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World J Clin Cases 2017 September 16; 5(9): 349-372



MINIREVIEWS

- 349 Primary prevention of cardiovascular disease in older adults in China
Yong J, Lin D, Tan XR

CASE REPORT

- 360 Conservative approach to the acute management of a large mesenteric cyst
Leung BC, Sankey R, Fronza M, Maatouk M
- 364 Recurrent aneurysmal bone cyst of talus resulted in tibiotalar calcaneal arthrodesis
Vosoughi AR, Mozaffarian K, Erfani MA
- 368 Anti-N-methyl-D-aspartate receptor encephalitis that aggravates after acinetobacter baumannii pneumonia: A case report
Wang CC, Li DJ, Xia YQ, Liu K

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Recurrent aneurysmal bone cyst of talus resulted in tibiototalcaneal arthrodesis

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Abstract

Aneurysmal bone cyst (ABC), a locally benign aggressive lytic lesion of either primary or secondary origin, seldom involves the talus. Herein, we present a 25-year-old man with recurrent ABC of the talus after curettage and bone grafting, which was managed by total resection followed by filling the defect using fibular graft and finally tibiototalcaneal arthrodesis due to articular surface involvement. At 18 mo postoperatively, no recurrence was detected. Arthrodesis might be a good option in cases with recurrent ABC of the talus especially with articular surface involvement.

Key words: Aneurysmal bone cyst; Tibiototalcaneal arthrodesis; Bone tumor; Talus

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Core tip: Despite rarity of aneurysmal bone cyst (ABC) of the talus, we present a case of recurrent ABC of the talus following curettage and bone graft. The tibiotalar and subtalar joints of the patient were fused after complete resection of the tumor.

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INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign aggressive bone lesion composed of expansible blood-filled cavities. The incidence is 0.14 per 100000 of the population per year with a slight female predominance^[1]. Although all parts of human skeleton can be involved, ABC of foot bones and particularly the talus is very rare^[2,3]. Several primary ABCs of the talus^[2,4-6] and secondary ABCs on giant cell tumor^[7] and Chondroblastoma^[8] were described in the literature. Recurrence of ABC of the talus after curettage and bone grafting is extremely rare^[5,6]. Surgical treatment of these lesions is essential to reduce the pain of the patient and to prevent the possibility of pathologic fracture. In the present case, we describe a case of recurrent ABC talus with articular surface involvement which resulted in tibiotalocalcaneal arthrodesis.

CASE REPORT

A 25-year-old man presented with swelling and pain in left ankle joint without any history of acute trauma. He had experienced another episode of severe swelling and pain in left ankle area without any preceding trauma about 2 years before arrival. After definite diagnosis of ABC by another surgeon in another hospital, He had undergone curettage and autologous bone grafting *via* medial ankle approach and detachment of deltoid ligament.

Eight months after previous surgery, pain and swelling had begun again and continued till the time of second operation. On examination, swelling and tenderness on medial side was clearly evident. The neurovascular status of left ankle and foot was intact. Range of motion of ankle was limited to 25 degrees in plantar flexion and 10 degrees in dorsiflexion in comparison to the right ankle. Moreover it was painful particularly in full plantar flexion. Left ankle plain radiograph showed a cystic lesion in posteromedial of left talus with invasion to the articular surface of tibiotalar and subtalar joints (Figure 1). As ankle CT scan clearly approved the size and intra-articular invasion of the lesion (Figure 2), magnetic resonance imaging was not requested.

Surgery was performed under general anesthesia in the lateral position after inflation of a thigh tourniquet using sterile conditions. Through direct lateral approach to the lateral malleolus, fibular osteotomy from 10 cm above tip of fibula was done and completely resected. The articular surfaces of tibiotalar and subtalar joints were removed. The ABC was carefully resected because of adhesions to the posteromedial structure of the ankle joint. The pathologic report confirmed recurrent ABC. After curettage, the bone graft from the excised distal fibula bone was impacted in the arthrodesis site. The correct position of tibiotalar and subtalar joints for fusion was prepared and fixed by a Steinmann wire temporarily. Finally, the fixation was achieved utilizing a compression screw and a PHILOS plate



Figure 1 Anteroposterior (A) and lateral (B) radiographs of left ankle show a cystic-like lesion on posteromedial of ankle joint with destruction of articular surfaces.

on lateral side from calcaneus to the distal of tibia. Short leg cast was applied till complete union at 3 mo postoperatively.

Follow-up imaging at 18 mo after the operation showed no apparent sign of recurrence (Figure 3). The patient has a little claudication due to triceps surae weakness. He also suffers from occasional pain after long-distance walking.

DISCUSSION

ABC is a benign tumoral condition peaking at the first two decades of life^[1]. Although different treatment options are available, commonly surgical treatment of ABCs including curettage with or without bone grafting in addition to different adjuvants or wide *en-bloc* resection of tumor is preferred^[6,9-11]. Although the presented case was initially treated using curettage and bone grafting, recurrence was seen after 8 mo following the primary surgery. Recurrence is not uncommon and the reported rates are as high as 59%^[12]. Local recurrence is particularly higher among young age male patients^[13].

On the other hand, wide *en-bloc* excision is an excellent option with the lowest rates of recurrence (95%-100% localized control)^[14]. Moreover, it leads to reduce risk of future malignant transformations. Complete resection should be limited to expandable bones like distal ulna or proximal fibula. Complete resection in the other parts is associated with high morbidity to the patient. Resection of a huge ABC with involvement of articular surfaces in the talus could result in instability and functional compromise; therefore arthrodesis might be an acceptable option to diminish the possibility of recurrence, as done in the presented case.

Tibiotalocalcaneal arthrodesis could be done utilizing retrograde hindfoot intramedullary nails or different plate and screw constructs. Although intramedullary nails are good options with higher stability and lower soft tissue damage^[15], we prefer to use plate with screws because of more access to the lesion from lateral approach and surgeon experience



Figure 2 Sagittal (A), coronal (B), and axial (C) cuts of computed tomography scan in favor of tibiotalar and subtalar joint involvement.



Figure 3 Anteroposterior (A) and lateral (B) radiographs of left ankle shows the bone healing without any obvious recurrence at 18 mo following surgery.

for this technique.

This case approved that despite rarity of ABC in the talus, recurrence of ABC of the talus with articular surface involvement after curettage and bone grafting could be treated by en-bloc resection and consequent arthrodesis.

COMMENTS

Case characteristics

A 25-year-old male presented to the authors' outpatient clinic with swelling and pain in left ankle joint with positive history of aneurysmal bone cyst (ABC) of the talus.

Clinical diagnosis

Recurrent ABC of the talus.

Differential diagnosis

Unicameral bone cyst, bone malignancies.

Imaging diagnosis

Plain radiograph and computed tomography demonstrated a big cystic lesion in posteromedial of left talus with invasion to surrounding articular surfaces.

Pathological diagnosis

The histopathology report confirmed recurrence of ABC of the talus.

Treatment

Complete resection followed by bone grafting and tibiotalocalcaneal arthrodesis.

Related reports

There have been very rare case reports of recurrent ABC of the talus treated

using arthrodesis methods.

Term explanation

PHILOS plate is an anatomical locking compression plating system for proximal humerus fracture fixation.

Experiences and lessons

Early definite treatment of massive bone cysts in hindfoot is strongly encouraged to prevent inadvertent complications like pathologic fractures.

Peer-review

This is an interesting case report.

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