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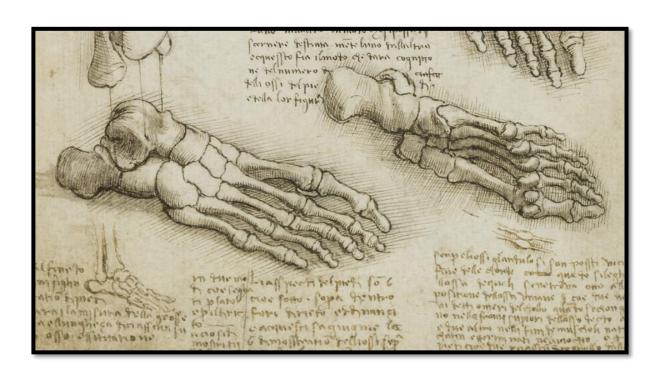
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# NORMAL AND PATHOLOGICAL FOOT BONES VARIABILITY IN HISTORICAL AND MODERN SERIES



## **EDUARDO SALDÍAS VERGARA**

**DOCTORAL THESIS, 2019** 

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# UNITAT D'ANTROPOLOGIA BIOLOGICA DEPARTAMENT DE BIOLOGIA ANIMAL, BIOLOGIA VEGETAL I D'ECOLOGIA DOCTORAT EN BIODIVERSITAT

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In memoriam to Eduardo Vergara and Domingo Saldías, my grandparents, who passed away during my PhD process. My best memories and their legacy always will be in my heart.

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"They called us resurrectionists, grave robbers."
"The miracle of life is granted, and how that miracle can be defective, is a nuance that I am most interested in understanding."
"I must know why five fingers are intended before I can discover the cause of six."
E.B. Hudspeth, "The Resurrectionist: The Lost Work of Dr. Spencer Black"

"Sapere aude"...

(Horatius, Epistularum liber primus)

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## 1. INTRODUCTION

"The human foot is a masterpiece of engineering and a work of art".

Leonardo da Vinci

#### 1.1. Abstract

Introduction: This thesis was devoted to the study of the foot, mainly in ancient collections. The foot is a forgotten bone set in many anthropological, paleopathological, and forensic studies. However, being the key structure that maintains the upright position, it is basic to know the life of a specific individual and the destiny of a population. To understand the structural and biomechanical changes of the modern foot, it is necessary to appeal to Paleoanthropology; the unique science that studies in detail the foot anatomy of our ancestors. Thus, through an interdisciplinary study, this analysis meets a series of scientific needs regarding foot morphology and the changes in its structure between populations.

**Aim:** We analyzed the normal and pathological variability of the foot bones, emphasizing mainly on factors that alter its structure. Moreover, we considered the particular features of each collection and their anatomical changes across time.

**Materials and methods:** To carry out our study, we used 890 samples that correspond to pairs of skeletonized feet of contemporary and ancient osteological collections from Spain, Italy, and Oman with different dating. We applied morphological and metric methods to check their differences, with different purposes. In addition, we used exploratory exams such as CT scans or RX cinematography in living patients.

**Results:** Six studies summarized all our results between 2016 to 2019. They are classified according to the etiology of the osteological changes in the foot. In regard to normal variability, we proposed equations to discriminate sex through metrical methods in the navicular bone. In addition, we observed the cuboid facet of the navicular bone, described as an occasional finding and present in the 52.7% of contemporary individuals in Spain, without significant differences between sexes and series. Taking into consideration the pathological variability, the most significant differences between the Spanish and Italian series were in the X-XIX centuries CE, with degenerative processes being the most common variables. On the other hand, we also proposed a new methodology to estimate non-pathological flat foot in dry bones; found new frequencies of bipartition of cuneiform bones exceeding the world average and, reported the oldest case of the Mueller-Weiss disease in the ancient Egypt.

**Discussion:** Considering the lack of bibliography in most of the studied topics, we proposed a series of new equations and methodologies to be applied in the laboratory. In addition, we suggested new focuses about foot morphology, which will be useful for setting biological profiles and aiding in population studies. Our results are the expression of osteological modifications, present since many centuries ago up to current day. Some factors could explain these changes. Footwear, diseases, genetics, or occupational stress are the most common. Nonetheless, despite technology being very present in the studies of many diseases that affect the feet, there is still much work to do.

#### 1.2. Hominization and bipedalism

From the metazoarian phase of the Life Evolution, deep in the Precambrian Period, displacement has been one of the basic needs of most animals, because it allowed the necessary activities for the survival and perpetuity in time. In the case of our old ancestors, their main evolutionary transformations caused by bipedalism are related with exploitation of natural resources, dietary modification and climate variations among other factors. Furthermore, they progressively acquired a bipedal locomotion after an almost exclusively human pattern in upright position, which allowed more efficiency during the gait.

Bipedalism is the adaptive capacity of some living beings to move, by the use of lower extremities or hind legs, which were originated by a complex evolutionary process in reptiles (since *Euparkeria capensis* from Carboniferous) [1], mammals and birds. In human cases, the bipedalism carried to an anatomical restructuring, produced by a progressive adaptation to the terrestrial environment, to the detriment of a previous arboreal life.

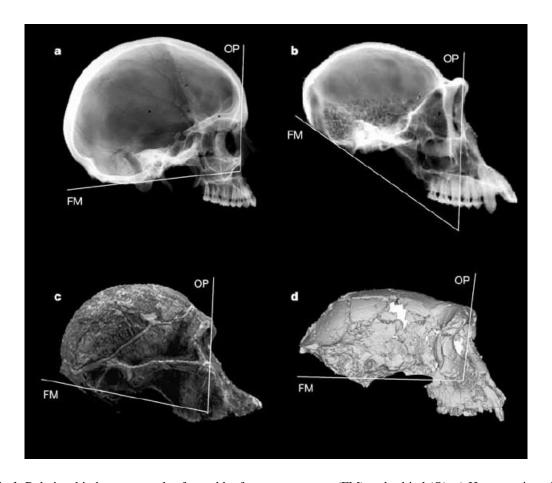
The study of the evidence of the bipedal adaptation of our ancestors is encompassed in Paleoanthropology, a science that studies human evolution, using the scarcely available hominin fossils [2]. Sometimes, the integrity of the remains is not optimal, and the information must be obtained from a single bone, using comparative anatomy of other primates or ancestors with similar features.

General characteristics of bipedalism imply a round skull in balance over the spinal column, curves of the axial skeleton, and a short and wide pelvis, with the isquion looking forward. In addition, we must consider the growth of a large femoral head, the biomechanical importance of the sacroiliac joints, hypertrophy of the great trochanter, considerable femoral length, adduction and knee valgus, great increase of the calcaneal volume, and an extremely and adduced hallux [3]. Some of the latest paleoanthropological discoveries have evidenced important changes in the features of the anatomical proportions of our older ancestors when acquiring a bipedal displacement. The latter is the most important milestone in the history of the human adaptation [4].

Zollikofer et al. (2005) inferred that gait could be a feature of the first hominids and that it could have arisen after the divergence between the chimpanzees and the lineage of the humans [5].

The oldest remains of the hominin evolutionary line in which bipedal evidences were found have been dated to 6-7 millions of years ago (Mya) approximately. According to the virtual reconstruction of skull remains of an individual (TM 266-01-60-1) found in Toros-Menalla, Chad, belonging to the *Sahelanthropus tchadensis* species, bipedalism was possible to determine by the position of the base of the skull.

This conclusion is mainly related to the existence of an almost perpendicular angle formed by the orbital plane and foramen magnum. The above could favor to an eventual upright position and bipedal displacement in this individual [6]. Despite the lack of postcranial evidences, Brunet et al. (2002) and Zollikofer et al. (2005) [5,6] suggested that *Sahelanthropus tchadensis* displayed the bipedal condition due the similarity of the basilar portion and facial bones with other posterior bipedal hominids (Fig.1).



**Fig 1**. Relationship between angles formed by foramen magnum (FM) and orbital (O). a) Homo sapiens. b) Pan troglodytes. c) Australopithecus africanus. d) Sahelanthropus tchadensis (reconstruction) (Zollikofer et al., 2005 [5]).

In 2001, hominin fossils belonging to *Orrorin tungensis* were found between 6.2 and 5.6 Mya in the Tungen mountain area, Kenya. Despite the absence of pedal remains [2], the appendicular skeleton was important to determinate its hypothetic bipedalism. According to Richmond & Lungers [7], the femur of this species is different than apes and Homo, being more similar to the *Australopithecus* and *Paranthropus* genera. The above is mainly because of the similarities in the biomechanics of the hip, suggesting that this feature was kept for around 4 millions of years until the Pliocene, with the irruption of the first Homo. Nonetheless, the total proportions of the femur and the rest of the lower extremity are unknown [8] due to its preservation state, so the anatomical dimensions correspond to estimations. Conversely, Almecija et al [9], by means of geometric morphometry in the proximal femur, unveiled that *Orrorin* had an intermediate shape, between Miocene apes and Australopithecus genus.

Ardipithecus kadabba is a species found in the Middle Awash area, Ethiopia, dated between 5.8-5.2 Mya, associated to an arboreal environment. According to the analysis of these remains, Haile-Selassie et al. (2001), indicates humans and ape-features in the teeth, which are shared exclusively with older hominins. The observed anatomy in the lower extremities, specifically in the articular surface of the foot phalanges, suggest that this species could move partially as bipedal, though more osteological evidences are needed to confirm this hypothesis [2].

On the other hand, Lovejoy et al. (2009) [10], indicated that the pelvis and femur of Ardipitecus ramidus (4.4 Mya), which were discovered in Aramis and Gona (Ethiopia), showed that this species dominated the arboreal movements and terrestrial walking displacing in a bipedal way, which suggests a transitional stage between both life styles. Even though, this movement was made in a more primitive way in comparison with the Australopithecus, whose bipedalism was related to cranio-caudal shortening of the ilium and the appearance of a lordosis in the lumbar spine, in contrast with other existent apes. These conclusions are similar to the results of the study by White et al. (2009) [11] which approached the paleo-biological aspects of this species and its osteological similarities with other hominids, considering dentition, craniomandibular and postcranial remains. Conversely, Sarmiento (2010) [12] criticized these approaches, arguing that the mentioned features of Ardipithecus ramidus exhibited in the fossil remains are not exclusivity human. Besides, the anatomy of this species demonstrated particularities shared with some early hominoids (Oreopithecus o Dryopithecus), gorillas, terrestrial quadruped primates or semiterrestrials (arboreal).

The beginning of the lineage of the australopithecines started with *Australopithecus* anamensis, species found in Kenya (4.2-3.9 Mya), which was suggested as the antecessor of *Australopithecus afarensis* [13]. The human remains correspond mostly mandible, upper limb and dentition, and to a right proximal and distal tibia. Regarding these tibia characteristics, some authors suggested that *Australopithecus anamensis* was a habitual biped, although kept some features primitive features in its upper extremities [14].

Australopithecus afarensis is a species that inhabited East Africa 3.7-3 Mya. The discovery of this species (since 1973) was a big milestone in the history of Paleoanthropology and human evolution. Australopithecus afarensis was the first discovery of our old ancestors, whose skeletal remains (femur, pelvis, foot and knee) demonstrated evidences of full bipedalism, before developing an increased brain. Therefore, bipedalism, and not the brain, was the definite characteristic of the human ancestor [15].

The well-preserved lower limb have helped to suggest a hypothesis about its locomotion, although some of the foot bones were found isolated [2]. Anyway, the pelvis shape, femur, knee joint and footprints show signals of upright position [15]. Besides, Ward et al (2011) [16] found similarities with transverse arch of human beings, that provide the propulsion and shock absorption during locomotion, like the longitudinal arch of the sole of the foot. The study yielded interesting results about the bipedalism and configuration of the foot structure since 3.2 millions of years ago.

Ward et al. (2011) suggested that it is unlikely that the natural selection had favored arboreal behavior of *Australopithecus afarensis*. They proposed that functionally, the foot of this species is similar to modern humans, supporting the hypothesis that a hominid was totally biped. Regarding the above, the transition from arboreal to terrestrial environment could have happened in the early hominin time (*Ar. ramidus*), when the natural selection kept adaptations to grip and climbing in an arboreal environment [16].

In relation to the vertebral column and pelvis, Lovejoy (2005) [17] mentioned similar features between humans and AL-288-1 (*Au. afarensis*) in the dimensions of the sacrum and L3, that developed a more coronal orientation of the column in comparison with apes. In respect to the pelvis, their features do not correspond to an intermediate rank between apes and modern humans, but a unique species that mixes the changes of the upper zone of the pelvis, which made bipedalism possible, with the most primitives features from the lower zone.

During the Pleistocene, the pelvis was indirectly modified by the locomotion, due to coxal bone transformations at birth, with a fetus being more encephalized each time. On the other hand, it is important to consider the elongation of the lumbar spine, which was previously shortened as an adaptation to the arboreal environment.

More recent fossils, such as *Australopithecus africanus*, provided other data. Barak et al. (2013) [18] studied the possible gait of *Au. africanus* (individuals Stw358, Stw389 and Stw567 from Sterkfontein, South Africa, dated between 2.8-2.6 Mya). The analysis revealed differences in the ankle angle between humans and chimpanzees, and the trabecular orientation of the distal tibia of these australopithecines. The *Au. africanus* gait had extended extremities and loaded in their ankles, being more similar to modern humans than chimpanzees. Nonetheless, these features could also locate *Au. africanus* in an intermediate point. Although the position of the ankle was a result of the load during tree climbing, it is very likely that occupied less time for this activity than modern chimpanzees, which demonstrates a minor degree of dorsiflexion of this species. Unfortunately, there are no foot bones associated with certainty with *Au. africanus* skeletons [2].

Despite the scarcity of the remains, *Australopithecus garhi* (dated between 3-2 Mya), found in the zone of the river of Middle Awash (Ethiopia), has allowed to suggest diverse hypotheses about its anatomical proportions. Asfaw et al. (1999) [19] indicated that a very long proportion of the upper extremities remained until the emergence of *Homo erectus/ergaster*, who was the first in exhibit a modern forearm. *Au. garhi* developed a long forearm in contrast with the humerus and femur. According to the size of the femur (remains of diaphysis and neck), the authors put to this species (individual BOU-VP-12-/1) between *Au. afarensis* (AL-288-1) and *H. erectus/ergaster* (KNM-WT-15000), which allowed to hypothesize that the femur could be long in posterior hominids, before the shortening of the forearm. In relation to the foot, the findings of a proximal phalanx are described as similar in size, long and curved to that of *Au. afarensis*.

Two partial skeletons belonging to *Australopithecus sediba*, (1.98 Mya, found in Malapa, South Africa [2]), show derivate and primitive features in its general biomechanics. Regarding to bipedalism, the findings correspond to foot and ankle bones. Although this species developed specific features, which allowed them to move in a bipedal way; there also existed some osteological features for arboreal behavior [20]. Additionally, DeSilva et al. (2013) concluded that *Au. Sediba* walked with a completely extended leg, and inverted foot during the oscillation phase in the gait.

*Paranthropus boisei* (2.3 to 1.2 Mya), is a species with great robustness in the molars, which inhabited East Africa and developed a clear sexual dimorphism between males and females. In spite of the apparent stockiness, the lower limb morphology (individual OH-80) shows similarities with the ones observed in *Homo habilis* (Domínguez-Rodrigo edt al., 2013) [21] This remark is supported with other femur remains possibly associated to *P. boisei* (SK 82 y SK 97).

Nonetheless, due to its robustness and cortical thickness, it is not possible to make a difference from individuals of gender Homo (*Homo erectus*), concluding that OH-80 has the thickest femoral diaphysis of the known hominids, over one million of years ago of antiquity. Considering that OH-80 radius is robust in contrast with individuals of *Australopithecus* (KNM-ER 20419, AL-288-1p, StW 431, StW 139) and possible *Paranthropus* (KNM-ER 1500, SKX 3699, SK 24601), whose length is superior to any hominid of Pliocene or early Pleistocene, it was suggested that this *P. boisei* walked in bipedal way, with arboreal moments.

Furthermore, *Paranthropus robustus*, a species that inhabited in South Africa approximately 2-1.2 Mya, was a bipedal hominid with a climbing trees component [22]. Susman & Brain (1988) [23] made a descriptive and comparative analysis of the first metatarsal bone (SKX 5017), dated with 1.8 Mya, attributed to *P. robustus*, suggesting that this species had a similar bipedalism as *Homo habilis*. According to Aiello & Dean [24], this bipedal species had a stride walk, but with a different weight transfer system than modern humans.

Homo habilis is a species that lived between 1.9 and 1.6 Mya, whose fossils have been found in Tanzania, Kenya, Ethiopia and South Africa. One of the most important findings about bipedalism process are a sub-adult remains (OH-8), found in Olduvai, Tanzania. They were early called as *H. habilis* for the presence of tools in the site. This species is characterized by the reduction of the size of the pelvis, favoring a less prolonged birth. Even, authors such as Napier (1964) [25], suggested that this species could have moved with a similar gait like the ducks, due to the inability to transfer the weight of the body from one foot to the other during displacement. OH-8 is the first hominid to present an insertion to the calcaneus-navicular ligament, main piece for the development of a medial arch.

Likewise, Ruff (2009) [26] compared the thickness of the humeral and femur diaphysis of a partial skeleton of OH 62 (*H. habilis*) with those of modern humans and chimpanzees, and two *Homo erectus* individuals (KNM-WT 15000 and KNM-ER 1808) and determinated that *Homo habilis* locomotion features are more similar to chimpanzees than modern humans. Thus, their gait could alternate with arboreal periods, in contrast with *Homo erectus* and *Homo ergaster* who dominated the open fields, developing a very similar corporeal structure with modern humans, mainly in the extremities.

Homo georgicus (1.8-1.6 Mya) corresponds to a species found in Dmanisi, Georgia. Through to postcranial evidence and studying the lower limb as an integrated skeletal-muscular system, Wallace et al [27] deduced that the tarsal and the talar neck have parameters similar to the modern human variability. Nonetheless, authors note difficulties to infer about bipedalism of this species, mainly about foot orientation, considering the lack of osteological evidences.

Homo ergaster (1.7-1.5 Mya) is a species found in Africa, with very similar characteristics with Eurasian *H. erectus*, and the first species without arboreal locomotion [28]. Considering that foot bones were not found, the measurements of long bones were used to suggest that *H. ergaster* shows similarities with modern humans from arid regions which is compatible with long-range full bipedalism [29].

Homo erectus (1.7-0.3 Mya) was the first species found outside Africa, although there are remains that belong to this continent. Furthermore, it was the first found species who adopted an upright position similar to modern humans. Its post-cranial skeleton and the lower limb bones suggest that was a habitual full bipedal species with human-like proportions [28]. According to Ruff (2009) [26], possibly, *Homo erectus*, facilitated by standing terrestrial locomotion, colonized regions outside Africa, unlike *Homo habilis* that was more geographically bound down to an arboreal environment.

Considering the other species of the genus *Homo* (*H. antecessor*, *H. heidelbergensis*, *H. neanderthalensis*), they were full bipedal individuals, although differences in some details of the locomotor structures might produce changes in the gait [28]. In contrast, *H. naledi* and *H. floresiensis* who also developed a mixture of human-like and primitive elements, were not able to endure walking for long distances [30–32]. Until our days, many hypotheses have

been built about these findings to explain these feature mosaics in both species. However, to understand the origin of the differences; the geographical environment, dating, and osteological elements in common with early *Homo* species, such as Dmanisi (*H.georgicus*) [2] should be taken into consideration.

#### 1.3. Foot evolution in hominids

Hominin evolution is a period with an extension of 7 millions of years, which is short in contrast with other species. During this time, many structural changes allowed our ancestors to adapt to the environment and settle in new territories. However, the first information about a primitive foot dates from 416-360 Mya. The *Sarcopterygii (rhipidistians fishes)*, a vertebrate aquatic species had an interesting complex of well-developed fins (lobe-finned, muscularised). Thus, in the Late Devonian (400 Mya), the tetrapods (i.e. *Ichtyostega*) developed an ancestral multi-rayed fin as a kind of foot, with at least five tarsal bones, seven toes and two-four phalanges [28]. Probably, the transitional change from fins to stiffer bones was due to an adaptation to the body loads in crawling movements in the depth of the ocean, which might produce an extra stress on the fins.

In addition, the evolutionary study of the foot has been studied in incomplete fossils of primates dated between Paleocene and Miocene (*Plesiadapis, Smilodectes, Notharctus, Propliopithecus, Aegyptopithecus, Proconsul, Dryopithecus, Sivapithecus*). These approaches cover the adaptation to the environment and their foot characteristics. These species could differ in the metatarsal size and sustained development of the tarsal bones, mainly in the calcaneus and talus. Furthermore, as an adaptation in an arboreal habitat, displacement was helped through to the presence of an opposable hallux in the feet to the grip [33].

Throughout the centuries and with the appearance of new species such as the first primitive reptiles (362-290 Mya) or the *Thrinaxodon* (a *Therapsid*, dated in 250 Mya), the foot morphology changes of its form, size and function, depending on the species and environment. With *Asioryctes* (164-45 Mya), their feet developed a structure similar to the basic human foot anatomy, with some differences in the proportions. Afterwards, with the emergence of early primates (55 Mya), their feet were adapted to the arboreal life, with an opposable hallux to the grasp [28].

Until our days, there is not much foot fossil evidences about our first ancestors, but there are many hypotheses about their total structure, movements and gait, which could have contradictions between them. Since 1960, the findings of the OH-8 remains (*Homo habilis*),

Lucy, Laetoli footprints and Hadar (site 333) (*Australopithecus afarensis*) have been contributing to the understanding of evolutionary process of full bipedalism, from an apelike foot to a modern human foot [2].

Fortunately, in the last 20 years, the findings have quadrupled, forcing the reinterpretation of our species history. Thus, to acquire a bipedal locomotion and an upright posture, the humans got a series of adaptations in the foot. As Zipfel et al. proposed [20], the foot, as a unit, is one of the anatomical portions that developed more specializations being responsible of the most important feature of the hominids, the bipedalism.

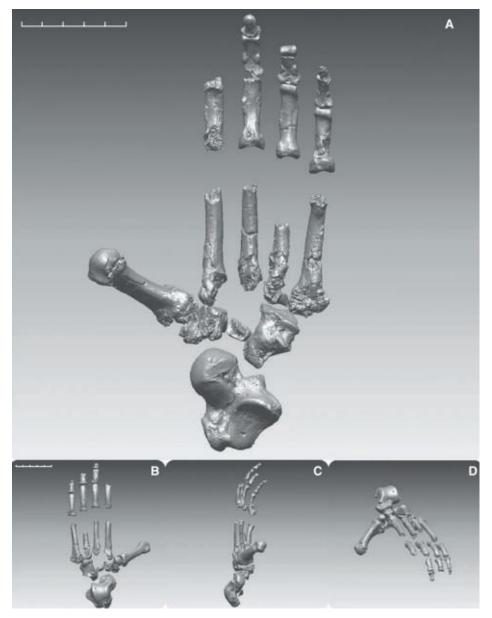
Considering the last findings, do not exist the presence of foot remains in the first hominids species (*Sahelanthropus tchadensis*, *Orrorin tugenensis*). The oldest evidences of the foot bones belong to the left fourth proximal pedal phalanx of *Ardipithecus kadabba* (5.8-5.2 Mya), whose longitudinal curve is compatible with the range of modern chimpanzees. Despite some studies about its locomotion, there are not enough remains to suggest a bipedal walk [2].

On the other hand, *Ar. ramidus* (4.4 Mya) is an important species to understand the first stages of the bipedalism. Despite to be considered as a unique species, individuals from Aramis and Gona could walk in different way. Aramis specimens show [2,10] an additional bone called *os peroneum*, present in old world monkeys and *Hylobates*, and it is not developed in the great apes (Fig.2). This bone is located in the tendon of the *peroneus longus* muscle, helping in its biomechanics. This muscle is the main support of the foot during the grip. *Ar. ramidus* has an opposable hallux in the foot. Thus, the ability to walk in an erect position was completely primitive as this species modified the other four fingers in the gait, holding the hallux to the grip. The above favored an alternation between a terrestrial and arboreal environment [10]. Nonetheless, the new discovery from Gona [34] demonstrated different pedal features. Gona foot would be more everted, and the head of MT1 dorsoplantar is taller than Aramis samples; that means a human-like landing and a better bipedal walker [2].

Anyway, the results of these studies about the feet of *Ar. ramidus* could stake out one of the most important hypotheses of the human evolution. Before the discovery of this species, bipedalism was considered produced by an obligatory transition, as an adaptation to plain

pastures environments, due to the disappearance of forests around 5 Mya. Nevertheless, *Ar. ramidus* remains were associated with an arboreal nature, which contradicts the previous proposal [15]. Undoubtedly, new discoveries about this species and our old ancestors could help to understand more about this transition in our locomotion.

Unfortunately, there is scarce information about the *Australopithecus anamensis* feet (4.2-3.9 Mya) to infer about its locomotion (Mt2 shaft and a proximal phalanx) [2]. The only source of information about the oldest Australopithecines is a complete tibia, which is more similar to *Homo* than *Australopithecus afarensis*. The tibial features might suggest that this species was bipedal [28] (see "Hominization and bipedalism" section).



**Fig. 2.** Aramis foot samples (ARA-VP-6/500. *Ardipithecus ramidus*). A) Plantar view. B-D) Dorsal view. (Lovejoy et al., 2009 [10])

Otherwise, Haile-Selassie et al. [35], described the remains of the right foot of a new individual (BRT-VP-2/73, dated 3.4 Mya), found in Burtele, Ethiopia, which has not been associated with a specific species and categorized as *Australopithecus deyiremeda* (?) [36]. The right metatarsal bones and phalanges differ from the *Cercopithecoidea* samples, for the height of the hallux base in relation to the length of the bone; the dorsoplantar height of the second metatarsal base, and other dimensions with the rest of the other found metatarsals. Even though some metatarsal lengths are more similar to *Cercopithecoidea* than apes or

humans, the principal component analysis was demonstrated that in general, its anatomy is more similar to gorillas and modern humans [35]. Although this species is contemporary with *Au. afarensis*, it differs in the general anatomy of the foot, and has locomotor adaptations more similar to *Ar. ramidus* than *Au. afarensis*. The features of Burtele specimens imply a short hallux and the lack of dorsal circumference of Mt1 head were observed. A long forth metatarsal is similar in size to the first and second metatarsals. The base of the second and forth metatarsals did not show a dorsoplantar expansion, visible in *Ardipithecus* or *Australopithecus*, which were associated with rugged ligamentous insertions able to resist a midtarsal and tarsal-metatarsal dorsiflexion, and a midtarsal break.

The probable *Australopithecus deyiremeda* (?) is similar to *Au. afarensis* to combine an opposable hallux and a direct medial torsion of the second metatarsal. The height of the base of the first metatarsal suggests that the development of the transversal arch of the foot preceded to the development of the longitudinal arch, although this feature is not observable in the samples. Finally, this species could keep a grip ability to dominate the arboreal environment and exploit it with better efficacy, although it did not exhibit a longitudinal arch and was not able to use the transversal metatarsal axis [2,36].

Nevertheless, according to the observations in the lateral metatarsal-phalanx complex, its bipedal capacity was optional and similar to *Ar. ramidus*. The differences with *Au. afarensis* are related to a major capacity to arboreal media and minor in the terrestrial environment [35].

Previously mentioned, *Australopithecus afarensis* is the most known old ancestor (3.7-3 Mya), mainly by AL-288-1 ("Lucy") [28], although there are many foot samples which have been not associated still with any species [2]. Concerning the foot bones, they are a mosaic of ape-like and human features. For instance, the phalanges shape are long and curved, similar to primitive features. Conversely, the hallux, although demonstrated to be opposable, was in lesser degree than apes. In addition, this species had an ankle joint similar to modern humans, like the ability of dorsiflexion in the toe. On the other hand, Desilva et al (2018) [2] indicated that lateral midfoot of *Australopithecus* was stiff and adapted to bipedalism,

considering the human remains found until the date.

Although there is a controversy about bone evidence and the presence/functionality of longitudinal arch (modern humans), footprints from Laetoli, Tanzania, reinforce the theory of full bipedalism. These prints (mostly "Laetoli G") looks like a "flat foot" impression in modern parameters. They showed the existence of a medial longitudinal arch, adducted hallux, stiff lateral midfoot and prominent heel-strike [2], which could have similar features with modern human gait.

Furthermore, the remains of a little foot belonging to the individual Stw573 ("Littlefoot") could correspond to *Australopithecus prometheus* (?) as suggested by some authors [2] or to *Australopithecus africanus* [25,28]. The skeleton was found in Sterkfontein, South Africa (dated 3.67 Mya) and it is one of the best-preserved human hominid remains until our time. "Littlefoot" corresponds to the oldest evidence of an adducted hallux in our ancestors (human-like), although keeps some primitive features [2].

Clarke & Tobias [37], who made the first report about the finding, described the presence of talus, navicular, medial cuneiform and first metatarsal of the left foot, which composed part of the middle longitudinal arch. They described the talus form, an elevated trochlear margin, a vertical groove near to the *hallucis longus* flexor muscle, and the absence of the depression of the medial malleolar facet as human-like features.

In contrast, the navicular shows a mix of human and ape features. The tuberosity resembles the OH8 (*H. habilis*) and differs from African apes, while the features from the lateral half suggest that the medial and intermediate cuneiform were orientated to the axis of an abducted foot. The medial cuneiform exhibits the facet to the intermediate cuneiform with "L" shape in its lateral face, human-like (two in apes), while the rest of the features are ape-like and allow the action of an abducted hallux.

The first metatarsal has a very concave proximal facet to receive the convex facet of the medial cuneiform, which helps the mobility of the metatarsal-cuneiform joint, suggesting

that the hallux differs from the rest of fingers. Even though the individual Stw573 shows an appropriate remodeling to a habitual bipedalism, the medial cuneiform and first metatarsal show ape-like features. The above suggests that the individual was in an intermediate rank between the arboreal hominid foot and human foot adapted to a habitual bipedalism [37]. However, is not possible to know how this individual developed its lifestyle in terrestrial and arboreal environments.

There are no foot bones associated with *Australopithecus africanus* skeletons, except the controversial Stw573 remains [2], to discuss its bipedalism and gait, in contrast to other hominids. Due to the deposit features (Member 4, Sterkfontein) and preservation conditions, there is no certainty in association an important number of foot bones with this species.

In relation to the next species in the phylogenetic tree, Prang (2014) [38] made a comparative analysis of the calcaneus of *Australopithecus sediba* (1.98 Mya). He compared it with primate calcaneus (*P. troglodytes*, *G. gorilla*, *P. pygmaeus*), hominins from Omo and Sterkfontein (Omo 33-74-896 y Stw 352), *Au. afarensis*, *Au. africanus* and *H. sapiens*. The results revealed that human calcaneus differed with big apes because of the presence of a robust tuberosity (increase of the posterior tuberosity), adapted to the big loads that must resist the heel during the bipedalism. Nonetheless, the analysis also demonstrated that the *Au. afarensis* heel developed a more robust structure than *Au. sediba* and showed features that were more ape-like. This characteristic reveals a functional advantage of *Au. afarensis* in the terrestrial environment, especially in the heel load over long distances, helped for the position of the plantar process. This structure provided a broader base of support that allowed a similar displacement to the modern humans.

In summary, this study could explain the phylogenetic pattern among the fossil species whereby the midtarsal features. *Au. africanus* (Stw 628 and Stw 485) has a midtarsal more similar to *Au. afarensis* (AL 333-160) and *H. habilis* (OH 8), with a flattened and dorsoplantar expanded forth metatarsal base. On the contrary, *Au. sediba* shows a convex metatarsal base like the big apes [38]. However, to understand the evolution between the different degrees of

kinship between taxa of hominid species, it is not possible to discard that different factors could produce a recoil or a development of characters, as was observed in the calcaneus.

The few remains of *Paranthropus robustus* (2-1.2 Mya) show that this species developed some modern features, which allowed a bipedal walk, but displayed different mechanisms of weight transfer during the gait [28]. The presence of a grasp hallux also is not clear [2].

In 1965, Day & Napier described the existence of a little well-preserved foot from Olduvai Gorge (OH 8), Tanzania [39]. These remains of *H. habilis* (1.9 to 1.6 Mya) were found complete, except the phalanges. The preliminary analysis indicates similarities to *H. sapiens* due to the presence of an articular facet between the bases of the first and second metatarsals, demonstrating the absence of divergent hallux. However, the fifth metatarsal was described as different from that observed in modern humans, concluding that the species was bipedal, but did not have an upright posture as *H. sapiens*.

