

CASE REPORT

Unilateral Painful Os Trigonum Syndrome in an Indian Housewife: A Rare Case Report

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ABSTRACT

The os trigonum is a tiny bone found on the posterolateral aspect of the talus. It has a separate ossification center which does not unite with the talus itself. It is difficult to make the diagnosis, as clinical features and physical examination findings mimic those that occur with conditions related to flexor hallucis longus, an accessory soleus muscle, peroneal tendons, tibialis posterior, or arthritis of the posterior subtalar or tibiotalar joints. Misdiagnosis may lead to inadequate management which includes early weight bearing and unrelieved symptoms. This syndrome is found mostly in ballet dancers, runners, and soccer players.

Keywords: Ankle pain, Os trigonum, Talus.

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INTRODUCTION

Several ossification centers are present in the developing human skeletal which do not fuse with the body of the corresponding bone. These are commonly considered as normal anatomical variations. The os trigonum is a tiny bone found on the posterolateral aspect of the talus. It has a separate ossification center which does not unite with the talus itself.^{1,2}

Usual age of appearance is 8 and 11 years, as a secondary center of ossification and fusion with the talus takes place within 1 year of its appearance.^{3,4} When it fails to fuse with talus and remains separate, it is referred to as the os trigonum. When a large posterolateral process is formed after the ossification center fuses with talus, it is known as a "fused os trigonum," a "trigonal process," or a "Stieda's process."^{5,6}

According to reports, the incidence of the os trigonum is 3 to 15%^{2,7,8} and bilateral os trigonum is more common than unilateral.³ Pain and sometimes swelling in the posterolateral aspect of the ankle are the characteristic features of the os trigonum syndrome. McDougal had described the two possible mechanisms of injury, either an episode of acute forced hyperplantar flexion or microtrauma from repetitive hyperplantar flexion.⁹ This syndrome is found mostly in ballet dancers, runners, and soccer players.

Clinical examination findings can raise the possibility of os trigonum syndrome, but the diagnosis is confirmed by radiographs showing the os trigonum and magnetic resonance imaging (MRI). Computed tomography (CT) and bone scans may be used to rule out other bony and soft-tissue involvement.^{10,11} Initial treatment is conservative. It includes rest, use of ice, and anti-inflammatory medications, cortisone injections, and physical therapy. When conservative care has failed to relieve the symptoms, surgical excision is recommended.

CASE REPORT

A 65-year-old housewife presented with right-sided ankle joint pain since 9 months. The pain was abrupt in onset while she was picking up a jar from the overhead rack standing on her toes. It was progressive in nature as she was unable to walk the next day and also noticed swelling of right ankle for which she was prescribed oral analgesics and activity limitation for 2 weeks. After 2 weeks, the pain subsided but as patient resumed her activities of daily living, there was a recurrence of pain in the same region which aggravated on movement.

So, an x-ray was done which showed an undisplaced fracture of right talus. She was treated conservatively with a crepe bandage for 3 months. During this time, the patient was walking with full weight bearing and no rehabilitation program was initiated. The pain was moderate in intensity, mechanical in nature, and used to get relieved on taking oral analgesics. When the patient tried to resume her complete preinjury activities of daily living, she again noticed similar symptoms in the ankle. At the time when she presented to our outpatient department, she was taking oral analgesics and was using crepe bandage for her right ankle. She was walking with full weight bearing.

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CLINICAL FINDINGS

Nine months after the initial trauma and the onset of symptoms, she was evaluated at our outpatient department. She presented with right ankle pain on posterolateral aspect which aggravated on walking. Pain was dull and aching in nature and was located in the posterolateral part of the right ankle. It was acute on onset, which aggravated on walking and relieved on taking rest and medication. There was no history of morning stiffness, nocturnal attacks, other joint involvement, or trauma. Severity of the mechanical pain, measured in visual analog scale, was 8/10. On physical examination, the patient was normotensive. Active and passive range of motion of talocrural, subtalar, and midtarsal joint was within normal range. Muscle strength testing of ankle did not reveal any muscle weakness. Tenderness of grade II was present over posterolateral region of right ankle. Sudden and forced plantar flexion was extremely painful. Considering the failure of previous modalities of therapy and the inconsistent nature

of ankle pain which increased on plantar flexion, she was advised an x-ray of both ankle joint in anteroposterior and lateral views. Blood tests for complete blood count (CBC), fasting blood sugar (FBS), thyroid-stimulating factor (TSH), liver function test (LFT), serum uric acid, calcium, 25(OH)D (vitamin D) were also advised.

X-ray of right ankle showed a fused os trigonum or a "Stieda's process" (Fig. 1). X-ray of left ankle was apparently normal (Fig. 2). An MRI was done to rule out other causes of posterolateral ankle joint pain. The MRI of right ankle showed fluid around tibialis posterior, flexor hallucis longus, flexor digitorum tendons, which was suggestive of peritendinitis (Fig. 3). It revealed the presence of a well-corticated bony/osseous structure behind the right talus, which was suggestive of Stieda's process (Fig. 4).

