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International Journal of Paleopathology

journal homepage: www.elsevier.com/locate/ijpp



Case study

A rare naviculocuneiform I coalition from Bronze Age Siberia

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ARTICLE INFO

Article history:

Received 2 February 2012

Received in revised form 22 March 2012

Accepted 25 March 2012

Keywords:

Tarsal coalition
Navicular
Cuneiform
Congenital
Siberia

ABSTRACT

Naviculocuneiform coalitions are among the least common types of tarsal coalition, a rare congenital anomaly characterized by incompletely separated tarsal bones. Only a handful of cases have been documented from the archaeological record, none of which have been recovered from north central Asia or date to earlier than about 2000 years B.P. Here, we present a case of nearly complete osseous coalition of the left navicular and cuneiform I from the early Bronze Age (ca. 5200/5000–4000 cal. BP) Lake Baikal region of Siberia (Russian Federation). We also provide substantial evidence for the congenital (rather than acquired) basis of this unusual condition and discuss, using modern clinical data, possible *intra vitam* complications.

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1. Introduction

Tarsal coalition, characterized by the incomplete separation of two or more adjacent tarsal bones (Fopma et al., 2002; Sartorius and Resnick, 1985), is a heritable and relatively rare congenital defect affecting 2–5% of most populations (Case and Burnett, 2010). The condition arises from abnormal cavitation of the mesenchyme (embryonic connective tissue) during prenatal development. Such coalitions have been observed on embryonic specimens as early as the eighth week of life (Gardner et al., 1959; Harris and Beath, 1948; Jack, 1954; Kawashima and Uhtohoff, 1990). In the majority of cases, the cartilaginous bridges uniting adjacent tarsals remain non-osseous (cartilaginous or fibrocartilaginous) throughout life, but a small proportion of coalitions, generally less than 5% (Case and Burnett, 2010), will ossify as individuals mature. Osseous coalitions are more likely to restrict normal tarsal movement and to be linked to potentially painful foot conditions such as peroneal spastic flatfoot (or rigid flatfoot) and tarsal tunnel syndrome, as well as to an increased susceptibility to trauma such as sprains (Case and Burnett, 2010; Harris and Beath, 1948; Jayakumar and Cowell, 1977; Kumai et al., 1996; Mosier and Asher, 1984; Sartorius and Resnick, 1985; Snyder et al., 1981; Takakura et al., 1991, 1998; Varner and Michelson, 2000). In most cases, pain and discomfort associated with coalitions appear to reflect mechanical stress at coalition site(s) and/or abnormal posture among adjacent tarsals,

rather than inflammation (Fopma et al., 2002; Kumai et al., 1998; Sakellariou and Claridge, 1998).

Tarsal coalition has been most frequently documented for the talus, calcaneus, and navicular, with talocalcaneal and calcaneonavicular coalitions accounting for over 90% of reported cases (Case and Burnett, 2010; Gregersen, 1977; Stormont and Peterson, 1983; Varner and Michelson, 2000). In fact, a probable case of calcaneonavicular coalition recently reported from Italy has been dated to as early as the Upper Paleolithic (Villotte et al., 2011). Coalitions also affects other tarsals, albeit much less commonly, as well as metatarsals and pedal phalanges (Case and Burnett, 2010; Case and Heilman, 2005; Le Minor, 1995; Regan et al., 1999; Stormont and Peterson, 1983). Isolated naviculocuneiform coalitions, the vast majority of which involve cuneiform I (or the medial cuneiform), are among the least common tarsal coalitions overall (Burnett and Case, 2005; Gregersen, 1977; Ross and Dobbs, 2011; Wiles et al., 1988). While they represent up to 30% of all coalitions reported from some 20th century Japanese and South African Bantu populations (e.g., Burnett and Case, 2005; Case and Burnett, 2010; Kumai et al., 1996), they are virtually absent from most European and/or American clinical samples and anatomical collections, generally representing less than 2% of documented cases (Burnett and Case, 2005; Case and Burnett, 2010). Only a handful of naviculocuneiform coalitions have been documented on prehistoric human remains from archaeological contexts (e.g., Barnes, 1994; Boshoff and Steyn, 2000; Burnett and Wilczak, *in press*; Burnett and Case, 2005; Regan et al., 1999) and, to the best of our knowledge, none have dated to earlier than 2000 cal. B.P. and none have been recovered from north central Asia. Here we present a case of nearly complete osseous coalition between

