Two cases of Paget's Disease of Bone from Poulton Chapel, Cheshire, UK

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Paget's Disease of Bone (PDB)

Today, PDB is the second most common metabolic bone disorder after osteoporosis that is frequently reported in older individuals (>55 years) affecting both men and women[1]. Skeletal elements affected by PDB can result in bone enlargement, fragility and deformity. Typically, PDB affects one or a few bones of an individual and is most frequently reported to affect the axial skeleton (e.g. skull, spine, pelvis and femur)[1,2]. Nowadays, clinicians have effective treatments to manage this disease however, the etiology is not completely understood[1,2,3]. On the other hand, genetic factors of PDB are well defined with many individuals sharing a family history1. This is somewhat reflected in the geographic distribution of PDB with the highest prevalence in the UK and, a regional concentration to the northwest of England[2,4,5,6,7]. Archaeological evidence seems to support the UK concentratation[8] however, few cases have been reported from this region[5].

Materials and Methods

In this study, we investigate two skeletons (SK463 & SK750) from Poulton Chapel, Cheshire (Fig. 1). Both individuals have been subjected to full 'head to toe' macroscopic and radiological examination to identify the skeletal distribution of PDB. Additionally, a single tooth was extracted from each skeleton for AMS radiocarbon dating, paleodiet reconstruction ($\delta^{13}C$ & $\delta^{15}N$) and, to investigate geographic origin ($^{87}Sr/^{86}Sr$ & $\delta^{18}O$).



Figure 1: Map of the UK with the County of Cheshire and approximate location of Poulton Chapel.

Results

Biological profiles of SK463 and SK750 are presented in Table 1. Macroscopic analysis identified distinct lesions of PDB across ~60% of their skeletons (Fig. 2a). For SK463, post-mortem breaks were apparent across the skeleton which provided an opportunity to observe the internal architectural skeletal changes of PDB (Fig. 3a). Radiographic analysis provided a more detailed review of the distribution and progression of PDB. Here, up to 75% of these skeletons are affected by PDB (Fig. 2b). In SK750, significant expansion of the right humerus in observed. Radiographic analysis has identified almost complete obliteration of the medulla cavity, especially when compared with the left humerus (Fig. 3b). In SK463, both tibiae exhibit distinct bony changes of PDB (marked trabecular thickening and bony expansion) but also present anterior bowing. The anterior bowing of the tibia observed in SK463 is apparent in this radiograph (Fig. 3c).

