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Original Article

Surgical Management of Pathological Fracture of Neck of Femur Following Unicameral Bone Cyst

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ABSTRACT

Unicameral bone cyst (UBC) is a benign cystic lesion most commonly diagnosed in the proximal aspects of the humerus and femur of growing children. Medullary venous obstruction is the leading pathogenesis theory, resulting in fluid accumulation, bone resorption, and cortical thinning. Most UBCs are asymptomatic and likely go undiagnosed, while the most common presentation is a pathologic fracture. UBC of the proximal femur exhibits unique characters and complications. Post-excision stabilization of the cyst is recommended to avoid malunion and facilitate post-operative rehabilitation and an earlier return to normal activities. In the present case report, surgical treatment of a pathological neck of the femur fracture following a unicameral bone cyst was managed with internal fixation of the proximal femoral plate with bone grafting, which showed a good clinical and radiological outcome.

Key Words: *Unicameral bone cyst, benign cystic lesion, children, humerus, femur*

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INTRODUCTION

Unicameral bone cyst (UBC) is defined as an atrophic degenerative osteolytic process consisting of a cavity filled with fluid and lined by a membrane [1]. Its highest incidence occurs in the first 2 decades of life [1], and 50% of upper femoral lesions occur between the ages of 17 and 54 [2]. It usually arises in the metaphysis of long bones immediately beneath the growth plate, and the most common location is the proximal humerus, followed by the proximal femur [1-5] which accounts for 27% of cases [6]. A vascular necrosis (AVN) of the proximal femoral epiphysis and collapse of the articular surface are reported as complications of UBC involving the proximal femur [7-9].

Similar to other benign bone lesions of childhood, most UBCs are asymptomatic and likely go undiagnosed unless there is an incidental finding or pathologic fracture. Symptomatic patients typically present with either an acute pathologic fracture or an insufficiency/stress fracture. In the latter group, the patient might present with mechanical symptoms, a painful range of motion, or limping. In a systematic review, more than three-quarters of the published UBC cases were diagnosed after an acute fracture, 15% with an insufficiency fracture, and the rest as an incidental finding[10].

UBCs appear as centrally located, well-defined, lucent meta physeal lesions on plain radiographs, with a narrow zone of transition, and often abutting the physis (active phase). With longitudinal growth, the lesion “migrates” away from the physis and becomes diaphyseal (latent phase). Larger lesions may lead to bone expansion and result in cortical thinning. A periosteal reaction or cortical destruction is absent unless a fracture occurs. Soft tissue extension is not a feature. The “fallen fragment sign” was first described by Reynolds to help distinguish UBC from other radiolucent but solid intramedullary lesions [11,12].

Among the wide range of different modalities described for treatment of UBC are: radical excision in the form of subperiosteal partial diaphysectomy and allograft [13], subtotal resection with [14] and without bone graft [15], curettage and bone graft [4], multiple drill holes [16, 17], intra-cystic prednisolone injection [3, 5, 7], and recently, intramedullary flexible nails [18, 19]. This case report is to assess the clinical and radiologic outcome of surgical treatment of a pathological fracture neck of the femur following UBC of the proximal femur.

Case report

A 32-year-old male presented with a history of left groin pain and an inability to bear weight over the left lower limb for 3 months. Examination revealed a left hip in an attitude of external rotation, tenderness over the left greater trochanter, and painful restriction of hip movements. After routine blood investigations and a plain radiograph of the pelvis with both hips AP and lateral, the following CT and MRI hip joints were done.

Fig. 1: A plain X-ray hip shows a centrally located, lytic lesion, well marginated outline and a breach in the cortex of the neck femur. X-ray also showed a fallen leaf sign

Fig. 2: CT pelvis scan suggests an osteolytic lesion in the neck and trochanteric area of the femur