Other studies demonstrated a controversy regarding the age of the individual [40] by noconsolidation of metatarsal epiphyses [41]. In addition, there is an extend discussion about the degree of bipedalism of the OH 8 remains. At first instance, it was stated that OH 8 foot exhibited a total adaptive development to bipedalism, while other investigations suggested that this species kept osteological features in an arboreal environment, which was checked through the study of other features of *H. habilis* [42].

For instance, Kidd (1998) [43] proposed that OH8 foot had a mix of human (cuboid, lateral longitudinal arch) and ape-like features (talus, navicular), which could favor a degree of grip adaptation, through divergent hallux. Considering these features, Kidd proposed that it is improbable that this species could move in an upright posture like modern humans. In addition, D'Aout & Aerts (2008) [28] mentioned that *H. habilis* foot had some similar features with *Au. africanus*, and modern structures such as non-opposable hallux and longitudinal arch.

From this point of the human evolution, foot of hominins displays more well-defined characteristics of modern human walking. The Dmanisi foot remains (*Homo georgicus*), despite showing a Mt1 with primitive features (ankle and the presence of longitudinal arch), developed modern human-like characteristics [2].

Due to the findings associated to postcranial skeleton of *H. erectus (ergaster)*, it was concluded that his bipedalism was upright and without arboreal adaptations. Besides, Bennett et al (2009) [4] made an analysis of hominid footprints, probably of *H. erectus* or *H. ergaster* in two layers of sediment, dated between 1.51 and 1.53 millions of years in Ileret, Kenya. The above is the oldest evidence of a modern foot with an adduct hallux, longitudinal arch and weight transfer before the impulse. The study compared the mentioned footprints with *H. sapiens* and impressions found in Laetoli, Tanzania (G1-33). They concluded that the Ileret footprints (FwJj14E) showed size differences in the instep and the presence of a functional longitudinal arch. In addition, a general similar size, an equal fingers takeoff with modern humans, and a probable speed of 0.63 m/s in the gait, compatible with a slow walk, like previous animal footprints in muddy terrain.

Considering the discovery of early *Homo* species and others more modern one, a big number of feet fossils have been found, especially in Sima de los huesos in the Atapuerca mountains (Spain). There are also some studies that checked species like *H. antecessor*; *H. heidelbergensis* and *H. neanderthalensis*, that suggested a completely bipedalism. Their foot morphology differs in some details from modern human (mostly in robusticity), but their displacement was made in upright position [25]. To date, there is a great number of human feet fossils that still have not been associated with skeletons and/or species [2,25,44].

Even though the mentioned species demonstrated very similar foot anatomy in contrast to modern humans, there are some exceptions between our ancestors. *Homo naledi* (South Africa, 335.000-226.000 years ago) and *Homo floresiensis* (Flores Island – Indonesia, 50.000 years ago) developed particular foot features, considering their environment and dating. Their morphology, despite a series of similar modern features, also developed primitive structures (i.e. talar head/neck declination, curved proximal pedal phalanges in *H. naledi*; long forefoot

and primitive navicular in *H. floresiensis*) [30,32]. As we mentioned before, these characteristics generated differences in the transfer of weight in the gait, not being able to go long distances and attain high-speed/efficient running [2,30].

Recently, in 2019 have discovered human remains of a probably new species (*Homo luzonensis*) in Callao Cave, Philippines (dated 67.000 years ago) [45]. Dentition study determinated that this finding might belong to three different individuals. Mt3 (CCH1) demonstrated a small base in relation to the bone length and a proximal articular facet with a marked convexity in the dorso-plantar plane. CCH3 is an intermediate foot phalanx, of which there is no-certainty about their comparative morphology due to the variability of this bone in *H. sapiens* and other hominins. On the other hand, proximal pedal phalanx (CCH4) has similar features with australopithecines such as a small bicondylar head and marked longitudinal curve, among others. Nonetheless, through 3D geometric morphometric shape, it is not possible to distinguish characteristics of *A. africanus* and *A. afarensis*. Thus, foot bone features indicate a mixture, an intermediate stage between great apes and modern human abilities [45]. The study also reveals that CCH4 could be compatible with *Homo* genus, but its specificity is not clear, which might indicate a new species with its own particularities.

Evolutionarily, the human foot features already had acquired an anatomical reduction and most of the abilities of the foot, like hyper-flexibility and grip were lost with the time. Besides, the midtarsal changed, causing the conformation of the longitudinal arch of the plantar vault [41], a fundamental structure for shock absorption and propulsion [42]. Forefoot also reduced its structure, shortening of metatarsals, cuneiforms and phalanges, with a parallel disposition of the first metatarsal, similar than the second. This makes a difference from the rest of primates because they do not correspond with a feature of a foot available to the grip, and a disposition of the calcaneus with talus similar to the humans, in relation to the transmission of load and movement [33].

Thereby, *H. sapiens*, with its developed brain and an upright walking, was able to exploit and administrate the sources of diverse ecosystems. The changes in the foot helped this species

to colonize the planet, emigrating to different parts of the globe in search of new and better life opportunities, developing in this way new forms of life, for example the agriculture. This new technique of resource management would change again the human diet, as well as the coexistence and interrelation between human groups [15]. In addition, taking in consideration different weather and geography, our species used animal leather to protect the body and the feet, thus creating the footwear.

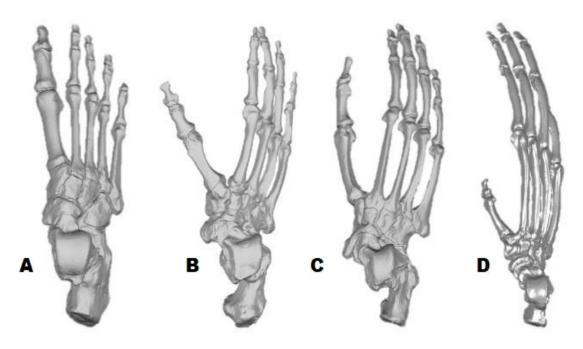
Thereby, the importance of the bipedalism and foot study, is that it not only provides interesting knowledge about the biomechanics of the individual displacement, but also allows learning about the lifestyle and habits. For instance, Kapandji (1998) [46] exposed that modern humans who live in cities, do not show significant degrees of adaptation. They walk over flat surface of the streets, resistant and without accidents, covering their feet with shoes. Considering the theory of the primate opposable hallux, he proposed that humans in the future could walk with reduced fingers like stumps, by the progressive atrophy of them, and the muscles that hold the feet.

### 1.4. Foot morphology in modern humans

In many vertebrates, foot is the distal portion of the lower extremities constituted by bones, tendons, cartilages, muscles and a large number of nerves/vascular endings. The function is not only limited to the displacement by movements, also it is characterized for shock absorption, body weight support and balance maintenance. In total, the modern human foot is composed of 26 bones in each foot, and divided in different portions: tarsal (7), metatarsal (5), proximal (5), intermediate (4) and distal phalanges [47,48]. In addition, under the first metatarsal head, it is possible to find the sesamoids, which extend into the foot tendons. According to the localization, the foot is divided in three big portions: hindfoot (calcaneus and talus), midtarsal (navicular, cuboid and cuneiforms), and forefoot (metatarsals and phalanges).

Jahss (1991) [33] stated that human foot differs from the rest of primates in three features. Firstly, the presence of longitudinal arch and a pronounced transversal arch, due to the disposition of the muscles and ligaments to disperse the body stress/weight in the foot. Secondly, the human feet are rigid, with narrow support ligaments, while the rest of foot primates and some ancestors show a major flexibility and musculature in the zone. Finally, the reduction of the foot size and non-adducted hallux are divergent features between humans and primates (Fig. 3). Despite the lost of the grip ability of the human foot, this acquired efficiency to transfer the body weight.

Considering the modern foot bones separately, the importance of the calcaneus [os calcaneum] in the bipedalism is related to different factors. One of them corresponds to its position in the articulation with talus and the ground, which determines if the foot will keep a neutral posture [33]. Otherwise, due to joint problems, it could acquire a valgus or varus position, originating an imbalance in the gait and progressively a pathology and deformation of the foot. Inserted on the anterior margin of sustentaculum tali is the plantar calcaneonavicular ligament (spring ligament) [49], which is extended to the plantar side of navicular [48]. This is the most important structure to maintain the stability of the longitudinal arch of the foot. Furthermore, the calcaneus of the modern human shows a wide and robust tuberosity, unlike other primates (Fig. 3) due to its importance in the bipedalism. Some ancestors like *Au. afarensis* and other posterior species also developed these features.



**Fig. 3.** Differences between primate feet. A) *Homo sapiens*. B) *Pan troglodytes*. C) *Gorilla gorilla*. D) *Pongo pygmaeus*. Notice the distribution of the phalanges, size of calcaneal tuberosity and localization of hallux. (DeSilva et al., 2019 [2])

On the other hand, the talus [astragalus] is an intermediate point between the muscles that are inserted between the calcaneus and navicular. Nonetheless, none is directly in it. Kapandji (1998) [46], indicated that the talocrural joint (tibiotarsal) is the most important articulation of the hindfoot because it allows the adaptability of the foot in irregular grounds. This articulation includes the flexoextension movements, and in a lesser degree, the adduction, abduction, pronation and supination of the foot. Thus, the ligaments involved in these movements are the lateral ligaments (internal and external lateral). Besides, upper and lower peroneo-tibials joints are related to tibiotarsal joint due to the biomechanical importance of the ankle.

The navicular [os naviculare] is an irregular bone, very important in the bipedalism, located in the middle part of the foot (midfoot). Its name is due by the deep posterior concavity (talar facet), which articulates with the talus head, similar to a ship (navium in Latin). The navicular has a flat dorsal surface with mild roughness, which articulates with cuneiform bones. It also shows a big lateral tuberosity where is inserted the musculus tibialis posterior, whose function is plantarflexion movement of foot and fingers and keep the stability of longitudinal

arch [48]. According to its position, the navicular forms the arthrodial (navicular-cuneiforms, navicular –cuboid) and enarthodial articulations (talus-navicular).

Likewise, the Cuboid [os cuboideum] articulates with the calcaneus in the posterior side, with the navicular and third cuneiform in the internal side, and with fourth and fifth metatarsal in anterior side [47]. The cuboid, navicular and the hindfoot bones are parts of one of the most important joints in the foot [46], the Chopart joint.

Also known as transverse joint of tarsus or midtarsal articulation, Chopart joint is an imaginary line in the midfoot [50] that articulates the calcaneus with talus, with navicular and cuboid [51], functioning through oblique and longitudinal combined axis. This flat joint is divided into two parts: an internal enarthrosis (talus-navicular joint) with frontal, sagittal and transverse movements, which function is the subjection with the internal longitudinal arch in the gait, and diffusion of the energy in the foot. The other part is external (calcaneus-cuboid), which shows flexion, adduction and extension movements [46].

The last bones that make up the midfoot are the cuneiforms [49], three short and quadrangular bones [os cuneiforme mediale, intermedium, laterale] that articulate with each other. They constitute the main union between midfoot and forefoot. Thus, the union of articular lines of cuboid-metatarsals and cuneiform-metatarsals joints forms Lisfranc joint (tarsus-metatarsals). They are composed of highly intricate arthrodes and a ligamentous system that provide solidity and movement to this joint [46].

The metatarsals [os metatarsi], sesamoid bones [ossa sesamoidea] and phalanges [phalanges digitorum pedis] form the forefoot. The hallux, formed by the first metatarsal and first/second phalanges, has undergone important changes in human evolution [2,28]. In the Mt1 head are the sesamoid bones, concave and round small structures inside the flexor hallucis brevis tendon, which employ constant and tense movements. Unlike the others metatarsals, Mt5 has a styloid process in its base, which does not articulates with any bone but provides space for peroneus brevis muscle insertion [47].

Furthermore, ligaments, muscles and bone articulations should join the tarsus with metatarsals and phalanges, allowing the movement of this structure and adding resistance to body weight (Fig. 4). This happens due to the flexibility and elasticity of the foot, favoring

the shock absorption and transmission of this energy to the ground. According to Kapandji (1998) [46], the most important joints of the foot are calcaneus-talus joint (subtalar), Chopart and Lisfranc (both imaginary articulation line), navicular-cuboid and first metatarsal-hallux (during the gait).

Foot muscles are classified into two big groups related to insertion and origin point. Intrinsic muscles have their origin in the plantar or dorsal surface of the foot and are divided into plantar and dorsal muscles. Extrinsic muscles have their origin in the leg and reach the foot bones [49]. All of these muscles contribute to the displacement of the individual and to foot movements (dorsiflexion, plantarflexion, inversion and eversion movements) [47]. Thus, considering skeletal and muscular systems, biomechanics of the modern foot is product of a long adaptation process to the environment, where natural selection, cultural and ambient factors are involved [2,25,28].

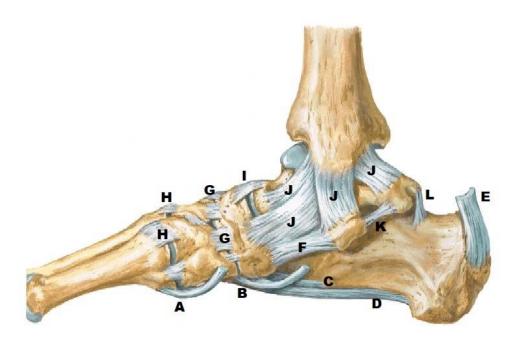


Fig. 4. Medial view of right foot. Tendons and ligaments. A) Tibialis anterior tendon. B) Tibialis posterior tendon. C) Short plantar ligament. D) Long plantar ligament. E) Achilles tendon. F) Plantar calcaneonavicular ligament (Spring). G) Dorsal cuneo-navicular ligaments. H) Dorsal tarso-metatarsal ligaments. I) Dorsal cuneo-navicular ligament. J) Medial ligaments of the ankle. K) Medial talo-calcaneal ligament. L) Posterior talo-calcaneal ligament. (Hansen, 2015 [48]).

## 1.5. Differences in foot structure: human variability and pathologies

Bipedalism requires of energy and the perfect conjunction of all joints. Considering our biomechanical structure, when we increase the velocity of the foot movements, we also change the type of gait (from walk to run). At each step, regardless of speed, feet are the only contact point between the body and the ground. Thus, the foot must hold on to the body weight and transfer the forces adequately, thrusting the lower limbs and generating the step [49].

There are four important biomechanical phases involved in the gait, which are helped by the flexibility and damping capacity of the foot. In conjunction with locomotor system, takeoff, swing, reception of load and medium support phases are implicated in the walk, supported by the performance of the agonist/antagonist muscles [33,49]. These stages not only allow the displacement of individuals, but also the balance with the rest of the body during the gait, and stabilization of the foot structure in contact with the ground. For instance, depending on the irregularity of the ground, the foot could adapt to the floor, deepening the plantar vault. In an inclined ground, the foot could keep a position to support in the surface and release the charge of the body, adopting an everted position, or flexion, cavus-varus position, supination, or supported in the anterior portion [33,46].

Nonetheless, the correct functionality of feet might be affected by a series of factors. In this regard it is important to understand the biomechanics and differences among different styles of walk, which could affect it in diverse degrees. In this line, despite the existence of a "normal gait" into ideal parameters, there are divergences in size, shape and mechanics for every person and every stage of the gait [49]. The above indicates that the particularity of every individual might influence the stability of the foot structure. In this sense, Jahss (1991) [33] mentioned that foot abnormalities not always have a strictly functional etiology, and systemic diseases could show the first physical evidence of the pathology.

Regarding to the skeletal system, bone is a dynamic complex, which could undergo natural morphological changes throughout the entire life [47]. Over the years, the displacement becomes increasingly difficult due to complications arising from foot joint wear, among other factors. However, problems that prevent a correct gait are not only related with aging, can

also be caused by genetic, neuronal and locomotor diseases, fractures, etc. Thus, a correct study could increase the knowledge of diseases to an adequate detection and treatment of individuals.

There is a big number of studies in paleoanthropology/bioarchaeology about osteological signs of diseases in our ancestors. Despite that some diseases have been eradicated, changed or endured over time; bones are clear testimonies of the deterioration in the health of individuals. In this line, Paleopathology, defined as "the study of diseases in ancient populations as revealed by skeletal remains and preserved soft tissues" [47], also provides individual and population information about the health of the people of the past.

Thus, anthropology has been able to understand some aspects of the humans, which could not be resolved with an analysis that lacks transversality. Paleopathology, influenced by several disciplines like dentistry, paleontology, medicine and anthropology [52], might provide data about health state of the individuals in a given context, diet, social status, occupation among other factors. In addition, Paleopathology must analyze the evolutionary and population effects between the affected individuals and their diseases [47].

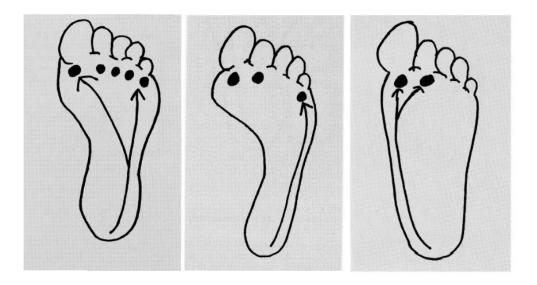
The physician and ornithologist R. Schufeldt applied this term for the first time in 1892 [52]. Nonetheless, in 1921 Sir Marc Ruffer popularized and extended this word to human studies, to refer to his studies on mummies. In 1985, Oxford English Dictionary, considering the use of this word, defined it as "the study of pathological conditions found in ancient human and animal remains" [52,53]. According to Ortner & Putschar (2003) [54], the first study in paleopathology in human remains corresponds to Warren in 1882, who compared artificial deformations of the skull of indigenous from North America. During the last decades, paleopathological studies have focused on differential diagnosis, on the rigorous classification and on identifying all the factors that influence diseases, considering new technologies for their approach [52,55].

Considering foot bones, it is not clear about the first paleopathological analyses in ancient populations. However, in 1741, Nicholas Andry, a French medic, published a book destined to parental care titled "Orthopedics, or the art to prevent and correct the child deformation", becoming the first medic to describe the foot under an orthopedic prism [33]. In this study, he analyzed the anomalies observed in the patients' feet. He detailed the following: "it is not

enough that legs and foot are free of these deformities, which we observed, also it requires of a certain grace to stand and walk on them". The analysis demonstrated that bipedalism and gait could suffer difficulties due to structural abnormalities. Even though there is an increase of interest in biological anthropology to study the foot bones, most of the research has been conducted during the last decade.

On the other hand, regarding the foot health, Brewer (2007) [49] mentioned that the latest studies on pathological gait have been focused on the "support phase" and the initial contact between the foot and the ground. In addition, "take-off phase" could affect the push of the individual. In detail, these anomalies are due to foot alterations that can affect the transversal, vertical or longitudinal axis with regard to with the ground, and they could influence more than one zone. These conditions could imply elevation in the middle longitudinal arc (*pes cavus*), inward-facing heel (*pes varus*) and heel orientated out of the middle line (*pes valgus*) [33,50,56]. Their signs and involved structures can vary depending on the caused damage, which could be visible in bones in severe states.

In the same classification, the three-dimensional alteration of the longitudinal arc of the foot, producing the contact between the sole and the ground is called flat foot (*pes planus*) [33,56]. This condition is well-described in the clinical literature, mostly in pediatrics and surgery, but almost not mentioned in regard to ancient populations. The only studies and reports that mention flat foot of individuals belong to the paleoanthropology field, based in comparative studies between ancient species, modern humans and apes [30,57,58] (Fig. 5).



**Fig. 5.** Plantar view of right feet, indicating the distribution of weight bearing in the metatarsal heads zone. Normal (left), cavus (middle) and flat foot (right). (Franco, 1987 [59])

Nonetheless, foot structures can also undergo changes caused by non-pathological factors. Under these parameters, occupational studies address these particular problematic factors. Even though, some occupations have changed across time with the introduction of new technologies, the usages and types of tools might generate skeletal variations. These features must be interpreted under anatomical and biomechanical terms [55].

Although pathologies related with activities also might also be considered, other indicators like enthesopathies or human variations are useful to unveil the activities and features of the population. Enthesopathies, bony lesions in the ligaments and muscles attachments [60], can provide important information about life and physical activities of ancient populations. Kennedy (1989) [61] defined this condition as "expression of bone plasticity under pressure of extracorporeal and internal forces that are not attributable to disorders of disease, metabolism, biochemistry, hormonal and enzymatic imbalances". Dutour (1986) [60] indicated that etiology of enthesopathies could be related to muscular hyperactivities, inflammatory or metabolic origin. In Spain, the first report of this topic using considerable series was made by Galera & Garralda (1993) [62] who studied 338 skeletons from Santa Maria de Hito. Analyzing enthesopathic markers, they checked that this population

developed hard daily tasks, including grazing, hunting and agricultural activities. In view of foot bones, Castellana et al (1996) [63] approached the occupational stress and feet adaptability to the ground in archaeological populations. These results suggest some aspects of the organization, social distribution, evolution and locomotion.

On the other hand, human variations imply a series of osteological non-pathological patterns in the skeleton, which are different between individuals and/or populations. Thus, some variables could influence this diversity. According to White & Folkens criteria (2005) [47], ontogeny, geography and individual (idiosyncratic) variation influence bone morphology. In this same category, sexual dimorphism is the main factor that influences the human normal variability, and also one of the most important analysis to configure the biological profile of individuals in forensic or bioarchaeological studies. The biological profile includes a set of important characteristics (sex, age of death, stature, ancestry, pathologies, etc.) that define our identity [64]. During the last decades, biological profiles have been very useful in forensic anthropology to compare information between victims/missed people and human remains. Nonetheless, in bioarchaeology and paleoanthropology, they have been used to study features of populations from the past. Until now, they have been confirming notorious morphological and morphometrical differences between sexes, with the skull and pelvis showing better accuracy classification (around 95%) [65–68].

However, taphonomical factors could be an important problem in anthropological investigations, essentially by the damage to the integrity and conservation of human bones obstructing sex diagnosis using the most dimorphic bones. Foot bones could be an interesting alternative. Usually, in contemporary contexts and some bioarchaeological cases, footwear and socks protect ankle and foot bones, which could keep the structures, delaying the decomposition and destruction by different agents [69,70]. Taking into consideration the foot bones, sexual dimorphism has been studied mostly in European and African populations [71–74], using the biggest bones (calcaneus and talus), probably due to their big size and the number of variables available for evaluation.

## 1.6. Aims

Considering the structure, dimorphism, biomechanical and evolutionary importance, and pathological implications, skeletonized feet are a good testimony of the life of individuals of the past. In order to analyze the feet information from contemporary and bioarchaeological populations, different studies were carried out to check these factors.

After to a process of observation in laboratory and archaeological excavations, we detected problems to configure the biological profile through the foot bones, which are one of the osteological structures with better preservation in burials. In addition, taking into consideration the morphologic particularities of these bones and morphological changes between populations, we found a scarcity of bibliography on this subject.

Thus, it was necessary to focus on the differences and structural diversity of the feet, by means of a populational study, using a significant number of individuals. The thesis aims to meet these needs, using new/adapted methodologies to discriminate between individuals and generating a new debate about previous investigations.

Therefore, the principal aim of this project is to approach the foot structure, proposing new morphological and morphometrical methods to grant new laboratory techniques in the biological profile configuration. All the morphometrical techniques used in this project (except a new own variable) [75] were previously suggested by other authors with different purposes [57,58,76,77].

Specifically, we suggested equations able to discriminate (with a high percentage of reliability) the structural differences of the foot bones, including non-pathological *pes planus*, sexual dimorphism, and presence of a cuboid facet, among others. In order to complement our results, we approached different secondary objectives in each of the articles, which are related to the changes of the frequencies of the populations across time (cuboid facet), bibliographic analysis and interpretation of the biomechanics of foot anomalies with scarce literature (bipartite cuneiform, Mueller-Weiss disease).

Conversely, the morphoscopic analysis included the observation and diagnosis of a series of osteological features, including normal variability, pathologies and enthesopathies, which were found in our collections [48,53–55,78,79]. This kind of analysis also implied statistical analysis to every one of their purposes.

All studies included in this project analyzed the foot bone structures from different points of view, especially the pathological changes, which characteristics are more visible and destructive.

## 1.7. Structure of the thesis

This thesis is structured in four chapters. The section "Results" compiled six of our research investigations, whose aim is to give an answer to the lack of foot anthropological studies in different research directions: sexual dimorphism, normal variability, population differences and paleopathologies.

The results are divided in two big subjects, which are related to the etiology of the morphological changes into the foot bones. Thus, the normal variability is deeply approached on the first three research investigations of the project, while the pathological or abnormal conditions are developed since the third to the sixth study.

Regarding the normal variability of the foot bones, the first research is dedicated to checking the size differences between sexes in navicular bones, from contemporary populations from Spain. Considering that many of the hind/midfoot studies have been focused in calcaneus and talus, and the existence of only one previous research on the topic before the year of our initial project [80], we consider this the priority of this work. In addition, in the chapter "Discussion" we included passages of the non-indexed research "Estimación del sexo a través del navicular en poblaciones españolas contemporáneas" of our authorship, to approach some aspects of the sexual dimorphism in non-Spanish collections.

The second study is about the morphological and biomechanical implications of the cuboid facet in the gait. Considering the lack of bibliographic information in this surface and the uncertainty of its etiology, we proposed a metric method in modern and historical series from Spain to check their presence and relationship with the tarsal morphology. Likewise, by means of exploratory exams in living patients during the gait, we analyzed the differences between individuals who developed this feature and who did not show it.

The third research focuses on the differences and evolution throughout the centuries of degenerative processes, enthesopathies and normal variation in human feet. We used foot bones from contemporary and bioarchaeological Spanish, Italian and Omani populations. Considering the particular characteristics of the series, this chapter evaluated every significant variable, its probable causes, and evolution over time. This study gathers most of the osteological changes mentioned in our project.

Concerning to the pathological variability, on the fourth, we focused on the differences between pathological and non-pathological flat feet in dry bones. Due to the lack of scientific tools to discriminate these individuals in laboratory, we proposed a new morphoscopic and morphometric methodology, which results have a considerable percent of good classification.

The fifth chapter studied a very rare particular condition called bipartite cuneiform, using historical and contemporary osteological series, and TC scans of living patients. Through a morphoscopic analysis, we checked the incidences and the probable limitations in the gait of the affected individuals. It was fundamental to the previous research of the subject and the conditions of the live patients to interpret the features and biomechanics.

The last and sixth chapter included a case report on the oldest individual with Mueller-Weiss disease, a condition well studied by clinical methods, but not in bioarchaeology. Furthermore, in the chapter "Discussion" we added extra material, delving into some aspects of the disease that were not included in the original publication for editorial reasons.

Finally, each of the research included in this project was prepared to approach the foot structure and its osteological changes under a different prism. The topics that make up the thesis were suggested considering the lack of information about them, and to propose new anthropological methodologies in the laboratory, with easy replication. Regarding the above, the papers were put in order from the most general to the most specific, considering the number of analyzed individuals and the subject.

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# 2. RESULTS

"Success is a science"
Oscar Wilde

M THE NAVICULAR BONE IN SPANISH SKELETAL COLLECTIONS
n the two genders? No. We came from outer space". Pet Shop Boys
estimation from the navicular bone in Spanish contemporary skeletal ce International. 2016: 267, pp. 229.e1 - 229.e6.



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# Sex estimation from the navicular bone in Spanish contemporary skeletal collections



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#### ABSTRACT

Sexual estimation is fundamental to reconstruct the biological profile of individuals, but postdepositional factors can alter the resistance of the bones, thereby preventing accurate diagnosis especially when the skull and the pelvis are absent. Navicular bones are usually well preserved in archeological and forensic contexts and can a good alternative to discriminate sex. On the basis of these as pects, the present investigation analyzed the sexual dimorphism in 231 pairs of navicular bones from documented contemporary collections from Spain. Receiver operating characteristic (ROC) curve analysis and binary logistic regressions were carried out in eight replicable linear measurements of the navicular bone. Each of the eight variables showed a significant sexual dimorphism in our sample.

The ROC curve results indicate that at least five out of the eight variables used have high ability for sex diagnosis, among which the maximum length of the cuneiform surface (LMAXCUN) showed a better performance (area under the curve value = 0.86). Moreover, we introduced regression equations with combination of measurements that correctly allocated the skeletons with 80% or greater accuracy. The equation with high allocation accuracy rate (83.4%) included a combination of the maximum height of the navicular (HMAX), maximum length of the cuneiform surface (LMAXCUN), and maximum length of the talar facet (LMAXTAL). The regression equations presented here are useful for the Western Mediterranean populations and offer better alternatives than formulas based on other population groups.

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#### 1. Introduction

There are diverse alternatives to estimate sex of adult human bones through metrical methods; nonetheless, the preservation condition of bones is not always optimal. In contemporary skeletonized remains, the short bones, especially from the feet, can be a good option for sexual diagnosis because their dimensions, forms, protection, and their own structures provide fewer probabilities of fractures or damage for different agents [1–4]. Considering this, analysis of sexual dimorphism in the calcaneus and talus of adult individuals has been conducted in different parts of the world, mainly in the European and African populations [5–12]. Despite this fact, the interest in study of the smaller bones

Therefore, the use of the navicular bone as a sex diagnostic tool is possible, although it has not been intensely studied. To explain

of the tarsus is not as high as that of the larger bones, and few studies are available. Specifically, few works exist about the

importance of the navicular bone for diagnosis of sex based on

metrical studies, Kidd and Oxnard [13] used nine measures of the

navicular to determine differences between sexes and populations

(Southern Chinese, Zulu from South Africa, Victorian Britain, and

Roman Britain). Moreover, Harris and Case [14], using Euro-

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American individuals from the William Bass Donated Skeletal Collection, analyzed a series of measurements in whole tarsal bones, including two navicular variables. They used logistic regressions to derive formulas for each bone of the foot as well as combined equations using different bones. Viwatpinyo et al. [15] proposed discriminant functions using 202 pairs of navicular bones from Thai population and obtained a high percentage of correct accuracy. Finally, Navega et al. [16] using machine-learning techniques constructed algorithms to estimate sex from the hind and mid-foot bones in a Portuguese collection (300 individuals).

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navicular sexual dimorphism, Eckstein et al. [17] proposed that the differences can be ascribed to the dissimilarity in the articular cartilaginous surface of the talonavicular joint, which is thinner and smaller in females. In addition, they attributed the sexual variation to differences in the foot structure, especially in the arch stiffness, which is lower infemales. These findings indicate that the female foot has more elasticity and ligament support in the articular surfaces, implying a great degree of adduction [15].

In addition, the studies of discriminant statistic analysis show that a function can be applied only to the population used to produce it or to those populations that have very similar sexual dimorphism [18,19]; however, to date, no Western Mediterranean series of navicular bones has been analyzed. To meet this need, the present investigation was focused on the navicular bone, searching for metrical parameters for a sexual diagnosis through the study of the contemporary documented bone collections from Spain.

#### 2. Material and methods

The materials used for this analysis were the navicular bones of both sides from 231 contemporary individuals of the 20th century with known sex and age (113 males and 118 females). The skeletal material was obtained from Universitat Autònoma de Barcelona Collection (cemeteries of Montjuic, Collserola, and Granollers, in Barcelona province), Universidad de Granada Collection (Cementerio San José of Granada), and Facultad de Medicina Collection from the Universidad Complutense de Madrid (Cementerio de Alcorcón and Cementerio Sur from Madrid), Spain (Table 1).

The specimens in a bad state of preservation or pathological specimens that prevented accurate measurements were discarded. Measurements were selected considering easily located landmarks in fragmentary or complete bones and replicability. All the landmarks have been described in the dassic anthropological literature by Martin and Saller [20] and Kidd and Oxnard [13], except one new dimension described here (measurement #7). To allow replicability with the study of other investigators, all the variables were measured to the nearest millimeter by a non-digital Vernier sliding caliper, except #1 and #2 that were measured using

Table 1

Number of individuals used from the three skeletal collections.

	Individuals			Age	Age			
	Male	Female	Total	Mean	Min	Max	SD	
UCM	63	62	125	654	20	94	16.9	
UAB-1	19	17	36	70.8	32	97	13.6	
UGr	31	39	70	68.4	22	90	13.0	
Total	113	118	231					

UCM, Universidad Complutense de Madrid; UAB-1, collection from Universitat Autònoma de Barcelona; UGr, Universidad de Granada.

the osteometric board. Therefore, considering the navicular in an anatomic position, the measurements are as follows (Fig. 1):

- Maximum width of navicular (WMAX) (Martin and Saller No. 1): Coronal maximum width. It is measured with an osteometric board where the specimen is laid down on the edges of the talar facet.
- Maximum height of navicular (HMAX) (Martin and Saller No. 2): Maximum distance between the tangent to the edge of the talar facet and the highest point of the cuneiform surface.
- Maximum length of the talar facet (LMAXTAL) (Martin and Saller No. 4): Defined as the maximum dimension of the talar facet.
- Sagittal length of the talar facet (LSAGTAL) (Martin and Saller No. 3): Perpendicular dimension to the LMAXTAL, measured in a central point.
- Maximum length of the cuneiform surface (LMAXCUN) (Martin and Saller No. 6): Defined as the maximum dimension of the cuneiform surface in a transverse plane.
- Maximum sagittal length of the second cuneiform facet (LSAGCUN) (Kidd and Oxnard No. 4): Maximum sagittal length of the second cuneiform facet, considering its limits without pathologies or little osteophytes that can confuse the observer.
- Width of the navicular tuberosity (WTUBER): Maximum dimension of the navicular tuberosity, considering anterior and posterior maximum points.

