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Identification of Tarsal Coalition and Frequency Estimates From Skeletal Samples

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ABSTRACT Tarsal coalition is a congenital defect that results when adjacent tarsals fail to separate properly during embryonic development. Anatomically, coalitions present as non-osseous bridges of cartilage or fibrocartilage – and occasionally as osseous bridges – between two neighboring bones. In skeletons, non-osseous tarsal coalitions are recognizable as matching lesions between two bones at predictable locations. These coalitions are of interest because they are known to be heritable and are therefore useful for tracing genetic relatives in archaeological cemeteries, because they can be misinterpreted in skeletons as trauma or joint disease, and because they can result in associated pathology. However, despite a considerable literature on tarsal coalition, estimates of coalition frequencies disagree considerably, perhaps due to biases inherent in clinical sampling. In order to gain a better estimate of tarsal coalition frequencies in human populations, data were gathered on 342 European-Americans from the Terry Collection (Smithsonian Institution), 536 South Africans from the Dart Collection (University of Witwatersrand, South Africa), and 756 medieval Danish skeletons (Anthropological Database, Odense University). The Danish skeletons are archaeological, with sample sizes by coalition type ranging from 366–507 individuals. Examples of eight different types of intertarsal coalition were identified among the 1634 skeletons examined. Overall frequency estimates for tarsal coalition ranged from 2.1%–3.5%. South Africans exhibited significantly higher frequencies in the midfoot, with naviculocuneiform I coalition (1.0%) the most common type. Conversely, no coalitions of the midfoot were found among the Euro-Americans or medieval Danes. Instead, these groups exhibited calcaneonavicular coalition as the most common type in the hindfoot (2.0% and 2.1% respectively), while calcaneonavicular coalition was among the least common in the South Africans (0.2%). Copyright © 2010 John Wiley & Sons, Ltd.

Key words: African; calcaneocuboid; calcaneus; Denmark; flatfoot; fusion; intercuneiform; talus

Introduction

Tarsal coalitions are highly heritable congenital defects that result when two adjacent tarsals fail to separate completely during the eight weeks of embryonic development (Leonard, 1974; Fopma & Macnicol, 2002). By the first month of the fetal period, affected tarsals will retain a small cartilaginous bridge that joins the two bones together (Trolle, 1948; Kawashima & Uhthoff, 1990). These bridges seem to remain primarily cartilaginous throughout the fetal period and much of childhood (Kawashima & Uhthoff, 1990; Kumai *et al.*, 1998). In a small percentage of cases, the cartilaginous bridge that joins the two tarsals will ossify during childhood or adolescence as the bones continue to grow and mature (Jack, 1954; Person & Lembach, 1985). However, such osseous coalitions (OSCs) are relatively rare, probably accounting for less than 5% of tarsal coalitions overall (e.g. Pfitzner, 1896; Cooperman *et al.*, 2001; Lysack & Fenton, 2004). In most individuals, the bridge appears to remain cartilaginous into adulthood, although it may become more fibrocartilaginous, probably due to activity-related mechanical stress resulting in microfracture and remodelling in the boundary area between the bridge and the bony tissue of the tarsals (Kumai *et al.*, 1998).

The most common types of tarsal coalition occur between the calcaneus and navicular (CN coalition), and between the talus and calcaneus (TC coalition). Many other types of tarsal coalition have been reported previously, but their frequencies tend to be much lower (Pfitzner, 1896; Stormont & Peterson, 1983; Fopma & Macnicol, 2002). Tarsal coalition has received considerable attention in the medical literature because of

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its association with clinically significant conditions in some patients, such as painful rigid flatfoot, pain at the coalition site after increased activity, severe limitation of subtalar motion, tarsal tunnel syndrome and a predisposition to trauma such as ankle sprains (Snyder *et al.*, 1981; Mosier & Asher, 1984; Takakura *et al.*, 1991; Varner & Michelson, 2000). Some of these conditions, such as rigid flatfoot, may result in bony changes that are recognisable in skeletal remains. Tarsal coalition is also occasionally reported in association with more serious congenital disorders such as manual symphalangism and clubfoot (Mosier & Asher, 1984; Spero *et al.*, 1994).

Tarsal coalition has received less attention from physical anthropologists, though apparently not because of its rarity. Estimated frequencies from clinical studies suggest that 1–2% of individuals from many European populations have some type of intertarsal coalition (Pachuda *et al.*, 1990; Kulik & Clanton, 1996; Sakellariou & Claridge, 1998), though affected individuals will often show no ill effects. Results from dissection studies suggest even higher frequencies of 5% or more (Pfitzner, 1896; Rühli *et al.*, 2003; Solomon *et al.*, 2003). The disparity between clinical and anatomical frequencies probably results from the fact that a large proportion of tarsal coalitions are asymptomatic and never come to the attention of medical practitioners (Leonard, 1974; Stormont & Peterson, 1983; Varner & Michelson, 2000). An even greater disparity exists for tarsometatarsal coalition between the third metatarsal and third cuneiform, where frequencies as high as 26% have been reported for some skeletal samples (Regan *et al.*, 1999), despite being hardly mentioned in the clinical literature (although see Day *et al.*, 1994; Stevens & Kolodziej, 2008). However, while tarsometatarsal coalitions are an interesting phenomenon and probably related to intertarsal coalitions aetiologically, they have been well described by Regan *et al.* (1999) and are not addressed here.

Adult individuals with tarsal coalition will exhibit either an osseous or non-osseous bridge between two or more adjacent tarsals within a given foot. OSCs are fairly obvious in dry bone because they cause two normally separate bones to present as a single element, though often with a cleft or partial joint space to indicate where the separation should have occurred (Figure 1). The main diagnostic challenge in cases of OSC is to eliminate other pathological mechanisms, such as trauma or inflammatory arthritis, that can result in tarsal ankylosis as opposed to congenital coalition. Non-osseous coalitions (NOCs) are much more common than OSCs, and much more likely to be present in skeletal samples. In dry bone, the bridge itself will be absent, but evidence of its presence will remain in the form of matching lesions at a very specific location between the two tarsals involved in the coalition (e.g. Figure 2, Supporting Materials fig. 1). These lesions tend to have certain characteristics in common (see 'Results and Discussion') that assist with identification regardless of the bones involved in the coalition.

Figure 1. Bilateral osseous coalition (OSC) of the second and third cuneiforms of a male individual from the Raymond Dart Collection (proximal view). Note the dorsal cleft indicating the proper border between the two bones. This individual was not encountered as part of our study and, therefore, is not included in frequency calculations.

Whether osseous or non-osseous, tarsal coalitions can occur through a joint space, or through some extra-articular point of proximity between two bones. Calcaneocuboid and naviculocuneiform coalitions, for example, occur through a joint between the two bones, but usually involve only a specific part of the joint. The remainder of the joint surface will form normally and usually show no evidence of pathology. Other coalitions, such as calcaneonavicular and third metatarsal–third cuneiform coalitions, are extra-articular, occurring at a point where two bones would normally lie very close together in life (Figures 1–2, Supporting Materials fig. 1). Some coalitions, such as talocalcaneal, are primarily extra-articular, but may involve a joint space as well in some cases (Kawashima & Uhthoff, 1990). Interestingly, some of the extra-articular coalitions occur at exactly the same site where anomalous synovial joints are commonly reported in dissection studies, such as between the calcaneus and navicular, and between the cuboid and navicular (Pfitzner, 1896; Rühli *et al.*, 2003). At least one example of an individual with CN coalition in one foot, and an anomalous synovial joint between the calcaneus and navicular in the other foot, is known (Pfitzner, 1896). Tarsal coalition sites are also frequently located where certain accessory bones are occasionally found (Beckly *et al.*, 1975; Ehrlich, 1982; Palladino *et al.*, 1991). This juxtaposition led Pfitzner (1896) to suggest that incorporation of accessory bones might be part of

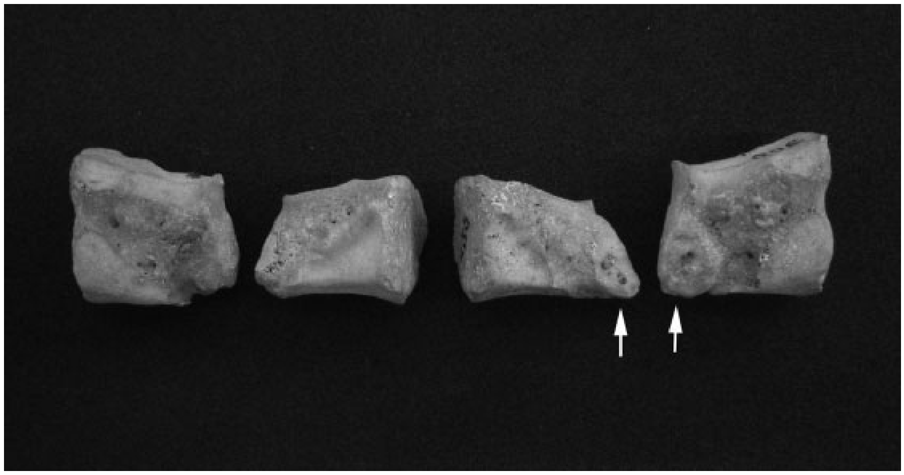

Figure 2. Lateral view of second cuneiforms and medial view of third cuneiforms of specimen A167 from the Raymond Dart Collection. Note the lesions on the plantar aspects of the proximal ends of the right second and third cuneiforms indicative of NOC (arrows). The coalition has affected the morphology of the plantar area by causing additional bone to build up as a base beneath the coalition. Such changes are common in coalitions that occur outside the confines of a joint space. The lesions are smooth at the edges and pitted toward the center, a common pattern seen in many forms of tarsal coalition. Note the small facets present in the same area on the left side.

the aetiology of tarsal coalition, though this idea has since been rejected (e.g. Ehrlich, 1982). The fact that coalitions, anomalous joints and accessory bones often occur at the same anatomical locations in different individuals does suggest that these coalition sites are subject to a greater degree of developmental plasticity than other parts of the tarsus.

