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A Patient with Tarsal Tunnel Syndrome Associated with the Flexor Digitorum Accessorius Longus Muscle

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Abstract

The flexor digitorum accessorius longus muscle (ALM) can be overlooked as the eliciting factor in patients with tarsal tunnel syndrome (TTS), an entrapment neuropathy of the posterior tibial nerve that elicits sole numbness and pain. Most elicitations are idiopathic, however, mass lesions within the tarsal tunnel can be also implicated. We report an 80-year-old woman whose flexor digitorum ALM led to the onset of bilateral TTS. She had suffered numbness in both soles for 3 years. Magnetic resonance imaging (MRI) of the bilateral tarsal tunnel showed that the posterior tibial nerve was compressed by the arteriovenous complex and in contact with the flexor digitorum ALM. We diagnosed bilateral TTS based on her symptoms and imaging findings, and performed bilateral decompression surgery of the posterior tibial nerve under local anesthesia. The artery on both sides was dislocated for nerve decompression. Because the posterior tibial nerve on the right side was strongly compressed in ankle plantar flexion we excised a portion of the tendon compressing the nerve. Postoperatively her symptoms gradually improved and she reported surgical satisfaction 6 months after the operation. In patients with flexor digitorum ALM-related TTS, the effect of dynamic factors on MRI findings and on surgical treatment decisions must be considered. Intraoperatively, not only the flexor digitorum ALM, but also other potential etiologic factors eliciting TTS must be kept in mind.

Keywords: decompression, dynamic, flexor digitorum accessorius longus muscle, surgery, tarsal tunnel syndrome

Introduction

Tarsal tunnel syndrome (TTS) is an entrapment neuropathy of the posterior tibial nerve at the tarsal tunnel that can elicit feelings of numbness and pain in the sole, a feeling of foreign objects sticking to the foot, and coldness. TTS affects the patient's quality of life and can be treated by less invasive surgery. However, as some patients are not satisfied with the surgical outcome, comprehensive preoperative information is needed.[1-3] In most patients the TTS-eliciting factor is idiopathic, however, mass lesions, e.g. ganglia, and accessory muscles within the tarsal tunnel, can be implicated.[1,4-6] We report a patient whose flexor digitorum accessorius longus muscle (ALM), one of the accessory muscles, led to the onset of TTS.

Case Report

This 80-year-old female had experienced numbness and coldness in her bilateral soles for 3 years. Because conservative treatment failed to improve her symptoms she consulted us. She reported severe numbness in both soles accompanied by feelings of coldness and of foreign objects sticking to her soles. Bilaterally the Tinel sign in the tarsal tunnel was negative.

Magnetic resonance imaging (MRI) of the right side in neutral position showed that the flexor digitorum ALM ran through the Achilles tendon of the arteriovenous complex in the tarsal tunnel (Fig. 1A, B, D). The posterior tibial nerve was entrapped between the arteriovenous complex and the accessory muscle (Fig. 1G, I). MRI of the tarsal tunnel with the left ankle in neutral position revealed that

