Ann Med Res

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Annals of Medical Research

journal page: www.annalsmedres.org

Bipartitism in tarsal bones: A retrospective analysis of clinical and radiological features

Muhammed Koroglu[a,*], Mustafa Karakaplan[a], Huseyin Utku Ozdes[a], Idris Coban[b], Ilkan Kisi[c], Firat Al[a], Omer Acet[a], Emre Ergen[a], Okan Aslanturk[a], Zeynep Maras Ozdemir[d]

[a] *İnönü University, Faculty of Medicine, Department of Orthopaedics and Traumatology, Malatya, Türkiye*
[b] *Viranşehir State Hospital, Department of Orthopaedics and Traumatology, Şanlıurfa, Türkiye*
[c] *Gaziantep University, Faculty of Medicine, Department of Orthopaedics and Traumatology, Gaziantep, Türkiye*
[d] *İnönü University, Faculty of Medicine, Department of Radiology, Malatya, Türkiye*

ARTICLE INFO

Keywords:
Bipartite
Tarsal bone
Foot deformity
Separation
Diastasis
Articulation

Received: Aug 22, 2024
Accepted: Sep 20, 2024
Available Online: 26.09.2024

DOI:
10.5455/annalsmedres.2024.08.174

Abstract

Aim: Bipartitism in the foot, especially in the tarsal bones, is a rare anatomical variant characterized by dividing one bone into two separate ossification centers. This condition can be congenital or acquired, often remains asymptomatic, and is only discovered incidentally during radiological imaging. This study aims to present a comprehensive retrospective analysis of bipartitism in tarsal bones, assessing its prevalence, type of articulation, and associated clinical features in a cohort of patients.

Materials and Methods: A total of 4,645 patients were retrospectively evaluated from 6,145-foot images consisting of 4,975 computed tomography (CT) and 1,170 magnetic resonance imaging (MRI) scans obtained between 2015 and 2023. 15 patients with tarsal bipartitism were identified and contacted through the hospital system for in-person medical history and examination. Radiological exams assessed partial and complete bipartition, diastasis, and exostosis in the tarsal bones. Age, sex, side, trauma history, pain, foot deformity, gait disturbance, and other clinical findings were also recorded.

Results: Bipartitism was detected in 15 patients (0.32%), predominantly affecting males (87%). The most common variant was medial cuneiform bipartitism, observed in 13 patients (0.28%). Talus and calcaneus bipartitism were each observed in one patient (0.02%). Bilateral involvement was present in 67% of the cases, with 60% of patients reporting chronic pain. Complete dissociation of bipartite bones was noted in 80% of cases. Accompanying bipartitism, two patients had pes planovalgus, and one had hallux varus deformity.

Conclusion: Although bipartitism of the tarsal bones is rare, it remains a diagnostic challenge due to variability in clinical presentation and possible association with other foot deformities. Accurate diagnosis through advanced imaging techniques is essential to differentiate these variants from fractures and guide appropriate treatment. This study contributes valuable insights into the prevalence and clinical impact of bipartitism in the tarsal bones, emphasizing the importance of early diagnosis and individualized treatment strategies.

Introduction

Bipartitism in the foot, particularly in the tarsal bones, is a rare anatomical variant where a bone is divided into two distinct ossification centers. This condition may be congenital or acquired, often remaining asymptomatic and unnoticed until it is incidentally discovered during imaging for other conditions [1]. The bipartite bones can vary significantly in their presentation, ranging from partial to complete separation, and the articulation between the bone segments may involve syndesmosis, synchondrosis, or a combination of these joint types [2]. Bipartitism has been documented in various tarsal bones, including the talus, navicular, cuneiforms, and calcaneus. While the clinical significance of these variants is often minimal, they can occasionally lead to symptoms such as pain, restricted motion, and in rare cases, conditions like tarsal tunnel syndrome [3]. The understanding of bipartitism is crucial in differentiating these anatomical variants from

*Corresponding author:
Email address: m.koroglu91@gmail.com (Muhammed Koroglu)

fractures, especially in the context of trauma where misinterpretation can lead to inappropriate management [4]. Despite its rarity, the presence of bipartitism has been observed across different patient demographics, with varying degrees of clinical impact. In some cases, surgical intervention is necessary when conservative management fails to alleviate symptoms associated with this condition [5]. However, the natural history of bipartite bones often shows a tendency towards spontaneous resolution of symptoms, particularly in younger patients [6].