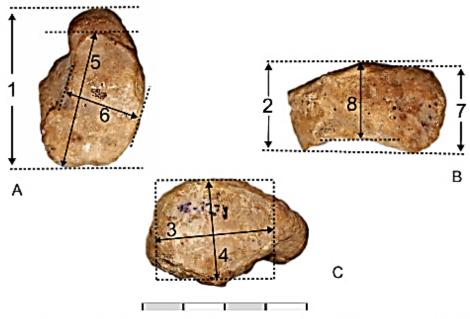


Fig. 1. Left navicular bone. Measurements numerated 1-8 according to the explanation in text. (A) Anterior view, (B) dorsal view, (C) posterior view. Scale bar: 4 cm.

 Sagittal length of navicular (LSAG NAV) (Kidd and Oxnard No. 8): Maximum dimension measured from the mid-portion of the navicular, in a sagittal plane.

To check inter- and intraobserver error, we randomly chose 21 individuals who comprised approximately 10% of the complete collection. The measurements were recorded twice by one observer (ES) in different weeks and once more by a second observer (AM) and were contrasted with the technical error of measurement (TEM) analysis and calculating the absolute and relative TEM [21].

Considering that some variables did not show a normal distribution, the non-parametric Mann-Whitney *U* test was applied to assess statistical differences between sides and sexes. Differences between the left and right naviculars were not significant; therefore, the left side measurements, and the right side ones when the left ones were absent, were used for the following analyses.

Binary logistic regression analysis was performed on a sample of 231 individuals (Table 1) to construct functions to discriminate sex from navicular metric measurements. Logistic regression was preferred instead of the commonly used discriminant analysis, as the former has much more relaxed data requirements (i.e., does not require the data to be normally distributed) and therefore was best suited for our data [22,23]. Moreover, receiver operating characteristic (ROC) curve analysis was used to compare the performance of the navicular measurements and the logistic regression equations to discriminate between males and females in our sample [24,25]. The result of ROC analysis is a curve that represents a plot of the "true positive proportion" on the y-axis against the "false positive proportion" on the x-axis. In the example of Fig. 2, the y-axis represents the male allocation accuracy - the number of males correctly identified divided by the total number of males in the sample, whereas the x-axis represents the proportion of females misclassified. The "true positive proportion" is generally known as the "sensitivity" of the test, and the false positive proportion is denoted as "1-specificity," where "specificity" is the proportion of females correctly identified [23]. Accuracy of sex diagnosis was measured by the area under the curve (AUC), which is the area under the ROC curve. The larger the AUC value, the better is the ability of the measurement or logistic regression equation to discriminate sex. When the variable cannot distinguish between the two sexes, the AUC will be equal to 0.5 (the ROC curve will coincide with the diagonal, see Fig. 2). The logistic regression equations of our analysis were constructed from measurements showing an AUC > 0.8. Combinations of pairs of measurements and a multiple combination of measurements using the stepwise method were selected for logistic regression analysis. On the other hand, ROC analysis allows the identification of threshold values within the measurement values as well as the logits produced by each regression equation. These threshold values would improve the allocation accuracy rate; however, it would exclude some

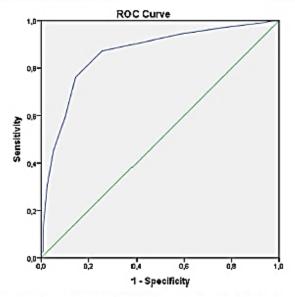


Fig. 2. ROC curve of LMAXCUN (blue line) versus random discrimination (green line) The area between the ROC curve and the green line is the AUC. The larger the AUC value, the better is the performance of the measurement to discriminate between males and females. The y-axis represents the male allocation accuracy, while the x-axis represents the proportion of females misclassified (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

individuals in the overlap region between the sexes [23]. Threshold values for all individual measurements and for the logits produced by the regression equations with a greater AUC are shown in our study.

Finally, the accuracy of the best functions, those with a larger AUC, was tested in a skeletal collection of 36 individuals that had not been previously used for this purpose. We used a sample of well-preserved skeletons of a contemporary collection housed at the Faculty of Medicine of the Universitat Autônoma de Barcelona (UAB-2), with known sex (22 males and 14 females), age (mean age = 79.9 years, min-max = 53-94), and death data (2012-2015) and belonging to the recent population from Northeastern Spain.

## 3. Results

A very low rate of error (relative TEM < 1.3%) was found between the measurements carried out twice by the first observer and an acceptable error (relative TEM < 3.7%) [21] in the measurements performed by the first and the second observer (Table 2).

Table 2
TEM analysis results to check the intra- and interobserver error.

	Intraobserver ana	lysis (ES)	Interobserver analysis (AM)			
	Absolute TEM	Variable average value	Relative TEM%	Absolute TEM	Variable average value	Relative TEMX
WMAX	0.2	38.8	0.6	0.4	38.9	1.0
HMAX	0.2	19.1	1.0	0.4	19.1	2.3
LMAXTAL	0.3	26.6	1.3	0.4	26.6	1.6
LSACTAL	0.3	20.1	1.5	0.4	20.1	2.0
LMAXCUN	0.2	33.1	0.7	0.4	33.0	1.4
LSAGCUN	0.2	17.2	1.2	0.5	17.3	3.4
WTUBER	0.2	18.4	1.0	0.2	18.4	1.2
LSAGNAV	0.2	16.6	1.3	0.6	16.7	3.7

Table 3
Descriptive statistical analysis according to sex (measurements in mm).

Me asure ment	Male				Female					
	N	Min	Max	Mean	S.D.	N	Min	Max	Mean	S.D.
WMAX	107	33	48	39.6	2.7	110	31	45	36.0	2.3
HMAX	111	16	24	20.1	1.6	114	16	21	18.2	1.2
LMAXTAL	110	23	34	27.5	2.1	115	21	30	248	1.6
LSACTAL	96	17	24	20.7	1.6	103	14	25	18.4	1.8
LMAXCUN	109	28	39	340	2.2	117	27	38	30.8	1.9
LSAGCUN	103	14	21	17.5	1.6	107	14	21	16.1	1.3
WTUBER	109	12	25	18.8	2.4	110	12	22	16.7	1.8
LSACNAV	113	14	21	18.0	1.6	118	14	20	166	1.2

Table 3 shows the descriptive statistics of the eight measurements used in this study according to sex. Overall, males have significantly higher means and variances than females (p < 0.01).

Table 4 shows the ROC curve results on the individual measures. LMAXCUN was the variable with the larger AUC (0.862); hence, it includes the best performance to discriminate between males and females. LSAGNAV showed the lower AUC value (0.738).

Useful threshold values for all individual measurements are also shown. With regard to the threshold values of LMAXCUN, it has a male threshold value of ≥32.50 mm with a sensitivity of 0.76 and a "1-specificity" of 0.14. This implies that 76% of males and only 14% of females would be expected to have a measurement of that size or larger. Thus, in a forensic context, we might consider that the probability of an individual of Spanish origin being male, with a LMAXCUN measuring 32.50 mm or larger, is approximately 86%. The female threshold value of LMAXCUN is ≤31.50 mm with a sensitivity of 0.74 and "1-specificity" of 0.12. Thus, the probability of the individual being female with a LMAXCUN of 31.50 mm or smaller is around 88%. Individuals in the overlap region between 31.50 and 32.50 mm would remain undiagnosed.

Results of the binary logistic regressions are presented in Table 5. Only the equations with a pooled allocation accuracy of 80% or greater are shown. We can see that except Equation #2, allocation accuracy rates were higher in females than in males.

Equation #9 that combined HMAX-LMAXTAL-LMAXCUN through the stepwise method shows high male allocation accuracy (81.6%), while Equation #4 (HMAX-LMAXTAL) high female allocation accuracy (86.6%). Considering the pooled allocation accuracy, Equation #9 displays the best rate (83.4%). The AUC values of regression equations are also presented in Table 5. All equations had an AUC  $\geq$  0.86. Equations with high AUC were #3 (0.88), #5 (0.88), and #9 (0.89). Useful threshold values for these equations are reported in Table 6. For instance, considering a male threshold value of 0.5 for the logits produced by regression Equation #9, 69% of males and only 9% of females would be expected to have a logit value  $\geq$  0.5. Thus the probability of an individual being male, with a logit value of  $\geq$ 0.5 for Equation #9, is approximately 91%.

The reliability of the mentioned best functions was tested in a skeletal sample (UAB-2) of 36 individuals of Spanish origin not used in the logistic regression analyses, whose pooled allocation accuracy rates were 86.2% for Equation #3 and 77.7% for Equation #5 and Equation #9.

#### 4. Discussion

The biological profile is a joint set of features that establish our identity and are defined as unique and unrepeatable. In both forensic and physical anthropology, a series of tools have been used to configure this profile through the study of human remains to

Table 4

ROC curve results. Useful threshold values with the sensitivity (true positive proportion) and the 1-specificity (false positive proportion) are presented for males and females.

Measurement AUC	AUC	Males			Females			
		Threshold	Sensitivity	1-Specificity	Threshold	Sensitivity	1-Specificity	
WMAX	0.850	≥38.50	0.645	0.155	≤37.50	0.773	0.168	
HMAX	0.802	≥19.50	0.613	0.167	≤18.50	0.596	0.153	
LMAXTAL	0.842	>26.50	0.709	0.130	≤25.50	0.730	0.182	
LSACTAL	0.816	≥20.50	0.573	0.126	≤18.50	0.602	0.104	
LMAXCUN	0.862	>32.50	0.761	0.145	<31.50	0.744	0.128	
LSAGCUN	0.751	>17.50	0.563	0.103	<1550	0.318	0.126	
WTUBER	0.747	>18.50	0.523	0.182	<1650	0.491	0.147	
LSACNAV	0.738	>18.50	0.398	0.076	<1550	0.212	0.062	

Table 5
Binary logistic regression analysis results.

Equation	Measurements	Logit equation*	Male %	Female %	Pooled %	AUC
1	WMAX - LMAXTAL	- 24,628 + WMAX*0.307 + LMAXTAL*0.499	81.0%	83.3%	82.2%	0.870
2	WMAX - LSACTAL	- 24,774 + WMAX*0.434 + LSACTAL*0.427	81.5%	80.4%	81.0%	0.877
3	WMAX - LMAXCUN	- 27,045 + WMAX*0.264 + LMAXCUN*0.526	79.8%	84.4%	82.2%	0.887
4	HMAX – LMAXTAL	- 26,373 + HMAX*0.539 + LMAXTAL*0.616	75.9%	86.6%	81.4%	0.874
5	HMAX – LMAXCUN	- 29,663 + HMAX*0.548 + LMAX*CUN*0.592	80.4%	81.1%	80.9%	0.889
6	LMAXTAL - LSAGTAL	-23,660 + LMAXTAL*0.659 +LSACTAL*0.329	79.8%	82.2%	81.0%	0.866
7	LMAXTAL - LMAXCUN	- 24,875 + LMAXTAL*0.408 + LMAXCUN*0.438	76.6%	84.2%	80.5%	0.873
8	LMAXCUN - LSACTAL	- 23,189 + LMAXCUN*0.536 + LSACTAL*0.295	76.6%	84.5%	80.7%	0.871
9	HMAX – LMAXTAL – LMAXCUN	-31,951 + HMAX*0.516 + LMAXTAL*0.353 + LMAXCUN*0.397	81.6%	85.1%	83.4%	0.891

Sectioning point = 0. A negative logit indicates female, while a positive logit indicates male.

Table 6
Useful threshold values of the logits produced by regression equation.

Equation	Equation Males				Females			
	Threshold	Sensitivity	1-Specificity	Threshold	Sensitivity	1-Specificity		
3	0.60	0.72	0.07	-0.83	0.71	0.09		
5	0.35	0.69	0.08	-0.83	0.76	0.09		
9	0.50	0.69	0.09	-0.99	0.73	0.08		

estimate ancestry, sex, age at death, and stature [26]. All these variables are useful in creating information that matches with missing people data for recent forensic cases or for specific interests in old populations.

To determine sex in adult human remains, a series of diagnostic variables were proposed, mainly concerning the morphoscopic analysis of the os coxa and skull where such general structures show a major divergence between sexes. It is interesting to mention the compiling works for the skull, mandible, and pelvis [27–30], which have been an object of constant actualization and application in the entire world. Additionally, metrical methodologies with a good and correct accuracy were proposed for postcranial bones, for example, hand and foot bone length [31], metacarpal bones [32], long bone circumferences [33], humerus, ulna, radius [34–36], and femur [37,38] among others. However, the human remains are not always in the best state of preservation, thereby preventing the use of the best elements for a sex diagnosis. For this purpose, feet bones are an interesting option.

Mainly during burials, from the archeological and forensic context, foot and ankle bones are often protected by socks and shoes, elements that provide a partial barrier against decomposition and destruction bone processes. Specifically, leather boots or shoes are often preserved in acidic soils that provide antiseptic effect against decomposition [1]; this results in longer preservation, as has been observed for the World War I field burials [2]. In addition, shoes and boots can protect against plowing, and they prevent access to invertebrates and other organisms, because artificial fibers are more resistant to breakdown, thereby giving long-term protection to the feet [3]. On the other hand, in forensic cases, carnivore scavengers can eat human remains, and the shoes are an element of protection against these factors [4].

As Eckstein et al. [17] proposed, we found high sexual dimorphism in the talonavicular joint (LMAXTAL, LSAGTAL). However, our study revealed that the weight of the navicular (WMAX) and, especially, the size of the cuneiform surface (LMAXCUN) show the highest differences between sexes. Regarding this finding, in the mid-foot, the cuneiform bone size (cuneonavicular joint) can significantly influence sexual dimorphism.

Considering the interpretation of sex differences in navicular bones and previous works on feet bones specifically, we propose to use the navicular bone for sex diagnosis through metrical analysis. The variables selected in this study were proposed for their easy technical replication, as demonstrated by TEM results, using measurements in areas that showed high level of preservation, such as the talar facet or the navicular's tuberosity. Because of the irregular state of preservation of the cuneiform surface, it was decided to take only two measurements in this area (LMAXCUN, LSAGCUN) that showed a good preservation state in these specific points. Nonetheless, the fragility of this area made it difficult to exclude any other options. As expected, no differences in laterality were found, and all the measurements analyzed showed sexual differences that permit their use in a logistic regression analysis.

The ROC curve analysis results showed that five out of the eight navicular measurements (WMAX, HMAX, LMAXTAL, LSAGTAL, and LMAXCUN) have high capacity (AUC > 0.8) to discriminate between males and females, with LMAXCUN showing better performance (0.86) among the variables. These measurements

were combined to create regression equations. The results showed the rate of reliability to discriminate sex was more than 80%, at least in the Western Mediterranean population.

Specifically, by using binary logistic regression, our study shows discriminant equations with high percentage of accuracy (83.4%) using three variables through the stepwise method (HMAX, LMAXTAL, and LMAXCUN) in a considerable number of individuals (231). Moreover, we obtained formulas that included other measurements with an almost similar percentage of reliability using two variables such as WMAX – LMAXCUN and WMAX – L-LMAXTAL (82.2%).

In general, allocation accuracy rates were higher in females than in males. In the female series, the percentage of correct classification was over 80% in all equations. A similar result was not obtained in the male series, where some equations had percentages less than 80%. The poor male allocation accuracy is related most likely to the greatest variance of the measurements in our male series.

Considering AUC values, equations with a higher performance to sex diagnosis were those combining WMAX – LMAXCUN (0.88), HMAX – LMAXCUN (0.88), and HMAX – LMAXTAL – LMAXCUN (0.89). These equations were tested in a skeletal collection of Spanish origin not used in the logistic regression analysis with variable results (77.7–86.2%), most likely explained by the low sample size (N = 36) of the test series.

Similar analysis with logistic regressions was conducted by Harris and Case [14] to determine sexual dimorphism in tarsal bones, including the navicular, by using 160 individuals (82 males and 78 females) of Euro-American origin. In this investigation, only two navicular measurements (NavBrd and NavLg, renamed WMAX and HMAX in our analysis) were used, and approximately an 84% of allocation accuracy was obtained. This percentage of correct dassification is very close to that obtained using our equation including HMAX-LMAXTAL-LMAXCUN in the Spanish population. These authors proposed discriminant functions with a high percentage of reliability using combined equations that include the navicular among other tarsal bones (88-91% of correct dassification at the most). However, it is interesting to note that they developed different equations for the right and left feet, and their most accurate formulas imply the use of measurements from other tarsal bones.

Recently, Navega et al. [16] used the same variables proposed by Harris and Case in tarsal bones and applied an algorithmic method in a contemporary Portuguese skeleton collection. They obtained correct accuracy of 88.3%, with calcaneus and talus being the ones that showed the greatest sexual dimorphism. However, the second cuneiform and navicular (including WMAX and HMAX in the analysis) were the less dimorphic bones. In our study, the same navicular measurements mentioned were included in the logistic regression equations with a higher performance to diagnose sex in Spanish population.

Viwatpinyo et al. [15] used eight measurements of the navicular bone (including five of the measurements that we used) to analyze 202 individuals from Thai population (Chiang Mai University Skeletal Collection). They obtained a high correct accuracy (90% and 92% for the left and right bones, respectively) using the measurement of the maximum tuberosity projection

height (tpH) and other variables. In our work, the diversity of forms of tpH resulted in a lower replicability and high error in the intraand interobserver analysis. The difficulty in the replication of the results caused the removal of this variable from our work.

Thus, few works focused specifically on the navicular bone, and none of them have studied populations of Western Mediterranean origin. It is known that the origin of the population is critical for obtaining a high accuracy in the diagnosis of sex. This statement is shared by different authors who have conducted studies on different bones, for instance, Bidmos and Daya! [9] who analyzed the talus of Black South African population; Spradley et al. [39] who analyzed the humerus of Americans, Mayas, and Hispanic populations; and Safont et al. [33] who studies the long bone circumferences of the Mediterranean population. With regard to tarsal bones, Kidd and Oxnard [13] using individual and integrated measurements showed sexual variation in different populations (Southern China, Roman Britain, Victorian Britain, and Zulu) with different patterns of discrimination. Therefore, the source country of the individuals is a factor to consider in this study field.

#### 5. Conclusions

The present investigation demonstrated the existence of sexual dimorphism in the measurements of the navicular bone, expressed through ROC curves and binary logistic regression analyses. In this analysis, formulas were developed that allowed the correct classification of sex of the individuals with a maximum accuracy of 81.6% in males and 86.6% in females and a general percent of correct classification of 83.4%.

Through binary logistic regression, the variables that mainly contributed to the sexual differences were HMAX, LMAXCUN, and LMAXTAL in a combined equation through the stepwise method. ROC curve analysis showed that the individual measurement with the best performance to discriminate between males and females is LMAXCUN. This study was performed with contemporary remains; thus, its application can be useful in both legal-medical cases of human remains and in recent bioarchaeological studies of Western Mediterranean population.

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2.2. MORPHOLOGICAL AND BIOMECHANICAL IMPLICATIONS OF CUBOID FACET OF THE NAVICULAR BONE IN THE GAIT
"All truly great thoughts are conceived while walking".  Friedrich Nietzsche

## Morphological and Biomechanical Implications of Cuboid Facet of the Navicular Bone in the Gait

Implicaciones Morfológicas y Biomecánicas de la Faceta Cuboídea del Navicular en la Marcha

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SUMMARY: The cuboid facet of the navicular bone is an irregular flat surface, present in non-human primates and some human ancestors. In modern humans, it is not always present and it is described as an "occasional finding". To date, there is not enough data about its incidence in ancient and contemporary populations, nor a biomechanical explanation about its presence or absence. The aim of the study was to evaluate the presence of the cuboid facet in ancient and recent populations, its relationship with the dimensions of the midtarsal bones and its role in the biomechanics of the gait. 354 pairs of naviculars and other tarsal bones from historical and contemporary populations from Catalonia, Spain, have been studied. We used nine measurements applied to the talus, navicular, and cuboid to check its relationship with facet presence. To analyze biomechanical parameters of the facet, X-ray cinematography was used in living patients. The results showed that about 50 % of individuals developed this surface without differences about sex or series. We also observed larger sagittal lengths of the talar facet (LSAGTAL) in navicular bones with cuboid facet. No significant differences were found in the bones contact during any of the phases of the gait. After revising its presence in hominins and non-human primates, and its implication in the bipedalism and modern gait, we suggest that cuboid facet might be related with the size of talar facet and the position of the talonavicular joint. However, other factors such as geographical conditions, genetics and stressful activities probably affect its presence too.

KEY WORDS: Cuboideonavicular joint; Population variability; Talar facet; Supination movement.

#### INTRODUCTION

The cuboid facet of the navicular bone (CFNB), or facies articularis lateralis anterior (Manners-Smith, 1907), is a small flat surface located next to the articular cuneiform surface, in the anterolateral area of the navicular, articulating with the cuboid bone (Fig. 1).

The CFNB is not always present in all modern humans but it is usually in non-human primates. In our species, the frequency of CFNB is ranked between 20 % to 70 % (Manners-Smith; Trinkaus, 1975; Pablos et al., 2018). In relation to hominids, Gomberg (1981) found that all great apes developed long and narrow facets while only 20 % of modern humans present this feature. Also, Sarmiento & Marcus (2000) evaluated the phylogeny and function of the naviculars of the fossils from Olduvai, apes and recent humans. The authors found the CFNB in all hominids and apes, noticing a low presence of this facet in modern humans. This analysis includes the CFNB area of every species, demonstrating that Gorilla beringei has the most similar results with Homo sapiens. In addition, they suggest that a

large combined cuneiform surface in modern humans, reflex an absent or small CFNB (Sarmiento & Marcus).

Despite that recent investigations in new hominin fossils approach the presence of this facet (Pomeroy et al., 2017; Pablos et al.), its causes and evolution are still insufficiently known. Considering these findings, we deemed it necessary to gather more information about the occurrence of the CFNB on contemporary and archaeological collections and delve into its function. On this basis, the aim of this study is to analyze the frequency of the CFNB within the Spanish population and to know under which circumstances the CFNB is present using morphometrical and radiological studies.

#### MATERIAL AND METHOD

We used the left navicular bones of 354 individuals (191 males and 163 females) from archaeological and

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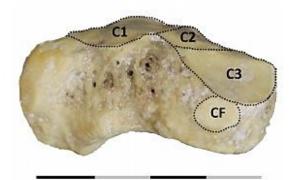


Fig. 1. Medial view of a left navicular bone. Cuneiforms and cuboid facets in discontinued lines. C1: Medial cuneiform, C2: Intermediate cuneiform, C3: Lateral cuneiform. CF: Cuboid facet. Bar scale: 4 cm

contemporary collections from Spain. The contemporary human remains (244) come from the documented collections of the Universidad de Granada, Universitat Autònoma de Barcelona and the Universidad Complutense of Madrid. The ancient remains (110) come from archaeological contexts in Catalonia belonging to the necropolis of the Esglésies de Sant Pere, Casserres and Castell de Termens (Table I). Only well preserved specimens were analyzed.

We considered the CFNB as a flat anterolateral surface next to the third cuneiform facet of the navicular bone (Fig. 1), which can vary in form, extension or breadth. An intra and inter-observer analysis for CFNB identification on 39 individuals from the UAB Faculty of Medicine collection was applied. The general frequency of the presence of the CFNB was obtained, considering the sex of the individuals and their corresponding collections. The eventual statistical differences were tested by means of the chisquared test.

To check if the dimensions of the talar bones influence the CFNB presence, nine metric variables were measured (Fig. 2). We considered two angles from the talar (1. Horizontal angle of the neck NECKANG and 2. Angle of torsion of the talar head TORSANG), and seven lineal measurements from different tarsal bones: two from talar (3. Head length LHEADT and 4. Head breadth BHEADT), two from navicular (5. Maximum length of the talar facet LMAXTAL and 6. Sagittal length of the talar facet LSAGTAL) and three from cuboid (7. Length LCUB, 8. Height HCUB and 9. Breadth BCUB).

To test the replicability, the intra observer error in the osteometric measurements was checked by one of the authors (ES) in approximately 10 % of the whole collection using the absolute and relative technical error of measurement test (TEM). We applied a two-way ANOVA analysis in one ancient and one contemporary series (Granollers) to study the possible effects caused by two factors (sex and CFNB occurrence) in the osteometric measurements.

Finally, the CFNB of the navicular was analyzed under biomechanical parameters. The presence or absence of the CFNB was evaluated in eighty-four patients without midtarsal pathologies from the Hospital Universitari Sagrat Cor from Barcelona (HUSC) using CT scans. Randomly, three patients with the CFNB and two without it were chosen to check the eventual contact between the navicular and cuboid bones in each phase of the step. The complete step was studied under X-ray cinematography (sagittal view) using a Canon Inc. CXDI Controller RF, EC: 2.00 mm, C: 500, A: 1500 and with a zoom of 160 %.

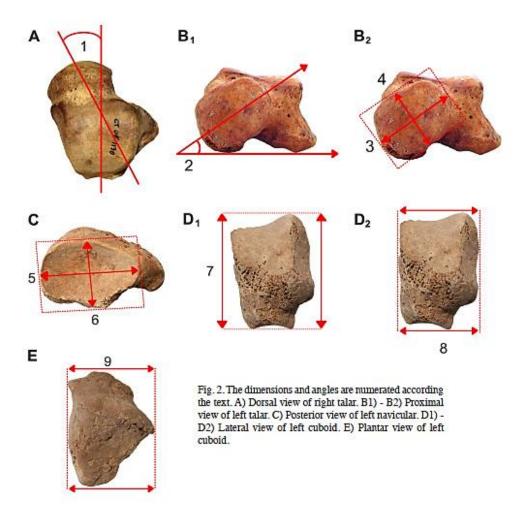
#### RESULTS

Considering the morphoscopic exam, the intra observer analysis did not show any differences between the first and the second observation and the interobserver analysis yielded the same results between the researchers. The CFNB frequency of the whole sample was 52.7%, being

Table I. Number of individuals used from the six skeletal collections.

	CONTEMPORARY			
Collections	Context	Male	Female	Total
UGR	Cemetery of San José, Granada.	31	39	70
UAB	Cemeteries of Collserolla, Montjuic and Granollers, Barcelona province.	28	21	49
UCM	Cemeteries of Alcorcón and Sur, Madrid.	63	62	125
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TER	Lleida, 18 <sup>th</sup> -19 <sup>th</sup> CE	7	3	10
CAS	Berga, 7th-9th CE	20	14	34
STP	Terrassa, 4 <sup>th</sup> -14 <sup>th</sup> centuries CE	42	24	66
TOTAL SERIES		191	163	354

UGR (Universidad de Granada) collection; UAB (Universitat Autònoma de Barcelona) collection; UCM (Universidad Complutense de Madrid) collection; TER (Castell de Termens) series; CAS (Casserres) series; STP (Esglésies de Sant Pere) series.



more frequent in the ancient series than in the contemporary ones (59 % vs 49.8 %), as well as being more frequent in women than in men (57.1 % vs 48.9 %). However, no significant differences were found in relation to chronology (c2 p=0, 116) or sex (c2 p=0, 132).

In relation to bone dimensions, the intra-observer analysis carried out in 50 feet from 30 individuals demonstrated a low rate of absolute TEM (<0.7 %) and an acceptable error in Relative TEM (Table II). Table III show the average of the seven osteometric linear dimensions and two angles measured. As expected, the two-way ANOVA analysis showed significant differences between sexes in all linear measurements, but not in the angles. Concerning the occurrence of the CFNB, the sagittal length of the talar facet (LSAGTAL) was the only metric variable showing

significant differences between naviculars with and without facets (p<0.05) (Table III). Specifically, this dimension was larger in naviculars with CFNB.

To observe the effect of the contact between the navicular and cuboid bones through the CFNB during the gait, we analyzed five patients from HUSC, which were previously characterized, with (three) and without (two) CFNB through CT scans. The movements of the foot during takeoff, swing, reception of load and medium support phase were analyzed by X-ray cinematography. Both individuals with CFNB and those who do not have it, show a gap between the cuboid and navicular bones. This gap remains during all the phases of step. Therefore, none of the walk phases show significant differences between groups that explain the presence or absence of this facet (Fig. 3).

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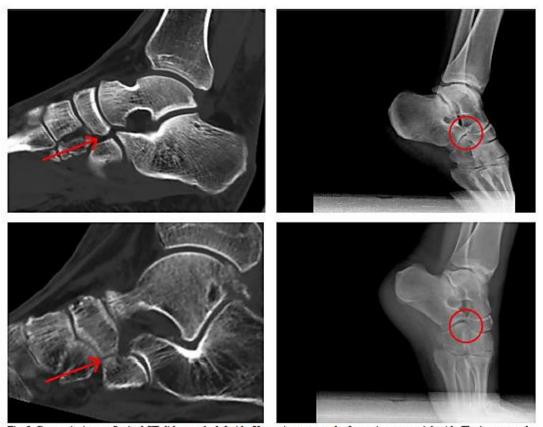


Fig. 3. Composite image. Sagittal CT slides, on the left side. X-ray cinematography freeze-images on right side. The images on the top belong to one individual with no joint between navicular and cuboid based on the CT images. The lower images are from a patient who developed cuboid facet. The images of the left shows no differences in the contact between navicular and cuboid.

	INTRA-OBS	ERVER ANALYSIS (ES)	
	Absolute TEM	Variable average value	Relative TEM %
NECKANG	0.7	13.5	5.6
TORSANG	0.7	31.4	2.4
LHEADT	0.2	25.8	0.8
BHEADT	0.3	8.6	3.7
LMAXTAL	0.1	16.7	0.7
LSAGTAL	0.1	13.6	1.1
LCUB	0.0	24.2	0.3
HCUB	0.1	18.3	0.7
BCUB	0.1	15.9	1.2

Table II. TEM analysis results to check the intra-observer error.

	CFNB PRESENCE		C	CFNB ABSENCE			TWO-WAY ANOVA P-value	
	N	Mean	SD	N	Mean	SD	Sex	CFNB
NECKANG	61	16.90	2.64	43	16.81	2.93	0.87	0.79
TORSANG	63	36.74	5.90	46	34.58	5.61	0.47	0.07
LHEADT	62	31.29	2.91	47	31.91	2.78	0.00	0.41
BHEADT	64	21.03	2.32	44	21.09	1.96	0.00	0.70
LMAXTAL	62	27.37	2.53	49	27.89	2.22	0.00	0.17
LSAGTAL	63	21.61	2.15	47	20.80	1.63	0.00	0.00
LCUB	67	36.20	3.45	48	36.43	3.59	0.00	0.86
BCUB	64	27.34	2.40	44	27.15	2.68	0.00	0.68
HCUB	65	23.64	1.94	47	23.89	1.82	0.00	0.69

Table III. Two-way ANOVA analysis. Osteometric measurements of tarsal bones (angles in degrees and dimensions in mm) by sex and CFNB occurrence.

#### DISCUSSION

The prevalence of the CFNB was around 50 % in the ancient and modern Spanish population. There are no significant differences among series or between sexes. Our results are similar to those found by other authors (Manners-Smith; Prang, 2016; Pablos et al.), and they move away from extreme values such as those Manners-Smith (70 %) and Gomberg (1981) (20 %).

The CFNB is an almost exclusive feature of Primates but hominins do not always present it. The tuberosity of the navicular and size/disposition of cuneiform surfaces are different between our ancestors and modern humans, but shape and localization of CFNB are similar (Jungers et al., 2009: Prang). For instance, Australopitecus afarensis show a big developed tuberosity of naviculars (AL 333-36, AL 333-47) (Prang), which suggests a development of the leg musculature related with plantar-flexion movements (inversion). According to this analysis, this species has a low longitudinal arch, with a mixture of ape and human features (Prang). Moreover, these samples show a semi-lunar (AL 333-47) or round-shape CFNB (AL 333-36), which could be considered similar in form, but bigger than in modern humans. The differences in sizes could be explained by diverse locomotor patterns. However, remains of "little foot" (Clarke & Tobias, 1995) from Sterkfontein (Stw 573, 3.5 millions of years) show unexpectedly, a navicular with a blend of human and apes traits but without CFBN. This bone developed apelike features with a narrow distance between talar and cuneiform facets and a marked angle between the lateral and intermediate cuneiform facets (Clarke & Tobias).

