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Talocalcaneal coalition in the female adult skeleton from the archaeological site of Perlek-Dioksid, Serbia (10th - 12th centuries AD)

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Abstract

In this case report, we will present a case of rare pathological condition, a talocalcaneal coalition, which was recorded in a female adult individual from the archaeological site of Perlek-Dioksid, dated in the period between 10th and 12th centuries AD. During the bioanthropological and paleopathological analyses it was noted the presence of a bilateral bony fusion of the calcaneus and talus of a female adult individual, suggesting an occurrence of talocalcaneal coalition. This was further confirmed by an X-ray examination. Talocalcaneal coalition (TC) occurs when adjacent tarsal bones, the calcaneus and talus, are partially or completely fused. A talocalcaneal coalition becomes symptomatic when, at the age of 12 to 15 years, a pre-existing coalition becomes ossified. In this case, TC was accompanied by degenerative changes of the spine and bilateral femoral neck anteversion. This condition did not require any form of health-related care for this person, although it probably caused occasional pain of varying intensity. This pathological condition is rarely described in bioarchaeological literature and it is, therefore, important to report every recorded case in order to improve our corpus of knowledge regarding such conditions.

Introduction

In this case report, we will present a case of a rare pathological condition, tarsal coalitions, specifically a talocalcaneal coalition, which was recorded in a female adult individual from the archaeological site of Perlek-Dioksid, dated in the period between 10th and 12th centuries AD.

Tarsal coalition is a condition of partial or complete fusion of adjacent tarsal bones (Fopma & Macnicol, 2002). Talocalcaneal coalition (TC) is one of the two most common subtypes, accounting for 45% Of all tarsal coalitions (Cowell & Elener, 1983; Amini et all, 2023). The talocalcaneal coalition was first defined by Emil Zuckerkandl, a Hungarian anatomist, in 1877 and it is believed to be the result of incomplete or faulty segmentation during development (Yun et all, 2015).

Etiologically there are two types of this condition: i) the congenital and ii) the acquired type (Kernbach, 2010). The most common type is congenital, whereby the talocalcaneal coalitions are a consequence of the failure in mesenchymal differentiation and segmentation (Taniguchi et all, 2003). Most of them are autosomal dominant with variable penetrance. The defect occurs when adjacent tarsal bones do not completely separate during the eight weeks of embryonic development (Fopma & Macnicol, 2002). By the first four weeks of the foetal period, the affected bones will retain a small bridge of cartilage joining them together. It appears that these bridges seem to remain mostly cartilaginous during the foetal period and into childhood (Kawashima & Uhthoff, 1990). The less common, acquired type could be a consequence of trauma, degeneration, inflammatory arthritis, or infections (Kernbach, 2010).

All three facets of the joint could be affected, however with the middle facet being the most frequently involved. There are three types of this condition, which depend on a tissue that bridges the two affected bones: i) syndesmosis (when fibrous tissue bridges the two bones); ii) synchondrosis (when cartilaginous tissue bridges the two bones); and iii) synostosis (when bony tissue bridges the two bones) (Crim & Kjeldsberg, 2004). Talocalcaneal coalitions can also be classified, based on the location of the bridges, into the four subtypes i) anterior facet; ii) middle facet; iii) posterior facet; and iv) extra-articular type (Yun et all, 2015). The extra-articular type may occur with or without os sustentaculi (Yun et all, 2015). The main association in the case of middle facet coalitions is pes planus while, in the case of posterior facet coalitions, it is pes cavus (Docquier et all, 2019). In modern clinical practice, the initial treatment consists of non-operative management, while conservative management is successful only in one-third of cases (Kernbach, 2010).

With a normal gait, the joint between the calcaneus and talus (subtalar joint) can be defined as a pronating and supinating joint. With the foot in the stance phase, the leg usually rotated internally. When the foot moves into the toe-off phase, the leg rotates externally as a result of closed chain movement. In a talocalcaneal coalition, this unique miter torque converter movement no longer functions leading to an increase in the level of stress on the neighboring joints, particularly the ankle joint and talonavicular joints. This leads to ligamentous laxity in the ankle joint, with traction spurs being a potential consequence of a break at the talar neck. Because of an accessory facet in a pediatric or young adult rigid flatfoot deformity, there is the potential for peroneal spasm (Martus et all, 2008).

