

Aneurysmal Bone Cyst of the First Metatarsal Reconstructed Using Non Vascular Free Fibular Graft Transfer - A Case Report

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Abstract: We present a case of aneurysmal bone cyst of the first metatarsal, which has been documented rarely in the literature. Its characteristics are similar to those of a variety of different bone tumours. While the radiographic appearance of this lesion frequently identifies it, microscopic examination is required for a definitive diagnosis. Depending on the size, age, and location of the lesion, there are a variety of therapeutic options available. In this report, we recommend total excision of the lesion and reconstruction of the defect by transfer of fibular segment. We obtained satisfactory clinical and radiological results, after 6 months of postoperative evaluation.

Keywords Aneurysmal bone cyst, Metatarsal, Fibula transfer

1.Introduction

An aneurysmal bone cyst is a benign osteolytic lesion that develops in the metaphysis of long bones, flat bones, and vertebrae. Jaffe and Lichtenstein were the first to describe it in 1942 ^[1].

Aneurysmal bone cysts account for approximately 1.3-1.5 percent of all primary bone tumours ^[1,2]. It originates as a primary tumour in about 70% of cases without any documented precursor bone lesion or as a sequel lesion in about 30% of cases when a prior osseous lesion can be identified ^[3].

Aneurysmal bone cysts in the metatarsal are extremely uncommon, with only a few cases recorded in the literature till date. The lesion is described as having a blown out, dilated cortex, and it is most commonly encountered in children and young adults. It first appears in the second and third decades of life. Females are slightly more likely to develop the lesion ^[4].

The clinical presentation varies, but the patient usually has localised pain, tenderness, and swelling, or a pathological fracture.

A radiolucent area with expansion and apparent septations can be seen on plain radiographs. The lesion is most commonly found centrally in the metaphysis, with equal cortical expansion on the periphery, leaving a thin bone cortex ^[2]. The aneurysmal bone cyst often shows a double fluid density on magnetic resonance imaging, with low intensity signals on T1 and high intensity signals on T2. These fluid-fluid levels can be very helpful in determining a diagnosis, but they are not diagnostic.

Blood-filled spaces with connective tissue septa between those spaces containing trabecular bone, osteoid tissue, osteoclastic cells, and the absence of endothelium are seen on histological examination. Histiocytes, fibroblasts, and

scattered multinucleated giant cells abound in the mesenchymal tissue ^[2].

As the lesion can have thin or eroded cortex, it's easy to mistake it for a malignant bone tumour like osteosarcoma. Aneurysmal bone cyst must be distinguished from giant cell tumour, unicameral bone cyst, hemangioma, chondromyxoid fibroma, eosinophilic granuloma, nonossifying fibroma, and osteoblastoma in the majority of cases. As a result, accurate microscopic evaluation is critical for determining a definitive diagnosis and prognosis.

Treatment options include curettage, curettage augmented by the use of a high speed bur, saucerization, and bone marrow injection. Resection of the tumor is advised when it is located in an expandable bone (e. g., fibula, rib) ^[2]. Present study report a rare case of **Aneurysmal Bone Cyst of 1st metatarsal showing Near total reconstruction of metatarsal after excision of rare bone tumor using non vascular free Fibular graft.**

Case report-13 year-old girl with complaints of pain and swelling over the dorsal aspect of her left foot of 7 months duration presented to department of orthopaedics, NSCB Medical college Jabalpur. The patient gave history of trauma 7 months back, which was diagnosed as sprain and managed conservatively with rest, ice and compressive bandaging. Local examination revealed a swelling over the dorsum of foot with surrounding indurated skin, tender on palpation and restricted ankle and toe movements due to pain. Rest of the local and systemic examination remained unremarkable. she presented with no other skeletal system complaint or abnormal laboratory studies.

Imaging studies included conventional radiographs and magnetic resonance imaging (MRI). Conventional radiographs showed fusiform dilation with a radiolucent area of multiple septations in the first metatarsal shaft (Figure 1). There was no interruption in the thin cortical lining. MRI revealed an expansile bone lesion with

multiple septations in the diaphysis of right first metatarsal bone causing significant expansion and thinning of overlying cortex. Small extension to the adjacent epiphysis of base of first metatarsal noted. This lesion was hypointense in T1 and hyperintense in T2 weighted images and no evidence of cortical breach, periosteal reaction or extraosseous extension noted (Figure 2). Needle aspiration biopsy was performed, which revealed osteoclastic multinucleated giant cells that indicated a benign lesion.



