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Case Reports and Series

Tarsal Tunnel Syndrome Due To Three Different Types of Ganglion During a 12-Year Period: A Case Report

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ABSTRACT

A 52-year-old male complained of numbness and radiating pain affecting the plantar region of his left foot. He was found to have recurrent tarsal tunnel syndrome due to posterior tibial nerve compression by 3 different types of ganglion during a 12-year period. To the best of our knowledge, a similar case has not been documented. At the first operation, flexor retinaculum release and simple excision of an epineural ganglion were performed without injuring the nerve fascicles; however, an intrafascicular ganglion developed approximately 2 years later. At the second operation, the ganglion cyst was resected completely to prevent recurrence, despite the risk of nerve fiber injury. The cyst originated from the subtalar joint; thus, the joint was closed, and a free fat graft was placed to prevent adhesion formation. However, an extraneural ganglion occurred about 3 years later. At the third operation, the cyst was resected completely, and a free periosteal graft was used to close the joint more effectively. No recurrence had developed at 6 years after the third operation. The findings of the present case show the need for long-term monitoring of patients with tarsal tunnel syndrome caused by a ganglion owing to the possibility of recurrence related to different ganglion types.

Tarsal tunnel syndrome is an entrapment neuropathy due to compression of the posterior tibial nerve in the tunnel formed by the flexor retinaculum behind and below the medial malleolus of the ankle. Although the etiology of tarsal tunnel syndrome varies, Takakura et al (1) reported that a ganglion was the most frequent cause. Nagaoka and Satou (2) reviewed 30 patients with tarsal tunnel syndrome caused by a ganglion. A rare case of tarsal tunnel syndrome caused by an intraneural ganglion of the posterior tibial nerve was recently reported (3–8). Just as in our patient, tarsal tunnel syndrome was initially caused by an epineural ganglion of the posterior tibial nerve. However, no previous cases have been reported of recurrent ganglion compressing the posterior tibial nerve in which an epineural ganglion, an intrafascicular ganglion, and an extraneural ganglion developed successively at the same site during a 12-year period.

Although magnetic resonance imaging (MRI) is useful for diagnosing different types of ganglion, the present case suggests that surgeons must also pay close attention to the relationship between the ganglion and its adjacent joint. If a connection is identified, it is necessary to completely separate the nerve from the joint and effectively close the communication with sufficiently strong tissue to prevent recurrence of the ganglion, especially if the lesion is in the foot and ankle region.

Case Report

On April 3, 2004, a previously healthy 52-year-old male presented to our hospital complaining of increasing numbness of the plantar region of his left foot of 3 months' duration, along with radiating pain that affected the great toe, second toe, and third toe. His job was to install air conditioners. He had no history of trauma. On examination, although no muscle atrophy or motor dysfunction of the left foot was present, a swelling was found behind the medial malleolus of the left ankle. Neurologic examination detected a positive Tinel's sign at the level of the swelling, and electroneuromyography of the lower extremities revealed left tarsal tunnel syndrome. MRI was performed to investigate the swelling. It revealed a single cyst inside the tarsal tunnel with uniform low signal intensity on T1-weighted images and high signal intensity on T2-weighted images (Figs. 1*A* and 2*A*). Based on these findings, tarsal tunnel syndrome due to a ganglion compressing the posterior tibial nerve was diagnosed.

On May 7, 2004, surgery was performed with the patient under general anesthesia. A zigzag incision was made superiorly,

