

CASE REPORT

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Tarsal tunnel syndrome secondary to osteochondroma of the calcaneus: a case report

Sung Hun Won^{1†}, Jahyung Kim^{2†}, Tae-Hong Min², Dong-Il Chun², Young Yi³, Sang Hak Han⁴ and Jaeho Cho^{5*} 

Abstract

Background: Tarsal tunnel syndrome is an entrapment neuropathy that can be provoked by either intrinsic or extrinsic factors that compresses the posterior tibial nerve beneath the flexor retinaculum. Osteochondroma, the most common benign bone tumor, seldom occur in foot or ankle. This is a rare case of tarsal tunnel syndrome secondary to osteochondroma of the sustentaculum tali successfully treated with open surgical excision.

Case presentation: A 15-year-old male presented with the main complaint of burning pain and paresthesia on the medial plantar aspect of the forefoot to the middle foot region. Hard mass-like lesion was palpated on the posteroinferior aspect of the medial malleolus. On the radiological examination, 2.5 × 1 cm sized bony protuberance was found below the sustentaculum tali. Surgical decompression of the posterior tibial nerve was performed by complete excision of the bony mass connected to the sustentaculum tali. The excised mass was diagnosed to be osteochondroma on the histologic examination. After surgery, the pain was relieved immediately and hypoesthesia disappeared 3 months postoperatively. Physical examination and radiographic examination at 2-year follow up revealed that tarsal tunnel was completely decompressed without any evidence of complication or recurrence.

Conclusions: As for tarsal tunnel syndrome secondary to the identifiable space occupying structure with a distinct neurologic symptom, we suggest complete surgical excision of the causative structure in an effort to effectively relieve symptoms and prevent recurrence.

Keywords: Osteochondroma, Tarsal tunnel syndrome, Calcaneus

Background

Tarsal tunnel syndrome is an entrapment neuropathy of the posterior tibial nerve or one of its branches within the tarsal tunnel on the medial aspect of the ankle [1]. Symptoms of burning, tingling, and pain along the foot plantar region can be triggered by either intrinsic or extrinsic factors that compress the posterior tibial nerve

beneath the flexor retinaculum [2]. Among the rest, a bony structure protuberated within the tarsal tunnel including talocalcaneal coalition, talar or calcaneal fracture, nonunion, or os sustentaculum tali have been identified as one of the significant factors to tarsal tunnel syndrome due to its firm mass effect [1, 3–7]. Osteochondromas, the most common benign bone tumor also characterized by a stalk or a flat protuberance emerging from the surface of the bone, however, rarely occur in tarsal bones [8–10]. Here, we report a patient who presented with tarsal tunnel syndrome secondary to osteochondroma of the sustentaculum tali and was successfully treated through surgical decompression.

* Correspondence: hohotoy@nate.com

[†]Sung Hun Won and Jahyung Kim contributed equally to this work and should be considered co-first authors.

⁵Department of Orthopaedic Surgery, Chuncheon Sacred Heart Hospital, Hallym University, 77, Sakju-ro, Chuncheon-si, Gangwon-do 200-704, Republic of Korea

Full list of author information is available at the end of the article



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Case presentation

Preoperative evaluation

A 15-year-old male presented with insidious onset of pain and a palpable mass on the left foot for 1 year. The burning pain was combined with paresthesia on the medial plantar aspect of the forefoot to the middle foot region, along the innervation of the medial plantar nerve. The symptoms were aggravated upon walking or prolonged standing and were relieved at rest. No previous trauma or surgical history was reported. On physical examination, there was a fixed, hard mass-like lesion palpated on the posteroinferior aspect of the medial malleolus. The range of motion of the lower extremity including subtalar motion was normal and no evidence of hindfoot malalignment was detected. Light touch and pin prick were performed and the patient complained of hypoesthesia of 80% the normal contralateral leg along the medial plantar foot area. A positive Tinel's sign was detected.

