Journal of Advances in Medicine and Medical Research

33(10): 84-89, 2021; Article no.JAMMR.67869
ISSN: 2456-8899
(Past name: British Journal of Medicine and Medical Research, Past ISSN: 2231-0614, NLM ID: 101570965)

Unusual Adult Bilateral Osteochondroma of the Talus with Severe Peroneal Tendinitis and Adult Bilateral Osteochondroma of Calcaneum with Haglund Syndrome – A Report of Two Cases and Review of Literature

A. Raviraj[1*], Manish Samson[1], K. Abhishek Sugumar[1] and Vidyasagar Maalepati[1]

[1]Apollo Hospitals, Bannerghatta Road, Bangalore -560076, India.

Authors' contributions

This work was carried out in collaboration among all authors. Authors AR and KAS designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Author MS managed the analyses of the study. Author VM managed the literature searches. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/JAMMR/2021/v33i1030912
Editor(s):
(1) Dr. Rameshwari Thakur, Muzaffarnagar Medical College, India.
Reviewers:
(1) Nuria Padrosflores, Universidad Miguel Hernandez, Spain.
(2) Carlos Martín Villa, Universidad Complutense de Madrid, Spain.
Complete Peer review History: http://www.sdiarticle4.com/review-history/67869

Case Report

Received 20 February 2021
Accepted 24 April 2021
Published 04 May 2021

ABSTRACT

We present two unusual cases of bilateral osteochondroma around the foot and ankle. The first case of a bilateral osteochondroma of the talus that presented with peroneal tendinitis and second of a case that presented as bilateral Haglund syndrome secondary to osteochondroma of the calcaneum. Both cases presented with an increase in size of the swelling in the sixth decade of life. This unusual presentation and bilaterality has not been reported in the literature.

Keywords: Osteochondromas; ankle surgery; peroneal tendinitis; haglund syndrome.

**Corresponding author: E-mail: orthoraviraj@yahoo.com;*

1. INTRODUCTION

Osteochondromas (Exostosis) are the most frequently occurring benign bone tumor accounting for 26.5% to 34% of all bone neoplasms [1,2,3]. The incidence of the tumor around the foot and ankle has been reported to be 3.9 and 4% in two studies [4,5] with the talus accounting for less than 0.5 % of the cases [6]. Osteochondromas around the foot and ankle including the calcaneum account for only 10% of the incidence of this tumor in practice [7]. They commonly occur around the distal metaphysis of the femur and proximal metaphysis of the tibia.

Although Osteochondroma of the hand and foot are quite rare, even small tumor of the talus produce biomechanical imbalance [3] and larger tumor can cause impingement of adjacent nerves and structures [8,9]. The lesion may also present with complications such as deformities, fractures, neurological or vascular involvement, bursa formation and rarely malignant transformation. Peroneal nerve entrapment caused by a tibial lesion is the most frequent and affects patients between the third and fourth decades of life, with no gender preferences. In advanced cases lesions show a bony outgrowth, with a base of separation between the lesion and the bone, due to the existence of cartilaginous tissue interposed between the two.

We would like to present 2 cases, first of a 57-year-old lady with Bilateral talus osteochondroma who presented with Bilateral severe Peroneal tendinitis and Second of 56year old lady who presented with Bilateral Haglund syndrome secondary to Bilateral calcaneal osteochondroma.

2. CASE REPORT

2.1 Case 1

A 57-year-old lady walked into the out-patient department of our hospital with complaints of pain and swelling over the dorsolateral aspect of both feet for two years. Pain was dull aching in character and aggravated on walking and eversion of foot.

The swelling on the right foot was approximately 3 X 2 Cm, located just distal and anterior to the tip of the lateral malleolus, skin over the swelling was normal. The swelling on the left foot was 4x2 cm, located just distal and anterior to lateral malleolus. Skin over the swelling was normal. The swellings were immobile and had an irregular surface. The dorsalis pedis artery (DPA) and posterior tibial artery (PTA) pulses were well felt, and ankle range of motion was normal bilaterally.

The Patient was radiologically evaluated using plain radiographs, CT and MRI scans. The MRI showed irregular bony outgrowth with marrow continuity, few adjacent fragments, edema arising from lateral talus on both sides. Secondary friction tendinosis of peroneal tendons seen. Degenerative changes were noted in the ankle and the subtalar joints. Laboratory work-up (Serum calcium, ALP, PO4) was within normal limits. A working diagnosis of Bilateral osteochondroma of talus was considered and patient was advised surgical excision of the tumor.

