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## Okayama Case Report

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# Tarsal Tunnel Syndrome Caused by a Schwannoma of the Posterior Tibial Nerve

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Schwannoma is the most common tumor of the peripheral nerve sheath. However, there have been few reports on schwannoma of the posterior tibial nerve causing tarsal tunnel syndrome. We report on a 60-year-old man with tarsal tunnel syndrome caused by a schwannoma of the posterior tibial nerve, which was first diagnosed as a ganglion cyst. After enucleation of this tumor, the patient was asymptomatic and had no related sequelae except for minor numbness in the plantar aspects of his digits. Although schwannoma of the posterior tibial nerve is rare, it should be considered even if a ganglion is clinically suspected.

Key words: tarsal tunnel syndrome, schwannoma, posterior tibial nerve

chwannoma is the most common tumor of the peripheral nerve sheath. It arises from Schwann cells and is rarely malignant [1]. Schwannoma typically occurs in patients aged 20 to 50 years and has no sex preference. Although it can occur anywhere in the body, it is found less frequently in the lower extremities [2] and rarely in the posterior tibial nerve [3,4]. We report here on a patient with tarsal tunnel syndrome caused by a schwannoma of the posterior tibial nerve.

## **Case Presentation**

A 58-year-old man initially presented to our hospital with a small mass on the posteromedial aspect of his left ankle. X-ray of the left ankle revealed no hind foot deformity such as post-traumatic arthritic spur or tarsal coalition. At that time, we diagnosed a ganglion arising from the subtalar joint. However, we could not aspirate any fluid and therefore left the mass untreated. The mass gradually grew and became more tender during 2

years. At 60, he presented again and complained of severe pain particularly when wearing shoes or walking. On clinical examination, the cystic mass was 3 cm in diameter and was soft, palpable, and fixed. He complained of pain and numbness radiating to the hallux with direct compression around the mass. However, no Tinel-like sign was elicited by finger percussion over the mass. Neurological testing revealed no obvious loss of sensation over the medial sole and no motor weakness.

Magnetic resonance imaging (MRI) of the left ankle revealed a mass with an eggshell-like appearance located distal to the medial malleolus. The mass appeared as a 22-mm region of homogeneous low intensity in T1weighted images and heterogeneous high intensity in T2-weighted images (Fig. 1). The lesion was located close to the posterior tibial nerve. The differential diagnoses included a ganglion and a benign tumor such as neurofibroma or a peripheral nerve sheath tumor.

Because the patient's pain was aggravated while wearing shoes, he elected to undergo surgery. Informed consent was obtained because the diagnosis of a ganglion was most likely and there was a possibility of a benign tumor. Surgical decompression by the enucleation of the mass under lumbar anesthesia was performed. A slightly curved incision, approximately 5 cm in length, was made along the medial ankle. A soft, ovoid, white-yellow mass was found within the tarsal tunnel. The mass was solid and seemed to be not filled with any fluid, and it continued to the posterior tibial nerve sheath. Exact enucleation was difficult, and the mass was bluntly dissected as an intralesional resection; however, the nerve sheath was spared using vascular loops (Fig. 2). Although the surface of the nerve sheath was identified and remained intact (Fig. 3).

The patient was able to walk and was discharged

from the hospital the day after surgery. At his first postoperative visit, he presented with mild numbness of the medial plantar nerve. No motor deficits were observed. On sectioning, the mass was encapsulated and had a white-yellow surface. Microscopic examination revealed an encapsulated lesion composed mainly of spindle cell areas with palisading. It contained a mixture of hypercellular Antoni A-type tissue containing spindle cells arranged in fascicles with characteristic nuclear palisading and hyalinized Antoni B-type areas with ectatic vessels (Fig. 4). Immunohistochemical analysis showed strong and diffuse positivity with an antibody against S-100 protein (Fig. 5). Microscopic findings suggested a schwannoma. After enucleation of this tumor, the patient was asymptomatic and had no related sequelae