The plain radiographs of both axial and lateral views of the calcaneus showed a large pedunculated mass located posteroinferomedial to the sustentaculum tali. In addition, corticomedullary continuity between the sustentaculum tali and the mass could be detected (Fig. 1). No other visible pedunculated lesion could be detected on the whole body x-ray. To clarify the lesion, a computed tomography (CT) was performed, which showed a 2.5×1 cm sized bony protuberance below the sustentaculum tali, demonstrating an apparent corticomedullary continuity with the underlying bone. Neither the talocalcaneal coalition nor fracture within the bony protuberance was detected (Fig. 2). Magnetic Resonance Image (MRI) scans confirmed the high T2 signal intensity mass extended from the sustentaculum tali, occupying the tarsal tunnel. The fatty bone marrow was observed on the central portion of the mass, continuously with the marrow cavity of the underlying bone, surrounded by thin

cartilaginous cap (Fig. 3). The Foot and Ankle Outcome Score (FAOS) was 81.11 points [11]. Electromyography was not performed due to typical signs and symptoms of tarsal tunnel syndrome and surgical intervention was indicated due to presence of tunnel occupying lesion.

Surgical procedure

Surgical excision of the bony mass was performed under the general anesthesia. The flexor retinaculum was released and 2.5×1 cm sized bony mass was found on the posteroinferior border of the sustentaculum tali, compressing the posterior tibial nerve underneath. No talocalcaneal coalition bar was detected. The bony lesion was completely excised from the sustentaculum tali using the osteotomes and rongeur until no remaining protuberance was seen within the tunnel, and the tension on the nerve was eventually released (Fig. 4). Excised bony mass was sent to the pathology department for histologic examination.

Postoperative progression

The histologic sample was confirmed to be osteochondroma (Fig. 5). A below-knee splint was applied for 2 weeks and the patient was allowed to fully bear weight and return to the regular activity after the stitches on the wound were removed. The patient's pain was relieved immediately after surgery and the hypoesthesia disappeared 3 months postoperatively. No complications or recurrence of symptoms were detected at the 2-year follow-up and the FAOS improved to 100 points.

Discussion and conclusions

Osteochondroma is the most common benign bone tumor accounting for more than 30% of all the benign bone tumors [12]. It is defined as a cartilage capped bony projection arising on the external surface of bone containing a marrow cavity that is continuous with that of the underlying bone [8]. It tends to be found in adolescent or children and no predilection among the gender exists. Most of the lesion occur as solitary, nonhereditary lesion, which is usually located at the metaphysis of the long bones, especially near the knee joint [8, 10]. Osteochondroma is seldom found in foot or ankle, excluding the case of Multiple Hereditary Exostoses [13].

In terms of differential diagnosis of the osteochondroma near the ankle, Dysplasia epiphysealis hemimelica (DEH) could also be taken into account because it is mostly affected in ankle and knee [14]. DEH is a rare developmental disorder defined as asymmetric osteochondral overgrowth of the cartilage of epiphysis or epiphyseal equivalent [15]. It is characterized as an intra-articular, irregular bony lesion with osseous continuity to the underlying bone arising from multiple



Fig. 1 Preoperative axial (a) and lateral (b) plain radiographs of the calcaneus. A large pedunculated mass is located posteroinferomedial to the sustentaculum tali. Corticomedullary continuity is observed between the mass and the sustentaculum tali (arrows)



Fig. 2 Preoperative CT scans of the left foot. Coronal (a), axial (b), sagittal (c), and three dimensional reconstructed (d) views show 2.5 × 1 cm sized bony protuberance below the sustentaculum tali, demonstrating an apparent corticomedullary continuity with the underlying bone (arrows). Neither talocalcaneal coalition nor fracture within the bony protuberance is detected

distinct ossification centers. Although the histopathologic study confirmed the lesion of this case report as osteochondroma, it is known to be difficult to discriminate osteochondroma from DEH both clinically and pathologically [16, 17]. In fact, calcaneus is an apophysal equivalent and DEH occurs predominantly on male in developmental ages, just as the patient in this case report [18]. Despite the definite diagnostic value of the molecular assays, these are costly and radiological findings are considered to be important in discriminating these two lesions [19]. In this case, the lesion was located under the sustentaculum tali, which could be considered extra-articular. In addition, the cortical and medullary continuity between the lesion and the sustentaculum tali was apparent compared with that of DEH [15]. Lastly, no additional lesion could be detected in other ossification centers on the whole-body x-ray,

excluding the generalized form of DEH. For these reasons, we thought it would be appropriate to regard the lesion as the osteochondroma.

