Síndrome de *Os Peroneum* Doloroso: Condição Rara ou Não Diagnosticada?

Painful Os Peroneum Syndrome: Rare or Underdiagnosed Condition?

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Resumo

Os Peroneum é um ossículo acessório localizado intra-substância no tendão *peroneus longus*, o qual pode ser responsável pelo aparecimento de dor na face lateral do pé. A síndrome *Os Peroneum* dolorosa resulta de um largo espectro de patologias, tais como *impingement*, fratura ou diástase do *Os Peroneum*, e que pode resultar em tenossinovite ou rotura do tendão *peroneus longus*. O diagnóstico diferencial de dor lateral no pé deve considerar esta síndrome. Embora o diagnóstico se baseie em características essencialmente clínicas, os exames imagiológicos apresentam uma importância acrescida na sua confirmação, assim como na orientação terapêutica. O tratamento descrito na literatura varia de acordo com a gravidade clínica, sendo maioritariamente cirúrgico em virtude da sua apresentação aguda típica - fratura do *Os Peroneum*. Os autores descrevem um caso atípico de apresentação crónica de síndrome *Os Peroneum* Doloroso sem fratura, documentado através de diferentes exames imagiológicos e tratado conservadoramente por meio de um programa de reabilitação individualizado, com resolução sintomática completa.

Palavras-chave: Doenças do Pé; Dor; Pé; Ossos Sesamoides; Síndrome.

Abstract

Os Peroneum is an accessory ossicle located within the substance of the peroneus longus tendon that can cause pain in the lateral aspect of the foot. The painful Os Peroneum syndrome arises from a wide variety of conditions, such as impingement, fractures or diastasis of the Os Peroneum and can lead to tenosynovitis or rupture of the peroneus longus tendon. The differential diagnosis of lateral foot pain should consider this syndrome. Although diagnosis is mainly based in clinical features, imaging plays a significant role both in confirming the diagnosis and in therapeutic orientation. According to the literature, treatment varies with clinical severity, but is mostly surgical due to the syndrome's typical acute presentation – the Os Peroneum fracture. The authors report an atypical case of chronic Os Peroneum syndrome without fracture that was documented by several imaging methods and treated conservatively by means of a customized rehabilitation programme achieving complete symptomatic resolution.

Keywords: *Foot; Foot Diseases; Pain; Sesamoid Bones; Syndrome.*

Introduction

Os Peroneum is an accessory ossicle located within the substance of the *peroneus longus* tendon in the lateral aspect of the cuboid,[1-3] formed by bone and fibrocartilage tissue.[4] Originated from the lateral fibula shaft surface, the *peroneus longus* muscle contributes to the plantarflexion and eversion of the foot and is innervated by the superficial peroneal nerve. The *peroneus longus* tendon is held in place by the superior and inferior peroneal retinacula and the long plantar ligament, as it courses through the posterior aspect of the peroneal malleolus, in the lateral wall of the calcaneus and cuboid gutter, and inserts in the plantar surface of the medial cuneiform and first metatarsal base.[4,5] Radiological and anatomic studies have identified *Os Peroneum* in 5% to 14% of the population,[1] as unilateral in 40% of cases and bipartite in 30%,[3,6] showing round margins in radiological studies in asymptomatic individuals,[7] and being the possible culprit for lateral midfoot and ankle pain.[6] The painful *Os Peroneum* syndrome (POPS), first presented by Sobet *et al*[1]

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results from a wide variety of conditions arising from a common denominator – pain in the lateral aspect of the foot –, from which ossicle fracture or diastasis of multipartite *Os Peroneum* stand out.[1-3,5] This way, and taking into account the predisposing causes, this syndrome may result in tenosynovitis or rupture of the *peroneus longus* tendon.[1,5] POPS must be considered in differential diagnosis of patients experiencing pain in the lateral aspect of the midfoot. Although most of the time clinical presentation and physical examination are enough when it comes to a correct diagnosis, imaging is usually needed both to confirm clinical suspicions and for therapeutic orientation, whether surgical or conservative.[8] This work aims to presenting a case report of a rare nosological entity – chronic POPS – as the cause of lateral midfoot pain non-associated with accessory ossicle fracture or *peroneus longus* tendon rupture.

Case Report

The authors describe the case report of a 32-year-old male, sports teacher, who went to our Physiatry consultation due to persistent, progressively worsening pain in the lateral aspect of his right ankle and midfoot in the last 6 months. The patient claimed no prior direct ankle trauma or sprain and his personal background and family history were irrelevant.