(Paleo)epidemiology

TC is prevalent in modern populations to an extent of 1 to 2%. However, cadaveric and radiological studies of the contemporary populations have suggested a prevalence as high as 13% (Park et all, 2022). A talocalcaneal coalition becomes symptomatic when, at the age of 12 to 15 years, a pre-existing coalition becomes ossified (Schenkel et all, 2010). A recent clinical study revealed that half of the patients with tarsal coalition also suffered from TC, and among those with TC 76% were a bilateral condition. TC is slightly more common in women, while it is most often recorded in the age group of 21 to 40 years (Park et all, 2022). In the archaeological population, the most comprehensive research was conducted by Case and Burnett (2010). This research included 23 studies of tarsal coalition from Prehistoric to Medieval period, of which 11 described only cases of TC (Case & Burnett, 2010). The frequencies of TC are reported for South African, Euro American and Danish cases, with the highest frequency noted in South Africa (0.8%). Aside from the case studies and works published by Case and Burnett (2010), very little information has been published regarding TC of past populations. Therefore, there are no comprehensive insights into paleoepidemiological picture of this condition. Given that this is a relatively rare condition in skeletal assemblages from an archaeological context, the vast majority of cases have been published in the form of a case report. However, this does not diminish the importance of publishing such case studies, as it still affords us some valuable insight into the distribution of this condition by sex and age, regardless of geographical region or period. As far as the authors of this study know, this is the first case study describing this condition in an archaeological assemblage from any period in the territory of modern-day Serbia.

Archaeological background

The archaeological site of Perlek-Dioksid is located in north Serbia, within the province of Vojvodina, in the South Bačka District, near Bečej (Figure 1). The skeleton from the grave No. 1/08, which is the subject of the current study, was orientated W-E, in a supine position with the upper limbs stretched along the body. It was buried in a rectangular grave pit, with no grave architecture (Figure 2). Personal items of the deceased, a needle, iron buckle, and glass beads were found in the grave. Based on the grave goods, this grave is dated into the period between the 10th - 12th centuries CE.

Osteobiography of skeleton from grave No. 01/08

The osteological material from grave 01/08, from the archaeological site of Perlek-Dioksid, in Bečej, was in a good state of preservation, with approximately 70% of

