Case Report

A rare occurrence of distal fibular giant cell tumor in adolescent: a case report

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ABSTRACT

Introduction: Most Giant Cell Tumors appear at epiphyseal area, predominantly at knee area or wrist. It is usually affecting people around the third to fourth decades, rarely below the twenties or above the sixties. The mainstay of treatment is surgery, purposely to eliminate the tumor, restore the function as near to normal. Diagnosis can be made by clinical, radiological decision and histopathology, yet the only gold standard is histopathology.

Methods: We present a rare case of giant cell tumor on the right distal fibula, in a female of eighteen years old. The clinical and radiological appearances were not pathognomonic, ultimately needing a biopsy to finally confirm it was a giant cell tumor indeed. A curettage was done, whilst the defect filled with bone substitutes and supported by a plate.

Results: Histopathology study after the surgery revealed the tumor was indeed a giant cell tumor, in accordance to microscopic and macroscopic findings. The patient was advised to not weight-bear for another 3 months, and partial weight-bear for the next 6 until 12 months. Diagnosing a giant cell tumor might be challenging, especially if the symptoms are not obvious, epidemiologically inconclusive, and radiologically insatiable. A biopsy will be needed, but a coherence with clinical judgement is advised. Finally, the proposed excision and reconstruction options should coincide with the need of functionality of the patient.

Conclusion: Given certain circumstances, it is hard to diagnose a giant cell tumor. A careful approach should be kept in mind, malignancy should be ruled out as soon as possible.

ABSTRAK

Pendahuluan: Giant cell tumor *paling banyak muncul di area epifisis, terutama di daerah lutut atau pergelangan tangan. Hal ini biasanya mengenai orang yang berusia sekitar dekade ketiga hingga keempat, dan jarang mengenai orang yang berusia di bawah dekade kedua atau di atas dekade keenam. Terapi utamanya adalah pembedahan, yang bertujuan untuk menghilangkan tumor dan mengembalikan fungsi mendekati normal. Diagnosis dapat dibuat berdasarkan keadaan klinis dan radiologis, tetapi pemeriksaan histopatologi adalah standar baku emas.*

Metode: *Kami menyajikan kasus langka* Giant cell tumor *di distal fibula kiri, pada wanita berusia delapan belas tahun. Penampakan klinis dan radiologis tidak patognomonik, tetapi pada akhirnya dibutuhkan biopsi untuk memastikan bahwa itu memang* Giant cell tumor. *Kuretase dilakukan terhadap pasien, sedangkan defek diisi dengan tulang pengganti dan diaugmentasi dengan plat.*

Hasil: *Studi histopatologi setelah pembedahan mengungkapkan tumor itu memang* Giant cell tumor*, sesuai dengan temuan mikroskopis dan makroskopik. Pasien disarankan untuk mobilisasi dengan tidak menahan beban selama 3 bulan, dan selanjutnya menahan beban sebagian selama 6 sampai 12 bulan ke depan. Mendiagnosis* Giant cell tumor *mungkin sulit dilakukan terutama jika gejalanya tidak jelas, secara epidemiologis tidak dapat disimpulkan, dan secara radiologis tidak memuaskan. Biopsi akan diperlukan, tetapi koherensi dengan penilaian klinis tetap disarankan. Akhirnya, pilihan eksisi dan rekonstruksi yang diusulkan harus sesuai dengan kebutuhan fungsionalitas pasien.*

Kesimpulan: *Mengingat keadaan tertentu, sulit untuk mendiagnosis* Giant cell tumor*. Pendekatan yang hati-hati harus selalu diingat, dan penyakit ganas harus disingkirkan sesegera mungkin.*

Keywords: Giant cell tumor, distal fibula, excision, reconstruction

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INTRODUCTION

Giant cell tumor of bone is one type of giant cell-rich lesion of bone. Most GCTs are located in the epiphyseal regions of long bones (distal femur, proximal tibia and distal radius). GCT of bone typically presents in persons aged 20 to 40 years. It is rare in adolescents and children, and <10% of cases are seen in patients >65 years old.[1] Patients with GCT typically present with pain. Physical examination usually reveals an area of direct tenderness to palpation, soft tissue swelling over the affected area, and the presence of either sympathetic or direct joint effusion. Radiographically, GCT of bone manifests as a large, purely lytic mass that frequently extends from the subchondral bone plate into the metaphysis and epiphysis. GCT may also have aggresive features, such as a wide zone of transition, cortical thinning, expansile remodeling, or even cortical bone destruction and an associated soft-tissue mass.[2] Depending on the extent of disease, the primary treatment of GCT is surgery. When surgery is not possible or would be associated with unacceptable toxicity, treatment with denosumab or radiation therapy may be useful.[3]

