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Paget's disease of bone in two medieval skeletons from Poulton Chapel, Cheshire, UK

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Abstract

Paget's disease of bone (PDB) is a chronic, metabolic disease disrupting normal bone turnover and is reported as one of the most common bone diseases after osteoporosis. PDB is characterised by excessive bone remodelling resulting in bone enlargement, fragility, deformity and additional complications. Typically, PDB affects one or a few bones of the axial skeleton and is commonly recorded in older individuals (over 55 years of age) affecting more males than females. Although PDB has been reported worldwide, there is a high concentration of reported cases in the UK, with a regional *hotspot* in the northwest of England. This study reviews an adult male (SK463) and female (SK750) with skeletal lesions of PDB from Poulton Chapel, Cheshire. Full macroscopic and radiographic analysis has identified the skeletal distribution of PDB, with up to 75% of both skeletons affected. SK463 presents noticeable anterior bowing to both tibiae, likely the result of PDB. AMS radiocarbon dating and stable isotope analysis performed on teeth samples confirmed that both individuals' dates were medieval, had a mixed/varied diet and were local to the northwest of England. This research adds to the emerging paleopathological literature on PDB, while providing additional support for the identification of a geographical hotspot observed in contemporary populations.

KEYWORDS

medieval, metabolic disease, northwest hotspot, osteitis deformans, osteoarchaeology, Paget's disease, radiology, stable isotopes

1 | INTRODUCTION

Paget's disease of bone (PDB) was first formally described by an English physician, Sir James Paget in 1876. Paget's monograph was published by the Royal Medical Surgical Society identifying a chronic condition of bone named as *osteitis deformans* (Paget, 1876). Here, observations of patients were made over several years, describing the distinct clinical features of PDB including bone pain, enlarging skull and bone deformity (Paget, 1876). Today, clinicians have an improved understanding of PDB with effective treatments to manage this disorder; however, it remains to be an enigmatic disease. At a cellular level, the pathogenesis of PDB involves three phases. Firstly, it begins with an osteolytic phase where bone turnover is markedly increased. This increased resorption occurs through the action of abnormal multinucleated osteoclasts giving the characteristic blade-of-grass appearance on X-ray (Tuck, Layfield, Walker, Mekkayil, & Francis, 2017). The second mixed phase of osteolytic and osteoblastic activity is dominated by rapid increases in osteoblast activity giving rise to abnormal bone

with irregular deposition of collagen fibres. Finally, in the last phase known as the sclerotic phase, bone formation again predominates. This results with newly formed bone being disorganized (woven) and mechanically weaker, overall resulting in skeletal lesions with abnormal bone architecture (Shaw et al., 2019). Typically, PDB affects one (monostotic) or more (polyostotic) skeletal elements, with research supporting a higher prevalence of the monostotic type of this disease (Winn, Lalam, & Cassar-Pullicino, 2016). Although PDB can affect any bone in the human skeleton, it is most frequently reported to affect the axial skeleton (e.g., skull, lumbar spine, pelvis, sacrum, femur and tibia; see Cundy, 2018; Ralston et al., 2019), and despite a steady decrease in secular changes (Abdulla, Naqvi, Shamshuddin, Bukhari, & Proctor, 2018), it is one of the most common metabolic bone disorders after osteoporosis (Vallet & Ralston, 2016).

Macroscopically, bones affected with PDB appear more porous and are heavier than normal. This is due to the mixed osteolytic–osteoblastic phase that leads to the formation of new woven bone. Essentially, cranial bones increase in thickness, whereas long bone diaphyses enlarge, thicken and may become bowed (Kesterke & Judd, 2018). When viewed in cross-section, bones are often described as *coral* or *pumice-like* in appearance (Ortner, 2003). Radiographically, the progressive changes of PDB can be observed more precisely. The early lesions are predominately lytic and osteoporotic and are often limited to either the endosteum or central layers of the cortex (Valenzuluela & Pietschmann, 2017). The later mixed osteolytic–osteoblastic phase leads to the formation of new woven bone, essentially the thickening of the cortex by endosteal and periosteal bone deposition, which is often observed progressing along the shaft of long bones (Tuck et al., 2017). Key radiological features include increased bone size, non-uniform increased density and cortical thickening (Cortis, Micallef, & Mizzi, 2011). Key features of PDB can include the *cotton wool appearance* on the cranium (Cortis et al., 2011), *picture frame vertebrae* (Brickley & Ives, 2010) and the *lytic wedge* (Cundy, 2018). However, not all individuals present these key features, and it must be noted that different phases of PDB can coexist in one bone at any one time (Winn et al., 2016). For example, a recent study by Shaw et al. (2019) reports 18 medieval skeletons from Norton Priory, UK, with different stages of PDB. Yet only some of these individuals presented one or more of these key features of PDB alongside the observed osteolytic and osteoblastic phases of PDB (Shaw et al., 2019).