Incision was marked centered over the swelling and along the posterior border of the lateral malleolus. The peroneal sheath was opened, and tendons retracted for better exposure of the tumor. Extensive cartilage capped masses were noted, and the tumor was completely removed at its base in multiple large and small pieces. Complete removal of the exostosis was confirmed under fluoroscopy. The peroneal sheath was repaired, and wound was closed in layers.

The excised mass was sent to histopathology which on multiple sections revealed similar histology. The tumor had a cartilaginous cap made up of hyalin cartilage arranged in ill formed lobules. There were intercommunicating bony trabeculae with intervening fibrous to fatty marrow beneath the cartilage cap with no evidence of malignancy.

The post-operative period was uneventful and post op radiographs showed completed excision of the tumor. Patient was encouraged to weight bear from 2nd post-operative day. Radiographs on two years follow-up showed no recurrence.

2.2 Case 2

A 56-year-old lady walked into our out-patient department with the complaints of bilateral heel pain and progressive increase in swelling in both heels for 6 months. The pain had increased in intensity from 3 days prior to presentation. Pain was described to be dull-aching in character and aggravated characteristically on walking in shoes. On palpation there were bilateral bony

hard swellings at the calcaneal tuberosity which were non-tender and immobile. Ankle and subtalar movements were normal and there was no alterations in gait.

The patient was evaluated with plain radiographs which showed bilateral enlargement of the posterosuperior border of the calcaneum and an irregular growth anterior to it. There was no evidence of lysis/sclerosis or cortical breach. Ankle and subtalar joints appeared normal. A working diagnosis of Bilateral Haglund syndrome was considered and patient was advised excision of the swelling.

The surgeries for each foot were carried out 3 weeks apart. The more symptomatic side was operated first. Under tourniquet control, the swelling was approached posterolaterally. Cartilage-capped swelling was excised using bone saw and osteotome. Retrocalcaneal bursa was excised.

The excised mass and bursal tissue were sent for histopathological examination. The tumor had cartilaginous cap with enchondral ossification. There were intercommunicating bony trabeculae with intervening fatty marrow beneath the cartilaginous cap with no evidence of malignancy.

The post-operative period was uneventful and post op radiographs showed completed excision of the tumor. Patient was encouraged to weight bear from second post- operative day. Radiographs on two-year follow-up showed no recurrence.

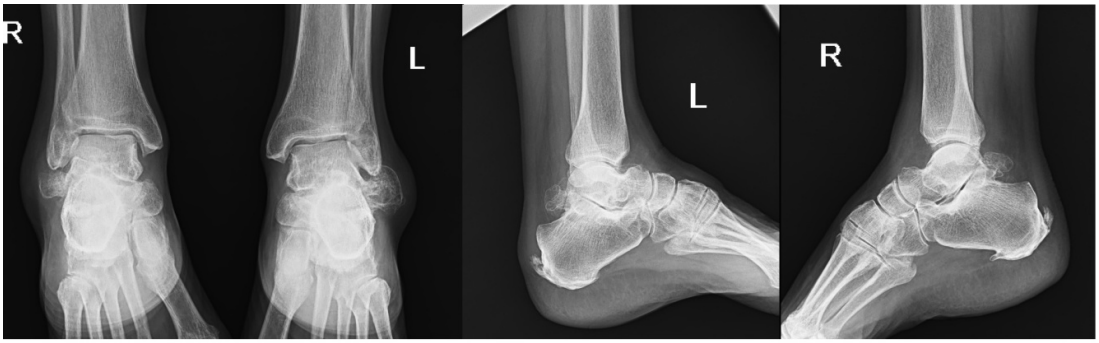

Fig. 1. The plain radiograph anteroposterior and lateral view of the osteochondroma arising from talus bilaterally