Strong evidence exists for the heritability of tarsal coalition (Wray & Herndon, 1963; Leonard, 1974; Ehrlich, 1982; Case, 2003:Table 4.2). Leonard (1974) studied the relatives of 31 patients from Edinburgh who had been treated for rigid flatfoot associated with either CN or TC coalition. Ninety-eight first degree relatives of these index patients were examined to determine whether they exhibited tarsal coalition, and whether the coalitions found in these relatives affected the same bones as those in the index patients. Among the relatives examined, 33% of parents and 47% of siblings exhibited either CN or TC coalition, perhaps indicating autosomal dominant inheritance as suggested by Wray & Herndon (1963). Evidence from other clinical studies involving sibling– and parent–child pairs with identical coalitions indicates that talonavicular and calcaneocuboid coalitions are also heritable defects (Rothberg *et al.*, 1935; Boyd, 1944; Hodgson, 1946; Challis, 1974; Zeide *et al.*, 1977; Pensieri *et al.*, 1985). Other forms of tarsal coalition can also be presumed to be inherited, but because they are likely to cause less dysfunction and because they tend to occur in much lower frequencies, they may simply have never been detected in first degree relatives by clinicians.

Because tarsal coalition has a high apparent heritability, it should be of interest to physical anthropologists for its utility as a marker of genetic relatedness in archaeological cemeteries. As an example, Case (2003) used a relatively high frequency of TC coalition among 108 skeletons from a medieval parish church in Odense, Denmark to argue for a familial relationship among four affected males. Two other males from the site exhibited CN coalition, and may also have been part of this family. Tarsal coalition should also be of interest to paleopathologists interested in trauma and joint pathology, because it is likely to be present in any sizable sample of skeletons from the past, and misidentification of OSCs, or non-osseous lesions, may result in incorrect interpretations. Finally, tarsal coalition should be of interest to paleopathologists in its own right, because it can lead to pain and even dysfunction, and can interfere with proper motion in the foot, causing early development of osteoarthritis. This appears to have been the case in the 9300 year old Kennewick Man skeleton (Case, in press).

Previous anatomic and skeletal studies

The best estimates of tarsal coalition frequencies come from dissection studies, where the different forms of coalition (osseous, cartilaginous and fibrocartilaginous) can be viewed directly and imaging problems associated with radiographs, computed tomography (CT) scans and magnetic resonance imaging (MRI) avoided (Beckly *et al.*, 1975; Emery *et al.*, 1998; Solomon *et al.*, 2003). Dissection studies permit estimation of overall frequencies of tarsal coalition as well as individual frequencies for each tarsal pair. The earliest and still the largest dissection study was carried out by Pfitzner (1896) on German cadavers. Reports of Pfitzner's numbers are somewhat confused in the literature, but tend to cite frequencies by the foot (e.g. Ehrlich & Elmer, 1991; Sarrafian, 1993:102;) rather than by the individual. Frequencies by the individual can be calculated from Pfitzner's raw data, which included 520 feet from 313 individuals. Excluding tarso-metatatarsal coalitions (see Regan *et al.*, 1999 for this information), an overall frequency for intertarsal coalitions in Pfitzner's study was 5.1% (16 coalitions). Nearly all cases identified by Pfitzner were non-osseous.

Since Pfitzner's study in 1896, only one additional anatomical study of all forms of tarsal coalition has been conducted (Rühli *et al.*, 2003; Solomon *et al.*, 2003). This study was carried out in two parts, and involved dissection of the feet of 62 Australian cadavers of European origin. Although the study only found CN and TC coalition, their combined frequency of 12.9% (8/62 individuals) for these two types, coupled with Pfitzner's (1896) frequency of 5.1% for his five types, suggests that clinical estimates of tarsal coalition prevalence are probably too low. The relative frequency of CN to TC coalition in Rühli *et al.*'s (2003) study strongly favoured CN coalitions (six CN, two TC), as was true of Pfitzner's study (11 CN, one TC).

Skeletal studies share some of the advantages of dissection studies, in that all surfaces of each bone can be viewed directly for evidence of OSC or NOC. Skeletal studies also have the advantage of allowing much larger samples to be examined in far less time than would be possible through dissection. Despite these advantages, a search of the literature identified only one large scale skeletal study of tarsal coalition. The study was carried out by Cooperman *et al.* (2001), and focused on CN coalition among 2900+ skeletons from the Hamann–Todd collection in Cleveland, Ohio. Cooperman *et al.* (2001) found a frequency for CN coalition of a little less than 1% (26/2900+ individuals) for their sample of European- and African-Americans, and found only one OSC out of 37 affected feet (2.7%).

Bioarchaeologists have identified a number of individual cases of tarsal coalition among various samples from around the world, including several examples of CN and TC coalition, as well as one or two cases each of naviculocuboid, naviculocuneiform I and intercuneiform II–III coalition (Table 1). These studies have also provided some useful photographs of tarsal coalitions, although they have mostly focused on OSC rather than the more difficult-to-identify NOC. Aside from these case reports, and recent work by the authors of the current study (Case, 2003; Burnett, 2005; Burnett & Case, 2005), little information has been published about tarsal coalition frequencies among past populations.

Table 1. Reported cases of tarsal coalition from archaeological contexts

Site/Location	Time period	Type	Form	Sources
France	Merovingian	Calcaneonavicular	OSC	Dastugue & Metz, 1977
Lerna, Greece	Mid. Bronze Age	Calcaneonavicular	NOC[a]	Angel, 1971
Necropole de Serra da Roupa, Portugal	Late Neolithic	Calcaneonavicular	NOC	Silva, 2005
Hipogeu de Sao Paulo II, Portugal	Chalcolithic	Calcaneonavicular	NOC	Silva, 2005
Larina Le Mollard, France	Medieval	Calcaneonavicular	NOC	Darton, 2007
Sintra, Portugal	Late Roman	Calcaneonavicular	NOC	Silva & Silva, 2010
Dorset, England	Late Roman	Calcaneonavicular	NOC	Dinwiddy, 2009
Almada, Portugal	Late Neolithic	Calcaneonavicular	NOC	Silva, 2010
Tikal, Guatemala	~AD 800–900	Talocalcaneal	OSC	Coe & Broman, 1958
Blain Mound, Ohio	~AD 1000	Talocalcaneal	OSC & NOC	Heiple & Lovejoy, 1969
Caen Saint-Martin, France	AD 800–1000	Talocalcaneal	OSC	Bonzom, 1976
Updown (Kent), England	~AD 600	Talocalcaneal	OSC	Calder & Calder, 1977
Roman Cemetery, Colchester	AD 200–300	Talocalcaneal	?	Birkett, 1980
Notre-Dame, France	?	Talocalcaneal	OSC	Spitery, 1983
Capitaine a Grillon, France	Chalcolithic	Talocalcaneal	OSC	Mahieu, 1984
Kona, Hawaii	Prehistoric	Talocalcaneal	OSC	Han *et al.*, 1986
Dalheim, Germany	~AD 1050	Talocalcaneal	OSC	Hofmann *et al.*, 2010
Coastal Chile	AD 1–500	Talocalcaneal	NOC	Gaytan & Tornow, 2009
Pueblo Bonito, New Mexico	AD 850–1150	Naviculocuneiform I	OSC	Barnes, 1994
Hamilton, South Africa	AD 1216–1267	Naviculocuneiform I	OSC	Boshoff & Steyn, 2000
Fairty Ossuary, Ontario	~AD 1400	Cuneiform II-III	OSC	Anderson, 1963
Kent, England	Iron Age	Naviculocuboid	NOC	Anderson, 1995
Almada, Portugal	Late Neolithic	Naviculocuboid	NOC	Silva, 2010

[a] Recognised as coalition from photograph in publication.

Purpose

Despite a relatively high frequency of tarsal coalition among some populations (Pfitzner, 1896; Rühli *et al.*, 2003), the non-osseous form of coalition has received relatively little attention by physical anthropologists until recently, when there has been a notable increase in documented archaeological cases of CN and TC coalition (e.g. Silva, 2005; Darton, 2007; Hofmann *et al.*, 2009; Dinwiddy, 2009; Silva, 2010; Silva & Silva, 2010). The earlier lack of attention to these coalitions is understandable, given that the morphology of NOC lesions in dry bone is difficult to ascertain from radiographs and other imaging technologies, and that photographs or illustrations of dry bone specimens have only been published for a few types (e.g. Pfitzner, 1896; Dwight, 1907:figs. 48, 65; Heiple & Lovejoy, 1969:figs. 1–3; Cooperman *et al.*, 2001:figs. 1–2; Rühli *et al.*, 2003:fig. 1; Solomon *et al.*, 2003:figs. 3–4; Burnett & Case, 2005:figs. 1–4; Silva, 2005:figs. 1–3). However, because the bias towards symptomatic patients in clinical samples is very difficult to overcome, only skeletal and anatomical studies are likely to provide reliable estimates of tarsal coalition frequencies in different populations, and relative incidences of the various coalition types. The few population studies that have been performed on skeletal and anatomical samples have either focused on a single type of coalition (Cooperman *et al.*, 2001; Burnett & Case, 2005), or have been carried out on relatively small samples of individuals (Pfitzner, 1896; Rühli *et al.*, 2003), making it difficult to obtain an accurate estimate of the overall frequency in various populations, and of the relative incidence of each coalition type. Our experience talking with physical anthropologists about these coalitions suggests that relatively few are aware of, or know how to recognise, these coalitions in skeletal samples. Therefore, the purpose of this study is to (1) describe criteria for identifying the various types of tarsal coalition in skeletal samples, based on 25 years of combined experience studying reported cases in the clinical and anatomical literature, and working to identify these coalitions in skeletal samples, (2) provide better estimates of tarsal coalition frequencies among several samples from different parts of the world and test for geographic differences and (3) test the hypothesis that some types of tarsal coalition exhibit a sex or side bias in light of our new data.

