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Case Report
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Atypical Presentation of Tibial Nerve Entrapment Coexisting with Flexible Flat Foot

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ABSTRACT

Introduction: tarsal tunnel syndrome presents with typical pain and paresthesia around the ankle and medial aspect of the foot. Flexible flatfoot results from posterior tibial tendon insufficiency can also present with above symptoms. The deformity in the early stages of flatfoot, may be overlooked due to its mild nature. Here we present a case of 45-year-old lady, presented with symptoms of tarsal tunnel syndrome associated with flexible flat foot. We highlight the importance of the good clinical acumen to gauge the tarsal tunnel syndrome coexisting with flexible flatfoot by the appropriate investigations and custom made management. It is essential to consider the co-existing deformities in the foot, when a patient comes with the tarsal tunnel syndrome like presentation and vice versa. It is also important to know the aetiology of each component along with the clinical assessment and appropriate investigations, before planning a management protocol. A custom made approach may be the answer, considering the patient's clinical, occupational, social conditions and radiological observations.

Keywords: Adult acquired flatfoot deformity; Posterior tibial tendon dysfunction; Tarsal tunnel syndrome; Flexible flat foot; Pes planus.

Introduction

Ever since the "tarsal tunnel syndrome" was termed by 'Keck' and 'Lam' for the first time, in the early sixties of 19th century, the disease has evolved in leaps and bounces in terms of its aetiology, clinical presentation and association with other conditions of foot and ankle. Tarsal tunnel syndrome by definition is 'the entrapment of posterior tibial nerve in the tarsal tunnel, causing discomfort in the form of pain and paresthesia around the ankle and foot'[1,2]. Varicose veins and space occupying lesions like ganglionic cyst are among the specific causes for the tibial nerve compression in the tarsal tunnel, which leads to compressive neuropathy. It is also not uncommon that the deformities in the hind foot, like varus and valgus heel can contribute to the incidence of compressive neuropathy with the incidence of 11% and 8% respectively[3]. When there are more than one cause exists in an individual overlapping the signs and symptoms, it is a challenge to the clinician to diagnose and plan the definitive treatment which is aimed at relieving the symptoms. Here we present a case of female patient aged about 45 years, presented with a flexible flat foot deformity on the right side associated with the overlapping symptoms of tarsal tunnel syndrome. The very purpose of this case study is to highlight the challenges in the clinical diagnosis, planning the appropriate investigations and also to reaffirm the concepts in the management of the case, when there are two conditions coexist in the same foot.

Case Report

A 45-year-old lady, home maker, presented to outpatient department with pain and swelling around the right ankle and foot for the period of 3 weeks. She was apparently normal 3 weeks back and gradually started having the dull aching pain, which was more around the medial aspect of ankle and radiating along the medial aspect of foot. Pain used to get aggravated, specifically on standing in the kitchen for long hours and it was getting relieved by taking rest. On clinical examination, there was "too many toes" sign present on the affected side with the hind foot valgus deformity compared to the normal contralateral side. Swelling was noted around the medial aspect of ankle just posterior to the medial malleolus. There was collapse of the medial arch height, which is a tell-tale evidence for the flat foot. The flatfoot was of flexible variety, and was confirmed by jack's test and double heel raise test. On palpation, there was tenderness along the tendon of tibialis posterior from a point just behind the medial malleolus till its insertion in the navicular bone. The 'single leg heel raise' test was painful while performing on right side, indicating the posterior tibial dysfunction. Hence a clinical diagnosis of flexible adult acquired flatfoot deformity (AAFD), secondary to the posterior tibial tendon dysfunction (PTTD) in the form of tendinopathy was made. Further corroboration was done by taking the appropriate views of plain radiographs. Supporting the above findings, standard weight bearing radiographs showed altered Meary's

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angle (Fig. 1A) and calcaneal pitch angle (Fig. 1B). These x-ray findings confirmed the diagnosis of flexible AAFD. An ultrasonography of right ankle and foot confirmed the posterior tibial tendinopathy without tear. The visual analogue scale (VAS) score and AOFAS ankle hind foot scale were recorded.