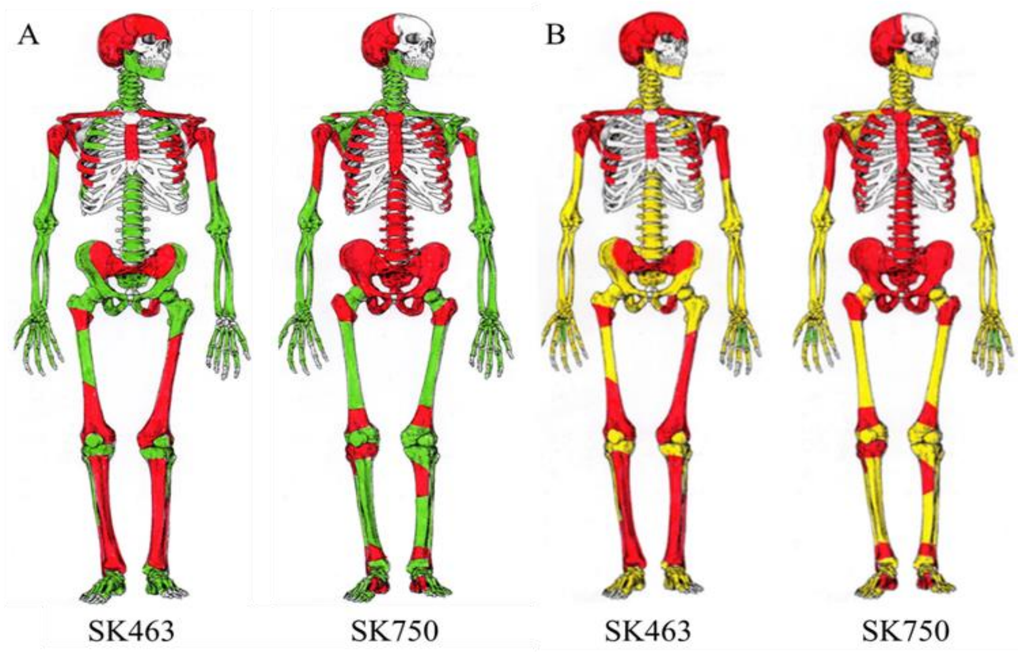


Figure 2: (A) Macroscopic distribution of bones affected with PDB (highlighted red) for each skeleton compared with unaffected bones (highlighted green). (B) Distribution of bones affected with PDB, Macroscopic changes (highlighted red), internal lytic changes identified during radiographic analysis (highlighted yellow) for each skeleton compared with unaffected bones (highlighted green).

Table 1: Biological profile of SK463 and SK750.

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

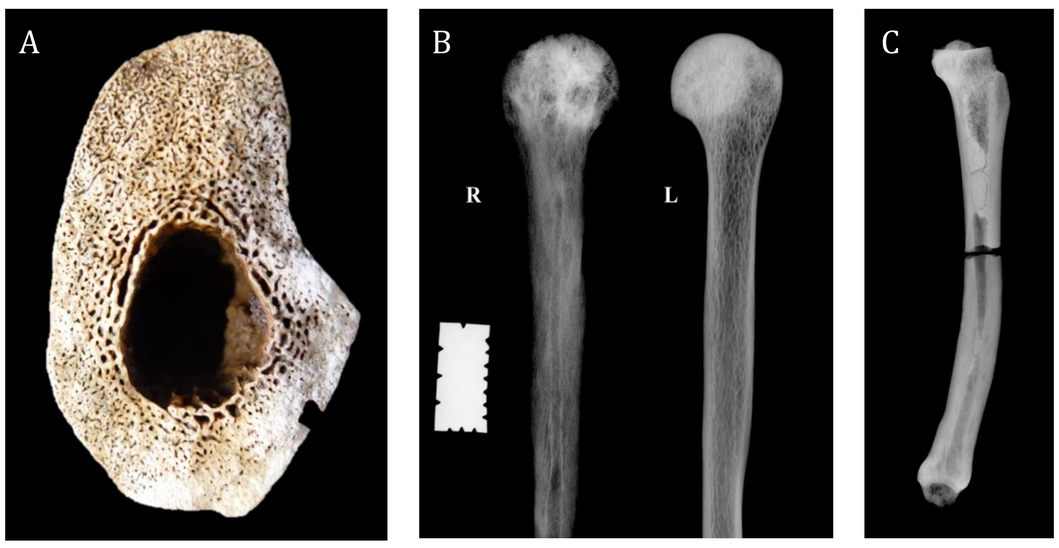


Figure 3: (A) Distinct architectural changes of PDB. (B) Radiograph of both humeri, the right displaying significant expansion and sclerosis with complete obliteration of the medulla cavity. (C) Radiograph displaying anterior bowing.

Results of the AMS radiocarbon dating identified that both skeletons are medieval in origin (AD 1275-1395). 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. 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. The $^{87}Sr/^{86}Sr$ and $\delta^{18}O_{VSMOW}$ values obtained indicate that both skeletons are local to the area, supporting the contemporary northwest concentration of PDB.

Conclusions

PDB has been identified in two medieval skeletons from Poulton Chapel. The current population prevalence is 0.5% of the adult sample (n=415). These analyses add to the emerging literature on PDB while providing additional support for the geographical hotspot observed in contemporary populations in the northwest of England. Manuscript with all the additional details is now available online!

Acknowledgments and References

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