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Management of aggressive giant cell tumor of calcaneal bone: A case report



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ABSTRACT

INTRODUCTION: Prevalence of giant cell tumor (GCT) at atypical locations like bones of the feet are rare, seen in <1% of cases. GCT may have aggressive features, including cortical expansion or destruction with a soft-tissue component. Difficult diagnosis most often followed with complicated management and high recurrence rate remains a challenge that is rarely reported.

PRESENTATION OF CASE: We presented a case of forty-six-year-old male patient with giant cell tumor of the right calcaneus Campanacci 3 with secondary aneurysmal bone cyst (ABC). Wide excision total calcaneotomy, followed by reconstruction bone defect using femoral head allograft and soft tissue coverage with sural flap had been done.

DISCUSSION: Conservative surgery with careful curettage and placement of bone cement should be considered the treatment of choice when feasible. However, aggressive GCTs may require wide excision and reconstruction or may be amputation. We decided to do salvage surgery since: traditionally curettage is not possible, adequately wide resection of local tumor could be achieved, neurovascular bundle was not involved, and also bone and soft tissue reconstructions could be done. In addition, he refused for amputation.

CONCLUSION: Wide excision total calcaneotomy, bone allograft reconstruction and soft tissue coverage with sural flap is a good option for surgical management in aggressive GCT of calcaneus instead of amputation.

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1. Introduction

Giant Cell Tumor (GCT) of bone is a benign neoplasm which consists of mononuclear stromal cells and characterized by multinucleated giant cells that exhibit osteoclastic activity. The tumor is typically appeared as an eccentric lytic lesion with a well-defined but non-sclerotic margin which can extend near the articular surface. However, GCT may have aggressive features, including cortical expansion or destruction with a soft-tissue component. Fluid-fluid levels, consistent with secondary formation of aneurysmal bone cyst (ABC), are seen in 14% of cases. GCT can mimic or be mimicked by other benign or malignant lesions at both radiologic evaluation and histologic analysis [1,2]. Most GCTs occur in the long bones of the lower extremity especially around the knee [1,3]. The bones of

the feet are considered atypical locations which is extremely rare and tumor diagnosis in these circumstances is often confusing [2,3].

The treatment of bone GCT remains a challenge, since there are no clinical, radiographic or histological aspects that allow one to accurately predict the trend of a single lesion to recur or to metastasize. Enneking's and Campanacci's classifications are helpful in planning the initial surgical treatment. Traditionally, GCT of bone has been treated surgically with curettage and placement of cement (polymethyl-methacrylate). However, the recurrence rates have been relatively high, ranging from 15% to 25% [2]. The trend of treatment in aggressive GCT cases, is heading towards limb salvage and amputation is reserved for recurrences and only rarely done [1,2]. In this report, we presented a case of 46-year-old male who was diagnosed with GCT of the right calcaneus Campanacci 3 with secondary aneurysmal bone cyst and was treated in our center by wide excision, bony reconstruction using femoral head allograft and sural flap.

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Fig. 1. Clinical pictures of the right heel showed a 7 × 10 × 6 cm lobulated mass over the right heel: (A) Lateral view. (B) Posterior view.



Fig. 2. A: lateral view radiograph of ankle showed a radiolucent lesion occupying the right calcaneus. B: magnetic resonance imaging was consistent with solid mass derived from calcaneal bone expanding to the soft tissue. C: there was small area of cystic component, most probably secondary aneurismal bone cyst.

2. Presentation of case

A 46-year-old male presented with right heel lump. The lump had been existed since 8 months prior and initially the size was as small as a marble, soft and painless. The patient did not have any history of trauma or infection. There was no history of such case or any mass before in his families. He went to a local general practitioner, who managed the patient conservatively for two weeks. After 4 months the lump was still persistent and he went to internist in another general hospital. He was told that the lump was due to

the uric acid and was prescribed uric acid-lowering drugs and at the follow up the uric acid level had decreased, but the lump still did not show improvement. Then, he consulted to a general surgeon and diagnosed as bone tumor. An open biopsy then was performed and the histopathologic examination showed to be GCT of tendon sheath. Unfortunately, the mass was getting bigger and became fungating through the biopsy tract, so the patient was difficult to walk. Subsequently, he was referred to Cipto Mangunkusumo Hospital.

