and moderate (*mean difference 48.6±12.0 minutes*, p=0.00) groups; but no statistically significant differences were evident between the moderate (*mean difference 15.2±11.3 minutes*, p=0.00) groups; but no statistically significant differences were evident between the mild and the moderate group (*mean difference 15.2±11.3 minutes*, p=0.57). Additionally, VPA was higher in the normal function group (*mean 14.6±1.9 minutes*) compared to the moderate limitation group (*mean difference 11.0±2.9 minutes*, p=0.00), but not statistically higher than the mild limitation group (*mean difference 6.5±2.6 minutes*, p=0.64). There was no difference in LPA between groups (p>0.05).

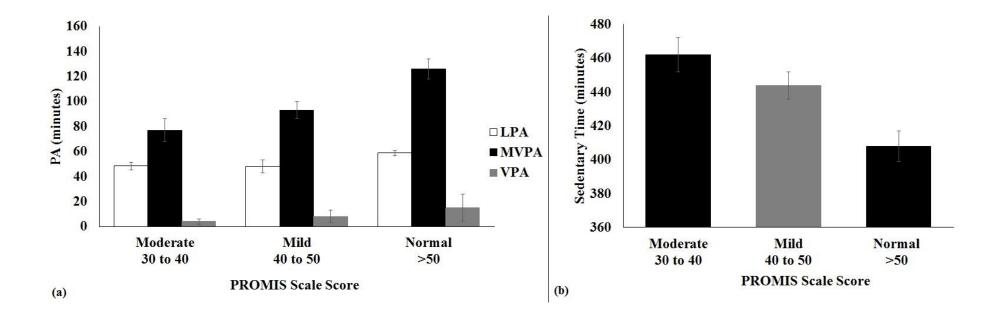


Figure 5: (a) Children's average daily MVPA divided by PROMIS Scale Score. (b) Children's average daily ST divided by PROMIS Scale Score. PA=Physical Activity; LPA =Light Physical Activity; MVPA=Moderate-to-Vigorous Physical Activity; VPA=Vigorous Physical Activity.

DISCUSSION

The aim of the study was to objectively assess physical activity level in children with hip disease and correlate physical activity with the PROMIS lower limb mobility score. The physical activity level of these children was generally higher than the average daily recommended by national guidelines for moderate to vigorous physical activity (19). LPA, MVPA and VPA were positively correlated to PROMIS Scale Score, children with normal mobility had higher physical activity levels and lower sedentary time.

This is the first study to investigate the correlation between hip mobility and physical activity level of children affected by hip disease. The results suggest that a lower PROMIS scale score (indicating impairment in lower limbs function) corresponded to a lower level of daily moderate and vigorous PA amongst children with hip disease. ST; average daily MVPA; and VPA were all correlated with the mobility score, whereby children with lower functions were the least active and children with normal functions were the most active. This study provides experimental data to provide external validity supporting the notion that the PROMIS Mobility Score can provide a general overview on the physical activity level of these children.

The PROMIS data from the current study population suggested that 70% of children had some degree of physical limitation and 30% had normal function. Understanding the limitations induced by hip disease is important as they have negative impact on the general quality of life of these children (3, 9), but whether this translates into reducing their ability to perform daily physical activity such as walking is unknown. Accelerometers were employed in the current study to measure physical activity level in children with hip disease and demonstrated that the average daily MVPA of these children is of 101 ± 37 minutes. The daily MVPA recommendation for children is at least of 60 minutes per day (19) and the obtained data suggest that these children were exceeding the recommended PA level. Previous observational studies, employing accelerometers to measure PA in healthy children in England (20) and in the North West of England (11) suggest recommended daily MVPA is reached in 22% of the English children and in 27% children from the North West. Therefore, the children affected by hip disease in this study seem to be more active than the general population, having an average daily MVPA 68% higher than the minimum suggested by the guidelines. Only one previous study has investigated the impact of childhood hip disease on physical activity, in children with Perthes' Disease in Sweden (9). This study used the International Physical Activity Questionnaire, and observed higher physical activity levels in children with Perthes' Disease compared to the national average. The authors linked the higher physical activity level in that study to hyperactivity related to attention deficit hyperactivity disorder (ADHD), a known association with Perthes' Disease (9, 21). It is perhaps also plausible that the high level of physical activity observed in participants in our study was an observer bias, whereby behaviour is modified in response to being observed.

