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Sever Hastalığı, Os Trigonum Sendromu ve Pes Planus Birlikteliği: Ayak Ağrısının Bermuda Şeytan Üçgeni

Coexistence of Sever's Disease, Os Trigonum Syndrome, and Pes Planus: The Bermuda Triangle of Foot Pain

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To the Editor,

Sever's disease (SVD), also known as calcaneal apophysitis, was first described by James Warren Sever in 1912 and is recognized as the most common cause of heel pain in children and adolescents (1). It is a clinical condition characterized by an insidious onset of pain, typically unrelated to trauma, but rather resulting from repetitive use, particularly in children participating in sports such as football and basketball. Sever's disease, which is frequently seen between the ages of 7-15, peaks between the ages of 10-12 (1). Histologically, it presents as inflammation or bone edema in the secondary growth center of the calcaneus, induced by repetitive traction forces exerted by the triceps surae muscle (2).

Posterior ankle impingement syndrome (PAIS) refers to a spectrum of conditions causing posterior ankle pain during plantar flexion, commonly observed in athletes. It can result from bony abnormalities, such as the Stieda process, os trigonum, and osteophytes, or soft tissue pathologies and anatomical variations (3). Os trigonum is the most common accessory ossicle in the ankle and is the most common cause of PAIS (2). Secondary ossification of the posterolateral aspect of the talus occurs between the ages of 8 and 15, typically fusing with the talus within one year. However, in 7% of cases, the os trigonum may not fuse with the talus (3). In addition to this defect, recurrent acute plantar hyperflexion and chronic microtrauma often result in a clinical presentation associated with flexor hallucis longus (FHL) tenosynovitis (2, 3).

Flatfoot (pes planus) is commonly observed in infants and children, typically resolving by adolescence, and is regarded as physiologic, being generally flexible, painless, and without functional impairment. However, in rare instances, it may become painful or rigid, suggesting the presence of underlying foot pathology, such as arthritis or tarsal coalition, although a universally accepted definition for pediatric flatfoot is still lacking (4). Symptomatic patients may present with complaints such as activity-related pain and rapid muscle fatigue. In some cases, pain is reported not only in the sole of the foot but also in the ankle. These symptoms may result from the lack of support provided by the natural arch and the reduced contact surface between the insoles and the ground (4).

All three conditions are common causes of foot pain that typically respond to conservative treatment, yet they can individually lead to significant issues in children and adolescents. In this article, we present a complex case in which three clinical conditions coexist, resulting in pain primarily in the ankle and heel regions.

A 12-year-old boy presented to the physical medicine and rehabilitation (PMR) outpatient clinic with bilateral heel pain, which was more pronounced on the right side. He reported experiencing pain in the back of his heel and ankle after playing football or prolonged walking, which persisted even with rest. His medical history was unremarkable, with no history of trauma or significant disease. Physical examination revealed pain with movement and tenderness on palpation in the bilateral ankles, Achilles tendon and heel, more severe on the right. Additionally, there was an appearance compatible with flatfoot on examination and walking test. Radiographic evaluation revealed sclerosis and fragmentation of the calcaneal apophysis, a possible os trigonum, and a possible arch drop suggestive of flatfoot (although not a weight-bearing examination) (Fig. 1). Ultrasonography (US) identified cortical irregularities in the calcaneus and findings consistent with Achilles tendinopathy (Fig.1).Although radiological and clinical evaluations suggested SVD and os trigonum syndrome (OTS), magnetic resonance imaging (MRI) was requested for further evaluation. MRI revealed edema in the calcaneal bone, confirming the diagnosis of SVD, as well as edema in the soft tissue surrounding the os trigonum, supporting the diagnosis of os trigonum syndrome. Additionally, MRI showed significant narrowing

of the calcaneal angle, suggesting a diagnosis of pes planus, while the detection of FHL tendinitis supported the diagnosis of OTS and PAIS (Fig. 1).