This study aims to provide a thorough retrospective analysis of bipartitism in the tarsal bones, assessing the prevalence, involvement characteristics, and associated clinical features in a cohort of patients. By examining the radiological patterns and clinical outcomes, the study intends to deepen the understanding of bipartite bones in the foot and provide valuable insights for clinicians in managing this anatomical variant.

Materials and Methods

We retrospectively evaluated 6,145 feet from a total of 4,645 patients for bipartitism in our study, which included 4,975 computed tomography (CT) and 1,170 magnetic resonance imaging (MRI) radiological images taken between 2015 and 2023. After retrospective screening, 15 patients with tarsal bipartitism were contacted via the hospital system, and their medical history and examinations were conducted in person. By radiological examination, we investigated partial and complete bipartition, diastasis, and exostosis in patients with bipartition in the tarsal bones. Age, sex, side, history of trauma, acute or chronic pain, foot deformity, gait disturbance, and additional clinical findings were also recorded. We considered acute pain as pain that has been present for less than 3 months and chronic pain that has been present for more than 3 months [7]. If the bone integrity consists of two completely separate parts, it is defined as complete articulation; if there is a bone bridge, even partial, between the parts, it is defined as partial articulation [8]. Diastasis was recorded when the separation of the two bone segments was larger than 2 mm [9]. Patients with a history of significantly altered foot anatomy due to severe trauma were excluded from the study because this situation could potentially lead to misinterpretation in the assessment and determination of bipartition during our scanning procedures. Additionally, CT and MRI scans that did not include all tarsal bones within the field of view, or those with artifacts due to excessive motion and metal, were excluded. However, cases with distal tibia, metatarsal, or tarsal fractures that did not interfere with the recognition of bipartition were included in the study.

Radiological examinations were performed on a digital workstation (Sectra Workstation IDS 7; Sectra AB, Linköping, Switzerland). MRI and CT assessments were performed by a radiologist with 15 years of MRI experience and 11 years of dedicated musculoskeletal imaging experience. CT and MRI scans were performed by four senior orthopedic residents briefly trained by the same radiologist. The typical MRI imaging protocol included axial T1, axial proton density (fat sat), sagittal T1, sagittal T2 fast spin echo (fat sat), coronal T1, coronal T2 fast spin echo (fat sat), and coronal proton density (fat sat) sequences. Foot CT scans were performed on CT scanners (Siemens, Munich, Germany) located in the radiology and emergency departments of our institution. Scans were obtained with tube voltages between 120 and 130 kV and tube currents between 72 and 104 mA. The slice thickness ranged from 0.5 to 2 mm. The ethical standards set out in the 1964 Declaration of Helsinki and its subsequent amendments were followed in this research.

This study was approved by our Inonu University's Institutional Review Board (2024/5323-18).

Results

Out of the 4,645 patients whose MRI and CT scans were examined, 2,203 were female (47.4%) and 2,442 were male (52.6%). Of the 6,145-foot images examined, 4,975 were CT scans (81%) and 1,170 were MRI scans (19%). Bipartitism in tarsal bones was detected in 15 patients (0.32%) out of 4,645; 13 of these patients were male (87%) and two were female (13%). The mean age of the patients was 36.3 years. Talus bipartitism was found in one patient (0.02%) (Figure 1), calcaneus bipartitism in one patient (0.02%) (Figure 2), and medial cuneiform bipartitism in 13 patients (0.28%) (Figure 3). Bipartitism was not observed in the navicular, cuboid, and other tarsal bones. The most common bipartitism was medial cuneiform bipartitism. Medial cuneiform bipartitism was symptomatic in seven patients (54%). While one out of six feet with partial medial cuneiform bipartitism experienced chronic pain, nine out of 15 feet with complete bipartitism had chronic pain. A total of 25 bipartite foot bones (0.4%) were detected, with bilateral involvement in 10 of the patients (67%). Trauma history was positive in four patients (27%). Nine patients (60%) had accompanying chronic pain. There was diastasis of more than 2 mm in bipartite bones in four patients (27%). Four patients (27%) with medial cuneiform bipartitism were found to have 1st tarsometatarsal joint exostosis. Additionally, one patient (7%) with medial cuneiform bipartitism exhibited navicu-