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Fig. 1. Left navicular and cuneiform I, dorsal view (distal end up and medial side on the right). Note the small cleft, representing a portion of the normal naviculo-cuneiform I joint space, visible on the lateral (left) side of the coalition. Image: A. Lieverse.

the left navicular and cuneiform I (Figs. 1 and 2) from the early Bronze Age (ca. 5200/5000–4000 cal. BP) cemetery of Khuzhir-Nuge XIV, located on the coast of Lake Baikal, Siberia (Russian Federation).



Fig. 2. Left navicular and cuneiform I, plantar view (distal end up and medial side on the left). Note the small cleft, representing a portion of the normal naviculo-cuneiform I joint space, visible on the lateral (right) side of the coalition. Image: A. Lieverse.



Fig. 3. Burials 57.1 (right) and 57.2 (left) in situ. Image: A. Weber (University of Alberta).

2. Materials

The individual in question, Burial 57.1, is a late adolescent-young adult (18–20 years of age at death) female excavated in 1999 (Fig. 3) from the early Bronze Age (ca. 5200/5000–4000 cal. BP) cemetery of Khuzhir-Nuge XIV. Khuzhir-Nuge XIV, located on the northwest coast of Lake Baikal (Siberia, Russian Federation), was excavated between 1997 and 2001 by joint Canadian-Russian archaeological teams that included the first author. Seventy-four graves yielding 84 individuals were recovered, all but one being radiocarbon dated to the early Bronze Age (Weber et al., 2007). Burial 57.1, in particular, has been directly dated to 4345–3945 cal. BP (Weber et al., 2006). Thirty-six of the 84 individuals recovered exhibited at least one preserved navicular or cuneiform I and, of these, Burial 57.1 was the only one to exhibit a naviculocuneiform I coalition. While one other individual (Burial 21.1) exhibited tarsal coalition, it was the talocalcaneal form (in this case, osseous and bilateral, see Fig. 4) that is generally more common than the naviculocuneiform I variety discussed here (Case and Burnett, 2010; Gregersen, 1977; Stormont and Peterson, 1983; Varner and Michelson, 2000).

The skeleton of Burial 57.1 was incomplete, fragmented, and partially commingled with a second burial (Burial 57.2) interred in the same grave pit, but the integrity of the specimen discussed here and its association with Burial 57.1 is firm. Not only was the postcranial skeleton largely intact and articulated (Fig. 3), but the grave was also excavated—as were all at the cemetery—with the involvement and close supervision of a biological anthropologist (the first author). Burial context, details regarding skeletal preservation, and age at death and sex assessment methodologies are all described by Lieverse et al. (2007). Other than the unusual case of osseous tarsal coalition described here, no other pathological or otherwise noteworthy conditions were observed on this individual.

