



Case report

Giant cell tumor with secondary aneurysmal bone cyst of the left calcaneus

Achmad Fauzi Kamal, Fahresa Hilmy*

Department of Orthopaedics and Traumatology, dr. Cipto Mangunkusumo General Hospital, Faculty of Medicine Universitas Indonesia, Jakarta, Indonesia

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ABSTRACT

Introduction and importance: Giant cell tumors (GCT) of the feet bones are rare, comprising of <1 % of cases. The lack of well-documented cases and similarity with other tumors under radiologic and histological evaluation makes diagnosis difficult. Current treatment modalities for GCT still result in a relatively high recurrence rate, making the overall management of the case a challenge. We reported a 27-year-old male diagnosed with GCT of the left calcaneus with secondary aneurysmal bone cyst (ABC) treated with curettage and femoral head allograft combined with bone cement application.

Case presentation: A 27-year-old male presented with lump on the left heel since seven months before admission. Physical examination demonstrated solid, palpable mass on the left heel region and limited ankle motion. Magnetic resonance imaging examination demonstrated expansile bone tumor at the left calcaneus with cystic components building fluid levels and hemorrhagic components, suggestive of giant cell tumors with secondary ABC. The patient is diagnosed with giant cell tumor with secondary ABC of the left calcaneus Campanacci grade 2. The patient was managed limb salvage surgery by curettage and subsequent mix of femoral head allograft and bone cement application to fill the defect.

Discussion: Conservative surgery via careful curettage is typically preferred for lower Campanacci grade lesions followed by bone reconstruction. In terms of filling bone defects, it is known that both bone cement and allografts have advantages and disadvantages. We hence decided to perform limb salvage surgery via curettage due to the size of the tumor and bone reconstruction using a mix of femoral head allograft and bone cement to fill the defect.

Conclusion: Curettage and bone allograft with bone cement reconstruction is an option for surgical management of lower Campanacci grade 2 GCT of the calcaneus.

1. Introduction

Giant Cell Tumors (GCT) of bone represent approximately 5 % of all bone tumors. GCTs generally are benign tumors histologically comprised of multinucleated giant cells with osteoclastic activity and mononuclear stromal cells as background [1,2]. Despite their benign nature, GCTs are known to express aggressive features such as cortical expansion, destruction, or recurrence after surgical resection [3]. GCTs typically arise in epiphyseal locations (90 %), rarely invade joint spaces, and have a well-documented predilection for the knee region, particularly the distal femur and proximal tibia [4,5]. GCTs of the feet bones however are relatively rare (observed in <1 % of cases), often making the diagnosis in these situations difficult [2,6].

Moreover, GCTs are known to demonstrate vague characteristics that would make them difficult to differentiate from other benign or malignant masses under histological analysis or radiologic evaluation [2,4].

Classification of GCT is usually done using the Campanacci grading, generally grouping the tumors into 3 grades (latent, active, and aggressive) based on radiological findings [4]. The goals of treatment for GCT are lower local recurrence and function maintenance, with various studies favoring intralesional curettage based on the good preservation of function [7–9]. Despite a relatively high rate of local recurrence compared to wide resection, modern imaging technologies and techniques of curettage can improve the surgical outcomes [10–12]. The procedure is typically followed by the application of a bone graft to fill the newly-formed cavity [4].

However, the rate of local recurrence despite these surgical efforts still ranges between 25 and 50 %, leading clinicians to implement a variety of adjuvants, some of which have proven beneficial in suppressing recurrence rates [13–16]. In this report, we illustrate the case of a 27-year-old male with Campanacci grade 2 GCT of the left calcaneus with a secondary aneurysmal bone cyst (ABC), who was treated in our

* Corresponding author.

E-mail address: dr.hilmy.penelitian@gmail.com (F. Hilmy).<https://doi.org/10.1016/j.ijscr.2022.107697>

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Fig. 1. Clinical pictures of the left foot revealed a solid palpable mass over the left heel, with an unclear margin and a circumferential size of 35 cm (contralateral 31 cm).