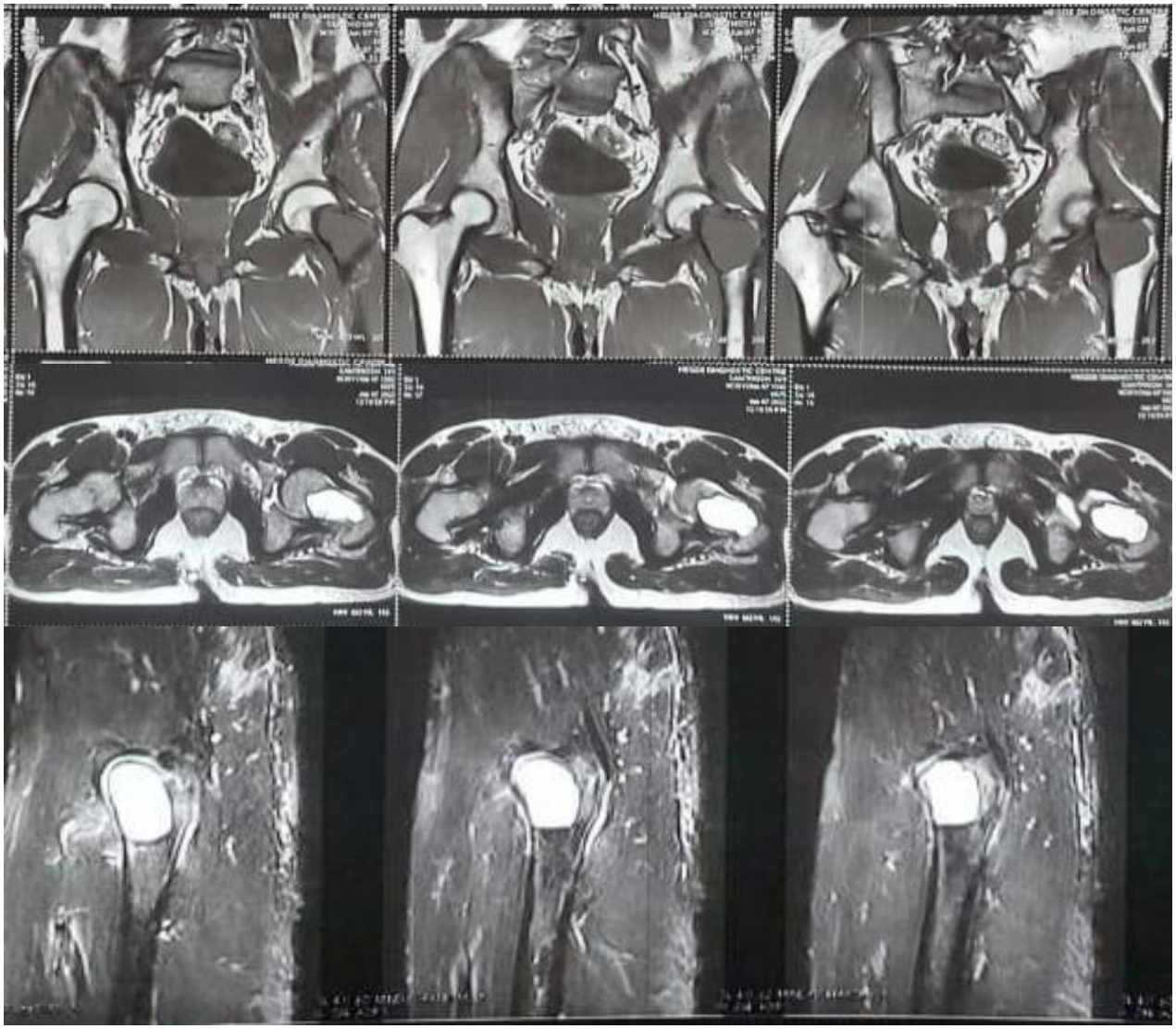

Fig. No 3 An MRI of the pelvis shows an expansile lytic lesion in the metaphysis of the neck of the left femur.

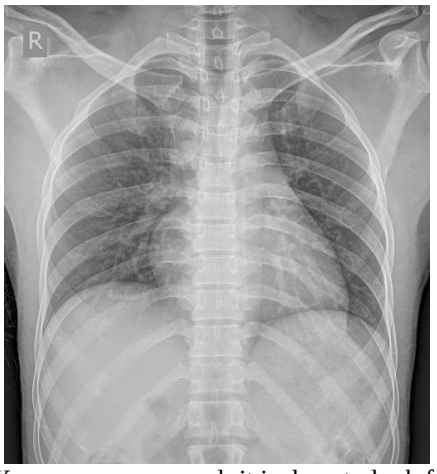

Fig. no 4: A chest X-ray appears normal; it is done to look for lung metastasis.

Following PAC clearance, the patient was taken up for intralesional curettage and Intra operative frozen section was done, diagnosis was confirmed. Contralateral iliac crest auto graft and freeze-dried allograft with internal fixation with locking plate was done under spinal anesthesia.