The diagnosis of contemporary PDB is rare before the age of 50 (Valenzuluela & Pietschmann, 2017), with the prevalence of the disease increasing with increased age (Ralston et al., 2019; Vallet & Ralston, 2016). PDB can affect both men and women, although it is slightly more common in males (Van Staa et al., 2002). Today, the aetiology of PDB is not completely understood, with theories exploring environmental factors with limited success (Lever, 2002; Reddy, 2006; Siris, 1996), and an autosomal dominant pattern of inheritance may be observed (Ralston et al., 2019; Shaw et al., 2019). The most notable gene is SQSMT1 (Hocking et al., 2002), which encodes the p62 protein involved (Ralston et al., 2019). Mutations in SQSMT1 have been identified in 40–50% of familial cases and in 5–10% of patients who do not have a family history (Hocking et al., 2004; Ralston et al., 2019). Most mutations impair the binding of the p62 protein leading to the activation of increased osteoclast activity (Jin et al., 2008). These genetic factors are reflected in the geographic distribution of PDB. Since the 1970s, radiographic survey of PDB has been the standard method of assessing the prevalence of the disease. Detailed surveys have taken place in the United Kingdom, Europe, North America, Australia, New Zealand and South Africa (Cundy, 2018). The UK has the highest prevalence of PDB in the world with a regional hotspot identified in the northwest of England (Barker, Chamberlain, Guyer, & Gardiner, 1980; Barker, Clough, Guyer, & Gardner, 1977; Cooper, Harvey, Dennison, & Staa, 2006; Cundy, 2018; Van Staa et al., 2002). It is also common in European countries such as France, Spain and Italy (Detheridge, Guyer, & Barker, 1982; Gennari, Merlotti, Martini, & Nuti, 2006; Lopez-Abente et al., 2003) and in people of European descent who have emigrated to other regions of the world such as Australia, USA and Canada (Corral-Gudino, Borao-Cengotita-Bengoa, Del Pino-Montes, & Ralston, 2013; Ralston et al., 2019). PDB is considered rare in China, Japan and Africa (Resnick, 1988).

Interestingly, this geographic distribution is reflected in the palaeopathological record. An extensive review by Mays (2010) provides a collated report of the paleopathology literature, supporting the UK concentration of this disease with the highest number of cases reported as medieval (AD 1066–1538). Although several archaeological cases of PDB have been reported across the UK (e.g., Aaron, Rogers, & Kanis, 1992; Farwell et al., 1993; Price, 1975; Rogers, Jeffrey, & Watt, 2002; Shaw et al., 2019; Stirland, 1991; Wells & Woodhouse, 1975), the largest number of cases reported are from London (Mays, 2010), which simply reflects the high concentration of excavations in this area in comparison with other areas of the UK. Nonetheless, the northwest hotspot is a noted geographic feature of contemporary PDB (Barker et al., 1977; 1980), and archaeological investigations in this area are scarce. However, at Norton Priory Cheshire, 18 skeletons have been reported with PDB (Shaw et al., 2019) affecting 16% of the adult sample (n = 114). A recent study of these remains have identified that the skeletal involvement of PDB is more extensive than previously anticipated, with up to 75% of their skeletons affected (Shaw et al., 2019). Additionally, radiocarbon dating and their stable isotopic signatures have acknowledged that these individuals are indeed local to the northwest of England, with the prevalence of the disease covering over 400 years of the site's history (Shaw et al., 2019).

This article presents two new cases of PDB from Poulton Chapel, Cheshire, an archaeological site in the northwest of England. Both individuals were subjected to macroscopic and radiological review identifying the full skeletal distribution of PDB in each individual. Additional investigations of these individuals included AMS radiocarbon dating and stable isotopic analysis on teeth ($\delta^{13}C$, $\delta^{15}N$, $^{87}Sr/^{86}Sr$ and $\delta^{18}O$). The results presented here offer a detailed review of the skeletal distribution of PDB with first-hand information about the period and geographical mobility of these skeletons while supporting the theory of a northwest hotspot for this disease.

2 | MATERIALS AND METHODS

The skeletons presented in this study are from the Poulton Chapel Collection, a continually growing collection of archaeological human skeletal remains associated with the ongoing excavations of the Poulton Research Project. The Poulton Research Project is a multiperiod site located in the rural village of Poulton in west Cheshire (Figure 1). The current focus of this site is the study of the remains of the medieval Chapel and its surrounding burial ground. Because excavations began since 1995, over 900 articulated human skeletons have been excavated along with large quantities of disarticulated bones. This collection is currently housed at Liverpool John Moores University, UK. Seven hundred and twenty-six skeletons have been subjected to a detailed osteological review, and the demographics of the sample are typical of a medieval rural cemetery with a total of 190 males, 187 females, 38 adults of undetermined sex and finally 311 non-adults (Burrell, 2018). During the demographic analysis, numerous skeletal pathologies were recorded within this collection. Two individuals (SK463 and SK750) were discovered with widespread skeletal lesions typical of PDB, along with additional pathological skeletal conditions (e.g., secondary osteoarthritis and bone deformity).