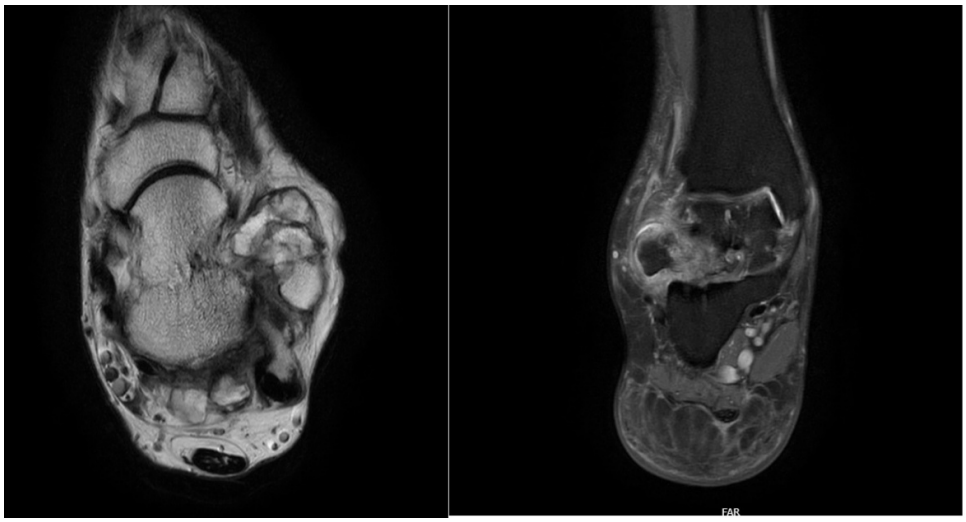

Fig. 2. The magnetic resonance image showing the talar osteochondroma

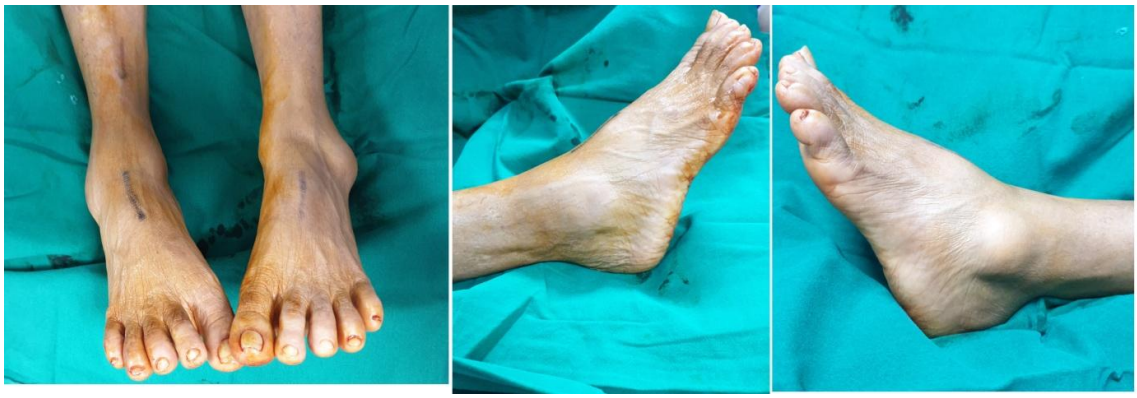

Fig. 3. Clinical picture of the bilateral osteochondroma talus

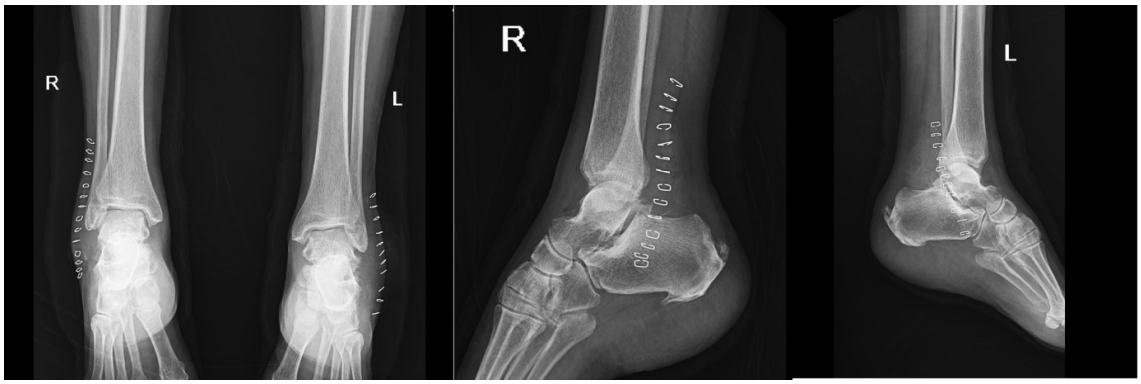

Fig. 4. The plain post-operative radiograph anteroposterior and lateral view of the excised osteochondroma arising from talus bilaterally