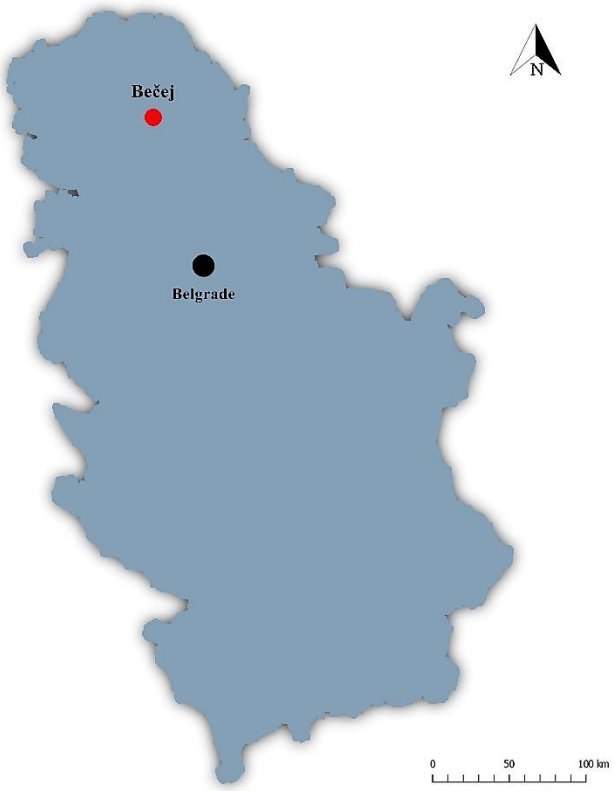

Figure 1. Location of the archaeological site of Perlek - Dioksid

the skeletal material preserved (Mikić, 1978). Almost the entire skeleton is preserved, except for part of the maxilla and several bones of the postcranial skeleton (Figure 3). However, the cortical parts of almost all bones were slightly damaged, porous and brittle. Based on the morphological features of the pubic symphysis, according to the Suchey and Brooks methodology (1990), the age-at-death was estimated at 35.2 ± 9.4 years. The analysed individual was female, as determined by the morphological characteristics of the skull and pelvis (Ferembach et all, 1980; Buikstra & Ubelaker, 1994). The reconstructed stature of the analysed individual is about 146 cm, based on the maximum length of the right femur and right tibia, according to Trotter & Gleser (1958).

The dental material is very well preserved, which enabled a good and precise analysis of the individual's dental status. The dental analysis was done based on the methodology proposed by Brothwell (1981). We noted the ante-mortem loss of two mandibular teeth (second and third mandibular molars from left side), but

Figure 2. Skeleton in situ

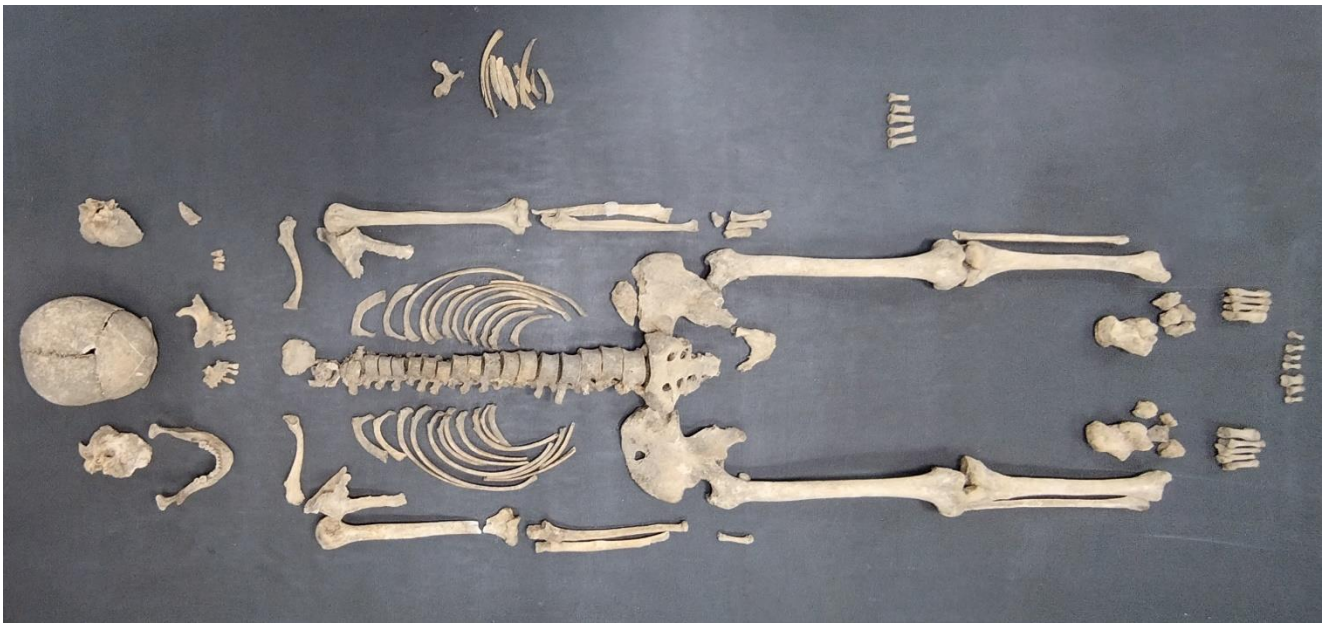

Figure 3. Skeleton during the bioanthropological analyses

also the complete absence of caries or any other dental diseases. Tooth abrasion, of a low degree, is present only on the mandibular incisors.

