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Case Study article

A rare probable chondroblastoma of the calcaneus in a pre-Columbian subadult from Illinois

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ABSTRACT

Discrete cystic or tumorous intraosseous lesions can arise from a variety of benign and malignant conditions as well as trauma and infection. They are clinically rarely observed in the calcaneus. A fourteen-to-seventeen-year-old subadult recovered from a Late Woodland (~AD 800-1100) period mortuary context in the Mississippi River Valley of central Illinois presents with a single lytic intraosseous lesion on the posterior right calcaneus that bilaterally perforates the cortex. The lesion, although primarily anterior to the epiphyseal plate, does breach it. There is also a small perforation of the outer cortex of the epiphysis above the insertion of the Achilles' tendon. The lesion is well-defined with a primarily spongy cancellous interior margin. On the body of the calcaneus, there is periostosis and a slightly expansive endosteal reaction. Comparative radiographic assessments undertaken to differentially diagnose the lesion indicate that it was likely not malignant. Based on the posterior location, the radiographic signature, the bilateral cortical perforation and the breach of the epiphysis, the lesion is best interpreted as a chondroblastoma.

1. Introduction

The occurrence of tumors (any abnormal mass of tissue) or cysts (air, fluids, or semi-solid tissue enclosed in a sac) in the calcaneus is clinically rare (e.g. Andermahr et al., 2004; Foo and Raby, 2005; Oommen et al., 2009; Polat et al., 2009; Weger et al., 2013) and most often identified in adults (e.g. Polat et al., 2009). Tumors or cysts can be the result of any one of a number of malignant or benign pathological conditions as can intraosseous lesions formed by sequestered infections or mechanical injury (Mascard et al., 2015; Oommen et al., 2009; Revenga Martínez et al., 2007). Certain malignant conditions, such as chondrosarcoma and Ewing's sarcoma, have a predilection for the calcaneus (Kilgore and Parrish, 2005). But, even if a calcaneal intraosseous lesion is benign, there may be tangible life-affecting biomechanical consequences to locomotion or other physical activities (Foo and Raby, 2005; Hafner et al., 2011; Hatori et al., 2001; Murani et al., 1989). Lesions of the calcaneus are very rarely described in the paleopathological literature (Curto and Fernandes, 2016; González-Reimers et al., 2015). A cortex-perforating lesion of the calcaneus is macroscopically visible in the skeletal remains of a prehistoric subadult from west-central Illinois. In addition to this case as the first reported calcaneal intraosseous lesion from pre-Columbian North America, the lesion enables a comparison of the radiographic signature to the reactive change on dry bone.

1.1. Site background

Schroeder Mounds (11He177) is a Late Woodland Period (~AD 800–1100) mortuary site located on a low bluff overlooking the eastern bank of the Mississippi River floodplain in Henderson County, Illinois (Fig. 1). The site was serendipitously discovered during the remodeling of a domestic structure and the impacted area was excavated in 1979 (Kolb, 1982). The site yielded 123 well-preserved adult and subadult burials (Nicosia et al., 2016; Mosher et al., 2015; Smith et al., 2016). Although there are no Late Woodland village or other domestic structures associated with the site, indirect archaeological evidence suggests a forager-farmer subsistence/settlement pattern (Esarey, 2000; Nansel and Green, 2000; Stoltman, 2000).

2. Case description

2.1. Age, sex, and preservation

The only long bone diaphyses of Burial 33 (Fig. 2) to exhibit epiphyseal fusion is the distal humerus which, preserving the