On physical examination there was absence of *cavus* foot or changes on the calcaneus inclination angle as well as absence of oedema or other inflammatory signs. Marked tenderness over the lateral aspect of the cuboid and first metatarsal plantar plate and pain exacerbation in active resisted plantarflexion were observed. The aforementioned complaints were refractory to a non-steroidal anti-inflammatory cycle treatment, reporting only partial relief.

Addicional clinical evaluation was performed. The oblique conventional X-ray of his right foot showed an accessory ossicle, adjacent to the cuboid, peroneal tendon-dependent, oval, with regular bone density and margins, and no evidences of fracture (Fig.1). The oblique contralateral X-ray of his foot showed no accessory ossicles (Fig. 2). The ultrasound studies of the ankle/foot showed the presence of one *Os Peroneum* with intact and regular hyperechogenic surfaces, as well as the thickening and heterogeneity of the *peroneus longus* tendon with signs of

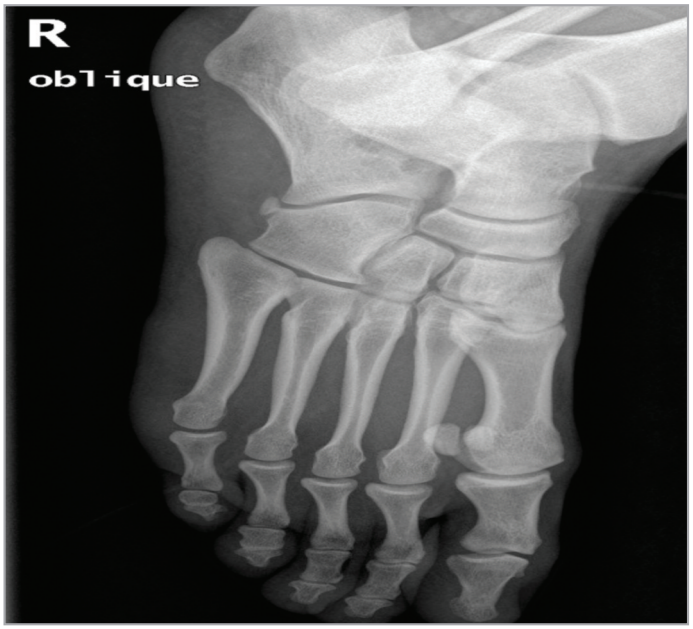

Figure 1 - Oblique view of the right foot showing an *Os Peroneum* with homogeneous density and regular margins at the level of the calcaneocuboid joint.

Figure 2 - Oblique view of the left foot showing the absence of accessory ossicles, adjacent to the cuboid, along the *peroneus longus* tendon.

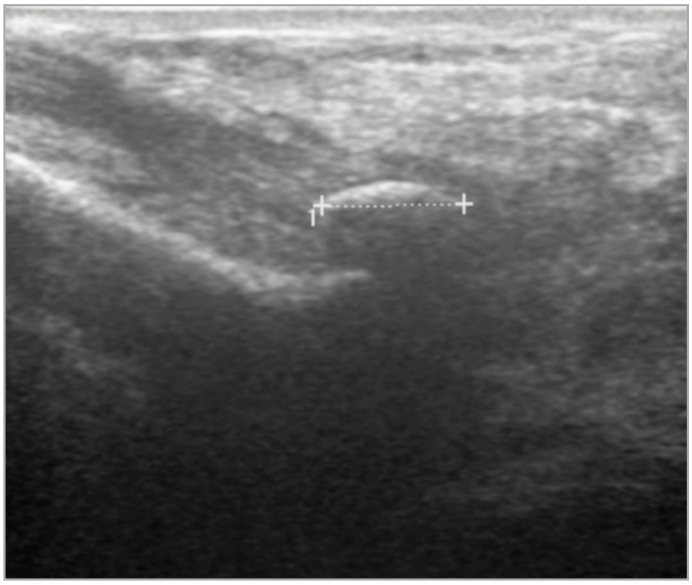

Figure 3 - Long axis view of an ultrasound showing an *Os Peroneum* with intact and regular surfaces and thickening and heterogeneity of the *peroneus longus* tendon with signs of tenosynovitis.