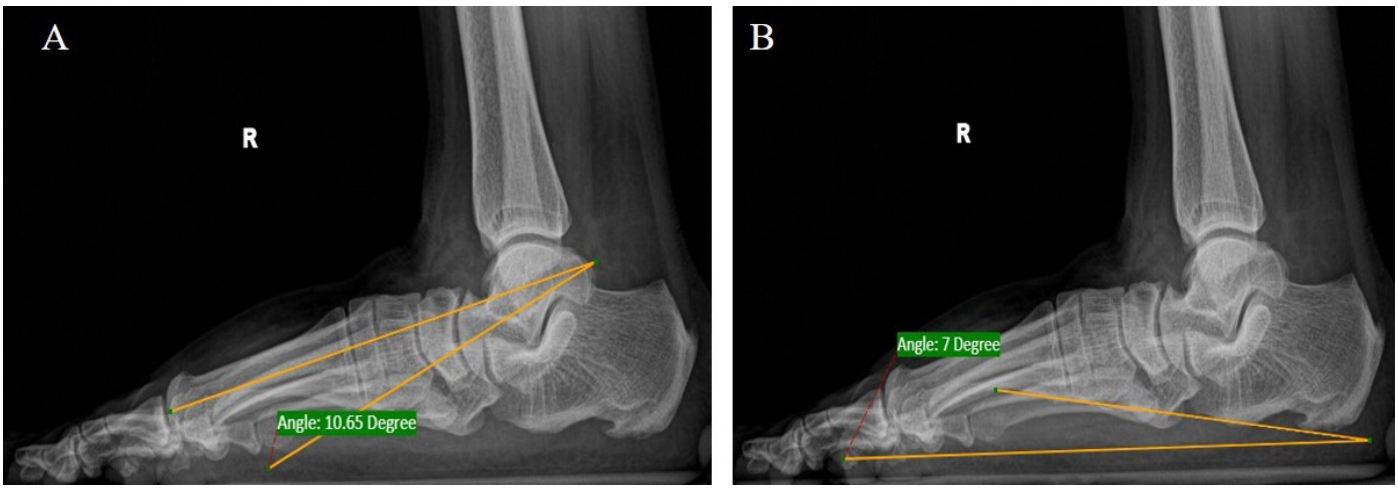

Figure 1. Radiographs showing the adult acquired flat foot deformity. A. Meary's angle is disturbed (10.65º, normal range 0-4º). B. Calcaneal pitch angle is decreased (7º, normal range 20-40º), indicating the collapse of the medial longitudinal arch.

Initially, conservative treatment was planned with anti-inflammatory drugs, rest and an UCBL orthosis (University of California biomechanics laboratory orthosis) for outdoor walking and footwear modification (medial arch support) for indoor walking. Patient was instructed for a review in 3 weeks. But the patient visited within a week, due to aggravation of symptoms in the form of pain, which was burning in nature, radiating along the inner aspect of the foot. The pain was more at rest and was disturbing the sleep. On examination, 'Tinel sign' was positive along the course of the posterior tibial nerve. The 'stress manoeuvre' was done by forced dorsiflexion and eversion, which generated pain along the course of tibial nerve and its branches in the foot. This time a clinical diagnosis of tarsal tunnel syndrome was made provisionally with the existing flexible flat foot with posterior tibial tendon dysfunction. To confirm the clinical diagnosis, a magnetic resonance imaging (MRI) was taken, which showed, 'a ganglionic cyst' in the tarsal tunnel between the posterior tibial nerve and artery, abutting the nerve along its medial aspect (Fig. 2A). MRI also showed posterior tibial tendinopathy with the partial tear of spring ligament (Fig. 2B).

The nerve conduction velocity (NCV) study revealed, the right sided posterior tibial nerve neuropathy, involving both the motor and sensory components, confirming the diagnosis of right sided tarsal tunnel syndrome. The aetiology was being the ganglionic cyst compressing the nerve. There existed a dilemma about treatment strategy, since both the conditions existed together. After discussing with the patient a common consensus was formed regarding treating the tarsal tunnel syndrome with surgical decompression and flexible AAFD without surgical reconstruction.