The analysis of the pathological profile of the individual from grave 01/08 was, at times, difficult due to the damaged cortical surface of the bones. However, based on the gross examination of the skeletal remains, we were able to identify several pathological conditions. The analysis of paleopathological lesions was mainly performed based on Buikstra's recommendation (Buikstra, 2019), however for the differential diagnosis of osseous talocalcaneal coalition we followed recommendation of Case & Burnett, (2010), while for the femoral neck anteversion we mostly followed the recommendations of Djukic et all (2014). A bilateral osseous talocalcaneal coalition was recorded on the skeletal remains. This condition manifests as the complete ossification of both the calcaneus and the

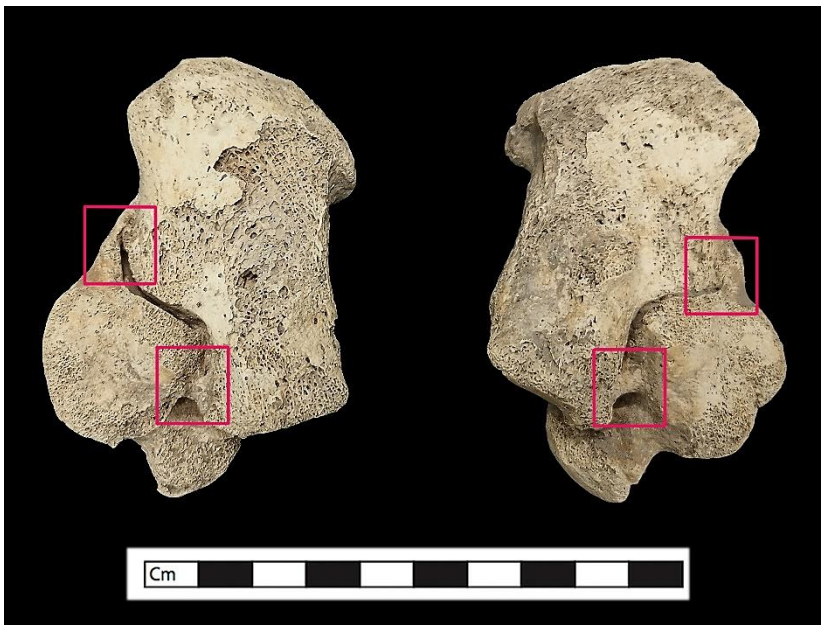

Figure 4. Complete bony fusion of all three facets of both talocalcaneal joints.

talus, with macroscopically visible bony fusions of all three facets of both talocalcaneal joints (Figure 4). Macroscopic changes were recorded and further analysed by way of an X-ray examination. The X-ray scanning was carried out in the Centre for Diagnostic Radiology, Faculty of Dental Medicine, University of Belgrade following standard, well established recommendation (X-ray readings were performed by Petar Milenković). X-ray images showed bilateral narrowing and destruction of the joint surface with visible sclerosis and bony bridges formed between the calcaneus and talus. The Bohler's, tuber-joint, angle was preserved bilaterally, with 33.6 degrees recorded on the left and 37.7 degrees on the right side (Figure 5). Both the macroscopic and X-ray analyses revealed bony synostosis in the region of all three facets of both talocalcaneal joints. The complete union of joint space was present on the lateral sides, while medially partial obliteration of tarsal sinus can be seen. Radiological diagnostics revealed that this condition could be defined as an osseous talocalcaneal coalition.

Given that in this particular case there were no signs of trauma, degeneration, inflammatory arthritis, or infections, we can speculate that this individual suffered from the congenital form of TC. This probably manifested in the early childhood of this individual and, as with most individuals, the bridge between the calcaneus and the talus appears to remain cartilaginous into adulthood. However, as a likely result of activity-related mechanical stress, which would result in microfracture and remodelling, more fibro-cartilaginous tissue may occur (Kumai et all, 1998). It is