Sedentary time in children participating in this study was higher than the ST reported among healthy English children (22). Children with moderate limitations reported a ST of 1.8 times higher than the average, while children with mild to normal functions reported a ST 1.6 times higher than the average. This may be due to the limitations induced by the hip disease (such as pain) which may increase the time these children spend sitting or lying, particularly during exacerbations (3). Increased sedentary time in children has been observed to be a risk factor for cardio-metabolic diseases, obesity and other comorbidities (23, 24). Sedentary-induced risk factors, such as obesity, may also play an additional role in the worsening of the symptoms and manifestation of the hip condition (25-27), and negatively influence the long-term cardiovascular risk of these children as has been previously shown amongst those with Perthes' Disease (28).

The choice of accelerometer, and proposed cut-points to define activity was a significant consideration, as it is well documented some researchers may overestimate or underestimate physical activity levels (16). Debate is ongoing on the best cut-points to adopt for accelerometers studies, however the cut-points defined by Freedson for ActiGraph accelerometers were employed in this study (17). These have shown significantly better classification accuracy for MVPA compared to cut-points defined by other authors (16).

The strength of our study is being able to correlate activity in the 'normal life' of a child over a 7-day period with patient reported outcomes, using an accelerometer that is extensively used in public health research. However, data were collected in a single UK centre from a relatively small sample of children, with a diverse range of hip disease. Furthermore, activity was almost certainly modified by observation (Hawthorne effect); though this is likely to be the case across the spectrum of hip disease therefore representing a uniform misclassification bias. We were unable to correlate radiographic severity of disease with physical activity, in part owing to the range of diseases included, and in part owing to the numbers of patients with each disease, though this is an area for which we plan future research.

CONCLUSION

In conclusion, this study offers strong experimental evidence to externally validate the paediatric PROMIS lower limbs mobility tool. PROMIS mobility score is correlated with physical activity levels of children with hip disease and provides a good general overview of everyday physical activity. Whilst, the general mobility of the study population was low, with most reporting moderate to severe limitations, daily PA levels were generally higher than daily minimum MVPA recommended for children.

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Appendix A. Patients' characteristics.

Patient ID	Gender	Age	Condition	Surgery	Operated	PROMIS	PROMIS
					Hip	Raw Score	Scale Score
#001	Boy	9	Perthes' Disease	Yes	Right	37	46
#002	Girl	8	DDH	Yes	Left	40	59
#003	Boy	10	Perthes' Disease	Yes	Left	34	41
#004	Girl	15	SCFE	Yes	Both	22	30
#005	Boy	14	Perthes' Disease	Yes	Left	30	37
#006	Girl	14	Perthes' Disease	Yes	Right	36	45
#007	Boy	11	Perthes' Disease	Yes	Left	37	46
#008	Girl	15	DDH	Yes	Right	32	39
#009	Girl	8	Perthes' Disease	No	Right	31	38
#010	Boy	12	DDH	Yes	Right	33	40
#011	Girl	10	Perthes' Disease	Yes	Right	37	46
#012	Girl	17	DDH with AVN	Yes	Right	24	32

#013	Girl	11	DDH	Yes	Right	40	59
#014	Girl	14	DDH	Yes	Left	33	40
#015	Boy	15	Perthes' Disease	Yes	Right	40	59
#016	Girl	12	DDH	Yes	Left	38	48
#017	Boy	8	Perthes' Disease	Yes	Right	39	52
#018	Girl	12	SCFE	Yes	Right	40	59
#019	Girl	14	DDH	Yes	Right	34	41
#020	Girl	11	DDH	Yes	Right	31	38
#021	Girl	8	Perthes' Disease	Yes	Right	27	34
#022	Girl	11	Perthes' Disease	No	N/A	40	59
#023	Girl	10	Perthes' Disease	Yes	Left	22	30
#024	Boy	10	Perthes' Disease	Yes	Right	40	59
#025	Boy	9	Perthes' Disease	No	N/A	39	52
#026	Boy	14	Perthes' Disease	No	N/A	39	52
#027	Boy	15	SCFE	Yes	Both	38	48
#028	Boy	13	Perthes' Disease	Yes	Right	40	59

DDH=Development Dysplasia of the Hip; SCFE Slipped Femoral Capital Epiphysis.