After inducing the spinal anaesthesia, the patient was positioned in supine position. A vertical curved incision measuring about 8 cm was given posterior to medial malleolus, at the midpoint between medial malleolus and Achilles tendon. The tarsal tunnel was

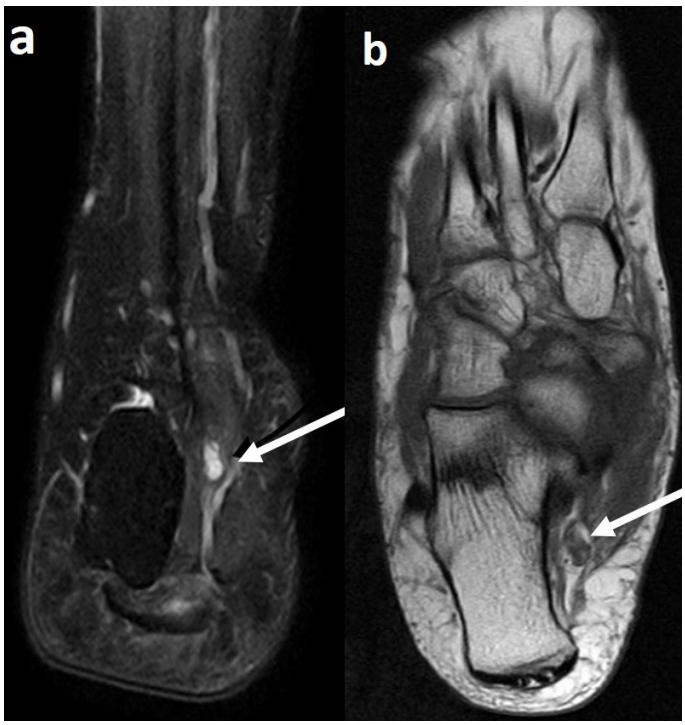

Figure 2. 2A. MRI films showing the 'ganglionic cyst' in the tarsal tunnel (↓) between the neurovascular bundle, abutting the nerve along its medial aspect; 2B. showing the tibialis posterior tendinopathy (↓).

approached in layers by blunt dissection to avoid the iatrogenic injuries to the superficial branches of the nerve. The flexor retinaculum is identified at its proximal end and a Mac Donald blunt ended dissector was passed underneath the retinaculum, protecting the vessels and nerves. The retinaculum was released to expose the posterior tibial tendon. The signs of synovitis were seen around the posterior tibial

tendon, and was debrided. On retraction of the tendon medially the ganglionic cyst was exposed (Fig. 3). The decompression procedure was performed, as the cyst was causing compression to the nerve. The nerve was identified through its course and the adhesions were released. The tarsal tunnel release was completed by decompressing the fascia distally over the abductor hallucis muscle belly. The muscle belly was retracted plantar aspect and the deep fascia was identified and released. There were no features of fibrosis around the posterior tibial nerve or its branches. The reconfirmation of decompression procedure of flexor retinaculum was done along its entire course, before the closure of the wound.

Figure 3. Ganglion cyst (↓) in the tarsal tunnel was exposed by retracting the tendon of tibialis posterior, medially.

Patient was put on a back slab for a period of 12 days, to take care of the surgical wound. The surgical wound sutures were removed at 12th post-operative day. Following which, the patient's leg was kept in a below knee CAM walker for encouraging the range of movements at the ankle and foot. This was accompanied with global strengthening of the muscles around the ankle and foot and strengthening the intrinsic muscles of foot. Patient was kept on partial weight bearing in a CAM walker for 12 weeks and following which, patient was given footwear with medial arch support and trained for resistance exercises for posterior tibial tendon, peronei and Achilles tendon.

Patient was regularly followed up at 3 weeks, 6 weeks, 3 months, 6 months, 1 year, 18 months and 2 years and once a year. The last follow up being at the end of 3rd year. At each visit, patient was evaluated with the VAS score for symptoms of pain. Patient was clinically examined for scar tenderness, presence of Tinel's sign and single leg heel raise. The AOFAS ankle hind foot scale score was also recorded at each visit. Patient had a drastic improvement in the pain, post operatively by 3 weeks. At 6 months, the VAS score came to 'zero' and the AOFAS ankle hind foot score was improved to 95 from 66 with minimum disability and occasional fatigue pain. Patient had complete relief of pain with no functional disability at the end of 3 years at the latest follow up.

