CASE REPORT

UNICAMERAL BONE CYST OF THE TALUS IN ADULT: A CASE REPORT

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ABSTRACT: We report a case of unicameral bone cyst of talus in an adult patient. The patient presented with pain and swelling following a twisting injury of ankle. Clinicoradiological diagnosis was unicameral bone cyst (UBC). The lesion was treated with curettage and autogenous iliac bone grafting. Diagnosis was confirmed by histopathology. Two years follow-up showed successful incorporation of autogenous bone graft and obliteration of lytic lesion.

KEYWORDS: UBC, Talus, Adult patient.

INTRODUCTION: The word "UNICAMERAL" derived from Latin: unus =one and camera = vault. It is also known as simple bone cyst. 80% of UBC occur in the proximal humerus and proximal femur.^[1,2] Other sites of involvement include ilium, calcaneum & talus, usually found in older patients. Most of UBCs occur in childhood where one third of the cases will resolve spontaneously by skeletal maturity.^[1,2] These lesions constitute approximately 3 % of all bone tumors, and usually involve the metaphysis of long bones.^[2,3] Rarely does a cyst progress after closure of the growth plates. Very few cases of UBC of talus in adulthood had been reported in world literature. We are presenting a case of UBC of talus in adult which was successfully treated with curettage and autogenous bone grafting.

CASE REPORT: A 37 years old male patient presented with pain and swelling of left ankle following a twisting injury. There was no other systemic symptoms. On examination there was swelling of the left ankle, no local rise of temperature, left talus was tender to palpate and ankle movements were painful. Routine biochemical tests, blood count and erythrocyte sedimentation rate (ESR) were within normal limits. AP and lateral radiographs of the ankle revealed a radiolucent lesion measuring about 19mm in diameter at the centre of the talus with round margins. There was no scalloping or septae formation. The percentage of the bone occupied by the cyst was more than 85% in both the radiological planes. Magnetic Resonance (MR) imaging of left ankle revealed a well-defined uniloculated osteolytic lesion with fluid filled levels involving the dome and posterior part of body of talus.

As the risk of fracture was high, curettage & autogenous bone graft was planned. We performed, excision-curettage followed by autogenous cancellous bone grafting. Under spinal anaesthesia the left talus was explored through the posterolateral approach. Talus revealed a thinned out posterior cortex. With the removal of posterior cortex, a reddish coloured fluid was exposed probably due to trauma which was evacuated to reveal a large cavity. The lining membrane of the cavity was curetted and the obtained material was sent for histopathological confirmation. The cavity was drilled at multiple sites into the talar neck to create vascular bed for the osteointegration of bone graft. The cavity was tightly packed with autogenous iliac crest bone graft, which was taken from the ipsilateral iliac crest. Postoperatively the operated limb was immobilized by a below knee slab for 3 weeks and converted to below knee cast for another 3 weeks.

CASE REPORT

Postoperatively, the patient was followed up every 6 weeks with x-ray. The patient was allowed to partial weight bearing after 12 weeks and full weight bearing after 16 weeks. The histopathology report confirmed UBC.

The patient was followed for 14 months at interval of 6, 12, 18, 24 weeks and at one year. At the latest follow up of two years, there was no evidence of any recurrence & there was complete osteointegration of graft with complete obliteration of the cavity. There was no evidence of collapse of articular surface with complete range of ankle movements and pain free ankle.

DISCUSSION: A unicameral (simple) bone cyst is a cavity found within a bone that is filled with straw-colored fluid. It is a benign condition. Natural history of these defects is that they are most active in children younger than age 10 years and later become inactive. Most diagnosed UBCs occur in childhood.^[1,2,3] UBC etiology is unknown.^[1,2,3] They account for 3% of all bone tumors, and usually involve the metaphysis of long bones, and have a predilection for the proximal humerus and proximal femur^[2,3] with the proximal humerus being affected two to three times more frequently than the proximal femur.^[4] Other sites of involvement include ilium, calcaneum & talus which is usually found in older patients. Lesions may be asymptomatic & identified incidentally. Some patients may present with swelling & stiffness of the adjacent joint. In our case cyst was inactive and diagnosed incidentally following a twisting injury.

UBC is not difficult to diagnose radiologically, but MRI has been reported as helpful in differentiating UBC & ABC (Aneurysmal bone cyst) in troublesome cases. Diagnosis is made by computed tomography (CT), magnetic resonance imaging (MRI), and histopathological study. The radiological features on plain x-rays include a well marginated, centrally located, uniloculated radiolucent, expansile lesion of the metaphysis. Cortical thinning without disruption is seen. Fallen fragment sign is virtually pathognomonic of a uniloculated bone cyst, which is rarely seen in talus.