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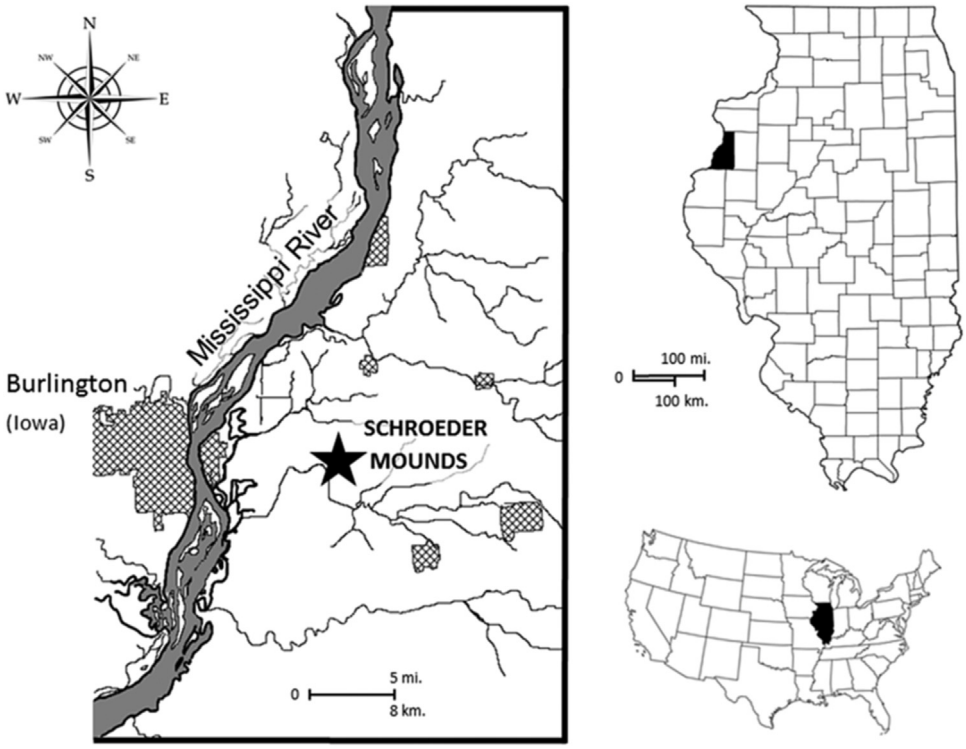

Fig. 1. Map of the location of the Schroeder Mounds site (11He177) in Henderson County, Illinois. The site is approximately six miles east of the present-day city of Burlington, Iowa.

epiphyseal line, suggests an age-at-death (Buikstra and Ubelaker, 1994; Schaefer et al., 2009; White and Folkens, 2005:372) of between fourteen and seventeen years. The level of molar crown-root development (Ubelaker, 1989:64) of Burial 33 is also consistent with this age-at-death. Based on the preserved pelvic anatomy (i.e., wide subpubic angle, quadrangular pubic corpus) (Buikstra and Ubelaker, 1994), Burial 33 is likely a female.

This subadult preserves most of the long bone diaphyses and epiphyses, twenty vertebrae, bones of the mid face, three small fragments of temporal squama, and the mandible (Fig. 2). The only evidence of reactive change on the long bones is an area of periostosis on the anterior left tibia (Fig. 3a and b). There is no evidence of tuberculosis in the Schroeder Mounds sample (Mosher et al., 2015) or other chronic infection in Burial 33 that might account for a focal lesion. The hands and feet are fragmentary. There are six manual phalanges (two are certainly distal), three identifiable metacarpals (left and right fourth, right fifth), four right carpals (capitate, trapezoid, lunate, and scaphoid) and two left carpals (lunate, scaphoid) (Fig. 2). The right fifth metacarpal exhibits periosteal plaque on the dorsal surface (Fig. 3c). There are no pedal phalanges and the metatarsals consist of two non-diagnostic distal epiphyses and a fragmentary left proximal metatarsal. The only tarsals present are the left and right talus, the left and right calcaneus, and the left cuboid. Although the left calcaneus is very fragmentary, the body preserves an epiphyseal surface suggesting an unfused epiphysis. The complete right calcaneus exhibits epiphyseal fusion with the epiphyseal line beginning to obliterate along the lateral margin.

2.2. The right calcaneus

The posterior calcaneal body (maximum length 65 mm, maximum width 39 mm) exhibits a well-defined, bilaterally perforating, lytic lesion (circa 15 mm × 15 mm orifice) anterior to the epiphyseal margin (Fig. 4a and b). The lesion breaches the epiphyseal plate at the unfused medial margin and is anterior to the fusing lateral margin (Fig. 4a). The lateral orifice is surrounded by expansile periostosis across the peroneal tubercle (Fig. 4b).

The medial and lateral orifices have a smooth cortical margin with a rounded rather than a sharp-edged perimeter. The defect has the appearance of a bore-hole with a slightly lobulated interior surface. The interior surface is macro-porotic on the anterior margin (Fig. 5a) and becomes smooth-walled at the posterior margin (Fig. 5b). The epiphysis has a small perforation at the area of the insertion of the Achilles tendon (Fig. 5c). These reactive changes on the lateral aspect of the calcaneus may have contributed to the fragmentation of the calcaneal epiphysis. That is, the lateral process, instead of being the area of initial epiphyseal fusion (Scheuer and Black, 2000:463) is a separate and unfused epiphysis (Fig. 5d). There is no evidence of any traumatic injury to the calcaneus. The only other preserved bone of the right foot is a fragmentary talus which exhibits no reactive change. The discrete area of periosteal plaque on the anterior left tibia and the right dorsal fifth metacarpal appear to have no relationship to the calcaneal lytic lesion.