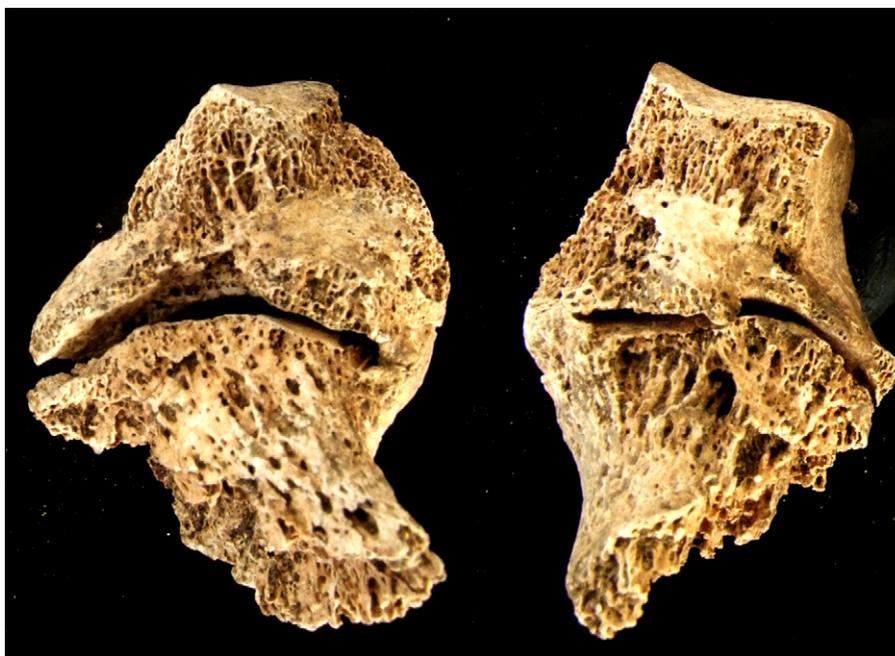


Fig. 4. Bilateral osseous talocalcaneal coalition also documented from Khuzhir-Nuge XIV (Burial 21.1), posterior view of left and right elements (dorsal surfaces up and medial sides adjacent to one another in the center). Note the short bony bridges uniting the medial aspects of the bones, affecting the middle and posterior talar and calcaneal facets. Image: A. Lieveise.

The specimen is an incomplete fragment representing approximately 75% of the left navicular and 50% of the left cuneiform I. Micro-CT imaging (SkyScan 1172; SkyScan, Kontich, Belgium) at a nominal isotropic resolution of 26 μm was performed to enable non-destructive 3D visualization of the internal trabecular architecture of the specimen. The two elements are united across most of the naviculocuneiform I joint space by a continuous bony bridge on its dorsal, medial, and plantar aspects (Figs. 1 and 2). Unfortunately, the extent of the medial union cannot be fully determined due to postmortem breakage on the medial portions of the bones. A definite cleft, representing the normal joint space between the two bones, is observable on the lateral side of the naviculocuneiform I joint, being interrupted only by a bony bridge on its plantar surface (Figs. 5 and 6). Most of the lateral surface of cuneiform I is intact, including much of the articular facet for cuneiform II (or the intermediate cuneiform), suggesting a normal articulation between

these two bones (Fig. 5). That the facets for both cuneiforms II and III (or the intermediate and lateral cuneiforms) on the navicular and for the first metatarsal on cuneiform I are also unremarkable indicates that adjacent bones were not involved in the coalition (Fig. 5).

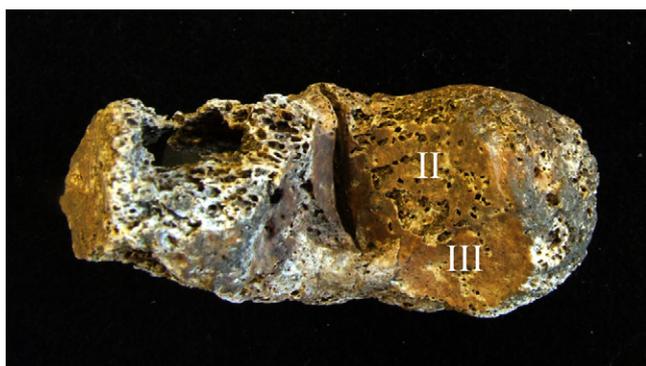


Fig. 5. Left navicular and cuneiform I, distolateral view (dorsal surface up and distal end, representing the cuneiform I-first metatarsal joint, on the left). Note the cleft on the lateral side of the coalition, representing a portion of the normal naviculocuneiform I joint space, as well as the small bony bridge across the plantar surface of the cleft. A portion of the facet for the cuneiform I-cuneiform II joint is located immediately adjacent to (on the left side of) the cleft. On the navicular, the facets for cuneiforms II and III are indicated by Roman numerals (II and III, respectively). Image: A. Lieveise.