Patients with osteochondroma usually do not have symptoms, excluding the discomfort caused by mechanical irritation of the mass. However, if osteochondroma occur near the neurovascular structure, mostly popliteal nerve and artery around knee, numbness, weakness, loss of pulse, and color change can be presented on the affected limb [20]. Patient in our case was put into a similar situation by the osteochondroma originated from the rarely affected structure, the sustentaculum tali. As a result, tarsal tunnel syndrome was provoked.

Surgical intervention for tarsal tunnel syndrome is indicated among symptomatic patients who do not respond to non-operative treatment or those who have space-occupying lesion within the tarsal tunnel [2, 21]. Besides, a positive Tinel's sign is considered one of the most predictable indicators to the satisfactory outcome

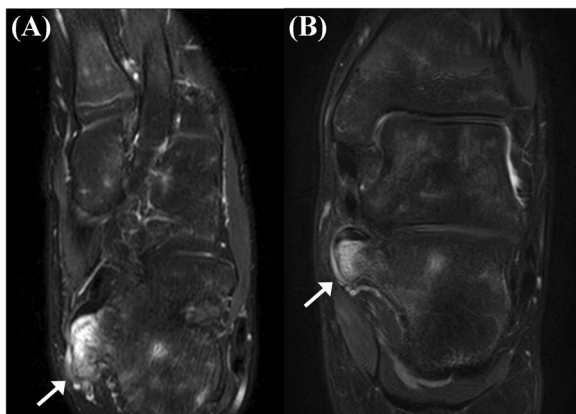


Fig. 3 Preoperative MRI scans show the high T2 signal intensity mass extended from the sustentaculum tali occupying the tarsal tunnel (a, b). The fatty bone marrow is seen on the central portion of the mass, continuously with the marrow cavity of the underlying bone, surrounded by thin cartilaginous cap (arrows)

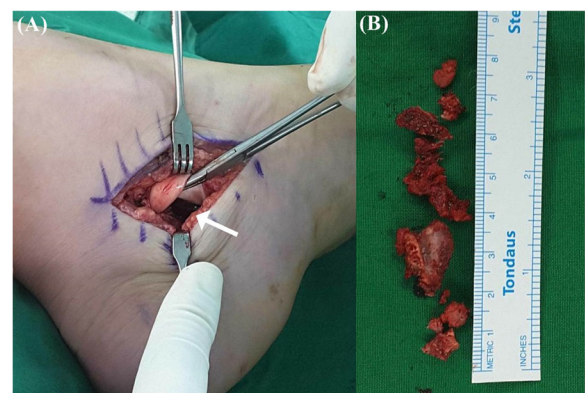


Fig. 4 Intraoperative photographs. a Sufficient blank space (arrow) under the retracted tendon, occupied after complete excision of the osteochondroma. b Bony mass meticulously excised from sustentaculum tali using the osteotomes and rongeur

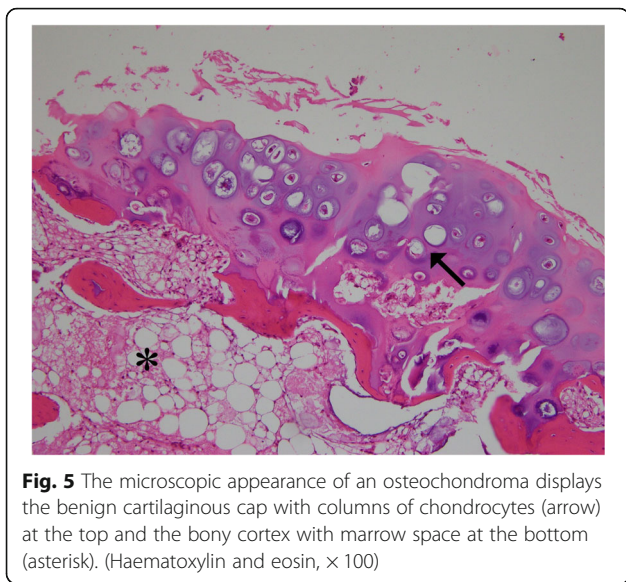


Fig. 5 The microscopic appearance of an osteochondroma displays the benign cartilaginous cap with columns of chondrocytes (arrow) at the top and the bony cortex with marrow space at the bottom (asterisk). (Haematoxylin and eosin, x 100)

