CASE REPORT

Bilateral tarsal tunnel syndrome attributed to bilateral fibrous tarsal coalition and symmetrical hypertrophy of the sustentaculum tali

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SUMMARY

Tarsal tunnel syndrome (TTS) is one of the frequently encountered entrapment neuropathies of the lower limb. Most often the aetiology is considered to be idiopathic. However, it has to be thoroughly investigated to rule out diverse extrinsic and intrinsic causes. The sustentaculum tali (ST) forms the superior part of the distal tarsal tunnel, and any increase in its size may result in irritation of the tibial nerve or its branches, the medial and lateral plantar nerves or decrease the overall volume of the tarsal tunnel culminating to clinical features of TTS. A hypertrophied ST is an extremely rare cause for TTS, and we report perhaps a first case of bilateral TTS in a young adult due to bilateral symmetrical hypertrophied ST which in turn was a result of bilateral fibrous tarsal coalition. Staged bilateral surgical excision of the hypertrophied part of ST and the underlying fibrous tarsal coalition ensured prompt symptomatic relief.

Morton's metatarsalgia, compartment syndrome of the deep flexor compartment, calcaneal spur, arthrosis, inflammatory changes of the fasciae and ligaments and ischaemia are some of the important differential diagnoses to be considered.¹

We present a first case of bilateral TTS in association with bilateral fibrous tarsal coalition and symmetrical hypertrophied sustentaculum tali (ST). In the index case, the diagnosis of TTS was suspected based on history and clinical examination, and the aetiology was identified as bilateral symmetrical hypertrophied ST based on MRI, CT scan and differential blockade. Until now, there has been only one report of a unilateral TTS as a result of hypertrophied ST.² According to the authors' knowledge, this is the first report of bilateral presentation of TTS attributed to bilateral symmetrical hypertrophied ST which, in turn, was the result of bilateral fibrous tarsal coalition.

BACKGROUND

Pain along the medial border and plantar aspect of the foot is one of the most common symptoms we come across in foot and ankle clinic. Tarsal tunnel syndrome (TTS) is one of the culprits, which is frequently misdiagnosed. Arriving at the diagnosis can be challenging as the clinical course is often protracted and the signs non-specific and ambiguous. A thorough history, clinical examination aided with radiological imaging techniques and nerve conduction studies are frequently warranted. There are numerous causes attributed as aetiology; however, most often the case remains idiopathic. Polyneuropathy, L5 and S1 nerve root syndromes,

CASE PRESENTATION

A 22-year-old student presented to our foot and ankle clinic with vague pain and numbness in his feet of 6 years duration. The pain and numbness was exacerbated on prolonged standing and walking and relieved on taking rest. This was associated with bilateral symmetrical swellings over the medial border of the foot. He noticed the swellings 6 years back which was insidious in onset and non-progressive. Prior to visiting us, he had undergone conservative management in the form of analgesics, silicone footwear supports and also below knee plaster cast. However, his symptoms persisted. There was no history of any trauma prior to the beginning of these symptoms.

On local examination, there were two bilateral symmetrical swellings, approximately 2×2 cm each, located approximately 1.5 cm below the medial malleolus and on the medial border of the foot (figure 1). Skin over the swelling appeared normal. There was mild bilateral hind foot valgus which was evident on weight-bearing. Foot arches were maintained. On palpation, the swellings were bony hard in consistency and non-tender. Tinel sign was positive bilaterally. Ankle and subtalar joint movements were unaffected.

INVESTIGATIONS

Plain radiographs (axial view of the heels) revealed bilateral symmetrical hypertrophied ST and

Figure 1 Clinical picture of the bilateral symmetrical hypertrophied sustentaculum tali. MM, medial malleolus.

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corresponding beaking of the talus (figure 2). CT scans revealed the presence of bilateral tarsal coalition (figure 3). MRI further revealed bilateral fibrous coalition of the middle (medial) talocalcaneal facet with hypertrophy and mild degenerative changes and hypertrophic ST with no other compressive pathology within the tarsal tunnel (figure 4A,B). There was neither any signal changes within the nerve nor any thickening. The nerve conduction studies were normal.

A diagnostic blockade using 2% lignocaine in the tarsal tunnel on both sides caused transient relief of his symptoms strongly pointed towards the diagnosis of TTS.

TREATMENT

Staged (right side first) surgical exploration and excision was contemplated for the ease of physiotherapy and mobilisation of the patient. Under the spinal anaesthesia and tourniquet control, the patient was placed in supine position. A 10 cm retromalleolar curvilinear incision was made over the right ankle. The tarsal tunnel was explored. The hypertrophied ST had changed the course of the tibialis posterior tendon making it cross above the ST. There was no other abnormality observed in the anatomy or content of the tarsal tunnel. The tibial nerve was found to be normal. The posterior part of the hypertrophied ST was excised, the tibialis posterior tendon was transposed back into the tarsal tunnel and the posterior tibial nerve was decompressed throughout its course (figure 5). The fibrous tarsal coalition was excised, and a local fat pad graft was interposed. A below knee cast was maintained for 1 month. Corresponding intraoperative findings were observed on the left side as well.

