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Research Paper

Distal Tibial Aneurysmal Bone Cyst Treated by Using Bone Graft Substitute and Autologous Cortical Iliac Crest Strut Graft: A Case Report

Medical Science

KEYWORDS : Aneurysmal bone cyst, distal tibia, bone graft substitute, hydroxyapatite

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ABSTRACT

Aneurysmal bone cysts are rare benign osteolytic skeletal tumors that most commonly occur in the first two decades of life, affecting the metaphysis of long bones. It presents as blood-filled cavitary septate spaces with blowout distension of the cortex, with often a thin rim of intact periosteum, and a fibrous cyst wall consisting of fibroblasts and histiocytes. The pathogenesis remains controversial; when occurring de novo, it is believed to be the result of local haemorrhage into bone. Treatment consists of extended curettage and use of bone graft and bone graft substitutes. Because of its excellent osteoconductive properties and mechanical strength, calcium hydroxyapatite has emerged as an excellent choice in treating benign bone defects. We present a case of a distal tibial aneurysmal bone cyst in a young female treated by using hydroxyapatite crystals and autologous cortical iliac crest strut graft.

Introduction

Aneurysmal bone cysts are rare benign osteolytic skeletal tumors that most commonly occur in the first two decades of life(1). Females are affected more than males in a ratio of 2:1. The most common location is the metaphysis of lower extremity long bones, more so than the upper extremity(2). Despite a descriptive history of more than 60 years, the nature, character, and optimal treatment of aneurysmal bone cysts remain obscure. The lesion was first described by Jaffe and Lichtenstein in 1942(3).

Intra-operative findings usually demonstrate a blood-filled cavity within an expanded region of the bone, and the cells that line the cyst wall show fibrous components, macrophages, giant cells, and islands of bone. The term aneurysmal seems to relate to the blowout distension, and the word cyst reflects the fact that the tumor often presents as a blood-filled cavity.

We present a case report of an Aneurysmal Bone cyst involving the distal end of tibia in a 20 years old female treated by curettage. Large calcium hydroxyapetite crystals were used to fill up the cyst cavity, and the eroded cortex was reconstructed using autologous iliac crest cortical strut graft.

Case report

A 20 years old female presented to us with pain around her left ankle for the past four months. Clinically, no swelling was palpable, and ankle movements were good. However, an X-ray of the ankle joint revealed an osteolytic expansile lesion involving the distal end of tibia. Cortex appeared eroded on the posterior and postero-lateral aspect; only a thin rim of bone was visible. A few thin septations could be appreciated within the cavity of the cyst. Articular surface of distal tibia appeared intact.

On getting an MRI done, presence of multiple locules containing free fluid inside the tumour cavity was confirmed. The cyst itself appeared benign with intact cyst wall and rim of articular cartilage. A provisional diagnosis of Aneurysmal Bone cyst was made.