About genus Homo, the navicular from OH8 (Homo habilis) shows a narrow and long CFNB that falls in the variability of modern humans (Kidd et al., 1996). This species probably had an arboreal capacity, with medial longitudinal structures adapted for grasping functions (navicular, talar), with terrestrial abilities (large plantar process of the cuboid), as an adapted longitudinal arch. Concerning Neanderthals, the frequency of CFNB moves from 71.4 % (Trinkaus et al., 2017) to 35.7 % (Pablos et al.). It is possible that the presence of CFNB were due to robust/broad size of the Neanderthal and the expanded tuberosity for the attachment of plantar calcaneus-navicular ligament and tibialis posterior muscle insertion (Pablos, 2015; Pomeroy et al.).

Homo floresiensis did not develop a CFNB and the cuneiform surfaces are smaller in proportion to other mentioned species (Jungers et al.). The navicular is the most primitive in shape to the genus Homo, which was not available to a high-speed or efficient running. The morphology of the locomotor skeleton of H. floresiensis exhibits features of a biped hominid but not exactly like the modern human.

In relation to ancient Homo sapiens, the presence of CFNB was estimated at 36.4 % in Early/Mid Upper Paleolithic humans, and 100 % in Middle Paleolithic Modern Humans (Trinkaus et al.). In contrast, Late Pleistocene H. sapiens 54.6 % of CFNB (Pablos et al.).

Because of bipedalism, joints of hominin foot became less mobile and the navicular bone developed an essential task in the movement of feet. It is especially important for the articulation between the talar head and the talar facet of the navicular, by being part of the peritalar joint complex (subtalar joint and partially the articulation of Chopart). This is one of the most mobile zones of the foot. The navicular bone has developed morphological changes such as the reduction of the tuberosity and the CFNB (Prang), which is absent in a large numbers of modern humans. However, the foot structure of non-human primates developed a different mobile configuration, allowing the use of the foot as a grasping tool, which is an important adaptation in arboreal environments. Their foot must be able to rotate inwards and towards the curved surface of a branch while grasping and/ or vertical climbing. These movements require that navicular and cuboid rotate laterally together (Gebo, 2014) creating a contact between them, forming the CFNB. Also taking part in these movements are strong supinator muscles, the movement of the subtalar joint (talocalcaneal joint) and, the transverse tarsal joint (articulation of Chopart).

The CFNB shows variability among different primate groups. The strepsirhine suborder, predominantly arboreal, quadrupeds, leaping/vertical clinging, show a broad area of articulation with a cuboid facet next to the ectocuneiform and mesocuneiform facets (Dagosto, 1988). This could explain a better adaptation to facilitate the grasp movement. Nonetheless, it is not clear if this strepsirhine condition is a derived character shared exclusively by these taxa (Fleagle & Key, 1994). For instance, in Archaeolemur and Daubentonia CFNB is located more laterally than toothcombed lemurs, being the medial lies in the union of the ecto and mesocuneiform facets (Sargis & Dagosto, 2008). In contrast, CFNB of omomyids, tarsiers and anthropoids that use diverse locomotion, including permanent bipedalism in humans, is related to the ectocuneiform facet and does not spread to the mesocuneiform facet nor the lateral edge (Dagosto; Fleagle & Key; Ni et al., 2013). In primates, all the phalanges and the entocuneiform (especially for its saddle-shape joint with the hallux) are involved in the grasp movement. Sarmiento & Marcus claim that a large cuboid

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facet would be involved in supinated postures, commonly performed by great apes in vertical climbing. For example, the relative big cuboid facet exhibit by African apes and Hadar specimens (AL-333) could be related to hallucal opposability.

However, the feet of modern humans show different biomechanical properties affecting CFNB. For instance, there is a "locking" in the articulation of Chopart that works as a rigid structure, transferring the force effectively during pushoff. This could be due by the plantar flange projecting from the cuboid in articulation with the calcaneus and the plantar ligaments (Lovejoy et al., 2009; Crompton et al., 2012; Prang). Thus, in the propulsive phase of gait, the foot becomes rigid, acting as a lever; meanwhile in the stance phase of walking, the human foot is flexible and adaptable to the ground through the cushing joints (DeSilva & Gill, 2013). This locking restricts CFNB development. Specifically a strong interosseous ligament binds navicular and cuboid bones, blocking the rotation and avoiding the collapse of the mid-transverse arch under load (Gomberg, 1985).

Sarmiento & Marcus explained that reduced CFNB or even its absence in modern humans reflects a foot with a tendency to pronate. Biomechanically, it could be explained as a foot loaded parallel to its long axis and rarely exposed to large midlateral forces. However, in our analysis of the human step with X-ray cinematography from normofunctional individuals, we found no significant differences in the contact of both bones during all the phases of the gait. The presence or absence of CFNB seems to not affect the normal step. This fact could explain why no homogenous frequencies were found in different ancient human and recent series.

Therefore, the differences of CFNB frequency shown in the historical series could be due to other factors such as geographical conditions, genetics, stressful activities and cultural uses, such as footwear use or barefoot conditions. For instance, inadequate shoes can produce deformity (Frey et al., 1993; Bálint et al., 2003), altering the normal gait and quality of life.

The presence of the CFNB could also be relate to some measurements of the midtarsal bones. The sagittal length of the talar facet (LSAGTAL) is the only variable that shows significant differences (p>0.05) among different bones, with or without the CFNB. This measurement affects the talo-navicular joint, one of the most important on the midfoot. This ball and socket articulation mainly allows movements of gliding and rotation, with a closed packed position to the supination (Magee, 2008). One of these

movements, which involves the sagittal length of the talar facet, may be responsible for producing the contact between the navicular and cuboid (as we observed in the X-ray cinematography analysis). The force of the talar head during the screw-like movement produced in the subtalar joint could push the medial or lateral area of the talar facet (Louie et al., 2014). If this impact is mainly in the medial area of the navicular, this bone could increase the contact with a rigid cuboid, which may produce a CFNB.

Taking all data into consideration, the evolution of the foot indicates that CFNB is produced by the movements related to the talar facet, possibly for a mild but constant and incomplete supination of the foot.

Summarizing, our results show that the total frequency of the presence of the cuboid facet is 52.7 %, without significant differences among populations and between sexes. Morphometrically, the sagittal length of the talar facet of the navicular (LSAGTAL) is the only one dimension related to the presence of the cuboid facet. Considering the movements in which the talar facet is involved, the position of the talo-navicular joint, could produce the contact between the cuboid and navicular, creating the facet.

In addition, taking into account the evolutionary history of the primate foot, the presence of CFNB should be related with supination movements that, in primates with a grasping foot, lead to a big cuboid facet. In correspondence to the above-mentioned data, the absence or small size of this structure in modern humans could be understood as the functionality and biomechanical of the gait. Otherwise, factors like footwear, geographical conditions, genetics, or stressful activities have an impact in the frequencies among different populations.

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SALDÍAS, E.; MALGOSA, A.; JORDANA, X. & ISIDRO, A. Implicaciones morfológicas y biomecánicas de la faceta cuboídea del navicular en la marcha. Int. J. Morphol., 37(4):1397-1403, 2019.

RESUMEN: La faceta cuboídea del hueso navicular es una carilla plana e irregular, presente en primates no humanos y en algunos de nuestros ancestros. En humanos modernos, no siempre está presente y es descrita como "un hallazgo ocasional" por la bibliografía. Hasta la fecha, no hay suficientes datos acerca de su incidencia en poblaciones antiguas y contemporáneas, ni una explicación biomecánica sobre su presencia o ausencia. El objetivo de nuestro estudio fue evaluar la frecuencia de la faceta cuboídea en poblaciones recientes y antiguas, su relación con las dimensiones de los huesos tarsales y su rol en la biomecánica de la marcha. Fueron estudiados 354 pares de naviculares y otros huesos del tarso provenientes de colecciones osteológicas de Cataluña, España. Aplicamos nueve medidas aplicadas al talus, navicular y cuboides para corroborar su relación con la presencia de la faceta. Para analizar sus parámetros biomecánicos, se empleó X-ray cinematography en pacientes hospitalarios. Los resultados mostraron que alrededor de un 50 % de los individuos desarrollaron esta carilla, sin diferencias entre sexos o series. Además, observamos que la longitud sagital de la faceta talar (LSAGTAL) es mavor en aquellas muestras con faceta cuboídea. No hay diferencias significativas en el contacto de los huesos en ninguna de las fases de la marcha. Después de revisar su presencia en primates no humanos, su implicancia en el bipedismo y en la marcha moderna, sugerimos que la faceta cuboídea podría estar relacionada con el tamaño de la faceta talar y la posición de la articulación talo-navicular. Sin embargo, otros factores como las condiciones geográficas, genética y stress ocupacional también podrían afectar su presencia.

PALABRAS CLAVE: articulación cuboideonavicular; variabilidad poblacional; faceta talar; movimiento de supinación.

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Pathological and normal variability of the foot bones in osteological collections

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**Highlights:** 

Analyzing foot bones variability, we got differences among series from Spain and Italy

Degenerative processes are the main features that show differences in the populations

Across the centuries, frequency of the variables rose only in Spanish collections

Differences between Spanish and Italian series are highest in X-XIX centuries CE

Lifestyle, occupations, footwear or geography could explain our results

**Keywords:** 

Foot diseases, skeletal variability, population studies, paleopathology.

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## **Abstract**

A wide number of factors could affect the osteological foot structure. In bioarcheology, few studies about feet anomalies include population comparisons and changes across time. Our aim was to identify normal and pathological variability that affected the foot in the recent history of West Mediterranean populations. Thus, we analyzed the evolution of pathological processes, enthesopathies, variability and their probable causes.

We studied 518 pairs of skeletonized feet dated since II-IX to X-XX centuries CE, from Catalonia (Spain) and the region of Lazio (Italy). Moreover, a series from Neolithic of Oman has been analyzed for contrast. We found that calcaneal spur, hypertrophic peroneal trochlea of calcaneus, periosteal reaction of talar neck, alteration of articular surface to the lateral cuneiform, displaced talar neck to medial plane, osteophytes in cuneiforms-navicular joint, fused phalanges, and forefoot eburnation showed significant differences among countries.

Contrasting by countries and dates, we noticed an increase in the frequencies of these variables from Spain across the centuries. Conversely, there are no differences among the Italian series. The period of X-XIX centuries CE demonstrated the highest differences between countries. Running, long walks, intense plantarflexion, osteoarthritis damage could be related with the presence of these osteological features.

## Introduction

Paleopathology is the science that studies the diseases in the past. It is an interdisciplinary discipline with the influence of medicine, molecular biology, genetic, histology, anthropology, archaeology, among others. To understand a pathological process, and to achieve an approach to its etiology, biological profile, morpho-anatomical changes, affected zone, and temporal/geographical frame were considered. The feet are susceptible to undergo muscular and skeletal changes by different factors, which could originate issues in their functionality and gait (Brewer, 2017). Commonly, chronic and progressive pathological conditions as arthrosis could occur with the age. They are characterized by losing cartilage and lesions by the direct interosseous contact. In addition, fractures, obesity, congenital and

neurological diseases, infections and cultural factors among others, could change the normal functions of the feet (Lee et al., 2005).

During the last decades, many researches (Chiavegatti et al., 2018; Isidro et al., 2015; Jashashvili et al., 2010; Laffranchi et al., 2015; Weiss, 2012) studied bioarchaeological collections to understand the lifestyle of the past through the health of the feet (Mays, 2005). Nonetheless, not all populations are affected in the same way, nor do they have the same anatomical variations. For example, foot anomalies are less present in barefoot populations (Balint et al., 2003; Isidro et al., 2015) whereas over a 70% of individuals from modern populations suffer pain and deformities in foot (Balint et al., 2003; Kapandji, 1998).

Thus, we decided to study two sets of skeletal series from two regions of the Western Mediterranean to check the main features of foot health and its variation over the centuries. For contrast, we added a Mesolithic series from the oriental coast of the Arabic peninsula in Asia, whose population was affected by sacral dysraphism in different degrees. The main aim of this research is to check the existence of population differences in foot pathologies/variability, and their changes through time. We would also to approach to the etiology of the main pathologies found, considering the features of the populations, such as occupational activities, geography or footwear among others.

#### Materials and methods

We analyzed 431 pairs of skeletonized feet from two countries, Spain and Italy, from different bioarchaeological contexts (Table 1). In the Spanish collections, we analyzed 215 remains from four archaeological sites in Catalonia: the Esglésies de Sant Pere, Casserres, Castell de Termens and Avinganya. Additionally, the documented collection of UAB that gathers 32 contemporary individuals from the cemetery of Granollers (Barcelona) was also studied.

Concerning to the Italian series, 216 individuals from ten archaeological sites from the Lazio were analyzed: Guidonia, Monterotondo CAR, San Ercolano, Crypta Balbi, Villa Gordiani, Castro dei Volsci, Colonna, Leopoli-Cencelle, Aquino and Allumiere. Besides, we studied

55 pairs of feet from Ras al-Hamra 5 (RH5), a Neolithic series belong to the Sultanate of Oman (Table 1). To contrast the changes across the time between countries and series, we divided chronologically the Spanish and Italian collections in two groups, considering cultural changes (the beginning of the High Middle Ages). The first group joins the sites between II to IX centuries CE, while the second is between X to XIX centuries CE. Considering the dating of Granollers and RH5 series, they were placed in specific groups. In addition, both groups have a balanced number of individuals.

Our analysis considered 37 variables clustered in three groups of features (Table 2), which correspond to all the osteological features that we found in the series. Every one of the foot bones were studied considering the affected zone using dichotomous record (presence or absence). According to their etiology, the first group of our classification includes osteophytes, non-osteophytic alteration of facets and bone deformation due to joint degenerative disease. The second corresponds to enthesopathies and third belong to the bone variability, including accessory bones and facet variability. For statistical purposes, Chisquare test was applied to compare groups and check for significant differences.

### **Results**

We have obtained the frequencies of all the variables by each population and grouped by countries (see Supplementary material, tables ST1, ST2 and ST3). The  $\chi 2$  results showed significant differences for some of these variables when the three countries were compared (Table 3). Most of the differences (8) (Fig. 1 and 2) correspond to degenerative processes (5), and the osteophytes in cuneiforms-navicular joint (ONCUN) showed the highest contingency coefficient values (Table 3). This value would better explain the differences between the collections.

To carry out a comparative analysis between pairs of countries, we used the previous significant variables. Spanish series demonstrated differences with the Italian series for all variables, but it did not show any differences when contrasted with the Omani series (Ras al-Hamra 5). Conversely, Italian collections showed differences with some Omani variables (DISHN, CUNOS, MERGE, EBANT) (Table 4).

To understand these results, we divided the collections by country and period (Table 5). Considering the Spanish collections, we found an increase of the frequencies of pathologies, enthesopathies or occupational stress markers across the time (higher values in X-XIX centuries CE group) in almost all the analyzed features, except DISHN and MERGE. Granollers series shows high frequencies in the majority of features, especially with SPURC, OMANK, EBANT, and MERGE, which are the highest frequencies among all the series (Table 5). Conversely, in the Italian series, there is not a clear evolution of the variables; for instance some occupational markers, SPURC, PERTR, OMANK, and EBANT are higher in the II-IX centuries CE group. Hypertrophic peroneal trochlea (PERTR) was present in all the populations. Despite low sample number (N=55), Ras al-Hamra 5 showed higher values than some Italian variables (DISHN, CUNOS, ONCUN, MERGE, EBANT) and the smaller number of incidences of SPURC in the total collection.

Contrasting by period inside every country (Table 6), Spanish populations demonstrated differences in some variables (calcaneus spur and periosteal reaction in the talar neck). Italian series showed uniformity among its variables. The comparison between Spanish and Italian showed more differences between X-XIX centuries CE groups (Table 6). Taking in consideration the variables, ONCUN showed the highest values of contingency coefficient again (0.307, see Supplementary material). Adding to Granollers to Spanish X-XIX centuries CE collections (renamed X-XX centuries CE), the significant differences inside the Spanish series and contrasted with the Italian X-XIX centuries CE group increased (Table 6)

## **Discussion**

The variables included in this work are mainly localized in hind/midfoot, although a small number of pathologies from forefoot were included. This is due to low frequencies of affected bones in this region that we observed, in contrast with the proximal region (calcaneus and talus). Considering the thirty seven variables of foot, eight showed significant differences in χ2 test: calcaneal spur (SPURC), hypertrophic peroneal trochlea (PERTR), periosteal reaction in the talar neck (OMANK), head/neck displaced to medial plane (DISHN), osteophytes in the surface to the lateral cuneiform of cuboid (CUNOS), osteophytes in

cuneiforms-navicular joint (ONCUN), forefoot eburnation (EBANT) and merged phalanges (MERGE).

By countries, Italian and Spanish series showed significant differences between them, but also when they were compared by periods. Paradoxically, Spanish and Omani series do not show differences, whereas Italian vs Omani series show differences in the half of variables. These results are very far from that expected, mainly in relation to Omani comparisons.

Most of our collections are situated during Middle Ages corresponding to the period between V-XV centuries CE, with some exceptions. Thus, the crisis and the fall of the Western Roman Empire (around IV century CE) affected to the villages and cities from Catalonia and Lazio, which belonged to this empire. Afterwards, diverse foreign invasions, sociocultural processes and overall great epidemics (i.e. Justinian plague) influenced this period (V-X centuries CE). During this period, important cultural changes by the Arabic world in Al-Andalus occurred (more than 80% of the Spanish current territory); while in Lazio occurred a social administrative crisis, by the succession of 23 Popes. Furthermore, in X century CE culminated the Early Middle Age (V-IX/X centuries CE), and began the High and Late Middle Ages (X-XV centuries CE) (Echevarría-Arsuaga and Rodríguez-García, 2013; Mitre, 2011; O'Malley, 2009; Romero, 1949). Due to the historical significance in Europe, we considered X century CE as limit to divide our collections. Afterwards, the annexation to the Crown of Aragon (XII-XVII centuries CE) and the War of the Spanish Succession (XVIII century CE) were the most important events that affected Catalonia. On the other hand, Lazio was part of the Byzantine Empire (Duchy of Rome, VI-VIII) and later of the Papal States (VIII-XIX centuries CE) (Echevarría-Arsuaga and Rodríguez-García, 2013; Mitre, 2004; Romero, 1949; Soldevilla, 1962).

In Spain, the series studied for both periods correspond to ecclesiastic or rural lifestyles. The first Spanish group (II-IX centuries) gathers two series from different context, Casserres and the first period of St. Pere (St Pere I). There is little documentation on the site of Casserres. It is a rural settlement from VII to IX centuries CE. Sant Pere I necropolis belongs to a Episcopal Seat necropolis (IV-VIII centuries CE) related with bishoprics, high social class which led the society (Ferran, 1987; Vives, 1990).

The second Spanish period includes the necropolis of St Pere II (IX-XII). After Muslim invasion, the socio-political relevance of St Pere churches decreased and it became a parish, giving rise to a balanced sex ratio, similar postcranial diseases, and possibly a major diversity in the social class of buried people. The changes in the status of the churches could also affect that kind of foot pathologies observed. The individuals from the first period might be linked with a high social status where degenerative diseases by aging, habits or kind of shoes among others could be responsible of foot pathologies. This data is supported by documents about secular and administrative functions performed by Bishops and functionaries, and because female group demonstrated low skeletal disorders (Jordana et al., 2010). However, pathologies and enthesopaties from the following period could be more related with their field labors and muscular stress activities.

This Spanish period also involved an ecclesiastic and a rural necropolis (Avinganya and Termens, respectively). Avinganya (XIII-XVII centuries CE) was a Trinitarian Monastery where a high prevalence of osteoarthritis in females has been highlighted (Fuentes-Sánchez et al., 2016). On the other hand, Termens was a necropolis located near the Termens castle, whose individuals are dated between 1830-1860 and correspond to an agricultural population.

Between these two periods of the Spanish collections, almost all variables (except DISHN and MERGE, both degenerative processes) demonstrated an increase of frequencies during the centuries. However, only the SPURC and OMANK variables show significant differences. Calcaneal spur (SPURC) defined as a bone spicule in the proximal attachment of the fascia plantar, is related with tensile and traction movements, high stress activities like ballet or running (Weiss, 2012), obesity (Wearing et al., 2006), microtrauma, repetitive tears (Benjamin et al., 2000), compression of plantar fascia and rheumatoid arthritis (Bouysset et al., 2011). It is present in a 16.6% in II-IX centuries CE, versus a 31.6% of the individuals from the X-XIX centuries CE group. Concerning periosteal reaction in the talar neck (OMANK), its etiology is not clear. It could be a reaction related with overcharge of movements of dorsal talo-navicular ligament (Hansen, 2015), responsible for glide movements in the foot. This zone is one of the most important areas of impingement in the dorsi-flexion of the ankle, that is also involved in the rotation and gliding motions (Magee,

2008), with a closed packed position to supinate. The OMANK variable, increased from 5.2% from II-IX centuries CE individuals until 16.8%, in X-XIX centuries CE. Although differences were not seen, almost in all the variables (except DISHN and MERGE, both degenerative processes) demonstrated an increase of frequencies during the centuries.

Foot movements could explain these results. Both variables (SPURC, OMANK), located in the talo-navicular joint and fascia plantar, might be related in intense and repetitive movements like long walks, running and plantar-flexion, occupational activities as strength works to move heavy objects from the floor. Besides, considering the features of the populations, kind of geography/ground, activities and footwear also could influence in these differences.

Finally, within the Spanish group, the Granollers / UAB collection (Saldías et al., 2016) was analyzed in a separate group due to its dating (XX century CE), and because the average age is over 60 years. The last factor is very important to understand is the probable etiology of their main foot pathologies related with degenerative process like osteoarthritis and its secondary signs, considering that age increases the risk of skeletal disorders (Glencross and Sawchuk, 2003). In addition, as a modern collection, footwear could be regarded as one of the main sources of stress in Granollers series. In general, non-suitable footwear could favor the development of feet damage (Balint et al., 2003).

The Italian collection gathers 10 series. Series from II to IX centuries CE includes from 8 to 35 individuals. They are a mixture of different lifestyles under the last years of the Roman and Byzantine empires, which are located inside the current city of Rome and the surrounding area, in the Lazio region. Crypta Balbi is an archaeological complex located in Rome, originally built around XIII CE as a theatre (Sagui, 1998). During the centuries this building and its territory was reused with different purposes. The stratigraphy and archaeological elements as bronze metalwork and glasses, demonstrated industrial activities into the roman urban life. To perform these jobs, the use of constant physical force and manual dexterity is required, in permanent contact with heat to melt the metals or glass. This was possibly to transfer heavy metal elements, the foot needs to make a greater effort in the fascia plantar and longitudinal arch to stabilize the body.

Likewise, Villa Gordiani, a monumental site composed by mortuary structures, religious and a residence complex, also is located in Rome. Through to archaeological findings, this place have been associated to roman imperial families (Palombi and Leone, 2008). According to the social status of the individuals, their feet features could be related to their affluent life, including special footwears (Van Driel-Murray, 2013), diet, genetic, and life span above average.

The rest of series correspond to populations located outside the current city of Rome (Guidonia, Monterotondo, San Ercolano, Colonna) whose lifestyle was animal husbandry and agriculture among others (Baldoni et al., 2018a, 2016; Rothschild et al., 2004; Rubini and Mogliazza, 2005). The necropolis from Castro dei Volsci could be considered an exception by their particular features. It corresponds to victims of the Justinian Plague (*Yersinia pestis*), a pandemic that affected a large part of the world population of the time. Previous anthropological reports (Rubini, 2009) indicated the evidence of notorious muscular insertions and small number of pathologies related with malnutrition or traumas. Furthermore, they could develop farming and pastoral activities, which were the base of the economy of the community.

The Italian series from X to XIX centuries include three collections, but one of them, Aquino, is testimonial because only two individuals could be studied. Leopoli-Cencelle (IX-XV centuries CE) (Baldoni, 2019) is a medieval city located atop of a hill, between Allumiere and Tarquinia. Archaeological findings revealed activities like warehouses, taverns, workshops and hen houses (Stasolla, 2018). On the other hand, population of Allumiere (XV-XVI centuries CE), located in the area of Tolfa Mountains, was a mining city with a high percentage of males (Baldoni et al., 2018b). To extract and process alum, individuals developed occupational skeletal stress corresponding with the alum extraction phases, and functional asymmetry and strong bones were observed. Workers who participated in excavation, moistening and lixiviation stages developed osteoarthritis in feet, while enthesal markers were found in individuals who excavated (Baldoni et al., 2018b).

Despite specificity of labors of Allumiere individuals, in contrast with the diversity of occupations of Leopoli-Cencelle series (X-XIX centuries CE), and the features of other Italian collections (from II-IX centuries CE), significant differences were not found.

Considering Allumiere stressful documented activities, it is possible that the miners developed other pathologies without significant differences. In addition, regarding to statistical study, due to the low and similar frequencies in some variables from Italian series, they could help to understand the uniformity of results.

However, the Spanish and Italian series show interesting differences. II-IX centuries CE groups exhibit divergences in three variables (PERTR, DISHN and ONCUN). On the other hand, differences between X-XIX centuries CE groups are the biggest found in this study. They showed significant differences in most of the variables: enthesopathies (SPURC, PERTR, OMANK) and degenerative processes (CUNOS, ONCUN). The highest frequencies correspond to Spanish population, with the exception of PERTR.

Hypertrophic peroneal trochlea (PERTR or retrotrochlear eminence) was the variable with more presence in the analysis, even over the number of absences of the total samples. This particular feature could be due, amongst several other causes, to a chronic peroneal tendonitis, which probably were common in our Italian series.

Adding Granollers to Spanish collections, the differences between Spanish and Italian series increased (Table 6). It is due to degenerative diseases, as damage by osteoarthritis (CUNOS, ONCUN, EBANT, MERGE), whose frequencies were higher in Spanish populations. For instance, forefoot eburnation (EBANT) might affect, in some cases, the gait and foot in repose with ground contact.

The last country analyzed was Sultanate of Oman. Ras al-Hamra 5 is a Neolithic series (3700-3400 centuries BCE) of sedentary fishermen who habited in the Arabian coast (Coppa et al., 1990, 1985). They collect mollusks from mangrove swamps and sea resources. The individuals analyzed in our study present different degrees of open sacral canal, which have been reported by Coppa (2012). Swaroop and Dias (2011) found a correlation between severe degrees of sacral dysraphism with deformations in joints. However, we did not find lumbar compromise in this collection. RH5 people shows the highest frequency of DISHN of whole the series, and important number in PERTR (83.3%) and EBANT (16.2%). In spite of Ras al-Hamra 5 and Spanish series belong to different chronological and geographical frame and probably develop different activities, they do not show significant differences.

After analyzing all series and countries, we hypothesize that observed differences might be due to specific activities of each population. The populations from X-XIX centuries CE Italian group (Leopoli-Cencelle and Allumiere) lived in the same period, habited in similar kind of geography (irregular soil), under the same cultural system and low pathological frequency. In contrast, X-XIX centuries CE Spanish group revealed a diversity of occupations, footwear, geography, dates and high pathological frequencies that could explain the results. Furthermore, SPURC and OMANK demonstrated significant differences inside Spanish series, which are related with human variability and occupational stress/style-life, implying muscles of plantar vault.

As mentioned before, footwear is an important socio-cultural factor to consider, which could have an influence on the observed osteological deformities. Ancient populations have a trend to develop less foot deformity than modern series, which could be related with inappropriate footwear and increase of life expectancy (Balint et al., 2003; Haines and McDougall, 1954; Kapandji, 1998). Depending on the historical period and culture; footwear can change of style, social context and use, being notorious their influence in increase of some particular foot disease. For instance, Frey et al (1993) observed that pain and foot deformities are mainly related to the use of shoes smaller than feet current women. In addition, Mays (2005) related to the influence of shoes/boots with the prevalence of hallux valgus in different populations throughout human history, due to constriction of shoes to toes (Haines and McDougall, 1954).

It is true that Spanish series are not homogeneous among them, and some differences can be found between the two periods. Conversely, the Italian series are statistically homogenous in relation to pathological affectation and anatomical variables. Partially it can be due to the few individuals of each Italian series, but it could also due to specific lifestyle. This is the main problem of the paleoepidemiology: it is not always possible select samples according their size or specific characteristics as age, sex, social status, lifestyle and so on. Therefore, it is not always possible to have a representation of a country, a geographical area or a specific time. Thus, we must consider that features of every archaeological site can explain these differences.

Summarizing, we found significant differences between countries and periods about frequencies of degenerative processes, enthesopathies and normal variability, which could be produced by diverse factors. Furthermore, we found an increase in the frequencies of the variables with significant differences from Spain (SPURC and OMANK), and uniform results among Italian collections. During the period X-XIX centuries CE, Spanish and Italian samples demonstrated divergences in most of the variables, whose frequencies are higher in the Spanish population, except with the presence of hypertrophic peroneal trochlea. Probably, there are differences among the ecclesiastic and rural life-style in Spanish series, which are not visible between agricultural labors and strength works from Italy. In addition, factors like geography, number of affected samples or footwear also could influence in these results.

Granollers and RH5 are series with particular features. Despite the sample of these populations being relatively small, they demonstrated high frequencies of some variables. Old age and genetic might explain these frequencies. Finally, the majority of the significant variables correspond to degenerative processes, some of them, increased their frequencies across the time, particularly in the Spanish series.

### **Conclusions**

Recent geographic and temporal human foot variability has not been widely studied from the point of view of paleopathology and morphoevolutive changes. The present study shows a comparative view of this topic from two regions, Catalonia and Lazio from Spain and Italy respectively. Both populations share a relatively similar history in the last two millennia as southern European countries, but obvious differences are seen when the comparison is performed town to town.

In these samples, the pathological and stress conditions mainly affect the hind/midfoot, while a very small number of pathologies from forefoot were found. From the 26 pathological conditions and 5 occupational stress markers appreciated in one or the other series, only 8 differences were found between countries: osteophytes in cuneiforms (ONCUN, CUNOS), alteration of the neck of the talus (DISHN), localized bone damage (MERGE, EBANT), and some enthesopathies (SPURC, PERTR and OMANK). These differences seem to be related to concrete activities of specific populations, because all could have a close-relative

explanation in the historical past of the respective populations: ecclesiastics, peasants, miners, traders, nuns, etc.

On the other hand, the bone variability shown as accessory bones or facets does not show the differences between countries or specific populations, illustrating the common populational substrate of both populations. Changes over time are minimal and related to specific series more than population of both countries.

In relation to the Omani series (ancient specific populations used by contrasting our series with an out layer either in age as in geographical area), it was selected to conduct an external comparison due to its very different historic-cultural and geographic context. It shows surprising differences with Italian group and similarities with Spanish ones. As in the case of Spanish and Italian series analyzed, the environmental characteristics of the sample are responsible for these unexpected results.

Specific lifestyle, involving daily work, occupations, journeys, or footwear for instance, and in the other hand geological and geographic characteristics of their environment, makes that every sample constitute a small world. All these factors are the true reason for the differences found among the populations.

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# **Tables**

TABLE 1. Number of individuals from every collection

COUNTRY	PERIOD	CENTURIES	SERIES	REFERENCES	N	PERCENTAGE OF THE TOTAL SAMPLE
		IV-IX CE	Sant Pere I	Jordana et al. (2010)	34	6.5
	II-IX CENTURIES CE	VII-IX CE	Casserres	Cascante & Farguell (2007)	58	11.2
		X-XIV CE	Sant Pere II	Jordana et al. (2010)	55	10.6
SPAIN	X-XIX CENTURIES CE	IX-XIV CE	Avinganya	Fuentes- Sánchez et al. (2016)	47	9
		XVIII-XIX CE	Termens	Alesan et al. (1996)	21	4
	CONTEMPORARY	XX CE	Granollers	Saldías et al. (2016)	32	6.2
		II-III CE	Guidonia	Baldoni, Ferrito and Martinez- Labarga (2018)	14	2.7
		II-III CE	Monterotondo CAR	Rubini & Mogliazza (2005)	11	2.1
		IV-V CE	San Ercolano	Rothchild et al. (2004)	15	2.9
	II-IX CENTURIES CE	V-VI CE	Villa Gordiani	Fornaciari et al. (1984)	15	2.9
ITALY		VI CE	Castro dei Volsci	Rubini (2009)	35	6.8
		VIII-X CE	Colonna	Baldoni et al. (2016)	25	4.8
		IV-V CE	Crypta Balbi	Sagui (1998)	8	1.5
		IX-XV CE	Leopoli- Cencelle	Baldoni (2019)	61	11.8
	X-XIX CENTURIES CE	XI CE	Aquino		2	0.4
		XV-XVI CE	Allumiere	Baldoni et al. (2018)	30	5.8
OMAN	NEOLITHIC	3700-3400 BCE	Ras al-Hamra 5 (RH5)	Coppa (2012)	55	10.6
	TOTAL	-	-	-	518	100

TABLE 2. Variables included in the analysis. Abbreviation codes in bold.