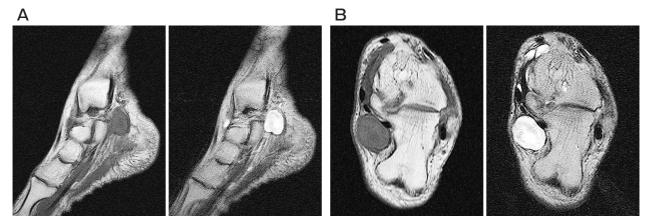


Fig. 1 A, Sagittal MRI images; B, Axial MRI images. A T1-weighted image (left) shows a homogeneous low-intensity eggshell-like mass located within the tarsal tunnel. A T2-weighted image (right) shows the lesion as heterogeneous and high intensity.

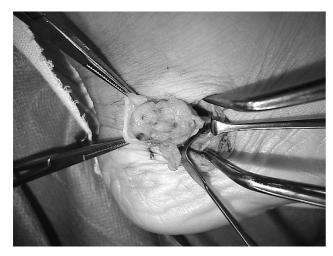


Fig. 2 A soft white-yellow mass is seen within the tarsal tunnel.

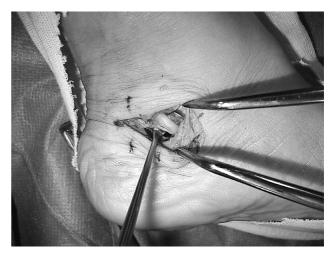


Fig. 3 The posterior tibial nerve was left intact.



Fig. 4 Histopathological sections of the schwannoma of the posterior tibial nerve reveal Antoni A-type and Antoni B-type tissues.

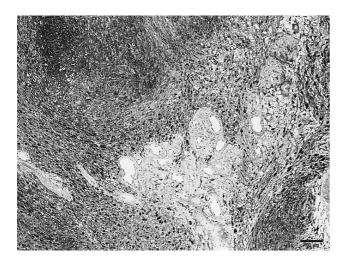


Fig. 5 Strong and diffuse positivity is seen with an antibody against the S-100 protein.

except for minor numbness in the plantar aspects of his digits at the 6-month follow-up. There was no evidence of recurrence on follow-up MRI at that time.

### Discussion

Space-occupying lesions may cause intrinsic compression of the posterior tibial nerve in the tarsal tunnel. Common causes include ganglion, post-traumatic arthritic spur, hind foot deformity, tarsal coalition, varicosity, lipoma, and schwannoma. Schwannoma is a benign tumor arising from the Schwann cells of a

peripheral nerve sheath [5]. The foot is affected in only 10% of all cases [6], and the diagnosis of schwannoma in a limb may be delayed. In a previous study, only 3 of 25 patients with schwannoma of the posterior tibial nerve were diagnosed within a year of onset, and the longest documented delay was 30 years [7]. Schwannoma of the posterior tibial nerve as a cause of secondary tarsal tunnel syndrome has been reported rarely in the literature [8-11].

To form a clinical diagnosis of tarsal tunnel syndrome, a typical history must be obtained and a physical examination must include positive provocative testing, such as that for a Tinel-like sign or for dorsiflexioneversion. These tests may vary in sensitivity and specificity depending on the degree of compression (disease stage) of the distal tibial nerve [12]. However, with mild compression, a patient could be negative for a Tine-like sign. When the sensory disturbance becomes persistent, the majority of these patients are expected to have a positive Tinel-like sign [13]. A systematic review concluded that the results of nerve conduction studies were abnormal in some patients suspected of having tarsal tunnel syndrome [12]. Schwannoma manifests as a cosmetic deformity or a palpable mass and/or with symptoms similar to those of a compressive neuropathy; however, neurologic symptoms tend to appear late [9]. The characteristic target sign on MRI T2-weighted images is present in 52% of cases, and it consists of a hyperintense rim and hypointense core, which correspond histologically to peripheral myxomatous tissue and central fibrocollagenous tissue, respectively [14].

