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Case report

Fragmented skeletonised remains: Paget's disease as a method of biological profiling using radiography

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ABSTRACT

Background: This study presents the incidental finding of Paget's disease within archaeological skeletal remains using radiography. The remains of a late medieval (14th-15th century AD) adult male were excavated in 2017 from St Albans Monks' Graveyard, Hertfordshire, United Kingdom. Upon visual inspection the skeleton exhibited widespread changes suggestive of Paget's disease which prompted the radiographic investigation. The resultant imaging demonstrated an expanded bone and coarse trabecular patterns characteristic of the pathology.
Discussion: Forensic radiography provides valuable information for the reconstruction of biological profiles of the deceased for victim identification. Characteristic medical conditions such as Paget's disease aid identification by excluding unaffected persons, as the pathology exhibits a higher prevalence among British white males of advanced age. This example, although archaeological in origin, illustrates how data available from human skeletal remains can be leveraged through radiographic imaging to glean biographical information which might otherwise be unavailable. In the presence of fragmentary skeletal remains, and the absence of DNA profiling, pathologies with characteristic radiographic appearances could be of forensic significance.
Conclusion: This example demonstrates the radiographic appearances of Paget's disease on fragmentary skeletal remains which may aid victim identification efforts. Consideration must be given to the limitations of Paget's disease including anthropological estimations and the potential for alternative diagnoses. Lastly, radiography may offer an accurate, permanent record of the deceased as secondary evidence for identification efforts.

Introduction

The discovery of skeletal remains poses a challenge for identification of the deceased, especially when fragmented or commingled. An account of identification efforts for World War II individuals in Berlin attests to this, highlighting the generally poor success rate despite the availability of deoxyribonucleic acid (DNA) analysis [1]. With the absence of DNA, fingerprint or dental comparative data, secondary evidence including biometric data and medical findings may be used to aid positive identification [2]. One method of secondary evidence collection is the application of radiography to complement macroscopic documentation of remains [3]. In some instances, radiographic evidence has been pivotal to the process by arrangement of the skeletal remains to emulate antemortem radiographs [4,5]. However, the diagnosis of pathologies demonstrated in the osteological record may also help to eliminate individuals known to lack the affliction. Additionally, specific pathologies may be predisposed to discreet portions of the population according to sex, age or ethnicity.

This study presents an archaeological example of Paget's disease in a fragmented individual recovered during excavation. For clarity, Paget's disease is described as disorganised focal bone remodelling with the development of lytic lesions and chaotic bone formation, which presents as a radiologically distinctive finding with expanded bone and coarsened trabecular pattern [6]. The radiographic imaging of this individual demonstrates the characteristic appearances of the pathology and highlights a potential medical finding that could be used as secondary evidence for identification. Furthermore, this paper discusses whether Paget's disease could assist biological profiling due to its affinity with specific demographics and the utility of radiography as a method of evidence collection with skeletal remains.

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Fig. 1. Remains of the adult male with Paget's disease (right) with comparable unaffected skeleton from the same excavation site (left). In particular, note the expansive nature of the right femur shaft typical of the disease.

Materials and methods

Twenty-five late Medieval (14th – 15th century AD) skeletons were recovered from an archaeological excavation in St Albans Monks' Graveyard, Hertfordshire (United Kingdom) in 2017. One individual exhibited widespread skeletal changes during visual inspection indicative of Paget's disease (skeleton 4817, Figs. 1 and 2) [7]. Age and sex estimation proved difficult across the assemblage due to predominantly moderate to poor preservation (n = 18), missing anatomy or fragmented elements. Notwithstanding, the assemblage included 23 adults and two sub adults (<18 years), with 16 males, three females and four of unknown sex. Skeleton 4817 was relatively well preserved (albeit fragmented) and osteological and anthropological analysis of the skull and pelvic bones support that the individual was biologically male [8,9]. Attempts to refine the age of the individual were thwarted by the lack of dentition, however, inspection of the epiphyseal fusion, left auricular surface (7T/category 8), left pubic symphyseal face and the size of the individual support an estimation of a mature adult with chronic affliction [9–11].