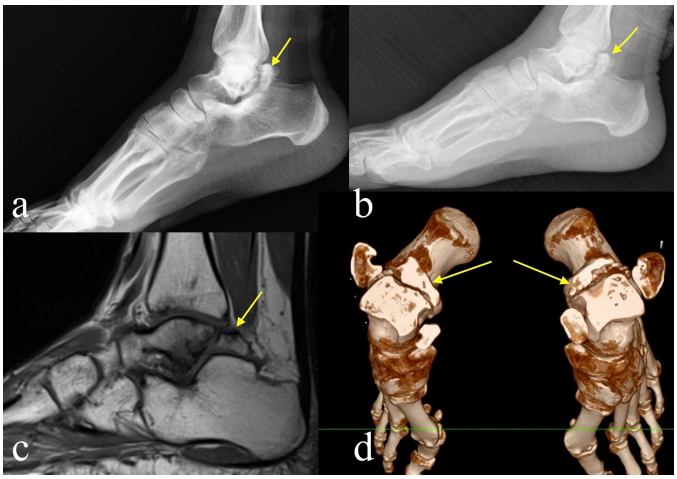

Figure 1. Right (a) and left (b) lateral foot radiography, magnetic resonance imaging (c) and three-dimensional tomography (d) images of bilateral complete symptomatic talus bipartition in a 30-year-old man.

Table 1. Clinical and radiologic data of the patients.

	Age	Sex	Side	Bone	Trauma History	Pain	Diastasis	Exostosis	Partial/Complete Articulation	Foot Deformity	Gait Disturbance	Additional Findings
P-1	37	M	Right	Medial Cuneiform	Positive	Chronic	Negative	Naviculocuneiform joint	Partial	Negative	Negative	ATFL rupture and Talus OCD
			Left	Medial Cuneiform	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None
P-2	26	M	Right	Medial Cuneiform	Positive	Negative	Negative	Negative	Partial	Negative	Negative	Phalanx fracture
P-3	31	M	Right	Medial Cuneiform	Positive	Chronic	Negative	1st Tarsometatarsal joint	Complete	Negative	Negative	None
			Left	Medial Cuneiform	Positive	Negative	Negative	1st Tarsometatarsal joint	Complete	Negative	Negative	Femur fracture
P-4	42	F	Left	Medial Cuneiform	Positive	Negative	Negative	Negative	Partial	Negative	Negative	Bimalleolar fracture
P-5	85	M	Right	Medial Cuneiform	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None
			Left	Medial Cuneiform	Negative	Negative	Negative	Negative	Complete	Negative	Negative	Total knee prothesis
P-6	26	M	Right	Medial Cuneiform	Negative	Chronic	Positive	Negative	Complete	Negative	Negative	None
			Left	Medial Cuneiform	Negative	Chronic	Positive	Negative	Complete	Negative	Negative	None
P-7	18	M	Left	Medial Cuneiform	Negative	Chronic	Negative	Negative	Complete	Negative	Negative	Os naviculare
P-8	13	M	Right	Medial Cuneiform	Negative	Chronic	Positive	Negative	Complete	Pes planovalgus	Positive	Calcaneal apophysitis
			Left	Medial Cuneiform	Negative	Chronic	Positive	1st Tarsometatarsal joint	Complete	Pes planovalgus	Positive	Calcaneal apophysitis
P-9	73	M	Right	Medial Cuneiform	Negative	Chronic	Positive	1st Tarsometatarsal joint	Complete	Charcot foot	Positive	Charcot neuropathy
			Left	Medial Cuneiform	Negative	Negative	Negative	Negative	Partial	Negative	Negative	None
P-10	12	F	Left	Medial Cuneiform	Negative	Chronic	Negative	Negative	Complete	Hallux varus	Positive	1st phalanx macrodactyly
P-11	48	M	Right	Medial Cuneiform	Negative	Negative	Negative	Negative	Partial	Negative	Negative	None
			Left	Medial Cuneiform	Negative	Chronic	Negative	Negative	Complete	Negative	Negative	Plantar fibromatosis
P-12	51	M	Left	Medial Cuneiform	Negative	Negative	Negative	1st Tarsometatarsal joint	Partial	Negative	Negative	None
P-13	50	M	Right	Medial Cuneiform	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None
			Left	Medial Cuneiform	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None
P-14	30	M	Right	Talus	Negative	Chronic	Positive	Negative	Complete	Negative	Positive	Peritalar arthritis
			Left	Talus	Negative	Chronic	Positive	Negative	Complete	Negative	Positive	Peritalar arthritis
P-15	3	M	Right	Calcaneus	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None
			Left	Calcaneus	Negative	Negative	Negative	Negative	Complete	Negative	Negative	None