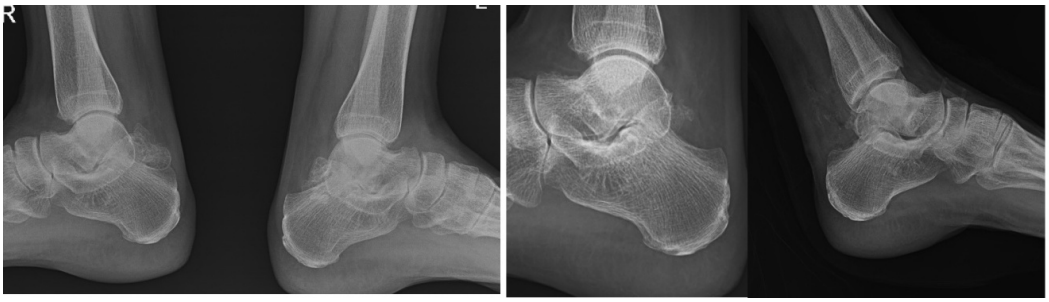

Fig. 5. Pre-operative and post-operative radiograph showing the bilateral calcaneal osteochondroma

3. DISCUSSION

Osteochondromas are chondrogenic lesions derived from aberrant cartilage, which can be solitary or multiple; sessile or pedunculated. The cortex and medullary cavity of the lesion is continuous with that of the native bone with an overlying cartilage cap.

Osteochondroma is the most common benign bone tumour, but exostosis around the foot and ankle are extremely rare and there have been no reports of bilateral exostosis in English literature. The first reports of a Talus exostosis were in 1947 by Robert Milch as osteochondroma of the astragalus and 1953 by Fredrick Rook as an

intra-articular osteochondroma of the astragalus [10,11].

In 2016 Suranigi et al reported a case of extensive osteochondroma of the talus presenting as tarsal tunnel syndrome [9]. Posterior ankle impingement caused by talus osteochondroma was reported in 2016 by Ercin et al [12]. Ozturk et all in 2019 presented a series of 10 cases of talus osteochondroma and found the anterior talus to be the most common site of the lesion [13].

MRI is the best method for visualization of structures surrounding the lesion, its effect on vascular and neural structures, complications (pseudoaneurysms, oedema) and the non-mineralized cartilage layer, which shows a high signal in T1 and high in T2, due to its water content, allowing these characteristics to adequate measure its thickness. MRI allows determinate the existence or absence of soft tissue involvement.

Open extra-periosteal resection of the of the lesion was the most common mode of management of these tumors. Kulkarni et al reported management of posterior talus osteochondroma with ankle arthroscopy [8].

Symptoms in cases of with osteochondroma is usually due to tendon or nerve irritation and mechanical block to motion. Extra-periosteal resection of the lesion with cartilage cap has been reported to be the most reliable method to prevent recurrence.

A giant Haglund's deformity caused by calcaneal osteochondroma has been reported in 2010 by Jung et al [14]. There are no other reports of a similar presentation in English literature. There are reports of calcaneal osterochondroma causing peroneal tendon impingement [15,16].

Haglund syndrome generally presents as posterior ankle pain associated with walking in shoes with a restrictive heel counter. It is often associated with retrocalcaneal bursitis and insertional Achilles' tendinitis / tendinopathy. It is often associated with an increase in calcaneal pitch secondary to a cavus foot, but our patient did not present with a cavus foot deformity.

Osteochondroma usually presents in the age group of 10-15yrs and gradually increases in size. The growth of the tumor typically halts with the closure of the physeal plates. There are isolated case reports of the same in literature by Krieg et all in [17] and Nogier et all in [18]. Koplay et al in 2009 reported a case of calcaneal osteochondroma with recurrence in a skeletally mature patient [19].

Both cases in our study presented with an increase in the size of the swelling and onset of symptoms in the sixth decade of life. The talar osteochondroma presented with peroneal tendinitis apart from the pain due to pressure on the surrounding structures. Excision gave complete relief of symptoms in our case. Bilateral presentation in the calcaneum and talus are rare. The calcaneal osteochondroma presented with Haglund spur like symptoms and they also got resolved after the excision.