OUTCOME AND FOLLOW-UP

There was significant improvement in the patient's symptoms. Two months later, the patient underwent similar surgical excision on the left side. At 14 months of follow-up, his American Orthopaedic Foot and Ankle Society hind foot scores bilaterally were 93.

DISCUSSION

TTS can be a frustrating condition for both patient and his clinician. In our experience, patients generally bear with the constant or nagging foot pain, only to seek a specialist after prolonged suffering. For clinician, it is at times very difficult to narrow down the exact aetiology of the TTS owing to the complex anatomy of the tarsal tunnel besides diverse foot and ankle conditions with similar clinical presentation. Hence, a thorough history taking, focused clinical examination, imaging and sometimes nerve conduction studies are considered essential for its diagnosis.

Figure 2 Plain radiographs axial view showing bilateral hypertrophy of the sustentaculum tali.

Figure 3 CT scan axial view further demonstrating the talocalcaneal coalition.

Clinical examination in the index case revealed several mixed interesting facts. There was a clinical dilemma in isolating the cause for patient's symptoms. Patient's age, bilateral presentation, vague chronic pain over the medial border of the foot, mild hind foot valgus deformity and MRI features were all in favour of tarsal coalition as the culprit. However, the vague pain associated with the numbness and paraesthesia in the distribution of tibial nerve branches was suggestive of TTS. On differential blocking, there was no improvement in symptoms when the local block was given at the site of the tarsal coalition, whereas the patient had substantial relief of symptoms when the local block was given within the region of the tarsal tunnel. Nerve conduction study is helpful in supporting the diagnosis in 80% of cases.³ However, in the index case, it was normal.

Tarsal coalition is an abnormal connection between the tarsal bones, which is thought to result from a congenital failure of differentiation in the developing foetal foot.⁴ It is bilateral 50% to 80% of the time.⁴ The coalition can be bony, cartilaginous or fibrous types.⁵ Clinical features markedly vary ranging from no symptoms to severe pes planovalgus deformity. The pain in this condition is frequently pointed to an area inferior to the medial malleolus, at the site of coalition. Plain radiographs, especially axial view of the foot, can identify the bony coalition. CT is useful to define the bony variety, and MRI is useful to detect the fibrous type besides outlining the relationship of the nerve with the lesion. Management of the condition persists to be ambiguous and a matter of debate. At first, a trial of conservative methods is usually employed. Surgical options range from

Figure 4 (A) MRI right side (photon density weighted, sagittal view) the tibial nerve (red arrow) kinking at the site of hypertrophied sustentaculum tali. (B) MRI right side coronal sections (proton density and T2 weighted), broadening and irregularity of the posterior sustentaculum tali and adjacent medial process of talus with an intervening hypodense area suggestive of fibrous tarsal coalition.

Figure 5 Intraoperative (right side); red arrow pointing at the tibial nerve (retracted with a silicone surgical loop); arrow head at hypertrophied sustentaculum tali.

resection of the bone bridge with or without interposition grafts to arthrodesis.

Lee *et al* reported a similar case of TTS caused by tarsal coalition, although unilateral. Ultrasonography and MRI showed that the medial plantar nerve was impinged at the coalition, and excision ensued significant clinical improvement.⁶ There was focal swelling of the medial plantar nerve at the site of impingement probably secondary to chronic irritation. Despite chronicity in the index case, the tibial nerve was found to be of normal calibre throughout the course bilaterally. It has been observed that failure to recognise the tarsal coalition and isolated decompression of the tibial nerve has resulted in persistence of symptoms.⁷

Although ST lies in close proximity to the tibial nerve course, attributes of ST as a cause of TTS are extremely unusual. A case of isolated fracture of the ST with formation of a non-union and subsequent migration and impingement of the tibial nerve has been reported.⁸ There is only one case of unilateral hypertrophied ST causing TTS documented in the literature.² Unlike

in these cases, besides bilateral hypertrophied ST, the associated bilateral fibrous tarsal coalition is unique in our patient.

MRI had ruled out other possible intrinsic and extrinsic factors for TTS. The fibrous tarsal coalition may have been responsible for the gradual hypertrophy of the ST which in turn led to the development of TTS. The surgical excision of the posterior part of the hypertrophied ST, decompression of the tarsal tunnel and excision of the coalition with fat graft interposition resulted in favourable outcomes bilaterally.

Learning points

- ▶ Bilateral tarsal tunnel syndrome (TTS) is a very unusual foot and ankle condition.
- ▶ Thorough evaluation is warranted to rule out an exhaustive list of extrinsic and intrinsic causes.
- ▶ Bilateral tarsal coalition in association with bilateral symmetrical hypertrophy of the sustentaculum tali which in turn is responsible for bilateral TTS has not been previously reported.
- ▶ Excision of both tarsal coalition and hypertrophied sustentaculum tali ensured prompt symptomatic relief in this case.

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