	DEGENERATIVE PR	OCESSES	OCCUPATIONAL STRESS MARKERS	VARIABILITY
OSTEOPHYTES	NON-OSTEOPHYTIC ALTERATION OF FACETS	BONE DEFORMATION/SEVERE BONE DAMAGE	ENTHESOPATHY	ACCESSORY BONES/FACETS VARIABILITY
Osteophytes in Cuboid-Metatarsal joint OCCJN	Flattening Calcaneus-Talus joint FLACC	Comma shape COMMAS	Calcaneal spur SPURC	Fusioned Sustentaculum tali facets STFUS
Osteophytes in the surface to the lateral cuneiform of cuboid CUNOS	Lipping in talar head LIPHD	Merged phalanges <b>MERGE</b>	Achilles entesopathy ACHIL	Cuboid facet of navicular CUBFC
Osteophytes in Cuboid-lateral cuneiform joint OCUCB	Lipping in cuneiform facets of navicular LPCUN	Hind/midfoot joint surface eburnation EBRME	Hypertrophic Peroneal trochlea PERTR	Extended talar tail <b>EXTTT</b>
Osteophytes in Cuneiform- Metatarsal joint OCMTT	Flattening Cuboid- Metatarsal joint FLCUC	Forefoot joint surface eburnation EBANT	Altered tuberosity of navicular EXTTT	Os trigonum <b>TRIGM</b>
Osteophytes in cuneiforms-navicular joint ONCUN	Plantar-medial sustentaculum tali PMSTA	Tarsal coalition COALT	Periosteal reaction in the talar neck OMANK	Trigonal articulation of calcaneus TRART
Osteophytic sustentaculum tali OSTST	Head/Neck displaced to medial plane DISHN	Hallux Valgus <b>HVALG</b>		Accessory navicular NAVAC
Osteophytic Calcaneal-cuboid joint CCLOS	Avascular necrosis in talar head <b>NECHD</b>	Fracture <b>FRACT</b>		
Osteophytic Calcaneus-Talus joint CTOST	Osteochondritis OCDRT			
Osteophytic talar tail <b>TAILO</b>				
Osteophyte in talar head <b>OHEAD</b>				
Osteophytic talar facet of navicular TFOST				

TABLE 3. Chi-Square test and contingency coefficient values of the whole collection. In bold, significant differences between countries/variables ( $\chi 2$ ), and the highest contingency coefficient.

VARIABLES	OSTST	PMSTA	SPURC	ACHIL	CTOST	FLACC	PERTR	TRART	OMANK	TTRNK	LIPHD	NECHD
N	366	361	320	327	365	336	259	339	431	426	422	424
χ2	0.117	0.119	0.012	0.138	0.691	0.073	0.005	0.376	0.012	0.614	0.490	0.066
Cont. Coef.	0.018	0.109	0.164	0.109	0.045	0.124	0.198	0.076	0.141	0.048	0.058	0.113
VARIABLES	OHEAD	DISHN	EXTTT	TRIGM	TAILO	TFOST	CUBFC	LPCUN	COMMAS	EXTUB	FLCUC	OCCJN
N	424	424	394	393	393	308	275	297	308	246	302	299
χ2	0.650	0.043	0.063	0.109	0.526	0.073	0.130	0.697	0.131	0.506	0.253	0.098
Cont. Coef.	0.045	0.122	0.118	0.106	0.057	0.129	0.121	0.049	0.114	0.074	0.095	0.125
VARIABLES	ONCUN	STFUS	COALT	MERGE	EBRME	EBANT	OCDRT	HVALG	CCLOS	OCMTT	FRACT	CUNOS
N	297	361	518	510	508	362	512	275	353	296	511	301
χ2	0.000	0.190	0.883	0.003	0.155	0.002	0.379	0.077	0.989	0.653	0.156	0.014
Cont. Coef.	0.236	0.096	0.022	0.148	0.085	0.183	0.061	0.135	0.008	0.054	0.085	0.166

TABLE 4. Chi-Square test values. Significant differences between pairs of series Spanish, Italian and Omani series are in bold.

COUNTRY	SPURC	PERTR	OMANK	DISHN	CUNOS	ONCUN	MERGE	EBANT
SPAIN/ITALY	0.012	0.002	0.003	0.044	0.004	0.000	0.001	0.003
SPAIN/OMAN	0.063	0.218	0.669	0.344	0.461	0.181	0.600	0.222
ITALY/OMAN	0.309	0.902	0.171	0.012	0.047	0.053	0.005	0.000

TABLE 5. Frequencies of variables in studied populations.

	SPAI	N II-IX CE	SPAI	N X-XIX CE	ITAL	Y II-IX CE	ITAI	Y X-XIX CE	RAS A	L-HAMRA 5	GR/	ANOLLERS
	N	Presence	N	Presence	N	Presence	N	Presence	N	Presence	N	Presence
SPURC	66	11	95	30	48	11	67	11	13	1	31	21
SPURC		16.6%		31,6%		22,9%		16,4%		7.7%		67,7%
PERTR	55	32	79	55	27	23	58	49	12	10	28	20
PENIN		58,2%		69,6%		85,2%		84,5%		83,3%		71,4%
OMANK	77	4	123	19	98	7	79	3	33	4	31	10
OWANK		5,20%		16,80%		7,10%		3.8%		12.1%		32.3%
DISHN	74	5	110	6	101	1	78	2	30	3	31	1
DISHIN		6.8%		5.5%		1.0%		2.6%		10%		3.2%
CUNOS	62	3	58	5	5	0	69	0%	31	1	29	2
CONOS		4.8%		8.6%		0%		0%		3.2%		6.9%
ONCUN	60	7	54	11	48	0	74	1	33	2	28	3
ONCON		11.7%		20.4%		0%		1.4%		6.1%		10.7%
MERGE	88	4	123	1	119	0	93	0	55	2	32	8
IVIERGE		4.5%		0.8%		0%		0%		3.6%		25%
EBANT	85	1	73	2	54	1	81	1	37	6	32	15
EDAINI		1.2%		2.7%		1.9%		1.2%		16.2%		46.9%

TABLE 6. Chi-Square test values. Spanish and Italian series divided by age. Significant differences in bold. Contingency coefficient between Spain vs Italy X-XIX centuries CE and variables in the last row. The strongest value in black.

COUNTRY/TIME	SPURC	PERTR	OMANK	DISHN	CUNOS	ONCUN	MERGE	EBANT
SPAIN II-IX/X-XIX CE	0.033	0.172	0.016	0.715	0.407	0.203	0.079	0.473
ITALY II-IX/X-XIX CE	0.384	0.934	0.339	0.417	-	0.421	-	0.772
SPAIN II-IX/ITALY II- IX CE	0.406	0.015	0.599	0.039	0.109	0.015	0.019	0.745
SPAIN X-XIX/ITALY X-XIX CE	0.029	0.044	0.005	0.333	0.013	0.000	0.383	0.500
SPAIN II-IX/X-XX CE	0.001	0.129	0.003	0.587	0.441	0.370	0.675	0.000
SPAIN X-XX/ITALY X-XX CE	0.001	0.041	0.001	0.391	0.016	0.001	0.018	0.001
CONT. COEF. SPAIN/ITALY X-XIX	0.169	0.169	0.197	0.070	0.216	0.307	0.059	0.054

CE

Figure 1

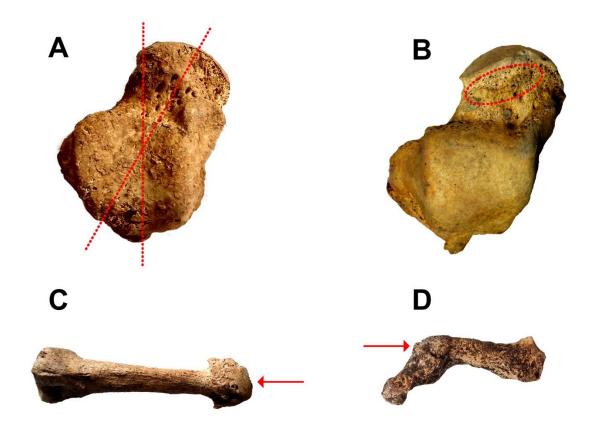
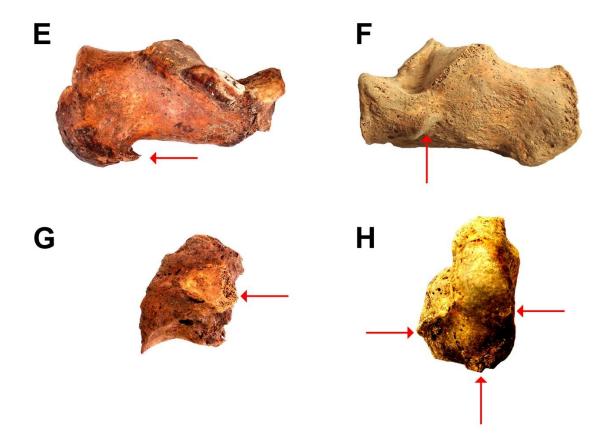


Figure 2



# Figure legends

Fig. 1. A) Head/neck displaced to medial plane (DISHN). B) Periosteal reaction in the talar neck (OMANK). C) Forefoot eburnation (EBANT). D) Merged phalanges (MERGE).

Fig. 2. E) Calcaneal spur (SPURC). F) Hypertrophic peroneal trochlea (PERTR). G) Osteophytes in the surface to the lateral cuneiform of cuboid (CUNOS). H) Osteophytes in cuneiforms-navicular joint (ONCUN).

# **Supplementary material**

TABLE ST1. Frequency of the presence for all the variables in the Spanish series.

Spanish Populations	CA	SSERR	ES	St P	ERE I	V-IX	St Pl	ERE X-	·XIV	AV	INGAN	ΥA	T	ERMEN	IS	GRA	ANOLL	ERS UAB
Variables	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%
OSTST	47	0	0.0	30	2	6.7	46	1	2.2	36	5	13.9	16	0	0	32	5	15.6
PMSTA	47	0	0.0	30	0	0.0	46	0	0.0	36	1	2.8	16	2	12.5	32	0	0.0
SPURC	44	8	18.2	22	3	13.6	44	9	20.5	36	17	47.2	15	4	26.7	31	21	67.7
ACHIL	44	27	61.4	26	21	80.8	45	33	73.3	36	33	91.7	15	8	53.3	31	26	83.9
CTOST	48	0	0.0	31	2	6.5	45	1	2.2	36	1	2.8	16	0	0	32	3	9.4
FLACC	42	0	0.0	25	2	8.0	45	0	0.0	36	1	2.8	15	2	13.3	31	2	6.5
PERTR	39	20	51.3	16	12	75.0	34	24	70.6	31	21	67.7	14	10	71.4	28	20	71.4
TRART	47	5	10.6	29	3	10.3	45	3	6.7	36	3	8.3	16	1	6.3	32	1	3.1
OMANK	48	0	0.0	29	4	13.8	46	5	10.9	46	11	23.9	21	3	14.3	31	10	32.3
TTRNK	48	0	0.0	28	0	0.0	42	1	2.4	46	0	0.0	21	0	0	31	0	0.0
LIPHD	45	0	0.0	29	1	3.4	43	0	0.0	45	4	8.9	20	1	5	31	3	9.7
NECKHD	45	0	0.0	29	0	0.0	43	0	0.0	46	0	0.0	20	0	0	31	1	3.2
OHEAD	45	0	0.0	29	0	0.0	43	0	0.0	46	0	0.0	20	0	0	31	1	3.2
DISHN	45	4	8.9	28	1	3.6	43	0	0.0	46	4	8.7	21	2	9.5	31	1	3.2
EXTTT	48	6	12.5	19	9	47.4	37	8	21.6	46	4	8.7	21	7	33.3	30	4	13.3
TRIGM	48	3	6.3	27	1	3.7	37	1	2.7	46	0	0.0	21	0	0	30	0	0.0
TAILO	48	0	0.0	27	1	3.7	37	1	2.7	46	5	10.9	21	0	0	30	1	3.3
TFOST	36	1	2.8	29	1	3.4	43	1	2.3	0	0	0.0	11	2	18.2	31	6	19.4
CUBFC	33	20	60.6	26	15	57.7	41	22	53.7	0	0	0.0	10	8	80	27	16	59.3

LPCUN	34	10	29.4	29	12	41.4	42	17	40.5	0	0	0.0	11	3	27.3	30	16	53.3
COMMAS	36	0	0.0	30	0	0.0	42	0	0.0	0	0	0.0	11	2	18.2	31	3	9.7
EXTUB	29	3	10.3	26	3	11.5	32	4	12.5	0	0	0.0	10	2	20	30	3	10.0
FLCUC	36	0	0.0	30	0	0.0	46	0	0.0	0	0	0.0	12	1	8.3	28	4	14.3
OCCJN	33	2	6.1	29	5	17.2	47	4	8.5	0	0	0.0	13	3	23.1	28	5	17.9
CUNOS	32	1	3.1	30	2	6.7	46	2	4.3	0	0	0.0	12	3	25	29	2	6.9
ONCUN	33	4	12.1	27	3	11.1	46	9	19.6	0	1	100.0	7	1	14.3	28	3	10.7
STFUS	49	25	51.0	30	9	30.0	46	22	47.8	35	22	62.9	16	8	50	31	19	61.3
COALT	58	1	1.7	34	0	0.0	55	0	0.0	47	0	0.0	21	0	0	32	0	0.0
MERGE	54	2	3.7	34	2	5.9	55	1	1.8	47	0	0.0	21	0	0	32	8	25.0
EBRME	54	0	0.0	34	1	2.9	55	1	1.8	47	0	0.0	11	1	9.1	31	6	19.4
EBANT	51	1	2.0	34	0	0.0	52	1	1.9	0	0	0.0	21	1	4.8	32	15	46.9
OCDRT	56	11	19.6	34	4	11.8	55	18	32.7	47	0	0.0	21	3	14.3	32	8	25.0
HVALG	51	1	2.0	34	2	5.9	55	0	0.0	0	0	0.0	21	1	4.8	31	6	19.4
CCLOS	49	0	0.0	30	1	3.3	47	3	6.4	29	6	20.7	17	0	0	31	3	9.7
OCMTT	33	1	3.0	28	1	3.6	45	5	11.1	0	1	100.0	7	0	0	28	3	10.7
FRACT	54	0	0.0	34	0	0.0	54	0	0.0	47	0	0.0	21	0	0	32	1	3.1

TABLE ST2. Frequency of the presence for all the variables in the Italian series

Italian Populations	C	ASTRC VOLS		MO	ONTERO CAF		AI	LUMIE	RE	G	VILLA GORDIA			LEOPOI CENCEL			CRYPTA BALBI	
Variables	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%
OSTST	18	0	0.0	11	4	36.4	26	1	3.8	11	1	9.1	47	0	0.0	3	0	0.0
PMSTA	18	0	0.0	11	4	36.4	25	1	4.0	10	0	0.0	45	0	0.0	3	0	0.0
SPURC	12	2	16.7	6	3	50.0	22	5	22.7	4	1	25.0	43	5	11.6	3	1	33.3
ACHIL	14	11	78.6	4	3	75.0	25	17	68.0	4	4	100.0	43	39	90.7	2	2	100.0
CTOST	18	2	11.1	7	1	14.3	27	0	0.0	11	1	9.1	43	3	7.0	3	0	0.0
FLACC	15	0	0.0	6	0	0.0	24	0	0.0	10	0	0.0	43	0	0.0	3	0	0.0
PERTR	7	7	100.0	5	4	80.0	17	15	88.2	3	2	66.7	40	34	85.0	3	3	100.0
TRART	15	1	6.7	6	0	0.0	24	3	12.5	10	0	0.0	41	1	2.4	3	0	0.0
OMANK	29	1	3.4	9	0	0.0	27	1	3.7	12	2	16.7	50	2	4.0	7	2	28.6
TTRNK	29	0	0.0	9	0	0.0	27	0	0.0	12	0	0.0	50	0	0.0	7	0	0.0
LIPHD	29	1	3.4	9	0	0.0	24	0	0.0	12	3	25.0	51	1	2.0	7	0	0.0
NECKHD	29	0	0.0	9	0	0.0	24	0	0.0	12	0	0.0	52	0	0.0	7	0	0.0
OHEAD	29	1	3.4	9	0	0.0	24	1	4.2	12	0	0.0	52	0	0.0	7	0	0.0
DISHN	30	1	3.3	9	0	0.0	25	0	0.0	12	0	0.0	51	2	3.9	7	0	0.0
EXTTT	27	5	18.5	8	0	0.0	28	6	21.4	10	2	20.0	48	2	4.2	7	0	0.0
TRIGM	27	3	11.1	8	2	25.0	28	1	3.6	10	2	20.0	48	0	0.0	7	0	0.0
TAILO	27	1	3.7	8	0	0.0	28	0	0.0	10	1	10.0	48	1	2.1	7	0	0.0
TFOST	16	0	0.0	3	0	0.0	24	0	0.0	8	2	25.0	42	0	0.0	7	0	0.0
CUBFC	12	6	50.0	2	1	50.0	21	4	19.0	7	3	42.9	39	23	59.0	1	0	0.0
LPCUN	13	7	53.8	1	1	100.0	23	5	21.7	8	7	87.5	42	16	38.1	1	1	100.0
COMMAS	16	0	0.0	3	0	0.0	24	0	0.0	8	0	0.0	42	0	0.0	1	1	100.0

EXTUB	7	1	14.3	2	1	50.0	14	0	0.0	7	0	0.0	33	5	15.2	1	0	0.0
	<u> </u>	1			1											1		
FLCUC	13	0	0.0	3	0	0.0	22	0	0.0	9	0	0.0	44	1	2.3	2	0	0.0
OCCJN	13	0	0.0	2	0	0.0	20	0	0.0	9	1	11.1	45	1	2.2	1	1	100.0
CUNOS	13	0	0.0	3	0	0.0	24	0	0.0	8	0	0.0	44	0	0.0	2	0	0.0
ONCUN	7	0	0.0	3	0	0.0	26	0	0.0	8	0	0.0	46	1	2.2	1	0	0.0
STFUS	18	11	61.1	7	5	71.4	27	19	70.4	11	7	63.6	45	21	46.7	3	0	0.0
COALT	35	0	0.0	11	0	0.0	30	1	3.3	15	0	0.0	61	0	0.0	8	0	0.0
MERGE	34	0	0.0	10	0	0.0	30	0	0.0	15	0	0.0	61	0	0.0	8	0	0.0
EBRME	35	1	2.9	10	0	0.0	30	0	0.0	15	2	13.3	61	0	0.0	8	0	0.0
EBANT	11	1	9.1	5	0	0.0	29	1	3.4	7	0	0.0	50	0	0.0	1	0	0.0
OCDRT	34	3	8.8	10	1	10.0	30	8	26.7	15	3	20.0	61	5	8.2	7	1	14.3
HVALG	2	0	0.0	4	1	25.0	17	0	0.0	7	0	0.0	22	0	0.0	0	0	0.0
CCLOS	15	0	0.0	6	0	0.0	25	1	4.0	11	1	9.1	44	5	11.4	2	1	50.0
OCMTT	7	1	14.3	3	1	33.3	26	2	7.7	8	0	0.0	46	2	4.3	1	0	0.0
FRACT	35	0	0.0	10	0	0.0	30	0	0.0	15	0	0.0	61	0	0.0	8	0	0.0

TABLE ST2 (Continuation). Frequency of the presence for all the variables in the Italian series.

Italian Populations	El	SAN RCOLA		G	UIDON	NIA	A	QUINO		C	OLONN	A
Variables	N	Presence	%	N	Presence	%	N	Presence	%	N	Presence	%
OSTST	4	0	0.0	10	0	0.0	2	0	0	17	1	5.9
PMSTA	4	0	0.0	10	0	0.0	2	0	0	17	0	0.0
SPURC	2	0	0.0	10	1	10.0	2	1	50	10	3	30.0
ACHIL	2	2	100.0	10	7	70.0	2	2	100	10	10	100.0
CTOST	4	0	0.0	10	0	0.0	2	0	0	16	0	0.0
FLACC	3	0	0.0	7	0	0.0	2	0	0	14	0	0.0
PERTR	1	0	0.0	2	1	50.0	1	0	0	6	6	100.0
TRART	1	0	0.0	8	0	0.0	2	0	0	11	1	9.1
OMANK	11	0	0.0	11	0	0.0	2	0	0	19	2	10.5
TTRNK	11	0	0.0	11	0	0.0	2	0	0	19	0	0.0
LIPHD	12	1	8.3	11	0	0.0	2	0	0	19	1	5.3
NECKHD	12	0	0.0	11	0	0.0	2	0	0	19	0	0.0
OHEAD	12	0	0.0	11	0	0.0	2	0	0	19	0	0.0
DISHN	12	0	0.0	11	0	0.0	2	0	0	20	0	0.0
EXTTT	7	2	28.6	6	0	0.0	2	1	50	18	0	0.0
TRIGM	7	0	0.0	6	0	0.0	1	0	0	18	3	16.7
TAILO	7	0	0.0	6	0	0.0	1	0	0	18	0	0.0
TFOST	5	0	0.0	6	0	0.0	1	0	0	16	0	0.0
CUBFC	4	3	75.0	6	2	33.3	1	1	100	14	9	64.3
LPCUN	5	1	20.0	6	4	66.7	1	1	100	16	6	37.5
COMMAS	5	0	0.0	6	0	0.0	1	0	0	16	0	0.0
EXTUB	4	1	25.0	6	0	0.0	1	0	0	11	1	9.1

EL CUC		0	0.0	5	0	0.0	1	0	0	1.0	0	0.0
FLCUC	6	Ü	0.0	3	0	0.0	1	0	0	16	0	0.0
OCCJN	6	0	0.0	4	0	0.0	1	0	0	16	1	6.3
CUNOS	4	0	0.0	6	0	0.0	1	0	0	16	0	0.0
ONCUN	5	0	0.0	7	0	0.0	2	0	0	17	0	0.0
STFUS	5	3	60.0	10	4	40.0	2	1	50	14	9	64.3
COALT	15	0	0.0	14	0	0.0	2	0	0	25	0	0.0
MERGE	14	0	0.0	14	0	0.0	2	0	0	24	0	0.0
EBRME	15	0	0.0	14	0	0.0	2	0	0	24	0	0.0
EBANT	2	0	0.0	12	0	0.0	2	0	0	16	0	0.0
OCDRT	15	1	6.7	14	1	7.1	2	0	0	23	5	21.7
HVALG	0	0	0.0	3	0	0.0	2	1	50	8	0	0.0
CCLOS	4	0	0.0	6	0	0.0	2	0	0	14	0	0.0
OCMTT	4	0	0.0	7	0	0.0	2	0	0	17	0	0.0
FRACT	15	0	0.0	14	0	0.0	2	0	0	24	0	0.0

TABLE ST3. Frequency of the presence for all the variables in the Omani series.

Omani Population	RAS AL-HAMRA 5						
Variables	N	Presence	%	Variables	N	Presence	%
OSTST	14	0	0.0	CUBFC	31	21	67.7
PMSTA	13	1	7.7	LPCUN	35	12	34.3
SPURC	13	1	7.7	COMMAS	36	1	2.8
ACHIL	15	10	66.7	EXTUB	33	6	18.2
CTOST	16	1	6.3	FLCUC	29	0	0.0
FLACC	15	0	0.0	OCCJN	29	3	10.3
PERTR	12	10	83.3	CUNOS	31	1	3.2
TRART	13	0	0.0	ONCUN	33	2	6.1
OMANK	33	4	12.1	STFUS	12	9	75.0
TTRNK	33	0	0.0	COALT	55	0	0.0
LIPHD	33	0	0.0	MERGE	55	2	3.6
NECKHD	33	1	3.0	EBRME	52	3	5.8
OHEAD	33	0	0.0	EBANT	37	6	16.2
DISHN	30	3	10.0	OCDRT	55	9	16.4
EXTTT	23	1	4.3	HVALG	18	3	16.7
TRIGM	23	1	4.3	CCLOS	14	1	7.1
TAILO	23	1	4.3	OCMTT	33	2	6.1
TFOST	36	3	8.3	FRACT	55	1	1.8

2.4. A NEW METHODOLOGY TO ESTIMATE FLAT FOOT IN
SKELETAL REMAINS. THE EXAMPLE OF MEDITERRANEAN COLLECTIONS
"Without laboratories, men of science are soldiers without arms".  Louis Pasteur
Bouls I distout
Saldías E, Malgosa A, Xavier Jordana, Martínez-Labarga C, Alfredo Coppa, Mauro Rubini, Isidro A. <i>A new methodology to estimate Pes planus from dry bones</i> . Submitted to: American Journal of Physical Anthropology (September 2019).

A new methodology to estimate flat foot in skeletal remains. The example of

Mediterranean collections.

Running title: New methodology to estimate Pes planus in dry bones

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## **Abstract**

# **Objectives:**

Flat foot (*pes planus*) is considered a defect in the alignment of foot sole, collapsing the longitudinal arch that is in contact with the ground. Depending on the degree of this anomaly, it could produce pain and difficulty in the gait. *Pes planus* has been well-studied in the clinical literature and paleoanthropology but has not been approached in extension in bioarchaeology. In this work, we propose a new metric and morphological method to discriminate non-pathological flat foot in dry bones.

### **Materials and methods:**

We studied 390 pairs of adult foot in a fair state of preservation from archaeological contexts from Spain, Italy and Oman. Morphological variability, angles and dimensions of the healthy and pathological flat feet were analyzed.

## **Results:**

We found a correlation between the presence of flat foot and some morphological and metric features, mainly in subtalar and Chopart joints. These results are expressed through a combination of morphological and metric variables, which are useful to discriminate between these two groups.

### **Conclusions:**

Not significant differences of pathological flat foot frequencies between the Spanish and Italian series were found across centuries. However, we noticed a notorious increase in the contemporary collection, probably due to the consequences of a life expectancy rise and modern styles of footwear.

**Keywords:** Pes planus, longitudinal arch, foot dimensions, archaeological collections.

**Declarations of interest:** none.

# **Research Highlights:**

Morphology and dimensions in dry bones are related with presence of flat foot

Historical series showed similar frequencies, but a notorious rise in modern pathological feet

Life span and modern footwear are key factors for the development of flat foot

## Introduction

In our species, the foot is one of the structures with more anatomical evolutionary specializations. Thus, the bipedalism and the stand posture correspond to the most important features of the hominins (Zipfel, B. DeSilva, J. Kidd, R. Carlson, K. Churchill, S. Berger, 2011). According to Isidro (Isidro, 1997), in the primate evolution context, the bipedalism is an anatomic adaptation to survive to the geo-climatic changes, involving mostly the morphomechanical re-design of the lumbar zone, pelvic girdle, knee and feet.

In modern humans, foot bones are strong structures able to hold on the weight of the body and allow a correct displacement. This bipedal locomotion allows a vertical posture and releasing the upper extremities. The foot arches (lateral longitudinal, medial longitudinal and transverse) compose a plantar vault and are the consequence of the bipedalism. The presence of these arches permit to modern foot to distribute the corporal weight through a better performance, involving propulsion, buffering and stabilizing movements in the gait (Fan, Fan, Li, Lv, & Luo, 2011).

The plantar vault is composed by the interaction of ligaments, muscles and bones. In detail, these bones are joint by are calcaneus-navicular ligament and calcaneus-cuboid ligament. The above, and other tendons like *tibialis anterior*, *tibialis posterior*, and *flexor hallucis longus* (Hansen, 2015; Kapandji, 1998; Parra, 2006) support to the talus-navicular joint. In relation with the height of the longitudinal arch of the plantar vault, we can classify the foot into three big categories, neutral (normal foot), high arch (*pes cavus*), and low (*pes planus* or flat foot). In children, the flat foot is normal because their arches are still not form (Pfeiffer, Kotz, Ledl, Hauser, & Sluga, 2006); however, flat and high arches are anomalous in adults. Flat feet be produced by diverse factors, for instance muscular injuries, fractures, and anomalous bone shape/position among others (Louie, Sangeorzan, Fassbind, & Ledoux, 2014).

Likewise, aging might increase the risk of skeletal disorders in the foot (Glencross & Sawchuk, 2003). With age, ligaments and joint structures start to lose their capacity to maintain a good performance during movements, producing wear and degeneration of the joints, deforming and changing the gait of individuals and the quality of movements. In this

sense, flat foot is one of the most common anomalies of the alignment of the plantar arch in the population. This condition in juvenile and adult individuals is due to the collapse of the longitudinal arch, producing the lack of the curvature, contacting the plantar vault with the surface. Kulowski (1939) was the first who observed the relationship between *tibialis posterior* tendon rupture and *pes planus* (Jahss, 1982). In the same line, to authors like Pomeroy and collaborators (Pomeroy, Pike, Beals, & Manoli, 1999), the dysfunction of the *tibialis posterior* tendon is one of the main causes of the adult flat foot and its clinical treatment has been profusely analyzed in the literature. *Pes planus* also can be produced for many other factors as bone or muscular traumas/dysfunction, some kind of obesity, degenerative diseases (arthritis), neuromuscular imbalance (spastic flat foot) and defects in the bone development (e.g. tarsal coalition) (Lee et al., 2005; Pandey, Pal, Kumar, & Singh, 2013).

The flat foot has been classified in four stages (Abousayed, Tartaglione, Rosenbaum, & Dipreta, 2016; Pomeroy et al., 1999) related with the degree of damage at level of muscles, ligaments and bones, and clinic signs. The stage I is the mildest and IV the most severe. According to the Pomeroy classification (1999), the fourth stage correspond to a painful phase, loss of flexibility in the hindfoot/midfoot, arthritic changes in the ankle and valgus deformation, among others features.

Therefore, a clear-cut diagnosis of *pes planus* only can be obtained in clinical patients, including radiological methods like Meary angle, calcaneal pitch angle, talo-navicular uncoveraged angle, among others (Lee et al., 2005; Lin et al., 2015). It is essential to observe the complete and articulated foot structure to evaluate this anomaly. According with Jahss (Jahss, 1991), there are a series of clinical characteristics (external examination, x-rays) to determine the existence of flat feet, implying forefoot abduction and pronation, pronounced valgus of the heel, increase of verticality of talus, rotation/abduction of navicular and the decrease of the medial arch.

In like manner, Jahss also considered wide osteological parameters to determinate *pes planus*, including five new morphological features, which could be associated to the severe damage stages, including anterior and/or posterior subtalar joint arthropathy in the hindfoot. In the midfoot, flat foot is related with the increase of the lateral joint surface of the head of the

talus/sentinel osteophyte in the neck of the talus, and alteration of the insertion of *tibialis posterior* tendon (navicular abducted/rotated/pronation). Additionally, in the forefoot, the alterations in sesamoid and first metatarsal head (sesamoid groove) suggesting an internal rotation of hallux, as other signs of *pes planus*. Nonetheless, there are not included normal variability, detailed pathological features or measurements, which could influence its presence.

Consequently, to diagnose a flat foot it is necessary to analyze the whole foot structure and it is not easy to obtain evidence of foot shaping from dry bones. In this sense, literature in bioarcheology about flat foot is very scarce, but some evolutionary approaches have been made. Comparative analyses between apes and humans suggest that an apelike foot has a structure compatible with flat foot due to its specialization and ability to arboreal life. Conversely, humanlike feet need the plantar arches to optimize an upright position and walk.

Therefore, the studies related to the biomechanical of our ancestors foot show a mixture of human and apelike features, compatibles with flat foot. For instance, according with morphology of modern humans, there are notorious differences with individuals of *Australopitecus afarensis* (DeSilva & Throckmorton, 2010). Lucy remains (A.L.288-1) had flat foot, but other remains from Hadar, Ethiopia (A.L.333-6 and A.L.333-7) show a possible rear-foot arch. In this line, considering research in the fore/midfoot, the Kromdraai talar remains (*Paranthropus robustus*) exhibit similar features to those of Olduvai Hominid 8 (OH8), and its horizontal angle is nearest to that of apes (*Gorilla gorilla, Pan troglodytes*) than humans (continental Europeans, Bushmen, Anglo-Saxon/Romano-British) (Day & Wood, 1968). On the other hand, Zipfel et al (Zipfel, B. DeSilva, J. Kidd, R. Carlson, K. Churchill, S. Berger, 2011) proposed that the locomotion of *Australopitecus sediba* show a unique form of bipedalism and a minor degree of arboreality, derived from his primitive and hominin foot. His feet show a cuboid facet in a similar angle than humans, which suggest foot arching and a smooth surface of calcaneal tuber.