In the talar osteochondroma case, being bilateral and benign pathology, we had the following as differential diagnosis: late presentation of epiphysealis hemimelica, hypertrophied ossicles, osteochondroma.

Though rare, osteochondroma should be considered as a differential diagnosis in adults presenting with swellings arising from talus and calcaneus. Presence of cartilage cap and Histopathological examination confirms the diagnosis. There was no recurrence in our cases.

4. CONCLUSION

Osteochondroma of the talus and calcaneum are rare pathologies and require a high index of suspicion for accurate diagnosis and timely management. Further studies are required to understand the pathophysiology of increase in the size of this tumor after skeletal maturity. Open extra-periosteal resection of the tumor remains the standard of care in these cases with good functional outcome and low recurrence rate.

CONSENT AND ETHICAL APPROVAL

As per international standard or university standard guideline participant consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Mercuri M, Casadei R. Tumours in the foot. Foot and Ankle Surgery2002;8:175–90.

2. Foo LF, Raby N. Tumours and tumour-like lesions in the foot and ankle. Clinical Radiology 2005;60:308–32.
3. Al Mutani M, Mahmood A, Chandrasekar CR. Giant osteochondroma of thetalar neck. Foot (Edinb). 2013;23:45–49.
4. Ozdemir HM, Yildiz Y, Yilmaz C, Saglik Y. Tumors of the foot and ankle: Analysis of196 cases. The Journal of Foot and Ankle Surgery. 1997;36:403–8.
5. Helm RH, Newman RJ. Primary bone tumours of the foot: experience of the Leeds bone tumour registry. The Foot. 1991;I:135–8.
6. Erler K, Oguz E, Komurcu M, Atesalp S, Basbozkurt M. Ankle swellingin a 6-year-old Boy with unusual presentation: Report of a rare case. J Foot Ankle Surg. 2003;42:235-239.
7. Giudici MA, Moser RP, Kransdorf MJ. Cartilaginous bone tumors. Radiologic Clinics of North America. 1993;31(2):237-260.
8. Kulkarni U, Kulkarni A. Posterior talus osteochondroma arare location, treated by posterior ankle arthroscopy. Foot Ankle Surg. 2015;21:e51-4
9. Suranigi S, Rengasamy K, Najimudeen S, Gnanadoss J. Extensive Osteochondroma of talus presenting as tarsal tunnel syndrome: Report of a case and literature review. Arch Bone Jt Surg. 2016;4:269-72.
10. Milch RA. Osteochondroma of the astragalus. Am J Surg. 1954;87:145–148.
11. Rook FR. Intra-articular osteochondroma of the astragalus. Am J Surg. 1953;85:807–810.
12. Ercin E, Bilgili MG, Gamsizkan M, Avsar S. Talarosteochondroma fracturepresenting as posterior ankle impingement. J Am Podiatr Med Assoc. 2016;106:225–228.
13. Öztürk R, Atalay İB, Yapar AE, Beltir G, Arıkan ŞM, Güngör BŞ. Talus localized osteochondromas: Treatment management and mid-term outcomes–Caseseries. Eklem Hastalik Cerrahisi. 2019;30(3):309-315.
14. Jung HG, Carag JA, Park JY, Bae EJ, Lim SD, Kim HS. Osteochondroma of the calcaneus presenting as Haglund's deformity. Foot Ankle Surg. 2011;17:e20–e22.
    DOI: 10.1016/j.fas.2010.08.007
15. Techner LM, De Carlo RL. Peroneal tubercle osteochondroma. J. Foot Surg. 1992;31:234.
16. Karakurum G, Oznur A, Atilla B. Bilateral peroneal tubercle osteochondroma of the calcaneus: case report. Foot Ankle Int. 1998;19(4):259–61.
17. Krieg JC, Buckwalter JA, Peterson KK, el-Khoury GY, Robinson RA. Extensive growth of an osteochondroma in a skeletally maturea patient: a case report. J Bone Joint Surg. 1995;77:269- 273.
18. Nogier A, De Pinieux G, Hottya G, Anract P: Case reports: Enlargement of a calcaneal osteochondroma after skeletal maturity. Clin Orthop Relat Res. 2006;447:260-266.
19. Koplay M, Toker S, Sahin L, Kilincoglu V. A calcaneal osteochondroma with recurrence in a skeletally mature patient: a case report. Cases J. 2009;2:7013.

Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sdiarticle4.com/review-history/67869