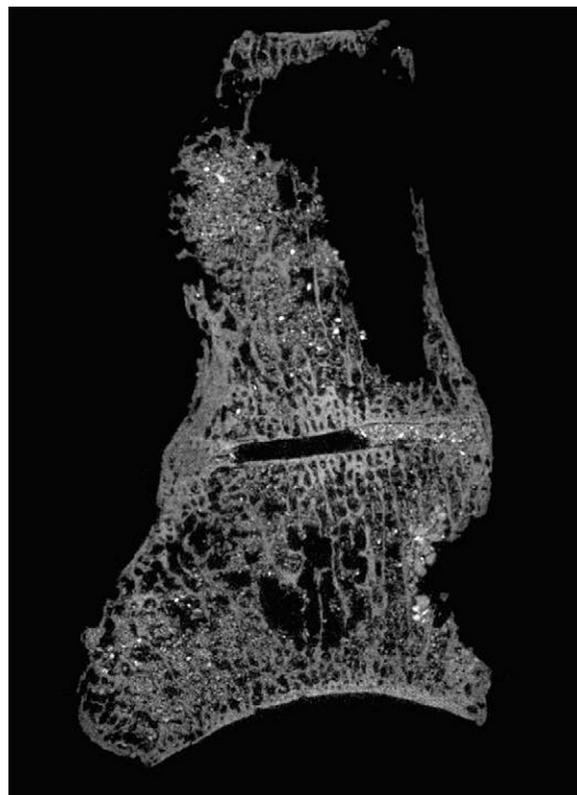


Fig. 6. Micro-CT scan of the left navicular and cuneiform I, sagittal plane (distal end up and dorsal surface on the right). Note the lateral cleft (also depicted in Fig. 5) interrupted by bony bridges on the dorsal (right) and plantar (left) surfaces. Image: D. Cooper.



Fig. 7. Left navicular, proximal view of the normal talar facet (dorsal surface up and medial side on the right). Image: A. Lieverse.

Likewise, the normal appearances of both the navicular's talar facet and its preserved lateral portion (Fig. 7) suggest the absence of any other associated coalitions (e.g., talonavicular and/or calcaneonavicular). Other tarsals recovered from the burial include the left calcaneus and talus and the right talus, all of which were typical in their morphology. Unfortunately, it is impossible to know whether or not this condition was present bilaterally, as no contralateral navicular or cuneiform bones were present.

3. Discussion

Unlike their non-osseous counterparts, osseous tarsal coalitions are, in general, easily identifiable by the presence of bony bridges uniting two normally discrete elements. In the case of naviculocuneiform I coalitions, bony bridges typically span part of the joint space, most often the plantar third or, less commonly, the dorsal third, leaving the unaffected joint surfaces to develop normally (Burnett and Case, 2005). Involvement of all or most of the joint space, such as the case presented here, is highly unusual, even in the clinical literature (e.g., Barnes, 1994; Gregersen, 1977; Marden et al., 2010; Ross and Dobbs, 2011; Wiles et al., 1988). In order to substantiate the congenital basis for osseous coalitions, a detailed differential diagnosis must first exclude other pathological conditions that may also cause ankylosis among one or more tarsal bones.