locuneiform joint exostosis. There was complete dissociation of bipartite bones in 12 patients (80%). Two patients (13%) had pes planovalgus, one patient (7%) had hallux varus, and one patient (7%) had Charcot foot deformity. Five patients (33%) had gait disturbances. Additional accompanying clinical information is detailed in Table 1.

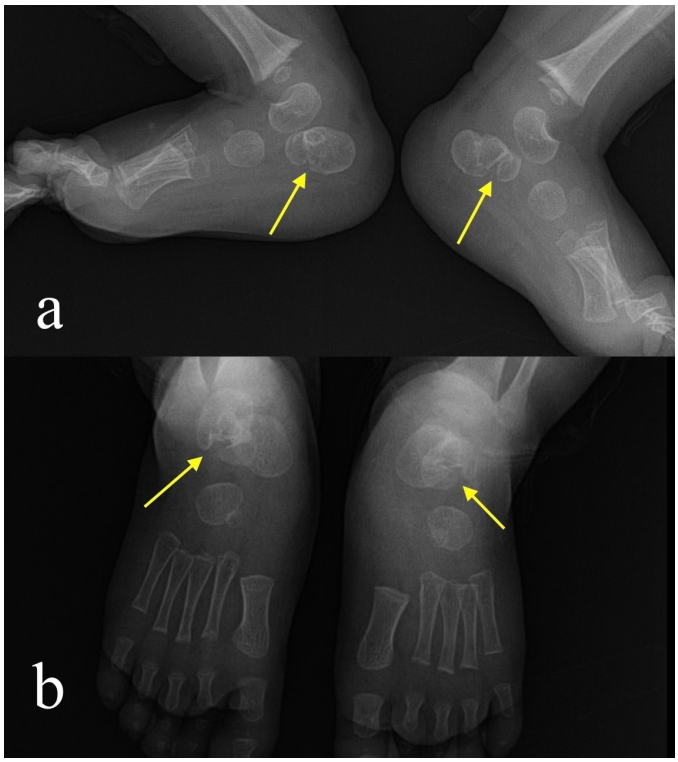

Figure 2. Lateral (a) and anteroposterior (b) foot radiographs of bilateral complete asymptomatic bipartition of calcaneus in a three years old child.

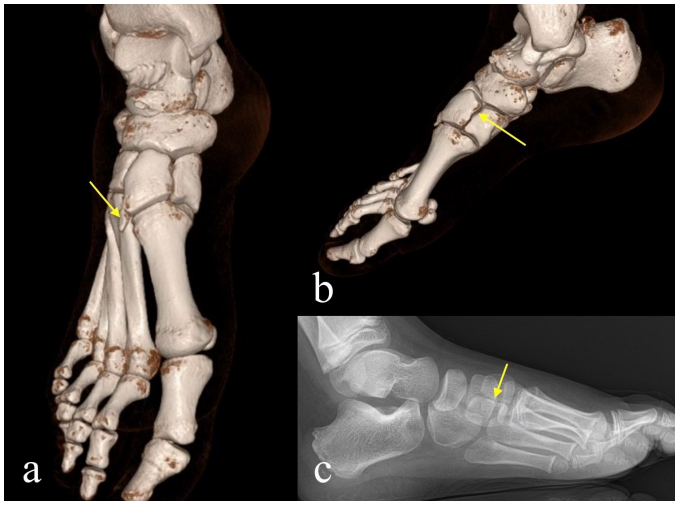

Figure 3. Three-dimensional tomography image of a 31-year-old man with symptomatic complete bipartition of the medial cuneiform (b) and exocytosis (a) of the 1st tarsometatarsal joint of the right foot. Lateral (c) foot radiography of thirteen-year-old male patient with symptomatic complete bipartition of the medial cuneiform.