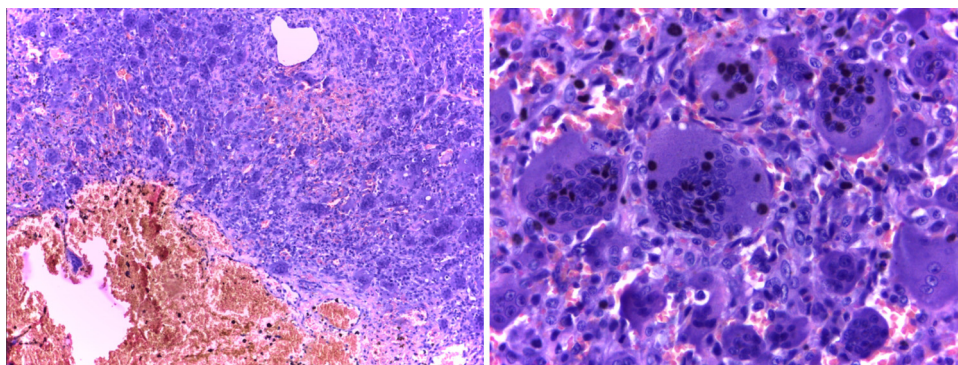


Fig. 3. The histologic section reveals tumor consist of numerous multinucleated giant cells, and area of secondary aneurysmal bone cyst (HE, 40 \times) B. The multinucleated giant cells contain nuclei showing similar feature to those of mononuclear stromal cells (HE, 400 \times).

On physical examination, there was a 7 \times 10 \times 6 cm fungating lobulated mass over the right heel (Fig. 1) which was tender (visual analog scale of 2–3) on palpation. Movements of the ankle joint were painful and restricted. Ankle radiograph showed an expanded osteolytic lesion in the right calcaneus (Fig. 2A). It was characterized as a grade 3 lesion according to Campanacci grading. Magnetic resonance imaging was consistent with solid mass derived from calcaneal bone expanding to the soft tissue. There was small area of cystic component, most probably secondary aneurysmal bone cyst (Fig. 2B, C).

The previous biopsy was reviewed by our pathologist which was consistent with GCT of bone with secondary ABC (Fig. 4A, B). Limb salvage surgery was performed by our oncology-orthopaedic consultant using wide excision, allograft reconstruction and sural flap. Patient was previously consented to the procedure and possible risk of surgery including the possibility of intraoperative amputation and a high risk of recurrences. After wide excision of GCT of calcaneal bone with surrounding soft tissue had been done, proximal femoral (neck and head) allograft was inserted into the defect created earlier and fixed into talus by screws. Subsequently, Achilles tendon lengthening was performed. We made new insertion of Achilles tendon into femoral head allograft. Soft tissue defect was covered by sural flap (Fig. 3). A below knee cast was applied for 6 weeks immobilization.

Five months after the surgery, there was no local recurrence and patient had good clinical and radiological appearance. He was able to walk partial weight bearing with axial crutches (Fig. 5). One year after surgery, he was able to walk full weight bearing. Upon asked, patient claimed to be satisfied with the management because he had no amputation and could walk with this kind of surgery.

3. Discussion

Giant cell tumor commonly occurs in patients with closed physes, or skeletally matured [5]. Seventy five to ninety percent of the cases occur on long bones, with majority found around the knee. The most common site of GCT is on the distal femur, proximal tibia, and distal radius [2]. GCT of calcaneal bone of the feet is very uncommon, with prevalence only less than 2% [6]. Campanacci *et al.* reported only 2 cases of GCTs in the calcaneus out of the total of 327 and Dahlin reported 4 out of 411 cases in his study [3,7].