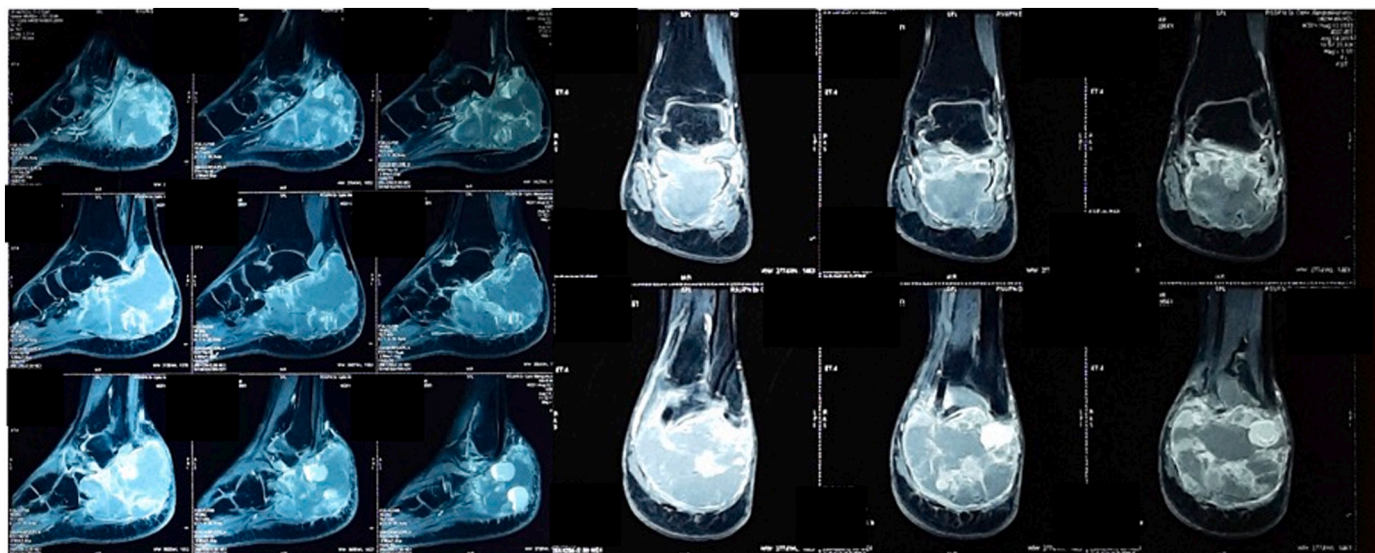


Fig. 2. Magnetic resonance imaging of the left calcaneus demonstrates an expansive bone tumor with fluid-fluid level and hemorrhagic component, indicating a diagnosis of GCT with secondary ABC. There was no talocrural or neurovascular involvement.

hospital via intralesional curettage followed by ethanol 96 % plus H₂O₂ 3 % femoral head allograft-bone cement composites. This case report has been reported in line with the SCARE Criteria [17].

2. Case presentation

A 27-year-old male presented with a lump on his left heel since 7 months prior to admission. The lump was initially the size of a chicken egg (4 cm in diameter) and painless. 1 month prior to admission, the patient's heel started to experience pain which was exacerbated with activity and did not subside with rest. At his local hospital, the patient was diagnosed with bone tumor based on radiologic findings and was referred to our hospital. He had a history of trauma about a year prior to admission, involving falling from a considerably height and landing on his heels. The patient received casting from an orthopaedic surgeon and was instructed to walk on a crutch. Instead, he proceeded to receive traditional massage three times. There was no other history of previous medication and family history of similar condition.

Physical examination of the left foot (Fig. 1) revealed a solid palpable mass over the left heel, with an unclear margin and a circumferential size of 35 cm (contralateral 31 cm), which was tender upon palpation (with a Visual Analog Scale of 2–3), classified as a grade 2 lesion based on Campanacci grading. Magnetic resonance imaging (MRI) of the mass demonstrated an 8.6 × 6.6 × 7.6 cm expansive bone tumor of the left

calcaneus with a cystic component, indicating GCT with secondary ABC as seen in Fig. 2. No talocrural intraarticular or neurovascular was involvement (Fig. 3).

Core biopsy revealed a diagnosis consistent with GCT of bone with secondary ABC. After patient consent was acquired, an extended curettage of the calcaneal mass was performed, followed by the application of ethanol 96 % plus H₂O₂ 3 % and femoral head allograft-bone cement composites and a below knee cast was applied for a 6-week-long immobilization. Radiology imaging 3 months after the operative procedure is shown in Fig. 4.