2.1 | Osteological analysis

Both skeletons included in this study are adults (over 18 years of age). In this study, assessment of age is achieved by observing degenerative changes of the pubic symphysis (Brooks & Suchey, 1990) and the auricular surface (Lovejoy, Meindl, Pryzbeck, & Mensforth, 1985). Sex is assessed through the examination of the skull (the nuchal crest, the mastoid processes, the supraorbital margin, the supraorbital ridge and mental eminence [Buikstra & Ubelaker, 1994]) and the pelvis (the greater sciatic notch [Walker, 2005], the subpubic angle [Rogers & Saunders, 1993], the ventral arc, the subpubic concavity and the ischiopubic ramus ridge [Phenice, 1969]). Additionally, maximum diameters of the femoral head, humerus and radius were studied (Stewart, 1979). The authors note that skeletal elements affected with PDB could affect the final assessment of age-at-death and sex in these individuals. This will be kept under consideration in the analysis. Alongside this, stature assessment was derived from regression formulas using different long bone lengths (Trotter & Gleser, 1958). After biological profiling, SK463 and SK750 were examined visually for macroscopic changes of PDB. Lesions included increased size, thickness or vascularity. Full radiographic reviews (anteroposterior and mediolaterally) were obtained using the Faxitron series X-ray system at the School of Natural Sciences and Psychology at Liverpool John Moores University, UK. The radiographs enable the observation of the progressive changes of PDB to be viewed more precisely within the human skeleton.

2.2 | Accelerator mass spectrometer (AMS) radiocarbon dating and stable isotope analysis

A single tooth was extracted from each skeleton and prepared for radiocarbon dating and isotopic analysis. The tooth roots were submitted to Beta Analytic, Miami, USA, for Accelerator Mass Spectrometer (AMS) radiocarbon dating, to establish their age, and stable isotopic analysis using carbon ($\delta^{13}C$) and nitrogen ($\delta^{15}N$), for paleodiet reconstruction. The crowns of the teeth were submitted to the Natural Environment Research Council Isotope Laboratory in Nottingham, UK, to study their enamel composition to obtain their strontium

FIGURE 1 Map of the UK with the county of Cheshire and approximate location of Poulton [Colour figure can be viewed at wileyonlinelibrary.com]

($^{87}Sr/^{86}Sr$) and oxygen ($\delta^{18}O$) isotopic values. The molars selected for analysis undergo crown development during 2.5 to 8.5 years of age (AlQahtani, Hector, & Liversidge, 2010), which allowed the investigation of geographic origin of the individuals during childhood. The selected teeth were well preserved and devoid of caries or other signs of decay.

3 | RESULTS

Skeletons SK463 and SK750 are in a good state of preservation with little disturbance to the burials in situ resulting in almost complete skeletons. The results of the biological profiles are presented in Table 1. Macroscopic analysis identified distinct lesions of PDB across ~60% of their skeletons (Figure 2a and Table 2). These elements present distinct periosteal changes and bony expansion and are heavy to hold. Although the pelvis and cranium are affected in both cases, the authors are confident with the age-at-death and sex estimations for both skeletons. Nonetheless, additional observations were conducted by professionals in our field and their conclusions reflected our results, confirming the results of the biological profiles presented in Table 1. In regard to macroscopic changes of PDB, notable and comparable differences in the size and thickness of the left and right humeri are observed in SK750 (Figure 3a). Figure 3b presents a close-up of the distinct porous lesions indicative of PDB. For SK463, post-mortem breaks were apparent across the skeleton, which provided an opportunity to observe the internal architectural skeletal changes of PDB. Figure 4 shows the marked internal changes of PDB to the left tibia. Additionally, both tibiae show distinct bony changes of PDB with marked trabecular thickening, bony expansion and anterior bowing.

Radiographic analysis provided a more detailed review of the activity, distribution and progression of this disease within each skeleton. Here, osteolytic lesions were evident for most elements where

TABLE 1 Biological profile of SK463 and SK750

Skeleton number	Sex	Age-at-death	Stature estimation	Burial location
SK463	Male	50–59 years	166.9 ± 3.0 cm	Northwest corner outside of Chapel
SK750	Female	45+ years	154.8 ± 3.5 cm	Southern side outside of Chapel

TABLE 2 Macroscopic assessment of elements affected with PDB for SK463 and SK750 with pathology

Skeleton number	Elements affected by PDB	Pathology and associated conditions
SK463	Cranium, clavicle (B), scapula (B), spine, humerus (B), ribs (B), pelvis (B, S), femur (B), tibia (B), fibula (B)	Anterior bowing of tibia (B)
SK750	Cranium, clavicle (R), humerus (B), ribs (B), pelvis (B, S), femur (B), tibia (B), fibula (R), tarsals (B)	Osteoarthritis with eburnation, to humeroradial joints (B), Schmorl's nodes

Note. L = left, R = right, B = both L and R elements, and S = sacrum.