Materials and methods

Criteria for identifying the various types of tarsal coalition were developed over many years of collecting drawings, photographs, and written descriptions from the medical and anatomical literature, and of studying suspected cases in skeletal samples. To assist with recognising non-osseous lesions versus those caused by other kinds of pathology, we examined examples of NOC from several other locations in the skeleton where cartilaginous or fibrocartilaginous tissue is sometimes known to abnormally connect two skeletal elements, such as between the third metatarsal and the third cuneiform (Supporting Materials fig. 2), between the acromion process of the scapula and the os acromiale accessory bone (Supporting Materials fig. 3), between the navicular tuberosity and the accessory navicular bone (Supporting Materials fig. 4), and between the third metacarpal styloid process and the os styloideum (Supporting Materials fig. 5). The purpose of these observations was to assess the range of variation that can be found in lesions left behind by cartilaginous or fibrocartilaginous bridging between bony elements.

In many cases, we relied on clinical or anatomical descriptions of tarsal coalitions to pinpoint the expected location of the bridge between a given pair of tarsals, then looked for individuals exhibiting lesions where non-osseous tarsal coalitions are known to exist and compared them to what we knew from these other parts of the skeleton as well as from other tarsal coalitions. These endeavours were occasionally aided by bilateral cases in which OSC was found on one side of the body, and a non-osseous lesion was present on the other. Once we felt confident in our ability to recognise NOCs, we could begin to assess variation in the lesions specific to a particular type of tarsal coalition by studying bilateral cases in which the non-osseous lesions were somewhat asymmetrical in their expression, such as when the lesion was large on one side, and smaller on the other, or when it exhibited pitting on one side, but only a simple smooth depression on the other.

For the purposes of frequency calculation and assessment of sex bias and laterality, one archaeological and two anatomical collections were surveyed for all types of intertarsal coalition (Table 2). The first sample consisted of 342 skeletons (171 female, 171 male) from the Robert J. Terry Anatomical Collection housed at the National Museum of Natural History. This sample represents individuals of European ancestry, most of who died in the St. Louis area during the first half of the 20th Century (Hunt & Albanese, 2005). The second anatomical sample consisted of 536 skeletons (238 females, 298 males) from the Raymond Dart Collection of Human Skeletons housed at the University of Witwatersrand in Johannesburg, South

Table 2. Tarsal coalition frequencies by coalition type

Sample	South Africans		Terry Euro-Am		Medieval Danes[a] (Population estimate)	
Coalition type	Freq. (%)	*N*	Freq. (%)	*N*	Freq. (%)	*N*
Calcaneonavicular	0.2	531	2.1	341	2.2	502
Talocalcaneal	0.8	532	0.0	342	0.6	507
Calcaneocuboid	0.4	532	0.0	341	0.0	467
Talonavicular	0.0	533	0.0	342	0.0	504
Naviculocuboid	0.0	529	0.0	341	0.2	*444*
Naviculocuneiform I	1.0	527[b]	0.0	341[b]	0.0	*433*[b]
Intercuneiform I-II	0.6	527	0.0	341	0.0	*387*
Intercuneiform II-III	0.2	524	0.0	341	0.0	*366*
Cuneocuboid	0.2	528	0.0	341	0.0	*393*
Naviculocuneiform II	0.0	525	0.0	341	0.0	*385*
Naviculocuneiform III	0.0	526	0.0	341	0.0	*395*

[a] Italicised numbers are estimated. All faces of all bones were examined for evidence of tarsal coalition, but inability to score these bones for certain forms of tarsal coalition was not noted when a particular coalition type was thought to be rare. The number of scorable individuals was estimated based on the presence of the relevant bone as well as the ability to score other traits on that bone. Because the criteria for entry were strict, the reported values are probably underestimates of the number of scorable bones that were actually present.

[b] These sample sizes are smaller than those reported in Burnett & Case (2005). Some of the difference is due to the older age minimum used in the present study, because some of the tarsals complete ossification rather late. In addition, some forms of coalition were not systematically examined in all of the South African and Terry skeletons. Those that were not examined for all types of coalition were excluded from this study. Finally, the St. Alban parish cemetery was removed from the Danish sample because the high frequency of talocalcaneal coalition in this sample likely results from several individuals within the sample being genetically related (see Case, 2003).

Africa. This sample represents individuals from Bantu speaking tribes in and around the Guateng province.

The archaeological sample is a group of skeletons excavated from several medieval church cemeteries in Denmark (Table 3). This sample is comprised of skeletons from nine different cemeteries on the large island of Funen and on the Jutland Penninsula. The data come from a larger, unpublished study of skeletal defects (Case, 2003). Our purpose in using the sample was to obtain an estimate of the population frequency of tarsal coalition among Danes from the medieval period (AD 1000–1536) using a sampling of skeletons from sites around the country.

Skeletons from the known-age anatomical samples were limited to individuals aged 13 or older, to insure that tarsal development would be sufficiently advanced for tarsal coalition to be visible. Because the Danish skeletons were from an archaeological context, individuals were considered to be old enough for the study if they had reached a level of skeletal maturity beyond the point where the tarsals should be fully ossified. To be included in this study, skeletons were required to exhibit one or more of the following: (1) fully fused digital epiphyses of the hands or feet, (2) a fused or fusing iliac crest, (3) a fused or fusing medial clavicle or (4) an age estimate based on the pubic symphysis, the auricular surface or both suggesting a minimum age greater than 30 years. Indicators 1–3 were sufficient for all but a few skeletons in the sample.

Because the Danish skeletons are not as well preserved as those from the anatomical samples, there is a fairly high degree of missing data. Some of the individuals from each cemetery had few or no observable tarsals, while others had most or all of them present. In order to tabulate frequencies for each coalition type, all skeletons with both bones of a given tarsal pair in at least one foot were scored for tarsal coalition. If only a single bone was present, the tarsal pair was treated as absent, since many coalitions require both bones for a certain diagnosis of coalition. Sample sizes for each tarsal coalition type varied considerably, from a low of 366 to a high of 507 (Table 2). In order to estimate the overall frequency of tarsal coalition for this sample, it was necessary to find a way to combine the results from the individual tarsal coalition types, as there were not enough skeletons with all tarsal pairs observable to provide a reliable estimate. When studying rare traits, smaller sample sizes tend to

Table 3. Description of Danish sites and proportion of excavated skeletons examined

Site name	Site type	No. provenienced individuals	City	Region	No. examined	Site %
Gray Friar	Urban monastery	362	Odense	Funen	317	88
Black Friar	Urban monastery	800	Odense	Funen	125	16
St. Jørgen	Leper hospital	1127	Odense	Funen	124	11
St. Knud	Urban monastery	175	Odense	Funen	35	20
Franciskaner	Urban monastery	152	Svendborg	Funen	15	10
Tirup	Rural parish	569	Bygholm	Jutland	50	9
Nordby	Rural parish	130	Viby	Jutland	25	19
St. Mikkel	Urban parish	300	Viborg	Jutland	59	20
Frederiksgade	Urban monastery	40	Aarhus	Jutland	6	15
Total		3655			756	

inflate apparent frequencies when at least one example of the trait is found in the sample, so use of the very lowest sample size ($n = 366$) to calculate frequencies would likely have overestimated the population frequency for tarsal coalition. Conversely, if we had used the largest sample size from among the individual type estimates ($n = 507$) it is likely that we would have underestimated the true frequency of tarsal coalition among the medieval Danes. We chose instead to calculate a median sample size from the samples for each individual tarsal coalition type to use in our calculation of the overall frequency. This median sample size should minimise our probability of seriously underestimating or overestimating the true frequency of tarsal coalition. Therefore, our sample size for the overall tarsal coalition frequency calculation was 433 individuals.

The two anatomical samples allow fairly confident assessment of overall tarsal coalition frequencies, laterality, sex bias and relative frequencies of the various types of coalition in their populations, because they have very few missing or damaged bones, and their sexes are known from mortuary records. The archaeological sample is a good representative of medieval Danes from Funen and Jutland, and can be used to better understand frequency variation within this northern European group.

All frequency comparisons were made using Fisher's Exact test. Two-tailed probabilities were used to test the null hypothesis of equal coalition frequencies among populations. All statistical tests were conducted at an α level of 0.05. Statistical testing was performed using web-based software written by Uitenbroek (2000).

Results and discussion

A total of eight different coalition types were found in the South African, Terry and Danish samples. Frequencies for each of these types are reported in Table 2. A single coalition type is sometimes referred to by more than one name in the literature such as cubonavicular and naviculocuboid coalition. We have chosen to use a simple naming convention here, in which the more proximal bone is named before the more distal, and the more medial bone before the more lateral. In most of the cases, this convention results in the most common name being used, except perhaps for naviculocuboid and cuneocuboid coalition.

The distribution of the eight types of coalition identified in our study was found to be non-random within the samples. For example, the South Africans exhibited seven different types of coalition spread throughout the foot, while the medieval Danes exhibited three types and the Terry sample exhibited only one type, with 21/22 coalitions involving either the calcaneus or talus in the latter two samples.

There were significant differences in the frequencies of individual types of coalition between the South African sample and the Danish and Terry samples. The most obvious difference is found in the frequencies of CN coalition. This coalition is the most common type among the samples of European ancestry, with frequencies of 2.1% in the Terry sample and 2.2% among the medieval Danish skeletons. These results mirror those of Pfitzner (1896) who found CN coalition to be the most common type by far in his German sample, with a frequency of 3.5%. Among the South Africans, however, CN coalition is among the least common types of tarsal coalition, accounting for only 1/17 cases and a sample frequency of 0.2%. The difference in frequency between the South African and both the Terry ($p = 0.007$) and Danish ($p = 0.003$) samples is statistically significant. Another difference that stands out between the European and South African samples involves coalition between the navicular and first cuneiform. Five examples were found among the South Africans, for a frequency of 1.0%, while none were found in any of the European samples. A significant difference between the South Africans and medieval Danes for this coalition has been reported previously (Burnett & Case, 2005).

A broader pattern evident in the data is a tendency for tarsal coalitions to cluster in the hindfoot among the two European samples, while they are more broadly distributed among the South Africans, and indeed seem to be more common in the midfoot than the hindfoot. For the purposes of this discussion, hindfoot coalitions are defined as those involving the calcaneus, talus and the proximal halves of the navicular and cuboid. These would include calcaneonavicular, talocalcaneal, calcaneocuboid, talonavicular and naviculocuboid coalitions, which seem to be the coalitions that are more clinically symptomatic. Midfoot coalitions are defined as intertarsal coalitions involving the three cuneiform bones. These would include all three types of naviculocuneiform coalition, both types of intercuneiform coalition, and cuneocuboid coalitions.