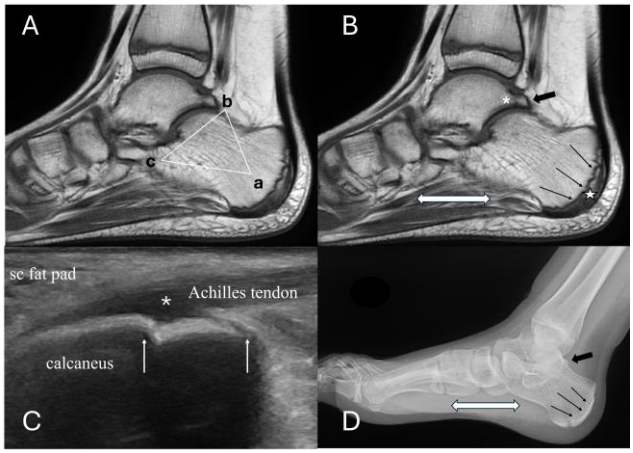

Figure 1 (Imaging of the right foot): Representation of three distinct diagnoses on MRI, reminiscent of the "Bermuda Triangle":

a) Sever's disease; **b)** Os trigonum syndrome; **c)** Pes planus

(A) MRI sagittal T1 image showing edematous areas (white asterisk) around the junction of the os trigonum (thick black arrow) and the talus; irregularities along the junction of the calcaneal tuberosity and calcaneal apophysis (thin black arrows); and an edematous area at the calcaneus (white star). Narrowing of the calcaneal angle consistent with flatfoot (double-headed white arrow)

(B) Longitudinal ultrasound imaging shows cortical irregularities in the calcaneus (white thin arrows) and thickening-edema in the Achilles tendon (white asterisk)

(**C)** Lateral radiograph shows os trigonum (thick black arrow), sclerosis and fragmentations of the calcaneal apophysis (thin black arrows), arch droop indicating flatfoot (double-headed white arrow)

(D) Sc; subcutaneous

The patient was prescribed nonsteroidal anti-inflammatory drugs, customized insoles with arch support and shock-absorbing properties, and home exercises. It is hypothesized that the specially designed insole may provide long-term benefits in the correction of flexible deformities, particularly pes planus. Additionally, the use of shock-absorbing materials has been recommended for the management of SVD to mitigate mechanical stress on the calcaneal apophysis. The prescribed home exercise program includes stretching exercises aimed at enhancing the elasticity of structures such as ligaments and tendons, as well as strengthening exercises designed to support the normal arch and intrinsic foot muscles, all within the patient's tolerance limits. For pain and inflammation management, NSAIDs have been prescribed for a defined period (regular use for 7–10 days, followed by as-needed administration for pain episodes). Furthermore, to prevent symptom exacerbation and structural deterioration in SVD, high-impact sports activities—including jumping, sprinting, running, and sports involving these movements, such as football and basketball—have been restricted.

Three important diagnoses to consider in the etiology of foot pain in adolescents are SVD, OTS, and pes planus (5). Although these three conditions can be detected radiographically, they do not have diagnostic value unless clinical signs are present and objective tissue damage is demonstrated on MRI (1, 3-5). For this reason, when multiple diagnoses are suspected, an MRI should be requested following radiographic evaluation to provide a comprehensive assessment of the foot. Additionally, due to its accessibility, ultrasonography can serve as a valuable tool for evaluating tendon, ligament, and muscle structures. In cases of heel pain, SVD should be considered; for posterior ankle pain, OTS; and for pain in the arch, pes planus should be suspected (1-5). However, to the authors' knowledge, there have been no reported cases in the literature where all three diagnoses coexist. Therefore, our report has two aspects. First, while rare, multiple diagnoses can coexist in adolescent foot pain, making management challenging. Second, a comprehensive assessment by specialists, primarily PMR physicians, may be necessary for effective diagnosis and treatment.

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