GCT of the calcaneus and other small bones of the feet and hand clinically showed non-specific symptoms such as local swelling (mass) and local pain. Therefore, many cases may be mismanaged as ankle sprain, infection or other disease. A history of preceding trivial injury may be present [8]. Initially, our patient had those symptoms, and diagnosed as inflammatory disorder. However, the mass was

acting like a benign aggressive tumor and fungating (more aggressive) after open biopsy procedure. This is the reason why the patient was referred to our hospital.

Normally, should GCT presented in the common age of the patient and location, it can be best identified with conventional radiograph. It shows a well-defined lesion with non-sclerotic margin. The location is eccentric, and extends to the nearest subchondral bone of articular surface [5,9,10]. The aggressive features may include wide zone of transition, cortical thinning, expansile remodeling, or even cortical bone destruction, with an associated soft-tissue mass [5]. Additionally, it may also contain fluid-fluid levels due to secondary ABC formation which may occur in up to 14% cases of bone GCT [11]. It may be differentiated from the primary ABC by the presence of an enhancing soft tissue component. Hemorrhage within cystic spaces may give an appearance of fluid-fluid levels on radiographic examination [2].

The treatment of GCT of calcaneal bone remains a challenge, because there are no clinical, radiographic or histological aspects that allow one to accurately predict the trend of a single lesion to recur or to metastasize. Enneking's and Campanacci's classifications are helpful in planning the initial surgical treatment. Conservative surgery with careful curettage and placement of bone cement should be considered the treatment of choice when feasible [4]. However, aggressive GCTs may require wide excision and reconstruction or may be amputation [1,2]. In this case, we performed wide excision, bone and soft tissue reconstructions. We decided to do salvage surgery since traditionally curettage was not possible, adequate wide resection of local tumor could be achieved, neurovascular bundle was not involved, and also bone and soft tissue reconstructions could be done. In addition, patient refused for amputation.

Osteochondral allograft reconstruction has also been described for an aggressive GCT of talus but there is paucity of literature on this particular modality of treatment. Moreover, to fill the large bone defect in the calcaneal bone, we used proximal femoral allograft. To date, there are many options for coverage, including distally based fascio-cutaneous flap, muscle flaps, septo-cutaneous flaps, axial flaps, local transposition flaps and free flaps. In this case, we opted to perform sural flap to close soft tissue defect after tumor resection. The superficial sural artery flap is one of the recently introduced therapeutic modality described by Masquelet and colleagues in 1992. The superficial sural artery based island flap has many advantages. The important advantage of the distal sural flap is that the blood supply is reliable, making this flap safe, even in patients with distal arterial insufficiency and there is no sacrifice of major arteries or nerves. The superficial sural island flap is a good choice to the management of exposed Achilles tendon. It has wide