When midfoot and hindfoot frequencies are compared among the different samples, some of the differences are statistically significant. Frequencies of tarsal coalition in the hindfoot are: South Africans (1.3%), Terry (2.1%) and Danes (3.0%). In the midfoot, these frequencies are reversed: South Africans (1.9%), Terry (0.0%) and Danes (0.0%). Differences in the hindfoot between the South Africans and the two samples of European ancestry do not reach statistical significance ($p = 0.419$ for the Terry comparison, and $p = 0.082$ for the Danish comparison).

In the midfoot, however, the South Africans show a significantly higher frequency than either the Terry sample ($p = 0.008$) or the Danish sample ($p = 0.006$). These results, as well as those outlined above for CN coalition, seem to indicate that hindfoot coalitions are more dispersed in the South Africans than in Europeans, though the overall frequency of hindfoot coalitions is essentially the same. In the midfoot, coalitions are again dispersed but clearly much more common in the South African sample.

Despite these differences in the locations of tarsal coalition within the foot, overall frequencies of tarsal coalition appear to be essentially the same among the three samples. Among the South Africans, intertarsal coalition affected, approximately, 3.2% of the sample (Table 2). This result does not differ significantly from that found for either the Terry Collection (2.1%, $p = 0.398$), or the medieval Danish sample (3.5%, $p = 0.858$). When these data are considered together with those reported by Pfitzner (1896), who found an overall frequency of 5.1% among his German sample, it would not be unreasonable to conclude that intertarsal coalition probably affects between 2 and 5% of most populations, with even higher frequencies possible in some cases (e.g. Rühli *et al.*, 2003). This is a substantially higher frequency than the 1–2% estimated from clinical research, and probably reflects a fuller reporting of individuals whose coalitions would not have been symptomatic in life.

The relative frequency of each type of coalition in groups of European descent can be estimated by combining the data from the various samples described here, as well as the samples studied by Pfitzner (1896) and Rühli *et al*. (2003). The results are presented in Table 4. These data indicate that about 75% of tarsal coalitions in populations of European descent are likely to be calcaneonavicular, followed by talocalcaneal at perhaps 10–15% and naviculocuboid at approximately 5–10%. The rest make up less than 5% of the total number of coalitions. These numbers are quite different from those found in the South African sample.

Sex bias

Table 5 lists the number of individuals of each sex who were found to be affected by each type of tarsal coalition when the various samples used in this study were combined. Both the total number of coalitions by sex, and the individual reports for each coalition type, tend to suggest parity among the sexes in tarsal coalition. The only type that stands out as possibly exhibiting a difference between the sexes is naviculocuneiform I coalition, with five males and no females affected in our samples. However, a recent study by Burnett & Case (2005) that included a slightly larger sample from the South African collection for naviculocuneiform I coalition found that even this type of coalition does not appear to exhibit a statistically significant sex bias ($p = 0.087$).

Most clinical studies of tarsal coalition report either equal incidence for both sexes, or a predilection for males (Leonard, 1974; Kulik & Clanton, 1996; Fopma & Macnicol, 2002; Lysack & Fenton, 2004). Conway & Cowell (1969), for example, reported an equal number of males and females for CN coalition, while for TC coalition males outnumbered females by four to one (16 males, four females). However, sex bias is another area in which clinical reliance on patients presenting with foot problems may influence the findings, because these studies rely on patient reports of pain which may in turn be affected by the types of activities engaged in.

Table 4. Relative frequencies of tarsal coalition by sample and ancestry

Sample	Terry	Danish	Pfitzner	Ruhli *et al.*	Total European		South African	
Coalition type	*N*	*N*	*N*	*N*	*N*	%	*N*	%
Calcaneonavicular	7	11	11	6	35	76.0	1	5.8
Talocalcaneal	0	3	1	2	6	13.0	4	23.5
Naviculocuboid	0	1	2	0	3	6.5	0	0.0
Talonavicular	0	0	1	0	1	2.2	0	0.0
Calcaneocuboid	0	0	0	0	0	0.0	2	11.8
Naviculocuneiform I	0	0	0	0	0	0.0	5	29.4
Intercuneiform I–II	0	0	0	0	0	0.0	3	17.6
Intercuneiform II–III	0	0	1	0	1	2.2	1	5.8
Cuneocuboid	0	0	0	0	0	0.0	1	5.8
Naviculocuneiform II	0	0	0	0	0	0.0	0	0.0
Naviculocuneiform III	0	0	0	0	0	0.0	0	0.0
Totals	7	15	16	8	46		17	

Table 5. Tarsal coalitions from this study by sex, side, laterality, and form

Coalition Type	Sample	Male	Female	Left	Right	Bilat	Unilat	Laterality?	Osseous[a]	Non-Oss[a]
CN Coalition	Danish	5	6	8	7	4	3	4	0	15
CN Coalition	Terry	4	3	6	4	3	4	0	0	10
CN Coalition	South African	1	0	1	0	0	1	0	0	1
TC Coalition	Danish	1	2	2	2	1	2	0	1	3
TC Coalition	South African	2	2	3	4	3	1	0	0	7
Calc-Cuboid	South African	1	1	2	1	1	0	1	0	3
Nav-Cuboid	Danish	0	1	1	0	0	1	0	0	1
Nav-Cun I	South African	5	0	1	5	1	4	0	0	6
InterCun I–II	South African	2	1	3	2	2	1	0	0	3
Intercun II–III	South African	1	0	0	1	0	1	0	0	1
Cuneocuboid	South African	0	1	1	0	0	1	0	0	1
Totals		22	17	28	26	15	19	5	1	51

[a] By coalition, not individual.

Most review articles on coalitions involving the calcaneus and talus agree that pain and dysfunction come from mechanical stress and interference with normal biomechanics of the subtalar joint (Kulik & Clanton, 1996; Sakellariou & Claridge, 1998; Fopma & Macnicol, 2002). Pain does not usually occur until the area around the coalition site has ossified. Some children and adolescents will experience pain as the coalition site itself begins to ossify, but for individuals with NOC, pain often only occurs after a change in lifestyle that leads to new physical demands, such as increased activity or prolonged standing, or after a traumatic event such as an ankle sprain (Rankin & Baker, 1974; Kulik & Clanton, 1996; Sakellariou & Claridge, 1998). Many people with tarsal coalition never become symptomatic, and some who have a symptomatic coalition in one foot will have an asymptomatic one in the other (Jack, 1954; Kumar *et al.*, 1992; Sakellariou & Claridge, 1998). Therefore, sexual dimorphism in body mass, and differences in activity level or intensity between males and females could lead to an over-representation of affected males in clinical samples.

Previously reported dissection- and skeletal-studies tend to agree with the findings of our research, that there does not appear to be a sex bias in overall tarsal coalition frequencies (Pfitzner, 1896; Rühli *et al.*, 2003). Based on results from Pfitzner's study on all types of intertarsal coalition, 6.5% of females (6/92) and 4.4% of males (9/203) exhibited some type of intertarsal coalition (Table 5). These percentages might seem to suggest a predilection for females, rather than males, but the difference is not statistically significant ($p = 0.568$, $\alpha = 0.05$). Limiting analysis to the study of CN coalition by Cooperman *et al.* (2001), frequencies for males were 0.9% and for females were 0.8%, which is also not a significant difference ($p > 0.999$).

Laterality

Our data do not indicate any significant side bias for tarsal coalition (Table 5) and they seem to suggest that coalitions are unilateral (57% of cases) slightly more often than they are bilateral (43% of cases). For CN coalition, which is the most common type in our combined samples, bilateral expression is found in 47% of cases. These results accord well with other clinical and anatomical studies.

Clinical studies based on properly positioned radiographs, MRI and CT scans have suggested that TC and CN coalition are frequently bilateral in their expression. In a recent review of six clinical studies by Fopma & Macnicol (2002), CN coalition was found to be bilateral in 25–57% of cases. A midpoint estimate of this range would be 41%. TC coalition was found to be bilateral in 30–64% of five clinical studies, with a midpoint estimate of 47%. Laterality estimates based on previous dissection and skeletal studies suggest a similar degree of bilaterality. A relatively unbiased estimate of laterality for CN coalitions can be assessed by combining the studies by Pfitzner (1896), Cooperman *et al.* (2001) and Rühli *et al.* (2003). These studies identified a combined 41 cases of CN coalition that could be assessed for laterality, of which 18 (44%) were bilateral. Taken together, clinical and anatomical studies seem to suggest that tarsal coalition, particularly those types affecting the calcaneus, tend to be bilateral in approximately 40–50% of affected individuals.

Skeletal identification of tarsal coalition

One of the primary challenges in studying tarsal coalition is accurate identification of NOCs versus other forms of pathology. Clinicians use various imaging techniques, including radiographs, CT scans and MRI, to diagnose tarsal coalition. Some of these observations, although not made on dry bones, are useful in understanding the morphology of NOC lesions in skeletal remains. Clinicians tend to divide NOC lesions into fibrous and cartilaginous forms, although most adult coalitions are probably a mixture of these two tissue types (Kumai *et al.*, 1998). The more fibrous coalitions are harder to detect with imaging technology, because they cause less change to the apposed surfaces of the two bones than do cartilaginous coalitions (Kumar *et al.*, 1992; Wechsler *et al.*, 1994). According to clinicians, cartilaginous coalitions are characterised by cystic joint irregularity or lytic-like lesions, while fibrous coalitions show more subtle narrowing and subchondral sclerosis (Hynes & Romash, 1987; Wechsler *et al.*, 1994). Sometimes, the cystic lesions will contain dense nodular bone formations as well (Hynes & Romash, 1987).

NOCs can also result in changes to the shape of the apposed surfaces (Newman & Newberg, 2000; Lysack & Fenton, 2004). A good example of such changes as seen in a CT scan is depicted in Hochman & Reed (2000: fig. 1). Minor expression on CT scans may appear as joint space narrowing with poor definition of the articulating facets and minimal reactive bone changes along the margins, while cortical hyperostosis, irregularity, and perhaps abnormal angulation of the involved facets may be the more typical expression (Warren *et al.*, 1990; Newman & Newberg, 2000). These clinical observations suggest that in dry bone, NOC lesions should exhibit slight to extensive disruption of the cortical surface, sometimes including lytic-like lesions and occasionally nodular bone formation within the lesions themselves. Skeletal research on tarsometatarsal coalition suggests that cartilaginous coalitions often include vascular canals within the floor of the lesion (Regan *et al.*, 1999), through which blood and nutrients are delivered to cartilaginous bridge.