Discussion

The tarsal tunnel syndrome is not an uncommon entity. The patients usually present with symptoms of pain and paresthesia, which spread diffusely around the medial aspect of the ankle. This pain radiates along the medial border of the foot up to the plantar aspect, involving the sole[4]. These symptoms may also exist in a patient with tibialis posterior tendinopathy. Tibialis posterior tendinopathy is one of the commonest causes of AAFD. In our patient, the symptoms were overlapping and causing ambiguity in the clinical diagnosis. When there is a co-existing condition, like flat foot associated with the tibialis posterior tendinopathy, this may lead to inflammation along the medial border of ankle and foot. Since tarsal tunnel syndrome can mimic the signs and symptoms of tibialis posterior tendinopathy, whether they are interrelated or not, it will be a challenge to diagnose clinically. PTTD is common in females and in age group between 35 to 70 years. Cimino[5] observed that, tarsal tunnel syndrome has slighter female gender predilection, which accounted for 56% and the most common cause included were trauma, space occupying lesion and deformities of foot. In the foot deformities, heel varus accounted for 11% and heel valgus accounted for 8%[4].

In our study, the patient is a lady in her forties with a flat foot deformity along with the heel valgus. She had difficulty and pain while standing alone on the affected leg, making the single leg heel raise test difficult, which is the pathognomonic of tibialis posterior dysfunction. This was confirmed by the MRI finding. There was also a space occupying lesion in the form of a ganglionic cyst, which was detected by the MRI. It was causing compression of the posterior tibial nerve, explaining the combined etiology of tibialis posterior dysfunction and tarsal tunnel syndrome. However, question arises whether the tarsal tunnel syndrome was contributed by the heel valgus or the ganglionic cyst. Hence we suggest that, whenever there is a coexisting condition, like in our present case, all etiological factors to be considered, while planning and executing the management protocol.

Relevant investigation becomes key for the definitive diagnosis. In our present case, there were two sets of investigations. One set to confirm the diagnosis of

flexible pes planus and the second set to confirm the tarsal tunnel syndrome. The standard weight bearing radiographs were taken to confirm the flat foot (pes planus). Important parameters like Meary's angle and calcaneal pitch angle were considered. The Meary's angle was reversed and the calcaneal pitch angle was abnormally decreased, confirming the diagnosis. The ultrasonography and MRI confirmed the tibialis posterior tendinopathy. The partial tear of spring ligament, further corroborated the etiology of the AAFD. The presence of a ganglionic cyst compressing the posterior tibial nerve at the tarsal tunnel and the electro diagnostic study revealing the abnormal sensory and motor conduction velocities confirmed the posterior tibial neuropathy. Hence after appropriate clinico-radiological and electro diagnostic study, a final diagnosis of flexible AAFD with tibialis posterior tendinopathy and tarsal tunnel syndrome, secondary to the ganglion cyst were made.

Few studies[5-7] reported that, in tarsal tunnel syndrome, electrodiagnostic studies are helpful in up to 90% in the presence of supportive history and physical examination. Frey and Kerr[8], in their study of tarsal tunnel syndrome observed that, 88% of their patients had a lesion, which was identified by MRI. It was concluded that the MRI is the most effective modality to assess the contents of the tarsal tunnel[8-10]. During the surgical planning, MRI becomes the most important diagnostic modality to localise the lesion causing the compression over the nerve and also to determine the extent and location in relation to the tibial nerve. This is supported by our study, where the MRI clearly visualized the contents of tarsal tunnel, being compressed by the ganglionic cyst, developing the tarsal tunnel syndrome. MRI also showed the posterior tibial tendinopathy and partial tear of spring ligament contributing in the symptoms of flatfoot. Wilemon[10] stated that the radiological investigations, including the weight bearing x-rays are useful in detecting the deformity of foot. In our study, the weight bearing radiographs were taken, which confirmed the presence of flat foot. The ganglion cyst, which was compressing the neurovascular bundle with the motor and sensory neuropathy of the tibial nerve, distal to the tarsal tunnel, contributed for development of tarsal tunnel syndrome. Hence there is a dilemma of co-existing etiology regarding the development of symptoms in the right foot.