Confirmation of the disease was sought by selective radiographic examination of the left tibia, right femur and pelvis fragments due to severity of bony change. Imaging was undertaken at Canterbury Christ Church University in 2019 by a diagnostic radiographer, using exposure factors of 60 kV and 4 mAs at a focal-film distance of 100 cm on a direct digital radiography system (Agfa Platinum Detector, Agfa Healthcare United Kingdom Limited) with a clinical x-ray table and ceiling-mounted x-ray tube (MULTIX TOP, Siemens Healthcare GmbH). Where possible, standard clinical views including anterior-posterior and medio-lateral projections were obtained to facilitate comparison with modern radiographic examples of the disease. Images were exported as DICOM files for interpretation using medical-grade monitors and viewed using RadiAnt DICOM viewer (RadiAnt Viewer version 2021.1, Poznan, Poland) [12]. A consultant radiologist was sought for definitive diagnosis. Complementary photographic imaging was also performed.

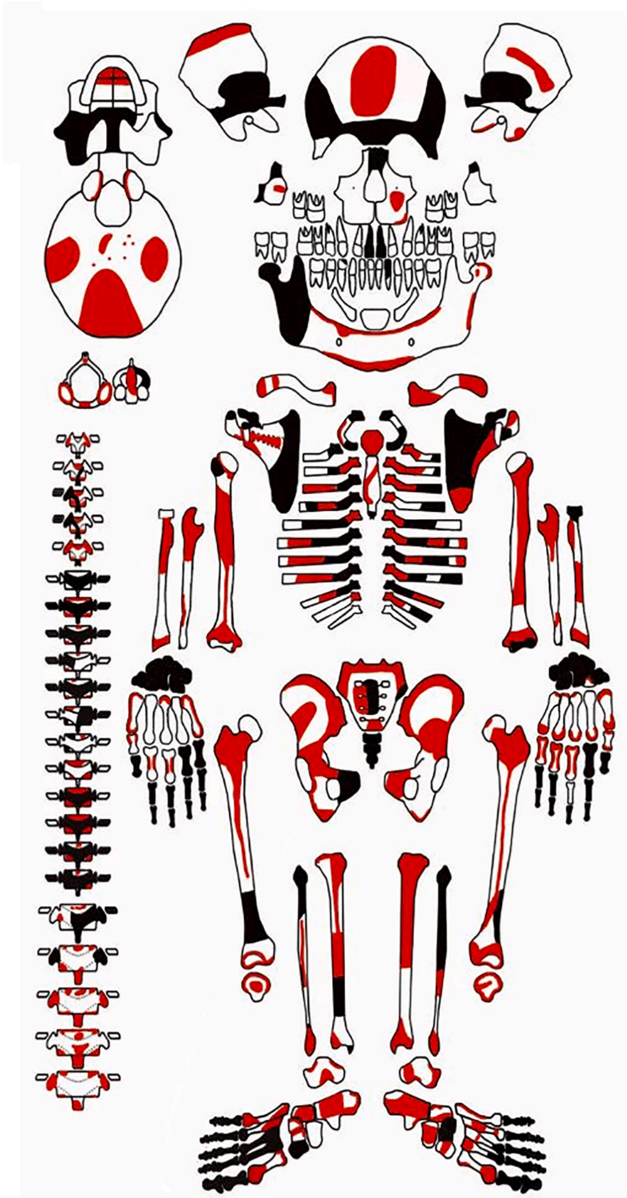

Fig. 2. Skeletal elements with areas of significant bony change highlighted in red. Black represents absent anatomy. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Results

Radiographic images demonstrated thickening of the cortex, coarse trabeculae, and expansion consistent with Paget's disease [6] (Figs. 3 and 4). Pathological changes are seen extending to the medullary cavity, lowering suspicion of changes due to trauma (ossified hematoma) or inflammation of the periosteum. Photographic imaging shows expanded bone and a rough bark-like appearance enveloping the proximal portion of the tibia (Fig. 5). Similar macroscopic appearances of Paget's disease were seen throughout the skeleton. All remains were visibly and radiographically free from soil infiltration, allowing clear visualisation of anatomy.