after the surgery [1]. The standard surgical approach to release the compressed posterior tibial nerve in the tarsal tunnel syndrome is known to be open tarsal tunnel decompression [1]. Our surgical success in this case seems to have been derived from radical excision of the bony mass because incomplete removal of the osteochondroma and inadequate tarsal tunnel release are together related to the recurrence and residual symptoms [12, 22].

One previous case report has demonstrated a similar case of tarsal tunnel syndrome secondary to osteochondroma of the sustentaculum tali [23]. However different from our case, the patient had undergone tarsal tunnel release prior to presentation and the talocalcaneal coalition was detected along with the osteochondroma. Scar tissues resulted from the previous operation and the mass effect of the talocalcaneal coalition may have affected the compression of the posterior tibial nerve in combination so the talocalcaneal bar resection was performed additionally. In this study we were able to effectively decompress the posterior tibial nerve by solitarily excising the osteochondroma followed by immediately diminished the symptoms.

In conclusion, we encountered a rare case of tarsal tunnel syndrome secondary to osteochondroma of the sustentaculum tali in pediatric patient. If the space occupying structure within the tunnel is identified with a distinct neurologic symptom, complete surgical excision of the causative structure can bring out significant improvement of preexisted symptoms without recurrence.

Abbreviations

CT: Computed Tomography; FAOS: Foot and Ankle Outcome Score; MRI: Magnetic Resonance Image

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Authors' contributions

JC was responsible for the clinical management of the patient and the drafting and editing of the manuscript. SHW and JK collected the patient's clinical data and wrote the manuscript. They contributed equally to the article. THM, DIC and YY participated in critical revision of the manuscript for intellectual content. SHH is a contributor in analyzing and explaining the pathology data. All authors read and approved the final manuscript.

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Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Ethics approval and consent to participate

Informed Consent form for participate of the case was obtained from the patient's parent due to children (under 16 years old). In addition, the present study was approved by the Institutional Review Board and Human Research Ethics Committee of Hallym University Chuncheon Sacred Heart Hospital (No.2020-04-007).

Consent for publication

Informed Consent form for publication of the case was obtained by written from the patient's parent due to children (under 16 years old). Also, the present study was approved the waiver by the Institutional Review Board and Human Research Ethics Committee of Hallym University Chuncheon Sacred Heart Hospital (No.2020-04-007).

Competing interests

The authors declare that they have no competing interests.

Author details

¹Department of Orthopaedic Surgery, Bone & Joint center, Soonchunhyang University Seoul Hospital, Seoul, Republic of Korea. ²Department of Orthopaedic Surgery, Seoul Hospital, Soonchunhyang University Seoul Hospital, Seoul, Republic of Korea. ³Department of Orthopaedic Surgery, Seoul Paik Hospital, Inje University, Seoul, Republic of Korea. ⁴Department of Pathology, Chuncheon Sacred Heart Hospital, Hallym University, Chuncheon, Republic of Korea. ⁵Department of Orthopaedic Surgery, Chuncheon Sacred Heart Hospital, Hallym University, 77, Sakju-ro, Chuncheon-si, Gangwon-do 200-704, Republic of Korea.

