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RESEARCH ARTICLE

Aneurysmal Bone Cyst Of Metatarsus : A Rare Case Report and Review of Literature

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Abstract

Aneurysmal bone cysts are locally destructive, blood-filled reactive lesions of bone and are not considered to be true neoplasms. Any bone may be involved, but the most common locations include the proximal humerus, distal femur, proximal tibia, and spine. Aneurysmal bone cysts (ABC's) in the foot are rare. We hereby report a case of Aneurysmal bone cyst in 2nd metatarsal of 12 year old boy. The patient was operated with En bloc resection , excision of articular cartilage of proximal phalynx and arthrodesis of 2nd metacarpophalangeal joint with non vascular Fibular grafting which was stabilized by K wires. The patient was followed up regularly and has had excellent functional recovery.

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Introduction:-

Aneurysmal bone cysts are locally destructive, blood-filled reactive lesions of bone and are not considered to be true neoplasms. Any bone may be involved, but the most common locations include the proximal humerus, distal femur, proximal tibia, and spine. Vertebral lesions, accounting for 15% to 20% of these entities, are located in the posterior elements with frequent extension into the vertebral body or to adjacent levels. Aneurysmal bone cysts (ABC's) in the foot are rare. Most occur in patients younger than 20 years old, and there is a slight female predominance. We hereby report a case of Aneurysmal bone cyst in 2nd metatarsal of 12 year old boy treated at our institute. Lesion characteristically showed fluid-fluid level , highly suggestive of Aneurysmal bone cyst on MRI .

Case Report:-

A 12 year old boy of Waghodiya , Vadodara came to our OPD with chief complain of swelling at right foot region of 6 months duration . The swelling was intermittently painful, gradually increasing in size, associated with pain and limp, with no history of trauma . On examination, the swelling was solitary, localized to dorsum of foot, tender, about 6cm*4cm in size , firm in consistency, with irregular surface and non pulsatile with normal distal neurovascular examination. On Xray examination (Fig 1.) an expansile lytic lesion of 2nd metatarsal was evident. On MRI (Fig 2), the lesion appeared hypointense on T1 weighted images and hyperintense on T2 weighted images with internal septations on enhancement views. Lesion characteristically showed fluid-fluid level , highly suggestive of Aneurysmal bone cyst .The patient was operated with En bloc resection , excision of articular cartilage of proximal phalynx and arthrodesis of 2nd metacarpophalangeal joint with non vascular Fibular grafting which was stabilized by K wires(Fig 3). On biopsy, microscopic examination showed multinucleated osteoclast like giant cells amongst vascular fibrocartilagenous stroma which confirmed the diagnosis of Aneurysmal bone cyst. Post operatively, patient was immobilized in BK slab for 2months. After 4 months post op, K wire was removed as graft got incorporated. At final follow-up patient had good functional recovery with near normal range of movements and no signs of recurrence (Fig 4 ,Fig 5).

Discussion:-

In 1942, Jaffe and Lichtenstein first described ABC as a distinct entity when they discovered "a peculiar blood-containing cyst of large size." Most ABC's will occur between the ages of 10 to 30 ^{1,5}. The precise

pathogenesis is uncertain; various theories have been suggested ranging from posttraumatic bony alteration , reactive vascular malformation , to genetic predisposition.⁷ ABCs of metatarsals are rare lesions with the predilection of this lesion for long bones². The lesions consist of blood filled spaces filled with connective tissue septa containing bone or osteoid and osteoclastic giant cells.^{1,2}

Most patients with aneurysmal bone cysts complain of mild to moderate pain that has been present for weeks to several months. It may also appear as a firm , slowly enlarging mass in adolescents .Sometimes rapid growth can occur and clinically may mimic a malignancy. Spinal lesions may cause neurological deficits or radicular pain^{2,7,8}.