2.3. Radiographic images

The radiographs of the calcaneus confirm the observations on the dry bone. In the lateral view (Fig. 6a), the lesion is visible as a well-defined radiolucent area anterior to the epiphyseal line. The contour is somewhat lobulated but not “soap-bubble” (i.e., Kuna and Gudena, 2011), and the margin is not sclerotic. The lesion does not alter the contour of the bone. The mottled appearance of the calcaneal body may reflect the periostosis evident on the dry bone. The radiograph also indicates the bilateral perforation of the cortex. In the coronal view (Fig. 6b), there is no radiolucent area indicating the presence of a lytic defect. However, despite the opacity generated by the articular facets, an area of diffuse (presumptive periostotic) mottling is evident.

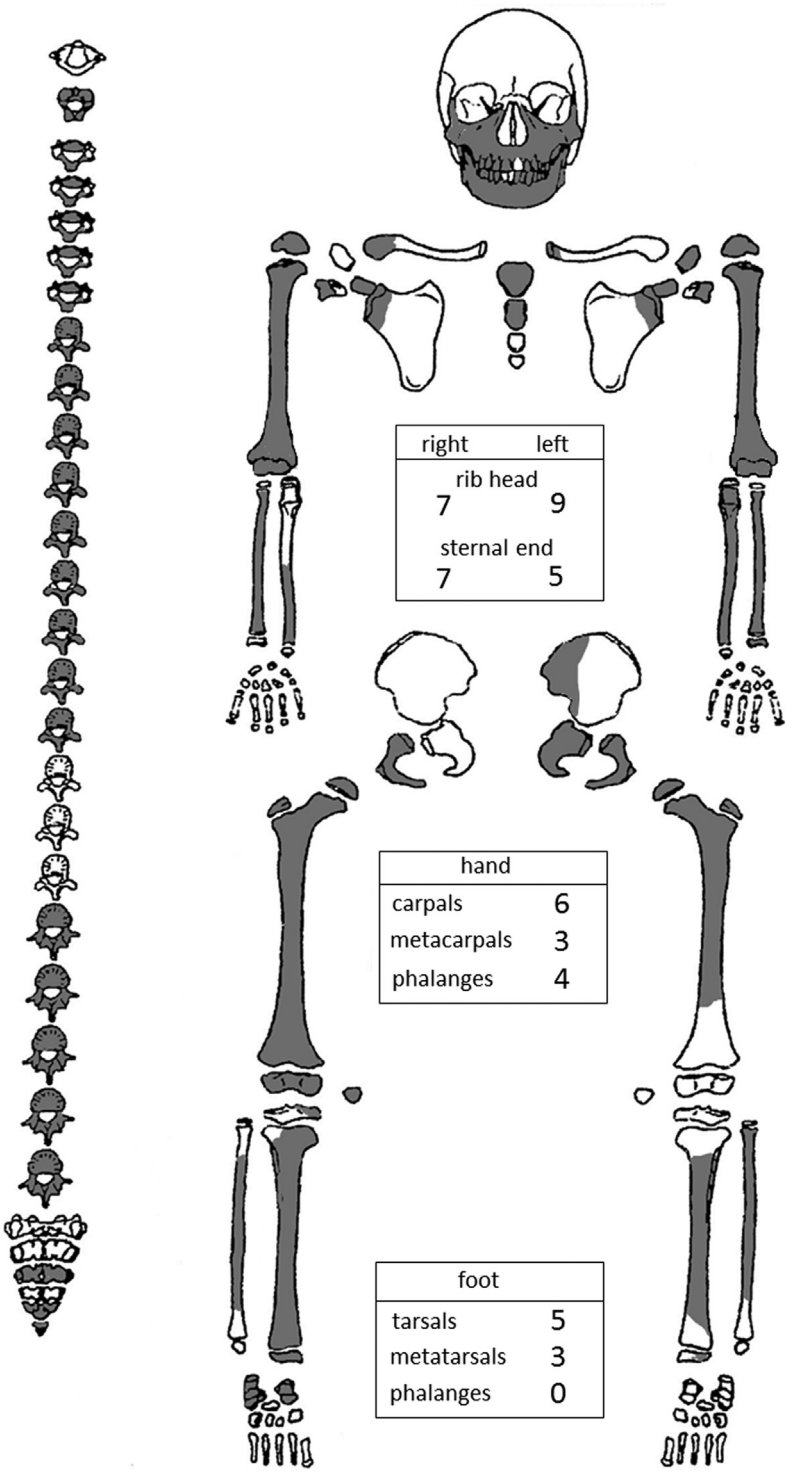

Fig. 2. The inventory of the skeletal elements attributed to Burial 33.

