Surgical and Radiologic Anatomy (2019) 41:1433–1439
https://doi.org/10.1007/s00276-019-02354-0

ORIGINAL ARTICLE

Incidence of symptomatic os trigonum among nonathletic patients with ankle sprain

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Received: 9 July 2019 / Accepted: 25 September 2019 / Published online: 14 October 2019

Abstract

Purpose Os trigonum syndrome is a rare condition, often affecting athletes. A paucity of data exists on the incidence of os trigonum syndrome in nonathletic population. The study aimed to determine the incidence and clinical characteristics of os trigonum syndrome in nonathletic patients with sprained ankles.

Methods The sample consisted of 798 adolescent and adult patients that attended the emergency department or Foot and Ankle Clinic with acute ankle sprain. Lateral and/or oblique lateral radiographs of the feet were screened for the presence of os trigonum in relation to age and gender. A cohort of 163 patients with os trigonum was followed up prospectively over a 48-month period to correlate the presence of the os trigonum with patient symptomatology.

Results Os trigonum was found in 20.4% (163/798) of sprained ankles. Patients aged 18–35 exhibited most os trigonum [42.3% (69/163)], with higher incidence in females. 5.5% (9/163) of the os trigonum patients developed an os trigonum syndrome after a standard treatment of an ankle sprain [3.8% (3/78) of males and 7.1% (6/85) of females]. Females aged between 18 and 35 years had higher incidence of os trigonum syndrome compared to males of a similar age.

Conclusion Os trigonum syndrome should be suspected in nonathletic patients with an ankle sprain unresponsive to standard treatment. About 1.1% of acute ankle sprain patients develop an os trigonum syndrome. This finding can help identify the source of a patient's symptoms, leading to an accurate diagnosis, appropriate treatment and reducing the potential chronic symptoms.

Keywords Os trigonum · Ankle sprain · Nonathlete · Posterior impingement · Anatomy

Introduction

Accessory bones are common skeletal variations in the foot and ankle. The os trigonum is one of the largest and most common accessory ossicles in the foot and ankle region [14, 18]. The os trigonum is a triangular ossicle located at the posterior aspect of the talus adjacent to the lateral tubercle. The posterior process of the talus is made up of two tubercles, the medial tubercle and the lateral tubercle. Between these tubercles is a groove that allows the passage of the

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flexor hallucis longus tendon from the posterior ankle to enter the tarsal tunnel. The lateral tubercle of talus is generally larger, and it is here that the secondary ossification center associated with the development of os trigonum is found. It begins to appear between the ages of 8–11 years in boys and 8–10 years in girls as a secondary center of ossification for the posterior process of talus. Normally it fuses with talus within a year [4]. If the ossicle fails to fuse, it is referred to as the os trigonum. Fusion results in the formation of a prominent lateral tubercle of the posterior talus, termed as a Stieda's process.

The frequency of occurrence of the os trigonum varies from 2 to 50% [2, 11]. Symmetry is not always present. Os trigonum was found to occur bilaterally in 2% of the general population and in 50% of os trigonum population [7, 20]. However, in Burman and Lapidus series, 14 cases had bilaterally free ossicles and 9 cases had one unfused and one fused ossicle on the other side [2]. On the other hand, examination of 813 dry-bone tali revealed no single case with a bilateral occurrence of os trigonum [11].

Os trigonum remains mostly asymptomatic. However, this anatomic variant is susceptible to fracture by virtue of its location [32]. Fracture and chronic inflammatory conditions of os trigonum are common among athletes and ballet dancers requiring forceful, repetitive ankle joint plantarflexion [5]. However, symptomatic os trigonum has been noted to occur in nonathletes [16, 19, 27]. Watson and Dobas described a classification scheme for the anatomical variations of the posterolateral tubercle of talus [30]. Type 1 was identified as a normal tubercle without clinical consequence. Type 2 is Stieda's process (an enlarged posterolateral process). Type 3 is an accessory bone, the os trigonum, which may be the source of discomfort because of repetitive trauma. Type 4 is a fused os trigonum, which forms a synchondrosis or syndesmosis with the talus.