Discussion

Recommendations in forensic radiology research have highlighted human identification and biological profiling (sex, age, stature and ancestry) as key areas for development [13]. Although the example presented here is from an archaeological context, the same principle may be applied to human skeletal remains of legal significance. The radiographic investigation of Paget's disease within the archaeological record has been reported elsewhere with medieval examples from Cheshire (United Kingdom) [14,15]. As with the individual presented in this case report, the Cheshire examples demonstrated widespread bone changes (up to 75%) indicating polyostotic disease (affecting multiple bones). To date though, the use of Paget's disease in forensic radiography as a form of evidence in human identification has lacked academic publication. Aside from being a discriminating factor for identity inclusion, the condition is more prevalent in specific demographic groups, occurring in 1-2% of white adults over the age of 55 [16]. Whilst the aetiology of Paget's disease is not entirely understood, its epidemiology is well documented. Research has shown the pathology to be more common in men of advanced age from a British white descent [17,18]. In contrast, Paget's disease is less common in individuals of African, Asian, Chinese, Indian, Japanese and Scandinavian descent [19].

Biological profiling based upon fragmented remains may prove problematic due to incomplete anatomy, as with this example with absent dentition. The diagnosis of characteristic pathologies, such as Paget's disease, may be instrumental in delineation of potential matches with antemortem data. However, there are limitations. Individuals without the pathology in their medical records can be excluded but the condition is frequently asymptomatic and may lack diagnosis [16]. Patients may exhibit the disease in single bones (monostotic) which, through taphonomic changes, grave disturbance, or other reasons, may be missing for analysis. Likewise, consideration must be given to the differential diagnosis for osteosclerosis demonstrated on radiographs, with osteoblastic metastases and osteopetrosis both providing alternative diagnosis [20]. Tumours, infections, traumas, medications and arthritic diseases may all lead to periosteal changes with a variety of radiographic appearances which may be considered [21].

The use of radiography has been established as a cornerstone of forensic imaging, with lower financial, logistical, training and data burdens when compared to computed tomography [22]. Within a hypothetical scenario of isolated and fragmented skeletal remains the use of radiography may provide confirmation of Paget's disease, as well as other pathological conditions, and obviate the need for more advanced imaging. Whilst DNA forms part of primary evidence for confirmation of identity, investigators may face an array of limitations including a lack of antemortem data, access to appropriate equipment or necessary finances. Radiographic imaging may provide an accurate, permanent record as part of the evidence collection process. The application of radiographic imaging is highly situation-specific and likely at the discretion of the forensic pathologist or forensic anthropologist [2].

Conclusion

Scenarios where collection of DNA, fingerprint or dental data are not available or possible may rely upon secondary evidence to aid identification efforts. This article demonstrates the use of radiography with fragmented skeletal remains recovered by archaeological excavation which exhibited widespread expansion of bone and remodelling. The characteristic radiographic appearances and distribution are highly suggestive of polyostotic Paget's disease. In addition to excluding individuals without the affliction, the disease is associated with white, British males of advanced age (≥55). Although the example presented is

