CASE REPORT

Recurrent Giant Cell Tumour with Secondary Aneurysmal Bone Cyst in Cuboid Bone: A Case Report

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ABSTRACT

This case report discusses Giant Cell Tumors of Bone (GCTB), constituting 5% of bone tumors, often found in long bones but rarely in foot bones (2.6%). These tumors exhibit a spectrum from benign to locally aggressive, with high recurrence rates despite surgery. Occasionally, GCTB leads to secondary Aneurysmal Bone Cysts (ABC), complicating diagnosis and treatment due to their similar presentation and high recurrence risk. The case involves a 25-year-old male with recurrent GCTB in the cuboid bone, initially excised. Subsequent evaluation revealed an expansive lesion, initially misinterpreted as other conditions. Histopathology confirmed secondary ABC arising in GCTB. Diagnostic biopsy, curettage, and tricortical bone grafting provided relief. Discussion highlights GCTB's aggressive behavior in foot bones, challenges in diagnosing secondary ABC, and the success of extensive treatment in managing GCTB with secondary ABC. Managing such lesions effectively is crucial due to associated severe morbidity. The case emphasizes the need for a comprehensive approach for diagnosis, treatment, and recurrence prevention, contributing to the evolving understanding of GCTB and secondary ABC interplay in different bone locations and to the growing body of literature on the challenging interplay between GCTB and secondary ABC.

KEY WORDS: Giant cell tumor, Aneurysmal bone cyst, Recurrent, Cuboid bone, Curettage, Tricortical bone graft.

Introduction

Cooper in 1818 first described Giant cell tumours of the bone (GCTB)[1], constitute about 5% of all bone tumours and typically affect the metaphysis and epiphysis of long bones.[2] GCTs in the feet are rare, with an incidence of 2.6%.[3] While overall GCT has a benign characteristic, but can display unpredictable behaviour, including local aggressiveness, bony destruction, and rarely metastasis, with recurrence rates between 25% and 50%.[4]

Aneurysmal Bone Cysts (ABC) are benign but locally aggressive lesions characterized by blood-filled cystic spaces.[4] Only a few cases of ABC secondary to GCT, particularly in short bones of the hands or feet have been reported.[5] The pathogenesis of secondary ABC remains partially understood, with RANKL, a cytokine involved in osteoclastogenesis, identified as a key factor.[2] Clinically, secondary ABC often mimics primary ABC, presenting with pain, swelling, and limited joint mobility.[5] Accurate diagnosis and management of secondary ABC require careful evaluation using advanced imaging and histopathological examination.[2]

We present a case of secondary ABC arising from recurrent GCT in a cuboidal bone, highlighting the need for a comprehensive approach to treatment and diagnosis.

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Case Presentation

A relatively asymptomatic 25 year old male patient belonging to lower socio-economical status presented to orthopedic outpatient department with complaint of pain in right foot and difficulty in walking since

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one year. Patient gave history of excision of mass from foot at same site one and half years back at an outside hospital, which was then diagnosed as Giant cell tumour of bone. The pain was insidious in onset, dull-aching, aggravated while walking and relieved with rest initially. Pain increased in severity for last four months and was continuous over the time. There was no history of night pain, fever or discharging sinus. On local examination, diffuse swelling was present over medial aspect of the midfoot, there was no redness or discharging sinus. On palpation, the swelling was diffuse, cystic and tenderness was present locally. The range of motion was restricted and painful.

X ray of left foot showed presence of well-defined expansile lesion with irregular margins and internal septations involving left cuboid bone, possibility of recurrent GCT was given.MRI showed presence of well-defined destructive, expansile, lytic, altered signal intensity lesion of size 23 x 25 x 28 mm with irregular margins and internal septations involving left cuboid bone. The lesion was hypotense and showed patchy diffusion restriction with peripheral blooming. No evidence of matrix mineralization/ periosteal reaction. Lesion was causing cortical erosion of adjacent cuboid bone with bone marrow edema. MRI was suggestive of: aggressive benign lesion / eosinophilic granuloma / osteomyelitis.

Biopsy from left cuboid bone was done and sent for histopathology. Histology was suggestive of Aneurysmal Bone Cyst with microscopic presence of predominantly large spaces, without any endothelial lining, filled with blood, with few osteoclast like giant cells.

Considering the previous history of GCT bone and radiological findings a diagnostic open biopsy & curettage was done with all aseptic precautions under spinal anesthesia using an antero-medial approach with a 4 cm incision to rule out recurrence. The cuboid bone was found to be almost entirely cystic with a breach in the dorsal cortex. Gross intra-operative appearance was suggestive of fibrous dysplasia or giant cell tumor of bone. Specimen was sent for histopathologic examination and pus culture & sensitivity. Microscopic Examination showed presence of uniformly distributed multi-nucleated giant cells interspersed between proliferating mononuclear stromal cell component. Also, there were presence of many large spaces, without any endothelial lining, filled with blood. They were delimited by fibroblast, myofibroblast, histiocytes and row of osteoclasts at places. Foci showing reactive bone formation were also seen. Diagnosis of Giant Cell Tumour with secondary Aneurysmal Bone Cyst formation was given.