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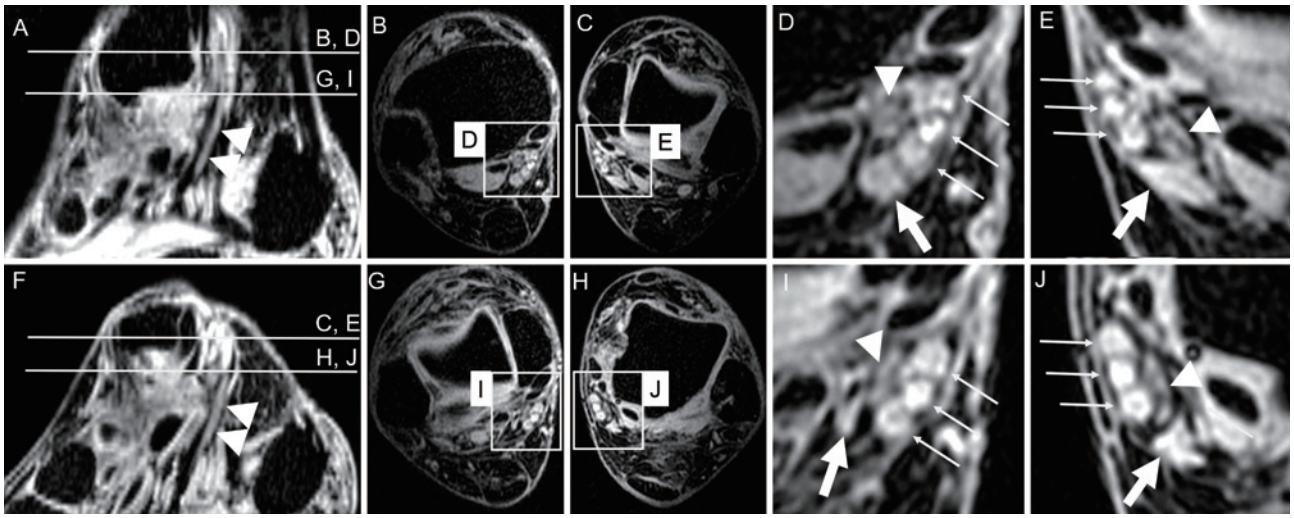

Fig. 1 MRI scans of the tarsal tunnel (fat suppression T2*).
Right side: Sagittal image (A) and axial images (B, D, G, I).
Left side: Sagittal image (F) and axial images (C, E, H, J).
A, B, D The flexor digitorum ALM (arrow) runs along the Achilles tendon side of the arteriovenous complex (thin arrows).
G, I The nerve (arrowhead) is compressed between the arteriovenous complex (thin arrows) and the accessory muscle (arrow).
C, E, F The flexor digitorum ALM (arrow) runs along the Achilles tendon side of the posterior tibial nerve (arrowhead).
H, J The distal portion of the nerve was strongly compressed by the arteriovenous complex (thin arrows).

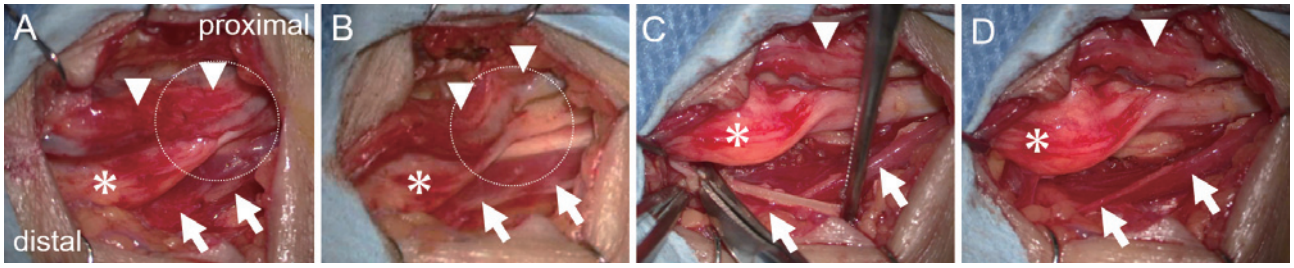

Fig. 2 Surgical field -Right-side tarsal tunnel.
A Ankle in resting position. The nerve (*) is sandwiched between the artery (arrowhead) and the flexor digitorum ALM (arrow) (dotted circle).
B Ankle in plantar flexion. The nerve (*) is strongly compressed by the tendon-like component of the flexor digitorum ALM (arrow) (dotted circle).
C Excision of the tendon-like component.
D After decompression.

the flexor digitorum ALM ran along the Achilles tendon side of the posterior tibial nerve (Fig. 1C, E, F); the distal posterior tibial nerve was strongly compressed by the arteriovenous complex (Fig. 1H, J). Electrophysiological examination demonstrated that the motor nerve conduction velocity of the tibial nerve was normal (41.4 ms on the left and 40.5 ms on the right), but sensory nerve conduction velocities of the medial and lateral plantar nerves showed no waveforms on either side.