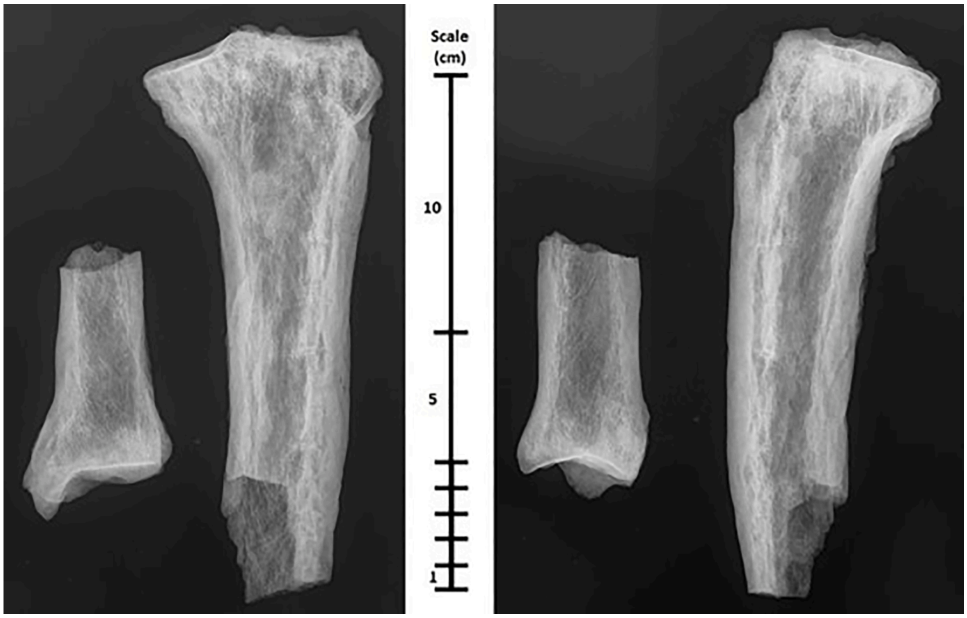

Fig. 3. Radiographs of proximal and distal fragments of the left tibia positioned in anterior-posterior (left) and medio-lateral (right) views. Radiographic features include thickened cortex and mottled medullary cavity, strongly suggestive of Paget's disease.

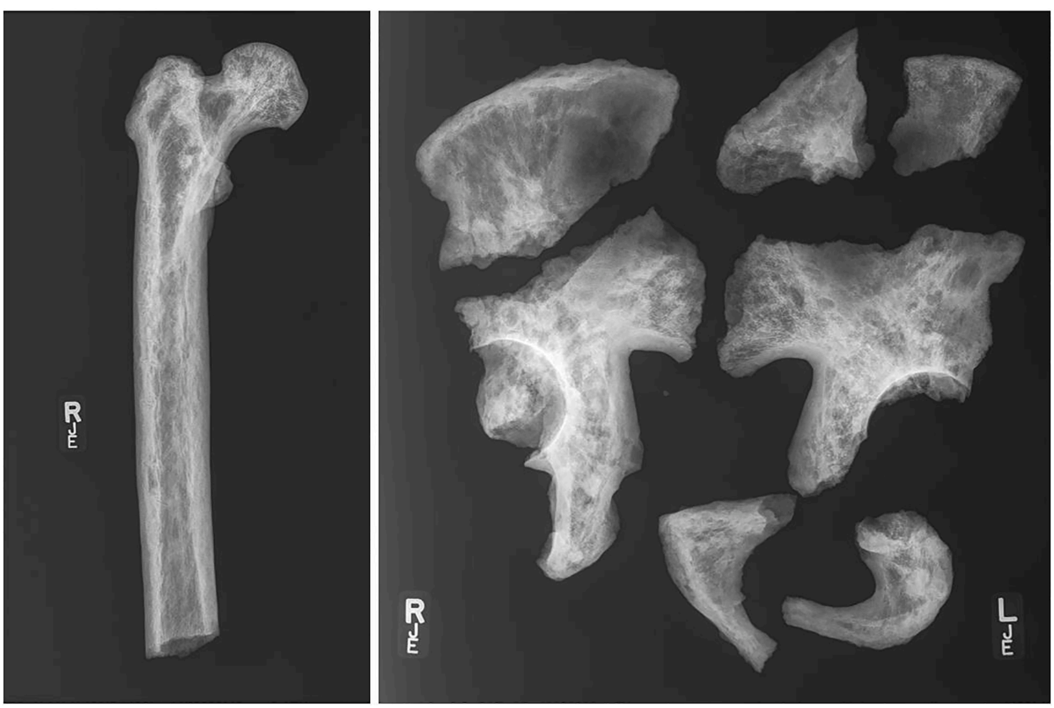

Fig. 4. Radiographs of the right proximal femur and pelvic fragments showing extensive bone changes.

of archaeological origin and therefore lacking legal impetus, the same principle may be applied to remains of forensic significance. Caution is advised though, as the pathological appearances may have alternative diagnoses and anthropological assessment may be limited (or hindered) by preservation and overall condition of bones. Nevertheless, radiographic imaging provides an accurate, permanent record of the deceased which may complement other methods of evidence collection and aid in victim identification in certain forensic settings.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