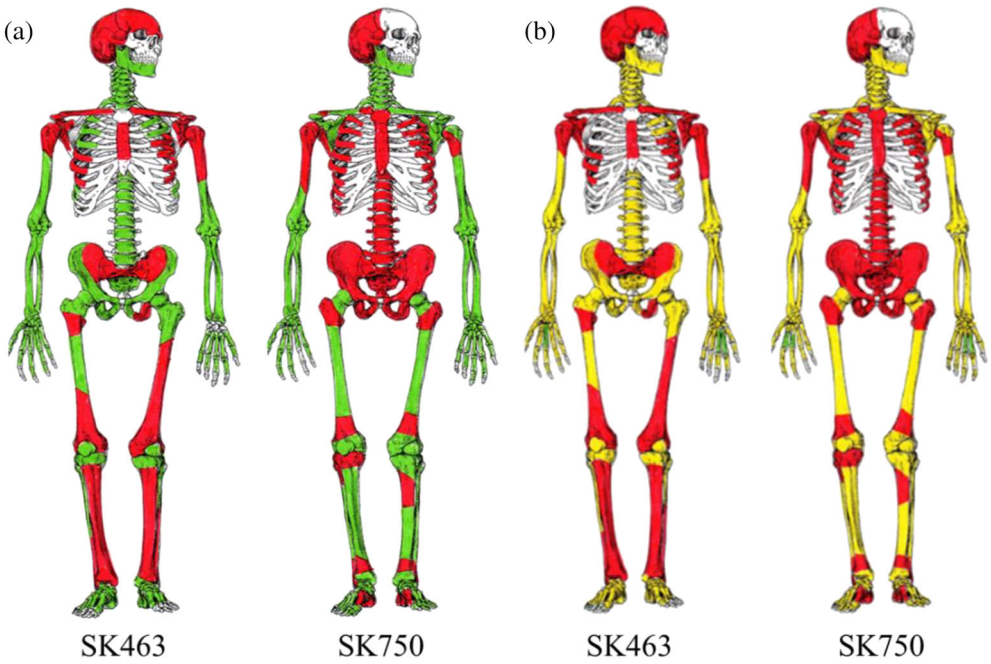

FIGURE 2 (a) Macroscopic distribution of bones affected with PDB (highlighted red) for each skeleton compared with unaffected bones (highlighted green). (b) Distribution of bones affected with PDB: macroscopic changes (highlighted red) and internal lytic changes identified during radiographic analysis (highlighted yellow) for each skeleton compared with unaffected bones (highlighted green) [Colour figure can be viewed at wileyonlinelibrary.com]

FIGURE 3 (a) Comparison between the left and right humeri of SK750. Note the bony expansion and bone enlargement to the right humerus. (b) Close up of the macroscopic changes of PDB to the proximal right humerus [Colour figure can be viewed at wileyonlinelibrary.com]

macroscopic lesions were not documented, identifying the earlier stages of PDB within these skeletons. The results identify that up to 75% of these skeletons are affected by PDB (Figure 2b). As an example, there was significant expansion and sclerosis of the right humerus of SK750 with almost complete obliteration of the medulla cavity when compared with the left humerus (see Figure 5). The coarse trabecular pattern is progressively evident towards the distal portion of the bone, which was not observed during macroscopic examination. For SK463, bony expansion and coarse trabeculation of the left femora and its corresponding radiograph is seen in Figure 6. The anterior bowing of the tibia observed in SK463 is apparent in this radiograph (see Figure 7). Here, distinct cortical thickening, expansion and coarsening of the trabecular pattern is noticeable.

3.1 | Accelerator mass spectrometer (AMS) radiocarbon dating and stable isotopic analysis

The good preservation of the dental samples allowed absolute AMS radiocarbon dates to chronologically place SK463 and SK750 within the Poulton Chapel timeline (Burrell, 2018). Both individuals were found to have lived between the late 13th century and late 14th century (Table 3). Dietary isotopic analysis ($\delta^{13}C$ and $\delta^{15}N$) identified a mixed diet focussed on C_3 plants, animal and marine sources, along with the occasional intake of freshwater fish protein (Table 4). The strontium ($^{87}Sr/^{86}Sr$) and oxygen ($\delta^{18}O$) isotopic values held within the tooth enamel reflect the geographic point of origin and the potential mobility of these individuals. These results were extracted from the tooth enamel as this preserves the isotopic signature during tooth formation during childhood. The $^{87}Sr/^{86}Sr$ and $\delta^{18}O_{VSMOW}$ values obtained (Table 4) indicate that both skeletons are local to the area.

4 | DISCUSSION

This paper presents two new cases of PDB from Poulton Chapel, Cheshire, a valuable addition to the growing palaeopathological literature of PDB but also in support of the reported northwest hotspot of this disease. Currently, the population prevalence of PDB for this sample is 0.3% (n = 726) or 0.5% of the adult sample (n = 415); however, the prevalence is likely to increase as additional skeletons from this collection are subjected to further analysis. Interestingly, similar results have been reported in another geographically local medieval collection, Norton Priory. A report from 2005 (Boyleston & Ogden, 2005) presented only six skeletons from Norton with PDB (~5%). However, new thorough macroscopic and radiographic analysis identified that these six individuals are more highly affected than previously anticipated, with skeletal lesions of PDB reported to affect up to 75% of their skeleton (Shaw et al., 2019). Alongside this, molecular analysis

FIGURE 4 Distinct architectural changes of PDB to the internal structure of the left tibia of SK463 [Colour figure can be viewed at wileyonlinelibrary.com]

of the same six Norton skeletons, inclusive with an additional 12 skeletons with PDB from Norton Priory (n = 18) confirmed an increased prevalence of PDB (14% [n = 130] or 16% of the adult sample [n = 114]; Shaw et al., 2019) and research into the remaining collection is still ongoing. As proposed here for the Poulton Chapel sample, it is very likely that PDB is underrepresented at this moment in time and, with further research, a greater understanding of PDB from the northwest of England will continue to emerge.