The anterior surface of os trigonum articulates with the posterolateral process of talus and the inferior surface articulates with the superior aspect of calcaneus. The posterolateral process serves as an attachment point for the posterior talofibular ligament and the posterior talocalcaneal ligament. The os trigonum communicates superiorly with the posterior capsule of the talocrural joint, inferiorly with the posterior talocalcaneal ligament, medially with the flexor hallucis longus tendon sheath and laterally with the origin of the posterior talofibular ligament [25]. The sheath of the flexor hallucis longus is often chickened in this area to accommodate the higher degree of friction encountered as it passes through the posterior ankle [23]. Such thickening may predispose this segment of tendon to repeated traction injury in patients with high physical demands. In extreme ankle dorsiflexion, the os trigonum is compressed between the flexor hallucis longus tendon and the posterior talofibular ligament. With end-range plantarflexion, the superior aspect of the calcaneus directly compresses the ossicle against the tibia. Extremes of motion in both plantarflexion and dorsiflexion may irritate the flexor hallucis longus tendon along its angled course at the posterior ankle [25]. Chronic inflammation of flexor hallucis longus and other local structures can create symptoms consistent with tarsal tunnel syndrome [8].

Posterior ankle impingement (PAIS) is a condition characterized by posterior ankle pain in plantarflexion. It is usually aggravated by repetitive plantarflexion and can result from either an acute injury or simple overuse [21]. Many causes of PAIS have been described, such as os trigonum, enlarged Stieda's process, loose body, avulsion fracture of posterior distal tibia, soft tissue origin (flexor hallucis longus tendinopathy, posttraumatic synovitis, ganglions, cysts), and joint origin (pigmented villonodular synovitis, osteophytes, osteochondritis dissecans) [21].

Patients with PAIS were more likely to have an os trigonum [33]. The onset of pain in os trigonum syndrome is often associated with an ankle sprain that is unresponsive to treatment. However, the coincidence of this injury with ankle sprains is likely underreported because this syndrome is rare and many general physicians are not familiar with its clinical presentation [25]. Based on our experience, we hypothesized that an os trigonum syndrome would be more common in nonathletes with sprained ankles than previously reported. The aim of this study was to elucidate the incidence of the symptomatic os trigonum in nonathletic patients with sprained ankles, thus providing a basis for future improvements in risk assessment, diagnosis, and early management of os trigonum syndrome.

Methods

The study was a prospective longitudinal study over a 48-month period made between 2015 and 2019. All patients seen at the Jordanian Royal Medical Services Emergency Room or Foot and Ankle Clinic, who reported at initial examination with an acutely twisted ankle, were included in the study. These patients were able to relate a specific traumatic event that occurred within the last week in which the ankle was not able to support the forces exerted on it. Excluded from the study were any patient with an ankle fracture, avulsions of the tibia or fibula, patients who underwent hindfoot surgery, patients with pathologies including osteolysis, severe arthritis, calcification and congenital deformations of the ankle joint.

The inclusion criteria were patients aged 14 years and older, and the availability of high-quality radiographs. This study was approved by the Academic Research Council and the Ethics Committee of the Faculty of Medicine at the University of Jordan and the Jordanian Royal Medical services.

Informed consent was obtained from all individual participants included in the study.

Lateral and/or oblique lateral radiographic views of 798 sprained ankles (440 men and 358 women) were included. The presence of an os trigonum bone was recorded by three researchers independently. Patient age and gender data were recorded. None of the patients was a professional athlete or a ballet dancer. The radiographs were of a single foot from each patient, either right or left. Of the 798 ankles, 163 had an associated os trigonum. The patients with os trigonum were followed up prospectively to correlate the presence of the os trigonum with patient symptomatology. MRI scans were obtained only from patients with suspected posterior ankle pathology to substantiate the clinical findings and diagnose any other associated pathologies.

The patients were divided into four separate age groups to ensure that the analysis includes all age ranges in the study population: teenagers (14–17 years), young adults (18–35 years), middle-aged (36–55 years) and elderly (56 years and over).