First described by Sir Astley Cooper in the year 1818, giant cell tumour of bone or osteoclastoma is the commonest benign bone tumour, It typically involves the epiphyseometaphyseal region of long bones. The commonest age is the 3rd or the 4th decade with a slight female predominance.[1]

Knee is the commonest site, followed by distal radius. The other less common sites are sacrum, distal tibia, proximal humerus, proximal femur and proximal fibula. Radiographically, GCT characterised as a lytic lesion occurring in the ends of bones. Current treatment modalities including meticulous curettage with extension of tumour removal using high speed burrs and adjuvant local therapy has significantly lowered the recurrence rates to less than 10% from 60% in the past with curettage alone. The incidence of giant cell tumour of distal fibula was found to be less than 1% of 1182 cases.[1]

The ankle joint is a complex and formed by lower end fibula, tibia, and talus along with complex ligamentous restraints. Lower end fibula forms an important component of ankle mortise. Any pathological lesion or ensuing deformity in lower fibula can lead to an unstable ankle resulting into lifelong debility. The management of any lesion in the lower end of fibula needs to address both the primary pathology management and restoration of the ankle joint for an adequate functional outcome. More than 50% of GCT occur around the knee joint. The upper end of the fibula is more commonly involved, 2.5% greater than the distal fibula. The reported incidence of distal fibular involvement is less than 1% of all GCTs.[1,3,4,5]

We present a case of a young female with GCT of distal fibula and was managed using an innovative reconstruction technique.

METHODS

Clinical picture

We present a female, eighteen years old, with pain in right ankle since 2 years ago. The pain felt by the patient when walking. 1.5 years later, a lump with a size of a marble grew on her ankle and was getting bigger progressively over time. There was no significant contributing history. (Figure 1)

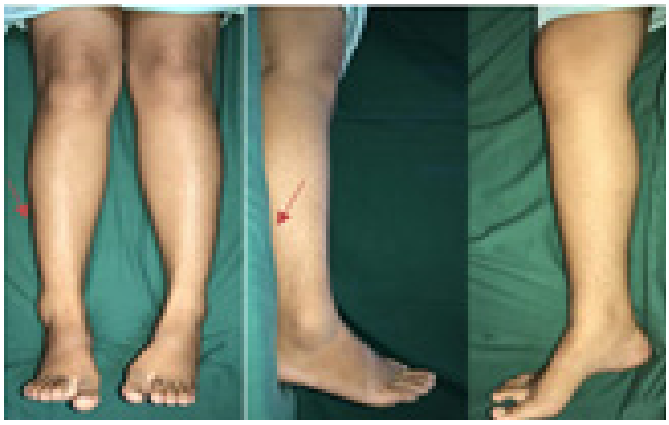


Figure 1. From the clinical picture, there is a lump on the lateral side of the right ankle.

Physical examination demonstrated a lump on the right distal fibula, with no venectation or sinus. There was palpable solid mass with tenderness on deep palpation. The circumferential diameter was 33 cm and contralateral side was 32 cm.

Radiograph findings in anteroposterior, lateral and mortise view showed single epimetaphyseal expansile lesion with soap bubble appearance. Magnetic resonance imaging revealed cystic bone tumour with septation in distal epimetaphyseal of right fibula, suggestive of Giant Cell Tumour. All routine haematological investigations were found to be normal. Core biopsy was performed; however, the results were inconclusive as typical cells

commonly found in giant cell tumour were not observed. (Figure 2-4)

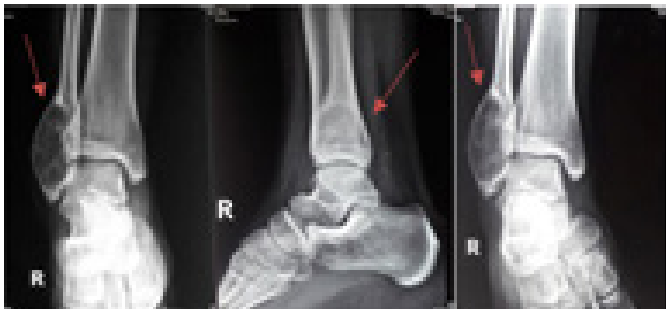
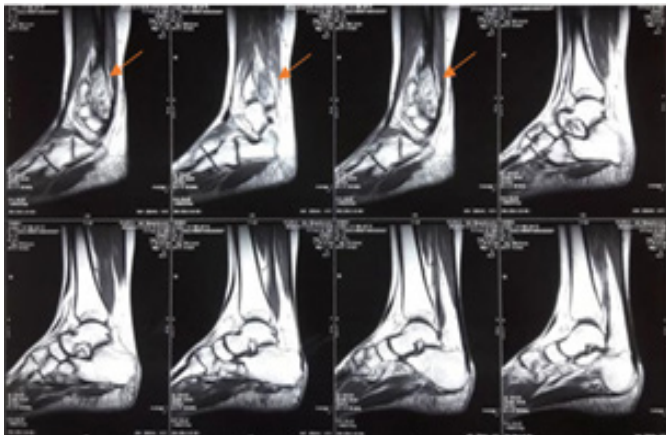


Figure 2. Ankle x-ray of the patient, from AP, lateral and mortise views, showed single epimetaphyseal expansile lesion with soap bubble appearance.