The association of macroscopic and radiological analysis in skeletons SK463 and SK750 reveals without reasonable doubt that these individuals have PDB. The authors do acknowledge that histological, CT and Micro-CT imaging is a frequent addition to the diagnostics of PDB in archaeological remains (e.g., Kesterke & Judd, 2018; Wade, Holdsworth, & Garvin, 2011). Unfortunately, these methods have not been attempted on the Poulton skeletons due to curatorial restrictions at this time. Nonetheless, the pathogenesis of PDB can be identified in the human skeleton through radiological imaging alone (Cortis et al., 2011), and as morphological features of PDB are distinct in the later phases of PDB (e.g., bone thickening and enlargement), such skeletal lesions have been identified in skeletons SK463 and SK750. Comparably, similar lesions have been reported at other archaeological sites (e.g., Aaron et al., 1992; Shaw et al., 2019; Stirland, 1991; Wade et al., 2011).

4.1 | Differential diagnosis of PDB

The authors have considered a differential diagnosis of PDB and some rare osteoclastic and osteoblastic forms of bone disorders that can be confused with PDB (e.g., hyperthyroidism, hyperparathyroidism, hereditary hyperphosphatasis, familial expansile osteolysis [FEO] and polyostotic fibrous dysplasia). The most noteworthy is hereditary hyperphosphatasis, sometimes referred to as *Juvenile Paget's disease*. Radiological changes of this metabolic disease mimic those of PDB with expanded osteoporotic long bones, coarse trabeculation and widened bones of the skull (Tüysüz, Mercimek, Üngür, & Deniz, 1999). Juvenile Paget's disease typically affects the whole skeleton as seen with the Poulton skeletons. However, this disorder, which begins in infancy, can lead to short stature, an enlarged head and the premature loss of teeth. Another disorder, FEO, shares many similarities with PDB with marked resorption and thickened trabecular bone. However, the radiographic features and natural history of this disease differ from PDB with multifocal changes seen within the same affected bone (Osterberg et al., 1988). Alongside this, FEO manifests during early adulthood (18 to 44 years of age), and the lesions observed are limited to the long bones, with no involvement of the skull or pelvis, which contradicts the typical skeletal distribution of PDB (Guyer, 1981). Similarly, polyostotic fibrous dysplasia can present changes with marked bone enlargement but does not present the thickened cortex observed in PDB (Kanis, 1992). Finally, hyperthyroidism and hyperparathyroidism are thyroid stimulating hormones that can affect bone metabolism resulting in the activation of osteoclastic bone resorption. The most common skeletal manifestation of hyperthyroidism is osteoporosis with radiographic findings often reported in the hands and feet alongside soft tissue swelling in patients. Although commonly reported in patients with PDB, it does involve bones of the axial skeleton as seen with PDB (Guyer, 1981). On the other hand, hyperparathyroidism also presents bone resorption in the hands but is described as lacelike in radiographic appearance. In the later stages, resorption can appear scalloped (Murphey, Sartoris, Quale, Pathria, & Martin, 1993). In the cranium, there is a decreased differentiation between the inner and outer tables of the skull with multiple circular lesions (Kar et al., 2001). In the long bones, the cortical resorption is often described as *smudged tunnelling* and is a prominent feature of hyperparathyroidism alongside brown tumours (Brown, Genant, Hattner, Orloff, & Potter, 1977). Hyperparathyroidism presents skeletal distributions similar to PDB, but the radiographic appearance differs. This review focuses on the clinical features of PDB where histology is rarely required to make a diagnosis as this can be established through radiography alone. To conclude, the authors can confirm the characteristics observed in the two Poulton skeletons are that of PDB.

4.2 | Comparison with the palaeopathological record

Exploration of the palaeopathological literature suggests that PDB has been suspected from as early as the Neolithic period (Fisher, 1935; Pales, 1929). Since then, several skeletons have been identified with