After the follow-up 19 months period, there was no sign of recurrence. The patient was also able to walk normally and was satisfied with his current condition. Radiographic examination in 19-months follow-up was shown in Fig. 5.

3. Discussion

GCT of bone is a generally benign tumor with the potential for recurrence and malignant transformation in about 1.5–13 % of cases, with the primary location of metastasis to the lungs [18]. GCT is most commonly found in the metaphysis and epiphysis of long bones, particularly the distal end femur and proximal end tibia, with a prediction for skeletally mature patients and a male:female ratio of 1:1.5 [4,19].

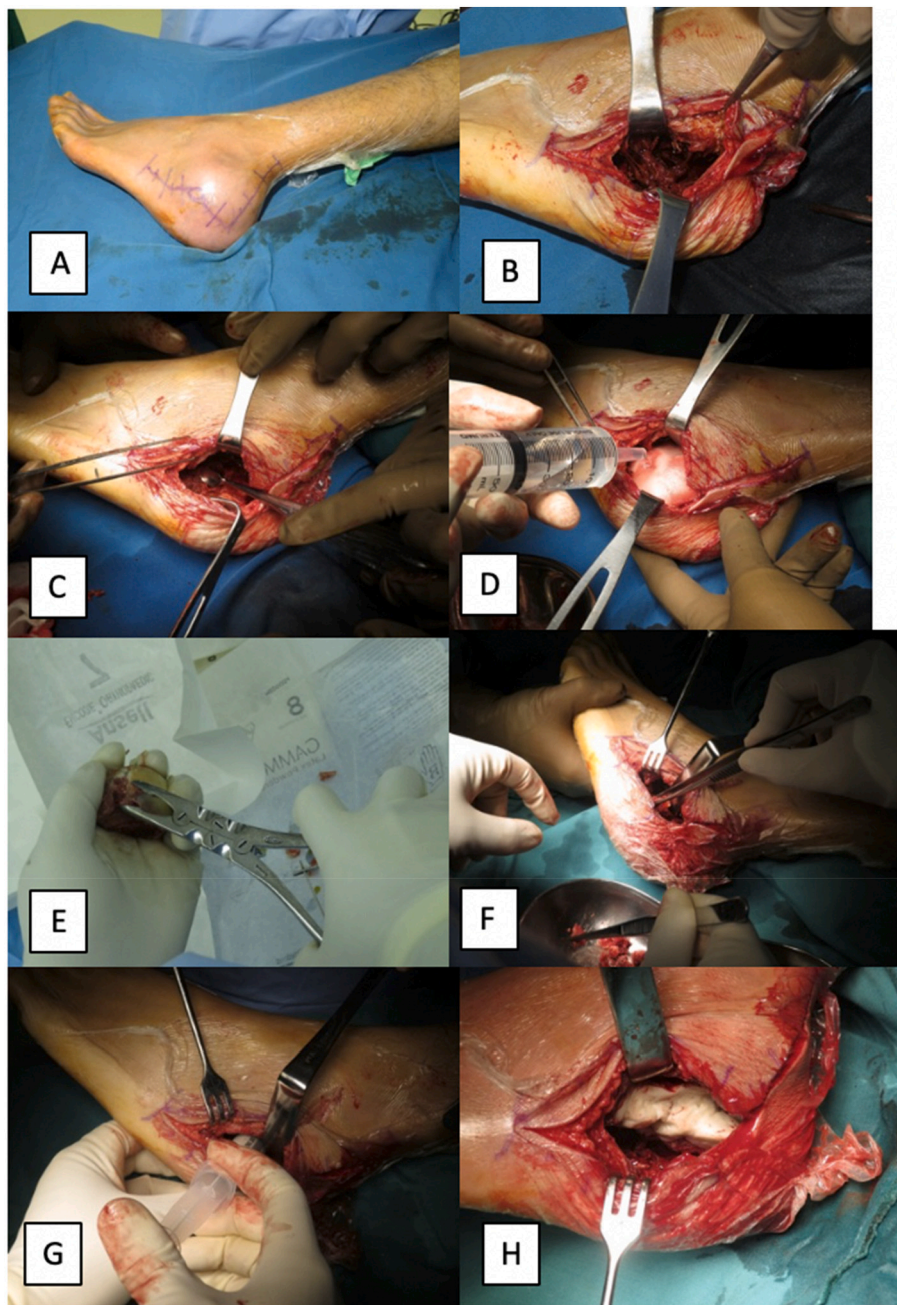


Fig. 3. Intra-operative documentation showing procedures from pre-operative markings to tissue reconstruction. (A) Pre-operative markings; (B) tumor exposure; (C) curettage; (D) extended curettage; (E) bone graft (femoral head allograft) preparation; (F) bone graft application; (G) bone cement application; (H) final tissue reconstruction.

Typical clinical presentations of GCT include pain related to mechanical insufficiency due to bone destruction, an observable soft tissue mass derived from tumor progression out of the bone. Due to its predilection near joints, GCT commonly presents with limited range of motion in the adjacent joint and more rarely, joint effusion and synovitis [4]. Upon diagnosis, pathologic fracture is usually found in 12 % of GCT patients, a feature thought to indicate a more aggressive disease with an increased risk of recurrence and metastatic