Fiqure 3. MRI Ankle Sagittal view revealed cystic bone tumour with septation in distal epimetaphyseal of the right fibula, suggestive of Giant Cell Tumour.

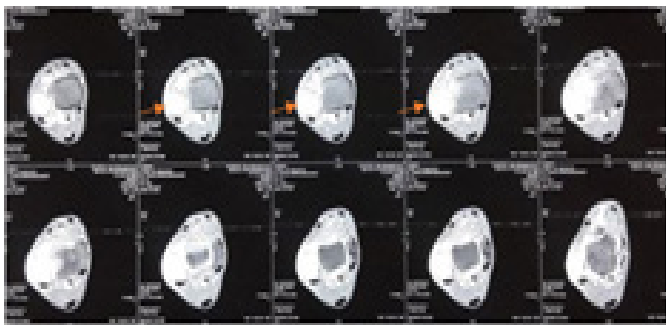
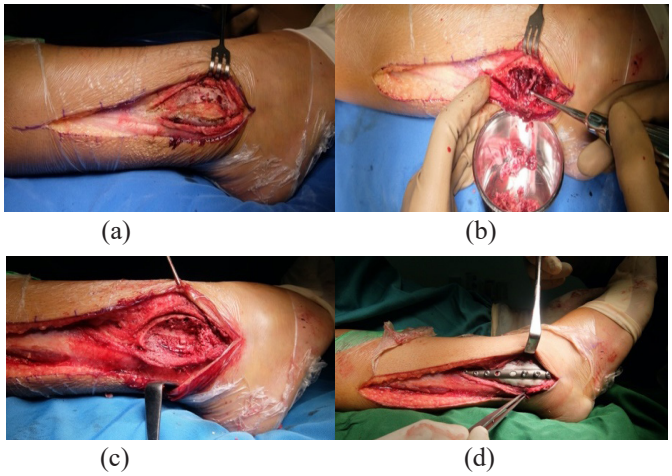


Figure 4. MRI Ankle Axial view.

After that, the patient underwent marginal excision by curretage with reconstruction using bone substitute application, plate and screw. The surgical steps for the operation can be seen in Figure 5.

Intraoperatively, the mass in the distal fibula was resected. We did hemicorticotomy. Curettage was performed followed by removing all of the tumor tissue completely on distal fibula. Fibular graft was taken from the fibular shaft as long as 5 cm and placed in the medial cortex of the distal fibula with bone substitute placement. We performed internal fixation in distal fibula with 1/3 tubular 6 holes and placed bone cement (Figure 6). Subsequently, the lateral ligament complex was sutured. Meticulous haemostasis was achieved after release of the tourniquet and the wounds were closed in layers. The specimen of the tumour was delivered to the pathology anatomy department.



(a) (b)

(c) (d)

Figure 5. Intraoperative condition: (A) Tumor exposed, (B) Bone curretage, (C) Bone graft and bone substitute application, (D) Final exposed

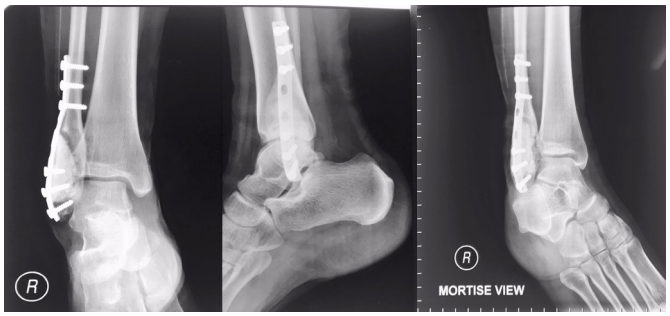


Figure 6. Ankle x-ray of the patient post-operative, from AP, lateral and mortise views.

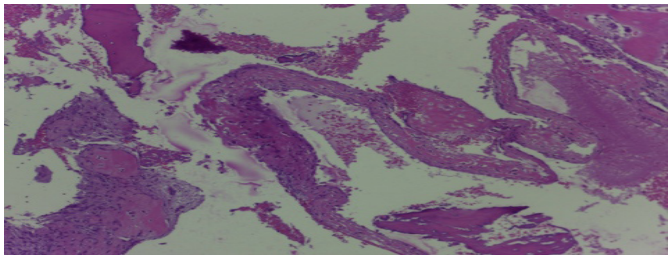


Figure 7. Intraoperative histopathological examination demonstrated multinucleated giant cells suggestive of giant cell tumor.