We diagnosed bilateral TTS based on the patient's symptoms and imaging findings. Because medication yielded little symptom improvement we initially performed right posterior tibial nerve ablation. A 25-mm skin incision above the right tarsal tunnel and opening of the flexor retinaculum revealed the flexor digitorum ALM on the side of the Achilles tendon of the arteriovenous complex. Dislocating the arteriovenous complex toward the medial malleolus showed that the artery indented the nerve. For its decompression, the artery was fixed subcutaneously on the medial malleolar side with three 5-0 nylon needles. Doppler ultrasound confirmed sufficient arterial blood flow. Although it should have been possible to decompress the pinched nerve between the artery and the flexor digitorum ALM by releasing the compression exerted by the artery, the flexor digitorum ALM was in contact with the nerve (Fig. 2A). However, when the patent positioned the ankle

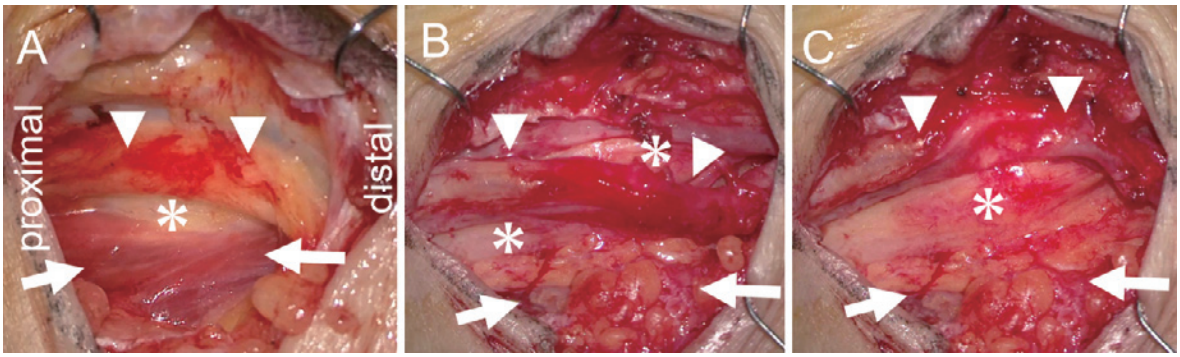

Fig. 3 Surgical field -Left-side tarsal tunnel.
A, B The nerve (*) is compressed by the artery (arrowhead) rather than the flexor digitorum ALM (arrow).
C The nerve (*) is decompressed by transposition of the artery (arrowhead).

in plantar flexion to obtain sufficient muscle contraction, the flexor digitorum ALM tendon touched and deformed the nerve (Fig. 2B). Resection of a portion of the implicated tendon released the dynamic entrapment (Fig. 2C, D).

Left posterior tibial nerve decompression was performed 3 days later. The procedures and findings were as on the right side (Fig. 3A). As the nerve was compressed by the arteriovenous complex (Fig. 3B), the complex was displaced to the medial malleolus side and fixed with three 5-0 nylon needles. Doppler ultrasound confirmed sufficient blood flow (Fig. 3C). There was no significant nerve compression by the flexor digitorum ALM when the patient intraoperatively placed either ankle in the resting- or plantar flexion position to obtain sufficient muscle contraction.

After the operation she was subjected to neither bed rest nor external fixation and her symptoms improved. Her numerical rating scale (NRS) grades for symptom improved from 7 to 4 the day after surgery; one month later the grade was 1. At the last follow-up 6 months postoperatively her NRS grades for symptom remains 1; her NRS grade for outcome satisfaction were 8. As her postoperative course was favorable, no additional electrophysiological studies were acquired.