Also, Gebo and Schwartz (Gebo & Schwartz, 2006) suggested that the talus from the zone of Omo (323-76-898, unknown taxon) is more similar with the samples of *Homo sapiens* and Koobi Fora (KNM-ER-813, a probably *Australopithecus boisei* or *Homo*) than *Pan*. Moreover, Jungers et al. (Jungers et al., 2009), which studied the remains of the *Homo* 

*floresiensis*, concluded that his feet show primitive features for the genus *Homo*, like long lateral toes, short hallucal metatarsal, re-evolving short hind limbs and flat foot, among others.

Despite these exceptional mentioned cases (isolated, individuals) it is complicate to obtain data about flat foot in historical remains. Moreover, it is difficult to extend this kind of analysis in populations. Therefore, the main aim of this work is to develop a protocol of diagnosis that might be useful for non-articulated and incomplete feet, as fossil and archaeological cases. Our work is based on the hypothesis those feet bones with pathological pes planus are related with several damage or even several anomalies in the talo-navicular and subtalar joints that will reduce the capacity of the muscles to keep the longitudinal arch. Therefore, morphology, angles and dimensions of involved bones might be observed.

For this purpose, we analyzed several archaeological populations. Considering osteological features of severe degrees of *pes planus* in feet bones (level III and IV) (Abousayed et al., 2016; Jahss, 1991; Lee et al., 2005; Pomeroy et al., 1999) and our proposals about bone metric and morphological features of flat foot bones, we want to create a new diagnostic tool to determinate non-pathological flat foot from dry bones.

### Material and methods

390 pairs of adult tarsal bones (206 males, 121 females and 63 indeterminate) coming from archaeological sites of Spain and Italy were used. The Spanish series come from Catalonia: Esglésies de Sant Pere, Casserres, Avinganya and Castell de Termens. The Italian series come from Lazio region: Monterotondo, Villa Gordiani, Colonna and Allumiere. Additionally, the Ras al-Hamra 5 collection from the Sultanate of Oman was considered due to its large number of individuals affected by sacral dysraphism. Besides, we added the contemporary series of Granollers/UAB in order to contrast with ancient conditions. The samples, mostly incomplete feet, showed a fair state of preservation. All the specimens with high degree of taphonomic damage were discarded (Table 1).

The whole collection was divided into three groups based on their pathologies and the affected area. The first group belongs to the pathological feet samples that meet the criteria of Level III and IV of flat foot. We considered as pathological flat feet to all the specimens

that have a great bone damage produced by degenerative diseases in hind/midfoot and/or forefoot (Jahss, 1991). Mostly, the samples show consequences from osteoarthritis in high degree as common features. The affected areas involved the Chopart joint and the calcaneus-talus joint.

The second group collect the remains of all the individuals who did not show pathologies that affect the foot (neither extreme measurements), which could be considered as normal feet. In the third group, we included all the indeterminate individuals that we could not confirm or discard the possibility that had flat foot because they were affected for one or more non-severe pathologies or anomalies.

To separate the groups, we used a series of anatomical features observed in the whole collection, located in the main joints of the foot bones (Table 2). Most of the variables belong to arthropathies from the subtalar joint, one of the most important articulations related with the presence of pathological *pes planus* (Jahss, 1991; Lee et al., 2005; Pomeroy et al., 1999). Furthermore, we added other variables to check their implication with the *pes planus* presence, which were present in a significant number of samples. These variables correspond to alteration in tuberosity of navicular (EXTUB) and the presence of *osteochondritis dissecans* in the foot joints (OCDRT). We have a particular interest in EXTUB because is the insertion point of the *tibialis posterior* tendon, one of the most important structures in the stabilization of longitudinal arch, (Jahss, 1991; Lee et al., 2005) (Fig.1) (Table 2).

To develop a method that allows us to discriminate flat foot of level I and II in dry bones (maybe some specimens of the group 3), we considered the morphological features in foot bone samples, which could be implied in this condition (group 1) and that be totally absent in the second group.

On the other hand, we checked the dimensions of the talus, navicular and cuboid, which belong to involved structures in the gait movements. Probably, due to their importance in the midtarsal joint, also they might be related with flat feet presence.

The measurements and angles were selected for its localization and easy replicability in the midtarsal, and they were proposed by other authors (Martin & Saller, 1957), (Day & Wood, 1968), (Gebo & Schwartz, 2006) (Harris & Case, 2012). We considered three dimensions

from cuboid (Breadth of the cuboid BCUB; Length of the cuboid LCUB; Height of the cuboid HCUB), two from navicular (Maximum length of the talar facet LMAXTAL and Sagittal length of the talar facet LSAGTAL), and two from talus (Length of the head of the talus LHEADT and Breadth of the head of talus BHEADT). In addition, we analyzed two angles in the talus (Angle of torsion of the talar head TORSANG, and Horizontal angle of the neck of the talus NECKANG) (Fig.2).

To determinate the influence of the populations in the results, and the relationship between *pes planus* and the morphoscopic variables, we applied a Chi-square test. Moreover, we applied a binary logistic regression to the significant features whose results would indicate the correct classification of the individuals with non-pathological condition.

To check the intra-observer error in bone measurements, 50 feet bones from 30 individuals were chosen randomly, whose number corresponds to the 14% of the complete collections. The first author (ES) checked twice the dimensions in different periods, contrasting with the Technical Error of Measurement test (TEM) (Perini, Oliveira, Ornellas, & Oliveira, 2005). In addition, we calculated the relative and absolute TEM.

Our samples did not show a normal distribution. The non-parametric Kruskal-Wallis test and Mann-Whitney U test was applied to check the existence of significant differences between populations, and to find a relationship between the metric variables and the presence of pathological *pes planus*.

Afterwards, considering these results, we used a Receiver Operating Characteristic (ROC) curve analysis to evaluate the performance of the significant variables. Its correct classification to discriminate *pes planus* is indicated by the area under the ROC curve (AUC), whose discriminative ability is higher when is more near to 1. When the AUC of a variable is 0.5, cannot distinguish the individuals with and without *pes planus*. Furthermore, the ROC curve results allow to choose cut-off points (threshold) with the best correct accuracy to discriminate between these two groups. However, all the individuals with values between these cut-off points are considered indeterminate. In our analysis, we only show the largest AUC and threshold values of the significant variables.

Finally, through to the morphological and metrical results, the cut-off points and the binary logistic regression results were applied to the indeterminate group (N: 176), to check the presence or absence of non-pathological flat foot (Level I and II). From the results, we got the probable frequencies of pathological and non-pathological *pes planus* in whole our series.

### **Results**

From the 390 pairs of adult tarsal bones checked from Spanish, Italian and Omani collections, individuals from pathological flat foot group (n: 29= 5.9%) and healthy feet group (n:185), were analyzed under metrical and morphological methods. From the pathological series, 15/254 individuals are from Spain (12%), 9/81 from Italy (11.1%), and 5 out 55 from Oman (9.1%) (Table 1).

The Chi-square test demonstrated that none morphological variables were influenced by the population factor (Table 3). Moreover, regarding that all these variables demonstrated to have a relationship with flat foot (p<0.05) (Table 3), we applied a binary logistic regression to check the variables with better performance to discriminate. According to the analysis, the combination of PMSTA, CTOST, TRART, OHEAD, DISHN, WITTT, TRIGM, EXTUB, OCDRT and CCLOS showed a 73% of correct allocation accuracy for *pes planus* individuals and a 98% to non-*pes planus*, with a 91% of reliability, in general.

On the other hand, the intra-observer analysis of the morphometric method indicated an acceptable Relative TEM and low rate of error in the Absolute TEM (<0.7) (Table 4).

Table 5 indicates the descriptive statistics of the nine metric variables, divided by the presence/absence of *pes planus*. The Angle of torsion of the talar head (TORSANG) show the highest variances in the pathological flat feet group.

Considering our samples demonstrated a non-normal distribution, we applied a Kruskal-Wallis analysis to ascertain the existence of significant differences between populations. The results indicate that the variables LHEADT, BHEADT and LCUB were influenced by this factor (Table 5), which were discarded to future analyses. Afterwards, Mann-Whitney U test was used to find a relationship between the metric variables and the presence of flat foot.

Angle of neck of talus (NECKANG) and height of the cuboid (HCUB) were the only variables that showed significant differences (p: 0.026 and 0.009) (Table 5).

Afterwards, we applied a receiver operating characteristics curve (ROC) to evaluate the discriminative performance of both variables (Table 6). This test allows the identification of the best threshold values (cut points) to every measurement, regarding their sensitivity (true positive proportion) and 1-specificity (false positive proportion). For instance, NECKANG has a value in individuals with flat foot of  $\geq 17.5^{\circ}$ , with a sensitivity of 0.571 and 1-specificity of 0.254. The above means that 57% of *pes planus* and 25% of healthy feet could have 17° or higher values for NECKANG. Thus, the probability that a talus specimen corresponds to have flat foot, with a NECKANG value of 17.5 ° or larger, is a 75%. On the other hand, all the values  $\leq$  to 16.5 ° are considered as without flat feet, and those between the mentioned cut-off points correspond to indeterminate individuals.

The variables NECKANG and HCUB demonstrated by separate, an acceptable correct accuracy to discriminate individuals with/without flat feet, but with low AUC and sensitivity (Table 6), probably by the small number of individuals with this alteration (N:29).

Finally, we applied the morphoscopic and metric results to 176 individuals from the indeterminate group (group 3). To increase the correct accuracy, we considered as individuals with non-pathological flat foot to all the samples that demonstrated positive matches through to at least two methods (morphoscopic, NECKANG, HCUB). All the individuals with values out from our parameters were consider as "without flat foot", and those with only one match, "indeterminate".

The results indicated that we found 30 of individuals with non-pathological flat foot and adding all the samples and affected bones from the collections, the total frequency of *pes planus* is a 15.1% (59/390) (Table 7). Furthermore, the flat foot condition, in any variant, is more common in male group in contrast with females  $(41 \frac{3}{14})$ . The latter means that sex ratio is 2.9.

Nonetheless, we must consider also that the number of males is higher (206/390=52.8%) than the females (121/390=31%) or the indeterminate individuals (63/390=16.1%) in our whole series (Table 7).

### **Discussion**

Morphological results from pedal elements from Spanish and Italian archaeological series indicated that the combination of PMSTA, CTOST, TRART, OHEAD, DISHN, WITTT, TRIGM, EXTUB, OCDRT and CCLOS variables are related with flat foot and provide a useful tool to discriminate between pathological flat foot and no affected foot. Analyzing the variables separately and taking in consideration the *pes planus* criteria by Jahss (Jahss, 1991), most of them are arthropathy of the subtalar joint (PMSTA, CTOST, TRART, TRIGM, DISHN, OHEAD, and WITTT). On the other hand, OCDRT and CCLOS are two features that were not considered into the Jahss classification. *Osteochondritis dissecans* (OCDRT) is a non-inflammatory condition that produce localized areas of necrosis in diarthrodial joints (Aufderheide & Rodriguez-Martin, 1998b), which could affect the foot joints; while Calcaneal-cuboid osteophyte (CCLOS) could be directly related with the morphometrical results of height of the cuboid (HCUB). We did not find anomalies with significant results in the forefoot.

Besides, as we hypothesized at first, EXTUB is related with the presence of *pes planus* due to its importance in the insertion of *tibialis posterior* tendon. This joint, and especially the *spring* ligament (plantar calcaneo-navicular ligament), provide the main support of the longitudinal arch. A dysfunction of this zone could tear the structure, producing a lack of stabilization of the arch and pain (Abousayed et al., 2016; DeSilva & Throckmorton, 2010; Franco, 1987).

According with our metric results, the horizontal angle of the neck of the talus (NECKANG) demonstrated a relationship with the presence of flat foot. This measurement has been worked on previous research about human evolution. The investigations of Day & Wood (Day & Wood, 1968) and Gebo & Schwartz (Gebo & Schwartz, 2006), who applied angles and measures to the talus, arrived at the same conclusions. Through a comparative study of *Homo sapiens* and different species, including ancestors and modern apes, they observed that modern humans have a small NECKANG, in contrast with apes (like *Pan troglodytes* or *Gorilla gorilla*), which developed a wide angle. In *Homo sapiens* samples, the media of their results is around 19°, but the divergence between modern human groups can change slightly.

Day & Wood observed that the maximum difference between groups was 2 degrees (18° in Barnett samples vs Cassington/Bushmen series with 20°)(Day & Wood, 1968).

High values of NECKANG usually suggest an opposable hallux, observation shared by Duckworth (Duckworth, 1904) and Volkov (Day & Wood, 1968). A small angle is related with an adducted hallux (*Homo sapiens*), and a foot with a toe able to the divergence is indicative of flat foot. In fact, Jahss (Jahss, 1991) proposed that the human foot differ with the rest of primates in three features: the reduction of the size of the foot and the presence of a non-opposable hallux, which allow the efficiency of the transmission of corporal weight. The third feature corresponds to the presence of longitudinal and pronounce transverse arch, due for the disposition of the ligaments and muscles to spread the weight of the body to the foot. These features indirectly give support to the relationship between the angle of the talus neck and flat foot. In our results, the cut-off point of NECKANG is 17.5° and the maximum value in the pathological flat foot group is 33°, which is indicative of a notorious angular anomaly or an anomaly in the configuration of the talo-navicular joint.

The modification of the talo-navicular joint in individuals with flat foot have been studied previously. Clinical studies, comparing patients with asymptomatic and symptomatic *pes planus*, show the change of position of the navicular regard to the talar head, with a lateral displacement of the navicular to lateral/plantar position (Louie et al., 2014; Moraleda & Mubarak, 2011; Peeters et al., 2013).

Regarding to the height of the cuboid (HCUB) and although the features of the cuboid usually are not mentioned as the causes of *pes planus*, we found a relationship with this condition. Our results show that HCUB is highest when foot is flat. A possible explanation could be that some factors, like consequences of osteological diseases, might modify the structure of this bone, generating altogether flat foot with other bone alterations. Other reason could be that decrease of the medial longitudinal arch, might produce a muscular effort of this zone, to stabilize the foot, which additionally could alter the insertions of the cuboid like the *peroneus longus* tendon, the calcaneus-cuboid ligament and the lateral longitudinal arch (Hansen, 2015; Kapandji, 1998).

Based on our observations, the affected areas by pathological *pes planus* are located mostly in subtalar and Chopart joint. The above might be due to the changes of calcaneus structure and in the calcaneus-cuboid joint, visualized in both results.

Currently, flat foot is an anomaly which is present in approximately the 3-19% of the population (Ferciot, 1972; Louie et al., 2014), being more frequently in women population (3:1), mainly with obesity and over 40 years old (Parra, 2006). There are some studies, mentioned in Lee et al (Lee et al., 2005), which confirm this rank. For instance, Harris and Beath (R. I. Harris & Beath, 1948) found a 6% of simple hypermobile flat foot with tight *fascia plantaris*, 15% of simple hypermobile flat foot and a 2% with tarsal coalition in 3619 soldiers of Royal Canadian Army. Even though we found a prevalence of flat foot in male group, we did not have a similar number of samples of females to contrast this tendency. Considering the nature of the osteological series, their information corresponds to estimations obtained through to anthropological methods. Granollers is the only documented collection from this study and flat foot was present in more females than males.

Despite the number of individuals affected with flat foot being small (N: 29) and we estimated 30 non-pathological *pes planus* in the group 3 (indeterminate), our final results (59/390 individuals, 14.8%) are into the world frequencies. Despite a small number of incidences, the methodologies classified with an acceptable percent of reliability, especially individuals with healthy feet. Thus, we recommend to consider at least two methods (NECKANG, HCUB, morphological features) with positive ranges of flat feet to determine it. Adding new pathological flat foot individuals might increase the percent of correct classification to discriminate this anomaly.

Excepting to Granollers from this consideration, the frequencies of pathological flat feet from bioarchaeological populations in Spain and Italy are the same (9 vs 9 individuals). Although is a small collection (determinates), one third of the feet remains from Villa Gordiani developed this pathologic alteration. This osteological series located in Prenestino quarter, Rome, Italy, is from a monumental complex which have been associated as propriety of imperial families (Palombi & Leone, 2008). In this archaeological site is possible to find structures related with different activities, including residence, religion, functional and

mortuary structures. Human bones were extracted from basilica-mausoleum, one of the only one structures that remains standing today. Considering the features of this collection, we hypothesized that high *pes planus* frequencies might be due to their imperial well-off life, which could allow to individuals to get high age over the mean. Despite, Roman footwear have been many changes through the years (Van Driel-Murray, 2013), in the first centuries, shoes (with some exceptions) had a contact almost direct with the ground. To Sebesta & Bonfante (Sebesta & Bonfante, 2001) the *calceus*, a kind of shoes /shoe-boots, made by soft leather and fastened in front with thongs, were worn specifically with toga, forbid to slaves. Thus, footwear is not a factor that could much influence to the presence of flat foot in Villa Gordiani. Besides, regarding that these remains belong to imperial family, genetic factors might explain the high frequencies.

Granollers is a contemporary series of individuals from Granollers cemetery, in the province of Barcelona, Spain (Saldias, Malgosa, Jordana, & Isidro, 2016). This collection is the only one from our study where the individuals have a mean age of over 60 years old. From the series with determinate flat foot, they have the highest values, 6/10 individuals showed compatible pathological features. Age is an important factor to develop degenerative diseases as osteoarthritis, which can influence to the presence of pes planus. However, modern footwear is an important factor that has been widely studied related with foot pathologies, mostly in females. To some cultures, the shoes were designed to an ostentatious display more than its functionality, or to validate a social status (Mays, 2005). Some authors (Août & Aerts, 2008; Kapandji, 1998) mentioned the differences between individuals who wear shoes and barefoot, especially in the distribution of the forces in the gait. Sachithanandam and Joseph (Sachithanandam & Joseph, 1995) studied static footprints of 1846 individuals over 16 years old (with a mean of 24) to check the age where the shoes started to affect in the presence of flat foot. Using their medical history, they proposed that there is a relationship between pes planus and the wear of shoes in the early childhood, finding prevalence between 3-4% in children wore shoes before 15 years old. In addition, due to narrow shoes and the compression of the fingers, the feet in adults also could be affected. Mays (Mays, 2005) studied two series from early and later Medieval period from Ipswich, England. He noticed that foot deformities as the presence of hallux valgus notoriously increased mainly in the

high-level society of the later period, probable due to the wear of pointed and narrow shoes, compressing the hallux, which is supported for documentary evidence found.

On the opposite part, we have the Ras al-Hamra 5 series. It is an osteological collection coming from the Arabian coast of the Sultanate of Oman, dating from Neolithic period (3700-3400 BCE). They were fishermen who collected molluscs and sea resources (Macchiarelli, 1989) that did not used footwear. Part of this series is located in Sapienza Università di Roma (Italy), whose individuals were affected by different degrees of open sacral canal, reported previously by Coppa (Coppa, 2012).

In our results, taking into consideration the individuals with this sacral defect, the frequency of pathological flat foot group (5/34) is equivalent to 14.7 %. Nonetheless, we could not confirm the relationship between this sacral defect and *pes planus* due we did not find severe vertebral compromise. Additionally, footwear is not a factor to consider because in these centuries did not exist footwear that constrict or modify the feet and age could neither be the cause of the alterations because the mean age of the population only is between 20 to 40 years old. There are no clear causes about this disease. However, some studies of sacral defects in ancient skeletal populations have related its developed with genetic-environment factors, high levels of arsenic, bio cultural isolation and endogamy (Aufderheide & Rodriguez-Martin, 1998a; Boston & Arriaza, 2009).

We did not find differences in the frequency of the affected individuals from Spain and Italy through the centuries, except for the contemporary series. The notorious increase of *pes planus* frequency in Granollers collection could be due to two factors that did not present the mentioned historical series. As we mentioned before, high increase of life expectancy (and their consequences) and modern footwear that constrict the feet, could explain our results.

Summarizing, the present study demonstrated the relationship between flat foot with metric variables, which were expressed through to ROC curve analysis, being the horizontal angle of the neck of talus (NECKANG) and the height of the cuboid (HCUB) the variables that yielded significant differences.

Considering the morphological results, the combination of PMSTA, CTOST, TRART, OHEAD, DISHN, WITTT, TRIGM, EXTUB, OCDRT and CCLOS variables demonstrated to discriminate between individuals with and without flat foot. Thus, the main affected zones in the foot involved subtalar and Chopart joints. Despite our study revealed that our frequencies in determinate/indeterminate *pes planus* groups are similar than world mean, the affected number of individuals are small. An increase of pathological *pes planus* could increase the correct accuracy to discriminate this anomaly.

In relation to population diversity, archaeological collections from Spain and Italy have similar frequencies, excepting the contemporary series (Granollers XX CE), which developed a high number of affected samples. According these results, even though *pes planus* could be produced by different factors like activity, genetic, diseases or age, the kind of footwear is also is an important cultural factor to be considered. Our methodological proposals might be useful in population studies or to define biological characteristics.

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# **Tables**

TABLE 1. Distribution of individuals with/without pathological flat foot. Divided by country, series, period and sex.

N: Total number of individuals analyzed.

			F	PATHOLOGICA	AL FLAT FOO	T		HEALTH	Y FOOT		INDET				TOTAL
COUNTRY	POPULATION	PERIOD	MALE	FEMALE	INDET	TOTAL	MALE	FEMALE	INDET	TOTAL	MALE	FEMALE	INDET	TOTAL	N
	Casserres	VII-IX c.CE	1	1	0	2	17	14	0	31	21	4	0	25	58
	St. Pere	IV-IX/X-XIV c.CE	3	0	0	3	27	13	0	40	31	21	0	52	95
SPAIN	Avinganya	IX-XIV c.CE	0	0	3	3	0	0	27	27	0	0	18	18	48
	Termens	XVIII-XIX c.CE	1	0	0	1	5	5	0	10	5	5	0	10	21
	Granollers	XX c.CE	2	4	0	6	1	3	0	4	14	8	0	22	32
	Monterotondo	II-III c.CE	0	1	0	1	1	4	2	7	2	0	1	3	11
ITALY	Villa Gordiani	V-VI c.CE	1	1	1	3	4	2	0	6	5	1	0	6	15
	Colonna	VIII- X c.CE	2	0	0	2	7	7	1	15	3	5	0	8	25
	Allumiere	XV-XVI c.CE	2	1	0	3	13	3	0	16	11	0	0	11	30
OMAN	RH5	3700-3400 BCE	4	1	0	5	12	11	6	29	11	6	4	21	55
TOTAL			16	9	4	29	87	62	36	185	103	50	23	176	390

c.CE: Centuries Common Era; BCE: Before Common Era

TABLE 2. Morphoscopic variables to determinate pathological flat foot. Features were divided by their location in the foot joints

Calcaneus-Talus	Calcaneus-	Talus-Navicular	Navicular-	Cuboid-	Other
	Cuboid		Cuneiforms	Cuneiform III	Pathologies
Sustentaculum tali	Osteophyte in	Neck/head	Osteophytes in	Osteophytes	Osteochondritis
displaced to plantar-	Calcaneus-	displaced to	navicular-	in Cuboid-	dissecans
medial (PMSTA)	Cuboid joint	medial (DISHN)	cuneiforms joint	Cuneiform III	(OCDRT)
	(CCLOS)		(ONCUN)	joint (CUNOS)	
Osteophyte/alteration		Osteophyte in			Alteration in
in Calcaneus-Talus		talar head			tuberosity of
joint (CTOST)		(OHEAD)			navicular
joine (C. Co.)		(0112/15)			(EXTUB)
Trigonal articulation		Necrosis-like in			(2/1105)
_					
(IIIAIII)					
		(IVECTIO)			
Os trigonum (TRIGM)		Lipping in talar			
		head (LIPHD)			
Wide talar tail		Osteonhytic talar			
(**************************************		lacet (IFO31)			
osteophyte (TAILO)					
		Periosteal			
		reaction of talar			
		neck (OMANK)			
(TRART)  Os trigonum (TRIGM)  Wide talar tail (WITTT)  Talar tail with osteophyte (TAILO)		head (LIPHD)  Osteophytic talar facet (TFOST)  Osteophyte in talar neck (ONECK) Periosteal reaction of talar			

TABLE 3. Chi square test. P values of the variables, contrasted with population and presence of pathological flat foot factors. Significant differences in bold.

		PMSTA	CTOST	TRART	ONECK	NECHD	OHEAD	DISHN	WITTT	TRIGM	TAILO	EXTUB	OCDRT	CCLOS
N		145	151	139	168	165	165	167	157	157	157	98	212	145
POPULATION/ VARIABLES	р	0.142	0.206	0.529	0.115	0.607	0.240	0.662	0.779	0.227	0.092	0.736	0.117	0.261
FLAT FOOT/ VARIABLES	р	0.000	0.000	0.000	0.001	0.020	0.001	0.000	0.005	0.000	0.000	0.000	0.005	0.000

TABLE 4. TEM analysis to check the intra-observer error (ES). Absolute TEM indicates a low error, while the relative TEM shows an acceptable error.

VARIABLES	ABSOLUTE TEM	RELATIVE TEM %	VARIABLE AVERAGE VALUE
BCUB	0.1	1.2	15.9
LCUB	0.0	0.3	24.2
HCUB	0.1	0.7	18.3
LMAXTAL	0.1	0.7	16.7
LSAGTAL	0.1	1.1	13.6
LHEADT	0.2	0.8	25.8
BHEADT	0.3	3.7	8.6
TORSANG	0.7	2.0	38.3
NECKANG	0.7	4.7	16.1

TABLE 5. Descriptive statistical analysis according to the presence pathological flat foot and healthy feet. In addition, Kruskal-Wallis test and Mann-Whitney p values are included. Significant differences in bold.

MEASUREMENTS		PATHO	LOGICAL	FLAT FOOT	Г		WITH	OUT FLA	T FOOT		KRUSKAL-WALLIS VARIABLES/POPULATION	MANN-WHITNEY U VARIABLES/FLAT FOOT
	N	MIN	MAX	MEAN	S.D	N	MIN	MAX	MEAN	S.D	р	р
BCUB	17	21	32	27.4	2.5	89	22	33	26.7	2.3	0.456	0.314
LCUB	17	30	43	36.9	3.3	97	28	43	35.3	3.2	0.032	-
HCUB	17	19	28	25.0	2.5	93	20	28	23.4	1.7	0.506	0.009
LMAXTAL	20	21	35	27.8	3.6	79	23	33	27.3	2.0	0.479	0.647
LSAGTAL	20	18	26	21.6	2.4	84	17	28	21.0	2.1	0.205	0.431
LHEADT	22	25	40	31.7	3.5	134	26	39	31.5	2.6	0.000	-
BHEADT	23	17	27	21.6	2.3	125	16	32	21.2	2.0	0.000	-
TORSANG	19	11	65	35.5	11.3	84	17	47	36.0	4.7	0.589	0.752
NECKANG	21	12	33	18.8	5.8	114	10	21	15.8	2.3	0.134	0.026

LCUB, LHEADT and BHEADT were excluded from the Mann-Whitney U analysis, because were influenced by the population factor.

TABLE 6. ROC curve results. Only useful threshold values to discriminate individuals with/without pes planus.

		F	LAT FOOT		WITHOUT FLAT FOOT						
<b>MEASUREMENTS</b>	AUC	THRESHOLD	SENSITIVITY	1-SPECIFICITY	THRESHOLD	SENSITIVITY	1-SPECIFICITY				
NECKANG	0.652	≥17.5°	0.571	0.254	≤16.5°	0.553	0.333				
HCUB	0.696	≥24.5 mm	0.588	0.258	≤23.5 mm	0.495	0.294				

TABLE 7. Final results of the pes planus classification, through to morphoscopic and metric methods. Number of individuals, divided by populations, condition and sex.

		PA	THOLOGICA	AL FLAT FO	ООТ	NON-PATHOLOGICAL FLAT FOOT				WITHOUT FLAT FOOT				INDETERMINATE FLAT FOOT				TOTAL
COUNTRY	POPULATION	MALE	FEMALE	INDET	TOTAL	MALE	FEMALE	INDET	TOTAL	MALE	FEMALE	INDET	TOTAL	MALE	FEMALE	INDET	TOTAL	N
	Casserres	1	1	0	2	4	0	0	4	25	16	0	41	9	2	0	11	58
	St Pere	3	0	0	3	12	3	0	15	36	26	0	62	10	5	0	15	95
SPAIN	Avinganya	0	0	3	3	0	0	0	0	0	0	40	40	0	0	5	5	48
	Termens	1	0	0	1	1	0	0	1	9	9	0	18	0	1	0	1	21
	Granollers	2	4	0	6	4	1	0	5	4	8	0	12	7	2	0	9	32
	Monterotondo	0	1	0	1	0	0	0	0	1	4	3	8	2	0	0	2	11
ITALY	Villa Gordiani	1	1	1	3	1	0	0	1	7	2	0	9	1	1	0	2	15
	Colonna	2	0	0	2	0	1	0	1	9	11	1	21	1	0	0	1	25
	Allumiere	2	1	0	3	1	0	0	1	17	3	0	20	6	0	0	6	30
OMAN	RH5	4	1	0	5	2	0	0	2	17	15	10	42	4	2	0	6	55
TOTAL		16	9	4	29	25	5	0	30	125	94	54	273	40	13	5	58	390

Figure 1

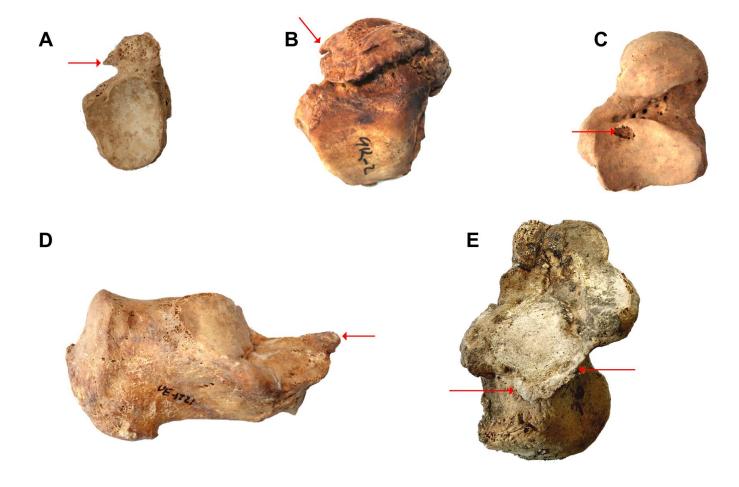
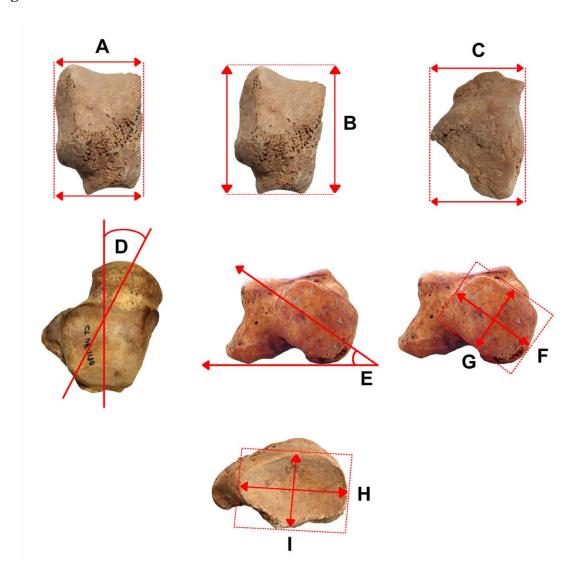


Figure 2



# **Figure Legends**

Fig.1. Some of the morphoscopic variables present in the pathological flat feet. A) Alteration in tuberosity of navicular (EXTUB). B) Osteophyte in talar neck (ONECK). C) *Osteochondritis dissecans* (OCDRT). D) Osteophyte in Calcaneus-Cuboid joint (CCLOS). E) Osteophyte/alteration in Calcaneus-Talus joint (CTOST).

Fig.2. Measurements to determinate flat foot. A) Height of the cuboid (HCUB). B) Length of the cuboid (LCUB). C) Breadth of the cuboid (BCUB). D) Horizontal angle of the neck of the talus (NECKANG). E) Angle of torsion of the talar head (TORSANG). F) Length of the head of the talus (LHEADT). G) Breadth of the head of talus (BHEADT). H) Maximum length of the talar facet (LMAXTAL). I) Sagittal length of the talar facet (LSAGTAL).

## **Supporting Information**

Description of the metric measurements, following the order of the Fig.2.