Statistical analysis

GraphPad Prism version 6.04 for Windows (Graph-Pad Software, La Jolla, CA) was used. The incidence of os trigonum and os trigonum syndrome was compared between genders (male vs. female) and age groups using the Fisher exact test. Odds ratios and 95% confidence intervals were estimated with logistic regression models. The significance threshold was set at 0.05.

Results

The average age was 36.5 (range 14–60) years. An os trigonum was found in 20.4% (163/798) of sprained ankles. The proportion of os trigonum was statistically higher among females [OR 0.69; 95% confidence interval (CI) 0.49–0.98, $p < 0.05$]. In males, an os trigonum was found in 17.7% (78/440) of sprained ankles. In females, an os trigonum was found in 23.7% (85/358) of sprained ankles. The incidence of sprained ankles and os trigonum in number and percentage is shown in Table 1.

An increased frequency of ankle sprain has been noted in the population ranging from 18 to 35 years, with male predominating. After the age of 35 years, the incidence was higher for females (Table 2). The highest incidence of os trigonum in sprained ankle patients was found in the young adult group (18–35 years) in both sexes. Os trigonum was identified in 7.1% (31/440) of the young adult males and in 10.6% (38/358) of the young adult females. The proportion of os trigonum was statistically higher among the young adult females ($p < 0.05$). No significant difference in os trigonum proportion in other age groups was observed. The lowest incidence of os trigonum was found in the elderly group. Os trigonum was identified in 0.2% (1/440) of the elderly males and not identified in the elderly females. The incidence of os trigonum in number and percentage in the different age groups is shown in Table 2.

Table 1 Incidence of os trigonum and os trigonum syndrome in patients with sprained ankles according to gender

	Ankle sprain	Os trigonum, *N* (%)	Symptomatic os trigonum, *N* (%)
All	798	163 (20.4)	9 (5.5)
Male	440 (55.1)	78 (17.7)	3 (3.9)
Female	358 (44.9)	85 (23.7)	6 (7.1)
Odds ratio		0.69	0.53
95% CI		0.49–0.98	0.13–2.18
p value		0.04*	0.49

*Significant $p < 0.05$, Fisher exact test

Of the 163 ankles with os trigonum, 5.5% (9/163) of the patients developed an os trigonum syndrome after a standard treatment of an ankle sprain. The overall incidence of os trigonum syndrome was higher in females. With the numbers available, no significant difference could be detected in symptomatic os trigonum proportions between genders in different age groups ($p > 0.05$). Of the 163 feet with os trigonum, 3.9% (3/78) of males and 7.1% (6/85) of females developed the symptoms of os trigonum syndrome (Table 1). Their symptoms varied between posterolateral ankle pain with passive ankle plantarflexion or dorsiflexion of the great toe to stiffness and swelling in the posterior aspect of the ankle. The symptoms were reproduced by forced ankle plantarflexion (a positive posterior ankle impingement sign). Pain was located at the posterolateral ankle in all patients, two patients had tenderness of flexor hallucis longus and a decrease in plantarflexion compared with the unaffected ankle. In all patients, the symptoms were aggravated by plantarflexion during certain stages of Muslim prayer.

An increased rate of os trigonum syndrome has been noted in the population ranging from 18 to 35 years. 2.6% (2/78) of the young adult males and 4.7% (4/85) of young adult females developed the symptoms of os trigonum syndrome following an ankle sprain. The incidence os trigonum syndrome in number and percentage in the different age groups is shown in Table 2.

On MRI, bone marrow edema in the os trigonum and the posterior aspect of talus was observed, along with soft tissue edema posterior to talus (Figs. 1, 2). Flexor hallucis longus tenosynovitis with fluid along the tendon sheath was noted on MRI (Fig. 1).