Discussion

In our study, the incidence of tarsal bipartitism was 0.32% among 4,645 patients, with medial cuneiform bipartitism being the most frequently observed variant. Specifically, we found an incidence of 0.27% for medial cuneiform bipartitism. Although the true prevalence of this condition remains uncertain, it has been reported in the literature to range between 0.1% and 7% [10]. In our research, we observed a notable male predominance of 87% for medial cuneiform bipartitism. The literature review indicated a higher incidence of medial cuneiform bipartitism in men [11]. Bipartitism of the medial cuneiform is one of the forms of tarsal bone bipartition. This condition typically presents as a horizontal or oblique division of the medial cuneiform into two distinct ossicles. The dorsal segment is usually smaller, and the articulation between the two fragments may consist of a synchondrosis, a syndesmosis, or a combination of the two [12]. In our study, of the 21 medial cuneiform bipartite feet, six were partial and 15 were complete bipartite. The rate of chronic pain in partial medial cuneiform bipartite feet was 17%, whereas it was 60% in complete bipartite feet. Clinically, this condition can present with midfoot pain, often following trauma, and may be mistaken for a fracture. MRI is particularly useful for identifying this variant and differentiating it from fractures and other conditions [13,14]. Steen et al. reported that symptomatic patients may have an exostosis over the first TMT of the foot [10]. Chronic pain was present in 10 of the 21 feet (47.6%) with medial cuneiform bipartitism, while 11 of the feet (52.4%) were asymptomatic in our study. We identified 1st tarsometatarsal joint exostosis in three patients with medial cuneiform bipartitism who had chronic pain (Figure 3). Additionally, we found naviculocuneiform joint exostosis in one patient with medial cuneiform bipartitism who had chronic pain. In two patients with medial cuneiform bipartitism who did not experience pain, 1st tarsometatarsal joint exostosis was detected. Steen et al published a report of a patient with a bipartite cuneiform with concomitant Lisfranc instability on the same side [10]. Therefore, it is important to evaluate the stability of the Lisfranc joint. There is limited evidence linking medial cuneiform bipartitism with specific congenital conditions, but it may coexist with general foot deformities such as flatfoot (pes planus), especially when there is a familial tendency towards these anomalies [15]. In our study, among patients with bipartite medial cuneiform, pes planovalgus was identified in two patients, hallux varus in one, and Charcot foot deformity in another. Of these patients, only four exhibited gait disturbances. Pain can often be alleviated with conservative approaches, such as oral analgesics and brief non-weight-bearing periods, which may resolve symptoms. If these measures are ineffective, surgery either removing the small fragment or fusing the two fragments can be considered. Successful outcomes are generally achievable in treating this condition [11].

Talus bipartitus was initially documented in a dissertation by Strehle at the University of Leipzig in 1928 [16]. The prevalence and etiology of talus bipartitus are unknown, there have been published case reports in the literature. It is believed that the talus usually develops from a single ossification center, and talus bipartitus may result from either the fragmentation of this center or the failure of fusion of a secondary ossification center [17]. Talus bipartitus can be congenital or result from trauma during the developmental phase. Imaging modalities such as MRI are essential in identifying this anomaly and distinguishing it from other conditions like os trigonum and talar fractures.