Three additional criteria can assist with the identification of NOC lesions in dry bone. The first is the location of the lesion. NOC lesions are nearly always found at a very specific location on each affected bone. For example, talocalcaneal lesions are usually centred on the posterior part of the sustentaculum tali, and have only rarely been reported at the posterior or anterior facets of the calcaneus (Beckly *et al.*, 1975; Kawashima & Uhthoff, 1990; Cohen *et al.*, 1996). Coalitions between the cuneiforms and adjacent bones tend to occur between the plantar one-third of the two bones, whether it be a naviculocuneiform I coalition (Burnett & Case, 2005), an intercuneiform coalition (Pfitzner, 1896, and reproduced in Sarrafian, 1993:fig. 2–80) or cuneometatarsal III coalition (Regan *et al.*, 1999). The size of the lesion may vary from case to case, but its location will remain constant in almost all affected individuals.

Second, a NOC lesion should normally be visible on both bones involved in the coalition. This fact helps to differentiate tarsal coalition lesions from similar-appearing lesions associated with osteochondritis dissecans (see for example Anderson, 2001:fig. 1). There should also be some degree of symmetry between the sizes of the matching lesions on the apposed bones. The lesions do not necessarily have to be equally deep or disruptive of the cortical surface, but their general length and width should match fairly well.

Finally, as noted by clinicians, some types of tarsal coalition result in a change in the shape or contour of one or both bones involved in the coalition. The best example of such change occurs along the plantar edge of the navicular in cases of CN coalition (Figure 3). Perhaps because this particular type of coalition is extra-articular, the navicular nearly always

Figure 3. Right and left CN coalition in an individual from medieval Denmark. Note the absence of an anterior facet on the calcaneus, the oblique angle that is formed between the calcaneus and navicular, the additional bone formation along the plantar aspect of the naviculars and the considerable difference in the morphology of the lesions on the right and left sides. The difference in lesion morphology may reflect differences in the amount of fibrocartilage making up the bridge on each side. Substantial pitting in the lesion on the left may indicate vascular channels and a higher proportion of cartilage in the bridge. The rim of bone on the left side probably results from greater mobility through the coalition on the left compared to the right.

has additional bone development along its plantar surface within which the NOC lesion is embedded.

In our experience, physical anthropologists occasionally confuse tarsal coalition with degenerative joint disease (DJD), particularly in coalitions involving articular facets. This is not surprising since DJD may also involve cystic lesions and joint surface or margin irregularity. Of course, DJD may be a co-occurring condition with coalitions, either independent of or secondary to mechanical complications arising from tarsal coalition. Nonetheless, we believe that DJD can be reliably excluded as a potential singular diagnosis in most cases of tarsal coalition for several reasons. First, DJD in the tarsus is infrequent and typically occurs as a result of trauma (Aufderheide & Rodriguez-Martin, 1998), which is unlikely to be bilateral. Second, the tarsus forms a complex set of interrelated elements in which DJD is unlikely to occur at a very localised site without accompanying signs of DJD on adjacent facets and tarsals. Finally, DJD, in general, is most likely to affect middle-aged to older adults, whereas tarsal coalition is present at birth and usually becomes recognisable in skeletal specimens by the end of adolescence. As a result, arthritis rates within a sample should increase with age while coalition rates should remain constant. Therefore, despite some rough similarities in appearance, we believe that tarsal coalition can be distinguished from DJD, particularly in cases involving younger individuals, an absence of trauma, bilateral lesions and/or highly localised joint surface lesions or irregularity. When the locational specificity of most articular coalition lesions is considered as an additional diagnostic clue, the probability of mistaking DJD for tarsal coalition would seem to be acceptably low.

Calcaneonavicular coalition

This is one of the easiest NOCs to identify in dry bone because of substantial changes to the inferior surface of the navicular and the anterior beak of the calcaneus (Figure 3). Effects on the navicular are most dramatic, and it should be possible to diagnose this particular form of coalition from the navicular alone. In place of a gently curving inferior surface, the affected navicular typically exhibits excess bone formation and a fairly linear, rather than the more normal convex, inferior edge. Some degree of pitting of the inferior surface is also common, probably in cases where the coalition is made up of primarily cartilaginous tissue. In some individuals, the inferior surface of the lesion may be roughened but not pitted. These roughened surfaces may exhibit the nodular formations described by clinicians using imaging technology. The less pitted lesions probably indicate coalitions composed of primarily fibrous rather than cartilaginous tissue. See Supporting Materials figs. 6 and 7 for examples of these different lesion types.

The affected calcaneus will typically exhibit an anterior border that is much more sharply angled posteriorly towards the medial side than is true for a normal calcaneus. Often the lesion will present a linear face at an angle of 30° or more from a line perpendicular to the long axis of the calcaneus (Figure 3, Supporting Materials figs. 6–7). There is typically no evidence of an anterior facet on the sustentaculum tali in affected calcanei, nor any space available on which one might have formed. Presumably, the segment of bone that would normally underlie the anterior facet becomes instead part of the inferomedial edge of the navicular during tarsal differentiation. The lesion along this sharply angled anterior border of the calcaneus will match in size that on the navicular and will have a similarly roughened or pitted appearance. Some affected calcanei exhibit an additional rounded extension of bone at the extreme lateral edge of the lesion (Supporting Materials fig. 7). These may be secondary centres of ossification that would normally form the beak along the inferior edge of the navicular, but which cannot form there because of the coalition (although see Supporting Materials fig. 8). Radiologists refer to the extension of the calcaneus as the 'anteater nose sign' when seen on lateral radiographs of patients with CN coalition (Chapman, 2007). A drawing of an osseous example of CN coalition can be found in Conway & Cowell (1969:fig. 1).

Talocalcaneal coalition

This coalition is somewhat more difficult to identify than CN coalition. TC coalition almost always presents as a bridge between the medial side of the talus, just anterior to the large posterior facet, and the posterior part of the sustentaculum tali, immediately adjacent to the posterior edge of the middle facet (see Sarrafian, 1993:fig. 2–74, for a drawing from Pfitzner's dissection study, and Figures 4–7, Supporting Materials figs. 9–10 from this study). The remaining joint surfaces usually form normally. When non-osseous TC coalition is present, there will often exist a small shelf of bone stretching from the posterior part of the sustentaculum tali posterodistally to the body of the calcaneus. It is on this shelf that the lesion will usually be visible (Figures 4–5). Sometimes the lesion will encroach upon the posterior edge of the middle calcaneal facet, but often that facet will be unaffected (Figure 5). These lesions may present as smooth

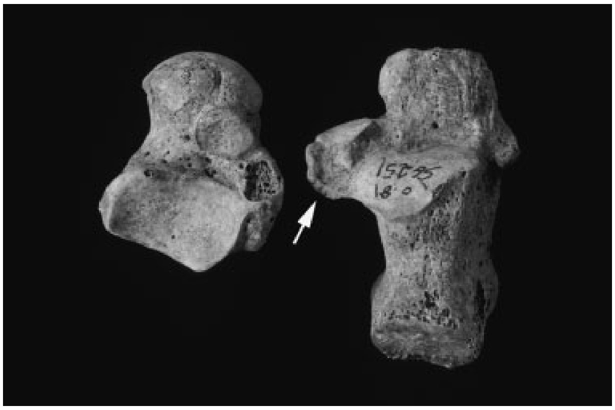

Figure 4. Example of TC coalition from the right foot of a medieval Danish skeleton. Note the smooth depression on the posterior part of the sustentaculum tali of the calcaneus, and the deep lesion on the matching aspect of the inferior talus, which resembles those often found between the third metatarsal and cuneiform. Note also the similar size of the two lesions. The depression on the calcaneus has only slightly impinged on the middle calcaneal facet area. The majority of the lesion is in the extra-articular space posterior to the middle facet.

depressions with little or no pitting, or as open lesions with a pitted floor, reminiscent of the NOC lesions found between the third metatarsal and third cuneiform (Regan *et al.*, 1999). In some cases, however, TC coalition may present as very large lesions that can alter the entire posterior facet area, presumably in response to abnormal motion at the coalition site (Figure 6). These large lesions resemble NOC lesions that we have occasionally seen between the medial tuberosity of the navicular and the accessory navicular bone. However,

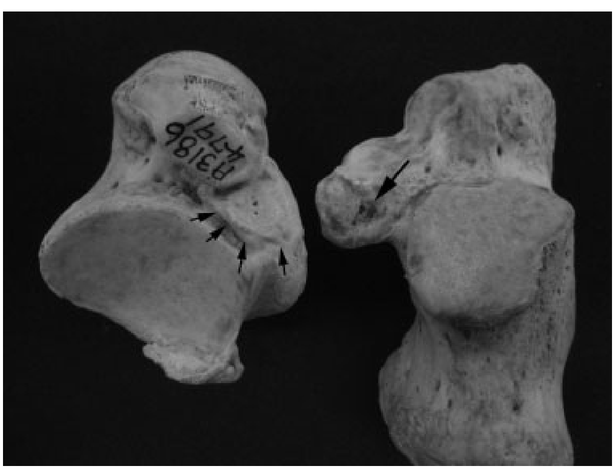

Figure 5. Example of TC coalition from the right foot of a South African individual from the Raymond Dart Collection. Note the slight rim around the lesion on the talus and the mild pitting at its center. Note also the slight rim around the lesion on the calcaneus, but the greater degree of pitting at the lesion's posterolateral corner.