Discussion

The flexor digitorum ALM is one of the accessory muscles around the ankle joint. Earlier studies[4,7-9] identified uni- or bilateral flexor digitorum ALM in 2 - 8% of cadavers. MRI of 100 asymptomatic legs revealed an incidence of 6%.[4] The flexor digitorum ALM originates at several structures in the proximal third of the leg, e.g the tibia, fibula, deep fascia, and tendon. A short head starting at the lower third of the leg has been observed.[4,7] On MRI scans of 20 sides of the flexor digitorum ALM, 8 muscle originated at lower-extremity muscles including the flexor hallucis longus, 6 at the flexor retinaculum, and in 6 the origin was unidentifiable.[4] In the tarsal tunnel, the nerve normally runs along the flexor hallucis longus muscle and the Achilles tendon side of the arteriovenous complex, exits the tarsal tunnel deep in the arteriovenous complex, and reaches the flexor hallucis longus tendon and the quadratus plantar muscle.[4,7,8,10] In plantar flexion, the nerve primarily affects the 2nd to 5th toes.[5,7,9]

Our earlier reports of consecutive surgical cases of TTS did not include patients whose TTS was associated with ALM.[2,11-14] We did not include flexor digitorum ALM-related TTS in our case series that examined adverse postoperative events in 257 patients,[13] or in our report on MRI findings on 28 feet of consecutive patients with TTS,[2] although others[1,5,7,8,15] reported that 11 - 16% of surgically-addressed TTS symptoms were associated with accessory muscles, in most instances the flexor digitorum ALM was implicated. Our active treatment of idiopathic TTS may account for the difference between their- and our findings. Cheung et al.[4] who examined 12 patients (14 legs) with flexor digitorum ALM reported that none of their patients presented with TTS and suggested that the presence of that muscle may not be a risk factor for TTS. Deleu et al.[7] proposed that besides TTS elicited by direct compression of the nerve by the flexor digitorum ALM, trauma or strenuous exercise may result in muscle edema, hypertrophy, and TTS. Their systematic review of 23 patients with TTS associated with the flexor digitorum ALM revealed that all were positive for the Tinel sign. Electrophysiological studies cannot be relied on to return diagnostic information.

Although axial MRI scans are diagnostically useful, on sagittal images it is difficult to identify the muscles unless they are quite large.[4] While the flexor digitorum ALM is implicated in the etiology of TTS, in some preoperative studies it was overlooked and discovered only intraoperatively.

Intraoperatively we found that ankle plantar flexion increased compression on the nerve. Elsewhere[14] we reported that MRI scans of the tarsal tunnel with the ankle in plantar flexion revealed increased nerve compression in patients with idiopathic TTS; an increase in arterial tortu-

osity in that ankle position was primarily implicated. As our current intraoperative findings showed that nerve compression by the ALM was exacerbated with the ankle in plantar flexion, we think that such strong nerve compression could be visualized on preoperative MRI scans and that preoperative plantar-flexion MRI may increase diagnostic accuracy in patients with idiopathic- and ALM-related TTS.

While nerve-compressing muscles can be addressed and the compressing flexor digitorum ALM is commonly resected in patients treated for TTS, it can be difficult to remove it at its origin or *in toto*.[1,5,7,9,15,16] Ettehadi et al.[10] obtained good treatment outcomes by nerve decompression without muscle resection. For successful decompression, dynamic compression elicited by muscle contraction must be evaluated intraoperatively.[8] In our TTS patient we noted no severe nerve compression on the left side by the flexor digitorum ALM. However, on the right side, ankle movement exacerbated nerve entrapment by the muscle. Therefore we removed it under local anesthesia, which made intraoperative dynamic observations possible.

While others[5,9,16] obtained good surgical results, unsatisfactory outcomes, possibly attributable to prolonged flexor digitorum ALM neuropathy, a congenital anomaly, have been reported.[7,8] Based on their study of operated TTS patients, Bouysset et al.[1] concluded that the presence of accessory muscles did not prevent effective tibial nerve release. Kinoshita et al.[5] addressed 84 feet with nerve decompression by removing the accessory muscles in 9 feet with TTS symptoms, in 5 of these the symptoms did not improve. They reported that unfavorable factors related to treatment failure were valgus and/or bone-nerve contact observed on imaging scans and that other TTS-eliciting factors must also be considered. Elsewhere our team[2,11,12,14] suggested that nerve compression due to tortuosity of the concomitant posterior tibial artery may lead to the development of idiopathic TTS. In our current patient, preoperative MRI and intraoperative findings also revealed that nerve compression by the posterior tibial artery had an effect on the onset of TTS and that by its decompression we obtained favorable treatment results.