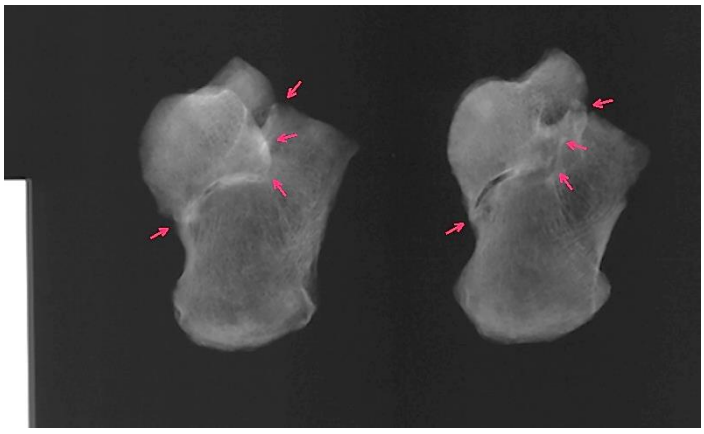

Figure 5. X-ray of both the calcaneus and talus.

macroscopically noted and radiologically revealed that, in this case, there is a bony synostosis in the region of all three facets of both talocalcaneal joints, suggesting that the bridge between the calcaneus and the talus ossified. The condition of tarsal coalition has received much attention in medical literature because of its connection with clinically significant conditions in some patients, such as pain from rigid flatfoot and at the coalition site after an increase in activity, significant limitation of subtalar motion, tarsal tunnel syndrome, and a tendency to injuries such as sprained ankle (Takakura et all, 1991; Varner & Michelson, 2000). Bony changes could potentially be the result of some of these conditions, for example rigid flatfoot (Mosier & Asher, 1984; Spero et all, 1994; Case & Burnett, 2010), which are recognisable in skeletal remains. Therefore, we could assume that this person probably suffered from this condition whole life and the presence of bony bridges in this region may suggest conditions such as rigid flatfoot. This would not have required any form of health-related care for this person, but probably caused occasional pain of varying intensity.

Degenerative changes in the form of osteophytes on the edges of the body of the C3 and L5 vertebrae were noted. Additionally, bilateral femoral neck anteversion (FNA) was recorded (Figure 6). The anteversion angle was reconstructed using Software KVI Popovac, version 2.2. Copyright Leica Imaging Systems, and it is 40.2º. Differential diagnosis (Djukic et all, 2014), suggested that it is bilateral symmetry (level C), with an increased FNA angle on both sides, which indicates that the deformity probably originated in childhood (Djukic et all, 2014). Previous research shows that the FNA angle at birth is about 40 degrees and that it gradually decreases with age, finally reaching about 8 to 15 degrees in adulthood (Fabeck et all, 2002; Fabry et all, 1973; Harkess, 2003; Hefti et all, 2007; Schoenecker & Rich, 2006). Based on paleopathological and clinical studies, as in cases of TC, in the cases with FNA it could be assumed that the individual suffered from a rigid flat foot (Mosier & Asher, 1984; Takakura et all, 1991; Varner & Michelson, 2000; Case & Burnett, 2010). The length of the leg bones and their thickness, in the investigated skeleton is normal. No hip