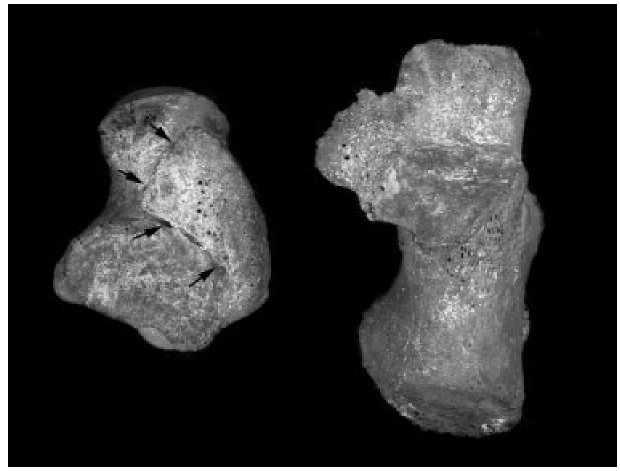

Figure 6. Example of TC coalition from the right foot of an individual from the Spitalfields Collection, Museum of Natural History, London. This case shows considerable alteration to the posterior part of the sustentaculum tali including absence of the entire middle facet. At first glance, this might appear to be the kind of destruction that would come from some traumatic event. However, the talus does not exhibit similar destruction. Rather, there appears to be excess bone along the inferior part of the talus that might represent part of the sustentaculum tali which may have been incorporated into the talus. Additionally, the talus and calcaneus exhibit pitting within their respective lesions as is commonly seen in NOCs. Note also that the posterior and anterior facets on the calcaneus are mostly unaffected.

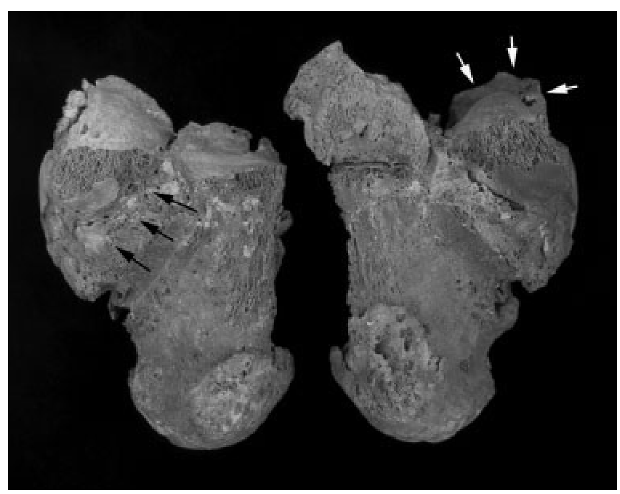

Figure 7. Medial view of bilateral osseous talocalcaneal coalition in an individual from the Terry Collection. This individual was not randomly encountered and, therefore, is not included in frequency calculations. The medial view shows an osseous bridge that involves nearly the entire sustentaculum tali on the left side, while the coalition on the right is restricted to the more typical posterior sustentaculum tali. Note also the effect of abnormal motion caused by the coalition on the morphology of the talar head.

even in cases of OSC, the remainder of the joint surfaces between the calcaneus and talus are usually unaffected by the coalition (Figure 7, Supporting Materials fig. 10; Hofmann *et al*., 2009). In rare cases, the coalition may occur at the margin of the posterior facet of the calcaneus, as was true for one of our South African skeletons (Supporting Materials fig. 11).

Talonavicular coalition

Clinical literature suggests that talonavicular coalition is the third most common type of intertarsal coalition (Feliu, 1991; Fopma & Macnicol, 2002). Dissection and skeletal studies (Table 4) suggest that it is quite rare. The difference in reported frequency may relate to the significant effect that talonavicular coalitions can have on a person's gait (Pontious *et al*., 1993), and therefore the likelihood that it will be noticed and diagnosed in the living. It may also relate to the fact that talonavicular coalitions are visible on standard radiographic views (Gold, 1971) and, therefore, might be more commonly discovered as an incidental finding than some other types. The authors have never seen this type of coalition in our skeletal research, despite having looked at the foot skeletons of perhaps 2000+ individuals. Clinicians have only reported OSCs as far as we can determine (e.g. Hodgson, 1946; Gill & Sullivan, 1985; Person & Lembach, 1985; Sarrafian, 1993; Frost & Fagan, 1995), and these should be readily recognisable. Pfitzner may be the only author to have reported a non-osseous talonavicular coalition (see Sarrafian, 1993:fig. 2–77 for an illustration). Unfortunately, his figure does not show the character of the non-osseous lesion itself.

Naviculocuboid coalition

Naviculocuboid coalition is extra-articular, making it easier to identify in dry bone because it can cause substantial change to the shape of each bone involved in the coalition, and will exhibit matching lesions at locations where the two bones would not normally be in contact. The point of connection on the navicular is through the plantar beak (Pfitzner, 1896:fig. 26a–b; Dwight, 1907:fig. 66; Johnson *et al*., 2005). In the one case that we have seen, the beak was substantially larger than normal, with a very large open pit embedded on the lateral side (Figure 8). The size of this plantar beak is similar to that seen when an accessory bone called the secondary cuboid fuses at this same location (see Dwight, 1907:fig. 58). The apposing lesion on the cuboid is located on the superomedial side of the bone where the cuboid beak should be located, and at the point where the two bones would normally lie closest to each other. As with CN coalition, it is possible that the styloid process of the cuboid has developed as part of the plantar surface of the navicular in affected individuals, explaining the abnormal mass of bone located there (Figure 8).

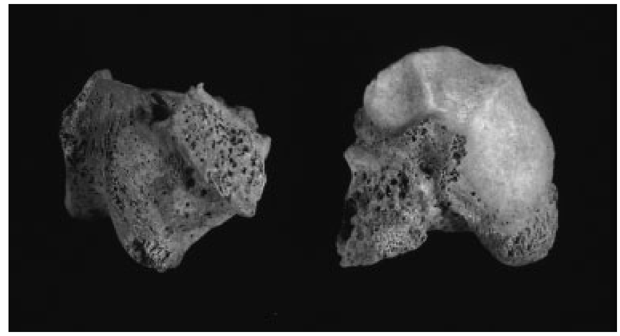

Figure 8. Naviculocuboid NOC in the right foot of a medieval Danish skeleton. Note the abnormal size of the navicular beak and the partial absence of the cuboid styloid process at the location of the NOC lesion.

Calcaneocuboid coalition

This type of coalition appears to be relatively rare, although this impression may be due to the fact that it usually does not cause dysfunction (Conway & Cowell, 1969; Craig & Goldberg 1977). Pfitzner (1896) did not find any cases in his sample of 313 individuals, and a search of the literature produced only 15 clinical examples, several of which were associated with other abnormalities (Conway & Cowell, 1969; Pensieri *et al*., 1985; Sarrafian, 1993; Frost & Fagan, 1995). However, the authors have seen three individuals with this kind of coalition in skeletal material. One was an osseous example in which the individual had both bilateral talocalcaneal and unilateral calcaneocuboid OSC (Figure 7, Supporting Materials fig. 10). Two examples of calcaneocuboid NOC were found in the Dart Collection from South Africa, one was bilateral (Figure 9) and the other was of unknown laterality because the other side was unobservable. The location of the lesion on the calcaneus is a small area on the medial aspect of the cuboid facet in both cases. The cuboid lesion is on the lateral side of the cuboid beak and the medial side of the cuboid's calcaneal facet.

Naviculocuneiform coalitions

Coalition between the navicular and first cuneiform appears to be rare in individuals of European ancestry, but significantly more common in native South African populations, and possibly Japanese populations as well (Kumai *et al*., 1998; Burnett & Case, 2005). This type of

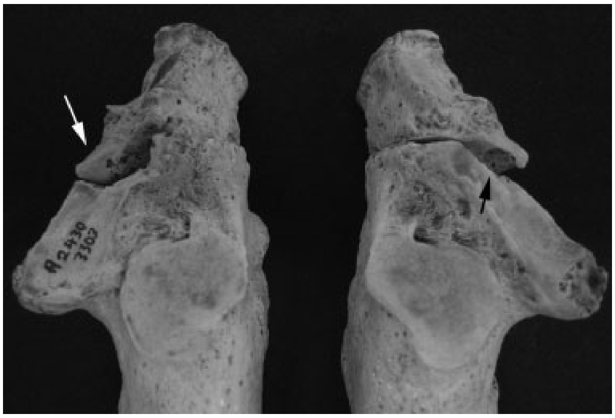

Figure 9. Bilateral NOC in a skeleton from South Africa. Note the location of the lesion on the medial side of the cuboid styloid process. The left calcaneus (on right side of photo) also exhibits a NOC-like lesion of the posterior part of the stustentaculum tali. However, there was no matching lesion on the talus, which would normally rule out tarsal coalition. It is possible that this lesion instead represents NOC between the sustentaculum tali and the os sustentaculi accessory bone.

coalition normally occurs between the plantar part of the navicular facet on the first cuneiform, and the plantar part of the first cuneiform facet on the navicular (Figure 10), although at least one example of coalition between the superior ends of each bone is known (Burnett & Case, 2005:fig. 4). Very few isolated cases of coalition between the navicular and the other two cuneiforms have been reported (Burnett & Case, 2005).

Cuneocuboid coalition

This coalition is apparently extremely rare. We were only able to find one report of this type of coalition, in a child who also exhibited talonavicular coalition

Figure 10. Naviculocuneiform I NOC in the right foot of a skeleton from South Africa. Note the plantar location of the lesion in both cases, and the presence of nodular bone in the lesion on the navicular. Reprinted from Burnett and Case (2005), with permission from Elsevier.

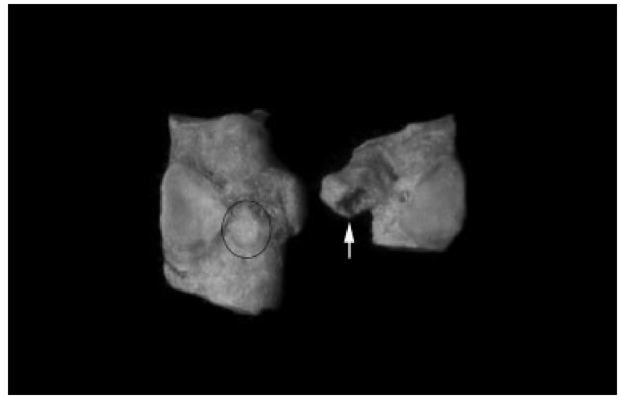

Figure 11. Cubo-cuneiform III NOC in the left foot of a skeleton from South Africa. Note the bony extension on the plantar surface of the third cuneiform. The associated lesion on the cuboid is circled.