Only two patients were successfully managed with the primary conservative strategy. Seven patients were

Table 2 Incidence of os trigonum and os trigonum syndrome in patients with sprained ankles according to age and gender

Age groups	Ankle sprain			Os trigonum			Symptomatic Os trigonum		
	All $N=798$ N (%)	Male $N=440$ N (%)	Female $N=358$ N (%)	All $N=163$ N (%)	Male $N=78$ N (%)	Female $N=85$ N (%)	All $N=9$ N (%)	Male $N=3$ N (%)	Female $N=6$ N (%)
14–17 years	228 (28.6)	147 (18.4)	81 (10.2)	40 (5.0)	23 (5.2)	17 (4.7)	1 (0.6)	0	1 (1.2)
p value					0.36			0.43	
18–35 years	314 (39.3)	202 (25.3)	112 (14.0)	69 (8.7)	31 (7.1)	38 (10.6)	6 (3.7)	2 (2.6)	4 (4.7)
p value					0.00*			0.68	
36–55 years	244 (30.6)	86 (10.8)	158 (19.8)	53 (6.6)	23 (5.2)	30 (8.4)	2 (1.2)	1 (1.3)	1 (1.2)
p value					0.19			1.00	
≥ 56 years	12 (1.5)	5 (0.6)	7 (0.9)	1 (0.1)	1 (0.2)	0	0	0	0
p value					0.42				

*Significant $p<0.05$, Fisher exact test

Fig. 1 A 35-year-old female presents with persistent posterior ankle pain 6 months following an ankle sprain. **a** Lateral X-ray of foot showing os trigonum. **b** Axial PD fat-saturated MR image showing fluid around the flexor hallucis longus tendon with faint hyperintensity within its substance consistent with FHL tenosynovitis. **c** Sagittal PD fat-saturated MR image showing os trigonum with edema in the posterior aspect of talus and the ossicle. **d** Sagittal T1W1 MR image showing os trigonum with hypointensity in the posterior aspect of talus and the ossicle. All MR images show soft tissue edema posterior to talus

operated for a symptomatic os trigonum using an open excision through a posterolateral approach. Patients were followed up every 3 months, with maximum follow-up of 24 months. Postoperative pain relief was obtained in all