spread [20,21]. However, GCTs of the feet are rare, and the clinical presentations of pain and swelling in this area are often confused with other, more common, etiologies such as ankle sprains, given that a history of trivial trauma in the feet is not an uncommon finding [22].

Radiologic examination of GCT commonly shows a lytic lesion with a well-defined but non-sclerotic margin, extends to the subchondral bone,

is eccentric in location, and occurs in patients with closed physes [9]. Some aggressive features that may be present include a wide transition zone, cortical thinning or destruction, expansile remodeling, and an associated soft tissue mass [23]. Histologic analysis would typically show giant cells with a benign spindle cell background [4].

Treatments of GCT should address local recurrences and function maintenance. The preferred technique for most GCT cases is curettage. Even though other techniques such as wide resection has lower local recurrence rate, recent studies of extended curettage with modern imaging techniques have shown an improved recurrence rate [7–9]. In our patient, after a 19-months follow-up, there was no sign of recurrence. The key to successful intralesional curettage is complete removal with adequate exposure to the lesion, this can be achieved by creating a large cortical window for tumor access. Failure in intralesional curettage may

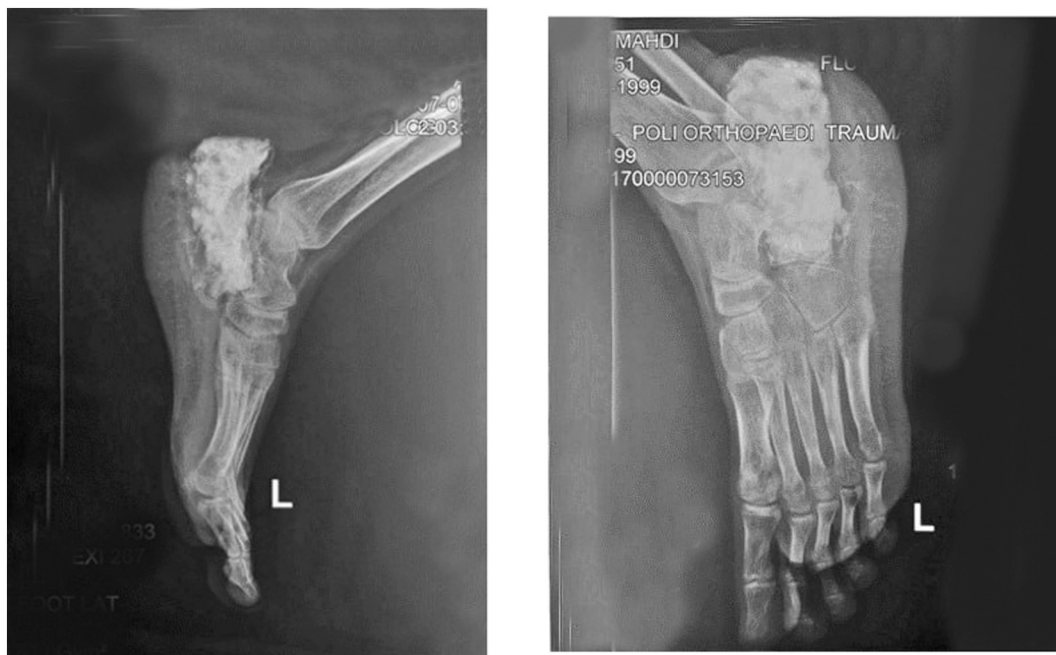


Fig. 4. Radiology imaging 3 months after operative procedure. No signs of local recurrent tumor, fracture, or operative site infection.