(Person & Lembach, 1985). However, we did find one example in our South African sample (Figure 11), suggesting that the condition may be more common than clinicians realise. The coalition is located between the plantar aspects of the two bones, connecting the medial edge of the cuboid tuberosity to the lateral aspect of the third cuneiform.

Intercuneiform coalitions

There are two types of intercuneiform coalition, intercuneiform I–II coalition, and intercuneiform II–III coalition. Both the clinical and anatomical literature suggests that these coalitions are extremely rare. We were only able to locate a single reported case of coalition between the second and third cuneiforms (Pfitzner, 1896:fig. 74, reproduced in Sarrafian, 1993: fig. 2–80), and none between the first and second cuneiforms. This lack of reporting is surprising, given that we identified four intercuneiform coalitions in our South African sample: three intercuneiform I–II (Supporting Materials fig. 12), and one intercuneiform II–III (Figures 1–2). It may be that intercuneiform coalitions are simply overlooked by clinicians, as are the very common cuneometatarsal III coalitions (Stevens & Kolodziej, 2008; Regan *et al.*, 1999) that occur in the same part of the foot. Intercuneiform coalitions tend to occur between the plantar one-third to one-half of the two adjacent bones, although one of our osseous cases exhibits coalition above the halfway mark (Figures 1–2).

Pathological effects of tarsal coalition

Tarsal coalition is the most common cause of a rare, but painful form of flatfoot called 'peroneal spastic flatfoot' or 'rigid flatfoot' (Slomann, 1921; Harris & Beath, 1948; Harris, 1965; Jayakumar & Cowell, 1977; Page, 1987). Rigid flatfoot is characterised by tightness of the peroneal muscles, pain in the space between the talus and calcaneus and limited subtalar motion (Jayakumar & Cowell, 1977; Mosier & Asher, 1984). Tarsal coalition can affect the peroneal muscles by placing strain on them through interference with normal joint biomechanics, resulting in spasm of the peroneus longus and brevis, and in severe cases, the extensor digitorum longus and peroneus tertius may also be involved (Harris, 1965; Mosier & Asher, 1984). Muscle spasm may be intermittent, with pain usually initiated by activity and relieved by rest, or continuous but with varying severity depending on the stresses acting on the foot (Harris, 1965). Spasm of the anterior and posterior tibialis muscles is also occasionally reported (Harris, 1965; Conway & Cowell, 1969).

The likelihood of developing rigid flatfoot as a consequence of coalition depends on the bones involved in the coalition, and the degree to which the coalition restricts normal motion. The vast majority of individuals with tarsal coalition will not develop rigid flatfoot. Stormont & Peterson (1983) identified only two cases in a sample of 54 patients with TC or CN coalition. It appears that the osseous form of TC coalition is by far the most likely type to lead to rigid flatfoot, because it virtually eliminates subtalar motion (Webster & Roberts, 1951; Harris, 1965). CN coalition is a less common cause because it generally has less effect on subtalar motion, although the larger and more complete the bridge between the two bones, the greater the restriction on movement (Jack, 1954). Rigid flatfoot is only rarely reported in association with the more distally positioned tarsal coalitions (Harris, 1965; Mosier & Asher 1984), presumably because the effect on normal motion between the talus and calcaneus is small.

Conclusion

In this study, we have described general criteria for identifying non-osseous intertarsal coalitions in skeletal samples, as well as some specific criteria for identifying particular coalition types. Using these criteria on three skeletal collections representing European Americans, native South Africans and medieval Danes, and combining the results with those from the dissection studies of Pfitzner (1896) and Rühli *et al*. (2003), we estimate that frequencies of tarsal coalition probably range between 2 and 5% or higher in most populations. This frequency is substantially greater than the 1–2% commonly reported by clinicians, and suggests that tarsal coalitions are common enough that they should be part of the fundamental knowledge base of human osteologists and paleopathologists.

This study also indicates that differences may exist in the relative frequencies of various tarsal coalitions among different populations. In our samples, individuals of European ancestry had the highest frequency of tarsal coalition between the calcaneus and navicular with over 2% of both samples affected. Other coalitions were much less common in both European samples, with intertarsal coalitions of the midfoot, affecting the three cuneiforms and their neighbours, completely absent. In contrast, CN coalition was among the least common types among the South African skeletons, with only a single case identified among 531 individuals. Instead, tarsal coalitions of the midfoot were significantly more common among the South Africans than among either of the samples of European ancestry, with naviculocuneiform I coalition exhibiting the highest frequency of all types identified among the South Africans.

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References

Anderson JE. 1963. *The People of Fairty: An Osteological Analysis of an Iroquois Ossuary*. Canada Department of Northern Affairs and National Resources: Ottawa.

Anderson T. 1995. The human skeletons. In *Iron Age Burials from Mill Hill*, Dean PK (ed.). British Museum Press: London; 114–144.

Anderson T. 2001. An example of unhealed osteochondritis dissecans of the medial cuneiform. *International Journal of Osteoarchaeology* **11**: 381–384.

Angel JL. 1971. *The People of Lerna: Analysis of a Prehistoric Aegean Population*. Smithsonian Institution Press: Washington, DC.

Aufderheide AC, Rodriguez-Martin C. 1998. *The Cambridge Encyclopedia of Human Paleopathology*. Cambridge University Press: Cambridge.

Barnes E. 1994. Polydactyly in the Southwest. *Kiva* **59**: 419–431.

Beckly DE, Anderson PW, Pedegana LR. 1975. The radiology of the subtalar joint with special reference to talo-calcaneal coalition. *Clinical Radiology* **26**: 333–341.

Birkett DA. 1980. Talo-calcaneal bridging in the town of Old King Cole. *3rd European Congress of the Paleopathology Association*, Caen; 117–119.

Bonzom Y. 1976. Pathologie de quelques populations anciennes de Basse-Normandie. *Thése de Doctorat en Médecine*, Université de Caen.

Boshoff W, Steyn M. 2000. A human grave from the farm Hamilton in the Limpopo River Valley (South Africa). *South African Journal of Field Archaeology* **9**: 68–74.

Boyd HB. 1944. Congenital talonavicular synostosis. *Journal of Bone and Joint Surgery* **26B**: 682–686.

Burnett SE. 2005. Developmental Variation in South African Bantu: Variant Co-occurrence and Skeletal Asymmetry. *Ph.D. Dissertation*, School of Human Evolution and Social Change, Arizona State University.

Burnett SE, Case DT. 2005. Naviculo-cuneiform I coalition: Evidence of significant differences in tarsal coalition frequency. *The Foot* **15**: 80–85.

Calder A, Calder J. 1977. Talo-calcaneal bridging in an Anglo-Saxon. *Medical History* **21**: 316–319.

Case DT. 2003. Who's related to whom? Skeletal kinship analysis in medieval Danish cemeteries. *Ph.D. Dissertation*, Department of Anthropology, Arizona State University.

Case DT. (in press). Bones of the hands and feet. In *Kennewick Man: Scientific Analysis of a Paleo-American Skeleton*, Owsley D, Jantz R (eds). Texas A&M Press: College Station.

Challis J. 1974. Hereditary transmission of talonavicular *coalition* in association with anomaly of the little finger. *Journal of Bone and Joint Surgery* **56**: 1273–1276.

Chapman VM. 2007. The anteater nose sign. *Radiology* **245**: 604–605.

Coe WR, Broman VL. 1958. *Excavations in Stella 23 Area*. University of Pennsylvania Museum Monographs. Tikal Report No. 25

Cohen BE, Davis WH, Anderson RB. 1996. Success of calcaneonavicular coalition resection in the adult population. *Foot and Ankle International* **17**: 569–572.

Conway JJ, Cowell HR. 1969. Tarsal coalition: Clinical significance and roentgenographic demonstration. *Radiology* **92**: 799–811.

Cooperman DR, Janke BE, Gilmore A, Latimer BM, Brinker MR, Thompson GH. 2001. A three-dimensional study of calcaneonavicular tarsal coalitions. *Journal of Pediatric Orthopaedics* **21**: 648–651.

Craig CL, Goldberg MJ. 1977. Calcaneocuboid coalition in Crouzon's syndrome (craniofacial dysostosis): Report of a case and review of the literature. *Journal of Bone and Joint Surgery* **59A**: 826–827.

Darton Y. 2007. Flatfoot: The palaeopathological diagnosis. *International Journal of Osteoarchaeology* **17**: 286–298.

Dastugue J, Metz F. 1977. Bloc calcaneo-naviculaire bilatéral sur un squelette Mérovingien. *Journal de medecine de Caen* **12**: 137–140.

Day FN, Naples JJ, White J. 1994. Metatarsocuneiform coalition. *Journal of the American Podiatric Medical Association* **84**: 197–199.

Dinwiddy KE. 2009. *A Late Roman Cemetery at Little Keep, Dorchester, Dorset*. Wessex Archaeology: Salisbury.

Dwight T. 1907. *Variations of the Bones of the Hands and Feet*. J.B. Lippincott Company: Philadelphia.

Ehrlich MG. 1982. Tarsal coalition. In *Disorders of the Foot*, Jahss M (ed.). W.B. Saunders Company: Philadelphia; 521–538.

Ehrlich MG, Elmer EB. 1991. Tarsal coalition. In *Disorders of the Foot and Ankle: Medical and Surgical Management* (Second edition). Saunders: Philadelphia.

Emery KH, Bisset GS, Johnson ND, Nunan PJ. 1998. Tarsal coalition: A blinded comparison of MRI and CT. *Pediatric Radiology* **28**: 612–616.

Feliu C. 1991. Cubonavicular synostosis: A case report. *Acta Orthopaedica Belgica* **57**: 306–308.

Fopma E, Macnicol MF. 2002. Tarsal coalition. *Current Orthopaedics* **16**: 65–73.

Frost RA, Fagan JP. 1995. Bilateral talonavicular and calcaneocuboid joint coalition. *Journal of the American Podiatric Medical Association* **85**: 339–341.