3. Discussion

In clinical contexts, the differential diagnosis of lytic lesions considers the age of the individual, the affected bone, the location on the bone (i.e., diaphysis, metaphysis, epiphysis), the number of lesions (single or multiple), and a suite of attributes visible on the radiographic image (Foo and Raby, 2005; Helms, 1995; Kilgore and Parrish, 2005; Miller 2008; Oommen et al., 2009). The radiographic attributes include the width and clarity of the radiopaque zone of transition from normal to abnormal bone (i.e., sharp margin, ill-defined margin, sclerotic margin, sclerotic mass [radiopacity]), the presence of diaphyseal or metaphyseal expansion (ballooning, "soap-bubble"), cortical perforation, the geometric shape of the radiolucency (simple, multi-lobular), the location within the bone

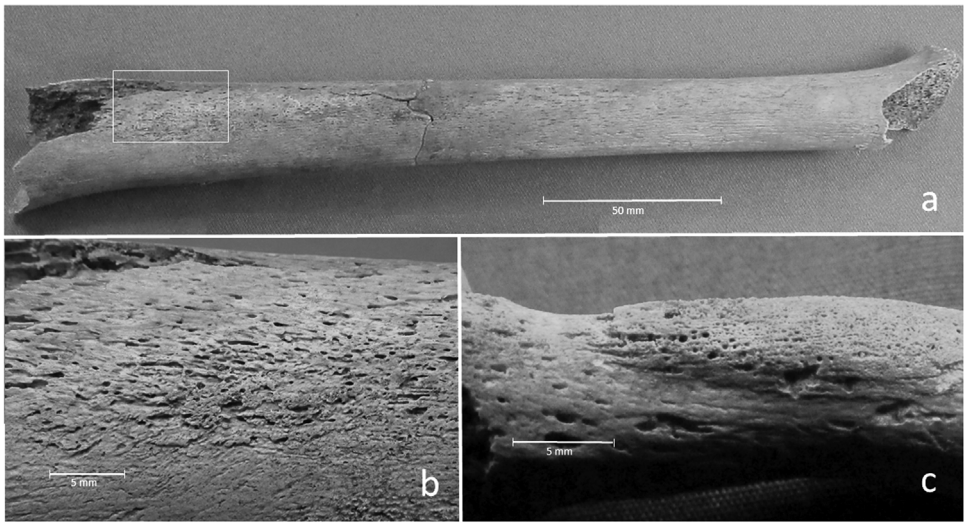

Fig. 3. The additional pathological features of the Burial 33 skeleton include: a.), b.) a patch of periostitis on the anterior left tibia; c.), a discrete area of woven bone on the dorsal surface of the right fifth metacarpal.

