Case report

OS TRIGONUM: A CASE REPORT OF A SYMPTOMATIC ANATOMICAL VARIATION AND ITS SURGICAL TREATMENT

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RESUMEN

Los huesecillos accesorios del pie son variaciones anatómicas normales de infrecuente importancia clínica. Se sitúan en los sitios de los centros de osificación secundarios del tarso y metatarso. Desde su primera descripción en la literatura científica en 1804, el os trigonum ha sido un frecuente tema de debate por diversos aspectos, de la etiología al tratamiento. Presentamos un raro caso de un os trigonum doloroso con hallazgos clínicos más evidentes que lo habitual. El huesecillo fue extirpado mediante cirugía abierta con alivio completo de los síntomas. Se discuten brevemente algunas peculiaridades históricas, el diagnóstico, el diagnóstico diferencial, los hallazgos clínicos y las modalidades terapéuticas.

***Palabras clave**: os trigonum; pie; cirugía*

ABSTRACT

Accessory ossicles of the foot are normal anatomical variations of infrequent clinical significance. They are situated at the sites of secondary ossification centers of the tarsus and metatarsus. Since its first description in the scientific literature in 1804, the os trigonum has been a frequent topic of debate in various aspects from etiology to treatment. We present a rare case of a painful os trigonum with richer than usual clinical findings. The ossicle was excised through open surgery with full relief of symptoms. Some historical peculiarities, the diagnosis, differential diagnosis, clinical findings, and therapeutic modalities are briefly discussed.

***Key words**: os trigonum, foot, surgery*

INTRODUCTION

The developing human skeleton has numerous ossification centers that, when fail to fuse with the main body of their corresponding bone, are commonly accepted as normal anatomical variations without definite clinical significance. However, there are different reports in the literature describing various pathological entities due to the presence of additional ossicles. The accessory os trigonum, described as a relatively common complementary ossicle of the foot, is such a normal roentgenographic variant that may cause clinical symptoms (Bellemans et al, 1993; Karasick and Schweitzer, 1996; Georgiev and Stokov, 2010; Köse, 2012).

In this report we present a case of a symptomatic os trigonum and briefly discuss its characteristics from anatomical and clinical point of view.

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CASE REPORT

A 51-year-old woman presented to our institution with a history of posterolateral ankle pain that had been progressively aggravating for two months. There was no history of prior trauma. Pain had been initially exacerbated by ankle plantar flexion and later on by both plantar and dorsal flexion and to a lesser extent by foot inversion and eversion. It was unaffected by passive or active toe motion with or without resistance regardless of foot position. Physical examination revealed pain on palpation behind the fibular tendons and moderate ankle joint effusion. Roentgenograms revealed a prominent os trigonum (Figure 1a). After conservative measures failed, including shoe-wear and activity modification, nonsteroidal anti-inflammatory drugs, and physical therapy, operative treatment was indicated. Surgery was performed through a posterolateral approach. A straight longitudinal incision was used, situated midway between the Achilles tendon and the lateral malleolus. The small saphenous vein and the sural nerve were retracted anteriorly. Dissection was carried out posterior to the fibular tendons through the pre-Achilles (Kager's) fat pad. The mobile os trigonum was reached, enclosed in the posterior ankle joint capsule, and having a strong fibrous connection with the talus anteriorly. The ossicle was dissected free from its soft-tissue attachments and was excised in its entirety. It measured 13×10×5 mm and had partial cartilage-like lining on both its plantar and dorsal surface, the former being visibly larger. (Figure 1b) The wound was closed over a drain in the usual manner. The excision was confirmed on a postoperative roentgenogram (Figure 1c).

Pain was completely relieved after surgery. Weight-bearing as tolerated and range-of-motion exercises were initiated on the second postoperative day. Follow-up after 8 months revealed normal clinical findings and return to full function.