Several reports of naviculocuneiform coalition presented in the clinical literature (e.g., Gregersen, 1977; Wiles et al., 1988) have suggested that the origin of those cases may have been acquired rather than congenital, possibly secondary to Köhler's disease. Köhler's disease, or osteochondrosis of the tarsal navicular, stems from an early childhood (<10 years) disruption to the bone's blood supply resulting in aseptic necrosis. The condition is rare, with a somewhat obscure etiology (including, possibly, trauma), and affects males five times more frequently than females. While complete recovery can occur with revascularization, especially with proper (modern clinical) treatment, Köhler's disease is typically identified by thinned/flattened, fragmented, biconcave, irregularly ossified, and/or sclerotic (radiologically dense) navicular bones (Aufderheide and Rodríguez-Martín, 1998:86; Ertel and O'Connell, 1984; Ortner, 2003:350; Williams and Cowell, 1981). The absence of any observable navicular deformity in this case makes it unlikely that Köhler's disease was responsible.

As pointed out by Case and Burnett (2010), osseous tarsal coalition may also be misidentified as a complication of degenerative joint disease (DJD), especially if it spans a synovial joint space, as is the case here. Tarsal DJD is not common and, when present, tends

to affect several adjacent elements and articular surfaces rather than one localized joint in the region. DJD is also associated with advancing age at death, being more common on older adults than younger ones, and with past trauma, particularly when occurring in the ankle and/or foot (Aufderheide and Rodríguez-Martín, 1998:95; Ortner, 2003:546–547). This individual's young age at death (18–20 years), the lack of any degenerative joint changes observable elsewhere on the skeleton (including the articular facets adjacent to the coalition site), and the absence of any indicators of past trauma to the left leg or foot all make it exceedingly unlikely that DJD played a role in the condition's etiology.

A number of other pathological conditions, including erosive arthropathies, infections, and traumatic injuries, have been implicated in previous discussions of osseous tarsal coalition (e.g., Case and Burnett, 2010; Chambers, 1950), but none of these seem a likely candidate in the present case. Erosive arthropathies may elicit sporadic osteoblastic responses and terminate with ankylosis, but they are primarily destructive and typically associated with osteoporosis (Albani, 1998:20.1; Ortner, 2003: 561–563; Resnick and Niwayama, 1995:972–977). Likewise, infectious conditions and traumatic injuries resulting in bony fusion typically cause substantial disfigurement to the joints involved (Ortner, 2003: 128–132, 222–223, 230–242). Other than the extensive bony bridge spanning most of the naviculocuneiform I joint in this specimen, there is no evidence of joint destruction or other deformities suggesting an erosive arthropathic, traumatic, or infectious origin for this condition. Thus, macroscopic examination has effectively ruled out an acquired basis for the coalition.

While our argument for the congenital nature of this condition cannot be strengthened by the presence of other congenital anomalies on the skeleton (e.g., see Burnett and Case, 2005; Fopma et al., 2002), it is substantially bolstered by our use of micro-CT imaging. A transverse slice through the navicular and cuneiform I at their point of union reveals trabecular architecture that continues uninterrupted from one bone to the other, and no evidence of cortical or subchondral bone representing the distal extent of one element and/or the proximal extent of the other (Fig. 8). The trabeculae themselves are arranged in a distinct pattern, radiating outwards from the dense compact bone comprising the lateral edge of the coalition. Recent research has demonstrated that the naviculocuneiform I joint is subjected to substantial rotation during locomotion—not unlike that experienced by the talonavicular joint and appreciably more than had been previously thought—so that a point of union between the two bones would likely experience considerable strain in the sagittal, transverse, and frontal planes (Arndt et al., 2007; Ross and Dobbs, 2011). The distinct pattern of radiating trabeculae (Fig. 8) may therefore reflect an *intra vitam* response to mechanical stress across the joint space. Micro-CT imaging demonstrates, unequivocally, the absence of two discrete bones or joint surfaces within the coalition site itself. This, together with the morphologically normal appearance of both elements, strongly supports a congenital basis for this condition.