Figure 1: Radiographs of the right foot showing fusiform dilation with a radiolucent area of multiple septations in the first metatarsal shaft

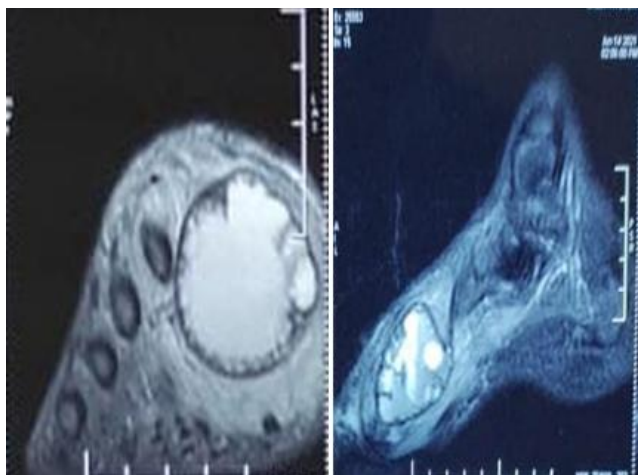


Figure 2: MRI revealed an expansile bone lesion with multiple septations in the right first metatarsal bone

The surgical plan was total excision of the lesion with incorporation of an autogenous interpositional graft using internal fixation. In the operation, a 8-cm linear incision was placed over the dorsum of first metatarsal. The incision passed through layers to the level of the periosteum. The periosteum was incised and reflected; exposing the full length of the metatarsal shaft. The first metatarsal revealed fusiform swelling, as anticipated from X-ray findings (Figure 3-a). The thin-walled tumor was resected en bloc with drilling and osteotomy (Figure 3-b). The defect was then replaced by transfer of ipsilateral fibula segment of 6 cm and fixed by krischnerwire across the osteotomy sites (Figure 3-c). The patient was kept on

below knee non-weight bearing cast for 8 weeks. The patient was then placed in a below-knee non weight bearing slab for 4 weeks. After 12 weeks of non-weight bearing, K-wires were removed. The patient was then placed in a below-knee slab for 4 weeks with partial weight bearing. During this time, range of motion of the first metatarsophalangeal joint was encouraged.

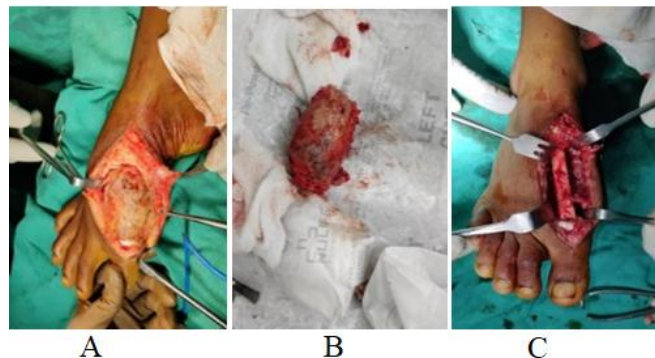


Figure 3: Excision of tumor (en-bloc) and transfer of ipsilateral fibula segment



Figure 4: Post op radiograph showing fixation of fibular graft with K wire

Histologically, the lesion was made up of benign reactive tissue with a fibrohistiocytic stroma, multinucleated giant cells, and reactive bone surrounding bloodfilled spaces. There were no significant numbers of mitotic cells found, and there was no evidence of nuclear atypia. Histopathological diagnosis was aneurysmal bone cyst.

Her 6 month follow-up radiographs showed the union with good graft incorporation and consolidation. There was no evidence of recurrence determined radiologically. The functional use of the metatarsophalangeal joint is full; the patient is able to walk freely.



Figure 5: Six month follow-up radiograph showed the good graft incorporation and consolidation

2. Discussion

Aneurysmal bone cyst is a benign tumor that may behave in an active, aggressive and destructive manner. The incidence is 1.4 cases per 1, 00, 000 individuals, and it constitutes approximately 1% of all bone tumors^[5]. Most of the patients present with swelling and pain, and often with pathological fracture.

The precise pathogenesis of ABC is unclear, but the most widely accepted pathogenic mechanism of ABC involves local circulatory disturbance, which results in an increase in venous pressure and the development of enlarged and dilated vascular components within the affected bone^[3].

The differential diagnosis of an aneurysmal bone cyst can include enchondroma, giant cell tumor at an early age, osteoclastoma, Ewing's sarcoma, giant cell reparative granuloma, nonossifying fibroma, fibrous dysplasia, ossifying fibroma, or eosinophilic granuloma. GCT is composed of mononuclear and osteoclast-like multinucleated giant cells, which have the potential to be locally aggressive^[6]. In GCT, the tumor is always eccentrically located in the epiphysis and metaphysis of the bone, and exhibits lytic expansion^[7]. GCRG is a rare, benign, intraosseous reactive lesion, histologically characterized by a predominance of giant and mononuclear cells in areas of hemorrhage^[8]. Brown tumors have been reported to have a considerably more lobulated architectural growth pattern; at differential diagnosis, hyperparathyroidism can be ruled out on the basis of serum calcium, parathyroid and phosphorus hormone levels (9). ABC, on the other hand, is known to be histologically composed of blood-filled cystic spaces separated by fibrous septa^[2].