In MRI, prolonged T1 and T2 relaxation times suggest a cyst, although T1 shortening may reflect proteinaceous content resulting in signal which is higher than water.^[5] MRI most accurately delineates the central fluid collection.

The most characteristic histopathologic finding is the thin membranous lining of the cyst, composed primarily of flattened to plump epithelium-like cells; the lining may also possess osteoclast-type giant cells, cholesterol cells and fat cells. Hemosiderin, fibrin, calcification, and reactive bone may be seen in focal areas of the cyst.^[6]

The differential diagnosis of lytic lesion in talus includes giant cell tumour, aneurysmal bone cyst, intraosseous lipoma and chondroblastoma. While radiologically Giant cell tumor appears as lytic lesion in the epiphysis, which bulges beyond the confines of the cortex and has multiloculated with soap bubble appearance. Aneurysmal bone cyst usually appears as eccentric, metaphyseal with thinning of cortex and multiloculated radiolucent lesion. Radiology and MRI imaging of intraosseous lipoma reveals a well-defined expansile lesion with fat density and central calcification. Chondroblastoma will show an osteolytic lesion with lobulated margin with MRI showing typical expansive peritumoral oedema in the entire bone.

The goal of management of UBC is the formation of a bone that can withstand the stresses of use by the patient without evidence of continued bone destruction as determined by serial radiographic follow up. There are no established guidelines for when and how to treat UBCs. The management of UBC varies from percutaneous needle biopsy, injection of local steroids,

CASE REPORT

demineralized bone matrix, and autogenous bone marrow infiltration have been reported as methods of treatment with various success rates.^[1,2] Standard surgical treatment consists of curettage & cancellous bone grafting (with autogenous or allogeneous bone).^[2] The main indication for surgical intervention is to prevent or treat a pathological fracture, to promote healing, to avoid cyst recurrence & refracture.^[1,2] The best treatment for UBC is yet to be identified. Curettage with bone grafting is indicated if the structural integrity of bone is at risk. If the percentage of the bone occupied by the cyst is more than 85% in both radiographic plane, the risk of fracture is very high and spontaneous healing usually do not occur.

Treatments for unicameral bone cysts vary depending on their location and patient age. For asymptomatic lesions with satisfactory cortical thickness, observation alone may suffice. For lesions with unstable cortical thinning (with or without pain), surgical intervention is necessary.

In children, about 15% of the cysts heal without treatment.^[7,8] Up to 15% of the cysts may heal after occurrence of a fracture with observation alone.^[8,9] Other treatments include intra-cystic injection of methylprednisolone acetate (to decrease the secretion of the synovial fluid and increase the rhythm of bone cell duplication),^[8] mechanical disruption of the cyst lining and/or wall by curettage, structural support with decompression with multiple drill holes.^[10] Intra-cystic injection of steroids achieves less morbidity and higher healing rates than curettage and bone grafting (70% vs. 53%).^[11] Nonetheless, a multicentre study (Glaser et al) suggested that such treatment is less effective in calcaneal and talus lesions, for which curettage and bone grafting may be a more predictable and successful procedure. A new minimally invasive technique that combines percutaneous decompression and grafting with medical grade calcium sulphate pellets has been reported.^[12]

CONCLUSION: Unicameral bone cyst is more common in children and very rare in talus. We reported a case of talar unicameral bone cyst which was incidentally diagnosed in an adult individual, and subsequently treated with excision-curettage and bone grafting successfully.

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CASE REPORT

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Figure 1: Pre-op radiograph showing well defined osteolytic lesion of talus & MRI showing well defined, uniloculated, osteolytic lesion with fluid level involving the dome and Posterior part of body of talus with intrinsic haemorrhage suggestive of unicameral bone cyst.

Fig. 1

Figure 2: Operative photographs showing poster lateral approach, revealing a large cavity on the posterior aspect of the body of talus which has been bone grafted with autologous iliac crest bone after multiple drilling of the cavity.

Fig. 2

CASE REPORT

Figure 3: Immediate postoperative radiograph showing the well packed autogenously bone graft & below knee POP plaster.

Fig. 3

Figure 4: Histopathology showing uniloculated cyst containing blood tinged fluid with thin, several cell thick lining with deeper layers of fibrous tissue, giant cells containing hemosiderin and cholesterol clefts and small nucleus with occasional foam cells and chronic inflammatory cells, suggestive of unicameral bone cyst.

Fig. 4

Figure 5: Six months follow-up radiograph & clinical photograph showing well consolidated bone graft and well defined articular surface with good range of movement of ankle.

Fig. 5

CASE REPORT

Figure 6: Two years follow-up radiograph showing complete obliteration of osteolytic lesion with no evidence of collapse of articular surface or avascular necrosis or recurrence.

Fig. 6

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