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On the importance of considering disease subtypes: Earliest detection of a parosteal osteosarcoma? Differential diagnosis of an osteosarcoma in an Anglo–Saxon female

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ABSTRACT

A case of potentially dedifferentiated parosteal osteosarcoma was found in the proximal humerus of an adult female buried in the late Anglo–Saxon cemetery of Cherry Hinton, Cambridgeshire, UK. Key features include a large, dense, lobulated mass attached to the medial metaphysis of the proximal humerus by a broad-based attachment, accompanied by cortical destruction and widespread spiculated periosteal reaction. Radiographic images confirm medullary involvement, lack of continuity between the cortex and external mass, a radiolucent cleavage plane and possible radiolucent zones within the bony masses. Differential diagnoses considered include osteochondroma, myositis ossificans, fracture callus, as well as the primary malignancies of osteosarcoma and chondrosarcoma, and their various subtypes. The macroscopic and radiographic analysis of the tumor is described and discussed within clinical and paleopathological contexts. One of only 19 uncontested examples of osteosarcoma from past human populations, most of which remain unconfirmed, this case represents what we believe to be the earliest, and probably singular, bioarcheological example of parosteal osteosarcoma in human history.

1. Introduction

Skeletal evidence for malignant bone tumors in archeological populations is characterized by its sparsity and limited analysis. Arising from either mesenchymal tissues (the sarcoma family) or epithelial structures (carcinomas), primary bone tumors are far less common than secondary metastatic lesions and constitute a mere 1% of all malignancies (Waldron, 2009). Osteosarcomas are the most common type of sarcoma, yet they are incredibly rare in archeological populations with the most recent comprehensive review estimating only 21 cases in existence (Strouhal and Němečková, 2009).

The gross appearance of osteosarcoma is varied, and reflects a spectrum of pathological initiation and progression that differs according to disease subtype. The most commonly discussed are intraosseous osteosarcomas, which as the name suggests originate in the medullary cavity and generally near a metaphysis (Ortner, 2003b). These are highly metastatic tumors whose oncogenesis drives rapid invasion by stimulating osteoclasts to enable bone resorption (Broadhead et al., 2011). Osteoblastic activity is stimulated by the body in response, and thus the morphological appearance of these tumors depends on the degree of sclerotic activity promoted by the body. The second important subtype is juxtacortical osteosarcoma, a group of rare tumors that arise within periosteal layers on the surface of long–bones and are typified by a more indolent period of growth and progression. These differences in sites of origin and pathogenic processes create distinctive morphological signatures and it is the exploration of these features which is considered essential to a thorough differential diagnosis in living patients (Ortner, 2003b; Suresh and Saifuddin, 2007). Yet despite these differences, and other important disparities such as significant differences in patterns of demographic and anatomical predilection, the subtypes of osteosarcoma have rarely been explored and discussed in the examination of archeological skeletal remains. Ancient osseous sarcomas deserve much more rigorous investigation if paleopathological endeavour is to contribute to our understanding of disease causation for this important group of malignancies which feature a modern 5–year survival of only approximately