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References

1. Doneddu PE, Coraci D, Loreti C, Piccinini G, Padua L. Tarsal tunnel syndrome: still more opinions than evidence. Status of the art. *Neurol Sci*. 2017;38(10):1735-9.
2. Franson J, Baravarian B. Tarsal tunnel syndrome: a compression neuropathy involving four distinct tunnels. *Clin Podiatr Med Surg*. 2006;23(3):597-609.
3. Guneri B, Kose O, Turan A, Acar B, May H. Tarsal tunnel syndrome due to Talocalcaneal tarsal coalition: a case report. *SM J Orthop*. 2016;2(3):1038-40.
4. Huri G, Atay AO, Leblebicioglu GA, Doral MN. Fracture of the sustentaculum tali of the calcaneus in pediatric age: a case report. *J Pediatr Orthop B*. 2009; 18(6):354-6.
5. Lee M, Chan P, Chau L, Yu K. Tarsal tunnel syndrome caused by talocalcaneal coalition. *Clin Imaging*. 2002;26(2):140-3.
6. Myerson MS, Berger BI. Nonunion of a fracture of the sustentaculum tali causing a tarsal tunnel syndrome: a case report. *Foot Ankle Int*. 1995;16(11): 740-2.
7. Stefko RM, Lauerman WC, Heckman JD. Tarsal tunnel syndrome caused by an unrecognized fracture of the posterior process of the talus (Cedell fracture). A case report. *J Bone Joint Surg Am*. 1994;76(1):116-8.
8. Bovée JVMG, Khurana J, Abdul-Karim F. Osteochondroma: IARC, 2002.

9. Saglik Y, Altay M, Unal VS, Basarir K, Yildiz Y. Manifestations and management of osteochondromas: a retrospective analysis of 382 patients. *Acta Orthop Belg.* 2006;72(6):748.
10. Solomon L. Hereditary multiple exostosis. *J Bone Joint Surg (Br).* 1963;45(2):292–304.
11. Lee KM, Chung CY, Kwon SS, Sung KH, Lee SY, Won SH, Lee DJ, Lee SC, Park MS. Transcultural adaptation and testing psychometric properties of the Korean version of the foot and ankle outcome score (FAOS). *Clin Rheumatol.* 2013;32(10):1443–50.
12. Kitsoulis P, Galani V, Stefanaki K, Paraskevas G, Karatzias G, Agnantis NJ, Bai M. Osteochondromas: review of the clinical, radiological and pathological features. *In Vivo.* 2008;22(5):633–46.
13. Herrera-Perez M, Aciego De Mendoza M, De Bergua-Domingo JM, Pais-Brito JL. Osteochondromas around the ankle: report of a case and literature review. *Int J Surg Case Rep.* 2013;4(11):1025–7.
14. Rosero VM, Kiss S, Terebessy T, Köllö K, Szöke G. Dysplasia epiphysealis hemimelica (Trevor's disease): 7 of our own cases and a review of the literature. *Acta Orthop.* 2007;78(6):856–61.
15. Tyler PA, Rajeswaran G, Saifuddin A. Imaging of dysplasia epiphysealis hemimelica (Trevor's disease). *Clin Radiol.* 2013;68(4):415–21.
16. Nowicki PD, Borders H. Dysplasia epiphysealis hemimelica (Trevor disease) of the ankle. *Orthopedics.* 2015;38(4):216–271.
17. Struijs PA, Kerkhoffs GM, Besselaar PP. Treatment of dysplasia epiphysealis hemimelica: a systematic review of published reports and a report of seven patients. *J Foot Ankle Surg.* 2012;51(5):620–6.
18. Ozan F, Doğar F, Gürbüz K, Ekinci Y, Bilal Ö, Öncel ES. Dysplasia epiphysealis hemimelica (Trevor disease) in the ankle. *Ther Clin Risk Manag.* 2016;12:545–7.
19. Kettelkamp DB, Campbell CJ, Bonfiglio M. Dysplasia epiphysealis hemimelica. A report of fifteen cases and a review of the literature. *J Bone Joint Surg Am.* 1966;48(4):746–65 discussion 765–746.
20. Van JO, Feyen J, Opheide J. Compartment syndrome associated with an osteocartilaginous exostosis. *Acta Orthop Belg.* 1996;62(4):233–5.
21. Ahmad M, Tsang K, Mackenney PJ, Adedapo AO. Tarsal tunnel syndrome: A literature review. *Foot Ankle Surg.* 2012;18(3):149–52.
22. Gould JS. Recurrent tarsal tunnel syndrome. *Foot Ankle Clin.* 2014;19(3):451–67.
23. Öznur A, Alpaslan A. Osteochondroma of the sustentaculum tali producing tarsal tunnel syndrome. *Foot Ankle Surg.* 2002;8(1):63–6.

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