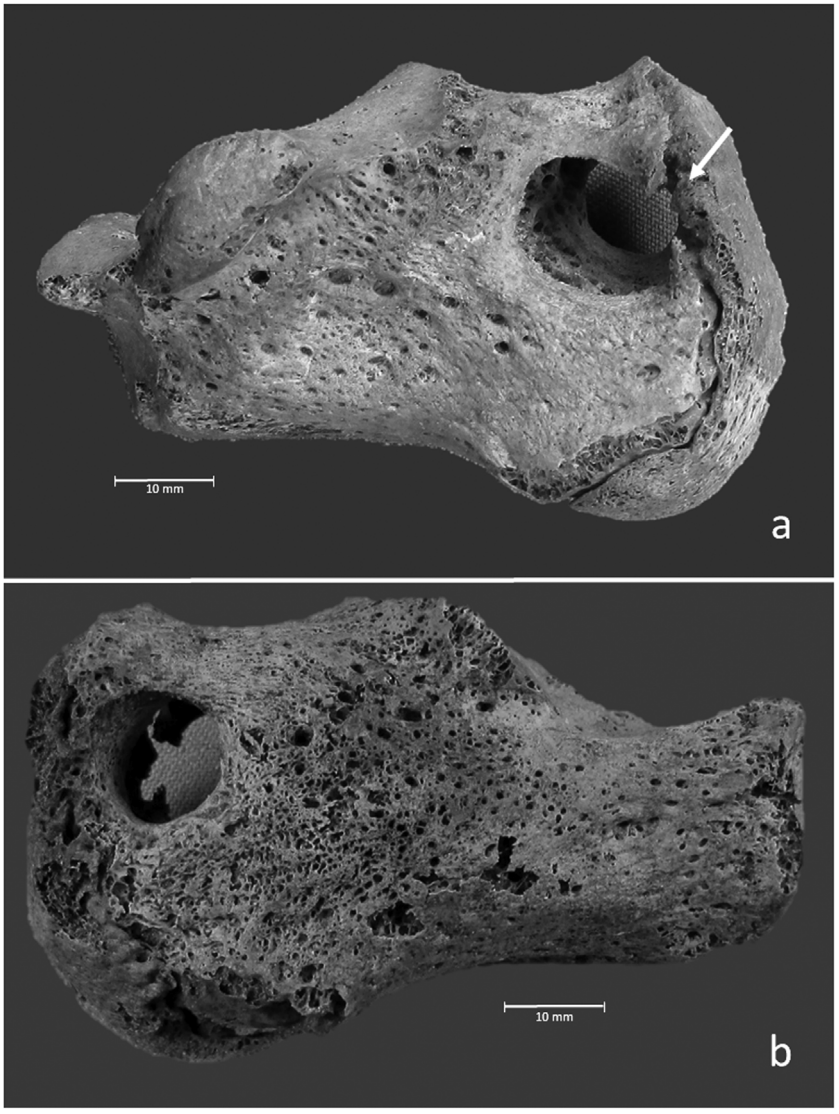

Fig. 4. The intraosseous lesion on the right calcaneus from the a.) medial and b.) lateral aspects. The perforation of the epiphysis is evident on the medial aspect (arrow). The lateral surface of the metaphysis exhibits periostotic reactive change.

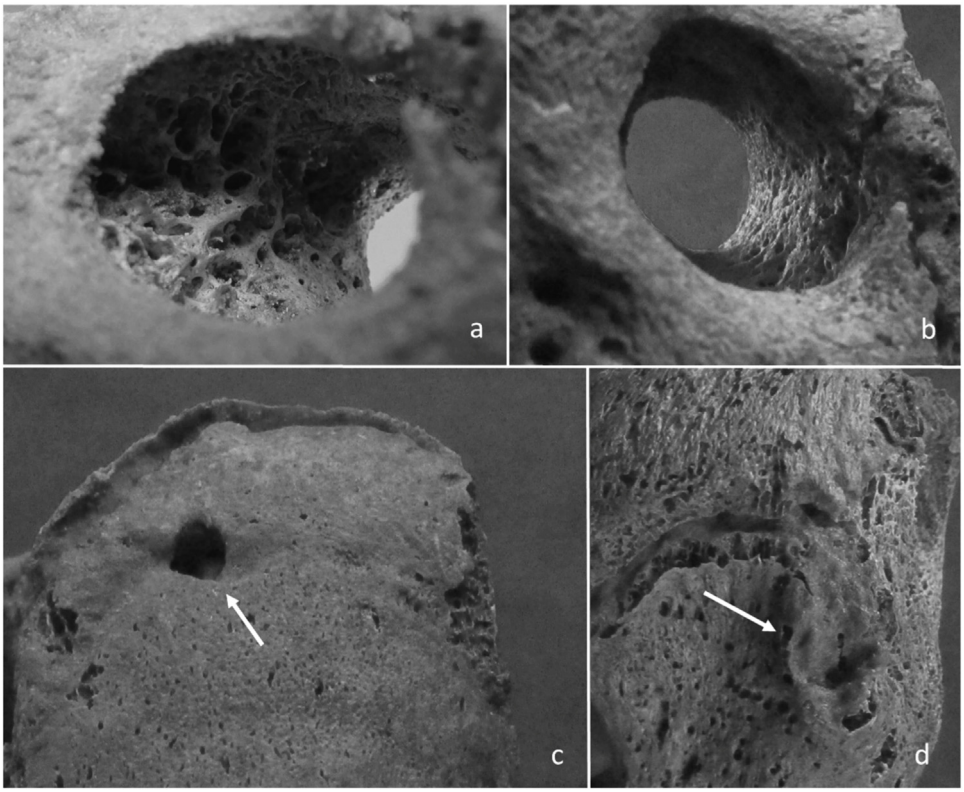

Fig. 5. Details of the reactive change on the calcaneus of Burial 33. The anterior wall of the lesion a.) is composed of coarse metaphyseal trabeculae and the posterior wall b.) is smooth. The lesion perforates the apophysis c.) just superior to the Achilles enthesis (arrow). The lateral process of the calcaneal apophysis d.) has a separate unfused epiphysis (arrow).