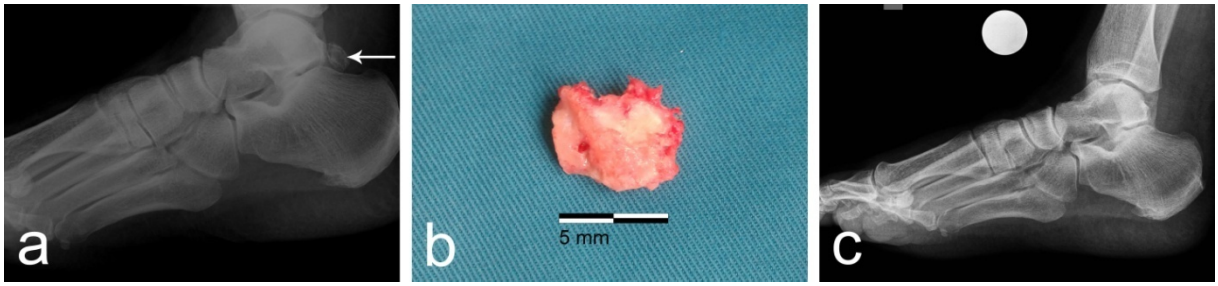

Figure 1 - a: Radiograph showing a prominent os trigonum (arrow); b: Photograph presenting the excised os trigonum (plantar surface); c: Postoperative roentgenogram confirmed the excised os trigonum.

DISCUSSION

A secondary ossification center appears between the ages of 8 and 13 at the posterolateral aspect of the talus. Usually, this ossification center fuses with the talus within one year of its appearance forming its posterolateral process, or Stieda's process, but fail to fuse in 1-25% of people forming a fibrous, fibro-cartilaginous or cartil-aginous articulation with the talus and is then called os trigonum (McDougall, 1955; Grogan et al, 1990; Köse, 2012).

Its first description as a separate ossicle is credited to Rosenmüller in 1804 (Yu et al, 2005). His opinion was supported by Gruber in 1864 and by Stieda in 1869 who assumed it resulted from a secondary ossification center of the talus that failed to fuse with its main body. Nevertheless, Shepherd in 1882 dismissed the idea of a second ossification center and described three cases of what he believed was a fracture of the posterior process of the talus which his name later became eponymous of. Ironically, he failed to reproduce such a fracture on anatomic specimens (Mc Dougall, 1955).

Symptomatic os trigonum does not prevail in either sex or in any age group, but its prevalence is higher in athletes who have to perform forced ankle plantar flexion (Yu et al, 2005; Rathur et al, 2009). The most frequent complaint from os trigonum is pain and tenderness often associated with swelling of the overlying soft tissues behind the ankle joint (Bellemans et al, 1993). Pain is localized behind the lateral, and less often the medial, malleolus and is typically increased by plantar flexion – the nutcracker sign – when the

accessory bone is crushed between the tibia and the calcaneus (Bellemans et al, 1993; Karasick and Schweitzer, 1996; Köse, 2012). We assume that the less frequently described pain on ankle dorsal flexion, as in our case, occurs after disruption of the soft-tissue connection between the os trigonum and the talus and is due to the movement of the ossicle as the joint capsule is being stretched. We associate pain on weight-bearing with movement of the subtalar joint during heel inversion and eversion. Sometimes the os trigonum could be mistaken for a fracture of the posterolateral, or trigonal, process of the talus (Köse, 2012). When os trigonum is suspected as the cause of chronic posterior ankle pain, plain radiographs should be acquired to confirm its presence. On plain radiographs it is usually triangular but may also appear round or oval. It is usually solitary and less than 1 cm in size but may be bipartite or even multipartite. The margins of the ossicle may be smooth or serrated (Karasick and Schweitzer, 1996). In addition, ultrasonography, magnetic resonance imaging, computed tomography, bone scans, SPECT/CT, and arthrogaphy with or without local anesthetic could confirm the diagnosis and differentiate from a fracture or soft-tissue afflictions: tendinitis, osteochondral fractures of the dome of the talus, tibial periostitis, tarsal tunnel syndrome, retrocalcaneal bursitis, etc. (Bellemans et al, 1993; Karasick and Schweitzer, 1996; Yu et al, 2005; Richards et al, 2010; Köse, 2012; Georgiev et al, 2013; Xaviour and Girijamony, 2013; Huang et al, 2014). Initial treatment consists of rest, application of ice, immobilization, non-steroidal anti-inflammatory medications, physical therapy, and local steroid injections. If non-operative treatment fails for 4-6 months, surgical removal of the os trigonum is indicated. It can be performed by a posteromedial, posterolateral, arthroscopic or endoscopic approach (Yu et al, 2005; Ahn et al, 2013).

In conclusion, clinicians should be aware of the presence of additional ossicles and should bear in mind that posterior ankle pain occurring after physical activity with or without trauma should be also examined for a painful os trigonum.

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