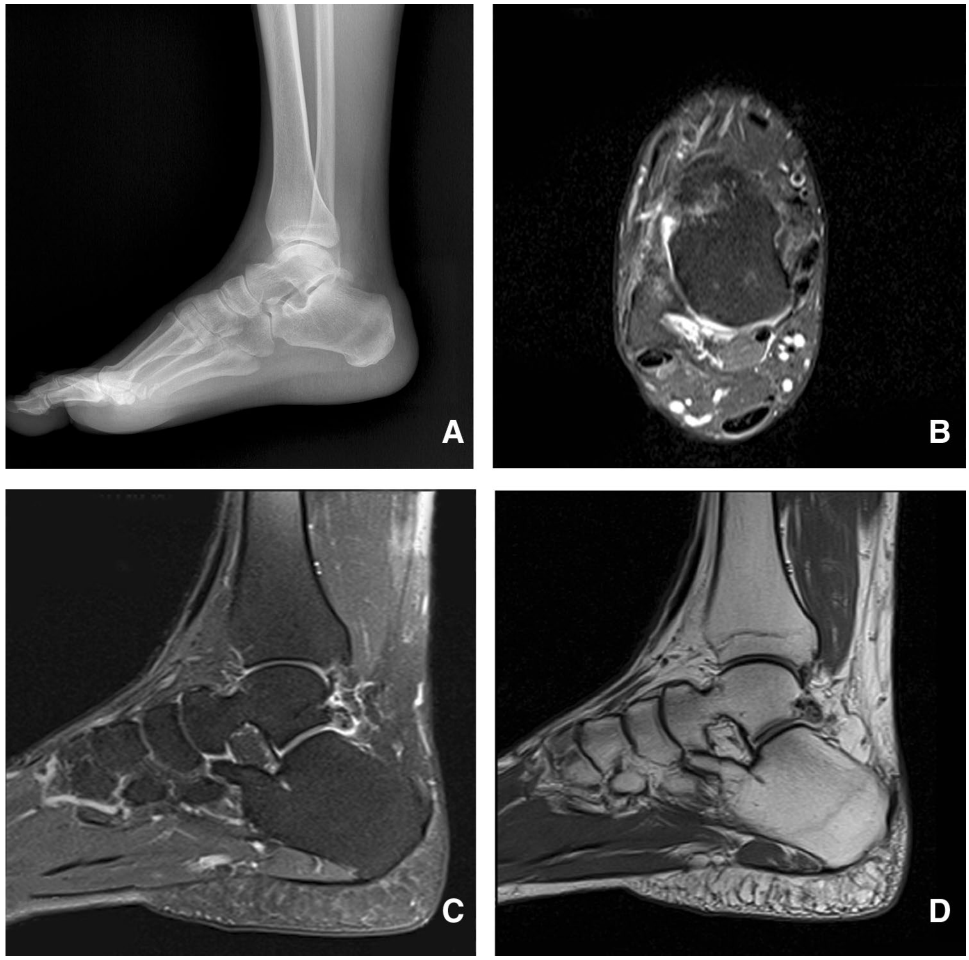

Fig. 2 A 32-year-old male presents with persistent posterior ankle pain 4 months following an ankle sprain. **a** Lateral X-ray of foot showing os trigonum. **b** Axial PD fat-saturated MR image showing fluid around the flexor hallucis longus tendon and around the posterior aspect of talus. **c** Sagittal PD fat-saturated MR image showing os trigonum with edema of the ossicle and the surrounding soft tissue posterior to the talus. **d** Sagittal PD image showing os trigonum with alteration of bone marrow signal intensity of both the ossicle and the posterior aspect of the talus

cases. All patients were able to return to unrestricted work activities within the first 6 months post-surgery.

Discussion

In our study population, an os trigonum was found in 20.4% of patients with ankle sprain. There is wide inconsistency in the literature regarding the incidence of the os trigonum. The lowest incidence reported in the literature was 1.7% [11]. Burman and Lapidus studied 1000 feet X-rays and reported a high occurrence of 50% but only 6% of them were truly separated [2]. In a recent cohort of 1256 ankles, an os trigonum was found in 32.5% of the patients [33]. In addition, a difference in incidence between populations was also reported. Os trigonum was not found in a cohort of Native Americans and Inuits [11]. In a study of eight different ethnicity groups, classified according to the country of birth or the country of birth of the patient's parents, Afro-Caribbean/Surinamese/Central African origin was associated with a lower rate of occurrence of an os trigonum [33]. In our study, the highest incidence of os trigonum in sprained ankle patients was found in the young adult group in both sexes and the lowest incidence of os trigonum was found in the elderly group. In line with our results, an os trigonum was more commonly detected in younger patients using CT imaging [33]. In the same study, an enlarged lateral tubercle of the posterior talar process was found in approximately one-third of the ankles without an os trigonum. Of the total cohort of 798 patients in this study, the enlarged posterior talar process was not present in any case.

Although there is some disagreement about the etiology of os trigonum, i.e., trauma-induced versus a secondary center of ossification, McDougall proposed three different mechanics for its development [12]: first, the persistence of a secondary center as a separate ossicle. A secondary center may occasionally be prevented from uniting with the body of the talus in active young persons. Second, detachment of the ossicle by repeated minor injury. The repeated impingement of the tubercle against the tibia may lead to separation of the tubercle from the body of talus. This mechanism was first noted in a number of football players with prominent posterior tubercles and their symptoms developed gradually. McMurray described its occurrence in football players when the football is kicked with the foot in equines and noted that the strain of the blow is borne largely by the anterior ligament of the ankle. He named the condition 'athlete's' ankle or 'footballer's' ankle [13]. Third, detachment of the ossicle by sudden violence. In this mechanism, the prominent process of talus is subjected to sudden uncontrolled violence and it is fractured with immediate symptoms of pain and tenderness deep to Achilles tendon [12]. It is worth noting that Shepherd's fracture (fracture of the lateral tubercle of posterior process of the talus) is sometimes mistaken for symptomatic os trigonum. Differentiation of a symptomatic os trigonum from Shepherd's fracture is not of clinical significance and is considered an academic exercise because the treatment remains the same [25]. A sharply defined fracture edge may be consistent with acute Shepherd's fracture, whereas the presentation of a smooth, well-rounded ossicle is indicative of an os trigonum [10, 25].