Although they are uncommon, several cases of isolated naviculocuneiform I coalition have been described in the clinical literature (e.g., Gregersen, 1977; Ross and Dobbs, 2011; Wiles et al., 1988), providing a reasonable proxy from which to extrapolate the extent of possible *intra vitam* complications such as pain and limitations to mobility. Most discomfort associated with tarsal coalition appears to reflect mechanical stress on the affected bones and/or associated postural anomalies among adjacent ones (Fopma et al., 2002; Kumai et al., 1998; Sakellariou and Claridge, 1998). In symptomatic cases of naviculocuneiform I coalition, patients typically report localized pain in the medial midfoot and limitations to normal flexibility (Kumai et al., 1996; Ross and Dobbs, 2011; Wiles et al., 1988). A large proportion (up to 25%) of naviculocuneiform I coalitions may be asymptomatic, especially early in life before

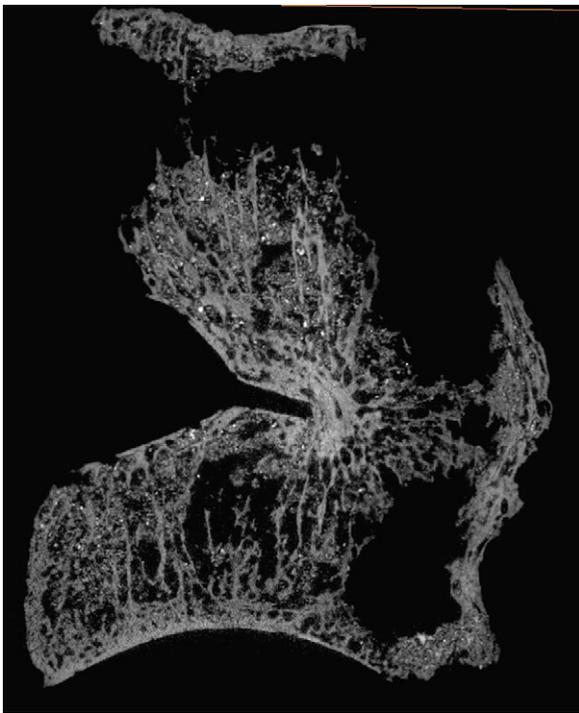


Fig. 8. Micro-CT scan of the left navicular and cuneiform I, transverse plane (distal end up and medial side on the right). Note the continuous trabecular architecture uniting the two elements. Image: D. Cooper.

the development of complications such as degenerative joint disease (Kumai et al., 1996; Ross and Dobbs, 2011), but this is more likely to be the case for non-osseous coalitions than osseous ones (Case and Burnett, 2010; Harris and Beath, 1948; Jayakumar and Cowell, 1977; Kumai et al., 1996; Mosier and Asher, 1984; Sartorius and Resnick, 1985; Snyder et al., 1981; Takakura et al., 1991, 1998; Varner and Michelson, 2000). Considering the young age at death and lack of any degenerative changes on the affected elements, it is certainly possible that this individual suffered no or only minimal symptoms related to the coalition.

4. Conclusions

This is the first documented archaeological case of naviculocuneiform I coalition from north central Asia. It is noteworthy not only because of its great antiquity—with a radiocarbon date of 4345–3945 cal. BP, it is perhaps the oldest case reported in the literature—but also because of the extensive and undoubtedly congenital nature of the union. While exploring the geographical distribution and relative frequencies of coalitions is far beyond the scope of this paper (for detailed discussions, see Burnett and Case, 2005; Burnett and Wilczak, in press; Case and Burnett, 2010), we hope that the presentation of this unique case will contribute to the growing body of literature on tarsal coalition in general and naviculocuneiform I coalition in particular.

Acknowledgements

This specimen was excavated as part of a long-term interdisciplinary research project funded largely by the Social Sciences and Humanities Research Council of Canada (principal investigator: Dr. Andrzej Weber, University of Alberta). We are grateful to all those who participated in the excavations of Khuzhir-Nuge XIV and to our gracious Russian hosts and collaborators. Finally, we wish to thank Dr. Scott Burnett, whose detailed command of the literature

brought the uniqueness of this specimen to our attention in the first place.

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