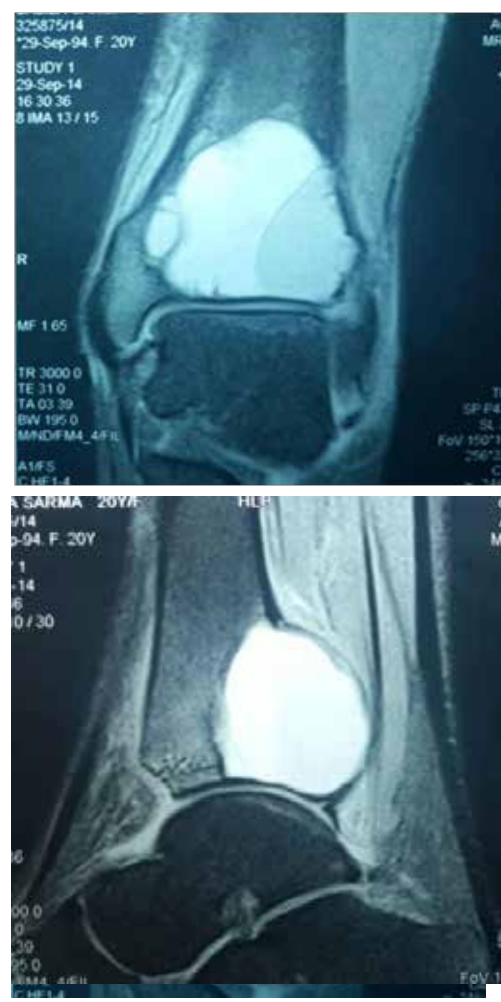

Fig. 1: T2 weighted MRI images showing homogenously enhancing fluid filled loculated cyst cavity in distal tibia. Articular surface appeared intact.

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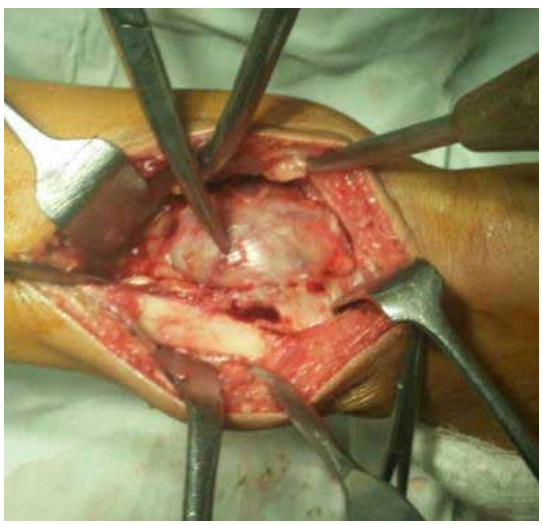
The patient was taken up for surgery with a plan to drain the cyst contents, curet out the wall of the cavity and fill up the defect with bone graft substitute. Commercially available calcium hydroxyapatite crystals were kept ready.

Patient was positioned prone under spinal anaesthesia. Tourniquet was raised. We went through a posterior approach going lateral to the tendo-achilles. Dissection was carried out maintaining proper hemostasis. Posteriorly, deficient distal tibial cortex was noted, and the posterior cyst wall was exposed. Needle aspiration of the cavity through the posterior wall resulted in about 10 ml of thick blood being evacuated.



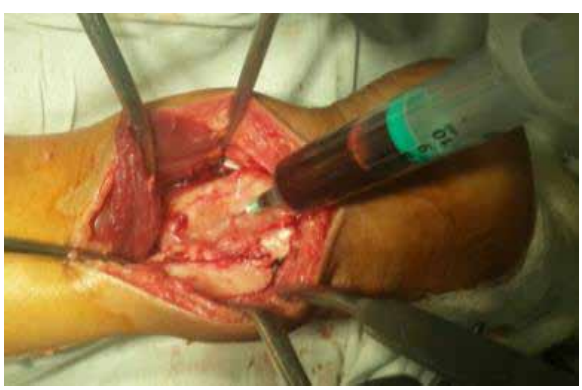


Fig 2: Intra-operative images showing deficiency in the posterior distal tibial cortex. The posterior wall of the cyst has been exposed alongside adjacent fibula. The tendo-achilles has been retracted medially. On attempting needle aspiration through the posterior wall, about 10 ml of thick blood was evacuated.

The cavity was opened through the posterior cyst wall. Cyst wall was found to be fibrous and friable. It was thoroughly curetted and rest of the thick blood evacuated. Bits of curetted wall tissue and blood were sent for histopathological examination. Roughly 6 cm by 4 cm cyst size was noted.

On palpating the distal tibia articular surface through the opening in the cyst wall, it was found to be intact and offering solid resistance throughout. After giving a thorough wash with hydrogen peroxide and normal saline, the cyst cavity was then packed with calcium hydroxyapapetite crystals. Ipsilateral cortical iliac crest graft was harvested, split, and used to reconstruct the deficient posterior wall of the distal tibia. The incised periosteum was sutured over the now reconstructed defect.