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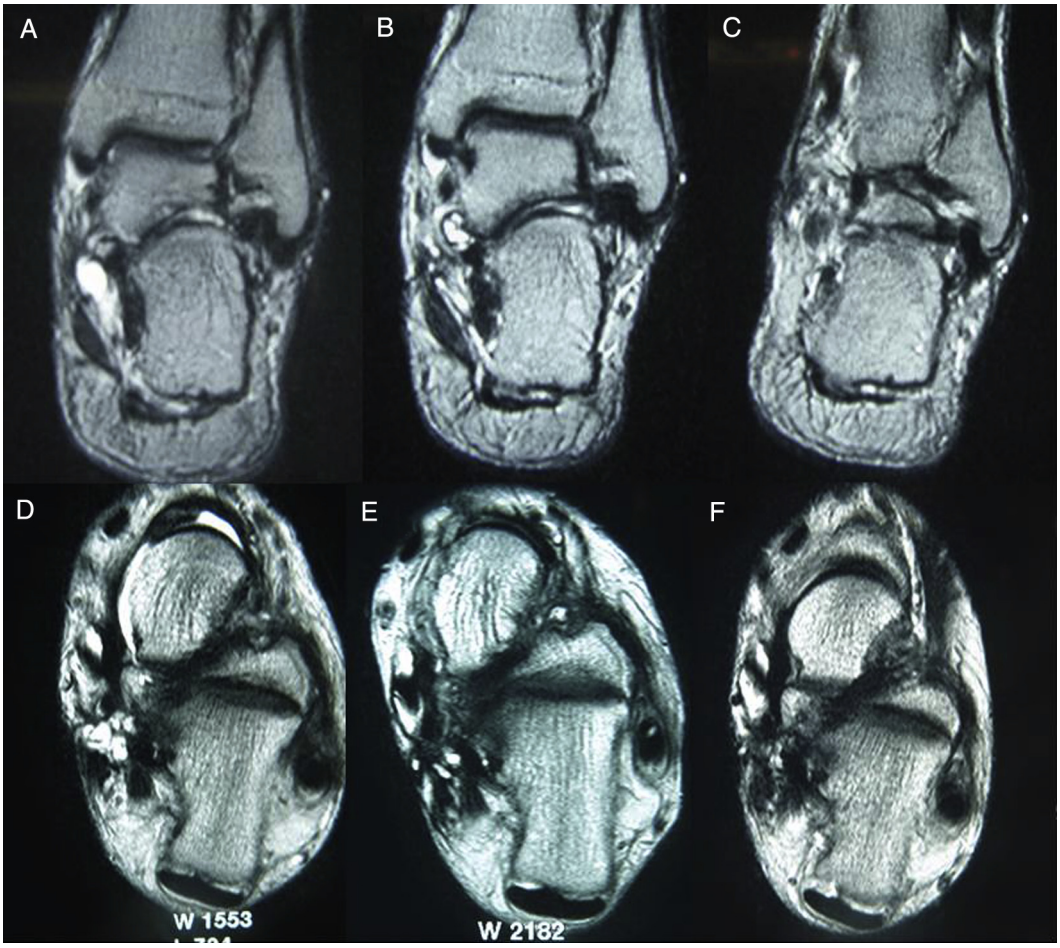

Fig. 1. T2-weighted coronal magnetic resonance images of the left ankle on (*A*) April 3, 2004, (*B*) October 10, 2006, (*C*) June 26, 2009, (*D*) November 17, 2009, (*E*) August 5, 2015, and (*F*) June 7, 2016.

posteriorly, and inferiorly to the medial malleolus. The flexor retinaculum was completely transected, and the abductor hallucis longus muscle was partially cut, after which the proximal border of the cystic mass was identified distal to the origin of the calcaneal branch of the posterior tibial nerve (Fig. 3*A*). Although the MRI findings seemed to show a pedicle connecting the mass to the subtalar joint, the relationship between the mass and the joint capsule or tendon sheath was unclear. The wall of the cystic mass in the epineurium was sharply excised (Fig. 3*B*). The fascicles of the posterior tibial nerve had been compressed by the lesion, but it clearly had not invaded the nerve. We diagnosed a ganglion cyst from the macroscopic features of the lesion and detection of clear colorless mucinous material on cyst puncture.

The patient had only slight residual numbness of the plantar region of the great toe postoperatively, and he did not attend outpatient follow-up examinations after April 3, 2005. However, he presented again on April 11, 2006, complaining of numbness of the left plantar region. Conservative therapy was tried for several months; however, radiating pain occurred and swelling was found behind the medial malleolus of his left ankle. MRI was performed because the symptoms seemed likely to be due to a recurrent ganglion compressing the posterior tibial nerve. MRI revealed a mass located within the posterior tibial nerve that showed a uniform low intensity on T1-weighted images and contained multiple, small, high intensity areas on T2-weighted images (Figs. 1*B* and 2*B*). These findings indicated that the posterior tibial nerve was being compressed by a lesion invading through the fascicles, which meant that the patient had an intrafascicular ganglion rather than an epineural ganglion as previously.

On November 9, 2006, surgery was performed with the patient under general anesthesia. An incision was made above the previous surgical scar. Beneath the posterior tibial artery near the site of scarring, enlargement of the posterior tibial nerve was identified proximal to the origin of its calcaneal branch. The lesion had invaded the fascicles below the epineurium of the posterior tibial nerve. The expanded epineurium was incised, and clear colorless mucinous material was drained from the cyst. Proximal and distal epineurotomy around the lesion was combined with neurolysis of the calcaneal branch, medial plantar nerve, and lateral plantar nerve for complete decompression (Fig. 4*A*). The subtalar joint was exposed under the posterior tibial nerve (Fig. 4*B*), and the joint was closed by suturing the surrounding connective tissue over it to prevent recurrence of the ganglion (Fig. 4*C*). Next, a free fat graft was placed to prevent adhesion of the posterior tibial nerve with neurolysis (Fig. 4*D*). We did not perform en bloc resection; however, the macroscopic findings, histologic findings (Fig. 5*A*), and drainage of clear colorless mucinous material from the lesion were all compatible with a ganglion cyst.

The patient only noted slight impairment of perception in the left medial plantar region postoperatively, and MRI showed no mass lesion. He did not return for follow-up examinations after June 26, 2009 (Figs. 1*C* and 2*C*). However, he presented again on November 12, 2009, complaining of numbness of 2 weeks' duration affecting both the plantar region and the great toe and second toe of his left foot.