Gaytan E, Tornow MA. 2009. A case of bilateral auditory exostosis from the Pacific coast of central-south Chile. *Presented at the 78th Annual Meeting of the American Association of Physical Anthropologists*, Chicago, IL.

Gill PW, Sullivan RW. 1985. Talonavicular coalitions. *Journal of the American Podiatric Medical Association* **75**: 443–445.

Gold GS. 1971. Tarsal coalitions: Clinical significance, diagnosis, and treatment. *Journal of the American Podiatry Association* **61**: 409–422.

Han TL, Collins SL, Clark SD, Garland A. 1986. *Moe Kau a Ho'oilo: Hawaiian Mortuary Practices at Keopu, Kona, Hawaii*. Bernice Pauahi Bishop Museum: Honolulu.

Harris RI. 1965. Retrospect: Peroneal spastic flat foot (rigid valgus foot). *Journal of Bone and Joint Surgery* **47A**: 1657–1667.

Harris RI, Beath T. 1948. Etiology of peroneal spastic flat foot. *Journal of Bone and Joint Surgery* **30B**: 624–634.

Heiple KG, Lovejoy CO. 1969. The antiquity of tarsal coalition: Bilateral deformity in a pre-Columbian Indian skeleton. *Journal of Bone and Joint Surgery* **51A**: 979–983.

Hochman M, Reed MH. 2000. Features of calcaneonavicular coalition on coronal computed tomography. *Skeletal Radiology* **49**: 409–412.

Hodgson FG. 1946. Talonavicular synostosis. *Southern Medical Journal* **39**: 940–941.
Hofmann MI, Böni T, Rühli FJ. 2009. Osseous talocalcaneal coalition in a medieval skeleton (ca. 1050 AD). *International Journal of Osteoarchaeology* DOI: 10.1002/oa.1080
Hunt DR, Albanese J. 2005. History and demographic composition of the Robert J. Terry anatomical collection. *American Journal of Physical Anthropology* **127**: 406–417.
Hynes RA, Romash MM. 1987. Bilateral symmetrical synchondrosis of navicular first cuneiform joint presenting as a lytic lesion. *Foot and Ankle* **8**: 164–168.
Jack EA. 1954. Bone anomalies of the tarsus in relation to peroneal spastic flat foot. *Journal of Bone and Joint Surgery* **36B**: 530–542.
Jayakumar S, Cowell HR. 1977. Rigid flatfoot. *Clinical Orthopaedics and Related Research* **122**: 77–84.
Johnson TR, Mizel MS, Temple HT. 2005. Cuboid-navicular tarsal coalition—presentation and treatment: A case report and review of the literature. *Foot and Ankle International* **26**: 264–266.
Kawashima T, Uhthoff HK. 1990. Prenatal development around the sustentaculum tali and its relation to talocalcaneal coalitions. *Journal of Pediatric Orthopaedics* **10**: 238–243.
Kulik SA, Clanton TO. 1996. Tarsal coalition. *Foot and Ankle International* **17**: 286–296.
Kumai T, Takakura Y, Akiyama K, Higashiyama I, Tamai S. 1998. Histopathological study of nonosseous tarsal coalition. *Foot and Ankle International* **19**: 525–531.
Kumar SJ, Guille JT, Lee MS, Couto JC. 1992. Osseous and nonosseous coalition of the middle facet of the talocalcaneal joint. *Journal of Bone and Joint Surgery* **74A**: 529–535.
Leonard MA. 1974. The inheritance of tarsal coalition and its relationship to spastic flat foot. *Journal of Bone and Joint Surgery* **56B**: 520–526.
Lysack JT, Fenton PV. 2004. Variations in calcaneonavicular morphology demonstrated with radiography. *Radiology* **230**: 493–497.
Mahieu E. 1984. Les fusions tarsiennes: un exemple de pont talo-calcanéen à l'Hypogée du Capitaine. *Bulletin et Mémoires de la Societé d'Anthropologie de Paris t. 1 série* **XIV**: 289–296.
Mosier KM, Asher M. 1984. Tarsal coalitions and peroneal spastic flatfoot. *Journal of Bone and Joint Surgery* **66A**: 976–984.
Newman JS, Newberg AH. 2000. Congenital tarsal coalition: Multimodality evaluation with emphasis on CT and MR imaging. *RadioGraphics* **20**: 321–332.
Pachuda NM, Lasday SD, Jay RM. 1990. Tarsal coalition: Etiology, diagnosis, and treatment. *Journal of Foot Surgery* **29**: 474–488.
Page JC. 1987. Peroneal spastic flatfoot and tarsal coalitions. *Journal of the American Podiatric Medical Association* **77**: 29–34.
Palladino SJ, Schiller L, Johnson JD. 1991. Cubonavicular coalition. *Journal of the American Podiatric Medical Association* **81**: 262–266.
Pensieri SL, Jay RM, Shoenhaus HD, Perlman M. 1985. Bilateral congenital calcaneocuboid synostosis and subtalar joint coalition. *Journal of the American Podiatric Medical Association* **75**: 406–410.
Person V, Lembach L. 1985. Six cases of tarsal coalition in children aged 4 to 12 years. *Journal of the American Podiatric Medical Association* **75**: 320–323.
Pfitzner W. 1896. Beiträge zur Kenntnis des Menschlichen Extremitätenskelets: VII. Die Variationen in Aufbau des Fusskelets. In *Morphologische Arbeiten*, Schwalbe G (ed.). Gustav Fischer: Jena; 245–527.
Pontious J, Hillstrom HJ, Monahan T, Connelly S. 1993. Talonavicular coalition: Objective gait analysis. *Journal of the American Podiatric Medical Association* **83**: 379–385.
Rankin EA, Baker GI. 1974. Rigid flatfoot in the young adult. *Clinical Orthopaedics and Related Research* **104**: 244–248.
Regan M, Case DT, Cleaves-Brundige J. 1999. Articular surface defects in the third metatarsal and third cuneiform: Nonosseous tarsal coalition. *American Journal of Physical Anthropology* **109**: 53–66.
Rothberg AS, Feldman JW, Schuster OF. 1935. Congenital fusion of astragalus and scaphoid: Bilateral, inherited. *New York State Journal of Medicine* **35**: 29–31.
Rühli FJ, Solomon LB, Henneberg M. 2003. High prevalence of tarsal coalitions and tarsal joint variants in a recent cadaver sample and its possible significance. *Clinical Anatomy* **16**: 411–415.
Sakellariou A, Claridge RJ. 1998. Tarsal Coalition: Aetiology, diagnosis, and treatment. *Foot and Ankle* **12**: 135–142.
Sarrafian SK. 1993. *Anatomy of the Foot and Ankle: Descriptive, Topographic, Functional*. J.B. Lippincott: Philadelphia.
Silva AM. 2005. Non-osseous calcaneonavicular coalition in the Portuguese prehistoric population: Report of two cases. *International Journal of Osteoarchaeology* **15**: 449–453.
Silva AM. 2010. Foot anomalies in the Late Neolithic/Chalcolithic population exhumed from the Rock Cut Cave of Sao Paulo 2 (Almada, Portugal). *International Journal of Osteoarchaeology* DOI: 10.1002/oa.1148
Silva AM, Silva AL. 2010. Unilateral non-osseous calcaneonavicular coalition: Report of a Portuguese archeological case. *Anthropological Science* **118**: 61–64.
Slomann HC. 1921. On the demonstration and analysis of calcaneonavicular coalition by roentgen examination. *Acta Radiologica* **5**: 304–312.
Snyder RB, Lipscomb AB, Johnston RK. 1981. The relationship of tarsal coalitions to ankle sprains in athletes. *American Journal of Sports Medicine* **9**: 313–317.
Solomon LB, Rühli FJ, Taylor J, Ferris L, Pope R, Henneberg M. 2003. A dissection and computer tomography study of tarsal coalitions in 100 cadaver feet. *Journal of Orthopaedic Research* **21**: 352–358.
Spero CR, Simon GS, Tornetta P III. 1994. Clubfoot and tarsal coalition. *Journal of Pediatric Orthopaedics* **14**: 372–376.
Spitery E. 1983. *La Paleontologie des Maladies Osseuses Constitutionnelles. Paleoecologie de L'Homme Fossile No. 6* Editions du Centre National de la Recherche Scientifique: Paris.

Stevens BW, Kolodziej P. 2008. Non-osseous tarsal coalition of the lateral cuneiform-third metatarsal joint. *Foot and Ankle International* **29**: 867–870.

Stormont DM, Peterson HA. 1983. The relative incidence of tarsal coalition. *Clinical Orthopaedics and Related Research* **181**: 28–36.

Takakura Y, Sugimoto K, Tanaka Y, Tamai S. 1991. Symptomatic talocalcaneal coalition: Its clinical significance and treatment. *Clinical Orthopaedics and Related Research* **269**: 249–256.

Trolle D. 1948. *Accessory Bones of the Human Foot: A Radiological, Histo-Embryological, Comparative Anatomical, and Genetic Study*. Munksgaard: Copenhagen.

Uitenbroek DG. 2000. Fisher's exact. SISA. http://www.quantitativeskills.com/sisa/statistics/ fisher.htm. [3/23/2009].

Varner KE, Michelson JD. 2000. Tarsal coalition in adults. *Foot and Ankle International* **21**: 669–6672.

Warren MJ, Jeffree MA, Wilson DJ, MacLarnon JC. 1990. Computed tomography in suspected tarsal coalition. *Acta Orthopaedica Scandinavica* **61**: 554–557.

Webster FS, Roberts WM. 1951. Tarsal anomalies and peroneal spastic flatfoot. *Journal of the American Medical Association* **146**: 1099–1104.

Wechsler RJ, Schweitzer ME, Deely DM, Horn BD, Pizzutillo PD. 1994. Tarsal coalition: Depiction and characterization with CT and MR imaging. *Radiology* **193**: 447–452.

Wray JB, Herndon CN. 1963. Hereditary transmission of congenital coalition of the calcaneus to the navicular. *Journal of Bone and Joint Surgery* **45A**: 365–372.

Zeide MS, Wiesel SW, Terry RL. 1977. Talonavicular coalition. *Clinical Orthopaedics and Related Research* **126**: 225–227.