Fig. 5. X-ray of digits after 19-months follow-up.

occur due to limited exposure to the lesion, which results in inadequate tumor removal. A study conducted by Trieb et al. further explained that adequate removal of the tumor is the sole predictor of recurrence rate in primary curettage [10–12].

Other than recurrence rate, functional outcome is also important in evaluating the patient's condition postoperatively. In our patient, after a

19-months follow-up, the patient was able to walk. A similar result was also reported in a study conducted by Kamal et al., in which a patient with the previous GCT of calcaneal bone treated with limb salvage surgery was able to walk full weight bearing after 1 year postoperatively.

Conservative surgery comprised of carefully extended curettage and

application of bone graft/bone cement or combinations both should always be considered when feasible, and wide resection is usually reserved for grade 3 Campanacci or aggressive lesions [24,25]. Not all cases of excision would require bone reconstruction. Small cavities have been known to fill up with blood clots that eventually ossify to form bone without additional intervention. Larger defects have several options including cementation or the use of bone grafts, each with its own advantages [4]. Bone cements have been known to produce a thermal effect which aids in destroying surrounding neoplastic cells. Bone cements can also be added with local cytotoxic agents such as methotrexate and adriamycin in an attempt to reduce local recurrence. Additionally, detection of recurrence is easier with bone cement [26]. However, bone cement is not a biological material, hence it is relatively weak towards shear and torsional forces, causing fractures through the cement when used in areas such as the head and neck of the femur [26]. Also, bone cement has been associated with the risk of damage to adjacent cartilage, causing articular degeneration with biomechanical changes [4].

Bone grafting, on the other hand, undergoes remodeling along stress lines, is more durable towards different types of forces, and is a more permanent form of reconstruction. The disadvantages of bone graft are due to the nature of its procurement, its quantity is usually both limited and expensive, and it causes donor site morbidity [26]. Using a mixture of bone grafts (auto and allograft) with bone cement has been reported to reduce the risk of articular degeneration of bone cement. Bone grafts are packed near subarticular tissue, with a gel foam layered over this. Bone cement is applied to the remaining cavity. This approach aims on reducing heat damage from the applied bone cement and limit articular complications from reconstruction using bone cement [4].

Another modality of treatment has also been used in GCT, such as chemotherapy, radiotherapy, or embolization. Although GCT has been shown to have a profound response to chemotherapy, there is still controversy regarding the use of chemotherapy in GCT. Radiotherapy is recommended in patients with aggressive tumors. Embolization is usually performed in unresectable GCTs, such as sacral or pelvic tumors, in which transcatheter embolization is applied to stop the tumor blood supply. Embolization is usually performed in monthly intervals aiming for significant clinical improvement such as pain. Embolization can be performed preoperatively to reduce blood loss during surgical intervention. Newer evidence also introduced the use of bisphosphonates, due to their osteoclastic characteristic that may limit tumor progression. However, further studies need to be conducted [4,5].

In our case, we performed extended curettage, bone and soft tissue reconstructions were possible, allowing us to apply a mix of bone allograft and bone cement to fill the defect.

4. Conclusion

In conclusion, conservative curettage, followed by local adjuvant, bone allograft application combine with bone cement is effective and safe to manage lower Campanacci grade 2 of giant cell tumors of the calcaneus without sacrificing joint function.

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Ethical approval

This case report did not intervene with patients' treatment plans and hence did not require ethical approval.

Author contribution

Achmad Fauzi Kamal: study concept or design, data collection, analysis and interpretation, oversight and leadership responsibility for

the research activity planning and execution.

Fahresa Hilmy: study concept or design, data collection, analysis and interpretation, oversight and leadership responsibility for the research activity planning and execution.

Guarantor

Achmad Fauzi Kamal, MD.

Research registration number

N/A.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

Nothing to declare.

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