Accurate diagnosis is essential, as overlooked large fragments may lead to clinical conditions like tarsal tunnel syndrome, where the etiology in the patient remains undefined [3,17]. In our study, talar bipartition was observed in one male patient (0.02%) out of 4,645 patients with symptomatic bilateral involvement. Zwiers et al. reported 23 patients diagnosed with this condition, highlighting a male predominance (61%) with a median age of 15.5 years at presentation in their literature review. The clinical presentation was primarily characterized by ankle pain, occurring in 96% of symptomatic cases, and restricted subtalar motion in 54% of cases [18]. The patient with talar bipartitus in our study reported no history of trauma and mentioned experiencing chronic pain due to an incorrect diagnosis over the past five years. MRI examination revealed diastasis of the talar bipartite along with arthritic changes in the peritalar region (Figure 1). The patient experienced painful movement in both the ankle and subtalar joints, which was found to cause a gait disturbance during the clinical examination. A surgical intervention plan was developed for the patient who did not show improvement following the initial stage of conservative treatment. If the fragment involves both the ankle and subtalar joint surfaces, fixation is recommended, as excision may reduce the joint surface area and lead to osteoarthritis. If only the subtalar joint is involved, the choice between excision and fixation is assessed based on the size of the fragment and any existing osteoarthritic changes [18,19]. Conditions that must be distinguished from talus bipartitus, such as os trigonum and posterior talar process fractures, are crucial for determining the appropriate surgical treatment. Unlike the more commonly occurring os trigonum, which generally measures less than 1 cm, the posterior fragment in talus bipartitus is significantly larger, reaching up to 4 cm [11,20]. The bipartite navicular is a rare clinical condition where the navicular bone consists of two fragments of unequal size, typically occurring bilaterally. A patient with bipartite navicular was not identified in our study. The first documented case of this condition was published by Muller in 1927. The etiology of the bipartite navicular is unclear [11,21]. It is often mistaken for conditions such as Köhler's disease; however, bipartite navicular is a separate and distinct condition. Patients with a bipartite navicular often present with midfoot pain, particularly after activity, and may show signs of pes planus due to the subluxation of the navicular on the talar head [22]. Therefore, it is a condition that should be considered when assessing the etiology in patients with pes planus. Bipartition of the navicular bone may also be associated with accessory navicular syndrome, in which an additional ossicle (os naviculare) can cause chronic pain and tenderness along the medial side of the foot [23]. Other conditions that can affect the navicular include Müller-Weiss Disease, a poorly understood disorder characterized by dorsolateral fragmentation and collapse of the navicular bone [24]. Although bipartite navicular is rare, it is essential to consider and differentiate overlapping clinical conditions based on the specific characteristics of the navicular bone to ensure an accurate diagnosis.

Calcaneus bipartitism, although rare, is significant because it can mimic other pathological conditions, such as fractures or accessory ossicles. Bifid os calcis was first described by Sever in 1930 [25]. This condition involves the division of the calcaneus into two distinct ossification centers, often separated by a cartilaginous or fibrous cleft. In our study, we detected a case of bilateral, asymptomatic calcaneus bipartitism in a 3-year-old male patient with no underlying genetic disease (Figure 2). The patient exhibited no foot deformity or gait disturbance. Calcaneus bipartitism is often bilateral and asymptomatic but may lead to biomechanical issues in the foot if the fragments do not fuse properly during development. It is important to distinguish this condition from others, such as calcaneovalgus foot deformity and conditions associated with skeletal dysplasias [26]. Calcaneus bipartitism has been linked to congenital conditions like fibrodysplasia ossificans progressive (FOP) and Larsen syndrome. FOP, a rare genetic disorder, causes abnormal bone formation in muscles and connective tissues, leading to progressive immobility. In FOP patients, calcaneal bipartitism, along with plantar calcaneal spurs, may be an early radiographic finding, aiding in diagnosis [27]. In Larsen syndrome, a genetic disorder that affects the development of bones, joints, and cartilage, bipartite or tripartite calcaneus may be seen, contributing to the overall foot deformity that characterizes this condition. Patients with Larsen syndrome often present with dislocations of large joints, clubfoot, and other congenital anomalies, making the management of bipartitism particularly challenging in these cases [27,28]. We should consider that calcaneal bipartition may be a key finding for the early diagnosis of genetic diseases and the differential diagnosis of associated conditions. This study has several limitations. First, it is a retrospective analysis. Second, due to the rarity of tarsal bone bipartitism, only small case series were available, which precluded statistical analysis. Consequently, the level of evidence is low; however, this study is expected to contribute valuable insights for future comprehensive research.

Conclusion

Bipartitism of the tarsal bones, including the talus, navicular, medial cuneiform and calcaneus, is a rare and complex anatomical variant that causes significant diagnostic challenges. Its variability in clinical presentation and association with congenital conditions necessitates accurate diagnosis through advanced imaging techniques to distinguish these variants from fractures and other foot pathologies. Understanding the natural history and clinical implications of tarsal bone bipartitism is essential for orthopedic surgeons and physicians, particularly when underlying congenital diseases or foot deformities are present. Differentiating bipartitism from accessory bones and recognising its potential as an indicator of greater skeletal anomalies are important steps in ensuring optimal patient care and management.

Data availability statement

The datasets generated during and/or analyzed during the current report are available from the corresponding author on reasonable request.

Funding statement

No disclosure.

Conflict of interest statement

None of the authors has financial interest in any of the products, devices, or drugs mentioned in this manuscript.

Ethical approval

This study was approved by our Inonu University's Institutional Review Board (2024/5323-18). Informed consent was obtained from all participants.

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