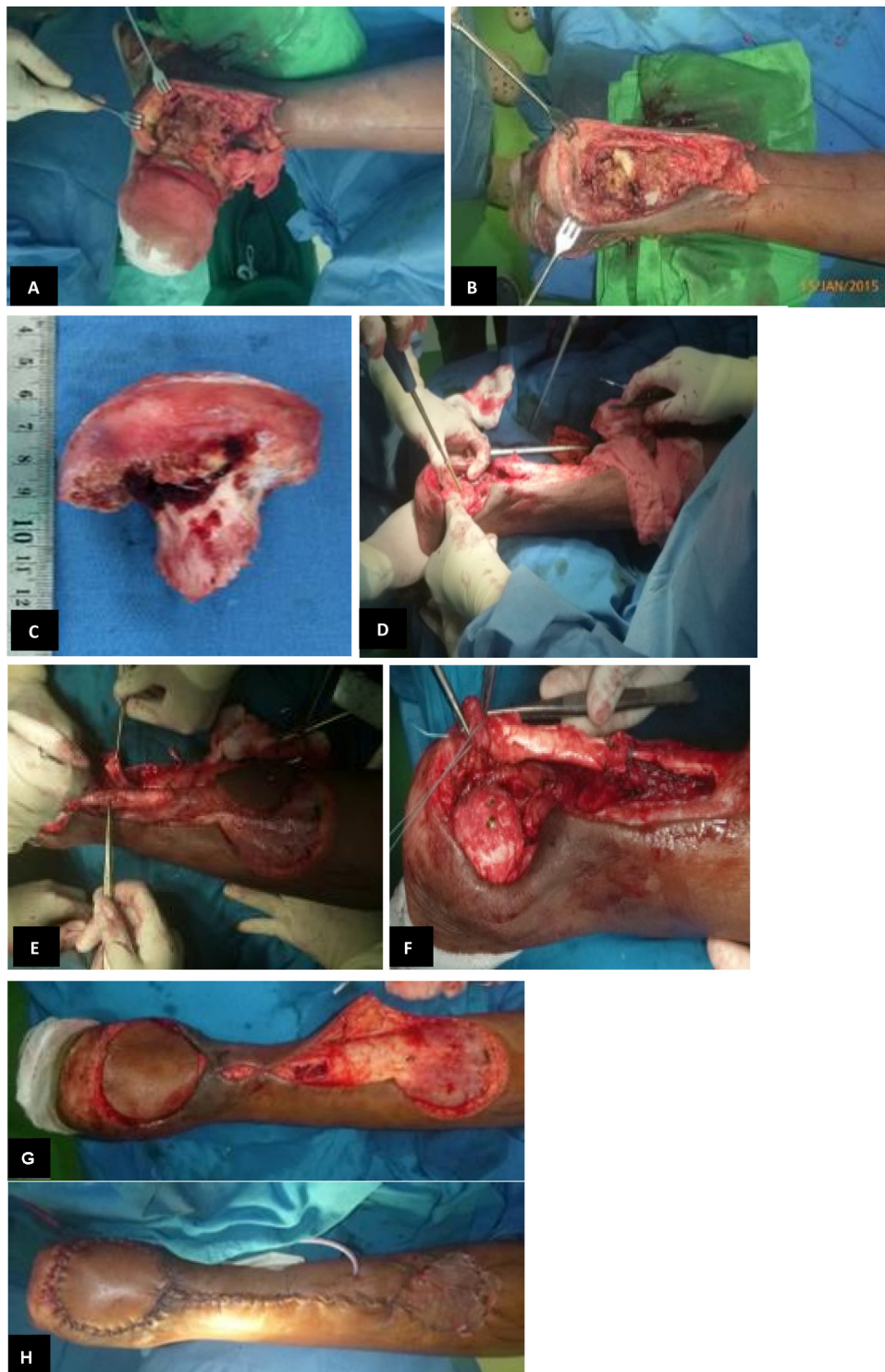


Fig. 4. The pictures showed surgical procedures from wide excision to bone and soft tissue reconstruction. (A) tumor excision; (B) post excision of the tumor; (C) femoral head allograft; (D) femoral head insertion to change the calcaneus; (E) achilles tendon lengthening; (F) achilles tenodesis; (G) soft tissue coverage with sural flap; and (H) complete surgery.

range of arc of rotation 180° for Achilles tendon coverage and is easy to perform. The success rate is high and minimal flap loss and other complications. Because this flap is fasciocutaneous, its durability is excellent, even in weight-bearing areas at the back of the heel on tendo-Achillis. The under surface of the flap provides a good surface for gliding of the tendon [12].

However, this flap has some limitations like maximum safe length-breadth ratio yet to be defined. There are no studies regarding maximum flap dimension (specifically, width) and safety, but usually a relatively large flap can be harvested with little donor site deformity or morbidity. Another disadvantage of this flap are the sacrifice of the sural nerve and the final scar, mainly when there



Fig. 5. Five months follow up after the surgery, (A) Clinical pictures (B) Radiographic pictures.

is a need for skin grafts to close the donor area. This is important particularly in women. When the donor area is closed directly, the final result is aesthetically more acceptable [13,14]

4. Conclusion

In conclusion, limb salvage surgery which consists of wide excision total calcaneotomy, bone allograft reconstruction and soft tissue coverage with sural flap is a good option for surgical management in aggressive GCT of calcaneus instead of amputation.