Fig 3: Intra-operative images showing the cyst cavity with intact distal tibial articular surface. Later, calcium hydroxyapatite crystals were used to fill up the defect, and split cortical iliac crest autograft was used to reconstruct the posterior wall.

Post-operative X-ray showed the cyst cavity to be completely packed with radiodense hydroxyapatite crystals, with hardly any dead space. Immediate post-operative period was uneventful. The involved ankle was immobilized in a below-knee Plaster of Paris slab, which was later converted to a fibreglass walking cast once the wound had completely healed and sutures were removed at two weeks after surgery. Toe touch weight bearing was started.

At 3 months follow-up the cast was removed. Surgical wound was healthy without any evidence of infection. Ankle movements were started; apart from the initial stiffness, it was for the most part painless. Radiographs showed signs of incorporation of the parent bone with the hydroxyapatite.

Fig 4: Fig 4: Post operative X-Rays showing the defect completely packed with calcium hydroxyapatite crystals. The posterior wall of the cyst has been reconstructed with cor-

tical iliac crest strut graft, with periosteum sutured over it.

Discussion

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.(4) In a review of 150 cases of aneurysmal bone cysts treated over a period of 20 years, the distal tibia was found to be involved in 7 patients (4.7%).(3) Most patients with aneurysmal bone cysts complain of mild to moderate pain that has been present for weeks to several months. Rapid growth can occur and clinically may mimic a malignancy. Spinal lesions may cause neurological deficits or radicular pain.

Pathogenesis of these tumors remains controversial. An aneurysmal bone cyst can arise de novo, but areas similar to an aneurysmal bone cyst are found in various other lesions, such as giant cell tumor, chondroblastoma, osteoblastoma, fibrous dysplasia, nonossifying fibroma, and chondromyxoid fibroma. Many studies believe it to be a result of haemorrhagic degradative events at these tumour sites. It is likely that primary aneurysmal bone cysts result from local circulatory disturbance leading to increased venous pressure and production of local hemorrhage. (4) End result is a cavitary lesion with blood-filled septate spaces, surrounded by a thin layer of bone covered by periosteum. The lining of the cavitary spaces consists of compressed fibroblasts and histiocytes.

Radiographic features include 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. Bone scan shows diffuse or peripheral tracer uptake with a central area of decreased uptake. 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.

Most aneurysmal bone cysts are treated with extended curettage and grafting with a bone graft substitute. In other studies, allograft chips, polymethymethacrylate and autograft have also been used.(3) Autograft implantation or utilization of intercalary allografts is mostly used for patients with lesions that are large or seem to threaten the integrity of the bone. Because the lesion may produce heavy bleeding, tourniquet control is advised. Marginal resection sometimes is indicated for lesions in expendable bones. Lesions in the spine or pelvis can be treated with preoperative embolization to minimize surgical blood loss. Arterial embolization has been used as definitive treatment of aneurysmal bone cysts in locations where curettage would be extremely difficult. Low-dose radiation has been reported to be an effective method of treatment, often associated with rapid ossification; however, it is not used routinely because of the potential for malignant transformation.

The use of calcium hydroxyapatite (HA) to fill and repair bone defects, either alone or used together with autograft, is well established.(5,6) Biological, histological and biomechanical surveys of its implantation into bone reveal that the material is safe and has excellent osteoconductive properties. Although it is biodegradable, HA is only very slowly replaced by new bone, so that its structural properties remain intact for long without weakening the bone. When used in periarticular lesions beneath the subchondral bone plate, it doesn't cause any joint degeneration. Hamson et al(7) found no significant difference between HA and autologous cortical bone in mechanical strength, six months after implantation. Vuola et al(8) reported that the compressive strength of HA was higher than bone marrow after 12 weeks. It has been reported that extraskeletal implantation of HA does not lead to osteoinduction. Although HA in the soft tissues can be a source of mechanical irritation, it very rarely causes a histological foreign body reaction. It is not reported to affect tumour recurrence rate. As a result of these properties, HA has replaced autologous bone graft and other substitutes in treating benign bone tumours.

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