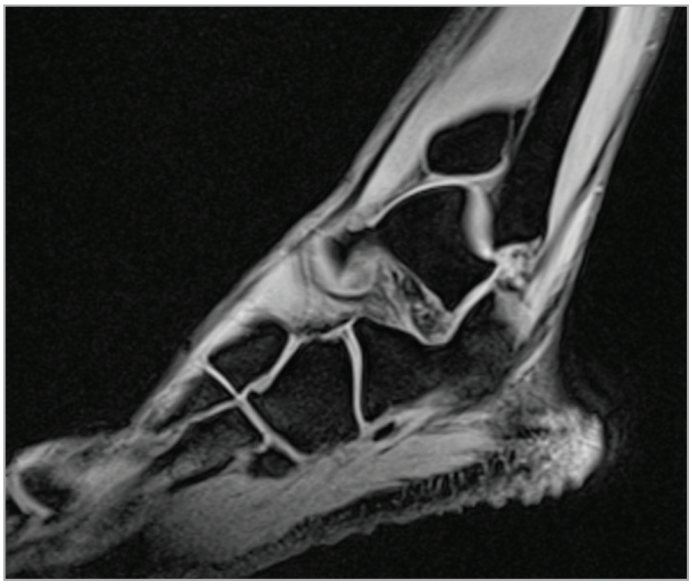

Figure 4 - T2 MRI scan with fat suppression (sagittal) showing an *Os Peroneum*, adjacent to the cuboid within the *peroneus longus* tendon, with inflammatory signs.

tenosynovitis (Fig. 3). The MRI showed one small accessory ossicle with regular margins and homogeneous density, and no cortical discontinuity or fragmentation; it also showed the inflammation of the *peroneus longus* tendon and no signs of tear (Fig. 4).

Clinical and imaging findings were compatible with painful *Os Peroneum* syndrome, as there was one accessory ossicle in the lateral aspect of the cuboid within the *peroneous longus* tendon, associated with signs of tendon sheath inflammation and absence of fracture of *Os Peroneum* or tear of *peroneus longus* tendon. The patient was treated conservatively with another cycle of non-steroidal anti-inflammatory drug (5 days) and physical therapy, including phonophoresis with piroxicam, preliminary analgesic massage and subsequent transverse deep friction massage, muscular dynamic strengthening exercises – concentric progressing to eccentric –, stretching exercises, always respecting patient's complaints regarding pain and proprioceptive training. Complete symptomatic resolution was achieved.

Discussion

Ankle and foot accessory ossicles are normal variants of bone development and are usually the result of failure in the union of the secondary ossification centres, adjacent to the main bone mass.[8] The literature describes several accessory ossicles, the majority of which are asymptomatic, even though they can become symptomatic due to fracture, dislocation, degenerative changes, osteonecrosis, osteoarthrosis, osteochondral injuries, avascular necrosis, tumours and adjacent soft tissue impingement.[9,10] *Os Peroneum* is an accessory round or oval ossicle that can be found within the *peroneus longus* tendon at the calcaneocuboid joint level. In average, this ossicle is 4 mm-thick and 13 mm-long.[8] Its development process is still controversial as there are authors claiming an embryonic development, supported by a recent study that showed an ossicle precursor in the foetal period, while other authors believe that *Os Peroneum* arises from a local response to repeated tendon stress that leads to its thickening and secondary ossification.[11,12] The association of the *peroneus longus* tendon and *Os Peroneum* with the lateral calcaneal aspect and the plantar area of the cuboid, as well as the anchoring of such ossicle to the several adjacent anatomic structures, namely the *peroneus brevis* tendon, the fifth metatarsal, the plantar fascia and the cuboid bone,[13] is described for over 100 years.