- A. Height of the cuboid (HCUB): Maximum distance between the dorsal surface of the cuboid to the most inferior point on the beak.
- B. Length of the cuboid (LCUB): Maximum distance between the proximal and distal point of the cuboid (Harris & Case No.5).
- C. Breadth of the cuboid (BCUB): From the superior view of cuboid, with the tuberosity pointing upwards, measure the maximum distance between the lateral and medial sides (Harris & Case No.6).
- D. The horizontal angle of the neck of the talus (NECKANG): Angle between the axis that passes through the middle of the head and the axis of the talus body that crosses in the middle of the trochlea (Day and Wood No.1  $\alpha$ ).
- E. Angle of torsion of the talar head (TORSANG): Angle composed by the horizontal plane on the talus (an imaginary line under the base of the talus), intersecting with an axis that spreads from the superior and inferior points of the talar head, dividing the zone in two similar portions (Gebo and Schwartz No.13).
- F. Length of the head of the talus (LHEADT): Maximum distance between the superior and inferior points of the head of the talus (Martin and Saller No.9).
- G. Breadth of the head of the talus (BHEADT): Maximum distance between the medial and lateral points of the head of the talus, usually located in the middle of the head (Martin & Saller No.10). Just the articular portion was considered.
- H. Maximum length of the talar facet (LMAXTAL): Maximum distance of the talar facet, localized in the navicular (Martin & Saller No.4).
- I. Sagittal length of the talar facet (LSAGTAL): Considering a central axis, it is the perpendicular dimension of the LMAXTAL (Martin & Saller No.3).

2.5. BIPARTITE CUNEIFORMS IN OSTEOLOGICAL AND SCANNER SERIES	
"The great art of life is sensation, to feel that we exist, even in pain".  Lord Byron	
Saldías E, Isidro A, Martínez-Labarga C, Malgosa A. <i>Bipartite cuneiforms in osteological and scanner series</i> . Submitted to: Journal of Foot and Ankle Surgery (March 2019).	
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Bipartite cuneiforms in osteological and scanner series.

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**Abstract** 

The bipartite cuneiform is a rare anatomical variant of cuneiform bones in which two

ossification centers grow separately resulting in two independent bones. In our study, we

analyzed its presence in 445 pairs of feet from 13 archaeological collections from Spain, Italy

and the Sultanate of Oman. Additionally, we observed 32 contemporary individuals from

Universitat Autònoma de Barcelona documented osteological series and 140 CT scans of feet

from the Hospital Universitari Sagrat Cor (Barcelona). The analysis showed one individual

with this anomaly in skeletal collections and four in clinical TCs.

The frequency and different degrees of segmentation of cuneiforms from the modern

osteological series were interpreted through descriptive and biomechanical parameters

during the gait, including associated pathologies observed in our morphological analysis.

According to previous clinical reports, the bipartite medial cuneiform diagnosis is mainly

discovered in high sport stress and incidental findings, which makes it difficult to know the

real frequency in the population. Therefore, in live individuals and depending on its bone

separation degrees and physical activity involved, bipartite cuneiforms could affect the

normal gait, producing an alteration on the joint configuration and structure of the adjacent

foot bones.

Level of clinical evidence: 5

**Keywords:** anatomical variant, congenital anomaly, imaging findings, os cuneiforme I

bipartitum, segmented cuneiform.

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### Introduction

Bipartition of the cuneiform bones is an unusual congenital anomaly that mostly affects the medial cuneiform. There are very scarce reports about affected intermediate cuneiforms and no cases concerning lateral cuneiforms (1). The bipartite medial cuneiform, also called *os cuneiforme I bipartitum* or BMC, is a variant of its structure in which the cuneiform is divided in two parts, corresponding to two centers of ossification.

The BMC was first recorded by Morel in 1957 (2). It is a well described but rare variant (0.33% to 2.4%) (3,4), with predominance in males (84.6%) (5) and a high occurrence of being bilateral (65.2%) (6). The majority of cases were described in anatomical reports before 1930 (7–9), and from clinical cases in recent times (3,4,6,10–13). Gruber in 1877 (7) conducted one of the oldest and largest studies using 2584 feet from anatomical cases dating from 1863 to 1876. He found 15 specimens with BMC from 10 skeletons: 0.27% accounting for full bipartition, and 0.39% accounting for incomplete bipartition. In later studies, Pfitzner in 1896 (5) found a 0.31% of complete BMCs, while Barlow in 1942 (8), made an analysis using a considerable number of feet (750 feet and 1000 respectively), finding a similar percent of incidences.

To date, the Burnett and Case study (2011) (5) is one of the biggest reports of BMC which includes collections from England (381), Denmark (927), Egypt (200), South Africa (1146), Japan (195), the American/Terry Collection (794) and from the Southwest of the US (239). In the whole set, they found 10 cases (two anatomical and five archaeological) of complete bipartition, and three with partial segmentation (two anatomical and one archaeological). Furthermore, they made a meta-analysis using results from previous studies with their own investigation. They estimated 71 complete BMC cases since 1757 until 2011, and 14 incomplete BMC cases, summarizing the cases from 1864 to 2011.

Nowadays, archaeological reports (5,14,15) or fossil cases of BMC from *Homo erectus* (from Dmanisi, Georgia) and *Australopithecus africanus* (Stw573c known as "Littlefoot"), are not

so frequent. This variant was also described in some primates specimens, such as *Pongo pygmaeus abelii* (sic) and *Pan paniscus* (16).

Regarding the clinical reports, some cases have been incidental (9,12,17,18), discovered through RX, CT, or Magnetic Resonance Imaging (MRI) (10,19), and observing a characteristic "E -sign" shape (12). However, the most frequent cases were symptomatic (3,4,6,10,11,19,20). Patients suffered pain that can be related to high physical stress on the zone, to fractures, or some temporary disabilities associated with the synchondrosis that connects both halves of the medial cuneiform.

The BMC can be classified into three categories: Complete bipartition (Type 1-TI), where the dorsal and plantar sections are completely separated; incomplete bipartition (Type 2-T2), where both portions are partially fused, with a notorious proximal-distal cleft (also observable as a bone bridge that joins both segments) on the lateral and medial surfaces; and division of the distal articular surface (Type 3-T3) (13,16).

Almost all information about the bipartition of the cuneiform bones are focused on the medial bone. The only exception is a study made by Brookes-Fazakerley et al (1), who described an extra intermediate cuneiform, located between the second metatarsal base and the second cuneiform. By using radiography and CT scans, the analysis showed a reduction of the size of the second and third metatarsal, and a possible segmentation of one of the involved bones. According to their conclusions, the described anomaly could be an additional bone or a bipartition of the intermediate cuneiform. On the other hand, there are no registered cases about the presence of bipartition of the lateral cuneiform.

To the present day, the causes of the segmentation of the cuneiforms are not clear. The ossification process of the cuneiforms commence between the first months after birth (lateral) to the second/third year (medial, intermediate) (21). The ossification can start as a nodule or as a compound center with double or multiple origins; a common double center of ossification generating dorsal and plantar segments (21). There are two possible explanations proposed for the bipartite anatomical variant related with the embryonic period (5,8,16).

First, a failure in the fusion of two primary ossification centers (dorsal and plantar) resulting in an incomplete coalition in the center of the medial cuneiform. Secondly, this malsegmentation could be due to an abnormal organization of the primordial mesenchyme due to the existence of two different portions of cartilaginous anlage (5), ossifying each center separately. Considering the BMC reports, the result in both cases was a cuneiform divided horizontally, resulting from a dorsal and plantar portion with the largest being the latest in most occasions. Both segments could be joined by an extra-articular joint or "bridge", described as a synovial joint and functionally similar to other tarsal bones, allowing glide movements; or as a cartilaginous synchondrosis (3,16).

In general terms, bipartite cuneiforms have been reported mostly in anatomical and clinical literature. Nonetheless, despite the existence of clinical reports about symptomatology and pain analysis in live patients, there are no detailed reports about the osteological consequences which could be developed in adjacent bones caused by the bipartite cuneiforms. Despite the existing knowledge of anatomical cases, there is not a clear incidence rate in contemporary cases, which might be due to the nature of the anomaly and its symptoms.

Regarding the above, we hypothesized that the frequency of bipartition is underestimated in the diagnosis. Due to the lack of knowledge and cases on this subject, we were interested to analyze a significant number of skeletonized individuals from bioarchaeological, contemporary and clinical series, searching previously described features to discriminate this variant in cuneiforms, focusing in its shape, degree of segmentation and nearby structures that could be affected.

#### Materials and methods

We analyzed the feet of 445 individuals from bioarchaeological contexts, belonging to Esglesies de Sant Pere, Casserres and Castell de Termens, from Catalonia, Spain; Monterotondo CAR, Guidonia, San Ercolano, Crypta Balbi, Villa Gordiani, Castro dei Volsci, Colonna, Leopoli-Cencelle, Aquino and Allumiere from the region of Lazio, Italy; and Ras al-Hamra 5 from the Sultanate of Oman. We also observed pairs of feet from 32 contemporary individuals (dated to 1991) with known sex and age from the Granollers cemetery, located in the province of Barcelona, Spain (22) (ES, AM, CML). The samples are in a good and regular state of preservation.

From clinical context, we analyzed 140 CT scans of adult patients from Hospital Universitari Sagrat Cor, Barcelona (HUSC), from 2010 to 2018. To consider the diagnosis of bipartite cuneiform by CT scans, we have taken into consideration three radiological facts: firstly, the presence of an intra-bone image in the sagittal view of the cuneiform, that could be consider as an anlage and different to a vessel image. In second place, the presence of, at least, one cartilage alteration either on the proximal or in the distal joint surface that could be related with the inner anomaly. Finally, a negative diagnosis of non-displaced bone fracture nor an osteochondritis of the damaged joint surface (Table).

In all of the positive CT scan diagnoses of bipartite cuneiform, the scintigraphy examine has been negative in this area. (*vs* 2BMC +). In the case of the bipartite lateral cuneiform, the MRI does not shows any kind of edema. The bone anatomical situation has been checked by a parallel coronal image of the CT scan in order to avoid mistakes in the levels between the three cuneiform bones.

None of the mentioned individuals were diagnosed or treated for compatible features with the bipartite cuneiform. Due to this reason, the only images available were those of the affected foot, making it difficult to know if the bipartition is bilateral or not. The scans were analyzed by three observers in different periods of time (ES, AI, AG). In addition, archaeological samples were analyzed by visual direct inspection.

### **Results**

The osteological analysis detected the presence of clear BMC characteristics in only one individual from the Granollers series (GR5), out of 477 cases in total. Moreover, from a clinical context and through the observation of CT scans, we found four out of 140 individuals with compatible signs of bipartition. They were found in the medial cuneiform (2), intermediate cuneiform (1) and lateral cuneiform (1).

The Granollers collection is a set of 32 individual contemporary human skeletons exhumed in 1991 belonging to the Granollers Cemetery in the province of Barcelona, Spain and is currently housed at the Physical Anthropology Laboratory of the Universitat Autònoma de Barcelona (UAB). The age of death for these skeletons extends from 32 to 91 years and of the 32 individuals, only one developed a malformation in the form of fusion from the two segments of the medial cuneiform.

This individual, GR5, was an adult female of 66 years who died in 1987. The individual presents antemortem fractures in a series of ribs on both sides and degenerative changes related with age. The feet show structural changes in the adjacent bones of the medial cuneiform due to its segmentation, as well as other unrelated diseases such as *osteochondritis dissecans*.

In the right foot, the medial cuneiform was partially divided into two portions (dorsal and plantar), observed by the presence of two distal facets and the complete fusion of the proximal and lateral surfaces (Fig.1). The medial portion of this cuneiform is also divided into two parts united by two bridges (Fig.1); the distal surface being larger than the proximal surface and the cleft or separation line finishing at the surface for the first metatarsal (MT1). The navicular bone shows slight eburnation in half of the surface that articulates with the medial cuneiform and also extends to the surface that articulates with the intermediate cuneiform. Macroporosity in the tuberosity and *osteochondritis dissecans* of the talar facet can also be observed (Fig.1). The first right metatarsal (MT1) shows a strongly divided

surface of its base (Fig.1), while mild signs of arthrosis were also observed in the head of this particular MT1.

The medial cuneiform of the right foot is entirely bipartite with the plantar portion being slightly larger than the dorsal one (Fig.2). The tissue observed in the divided portion is flat, and very congruent with the articular surface while some signs of pitting on the segmented surfaces were also found (Fig.2). The left foot shows a navicular with eburnation and a depression on the surface that articulates with the medial cuneiform.

The first metatarsal developed a marked and divided surface to accommodate the two portions of the medial cuneiform; the dorsal portion being larger at 13 mm and having a rounded and compact form while the plantar portion is smaller at 11 mm and extending in a diamond like shape (Fig.2). Additionally, signs of arthrosis such as slight eburnation and small osteophytes can be observed around its head.

The CT scans of the Hospital Universitari Sagrat Cor displayed four individuals that developed the anomaly in different degrees and in different cuneiforms, with prevalence in the medial (two cases *vs.* one in the intermediate and one in the lateral cuneiform). According to these findings, every one of the cases are female. None of them belong to *T1* type.

The sagittal view of the CT scans provides the best images to analyze the presence or absence of bipartition in the zone. Bipartite cuneiforms were found in four cases, though no cases of complete bipartition were observed. The diagnosed cases showed a line of separation not arriving completely on both extremes of the cortical bone (Fig.3). We did not observe the presence of degenerative diseases, osteophytes or other anomalies between the contact surfaces of the cuneiforms, metatarsals and naviculars. The only exception was the case of a female patient, who displayed a possible sign of arthrosis in the base of the first metatarsal.

### **Discussion**

There is not a clear frequency of bipartite cuneiforms in the general population. The majority of incidences affect the medial cuneiform, while reports of the intermediate and lateral cuneiforms are very scarce. The general incidence of BMC is 1 in 320 (17) and the frequency is considerably less in females than in males in the population (15.4% v 84.6%) (5,12). Considering the type of bipartition, cases of partial BMC (T2 and T3) are more common in males than in females (70% vs. 30%) and the same is true for complete bipartition cases (T1) (84.1% vs. 15.9%) (5). In relation with laterality, the cases where both sides were affected are more common (81.8%) (5). In separating them by type; partial BMC is bilateral in 66.7% of cases and unilateral in 33.3% of cases while the complete bipartition of the medial cuneiform (T1) is bilateral in 86% of cases and is unilateral in 14% of cases.

The incidence of BMC came from osteological/archaeological studies, incidental asymptomatic clinical cases, or derived from pain in the foot as we previously mentioned. Nonetheless, we do not know the real frequency of BMC considering the size of collections and the origin of the current series. The latter mainly come from a hospital where a high number of individuals that were diagnosed with this condition practiced high-level sports or partook in constant physic activity such as runners or soldiers (3,6,9–13,19). Therefore, asymptomatic cases were almost not considered. Of the clinical cases, 66.6% are related with sport episodes, while 33.3% are related with incidental cases or with patients who suffered chronic pain not associated with a sport event. In the majority of RX services, BMC is not a routine reportable trait.

Regarding our samples, the BMC Granollers case is of a woman (GR5) with a partial fusion in the right foot and complete fusion in the left. The features shown by individual GR5 are quite similar to the previous descriptions of archaeological cases, observing cortical tissue in the affected zone and size differences between the dorsal and plantar portions (5,15,16).

The right foot corresponds to an incomplete bipartition (T2) that is more visible in the medial surface where a cortical bridge joins both portions of the bone. In cases like this, we suppose

that they could be asymptomatic. The joint is not weak, suggesting stability of the medial cuneiform and resistance to movements and impacts. Meanwhile, the navicular of individual GR5 shows eburnation, suggesting some degree of pain in this zone.

On the other hand, a complete bipartition (T1) was found in the left foot. This kind of segmentation is the most common in all cases (6). While the overall shape is conserved, the size of the two combined bones is larger than that of a normal medial cuneiform and has been observed in other similar cases (14,16,20). The osteological damage of the GR5 case prevents analysis to infer if it was symptomatic, and if the pain could mainly be a consequence of the damage in the adjacent bones, i.e. eburnation and osteophytes. However, from clinical literature we know that episodes of pain and discomfort due to the presence of BMC are related to stress activity (3,10,12) or derived from sport injuries (6,19). Depending of the degree of activity and damage of the patients, BMC could be very painful and in some cases surgical procedures was necessary (3,10,13). Similarly, clinical symptoms were also reported in the bipartition of the intermediate cuneiform (11).

To understand walking alterations of individual GR5 from Granollers, it is very important to contextualize the damage or changes observed in the bones that articulate with the medial cuneiform. We must note the division on the MT1 surface and the eburnation on the surface of the navicular. This condition could cause pain and inconveniences in the gait, mainly in the movements where the MT1 and hallux are involved. Even though individual GR5 developed other degenerative features in the column and pelvis, the eburnation on the left foot is a consequence of the form and position of the medial cuneiform in articulation with the navicular and is more severe than that seen in the right foot. Other authors observed bone damage produced by the segmentation of the cuneiform, as seen in a case of a 36 year old male patient with cystic changes in the bipartition, producing degenerative arthritis and abnormal motion (12).

The reason of degenerative changes in these cases can be found in the biomechanical function of cuneiforms. The medial cuneiform is part of the navicular-cuneiform joint, which is related with the modification of the form and curve of the plantar vault, making the foot adaptable

to different kinds of ground. This joint is also related with the orientation of the feet with the longitudinal and vertical axes (23). An affection of the medial cuneiform and MT1 zone could damage the Lisfranc joint or the lateral long peroneus tendon. Moreover, it might alter the transmission of forces to the metatarsals, generating problems with the adaptability of the foot with the ground, in the stability of the arch, in the eversion of foot, and in the plantarflexion of the ankle (24).

Concerning current population, the HUSC does not register all cases of BMC as a standard rule, but only those cases related with a specific pain in the region. Therefore, we revised 140 CT foot scans searching for this specific anomaly. We did not find a complete bipartition in any sample, but some incomplete BMC (T2 and T3) were detected. These cases exhibit an incomplete segmentation line, which does not go through both extremes of the cortical bone.

We found four BMC samples, in a total of 617 feet (2 osteological and 2 CT scans). As we expected, this frequency is higher than the incidence ratio mentioned in studies (1 in 320) and it could be due to different factors such as genetic origin. Cvrček et al (25), identified family relationships based on skeletons (44 adult individuals) of 5 different branches of one family from the country of Bohemia, (between the 19th–20th centuries). They reported a division between the medial cuneiform and first metatarsal articular surfaces (probably a partial bipartition) in some members of the family. The above suggests that there is probably a genetic factor which could be inherited.

Another hypothesis to explain this high frequency could be related to the underestimation of the BMC incidence until now. Considering that many of the bipartitions are asymptomatic and therefore only have been discovered by incidental detection, the lack a systematic study could reduce the number of BMC detected. In fact, BMC and other cuneiform segmentations are rare variants and many professionals might focus on other causes for pain, obviating a bipartition diagnosis.

The low findings on intermediate and lateral cuneiforms could have the same reason. The only case of a possible bipartition of the intermediate cuneiform (1), reported pain in the

segmentation zone. Nonetheless, not one of the CT cases studied in our investigation reported pain in the cuneiforms area, which suggest that they were asymptomatic.

In light of the many incidental cases and the few clinical occurrences of chronic pain not deriving from contact sport events, we proposed that bipartite cuneiforms are generally asymptomatic cases, according to Dellacorte et al. (17) and O'Neil et al. (18). Therefore, the pain in Bipartite Medial Cuneiforms could be related mainly to physical stress, strong contact in the zone, and prolonged activity where the foot is involved.

In summary, we analyzed 15 osteological collections, finding one contemporary Bipartite Medial Cuneiform case (Granollers) and four contemporary individuals from Barcelona through CT scan images (two BMC, one intermediate and one lateral cuneiform). The contemporary individual developed a bilateral condition, exhibiting partial and complete bipartition.

According to previous clinical cases, BMC has mostly been incidental with some cases associated to high physical stress and arthrosis. According to our criteria, every one of the CT scan findings corresponded with incomplete bipartitions, which would be asymptomatic because not one of the patients were treated in the zone for this affection.

Complete bipartition could affect consecutive bones, generating a degenerative and progressive decrease of the bone state that might produce pain, as observed in the individual from Granollers. Although the detected frequency of BMC in osteological series is similar with previous reports, the CT scan findings are much higher than the average. This frequency could be explained by different factors related with the origin of the samples, their genetics, or an underestimation of clinical frequency.

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## 9. Tables

 $TABLE.\ Number\ of\ individuals\ used\ for\ BMC\ analysis\ from\ the\ skeletal\ collections.$ 

COUNTRY	SERIES	DATATION	REFERENCES	N
	Sant Pere	IV-XIV CE	Jordana et al.(26)	95
Spain	Casserres	VII-IX CE	Cascante and Farguell (27)	58
	Termens	XVIII-XIX CE	Alesan et al.(28)	21
	Monterotondo CAR	II-III CE	Rubini and Mogliazza (29)	11
	Guidonia	II-III CE	Baldoni, Ferrito and Martinez-	14
			Labarga (30)	
	San Ercolano	IV-V CE	Rothchild et al. (31)	15
	Crypta Balbi	IV-V CE	Sagui (32)	8
Italy	Villa Gordiani	V-VI CE	Fornaciari et al. (33)	15
	Castro dei Volsci	VI CE	Rubini (34)	35
	Colonna	VIII-X CE	Baldoni et al.(35)	25
	Leopoli-Cencelle	IX-XV CE	Baldoni (2019)	61
	Aquino	XI CE		2
	Allumiere	XV-XVII CE	Baldoni et al.(37)	30
Sultanate of	Ras al-Hamra 5 (RH5)	3700-3400 BCE	Coppa (2012)	55
Oman				
Spain	Granollers	Contemporary	Saldías et al. (2016)	32
Spain	HUSC	Contemporary		140

# 10. Figures

Figure 1

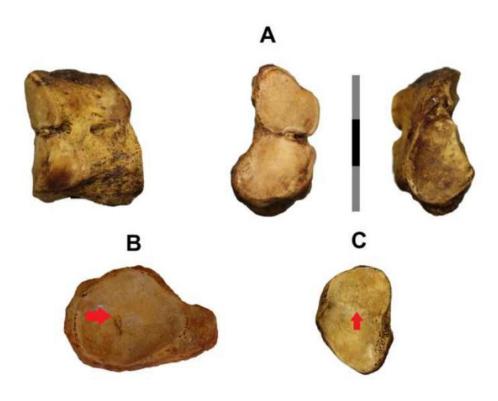


Figure 2

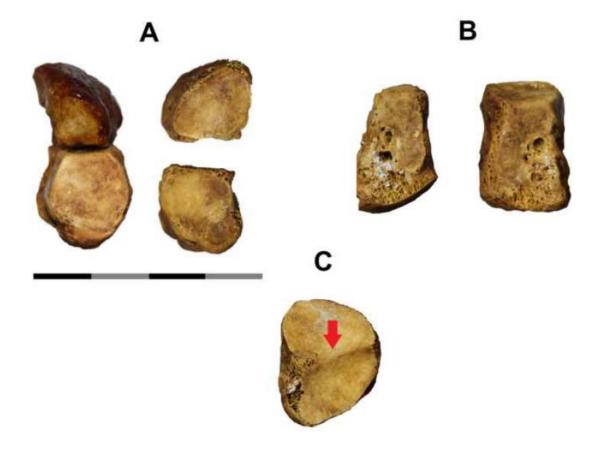
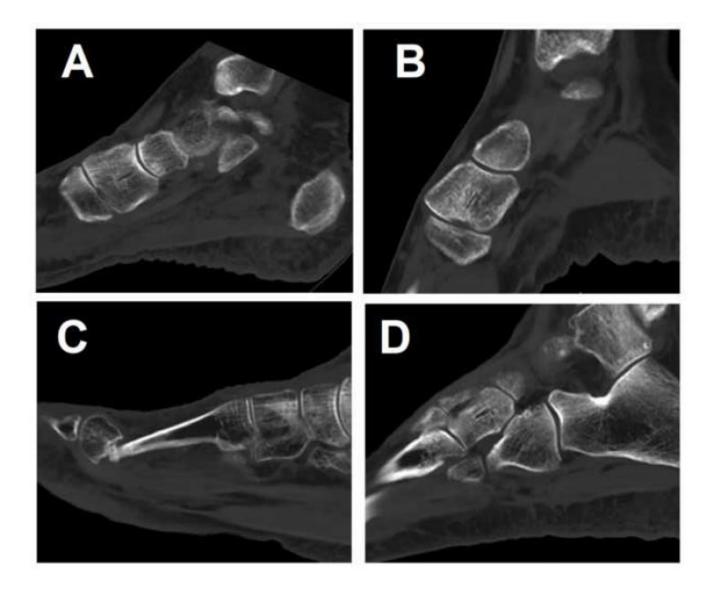


Figure 3

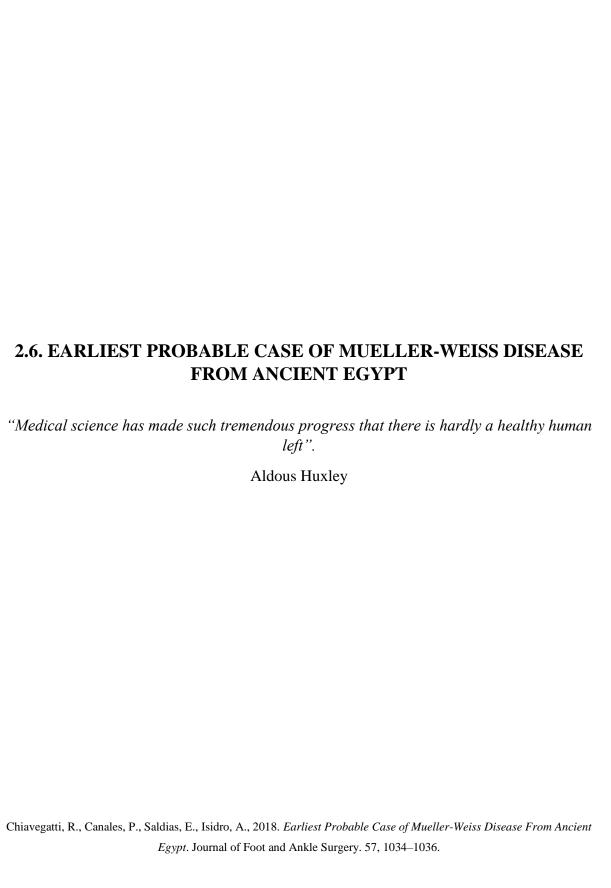


### Figure legends

**Fig. 1.** A) Medial, distal and proximal view of right medial cuneiform (GR5). B) Talar facet of navicular, affected by osteochondritis dissecans (arrow). C) Base of MT1. Red arrow indicates the separation related with the segmented facets of medial cuneiform.

**Fig. 2.** A) Complete bipartited left medial cuneiform (GR5). Proximal (articulated) and distal view (separated). B) Non merged portions of medial cuneiform. C) Base of MT1. The red arrow indicates the separation due to BMC.

**Fig. 3.** Composite image. A) Bipartite Medial Cuneiform: In the center of the bone is present an anlage that corresponds with the distal joint prominence. B) Bipartite Medial Cuneiform: Near to the center of the foot, is visible a double image that correspond to a T3 bipartition of the cuneiform bone. This condition is supported by the presence of a distal joint prominence. C) Bipartite Intermediate Cuneiform: The image shows an anlage in the intermediate cuneiform. The neck of the second metatarsal bone is affected by a lack of fusion, with a dorsal displacement, after a percutaneous surgical procedure. D) Bipartite Lateral Cuneiform: A clear anlage of the lateral cuneiform bone was observed. In this case, the joint surfaces do not present any alteration.

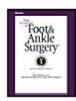




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## Earliest Probable Case of Mueller-Weiss Disease From Ancient Egypt



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#### ABSTRACT

Mueller-Weiss disease is an alteration of the tarsal navicular that is primarily due to anomalous ossification of the bone and lateral deviation of the talar head associated with screw-like movement through the axis of the subtalar joint. This syndrome tends to be associated with various degrees of flatfoot and hindfoot valgus combined with subtalar joint varus. Ancient cases of Mueller-Weiss disease have not been described in specialized literature. We present the case of an adult male from the Hellenistic period (Ptolemaic dynasty; fourth to first century BC), the skeleton for which was found inside a sarcophagus in the archaeological site of Sharuna (middle Egypt) with Mueller-Weiss disease. The specimen is, in all likelihood, the earliest case of this type of foot pathology in the archaeological record.

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Tarsal scaphoiditis, or Mueller-Weiss disease (MWD), is a deforming disease of the navicular bone in adults. It tends to be associated with severe flatfoot, with the navicular displaying medial deviation. Clinically, MWD can cause severe pain in the midtarsal area of the foot and is a cause of mechanical pain with both walking and standing. MWD is more common in females (73% of cases) than in males (1), Our understanding of the archeological record indicates that ancient cases of navicular pathologies are very unusual. In the present report, we describe what we believe to be the earliest case of MWD in an ancient male skeletal specimen. Because MWD is often associated with clinical symptoms in living humans, we thought that foot and ankle surgeons would be interested to know that archeological evidence exists of the disease in the skeletal remains of an individual who lived –2300 years ago, during the Hellenistic period of ancient Egypt.

#### Case Report

Since 2006, the Spanish-German mission from the Museu Egipci de Barcelona (Spain) and Eberhard Karls Universitat (Tübingen, Germany), has been working in the necropolis of Sharuna, Middle Egypt, 200 km south of El Cairo, on the east bank of the Nile river. This vast

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Conflict of Interest: None.

Address correspondence to: Albert Isidro, PhD, MD, Department of Orthopedic Surgery, Hospital Universitari Sagrat Cot, Viladomat 288, Barcelona 08029, Spain. E-mail address: aisidro.cot@gmail.com (A. Isidro). necropolis spanned from the third dynasty of the Old Kingdom (2650 to 2575 BC) to the beginnings of the Coptic period (fourth to seventh century AD). To date, the skeletal remains of 438 individuals have been identified, and their pathologic features studied (2). Most (84,37%) of these individuals were skeletons found with poor anatomic positioning, although the bones were well preserved, primarily owing to a long history of plundering in search for jewelry and other goods. Nearly 16% of the remnants were found partially or totally mummified,

In the 2008 archaeological season, a different chamber buried in the bottom of a shaft from the Ptolemaic dynasty was identified. A total of 12 sarcophagi from this chamber were studied, 1 of which (U.E. 2569, sarcophagus number 7) belonged to an adult male, as determined using anthropologic methods such as the right angle of the jaw, pronounced supraorbital torus, closed sciatic notch, narrow pelvic ring, and robustness index of the long bones. The individual showed some enthesopathies in the upper extremities (deltoid and large pectoral muscle adhesions in both humeri, a bicipital tuberosity in the proximal third of the radius), which might be an indication of hard work with the arms. He also had moderate to severe dental attrition because, in ancient Egypt, bread was mixed with large amounts of sand particles, which would explain the poor oral health in almost all individuals. He was situated in the decubitus supine anatomic position with his left femur migrated through his thorax (Fig. 1). The femur was 435 mm long; thus, his estimated height was 164,8 cm. Moreover, from these findings, our best estimate of his age at the time of his death was 21 to 30 years, owing to the total absence of growth lines,

The sarcophagus housing U.E. 2569, sarcophagus number 7, was anthropoid shaped, complete, and constructed of white limestone. This



Fig. 1. Photograph of individual UE.2569/S7 from the necropolis of Sharuna (Middle Egypt) dated to the Ptolemaic period. Arrows indicate the prominence on the talar neck.

type of sarcophagus dated from the end of the Late period and the beginning of the Hellenistic period (380 to 150 BC). The only vestiges of decoration appeared in the cover stone, The sarcophagus head was in high relief that extended to the thorax, which it could represent the remains of a portrait of the deceased. With rough lines, a figure of the individual was displayed, depicting a false beard and the nemes crown, similar to that seen in association with royal individuals. Under this, the cover stone was smooth, and no other signs of decoration appeared. In the late Ptolemaic period, an archaizing tendency was present, inspired by the New Kingdom, such as in the Pyramid Texts; thus, the first sarcophagi from the Ptolemaic period present with this archaic shape from the Late period (3).

The inner part of the sarcophagus was long and narrow, too small and insufficient to harbor a wood receptacle about the remains of the human. Inside the sarcophagus, neither cartonnage nor grave goods were present, which might have been related to the traces of spoliation, although this had not altered the skeleton, with the exception of the misaligned left femur as described. Furthermore, the decline of the practice of mummification was obvious at the end of the Ptolemaic period. Also, during the second century BC, nostril extraction of the brain was lost, Hence, in our specimen, the brain had been left inside the skull, tampons had been inserted in the nose, and the viscera were harbored inside nearby canopic jars.

Close inspection of the remains of the individual showed no gross pathologic features, except for signs of avascular necrosis in the right humeral head and both feet, abnormalities that were evident only after opening the sarcophagus. The talar neck, bilaterally, showed a prominent dorsal osteophyte and, after cleaning the remaining bones of each foot, which were covered with debris. Each tarsal navicular displayed abnormal morphology that included flattening of the external

par and a distinct osteophytosis (Fig. 1). Both tarsal naviculars showed distinct thinning owing to lateral compression, and each talonavicular joint was degenerated. These macroscopic, bilateral, symmetric abnormalities of the tarsal navicular bones strongly suggested that these were the remains of an individual with stage 1 to 2 MWD (Fig. 2). Moreover, the evidence of right humeral head degenerative changes suggestive of avascular necrosis, along with the abnormalities of each tarsal navicular and talonavicular joint, suggested that this young individual might have experienced the clinical symptoms associated with the deformity and pain related to these osteochondroses.