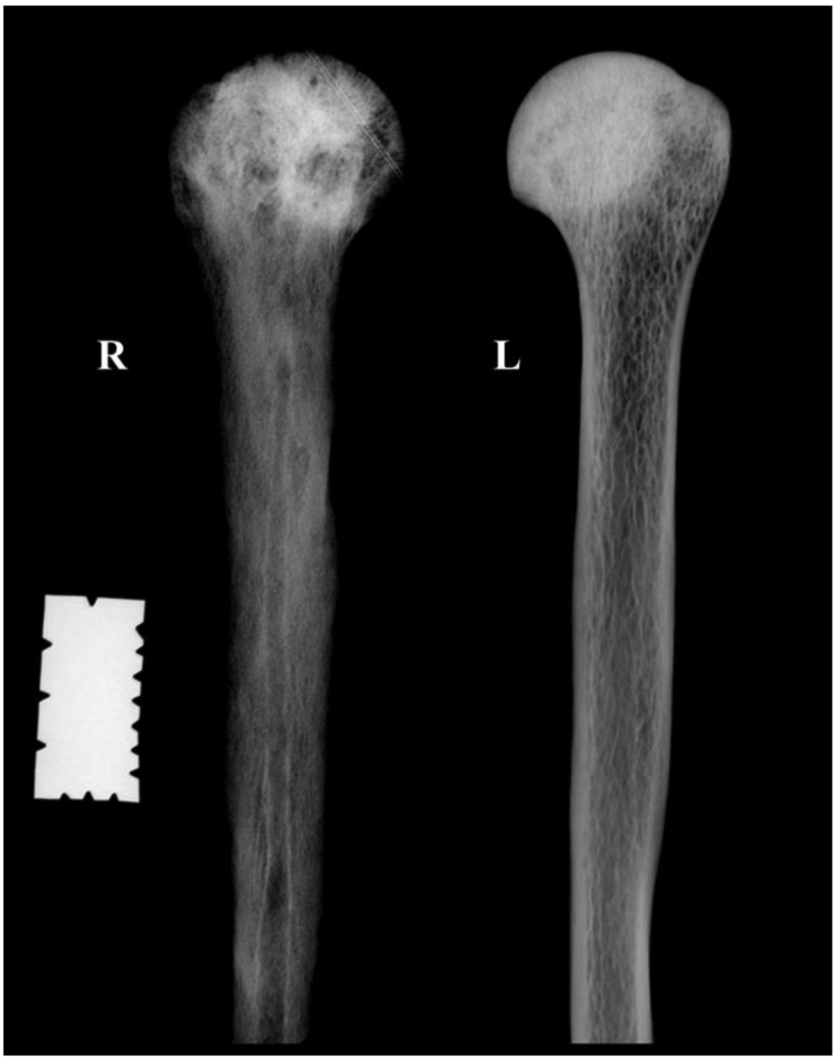

FIGURE 5 Radiograph of the left and right humeri of SK750. Significant expansion and sclerosis of the right humerus with complete obliteration of the medulla cavity is noted

PDB in the palaeopathological record with macroscopic and radiographic imaging alone (e.g., Farwell et al., 1993; Molleson & Cox, 1993; Price, 1975; Rogers et al., 2002; Stirland, 1991; Wells & Woodhouse, 1975). However, only the skeletal elements with identifiable macroscopic lesions of PDB are subjected to radiological review, not the entire skeleton. With the results presented in this study, SK463 and SK750 show macroscopic lesions across ~60% of their skeleton. Alongside this, full radiological review identified earlier lesions of PDB identifying that up to 75% of their skeleton is affected with PDB. Similar results have been identified in recent studies (Shaw et al., 2019) and, with advisory caution from earlier research (Mays & Turner-Walker, 1999), care must be taken in the interpretation of the skeletal distribution of PDB in the reported samples as valuable information could be missing.

This study highlights the importance of radiological imaging alongside macroscopic review in the diagnosis of PDB. The high number of skeletal elements affected in SK463 and SK750 suggest a progressive polyostotic type of this disease, which is unusual in comparison with contemporary PDB (Winn et al., 2016). A recent study from Norton Priory provided strong evidence for performing full radiographic analyses with the skeletons from Norton displaying lesions of PDB affecting up to 75% of their skeletons but with an early age onset (Shaw et al., 2019). Additional molecular work conducted on these samples identified an abnormal SQSTM1 (p62) protein in the archaeological remains suggesting that the PDB observed at medieval Norton Priory is an ancient precursor of contemporary PDB (Shaw et al., 2019). Mutations of the SQSTM1 have been identified in individuals who share a family history, and this is reflected in the burial spatial distribution of the skeletons from Norton Priory. Within the priory, specific areas of burial are reserved for family members. For example, the north-east chapel was funded by the Dutton family, important benefactors of the Priory during the 12th to 13th centuries (Brown & Howard-Davis, 2008). Interestingly, this is where majority of the affected skeletons from Norton with PDB are buried (Burrell, 2018; Shaw et al., 2019).

Due to the extensive skeletal distribution of PDB in the Poulton skeletons (Figure 2) and the geographic location of this site to Norton Priory (north west area), it is possible that SK463 and SK750 could also be presenting an ancient precursor of the contemporary PDB protein. Based on historical records and the structure of the current landscape, Poulton is a rural farming community located in a small nucleated village. Here, several small holdings gather around a green