Parameters like CBC, FBS, LFT, TSH, uric acid, and calcium all came out as normal. But, serum 25(OH)D was at a deficiency level (7 ng/mL).



Fig. 1: X-ray of right ankle



Fig. 2: X-ray of left ankle

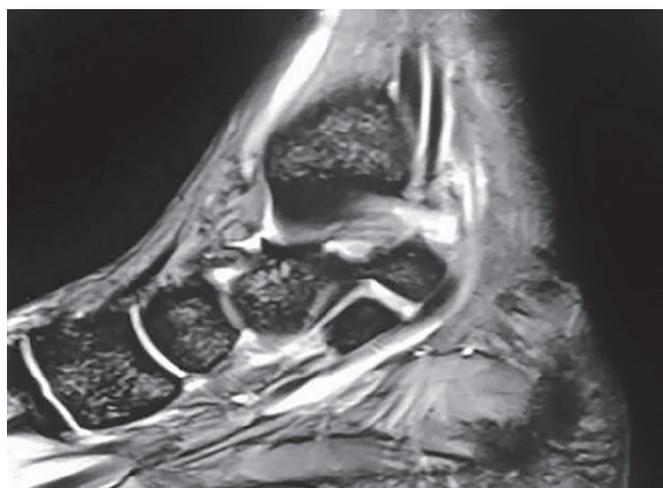


Fig. 3: Magnetic resonance imaging of right ankle



Fig. 4: Magnetic resonance imaging of right ankle: Stieda's process

TREATMENT

Surgical option for os trigonum syndrome was discussed with the patient and was referred to orthopedics surgeon. She refused to get any surgical intervention. The patient was treated at our department with a below knee plaster cast in neutral position of ankle. Along with that analgesic medication and Vit D₃ supplementation were prescribed. After immobilization for 3 weeks, the cast was removed and active range of motion of ankle joint along with weight bearing and physical therapy for range of motion, functional rehabilitation, intrinsic foot muscle stretching and strengthening were prescribed. Use of open counter shoes with slight heel raise and soft sole was ensured. The patient was educated about the lifestyle modification to avoid trauma, hyperplantar flexion, and barefoot walking.

After 3 weeks of rehabilitation, there was no tenderness over the posterolateral aspect of the ankle. Visual analog scale score for pain intensity was reduced to 1/10. The patient was followed up for 2 months. She had no complaints of pain or swelling on ambulation and had resumed her complete activities of daily living comfortably.

DISCUSSION

The os trigonum is one of the most common accessory bones located in the foot and is present posterolateral to the talus bone.⁶ Most of the times it is asymptomatic; however, rarely it may be associated with persistent posterior ankle pain which is called os trigonum syndrome.¹² Os trigonum syndrome may be commonly misdiagnosed as a fracture of posterior process of talus bone.^{3,4,6}

The mechanism of injury is the excessive or hyperplantar flexion of the ankle which compresses the os trigonum between the posterior malleolus of the tibia and the tuber calcaneus.¹³ Common clinical features include pain elicited on forced plantar flexion of the ankle, i.e., nutcracker sign, which was positive in our case. In our reported case, hyperplantar flexion of ankle was the cause of injury.

The diagnostic modality of choice or gold standard to diagnose for os trigonum as well as posterior talus structure fractures is CT scan.^{4,6} In some case reports, technetium bone scan has been shown to be helpful in diagnosing symptomatic os trigonum as well as united posterior process fracture by demonstrating increased uptake in the region of os trigonum.^{2,14} However, it is costlier and not freely available. Magnetic resource imaging can also identify a disrupted cartilaginous synchondrosis by demonstrating the presence of fluid between the os trigonum and the lateral talar process. Associated

problems like flexor hallucis longus tenosynovitis, which is mostly stenosing, can also be assessed. Degenerative joint changes about the synchondrosis or between the os trigonum and the adjacent calcaneus can also be seen.

Injuries due to excessive or hyperplantar flexion ankle may result in fractures in the posterior ankle compartment, but these are often missed and mistreated as simple ankle sprains.¹⁴ Fracture of the os trigonum is an uncommon and extremely rare entity; therefore, a high index of suspicion is important to diagnose fracture of os trigonum due to severe plantar flexion of the ankle. Literature shows that conservative treatment is generally successful, although recalcitrant cases like nonunion have also been reported.^{3,12}

Though in this case os trigonum fracture was not present, pain was due to tendinitis and painful os trigonum syndrome. As per evidence, painful os trigonum may be a possible source of recalcitrant posterolateral ankle joint pain. The index of suspicion should be high when the patient's symptoms are reproduced with passive plantar flexion of the foot or palpation of the posterior aspect of the subtalar joint between the Achilles tendon and the lateral malleolus. As demonstrated by this case, the painful os trigonum can be effectively managed by conservative method and appropriate physical therapy.

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