It becomes crucial to plan a treatment protocol, considering the dual etiology for the presenting symptoms in the same lower limb. Non-steroidal anti-inflammatory drugs and corticosteroid local injections are the accepted non operative management documented in many studies for the symptomatic tibialis posterior tendinopathy[3,11]. But it is very important to avoid direct injection to the tendon substance to prevent its rupture. Whenever there is a co-existing flexible heel valgus with tarsal tunnel syndrome, non-operative treatment like orthosis either in the form of medial arch support or rigid orthosis like, UCBL heel cup is recommended[3, 5-6, 12]. In our patient, at her first visit with signs and symptoms of AAFD due to posterior tibial tendinopathy, she was advised a medial arch support as a footwear modification, along with rest and anti-inflammatory drugs. After a period of three weeks, patient was reassessed for recommendation of exercise therapy, which is a routine protocol in the treatment of AAFD due to posterior tibial tendinopathy. Since the patient visited within 10 days, with aggravation of symptoms, the reassessment was done. Appropriate investigations revealed, existence of tarsal tunnel syndrome along with the posterior tibial tendinopathy. The aggravation of symptoms, after the use of medial arch support in cases of tarsal tunnel syndrome is also mentioned by Radin[13]. In a cadaveric study by Lau and Daniels[14], the pes planus was created in the lab and it was found that, surgical release of the tarsal tunnel will lead to increase in the tibial nerve tension, which may worsen the AAFD, and lead to failure of tarsal tunnel release. This indirectly states that, tarsal tunnel decompression is contra-indicated, when there is co-existence of flexible pes planus. A study by Feiffer and Cracchiolo[15] suggested that, unless there is a space occupying lesion like ganglion cyst, which is compressing the nerve, the surgical decompression should not be executed. In 24 months follow up, they observed that only 44% of their cases had beneficial effect by surgical decompression. In our case, aetiology of the tarsal tunnel syndrome was a space occupying lesion, which was obvious in the MRI, but there was also a pes planus deformity. This might have also contributed to the increased tension over the nerve. As per the opinion of Lau *et al.*[14], question arises about the management protocol to decompress the ganglion cyst and release of tarsal tunnel in isolation, against adding the reconstruction of the heel valgus and medial arch collapse as a surgical remedy for pes planus. The management of adult acquired flat foot is based on the stage at which it is present[16]. In our case, the AAFD is of flexible variety and eligible to be grouped as stage 2A, according to Johnson and Strom classification modified by Deland[17]. Accordingly, non-operative management with orthosis and exercises are recommended as treatment of choice. The surgical options are discussed only if the non-operative measures fail after 3 months to 6 months' span. The dilemma of treatment in our case is to go ahead either with an aggressive reconstructive approach, for AAFD, with medial displacement osteotomy of the calcaneum and transfer of flexor digitorum into the navicular bone combined with the decompression of the ganglion cyst and release of the tarsal tunnel. The alternative approach is to go ahead with surgical decompression of the tarsal tunnel and excision of the ganglionic cyst, and the AAFD is managed with

footwear modification and braces followed by exercise therapy for ankle and foot.

In this present case, the counselling of patient was done with discussion about the options available. It was agreed to go ahead with the surgical excision of ganglion cyst with decompression of the tarsal tunnel and opt for a conservative approach for the flexible AAFD. As a result, 36 months follow up showed no recurrence or aggravation of symptoms. And the flexible AAFD did not progress.

Conclusion

It is very important to consider the co-existing deformities in the foot, when a patient comes with tarsal tunnel syndrome like presentation and vice versa. It is also equally important to know the aetiology of each component with the clinical assessment and appropriate investigations, before planning a treatment protocol. A custom made approach may be the answer, considering the patient's clinical condition and radiological observation with that of the patient's occupational and social conditions.

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Ethical Approval

Institutional ethical committee clearance taken: IEC 507/2020.

Informed Consent

Written informed consent taken for the scientific publication. The images are consented to be published in scientific articles from the patient.

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