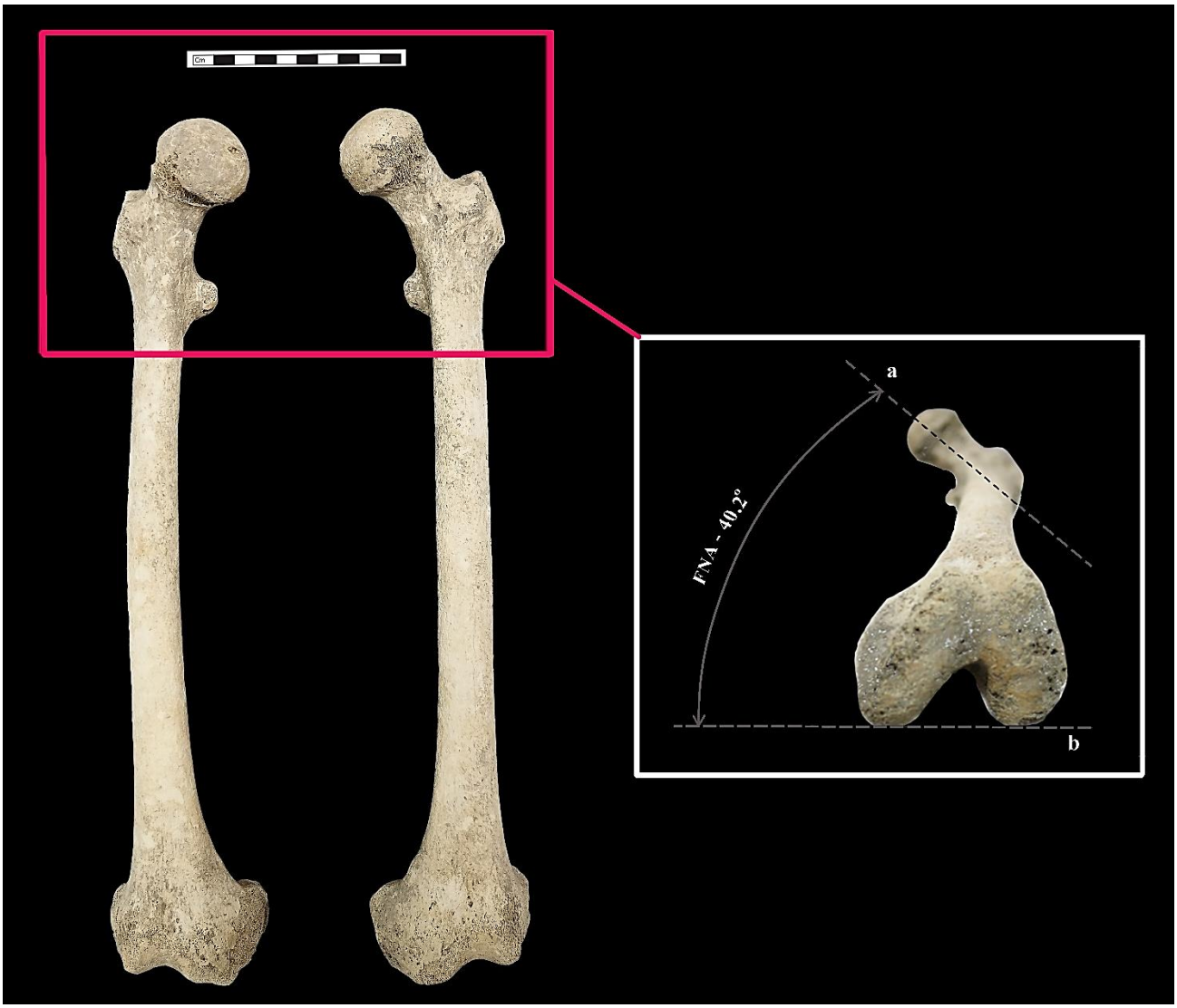

Figure 6. Femoral neck anteversion recorded on the skeleton

subluxation/dislocation, and no hip joint diseases were recorded. However, it is still unclear whether these two pathological conditions (TC and FNA) were connected, or if FNA could be a consequence of a specific gait due to the potential pain caused by TC.

Conclusion

In this study we presented a rare paleopathological congenital defect, which was manifested as talocalcaneal coalition on an adult female individual from the archaeological site of Perlek-Dioksid, dated into the period between the 10th and the 12th centuries CE. Through differential diagnosis and comprehensive paleopathological analysis, it was revealed that the examined individual had impaired movement, probably pain at the coalition site, especially after increased activity, and severe limitation of subtalar motion, with a high predisposition to trauma, such as ankle sprains and osteoarthritis. This person probably suffered from this condition (TC) throughout their whole life, with the presence in this region of bony bridges and FNA suggesting conditions such as rigid flatfoot. This person would not have required every-day health-related care, but would probably have experienced occasional pain of varying intensity. Degenerative changes in the spine, as an accompanying paleopathological condition, was an incidental finding and it is not considered to be directly connected with TC and FNA.

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