Following the biopsy report, extensive curettage of the tumor followed by Tricortical bone graft (taken from ipsilateral iliac crest) & plating was done. Following surgery patient was stable and had relief from pain in the foot.

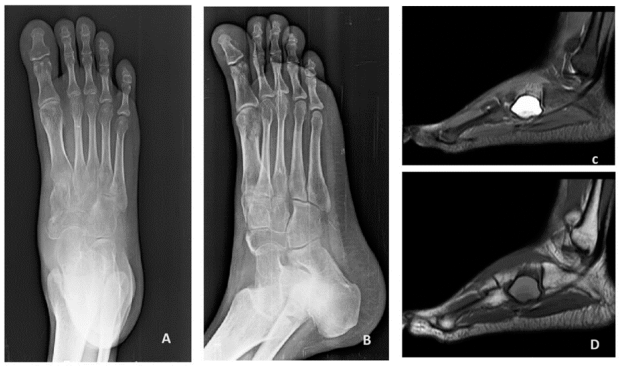

Figure 1: A & B: X ray of left foot. Left cuboid bone shows tiny internal sclerotic and lytic areas. Few tiny bone fragments are noted around cuboid bone; C & D: MRI: Sagittal view of left foot. Presence of well-defined expansile altered signal intensity lesion of size 23x25x28 mm with irregular margins and internal septations is noted involving left cuboid bone, destructive expansile lytic lesion is noted involving cuboid bone

Discussion

Giant cell tumor of bone (GCTB) is a common, locally invasive benign tumor typically found in the metaphyseal-epiphyseal region of long bones after skeletal maturity.[6] Although rare, GCTB can be more aggressive when located in the small bones of the hand, foot, and ankle.[6] An aneurysmal bone cyst (ABC) secondary to GCTB is a rare condition, with an incidence of 0.011 to 0.053 per 100,000 annually.[5] ABC is a benign, expansile cystic bone lesion characterized by blood-filled spaces separated by connective tissue septa, with osteoclast-type giant cells and reactive woven bone.[4] On radiographs, ABC often presents with a 'soap bubble' appearance, similar to GCTB.

ABCs can be primary or secondary to existing bone lesions or trauma. Secondary ABCs are associated with various conditions, including GCTB, chondrob-

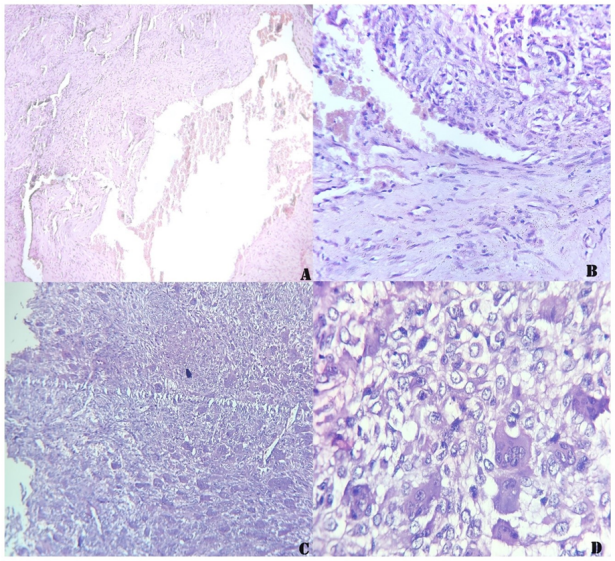

Figure 2: A: H & E stain 10x view, large spaces, without any endothelial lining, filled with blood; B: H & E stain 40x view, Endothelial lining is absent. They were delimited by fibroblast, myofibroblast & histiocytes suggestive of ABC; C: H & E stain 10x view, uniformly distributed multi-nucleated osteoclast like giant cells interspersed between proliferating mononuclear stromal cell component. Suggestive of GCT; D: H & E stain 40x view, uniformly distributed multi-nucleated osteoclast like giant cells

lastoma, and osteosarcoma.[5] Histopathologically, the presence of both GCTB and ABC is crucial for diagnosis.[7,8] Although the risk of malignant transformation is low, secondary ABCs can lead to significant morbidity, as illustrated by a 2022 case involving the cervical spine in a pediatric patient.[9]

The management of GCTB is critical due to a reported recurrence rate of 24.4%, highlighting the need for effective treatment strategies.[10,11] The standard approach includes extensive curettage, which was performed in our case, followed by adjunctive treatments.[8]

Recent case reports, including those by Kamal and Hilmy (2022) and Li et al. (2020), demonstrate the need for heightened awareness of GCTB's potential to affect various bones, such as the calcaneus and hand phalanges.[2,12] Identifying secondary ABC is crucial for accurate diagnosis and treatment, which typically involves addressing the underlying GCTB.

Conclusion

In conclusion, the coexistence of giant cell tumor (GCT) and aneurysmal bone cyst (ABC) poses diagnostic and therapeutic challenges, necessitating a comprehensive understanding of this entity for proper management and prevention of morbidity and recurrence.

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