The patient was kept non-weight-bearing for three

months and plan to full weight-bearing at six months after the removal of screws. Intraoperative histopathological examination showed multinucleated giant cells that were suggestive of giant cell tumor. (Figure 7)

RESULTS

There were no significant postoperative complications. The wound was healed, and the patient was kept at non-weight-bearing for at least 3 months. The patient denied any complaint on ankle move restrictions, pain, or decrease of functionality. In general, the result was satisfying.

DISCUSSION

Giant cell tumors are benign aggressive tumors that may predilect at variable areas. It may appear at bones or other tissues. While knee area is thought to be one of the main predilection locations of the tumor, there is high possibility that the tumor may appear at other area. while the pathognomonic appearance may assist in establishing diagnosis, it may also cause confusion for clinician in term of diagnosis, especially in the late stage of giant cell tumor. Characterized by the giant cells-containing tissue, histopathology study may be the only way to clearly differentiate the giant cell tumor of the bone from the other kind of lesions.[6,7]

We reported a case of a female with a lump on her right ankle. Imaging showed the characteristic of expansile lesion that is usually seen in giant cell tumor. However, the diagnosis was halted by the fact that the patient was at her second decade of life, which is quite rare of the tumor. Even more, the location was quite unusual. Finally, after finding the typical giant cell-containing tissue that the diagnosis was then established. Diagnosing bone tumor is hard as it is tricky. Especially in cases of giant cell tumor, care is needed to even establish between whether this lesion is an infection (osteomyelitis), benign tumor, or even an aggressive tumor as each of the lesion only accepts a totally different treatment.[1,8]

In Cipto Mangunkusumo Hospital, every week a clinicopathology conference is done, attended by experts from orthopaedic-oncology, internal medicine, pediatrician, pathology anatomist, radiologist and other related department. The aim for this conference is to study cases and decide the diagnosis and treatment on the basis of collective knowledge from each branch of medicine. This is what this patient had underwent before advancing to treatment.[7,8]

The aim for treatment of bone tumor is total tumor resection, minimize the risk for recurrence, and promote maximal functional outcome. To this patient, we did tumor curettage, completed with adjuvants such as H2O2 and 96% alcohol, and bone grafting to fill in the defect. We decided to do this approach as for several reasons, the first was that this patient had not experienced any tumor cortical breakage, thus allowing to save the surrounding bone and soft tissues, while the second reason was to save the still-fully-functional ankle and not sacrificing any motion. In distal fibular resection without reconstruction, the stabilising effect of the lateral malleolus is lost. Soft-tissue reinforcement, even when it is possible, cannot fully compensate for the loss of stability. Resection of the lateral ankle can cause varus instability or a collapse into valgus. Resection and reconstruction provide good oncological clearance and better functional outcome and recommended in young active patients requiring resection of distal fibula. Intraoperatively, we did not sacrifice the syndesmosis as the stability was still good, and we did not sacrifice any of the lateral ankle ligaments.[9]

The distal end of the tibia and fibula are uncommon sites of occurrence for primary bone tumors. Limb salvage surgery at the ankle joint level is challenging due to scarce soft tissue coverage in this area and difficulty in achieving wide margins of resection because of this kind of problem of limb-sparing surgery for malignant and aggressive benign tumors about the ankle has become more feasible. Moreover, limb salvage surgery is certainly indicated in benign bone tumors. A case report by Nadkarni *et al*. stated that the use of modern imaging techniques and extended curettage through the use of power burrs and local adjuvants have improved outcome with reduced recurrence rates. Phenol, liquid nitrogen, bone cement, hydrogen peroxide, zinc chloride and more recently, argon beam cauterization have been employed as local adjuvants. Distal tibia tumor reconstructive options include custom made prostheses, osteoarticular allografts and ankle arthrodesis by the use of free or vascularized bone grafts. In this case, we did bone curretage, ORIF composite, bone substitute and bone grafting. Our technique is similar with a study conducted by Campanacci *et al*., where they did resection of the distal tibia and arthrodesis of the ankle with non-vascularized structural bone grafts combined with autologous bone chips, which

can be an effective procedure in bone tumor surgery with durable and satisfactory functional result (87% of MSTS functional score) and with no local recurrence, and histological examination revealed a successful wide surgical margin with an average of 53.5 months of follow-up.[10-12]

CONCLUSION

Giant cell tumor, as one of the most common bone tumors, may appear as an out-of-ordinary lesion and cause confusion in diagnosis. A careful approach, thorough history taking, complete imaging, and a state-of-the-art biopsy may be needed in order to establish a diagnosis. A well-thought-out treatment plan is needed by taking into account the risk of recurrence and maintaining the functionality of the musculoskeletal structure.

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