Surgical enucleation of the mass and decompression of the posterior tibial nerve are the recommended treatments, particularly when neurogenic pain is present. Although the recurrence rate is less than 5%, complete resection is recommended. Preservation of the fascicle is the key to preventing neurological complications [15], and longitudinal incision on the perineurium at the mid-portion of the tumor is reportedly less invasive to the nerve [8]. When results were classified by cause, patients with coalition or tumor were reported to fare better, whereas those with idiopathic or traumatic causes fared worse [16]. We herein describe a rare case of tarsal tunnel syndrome caused by a schwannoma arising from the posterior tibial nerve located on the posterior medial ankle. After enucleation of this tumor, the patient was asymptomatic and had no recurrence or related sequelae except for minor numbness in the plantar aspects of the digits at the 6-month follow-up. Limitations of this report are the lack of electrophysiological information and the short follow-up period. Although schwannoma of the posterior tibial nerve causing tarsal tunnel syndrome is rare, it should be considered even if a ganglion is clinically suspected.

#### References

- Knight DMA, Birch R and Pringle J: Benign solitary schwannomas: a review of 234 cases. J Bone Joint Surg Br (2007) 89: 382–387.
- Birch R, Bonney G and Wynn Parry CB: The peripheral nervous system and neoplastic disease. Surgical disorders of peripheral nerves (1998) 335–352.
- Grossman RG, Mandracchia VJ, Urbas WM and Mandracchia DM: Neurilemmoma of the posterior tibial nerve with an uncommon case presentation. J Foot Surg (1992) 31: 219–224.
- Mangrulkar VH, Brunetti VA, Gould ES and Howell N: Unusually large pedal schwannoma. J Foot Ankle Surg (2007) 46: 398–402.
- Graviet S, Sinclair G and Kajani N: Ancient schwannoma of the foot. J Foot Ankle Surg (1995) 34: 46–50.
- Odom RD, Overbeek TD, Murdoch DP and Hosch JC: Neurilemmoma of the medial plantar nerve: a case report and literature review. J Foot Ankle Surg (2001) 40: 105–109.
- 7. Nawabi DH and Sinisi M: Schwannoma of the posterior tibial

- nerve: the problem of delay in diagnosis. J Bone Joint Surg Br (2007) 89: 814-816.
- Kwok KB, Lui TH and Lo WN: Neurilemmoma of the first branch of the lateral plantar nerve causing tarsal tunnel syndrome. Foot Ankle Spec (2009) 2: 287–290.
- Boya H, Ozcan O and Oztekin HH: Tarsal tunnel syndrome associated with a neurilemmoma in posterior tibial nerve: a case report. Foot (Edinb) (2008) 18: 174–177.
- Milnes HL and Pavier JC: Schwannoma of the tibial nerve sheath as a cause of tarsal tunnel syndrome-a case study. Foot (Edinb) (2012) 22: 243–246.
- Hallahan K, Vinokur J, Demski S, Faulkner-Jones B and Giurini J: Tarsal tunnel syndrome secondary to schwannoma of the posterior tibial nerve. J Foot Ankle Surg (2014) 53: 79–82.
- Patel AT, Gaines K and Malamut R: Usefulness of electrodiagnostic techniques in the evaluation of suspected tarsal tunnel syndrome: an evidence-based review. Muscle Nerve (2005) 32: 236– 240.
- 13. Dellon AL: Tinel or not Tinel. J Hand Surg Br (1984) 9: 216.
- Varma DG, Moulopoulos A, Sara AS, Leeds N, Kumar R, Kim EE and Wallace S: MR imaging of extracranial nerve sheath tumors. J Comput Assist Tomogr (1992) 16: 448–453.
- Woodruff JM, Selig AM, Crowley K and Allen PW: Schwannoma (neurilemmoma) with malignant transformation. A rare, distinctive peripheral nerve tumor. Am J Surg Pathol (1994) 18: 882–895.
- Takakura Y, Kitada C and Sugimoto K: Tarsal tunnel syndrome. Causes and results of operative treatment. J Bone Joint Surg Br (1991) 73: 125–128.