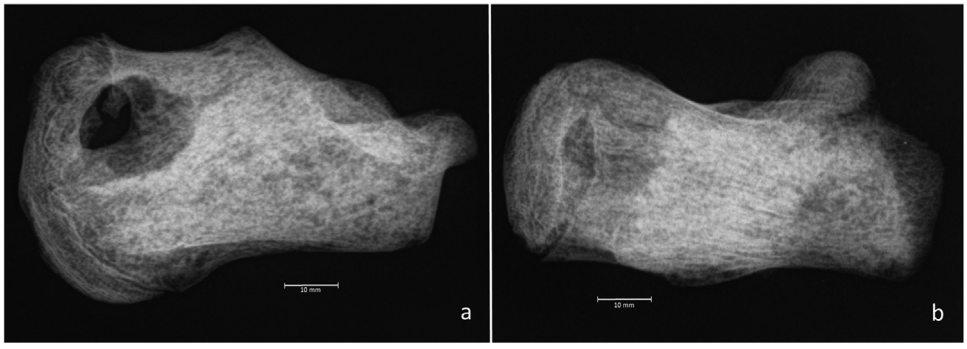

Fig. 6. The radiograph of the medial aspect a.) indicates a sharp-margined, perforating, mildly lobulated, lytic area with no sclerotic margin. Periostotic change is evident from the radio-dense mottling on the diaphysis. The inferior aspect b.) reveals the mottling of localized periostosis but no contours of a lytic lesion.

(central, eccentric [lateral], cortical), and the presence of periosteal reaction (Helms, 1995; Kuna and Gudena, 2011; Miller, 2008).

The circumstance of assessing the dry bone case in tandem with the radiograph provides three-dimensional anatomical details not visible on a two-dimensional radiograph that may have diagnostic relevance. Therefore, the perforation of the epiphyseal plate, the perforation of the epiphysis, the bilateral perforation of the cortex, the presence of periostosis, and the structure of the internal margins of the lesion (both spongy and smooth-walled) are important for the differential diagnosis of this lesion.

3.1. Malignant and infectious processes

A small number of malignant tumors and infectious processes (e.g., osteomyelitis) can occur in the calcaneus of subadults. The radiographic signature of the lesion of Burial 33 (a sharp zone of transition) is qualitatively different from the reactive change indicative of sarcomas such as ill-defined margins and sclerotic masses (Agarwal and Bin Sabir, 2008; Helms, 1995; Miller, 2008). This suggests that the calcaneal lesion is likely due to a benign process.

Although the prevalence of (hematogenous) osteomyelitis has been reported to be as high as eight percent in children (Puffinbarger et al., 1996; Rasool, 2001), it may also be diagnostically eliminated as the dry bone reactive change in the calcaneus is not consistent with a suppurative pyogenic process (González-Reimers et al., 2015; Rogers and Waldron, 1989), and it does not exhibit the mixed radiographic reactive change of osteomyelitis (e.g. sclerosis and/or sequestration) (Puffinbarger et al., 1996; Rasool, 2001). A form of subacute pyogenic osteomyelitis common to subadults is the Brodie's abscess (Bogoch et al., 1984; Kozlowski, 1980), but it, too, is an unlikely interpretation as the lateral radio-

graph of Burial 33 lacks the wide zone of sclerotic, often scalloped, bone characteristic of the fully formed Brodie's abscess (Bogoch et al., 1984; Pöyhiä and Azouz, 2000).

3.2. Monostotic benign lesions

The benign intraosseous lesions included in the differential diagnosis are all sharp-margined, have a subadult age of onset, and occur in the calcaneus. The likelihood of a correct diagnosis of the lesion in Burial 33 depends on the diagnostic value of the posterior location, the absence of cortical expansion (e.g., ballooning), the absence of a sclerotic margin, the presence of cortical perforation, the presence of periosteal reaction, and the breach of the epiphyseal plate. The three most common intraosseous lesions of the calcaneus in subadults are the unicameral or simple bone cyst (UBC), the intraosseous lipoma (IL), and the osteoid osteoma (Aycan et al., 2015; Kumar et al., 1991; Mascard et al., 2015; Oommen et al., 2009; Weger et al., 2013).