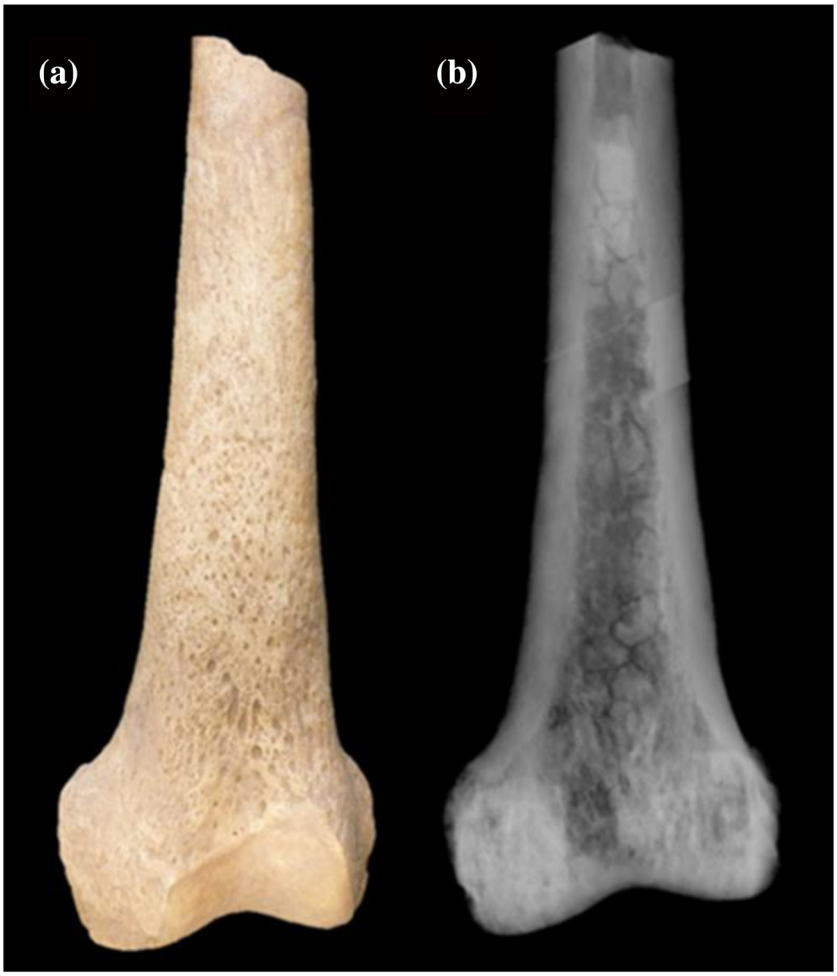

FIGURE 6 (a) Left femur of SK463 showing macroscopic changes of PDB to the distal femur and (b) corresponding radiograph [Colour figure can be viewed at wileyonlinelibrary.com]

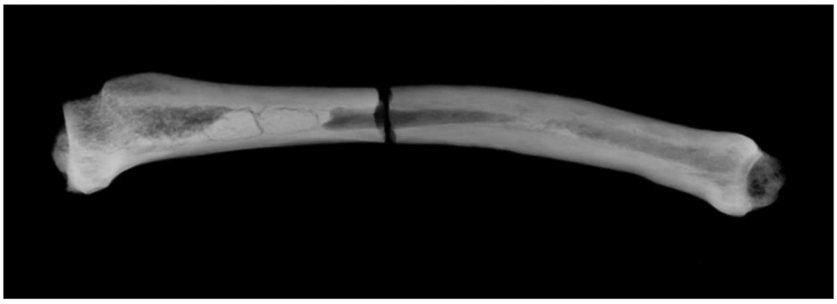

FIGURE 7 SK463 Radiograph of the right tibia showing marked anterior bowing. No fissure fracture is evident

TABLE 3 AMS radiocarbon results attained from the dental collagen of SK463 and SK750

Skeleton number	Tooth selected	Lab number	Radiocarbon age BP	AMS radiocarbon date 2 sigma calibration
SK463	First mandibular premolar	BETA-425289	680 ± 30	AD 1275–1310 & AD 1350–1390
SK750	Second mandibular molar	BETA-425290	640 ± 30	AD 1285–1330 & AD 1340–1395

TABLE 4 Stable isotope results attained from SK463 and SK750

Skeleton number	C $\delta^{13}C$	N $\delta^{15}N$	Sr $^{87}Sr/^{86}Sr$	O $\delta^{18}0_{VSMOW}$
SK463	−20.0‰	11.6‰	0.71165	17.5
SK750	−20.1‰	11.8‰	0.71226	18.1

near a church or chapel suggesting a structured family-based community with approximately 30 villagers at any one time (Burrell, Canavan, Emery, & Ohman, 2018). More recent research applied genetic non-metric traits through burial spatial analysis as a proxy for identifying plausible familiar relationships within the burial ground (Burrell, 2018). Unfortunately, the results were limited, but this is likely due

to the long-term use of this cemetery with burials taking place over several centuries as opposed to the high frequency of unusual skeletal variants (e.g., vertebral anomalies; Burrell, 2018). Nonetheless, due to the seclusion of this site, it is likely that the individuals buried at Poulton are likely to be closely related genetically anyway. As suggested, it is possible that SK463 and SK750 could also be presenting an ancient precursor of the contemporary PDB protein, and this aspect of research is being considered for future analyses of the Poulton skeletons.

4.3 | Comparison with modern populations

Today, PDB affects both men and women over the age of 55 years (Valenzuluela & Pietschmann, 2017; Vallet & Ralston, 2016) with the prevalence of PDB increasing with increased age. Skeletons SK463 and SK750 are both considered as "older" adults (over 45 years of age; see Table 2), reflecting the modern aetiology of this disease. Contemporary PDB typically affects the axial skeleton with only one or two bones affected (Kanis, 1992; Winn et al., 2016). However, skeletons SK463 and SK750 exhibit PDB changes across ~75% of the skeleton, although the anatomical distribution of this disease does not differ from what is seen in modern patients with the axial skeleton affected in both cases (Figure 2a,b). SK463 and SK750 are polyostotic, with multiple sites extensively affected by this disease. This is very unusual in comparison with modern patients where only one or a few bones are affected (Winn et al., 2016). Nevertheless, similar extensive skeletal distribution of PDB has been observed in the medieval skeletons from Norton Priory, Cheshire (Shaw et al., 2019).