There are several causes for pain in the lateral aspect of the midfoot, including dislocation or subluxation of the peroneal tendons, anterior talofibular ligament or calcaneofibular ligament injury, or fractures of the fifth metatarsal or the anterior process of the calcaneus or cuboid.[1,10] The underlying etiopathogenic richness of lateral foot pain represents an additional challenge for POPS diagnosis, because its rarity along with the absence of trauma, especially in chronic situations, may impair early detection. POPS manifests in two main forms: acute and chronic.[1] The acute form arises from trauma, usually ankle supination and/or ankle inversion strain that results in the fracture or diastasis of *Os Peroneum* with high prevalence of tear of the *peroneus longus* tendon.[1,3,10] The chronic form usually mimics the after-effects of an ankle sprain. Complaints can persist for several months or even years until attention is drawn to the problem after an ankle inversion strain and/or worsening of pain in the lateral aspect of the foot. This type of POPS is typically related to the impingement of the *peroneus longus* tendon, diastasis of the *Os Peroneum* or with the consolidation of a fracture of the *Os Peroneum* with remodelling.[1,3,8,11,14] The combination between the anatomic topography of the *peroneus longus* tendon within the lateral aspect of the cuboid and the mechanical stress factors gives rise – as expected – to local friction and subsequent mechanic conflict between both structures,[1,14] enhanced by the practice of sports activities or ankle oversupination.[15] Clinically, this is characterized by an elective pain in the lateral midfoot along *peroneus longus* tendon at the level of the distal portion of the fibula and the cuboid tunnel that can eventually irradiate to proximal areas.[1,14] Dysesthesias may be present in the setting of sural nerve compromise.[16] Also, patients can experience weakness or pain in eversion and a feeling of "stepping on a pebble".[1] Physical examinations show pain caused by palpation of the adjacent area to the cuboid bone, as well as pain in resisted plantarflexion at the level of the first ray and the final stage of support in the gait cycle.[1] Clinical methods not always result in accurate diagnosis for the underlying condition that can vary between tendinosis, tenosynovitis or the tear of the *peroneus longus* tendon. Some authors have suggested that the presence of *Os Peroneum* at the level of the cuboid gutter in association with the tendon's sliding movements results in a predisposition to significantly increase the mechanical stress at this level with consequent functional repercussions.[17,18] Regarding the complementary study, imaging is the pinnacle of *Os Peroneum* diagnosis. This ossicle can be identified by several imaging techniques like the conventional radiography, ultrasound, CAT scans, magnetic resonance imaging (MRI) and skeletal scintigraphy. The conventional radiography is currently the first exam to be made, because it usually allows identifying the accessory ossicle, adjacent to the calcaneofibular joint.[10] As to conventional radiography, this ossicle is best seen in the oblique view, and the proximal migration is usually a sign of distal tear of the *peroneus longus* tendon.[10,18] In the case described above, the patient was experiencing pain in the lateral aspect of the midfoot that lasted for more than 6 months, despite absence of previous trauma. The oblique view of the conventional radiography showed one accessory ossicle, adjacent to the cuboid, with no evidences of fracture. The struggle in the differential diagnosis between the ossicle, an avulsion fracture or tendon calcification is real and challenging for patients complaining of lateral foot pain. There are reported cases of calcific tendinitis of the *peroneus longus* tendon that may as well be potential causes of chronic POPS situations. According with Sobel *et al*, the radiological features of the cases describing *Os Peroneum* fractures are coincident, in their location, size, shape and radiodensity, with the subsequent formation of a bone callus, remodelling and elongation.[1,3] Also, the "calcific tendinitis" of the *peroneus longus* was never proved in histological terms.[1] For the past decades, ultrasound and MRI have become more relevant in the diagnosis of this nosological entity. The ossicle can be easily identified in ultrasounds, due to its histological features – typical curved echogenic focus with posterior acoustic shadow,[10] which can be used for therapeutic orientation, namely interventional procedures like corticosteroid injection in the peroneal tendon sheath and calcaneocuboid joint.[8] In the case presented above, the ultrasound showed a hyperechogenic oval structure with well-defined, smooth margins, associated with a tenosynovitis of the *peroneus longus*, adjacent to the cuboid. Despite there were no consensual ultrasound features leading to the diagnosis of fracture, the presence of a 2 mm-interval between both parts of the ossicle, irregular margins, and pain or discomfort when placing the ultrasound probe, suggest the fracture of the *Os Peroneum.*[15] It is at the level of inflection at the cuboid bone that the *peroneus longus* tendon is more predisposed to injury,[14] due to the significant increase in supination and foot inversion forces. Chronic POPS can lead to the fracture of the *Os Peroneum* and rupture and/or tenosynovitis of the *peroneus longus* tendon, due to the persistent compression forces. In this case, the MRI showed one accessory ossicle with regular margins and no signs of fracture, as well as signs that are compatible with a tenosynovitis of the *peroneus longus* tendon. This imaging method showed an ossicle signal similar to the signal of the adjacent bone, increasing within the substance of the *peroneus longus* tendon.[10]

Several therapeutic options have been postulated for approaching this pathology, wherefore injury chronicity and patient activity level are key factors for the clinical decision. The conservative treatment includes non-steroidal anti-inflammatories, cast immobilisation (*Os Peroneum* fracture*)*, orthoses (weight-bearing and/or alignment of calcaneal plantar axis), corticosteroid injection in the *peritendineum,*[1,3,14] and customised rehabilitation

programmes. Surgical treatment is usually reserved for acute cases and thoses that cannot be handled by conservative means, showing good results in regard to function and mobility for all cases described.[20] Literature describes several surgical approaches like debridement, repair of *peroneus longus* tendon-related changes and tenosynovectomy, excision of the accessory ossicle and possible fragments, and resection of the hypertrophic peroneal tubercle.[1,3,14]

In conclusion, the authors have described a rare case of painful *Os Peroneum* syndrome with tenosynovitis of the *peroneus longus* tendon that showed no signs of accessory ossicle fracture, presenting with chronic pain in the lateral aspect of the midfoot.

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