3.2.1. Unicameral or simple bone cyst

The unicameral bone cyst (UBC) is a benign lytic lesion that develops in subadults between the ages of four and ten years and, as its name suggests, is a fluid-filled cavity (of serum and blood) with a fibrous wall (Mascard et al., 2015; Lenze et al., 2015; Javdan et al., 2012). The UBC constitutes three percent of all bone tumors (Aycan et al., 2015), six percent of which occur in the calcaneus (Kadhim et al., 2014). It is of uncertain origin with an incompletely understood pathogenesis (Lenze et al., 2015). The UBC arises adjacent to the growth plate (Pogoda et al., 2004) and, in non-calcaneal contexts, does occasionally breach it (e.g., Capanna et al., 1986; Gupta and Crawford 1996; Malawer and Markle 1982; Ovadia et al., 2003). The UBC appears radiographically as a single oval-shaped radiolucency with a well-demarcated border, a narrow zone of transition, minimal loculations, and a mildly expansile periosteal response (Johnson 2011; Mascard et al., 2015; Lenze et al., 2015; Lasanianos et al., 2011; Kacki et al., 2010). Although the typical location of the UBC is in the anterior third of the calcaneus inferior to the subtalar joint within a triangle of reduced trabecular density (*trigonum calcis*) (Andermahr et al., 2004; Foo and Raby 2005; Lasanianos et al., 2011), it can occur in the posterior calcaneus (Oommen et al., 2009). Although the UBC may span the entire width of calcaneus (Pogoda et al., 2004), the clinical literature does not report cortical perforation (Kadhim et al., 2014). Because of the bilateral perforation of the calcaneal cortex in Burial 33, and the lack of specific clinical evidence for breaches of the calcaneal epiphysis, the unicameral bone cyst is an unlikely diagnosis.

3.2.2. Intraosseous lipoma

The IL, a rare intraosseous cystic lesion (circa 0.3%) of which one-third occur in the calcaneus (Foo and Raby 2005; Oommen et al., 2009), is composed of mature fat cells (Gonzalez et al., 1997; Revenga Martínez et al., 2007). Radiographically, it is an oval with well-defined margins but, unlike Burial 33, it is most often surrounded by a sclerotic rim (Hatori et al., 2001). The majority of calcaneal lipomas are located within the triangle of reduced trabecular density (*trigonum calcis*), making it radiographically difficult to distinguish from a late stage UBC (Foo and Raby, 2005; Kumar et al., 1991; Polat et al., 2009). However, despite the clinical frequency of the subadult calcaneal IL, the epiphyseal location of the lesion in Burial 33 negates it as a diagnosis.

3.2.3. Osteoid osteoma

The osteoid osteoma is a bone-forming vascular tumor that is also radiographically described as an oval or round radiolucency with no marginal sclerosis (Foo and Raby, 2005; Hamada et al., 2016; Pogliacomi and Vaienti, 2003). It accounts for approximately ten percent of all intraosseous tumors and circa twelve percent of calcaneal lesions (Foo and Raby, 2005; Hamada et al., 2016; Pogliacomi and Valieti, 2003). Attributes of the osteoma that rule it out as a diagnosis include its size (it rarely exceeds 1.5 cm), a predisposition to the subtalar area of the calcaneus, and a perimeter of a radiopaque zone of reactive bone (Foo and Raby, 2005; Kumar et al., 1991).

3.3. Alternative diagnoses

Four other lesions occur in subadults and radiographically present as well-demarcated lytic radiolucencies in the calcaneus (Foo and Raby, 2005; Kumar et al., 1991; Oommen et al., 2009). These include, in order of diagnostic likelihood, the chondroblastoma, the non-ossifying fibroma (NOF), aneurysmal bone cyst (ABC), and the chondromyxoid fibroma (CMF).