Alongside this, skeletal complications of PDB are often reported in contemporary populations. Complications can include osteoarthritis, deafness, fractures, deformity and sarcomata's change (e.g., Melton, Tiegs, Atkinson, & O'Fallon, 2000; Valenzuluela & Pietschmann, 2017; Van Staa et al., 2002; Wermers, Tiegs, Atkinson, Achenbach, & Melton, 2008). SK750 presented osteoarthritis, with eburnation, to both left and right humeroradial joints. However, only the proximal portions of both humeri exhibited advanced lesions of PDB with earlier osteolytic changes observed in the distal portions of the humeri and in both radii and ulnae (see Figure 2). The presence of osteoarthritis is likely the result of increased age and other extrinsic and intrinsic factors rather than secondary osteoarthritis to PDB. However, SK463 showed marked anterior bowing to both tibiae. In contemporary samples, there is a high incidence of fissure fractures with marked bowing deformity (Redden, Dixon, Vennart, & Hosking, 1981) with fissure fractures common on the anterior aspect of the tibia. However, both anteroposterior and mediolateral radiographs did not identify a fracture in either tibiae (see Figure 7). Deformation is typically reported to occur in the long bones but also to the cranium and facial bones (see Kanis, 1992). Deformity of the femur results in lateral bowing, which is sometimes referred to as the *shepherds hook*, whereas deformity of the tibiae results in anterior bowing. In extreme cases, patients reported with marked changes to both femora and tibiae are unable to walk due to crossed-over legs (Kanis, 1992). Fortunately for SK463, only the tibiae exhibit such deformity. Although this suggests this individual was able to walk, it does mean that this individual may have experienced notable bone pain and discomfort during movement and at rest.

Results of the dental samples have identified that both individuals are medieval (13th–14th century), corresponding with the chronology of Poulton Chapel. The dietary isotopic analysis ($\delta^{13}C$ and $\delta^{15}N$) identified a mixed diet (Table 4), and the strontium ($^{87}Sr/^{86}Sr$) and oxygen ($\delta^{18}O$) isotopic values indicate that both individuals were local to the area (Table 4). Comparison of the $^{87}Sr/^{86}Sr$ against the isotopic map published by Evans, Montgomery, Wildman, and Boulton (2010) indicates that our results are mainly associated with rocks (Triassic Sandstone and Carboniferous Limestone) found in the northwest, west and central part of the UK. Additionally, the mean value for $\delta^{18}O_{VSMOW}$ for Western UK is 18.2% +/− 3, due to high levels of rainfall (Evans, Chenery, & Montgomery, 2012). Overall, our results are consistent with a Western UK origin for the studied teeth samples. However, more recent work by Evans, Royse, and Chenery (2019) has led to the redevelopment of the isotopic biosphere map (Biosophere Isotope Domains GB, Version 1), which includes additional and new data from across the UK. Direct input of the $^{87}Sr/^{86}Sr$ and $\delta^{18}O$ values for SK463 suggests that this individual is local to the area, originating from central Wales, slightly west from Poulton. For SK750, the isotopic values suggest a likely origin from the western border of Wales. Together, the $^{87}Sr/^{86}Sr$ and $\delta^{18}O$ isotope results are broadly consistent with a local provenance, reflecting the high concentration of contemporary PDB in northwest England (Barker et al., 1977; 1980; Van Staa et al., 2002).

The authors would like to note that the current occurrence of PDB for this sample is 0.5% of the adult sample (n = 415). However, during demographic review of the whole skeletal collection, other individuals presented skeletal lesions typical of PDB and are now the subject of further study (Burrell, Gonzalez, Smith, Emery, & Irish, 2016). Essentially, the frequency of PDB within this collection is likely to increase in the future as the confirmation of this disease is obtained from these additional skeletons.

5 | CONCLUSIONS

The authors have identified two cases of PDB in the medieval Poulton Chapel Collection. Both skeletons have been subjected to a full macroscopic and radiological review. Characteristic lesions of PDB have been documented, with a population prevalence of 0.5% for the adult sample. Interestingly, these skeletons are polyostotic, presenting marked skeletal changes of PDB across 75% of their skeleton. This aetiology is somewhat different in comparison with contemporary PDB, where only one or a few bones are affected. It is possible, as reported in the skeletons from a neighbouring medieval site, Norton Priory, that the skeletons from Poulton Chapel present an atypical form of contemporary PDB, which has likely evolved over time. Molecular studies (ancient proteins and ancient DNA) are required to explore this hypothesis. Nonetheless, new radiocarbon dating (^{14}C AMS) and stable isotopic analysis ($\delta^{13}C$, $\delta^{15}N$, $^{87}Sr/^{86}Sr$ and $\delta^{18}O$)

have shown that PDB was prevalent during the 13th and 14th centuries at Poulton and that the individuals originated broadly from the northwest of Britain. The geographic origin of these individuals further supports the northwest hotspot of contemporary PDB. These analyses add to the emerging paleopathology literature on PDB while providing additional support for the geographical hotspot observed in contemporary populations in the area.

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AUTHOR CONTRIBUTIONS

All authors provided input to the intellectual content of this paper. Carla L. Burrell performed and interpreted macroscopic and radiographic analyses of skeletal samples and facilitated AMS radiocarbon and stable isotopic analyses. Silvia Gonzalez interpreted the results and the discussion of the radiocarbon and stable isotopic analysis. Michael M. Emery organized the analyses of the skeletal remains from the Poulton Chapel Collection and provided information on the history of the site. All authors agreed on the conclusions presented in this manuscript.

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DECLARATION OF INTERESTS

The authors declare no competing interests.

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