Radiographs reveal an expansile lytic lesion that elevates the periosteum but remains contained by a thin shell of cortical bone. An aneurysmal bone cyst can have well-defined margins or a permeative appearance that mimics a malignancy. It is most often eccentrically located in the metaphysis. CT is particularly helpful in delineating the cyst in areas of complex anatomy, such as the spine or pelvis. In addition, the thin rim of bone surrounding the cyst can be identified. MRI shows the multiloculated cavities and fluid levels. When differentiating between a unicameral and aneurysmal bone cyst using MRI, the presence of a double-density fluid level and intralesional septations usually indicates an aneurysmal bone cyst. However this finding is also noted sometimes in Giant cell tumor, telangiectatic osteosarcoma as well as osteoblastoma^{1,2,7}

Differential diagnosis to an aneurysmal bone cyst include various lesions such as Giant cell tumor, Giant cell reparative granuloma ,Chondroblastoma, osteoblastoma, Brown's tumor, telangiectatic osteosarcoma , angiosarcoma and chondromyxoid fibroma. Grossly, an aneurysmal bone cyst is a cavitory lesion with blood-filled septate spaces. It is surrounded by a thin layer of bone covered by a raised periosteum. The microscopic appearance is of hemorrhagic tissue with cavernous spaces separated by a cellular stroma. The lining of the cavitory spaces consists of compressed fibroblasts and histiocytes , and multinucleated giant cells also are present . Sanerkin et al described a 'Solid variant' of ABC with unusual noncystic intralesional features.^{1,2,7}

There are variable techniques used to treat ABC's . Most aneurysmal bone cysts are treated with extended curettage and autologous bone graft . Adjuvant therapies have also been attempted post curettage including the use of liquid nitrogen and phenol. Once the cyst is evacuated, a number of various materials have also been used to fill the void. The cavity can be packed with either autograft, heterograft and even polymethyl-methacrylate and can be done endoscopically as well if the site permits¹⁴. Because the lesion may produce heavy bleeding, tourniquet control is advised^{1,2,12}. The recurrence rate after curettage of an aneurysmal bone cyst is 10% to 20%. Recurrence has been correlated with age younger than 15 years, centrally located cysts, and incomplete removal of the cystic cavity contents. Recurrent cysts can be treated with the same approach as the primary lesion.^{1,2} In a study by Gibbs, et al., thirty-four of the forty patients with ABCs reviewed had curettage of bone using a high-speed burr. Of the twenty-two patients who had cancellous bone autogenous grafting, only 12% had local recurrence. Of six patients who had resection of the cyst through its margin, none had recurrence.^{1,4} In 1997, Schreuder, et al., reported their results on curettage with the adjunctive use of cryotherapy. They found similar results to those of marginal resection.^{1,6,7}

Marginal resection or En Bloc resection sometimes is indicated for lesions in expendable bones such as metatarsals . Such lesions, post resection, have been treated with tricortical iliac crest graft⁹, tibial diaphyseal cortical graft¹⁰ or Non vascularised Fibular autograft^{11,12}. En bloc resection so far has been reported with lowest recurrence rate.^{7,9,10,13}

Percutaneous injection of sclerosing agent like alcohol is a viable alternative to more invasive procedures for treatment of ABCs .In a study by S. Rastogi et al , 72 patients with a histologically-proven diagnosis of aneurysmal bone cyst, at various skeletal sites , were treated with percutaneous sclerotherapy using Polidocanol . The mean reduction in size of the lesion (radiological healing) was found to be 76.6% (61.9% to 93.2%) with a mean clinical response of 84.5% (73.4% to 100%).¹⁵ Adamsbaum et al. had also reported good results after percutaneous injection of Ethibloc in benign bone cyst cases .¹⁶

In the present case of 2nd metatarsal ABC , a rare lesion at this location, as the tumor encompassed from the base to neck of 2nd metatarsal the only viable option left was Enbloc resection and reconstruction with non vascularised fibula graft . The fibula graft was inserted into the cancellous bone of middle cuneiform proximally and articular cartilage of proximal phalynx was removed for arthrodesis of 2nd Metatarsophalangeal joint .This graft was then stabilized with two K wires; one intramedullary creating athrodesis of 2nd metatarsophalangeal joint and the other in transverse transmetatarsal direction for maintaining the metatarsal arch. The transmetatarsal K wire insertion helped to retain the transverse arch of foot and has not been mentioned in any previously available literature . The patient was followed up regularly and has had excellent functional recovery without any complaints after 6 months post operatively ,and is able to walk and run normally .

In conclusion, ABCs of metatarsal is a rare lesion to be reported in current literature and treatment with enbloc resection and non vascularised fibular graft stabilised by K wires may be considered as a viable option for its treatment, with minimal risk of recurrence .