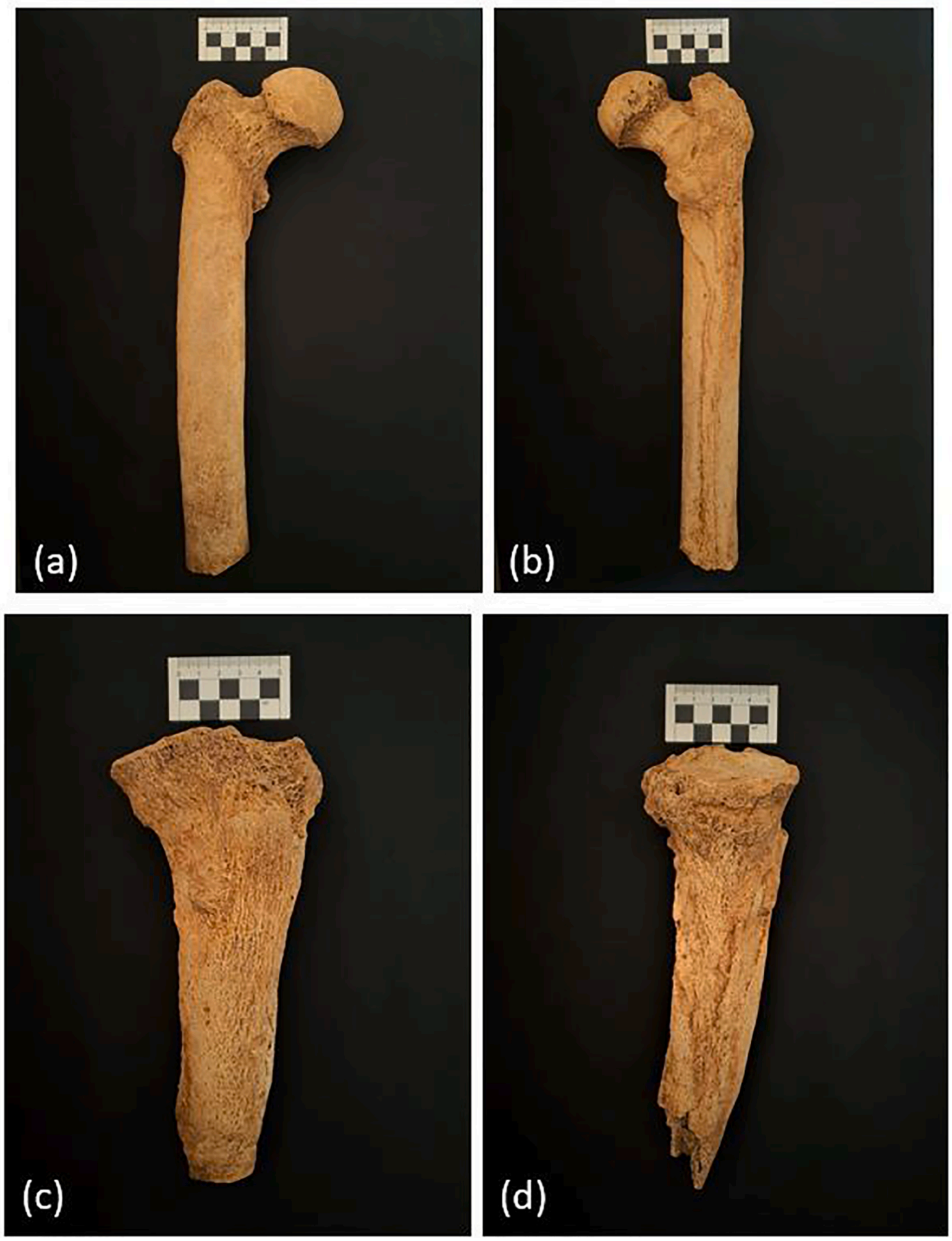

Fig. 5. Photographs of the right proximal femur anterior (a) and posterior (b) aspects. Left tibia anterior (c) and medial (d) aspect. Images display expanded shafts and mixture of lamellar and woven bone.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.fri.2023.200534.

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