Historically, the most common form of treatment for aneurysmal bone cyst is curettage and packing of bone graft^[10]. This method employs a cortical window in the lesion with aggressive curettage and irrigation. It is followed by the packing of the lesion with bone chips and replacement of the window. A problem with this method

is that osteoclastic activity can reabsorb the graft material, depending on the aggressiveness of the lesion, which is variable and unpredictable. Another limitation of this method is that if incorporation occurs, the original size of the lesion still is present and, if large, can take years to remodel^[10].

Radiation has been offered as an alternative treatment, especially when it involves more inaccessible areas such as vertebral bodies. The sclerosing factor of radiation to vessels appears to be beneficial in treating the lesion. However, there have been reports of malignant degeneration to sarcomas occurring with this method^[11].

If the lesion is aggressive and the function would not be compromised, total excision is recommended^[10, 2]. Total excision of the lesion with graft incorporation is suggested to prevent a recurrent deformity. Tibial autogenous grafts have been used to replace the first metatarsal aneurysmal bone cyst. However, graft incorporation may be difficult. Bone grafts from this area produce primarily cortical bone and create a structural weakness in the donor site^[10]. Iliac grafts provide an excellent source of corticocancellous material and have been used for metatarsal grafting^[10, 2]. Significant bleeding can occur along with disability from harvesting the material from this site.

Fibular grafts can be whole or partial and offer a good source of corticocancellous bone. They also do not lead to much structural compromise^[12]. Adjuvants to surgery such as chemical cautery (phenol) or cryotherapy (liquid nitrogen) have been used by a few authors to decrease the recurrence rates, but they result in an increased risk of postoperative fractures^[2]. Minimally invasive procedures such as the percutaneous injection of steroid or calcitonin have been reported in small series with good results. This may be indicated in lesions with difficult surgical approaches (i.e., C1 spine or pelvis), but the presence of intracystic septa increases the risk of incomplete healing.

3. Conclusion

Diagnosis of ABC is challenging and, it must be stressed that neither radiographic nor clinical finding should be relied on for diagnosis of a lytic bone lesion. Histopathological examination combined with relevant clinical history, X-rays, and pathological information is critical in obtaining an accurate diagnosis. En bloc resection and replacing the bone defect with fibular graft is a safe procedure with minimal risk of recurrence and may be considered treatment of choice if the resection is possible.

References

- [1] Jaffe HL, Lichstentein L (1942) Solitary unicameral bone cyst, with emphasis on roentgen picture, pathologic appearance and pathogenesis. Arch Surg 44: 1004-1025
- [2] Ramirez AR, Stanton RP (2002) Aneurysmal bone cyst in 29 children. J Pediatr Orthop 22: 533-539

- [3] Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. *Arch Orthop Trauma Surg* 127 (2): 105-114, 2007.
- [4] El-Khoury GY, Seaman RW (1980) Case report 125. *Skeletal Radiol* 5: 201-203
- [5] Ozaki T, Hillmann A, Lindner N (1997) Cementation of primary aneurysmal bone cysts. *Clin Orthop* 337: 240-248
- [6] Foo LF and Raby N: Tumours and tumour-like lesions in the foot and ankle. *Clin Radiol* 60: 308-3332, 2005.
- [7] Futamura N, Urakawa H, Tsukushi S, Arai E, Kozawa E, Ishiguro N and Nishida Y: Giant cell tumor of bone arising in long bones possibly originates from the metaphyseal region. *Oncol Lett* 11: 2629-2634, 2016.
- [8] Cook DL, Rosenthal DC and Shikoff MD: Giant cell reparative granuloma of the middle phalanx of the foot: A review and case report. *J Foot Ankle Surg* 47: 589-593, 2008.
- [9] Hanna BG, Donthineni R, Majid K, Parekh S, Shin JS and Lackman RD: Leg mass in a 61-year-old man. *Clin Orthop Relat Res* 406: 298-307, 2003.
- [10] Hatch DJ, Buckholz JM (1992) Aneurysmal bone cyst of the second metatarsal. *J Foot Surg* 31: 129-133
- [11] Dabska M, Buraczewski J (1969) Aneurysmal bone cyst: pathology, clinical course, radiologic appearances. *Cancer* 23: 371-377
- [12] Hexner GU, Jacob HA, Middendorp J (1998) Reconstruction of the first and second metatarsals with free microvascular fibular bone graft after resection of a Ewing sarcoma. *J Pediatr Orthop B* 7: 239-242