3.3.1. Chondroblastoma

Chondroblastomas (CB), or Codman tumors, are cartilaginous defects that predilect the epiphysis in the long bones of subadults (Blitch and Mendocino, 1996; Erickson et al., 2001; Fink et al., 1997; Kricun et al., 1977). The CB has been observed to be non-epiphyseal in the calcaneus (Davila et al., 2004). Potentially malignant, they account for less than two percent of all bone tumors but seven percent of these occur in the calcaneus (Blitch and Mendicino, 1996; Foo and Raby, 2005; Otsuka et al., 2002; Tsai et al., 2010), and they account for less than seven percent of all calcaneal lesions (Young et al., 2012). They are found in two locations: the talocalcaneal joint and the posterior calcaneus (Dutt et al., 2015; Fink et al., 1997; Kricun et al., 1977; Kumar et al., 1991; Tsai et al., 2010). Diagnostically relevant, they have well-defined radiolucencies with a thin sclerotic border, usually multi-lobular (often scalloped) but can be smooth, and are associated with periosteal reaction (Blitch and Mendocino, 1996; Fink et al., 1997). Unlike the UBC, which is routinely cited as an alternate diagnosis, there is clinical data to support cortical perforation in the calcaneus (Bloem and Mulder, 1985; Liu et al., 2013; Turcotte et al., 1993).

3.3.2. Non-ossifying fibroma

The NOF (or fibroxanthoma) is a metaphyseal defect arising from the cortex that is extremely common in subadults with as many as one-third of otherwise normal children exhibiting at least one somewhere in the skeleton (Helms, 1995; Herget et al., 2016; Kilgore and Parrish, 2005; Oommen et al., 2009). It typically occurs in long bones, particularly in the distal tibia (Bowers et al., 2013; Herget et al., 2016), but diagnostically important, it does present in the calcaneus (Foo and Raby, 2005). It can perforate the cortex and become large in size (Bowers et al., 2013; Schajowicz and Gallardo, 1953). Consistent with Burial 33, they arise in the metaphysis adjacent to the epiphysis; eventually they do migrate away from the growth plate. However, contrary to Burial 33, the epiphysis is never affected, there is no accompanying periostosis, and there is usually a thin sclerotic margin. The most important exclusionary attribute of the NOF is that it routinely presents as a multi-lobular defect.

3.3.3. Aneurysmal bone cyst

The aneurysmal bone cyst (ABC) is a benign, osteolytic, expansile, and hemorrhagic lesion that is locally destructive and accounts for six percent of all benign bone lesions (Mascard et al., 2015; Babazadeh et al., 2011; O'Connor et al., 2004). Consistent with the age and sex of Burial 33, the ABC usually presents in the first two decades of life with a slight female predominance (Johnson, 2011) and occurs in, but does not predilect, the calcaneus (Hertzanu et al., 1984). The cyst can either be primary or secondary to other lesions (e.g., chondroblastoma) (Ly et al., 2004). Although radiographically the ABC has well-defined margins, key characteristics

are different from those of Burial 33: the ABC is expansile, often "soap-bubble," and eccentric (Foo and Raby, 2005; Johnson, 2011; Kuma and Gudena, 2011).

3.3.4. Chondromyxoid fibroma

The CMF is a very rare defect, reflecting less than 0.05% of all bone tumors and very rarely presents in the calcaneus (Jamshidi et al., 2013; Khodamorad et al., 2013; Oommen et al., 2009). Of diagnostic importance, it originates in the marrow space of the metaphysis close to the epiphysis and may extend into the epiphysis (Schajowicz and Gallardo, 1953). The CMF can be misdiagnosed for a UBC (Roberts et al., 2013). However, it is generally subarticular, has an eccentric location, and the contours of the CMF are described as "soap-bubble" with sclerotic margins (Jamshidi et al., 2013; Khodamorad et al., 2013; Schajowicz and Gallardo, 1971; Tsai et al., 2010). These radiographic attributes are inconsistent with those of Burial 33 and is therefore an unlikely diagnosis.

4. Conclusion

Defects in the calcaneus are clinically rare and virtually invisible in the paleopathological record. The case of Burial 33, a subadult presumptive female from the pre-Columbian North American site of Schroeder Mounds, presents a unique diagnostic paleopathological context. The lesion on the posterior body of the dry bone bilaterally perforates the cortex revealing the coarse trabecular bone of the interior margin as well as a breach of the epiphyseal plate. The radiograph of the lesion exhibits a sharp non-sclerotic margin, and minimal lobulation consistent with the radiographic signatures of several benign intraosseous lytic defects. Of these, the chondroblastoma (CB), a rare benign tumor of bone and which accounts for maximally seven percent of all intraosseous calcaneal lesions, is the best diagnostic fit.

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