Fig. 1 Pre Op Xray

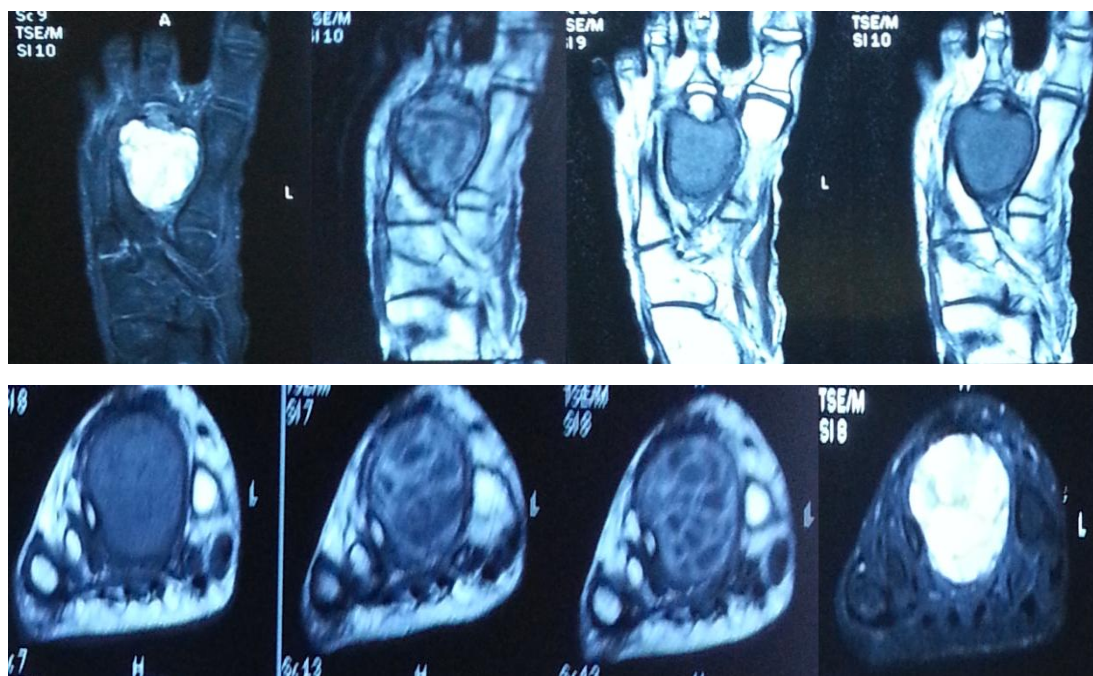


Fig 2. MRI Right Foot

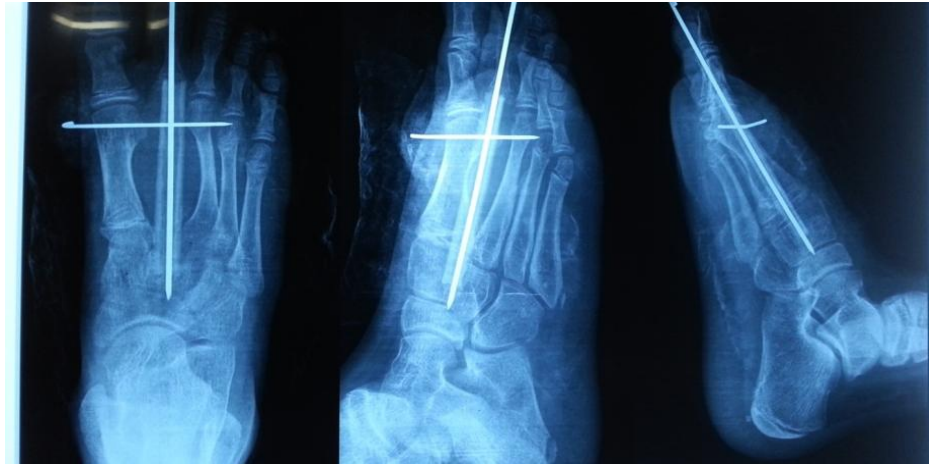


Fig 3 .Post Op Xray



Fig. 4 Final Follow up X-ray at 7 months



Fig 5: Functional outcome at Final follow-up at 7 months

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