Conflict of interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Sources of funding

The authors declare that sponsors had no such involvement.

Ethical approval

The patient received an explanation of the procedures and possible risks of the surgery, and gave written informed consent.

Author contributions

AFK contributed to perform the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval

for publishing; AW contributed to assist the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval for publishing; ZE contributed to assist the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval for publishing; EK contributed to data collection, analysis and interpretation of histopathology, manuscript drafting, revising, and approval for publishing.

Guarantor

AFK is the Guarantor.

References

- [1] A. Bhattacharyya, R. Das, R. Darwani, Giant cell tumor of the talus: a case report and review of literature, *Foot Ankle Online J.* 3 (8) (2010) 2.
- [2] C.J. Chakraborty, D.M. Forrester, C.J. Gottsegen, D.B. Patel, E.A. White, G.R. Matcuk Jr., Giant cell tumor of bone: review, mimics, and new developments in treatment, *Radiographics* 33 (1) (2013) 197–211.
- [3] D. Gotecha, N. Bhalerao, N. Gadre, U. Pote, Giant cell tumor of the calcaneus: a case report, *Int. J. Sci. Study* 3 (2) (2015).
- [4] C. Errani, P. Ruggieri, M.A. Asenzio, A. Toscano, S. Colangeli, E. Rimondi, et al., Giant cell tumor of the extremity: a review of 349 cases from a single institution, *Cancer Treat. Rev.* 36 (1) (2010) 1–7.
- [5] M.D. Murphey, G.C. Nomikos, D.J. Flemming, F.H. Gannon, H.T. Temple, M.J. Kransdorf, From the archives of AFIP. Imaging of giant cell tumor and giant cell reparative granuloma of bone: radiologic-pathologic correlation, *Radiographics* 21 (5) (2001) 1283–1309.
- [6] R. Biscaglia, P. Bacchini, F. Bertoni, Giant cell tumor of the bones of the hand and foot, *Cancer* 88 (9) (2000) 2022–2032.
- [7] M. Campanacci, N. Baldini, S. Boriani, A. Sudanese, Giant-cell tumor of bone, *J. Bone Joint Surg. (Am.)* 69 (1) (1987) 106–114.
- [8] S. Sharma, I.H. Wani, N. Gupta, N. Mahajan, Salaria AQ. Giant cell tumor of talus: a case report, *Cases J.* 2 (2009) 74.
- [9] R.E. Turcotte, Giant cell tumor of bone, *Orthop. Clin. N. Am.* 37 (1) (2006) 35–51.

- [10] R.T. Arnold, M.T. van Holsbeeck, T.G. Mayer, M.P. Mott, S.R. Koch, Best cases from the AFIP: Necrotic giant cell tumor of bone manifesting with pathologic fracture, *Radiographics* 31 (1) (2011) 93–98.
- [11] C. Anchan, Giant cell tumor of bone with secondary aneurysmal bone cyst, *Int. J. Shoulder Surg.* 2 (3) (2008) 68.
- [12] J. Tharayil, R.K. Patil, Salvage of foot with extensive giant cell tumour with transfer of vascularised fibular bone graft, *Indian J. Plast. Surg.* 44 (1) (2011) 150–156.
- [13] M. Alam, M. Shaheen, S. Hossain, S. Anam, S. Rahman, Sural island flap – a good option for coverage of the exposed heel (tendoachillis), *J. Dhaka Med. Coll.* 19 (1) (2010) 19–24.
- [14] M.A. Kalam, S.R. Faruquee, S.K. Karmokar, P.B. Khadka, Superficial sural artery island flap for management of exposed Achilles' tendon – surgical techniques and clinical results, *Kathmandu Univ. Med. J.* 3 (4) (2005) 401–410.

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