#### Discussion

Referring to the condition as "double navicular" in 1927, Mueller (4), a surgeon from Leipzig, Germany, described the case of a patient with a severe deformity of the tarsal navicular in both feet. In that same year, Weiss (5), an Austrian radiologist, described 2 similar clinical cases. Since then, this pedal malady has been known as MWD, It was Schmidt (6), in 1925, who described this same foot alteration in a patient with pluriglandular insufficiency, an endocrine disorder. In 1939, Brailsford (7), after studying 9 cases, was the first to describe MWD as an osteochondritis. In 1948, Fontaine et al (8) first referred to MWD as "tarsal scaphoiditis of the adult." It is also interesting to consider that, although they can be similar in morphology, no direct relation has been found between Koehler disease in children and the development of MWD in adults (9).

From the beginning, the precise etiology of MWD has been an issue of controversy, and the first investigators to describe the condition suggested 2 different etiologies, Although Mueller (4) thought that it was a degenerative sequela of childhood disease, Weiss (a disciple of Kiemböck) (5) was a proponent of osteonecrosis, Subsequent investigators suggested the following etiologies to explain MWD: post-traumatic degeneration (10), congenital bipartite navicular (11), Koehler-like avascular necrosis (9), and osteochondrosis (12).

Currently, delayed navicular ossification and lateral navicular compression for lateral weightbearing transfer are thought to be the key elements that result in MWD, Hindfoot varus coexists in cases of MWD, such that the lateral portion of the head of the talus redistributes the compressive stress to the lateral part of navicular and the cuneiforms. In addition, a short first metatarsal contributes to this abnormal pattern of force distribution (1). In some anthropologic studies, MWD has been



Fig. 2. Photograph of the talonavicular joint. The talar head presents with a dorsal osteophyte, and the navicular shows alterations compatible with Mueller-Weiss disease.

associated temporally with episodes of nutritional stress, with evidence of Harris lines, dental enamel hypoplasia, long bone growth arrest lines, and cribra orbitalia described in association with individuals with MWD (13,14). However, the conditions of our archaeological fieldwork did not allow us to reinforce our gross findings with radiographic inspection of the skeletal remains, which could have potentially provided further information on our findings in terms of sclerosis of the cortical bone and/or the presence of intra-bone cysts.

In conclusion, few examples of tarsal navicular pathology are available in the archaeological record, and reports of midtarsal bone pathologies are very scarce in the specialized literature, although references to the calcaneus and talus and to forefoot bony abnormalities are more common. Furthermore, references to medial cuneiform osteochondritis (15), cuboid osteochondritis (16), Early Medieval Period bipartition of the medial cuneiform (17,18), 2 cases of prehistoric calcaneonavicular nonosseous coalition (19), and 2 cases of navicular osteochondritis from ancient Egypt (20) are noteworthy. To date, however, and to the best of our efforts to search the reported data, no cases of MWD have been reported in the paleopathological record.

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## 3. DISCUSSION

"The important thing in science is not so much to obtain new facts as to discover new ways of thinking about them".

William L. Bragg

### 3.1. General considerations

In this project, we have studied the variability of some bone elements and the skeletal structure of foot from different points of view. Feet vary in size depending on age, sex or population. Furthermore, the variation affects morphology between normal and pathological conditions.

To analyze this variability, we compiled morphological and metric data from different archaeological and current skeletal collections. Most of the osteological measurements were proposed by Martin & Saller (1957) [1], and has been used in the thesis for different research. They were applied to check the sexual dimorphism in navicular bone, to check their relationship with the cuboid facet of the navicular (CFNB), and to identify non-pathological pes planus. The original project included a wider list of measurements in each research, but some were discarded for their low reliability or lack of interest.

Concerning morphological methods, the original list of features used in our research, included a big number of osteological signs mentioned in the literature. Nonetheless, there is a small number of them that were discarded, because they were not present in the samples. Mostly, they correspond to additional bones and diseases that imply severe bone deformations. Furthermore, other variables were added in the observation process.

In general, the collections were chosen for their particular features and their optimal level of preservation. Due to legal aspects that vary among countries, the access to contemporary series was not simple. Thus, we count with a major variety and number of samples from archaeological collections, in contrast with the modern, only obtained from Spanish institutions.

The chronology of the archaeological series allows us to compare the cultural changes in concrete geographical zones (Catalonia in Spain, and Lazio in Italy). In order to categorize the collections under the importance of their historical periods, we decided to take the X century CE as the period to divide the total series into two groups (See Results, "Pathological and normal variability of the foot bones in osteological collections"). This century might be considered as transitional due to important cultural changes, especially in the Arabic world and its influence in current territories from Spain (Al-Andalus). In addition, during this

century the end of the *Early Middle Age* (V CE-IX/X CE) occurs, a period when two big empires emerged under the influence of religion: Christianity (East Romans) and Islamic (Caliphate). Afterwards, the *High* and *Late Middle Ages* (X-XV CE) started, a period that began with an economic, cultural and demographic renaissance. Here, it occurred the transition from feudalism to capitalism and the discovery of America, among other significant events [2–4].

Hereunder, we present the discussion of the thesis, divided by each of the articles that compose it, and general considerations on aspects that involve the project and its aims.

### 3.2. The study of the foot

The feet have received little attention from anthropologists. Certainly, evolutionary scholars have profusely studied it, but the variability of *Homo sapiens* foot has been looked over just a few. The main studies are about specific pathologies from particular individuals, but population studies are not frequent. Therefore, despite the fact that a normal foot is essential to the daily life of the humankind, anthropological studies have focused on other zones, for instance, the skull [5].

Thus, we approached this topic according to the features of the collections and scarcity of literature on these topics: sexual dimorphism and the biomechanical implications of the cuboid facet of the navicular, pathological analysis, determination of pes planus, bipartition of cuneiform bones and the oldest registered case of Mueller-Weiss disease.

### 3.2.1. Normal variability

Every individual is different from any other person in many biological aspects, including the skeleton. It is possible to use the skeletal elements to distinguish some individual characteristics and even to identify a person in specific cases. This variability has different origins and one of its most important sources is sex. For this reason, we began to study the sexual dimorphism in relation to the foot. Specifically, we focused on the study of the navicular, a foot bone that a few researchers studied from this point of view.

Like sexual dimorphism, the growth and development of foot bones also produce important changes in the structure, which will variate in a particular way in every bone and zone. According to Scheuer & Black (2004) [6], mature foot length is achieved at around 14 years in females and 16 in males, which coincides with the stop of the epiphyseal growth. Foot bone development begins around day 37 in the intrauterine life, when it is possible to identify the footplate in the lower limb bud, and during the end of the week 7, tarsal and metatarsal bones start the chondrogenesis, with single nucleus or separate centers. At day 57, the embryonic life of the foot finishes, and chondrogenesis starts in all the tarsal/metatarsal/phalanges and other structures. Anomalies as oligo/polydactyly, supernumerary bones, tarsal coalition and clubfoot are relating to the first days of life. In the fetal phase, there are 46 centers of ossification (26 primary and 20 secondary), arising some primary centers during this stage and others after birth. The first group of ossification centers includes the metatarsals, distal, proximal and middle phalanges, while the second group imply the calcaneus, talus and cuboid. After birth, ossification continues with lateral, medial and intermediate cuneiform [6,7]. During this period (2-3 years old of life), bipartition of the medial cuneiform can occurs as we explained in "Bipartite cuneiforms in osteological and scanner series".

Despite being considered one of the biggest foot bones, the formation of navicular is completely particular. Chondrification begins at week 8 (two different nuclei) being the last tarsal bone to start this process and its ossification is the last to start, and one of the last to merge completely, at around 9-13 years old of life [6,7].

On the other hand, variability can be found in every morphological detail of the bones. Therefore, the morphological spectrum to analyze is enormous. We chose a little facet of the navicular bone, the cuboid facet, to understand its variability, because very few works have been done on it and their results do not offer an unambiguous explanation.

Finally, the congenital variability also is considered normal variability. As an example of this category, we examine the question of bipartite cuneiforms. Usually, bipartite cuneiform is interpreted as a variability of cuneiform, asymptomatic. Nevertheless, there are some cases with a total division of the bone, which could lead to some problem of discomfort or pain.

Precisely, we studied a similar case, the reason why we located this article inside the pathological conditions.

Next, our researches about normal variability were discussed, including important aspects that were not addressed at the time of publication.

"Sex estimation from the navicular bone in Spanish contemporary skeletal collections"

The first research approached sexual dimorphism through osteological dimensions of the navicular bone. As compared to the other researches included in this project, this study analyzed only contemporary series from Spanish collections (Granada, Madrid and Barcelona). We concluded that our equations might be applied to Spain or similar populations [8,9], but until our date, there are no additional studies with the same focus.

During 2018, we published "Estimación del sexo a través del navicular en poblaciones españolas contemporáneas" [10] (a non-indexed research, not included in this thesis), an adaptation of our research to the Chilean context. Here, we consider the latest genetic analyses from Chile to check the components of the current population and the possibility to apply our previous metric results [11] to the Chilean population. As molecular analyses shows, there is a significant European genetic baggage. Homburger et al [12] who made a genetic study in South American countries (Chile, Argentina, Colombia, Ecuador and Peru), determined that Chilean population has a 57.2% of European origin (mostly Iberian peninsula and the Mediterranean), 38.7% Amerindian, 0.02% from West Africa and 0.01% from East Asia. Just as above, Fuentes et al [13] analyzed 913 Chileans, finding a 51.85%  $\pm$  5.44% of European component (mostly in central Chile), a  $44.34\% \pm 3.9\%$  Amerindian (high in the north of Chile and Araucanía and Aysén regions) and 3.81%  $\pm$  0.45 African (mostly in the north of Chile). Similar results got Eyheramendy et al [14] (52.25% with European origin, 44.74% Amerindian and 3.01% African) who analyzed 313 individuals from continental Chile. Thus, these studies demonstrated that more than 50% of the genetic components of Chileans came from Europe, mainly from the Iberian Peninsula. The above is related with historical documents that mentioned migratory movements during the conquest and reconquest of Chile (Basques, Andalusians and Castilians among others) and the last century, mostly during the civil Spanish war [15,16].

Due to the scarce investigations with a significant number of skeletonized individuals in contemporary Chilean collections and the high genetic similarity with Spain, we proposed to contrast our method on naviculars from Chile to check the correct classification. Although we did not test our method in Chilean series, we hypothesized that results must be similar by the affinity with the genetic charge from Spain in Chile. The importance of proposing populational studies to determine sex through bone dimensions is to increase the correct accuracy in specific series.

"Morphological and biomechanical implications of cuboid facet of the navicular bone in the gait"

In this chapter, we studied the frequencies and probable causes of the normal variability of the cuboid facet of the navicular bone (CFNB) in contemporary series from Spain. Morphologically, we considered the CFNB as a flat anterolateral surface next to the third cuneiform facet of the navicular, which can vary in form, extension or breadth.

In the cuboid bone, the articular contact with the navicular bone (CFNB) can be seen by the presence of the "navicular facet", which is located below the surface for the third cuneiform [17]. These surfaces could change, especially in the size, depending on the contact with the third cuneiform and navicular. However, unlike the cuboid facet, the limits of the navicular facet of the cuboid bone are not well defined and are irregular, and sometimes look like they merge with the surface of the third cuneiform, which makes it difficult for the observer to determine its presence.

Conversely, the cuboid facet showed different shapes and always displayed a well-defined limit. CFNB could develop a wedge-shaped, almost square form, as an extension of the ectocuneiform, but in some few cases, they were separated from cuneiform facets. The sizes were different between individuals, with the cuboid facet being short or long. However, it is difficult to make a detailed classification of every sample, due to wide variability of the cuboid facets.

In the Spanish series, the CFNB shape was almost square as an extension of the ectocuneiform. However, there are different forms. Already in 1907, Manners-Smith [18] studying an Egyptian collection, made the first classification considering the shape of the

CFNB. He described the most common cuboid facet form as "four-sided, with the posterior angles rounded off". He also classified other shapes including almost square, rhomboidal, semilunar, oblong and wedge. Generally, we coincided with the description made by Manners-Smith about the varied form of the cuboid facet. Nonetheless, we could not classify this surface into precise categories, because there were a great number of samples, whose cuboid facets showed a mix of features. Taking into account that the cuboid facet could be produced by some degree of contact between the navicular and cuboid bones in the foot movements, shape and size of this surface can suggest the degree of contact of these bones. If so, this could explain its presence in primates and old ancestors in contrast with modern humans due to their differences in foot specializations.

According to the results of our study on CFNB, the sagittal length of the talar facet (LSAGTAL) was the only variable that demonstrated a relationship with the presence of CFNB. Likewise, in "Sex estimation from the navicular bone in Spanish contemporary skeletal collections" the same variable showed one of the highest AUC (as a single variable or into an equation) and one of the best performances to discriminate between sexes, mostly to classify female bones. Talar facet of the navicular is a concave surface, which articulates with the talar head of the astragalus, and together form the talo-navicular joint. Despite this articulation has a closed packed position to supination [19], the movements of this joint (where could be involved the LSAGTAL) could contact to a rigid cuboid, producing the CFNB. As we previously explained, these measures would be smaller in the female group. Furthermore, other hypotheses about the presence of CFNB might be related with a remnant characteristic of human evolution, because it is present in some ancestors and primates, or due to congenital variability in the configuration of midtarsal joints.

On the other hand, CFNB was part of the variables that previously were discarded in "A new methodology to estimate flat foot in skeletal remains. The example of Mediterranean collections" because they did not show a significant relationship with the presence of pathological flat foot. The above could be interpreted as the pathological pes planus produces injuries in the muscular and skeletal systems, forcing to maintain obligate pronation by the dysfunction of the longitudinal arch. Thus, mild but constant and incomplete supination

movements would be more difficult to perform, avoiding the contact between navicular with the cuboid.

### 3.2.2. Pathological variability

Many factors could produce different osteological features in individuals and populations. As we mentioned before, biological processes such as growth and development, sexual dimorphism, among others, could be responsible for this diversity. Nonetheless, osteological modifications do not correspond to normal variability only.

Furthermore, there are osteological features related to diseases, old age and traumatic events that also are expressed in the bones. These ailments that people suffer during the life, have been widely studied by paleopathologists in some bones, but we noticed a lack of information on the feet bones. Despite the fact that we use feet constantly, they could undergo modifications, limiting our movements in the walk, or during the repose.

Hereunder, we discuss every research that involved pathological variations, adding new information and interpreting these results under biomechanical parameters, among others.

"Pathological and normal variability of the foot bones in osteological collections"

This research analyzed degenerative diseases, enthesopathy and human variability of feet in archaeological series from Spain, Italy and Oman, and a contemporary collection from Barcelona, Spain. The results indicated significant differences in some variables among populations: Calcaneal spur (SPURC), Hypertrophic Peroneal trochlea (PERTR), Periosteal reaction in the talar neck (OMANK), Head/Neck displaced to medial plane (DISHN), Osteophytes in the surface to the lateral cuneiform of cuboid (CUNOS), Osteophytes in cuneiforms-navicular joint (ONCUN), Forefoot joint surface eburnation (EBANT) and Merged phalanges (MERGE). These differences are more notorious between the populations dated in X-XIX centuries CE from Spain and Italy, and the frequencies only increased across time in the Spanish series.

Despite the fact that Catalonia and Lazio are situated close to each other, sharing a relatively similar history during the last two millennials, we found differences between the series, probably by such factors as lifestyle, geography, or occupation. If we consider Granollers as

part of the Spanish collections, their frequencies increase more. We found the same situation in our second work "A new methodology to estimate Pes planus from dry bones" where Granollers series shows the highest percentage of the flat foot among all our series.

In both research, we suggested two probable hypotheses to explain the high frequencies of pathological conditions in this contemporary collection, which are focused on the increase of the age-life (mean over 60 years old) and the influence of modern shoes. The relationship between old age and osteological damage has been widely studied [20], but the damage caused by inappropriate shoes in historical, even contemporary populations, was not.

Footwear is a cultural element that reflex diverse aspects of society. Footwear appeared after the full modern foot was formed (Août & Aerts [21]). The oldest chalcolithic footwear in the world is a right leather shoe from Areni-1 Cave, Vayots Dzor province, Armenia (3627-3377 centuries BCE), although the oldest archaeological findings of fiber and/or leather sandals where found in Arnold Research Cave, USA (7420 ±50 centuries BCE) [22]. Another example of the oldest footwear in the world corresponds to the right leather shoe belonging to the natural mummy called the "Hauslabjoch man" (Ötzi) [23], dated between 3300 centuries BCE. This specimen is a well-preserved and sophisticated shoe made of fur and leather, designed to walk on the snow, with a water-repellent effect [24]. Moreover, in the archaeological site "La Cueva de los murcielagos", located in Albuñol, Granada, Spain, an important number of baskets and sandals of esparto (5200-4600 centuries BCE) was found, which were woven through a braid system [25]. They correspond to the oldest archaeological elements of ropes and baskets found in the Iberian Peninsula.

Zipfel & Berger (2007) [26] who studied shod and unshod populations, discovered that prepastoral groups with unshod lifestyle demonstrated low frequencies of osteological anomalies, in contrast with recent groups. There are some studies [21,27–29] that mentioned the negative effects of footwear on the health of the foot in the populations.

For instance, 90% of patients with hallux valgus from Britain are female wearing fashion shoes/boots, that tighten the toes [28]. In St. Helena (South Atlantic Ocean) Shine [30] found a correlation between shoes and women, and high ratio of Hallux Valgus (48% of women who wore shoes for more than 60 years). Likewise, Japanese women who changed traditional

footwear for constrictive western-style shoes developed an increased frequency of hallux valgus during the second half of the XX century CE [28].

In contrast, walking barefoot favors a short step, causing a soft impact in the heel and comfortable pronation, without limitation caused by shoes. Also, running barefoot helps to initial contact with tiptoe, which favors an upright position. The above produces a ground reaction and shock absorption in different muscles of lower limbs, avoiding progressive damage in a short time [31]. The contact between foot and ground is favored by deepening of the plantar vault, which is able to adapt to different kind of grounds [29].

During the last years, we count with new technologies to design a comfortable footwear. However, despite developing a high industrial scale of shoes and many style options, some studies show that damage in feet and lower limb caused by modern shoes continues [26,32].

Like Granollers, Ras al-Hamra 5 (also named RH5, Neolithic) also demonstrated particular features in "Pathological and normal variability of the foot bones in osteological collections" and "A new methodology to estimate Pes planus from dry bones". RH5 series were sedentary fishermen who collected mollusks and sea resources in the Arabian coast of Oman. According to Coppa [33–35] and our own investigation, they present a high frequency of open sacral canal and other anomalies of the posterior arch of the sacral vertebrae, without lumbar dysraphism compromise. In RH5, we found the highest horizontal angle of the neck of talus from whole our collections (NECKANG, 33°), and feet demonstrated severe deformations. There are studies that relate defects of this canal to foot skeletal and muscular deformations [36]. However, in the whole collection, we did not find evidences to confirm a significant relationship between severe degrees of pes planus by the open sacral canal. Likewise, other factors such as endogamy could be a factor related to genetic anomalies, like sacral dysraphism cases and/or degenerative diseases/pathological flat foot.

"A new methodology to estimate flat foot in skeletal remains. The example of Mediterranean collections"

During this investigation, we emphasized the scarce bibliography and problematic about the determination of the non-pathological flat foot through bone remains, belonging to modern and historical collections. To build our new methodology, it was necessary to refer to paleoanthropology and clinical medicine, which research were useful to choose the parameters to follow. Thus, we proposed to estimate pathological flat foot by new metric and morphoscopic methods, based on clinical references, and on our hypothesis to find results in the talo-navicular and subtalar joints.

According to the results of the morphoscopic analysis, the presence of some variables in the foot bones helps to demonstrate a 73% of correct classification for *pes planus* and a 98% to non-*pes planus* (a 91% of accuracy in general). These features correspond to: Sustentaculum tali displaced to plantar-medial (PMSTA), Osteophyte in Calcaneus-Talus joint (CTOST), Trigonal articulation (TRART), Osteophyte in talar head (OHEAD), Neck/head displaced to medial (DISHN), Wide talar tail (WITTT), Os Trigonum (TRIGM), Alteration in tuberosity of navicular (EXTUB), Osteochondritis dissecans (OCDRT), and Osteophyte in Calcaneus-Cuboid joint (CCLOS). However, the number of characteristics that we observed at the beginning of our investigation was higher but they were discarded as non-relevant due to presence of *pes planus* (Chi-square analysis).

Therefore, we need a combination of nine morphoscopic variables to determine flat foot. Nonetheless, most of these features are a consequence of the osteoarthritis and are located almost in the same place (calcaneus-talus and talus-navicular joints), which is not difficult to find.

In addition, in order to increase the robusticity of the statistic results and the correct accuracy in future investigations, we proposed to increase the number of pathological flat foot individuals in future investigations.

"Bipartite cuneiforms in osteological and scanner series",

The fifth research included in this project approached different aspects of the bipartition of the cuneiform bones. We analyzed their frequency in live patients through CT scans and in historical/contemporary osteological series. These results were interpreted under descriptive and biomechanical parameters in the gait.

Considering each research that composes the thesis, this study has the smallest number of bibliographic references, which made it difficult to discuss its symptomatology and the world frequency. The bipartition of the cuneiform bones is a very rare condition, being the findings in the medial cuneiform almost the only variant registered.

However, our results (4/617 feet) demonstrated higher incidences of bipartition than the world frequency (1/320). We analyzed some factors that could explain these findings, such as the genetic or the origin of the samples, but probably the cause is the underestimation of this condition. Both, the lack of knowledge about the existence of this osteological bipartition, and the fact that most of the cases are asymptomatic, could considerably influence the number of current registers.

In relation to the other two cuneiforms, only one case of a probable bipartition of an intermediate cuneiform was registered by literature [37], and no one report of lateral cuneiform. Conversely, we observed one case in each of these bones in our series.

Even though it is known that bipartition is produced in the embryonic period, there is not clear why is much more frequent in the medial cuneiform than the others. In addition, the scarce reports of bipartition of the intermediate and lateral cuneiforms might be also by underestimation or bad diagnosis.

"Earliest Probable Case of Mueller-Weiss Disease from Ancient Egypt"

The last research of this thesis analyzed a pathological condition that could produce a severe osteological damage, especially in the talus and navicular bones, called Mueller-Weiss disease. In 2018, we reported an incidence of this pathology in a skeletonized foot from an adult male (individual 2569) who lived during the Hellenistic period, in Sharuna, Egypt. Due

this research is the least extensive of the thesis because it is only a report case, it will be one of the most widely discussed in this chapter.

Mueller-Weiss disease (MWD) is an uncommon bilateral disease, which is known by a spontaneous osteonecrosis of the tarsal navicular bone exhibit in adults, mainly females (6:4). It is characterize for its aspect of "comma-shaped" by the collapse of the structure, due the compression of the lateral portion. In living patients, this pathology is associated with pain and progressive deformity of the area.

This disease was mentioned for first time in 1925 by Schmidt (1925) who described a patient who suffered endocrinopathy [38] [39]. Nevertheless, the name "Mueller-Weiss disease" is due for the studies by the German surgeon Walther Müller (1927), who described the "double navicular" as an osteological manifestation in a patient who developed a severe deformity of the navicular bone in both feet. Müller attributed its etiology as a congenital origin, not traumatic, being the compression the cause of this alteration [40] [41]. After, in 1929, Konrad Weiss, described two similar cases, but implying osteonecrosis in his new definition, concept supported by current radiologists.

Between 1939 and 1998 few works were published about this disease [40,42–44]. These reports described osteonecrosis in the talo-navicular joint, arthritic changes and loss of volume in the midtarsal joint, dorsal protrusion, collapse in the lateral portion, fracture lines and fragmentation of the navicular. In the decade of 2000, studies increased [38,45–49], whose observations and findings were similar with the old reports, especially about the pre-disposition of the feet to injuries and pain. During this period, it was proposed that the bilateral deformation is more common than unilateral deformity, although the last mentioned is present in young patients [38].

The etiology of this disease is not still clear and there are many hypotheses. Maceira & Rochera (2004) [39], studied a considerable number of patients with MWD (101), who were mostly manual workers. They suggested that this high incidence is due to stressful conditions like poverty, malnourishment or war in the ossification/growing bones period. Therefore, they attributed the cause of the MWD to the combination of the abnormal force distribution in the navicular and a delayed ossification of this bone. In addition, this disease has been

associated with temporal episodes of nutritional stress and markers as Harris lines, hypoplasia of the dental enamel and *criba orbitalia*. Conversely, Doyle et al (2012) [45] who worked with 12 patients and 19 altered feet, did not find the same extreme factors in any patients with MWD, in contrast with the Maceira & Rochera conclusions.

Considering their osteological features, sometimes MWD is confused with the Kohler's disease (KD), a kind of osteochondrosis that affect to the talus and navicular in children. Nevertheless, both have distinguished features to discriminate. In first case, KD is unilateral (75%-80% with male predominance 6:1) and mainly affects to children between 3-7 years, associated to minor symptoms, with or without clinical pathology [40]. Furthermore, it must be distinguish from rheumatoid arthritis, diabetes, secondary osteonecrosis for trauma, steroid therapies among other conditions [42].

Other of the aspects not-widely approach our report case (See Results section) are the particular features of this foot and the probable damage in the gait. We considered his age, the bilateral damage, clear osteophytes, navicular comma-shaped, deformation of the mentioned talar bones (mainly in the dorsal portion), and the absence of fractures or external factors that could produce this damage. Regarding these not severe osteological damage, his most probable diagnosis is a stage II of MWD, according with the classification by Maceira & Rochera [39].

Due to the deformation of the bone structures, the analyzed individual in "Earliest Probable Case of Mueller-Weiss Disease From Ancient Egypt" (individual 2569 from Sharuna, Egypt) probable suffered a series of changes in the gait and displacement. Regarding its bibliography, we inquired in the probable symptoms in live. For instance, Fornaciari et al [48] and Wang et al [46] described the symptoms from patients with stage III-V of MWD, which demonstrated osteological deformity, ankle/medial foot pain during walking, and collapse of the medial longitudinal arch in the severe degrees.

Our analysis mentioned that Individual 2569 exhibit MWD features (stage II). Probably, he did not developed severe degrees of intense pain or bone destruction. Nevertheless, the navicular shows loss of volume and osteophytes, which could produce pain and tenderness in the mid-tarsal zone in the charge and in the gait.

This finding is the oldest case of the presence of Mueller-Weiss disease registered until our days. This is fundamental to deduce that, at least, this pathology have been present during millennia. Thus, humans have had to live with foot bone diseases for many centuries, whose solutions, in many cases, do not exist still or have not studied in depth.

### 3.3. Final considerations

This project proposed the study of the normal and pathological variability of the foot bones because there is a lack of knowledge from anthropological point of view. To carry out our aim, we set different researches that address specific aspects within this broad field.

Thus, we analyzed the sexual dimorphism of the navicular in contemporary populations and the biomechanical implications of the CFNB in the gait. Moreover, we studied the morphological differences and changes across time, the presence of non-pathological flat foot in dry bones, the frequencies of bipartite cuneiforms and the oldest case registered of Mueller-Weiss disease.

During this process, we proposed new morphological and/or metric methodologies to discriminate in laboratory (flat foot and sexual dimorphism). In both cases, we found significant differences to discriminate between groups, although we suggested to increase the number of pathological pes planus to strengthen the correct accuracy.

Considering each results of our research, we found that most of the osteological changes, especially the pathological, are visible in the Chopart joint. This articulation is an imaginary line that cross the talo-navicular and calcaneus-cuboid joints, which are one of the most mobile areas of the foot in the gait. Functionally, as we explained in "Morphological and biomechanical implications of cuboid facet of the navicular bone in the gait", Chopart joint suffered important functional changes in the human evolution. This change involved the calcaneus-cuboid joint, avoiding a wide supination as the primates and some of our ancestors [50–53]. Moreover, the glide and rotation movements produced in the talo-navicular joint, could be affect by factors as footwear, old age among others, producing changes in the osteological structure as degenerative diseases and/or enthesopathies.

Summarizing, the principal aim of this project that was the study of the normal and pathological variability, using historical and contemporary series. Here, we proposed new morphological and metric methods to improve the biological profile through the analysis of the foot structure. We obtained formulas with easy replicability that have a high percent of reliability and can be apply to diverse scopes of the feet. The cut-off points and equations help to determinate the non-pathological flat foot, and sexual dimorphism, between others.

Taking in consideration the scarcity of bibliography in some alterations and variants of the foot bones (bipartite cuneiform, MWD, cuboid facet), we improved the knowledge about these topics searching in their biomechanic, probable damage and consequences in the gait of the individuals. Currently, we have achieved a part, but much remains to be done.

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## 4. CONCLUSIONS

"A scientist is as weak and human as any man, but the pursuit of science may ennoble him even against his will".

Isaac Asimov

### **Conclusions**

This project analyzed the normal and pathological variability of the bones of the foot, focusing on factors that alter its anatomy. We studied a total of 890 samples, including mostly pairs of skeletonized feet of bioarchaeological and contemporary contexts of Madrid, Granada and Catalonia (Spain), Lazio (Italy) and Ras al-Hamra 5 (Oman) that allow us to conclude the following:

### 1- In relation to the normal variability:

- a. Sexual dimorphism can be observed in contemporary Spanish populations through metric variables in the navicular bone. Overall, the dimensions showed the highest values in the male group. The results, expressed through equations (binary logistic regression) and cut-off points (ROC curve), demonstrated different percentages of reliability to classify men and women, ranging from 76% to 93%. We suggest that our formulas could be useful in contemporary studies, bioarchaeological contexts of the Western Mediterranean population or series with similar characteristics.
- b. The cuboid facet of navicular bone (CFNB) is present in 52.7% of contemporary individuals in Spain, whose results showed no significant differences between collections and sexes. In addition, using dimensions of the talus, navicular and cuboid, we found that the sagittal length of the talar facet of the navicular (LSAGTAL) showed a relationship with the presence of CFNB. The latter could be explained by the movements that the talar facet is involved and by the position of the talo-navicular joint. The literature highlights its important role in supinatory movements in human evolution and primates. On the contrary, we did not find significant differences between living patients with / without CFNB through X-ray cinematography. Footwear, genetics (also as a recessive feature) or stressful activities are factors to consider when interpreting their presence in naviculars.

### 2- In relation to the pathological variability:

- a. We found significant differences in the normal and pathological variabilities of the foot bones, which are more notorious in the X-XIX centuries CE. The Italian series showed uniformity across time, conversely with Spanish collections that demonstrated an increase in the frequencies during the centuries. The average age of the population, lifestyle or the diversity of geography might explain these results.
- b. Regarding to the enthesopathies, pathologies and normal variabilities, we found that the degenerative processes are the main characteristics that demonstrated differences between Spanish and Italian collections. These differences increase with the inclusion of the contemporary series (Granollers). Factors as the footwear, old age, genetics or occupational stress might be involved in their causes.
- c. In relation to the study of the pathologic *pes planus*, we demonstrated the existence of a relationship among the morphological variables and the non-pathologic flat foot samples. They were expressed as a combination of features, which are mostly localized in the Chopart and subtalar joints. Likewise, two metric variables (The Horizontal angle of the neck of the talus NECKANG and Height of the cuboid HCUB) also discriminate between individuals with *pes planus* and healthy feet. Furthermore, considering our collections, we found great differences in the frequency of this condition, which could be influence by inadequate footwear, genetics, and increase of the life expectancy among others.
- d. Through exploratory exams in patients and observation of skeletal series, we found the presence of four feet affected by bipartition in their medial cuneiforms (4/617). In addition, we found one intermediate and one lateral bipartite cuneiform through the same exploratory method. These frequencies

are much higher than the world average, which might be explained by diverse factors as genetics or population, but it was most likely due to the underestimation of clinical frequencies of the samples.

e. Our investigation on Mueller-Weiss disease unveiled the oldest dating of this condition. Currently there is a limited bibliography in historical collections, which contrasts with the considerable number of clinical incidences. Nevertheless, although there are a growing interest in the study of this disease, its etiology is still unclear.

Finally, through analytic, comparative and descriptive analyses, this project proposed new approaches about the knowledge of the normal and pathological variability of the foot bones. Considering historical or modern burials and the often good condition of preservation of these kinds of samples, feet could be a source of valuable information of the past. Depending of the context, our methodologies might be useful to configure the biological profile of the individuals or in population researches.