Most studies point to ankle injury as the most common cause of symptomatic os trigonum often affecting ballet dancers and athletes, underreporting the nonathletic population. Fallat et al. examined 639 sprained ankles, only 0.2% suffered from os trigonum syndrome [6]. However, the coincidence of this syndrome with ankle sprains is likely underreported because os trigonum syndrome is unfamiliar to most general physicians [25]. A higher incidence was observed in our study. About 1.1% of acute ankle sprain patients develop an os trigonum syndrome. The most important finding of this study is that the os trigonum syndrome is more common in nonathletic population than previously reported. 5.5% of the os trigonum patients developed an os trigonum syndrome after a standard treatment of an ankle sprain. The overall incidence of os trigonum syndrome was higher in females. An increased rate of os trigonum syndrome has been noted in the population ranging from 18 to 35 years.

Os trigonum syndrome is frequently observed in athletes where the mechanism of injury is either overuse or direct trauma [26]. The incidence of os trigonum syndrome in the athletic population ranges between 1.7 and 50%, with no gender or age group differences in its prevalence [26]. In a study of 19 athletes diagnosed with a post-traumatic overload syndrome of os trigonum, the authors concluded that after chronic athletic activity or an acute ankle sprain the os trigonum may undergo mechanical overload [15]. An acute trauma in plantarflexion may result in contusion, compression, or fracture of the os trigonum, causing an overload post-traumatic syndrome of the os trigonum. The os trigonum becomes painful; however, it appears undisrupted on the lateral X-rays [1, 15].

A recent study examined the association between the injuries to the lateral ligament complex of the ankle and the clinical os trigonum syndrome. A chronic lateral ligament injury increases the likelihood of os trigonum syndrome surgery by ten times compared to those with an acute lateral ligament injury. The study provided new insights into the role of ankle instability in the etiology of os trigonum syndrome in the professional athletes [3]. Lateral ankle instability was previously linked to mechanisms leading to PAIS [17]. Some reports explained the pathophysiology of os trigonum syndrome; for example, in a lateral ankle sprain, the talus can rotate more anteriorly under the tibial plafond, resulting in the ossicle impingement between the posterior edge of distal tibia and the talus [22, 24].

Ankle sprains occur usually through excessive stress on the ligaments of the ankle. The usual mechanism of injury is inversion and adduction of the plantarflexed foot [29]. Considering that plantarflexion of ankle and foot during different postures in prayer reproduced the symptoms in our patients, the repetitive trauma occurring during the physical movement in Muslim prayer following an ankle sprain might cause an overload of the os trigonum. Consequently, an os trigonum becomes painful owing to irritation or impingement of adjacent soft tissues.

Bone scanning was recommended to differentiate symptomatic from asymptomatic os trigonum [10, 31]. Technetium bone scan shows increased uptake in the region of the os trigonum [9, 10]. However, a study of bone scintigrams of soldiers on active duty suggested that increased uptake of Tc-MDP in the os trigonum region is a frequent finding among active soldiers and is of limited value in detecting symptomatic os trigonum [28]. In their study, 27 feet showed an increased uptake of Tc-MDP in the os trigonum region. Only 10 of these 27 feet (37%) had a symptomatic os trigonum.

In conclusion, symptomatic os trigonum should be held accountable for chronic ankle pain in nonathletic population following an ankle sprain unresponsive to standard treatment. A high index of suspicion is necessary for the diagnosis of os trigonum syndrome. Being familiar with the anatomical variations and their possible associated pathology, besides the detailed clinical and radiological assessment, is necessary to diagnose this injury. Our data may help guide effective prevention strategies and inform effective clinical practice.

Author contributions KH: manuscript writing. AO: data collection and management. AM: manuscript writing and analysis design. MY: literature review and data collection. SJ: data collection. AT: data analysis. AD: data analysis. AMH: project development.

Compliance with ethical standards

Conflict of interest The authors declare that the research was conducted without any commercial or financial relationships that could be seen as a potential conflict of interest.

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