Conclusion

The flexor digitorum AML can elicit TTS. Although preoperative tarsal-tunnel MRI scans can reveal the site of nerve compression, it may appear slight when it is elicited dynamically. Preoperative MRI scans with the ankle in plantar flexion may be of diagnostic value. However, in TTS patients scheduled for surgical treatment not only the muscle's presence, but also other TTS-inciting factors must be considered.

Conflicts of Interest Disclosure

The authors declare that they have no conflicts of interest in relation to our submitted manuscript and no commercial relationships or financial support from pharmaceutical or other companies. All authors are members of The Japan Neurosurgical Society (JNS) and have submitted signed, self-reported COI Disclosure Statement Forms available at the website for JNS members.

References

1) Bouysset M, Denarié D, Coillard JY, et al.: Predictive factors of effective tibial nerve release in tarsal tunnel syndrome. *Foot Ankle Surg* 28: 610-615, 2022
2) Kim K, Kokubo R, Isu T, et al.: Magnetic Resonance Imaging Findings in Patients with Tarsal Tunnel Syndrome. *Neurol Med Chir* 62: 552-558, 2022
3) Kokubo R, Kim K, Isu T, et al.: Patient satisfaction with surgery for tarsal tunnel- and carpal tunnel syndrome. Comparative study. *Neurol Med Chir* 63: 116-121, 2023
4) Cheung YY, Rosenberg ZS, Colon E, et al.: MR imaging of flexor digitorum accessorius longus. *Skeletal Radiol* 28: 130-137, 1999
5) Kinoshita M, Okuda R, Morikawa J, et al.: Tarsal tunnel syndrome associated with an accessory muscle. *Foot Ankle Int* 24: 132-136, 2003
6) McSweeney SC, Cichero M: Tarsal tunnel syndrome - A narrative literature review. *Foot* 25: 244-250, 2015
7) Deleu PA, Bevernage BD, Birch I, et al.: Anatomical characteristics of the flexor digitorum accessorius longus muscle and their relevance to tarsal tunnel syndrome. A systematic review. *J Am Podiatr Med Assoc* 105: 344-355, 2015
8) Sammarco GJ, Conti SF: Tarsal tunnel syndrome caused by an anomalous muscle. *J Bone Joint Surg Am* 76: 1308-1314, 1994
9) Wittmayer BC, Freed L: Diagnosis and surgical management of flexor digitorum accessorius longus-induced tarsal tunnel syndrome. *J Foot Ankle Surg* 46: 484-487, 2007
10) Ettehadi H, Saragas NP, Ferrao P: A Rare Case of Flexor Digitorum Accessorius Longus Muscle Presenting as Tarsal Tunnel Syndrome. *Arch Bone Jt Surg* 10: 112-116, 2022
11) Fujihara F, Isu T, Kim K, et al.: Transposition using indocyanine green for tarsal tunnel decompression. *World Neurosurg* 141: 142-148, 2020
12) Kim K, Isu T, Morimoto D, et al.: Neurovascular bundle decompression without excessive dissection for tarsal tunnel syndrome. *Neurol Med Chir* 54: 901-906, 2014
13) Kim K, Isu T, Morimoto D, Kokubo R, Fujihara F, Morita A: Perioperative Complications and Adverse Events after Surgery for Peripheral Nerve- and Para-lumbar Spine Diseases. *Neurol Med Chir* 62: 75-79, 2022
14) Tajiri T, Kim K, Isu T, et al.: Impact of dynamic change of meandering of parallel artery to the idiopathic tarsal tunnel syndrome. *Neurol Med Chir* 63: 165-171, 2023
15) Lalevée M, Coillard JY, Gauthé R, et al.: Tarsal Tunnel Syndrome: Outcome According to Etiology. *J Foot Ankle Surg* 61: 583-589, 2022
16) Lin D, Williams C, Zaw H: A rare case of an accessory flexor hallucis longus causing tarsal tunnel syndrome. *Foot Ankle Surg* 20: e37-39, 2014

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