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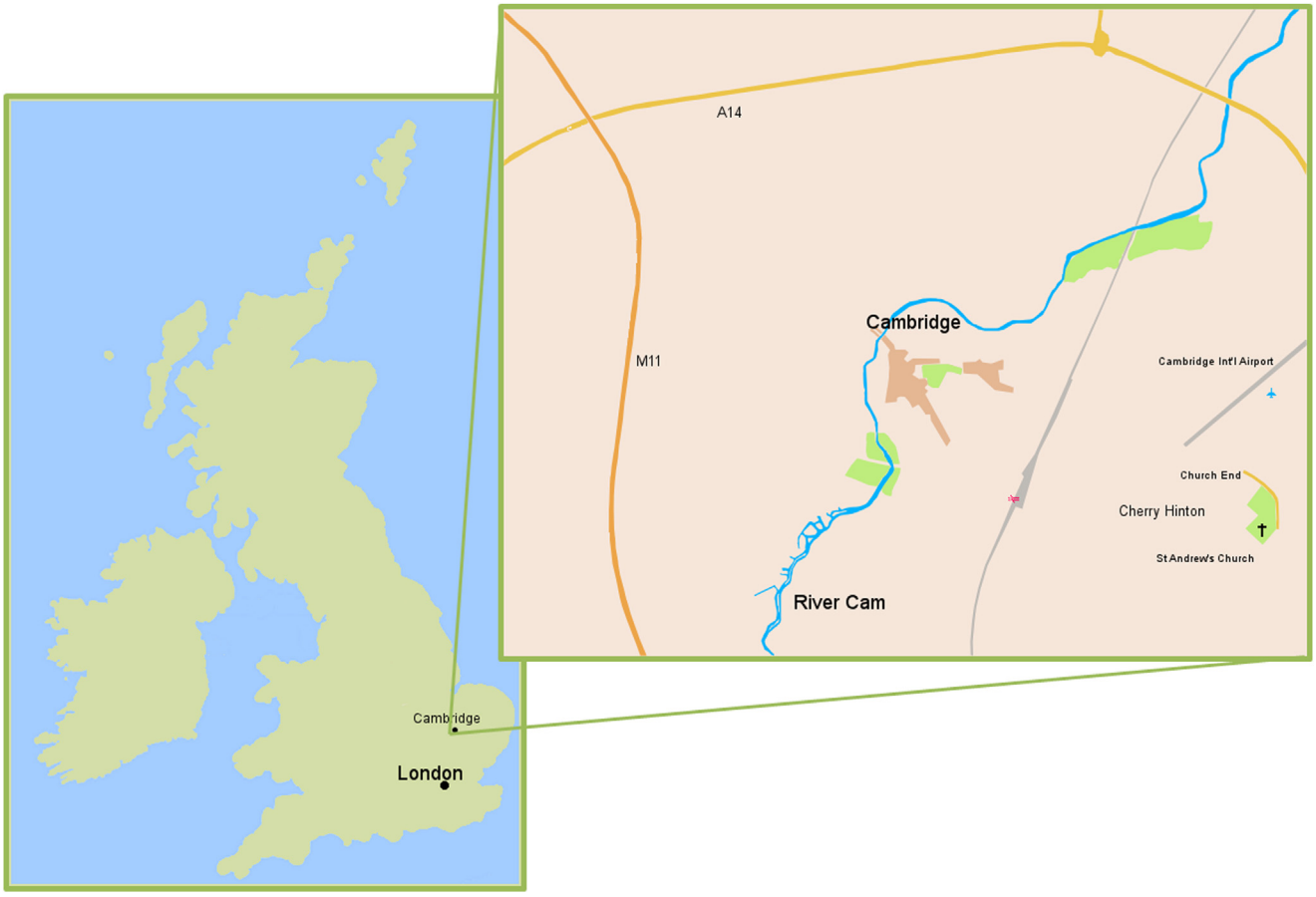

Fig. 1. Map of the United Kingdom, showing the location of Cherry Hinton, Cambridgeshire.

60% (National Cancer Intelligence Network and Public Health England, 2010). A more precise understanding of ancient prevalence, as well as a more comprehensive discernment of the true age and sex distribution of subtypes, could provide unique insight into the disease mechanisms underlying primary osteosarcomas.

In this paper we present a rare case of osteosarcoma: what we believe to be the earliest example of parosteal osteosarcoma (a juxtacortical type of osteosarcoma) so far identified, and one of only two cases (Czarnetzki and Pusch, 2000) reported from an archeological population. The lesions provide a distinctive example of malignant bone cancer, in an untreated individual with advanced oncogenic pathological change who died approximately one thousand years ago. We discuss key features discovered through in–depth gross analysis and radiological examination of the skeletal remains, and examine the differential diagnosis of various osteosarcoma subtypes.

2. Materials and methods

The subject of the present study was retrieved during excavation of a large Anglo-Saxon cemetery in Cherry Hinton, Cambridgeshire, England (OS NGR TL 488 575)(Fig. 1). Graves were oriented East–West, had no grave goods, and were distributed in rows around a central structure thought to be a Saxo–Norman church. These are all features commonly seen in Mid-Late Anglo-Saxon cemeteries in England (Taylor 2001), which represent the first Christian cemeteries after the Final Phase Period. Ceramic and stratigraphic evidence suggest the cemetery was created in the 9th Century AD, and fell out of use during the 12th Century AD, at which point the medieval church of St. Andrews was founded at the modern centre of Cherry Hinton village (Murray and Vaughn, 1999).

In total 683 individuals were disinterred, of which a little over 66% (92/278 sub-adults and 361/405 adults) presented evidence for abnormal antemortem change to at least one bone or tooth. While the most common types of affliction were dental disease (46%) and arthropathy (42%), osseous evidence for neoplasm was observed in 3% (8 button osteoma, 10 meningioma, 1 primary and 1 secondary malignancy) (Ferrante di Ruffano and Waldron, 2006). We discuss skeleton 4411 (SK4411), the sole individual to demonstrate clear signs of a malignant, primary bone tumor.

The degree of preservation was fair–poor with half the skeleton remaining, including an intact cranium, most vertebrae, partial pelvis and the right appendicular skeleton (Fig. 2). Other than the cranium, existing elements had suffered severe fragmentation and surface abrasion by post-depositional factors. Due to absence of the pubis, sex was determined by morphology of the cranium while age-at-death was estimated using morphological observation of the auricular surface (Lovejoy et al., 1985) and patterns of molar attrition (Brothwell, 1981). An estimation of stature was calculated from the maximum length of the radius using the standard formula for a Caucasian female published by Trotter and Gleser (1958, 1977).

Pathological bones were subject to macroscopic analysis and radiographs were taken with a Todd x-ray cabinet using a 0.2 mm aluminum filter, set at a constant power of 3 mA. Exposure was 50 Kvp at 35 and 45 s. No other bones were radiographed.

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