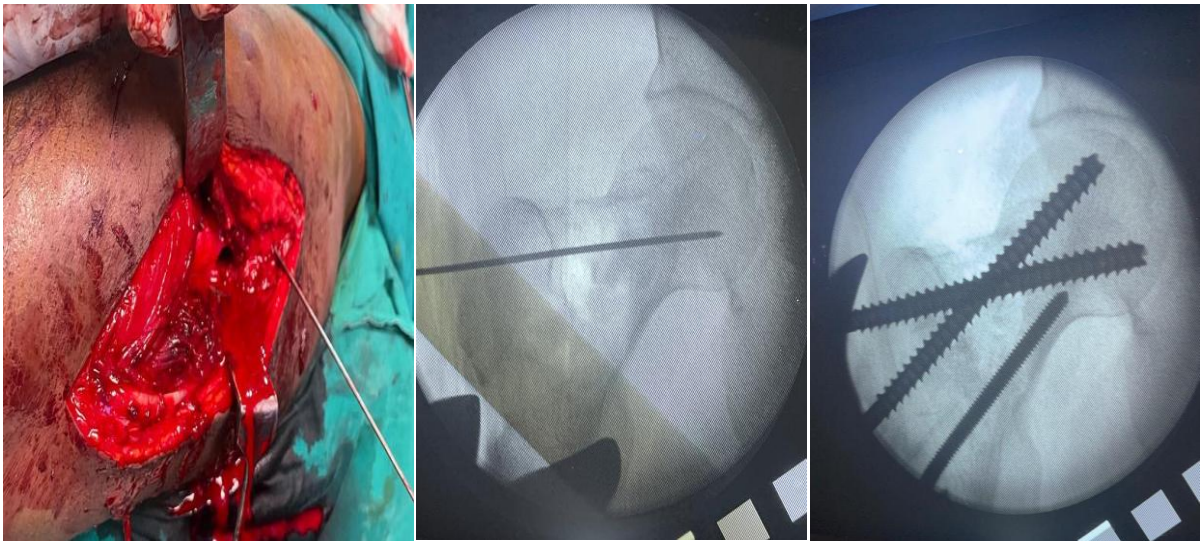

Fig.No 5: Intra operative gross and c arm pictures

Postoperatively, the histopathological diagnosis was confirmed as a unicameral bone cyst. The patient was followed up at regular intervals and at a 4-month follow-up with full functional recovery. A 4 months postoperative X-ray showed incorporation of the bone graft and complete healing of the fracture with the implant in situ. 4 months postoperative X-ray revealed incorporation of bone graft and complete healing of fracture with implant in situ.

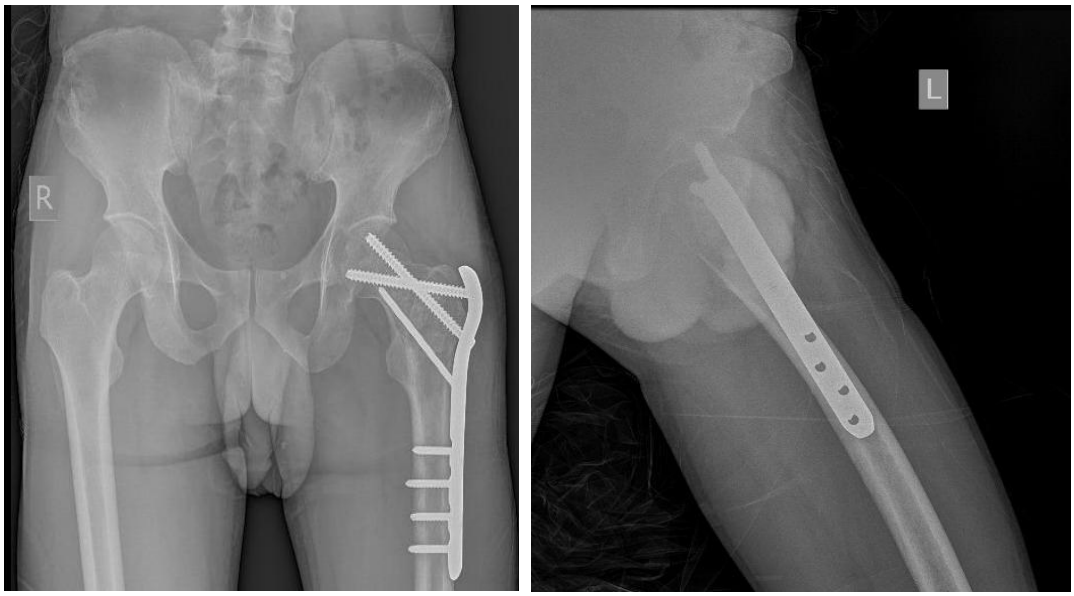

Fig. no: 6 Immediate Post-operative Hip and Pelvic X-ray (AP and Lateral)

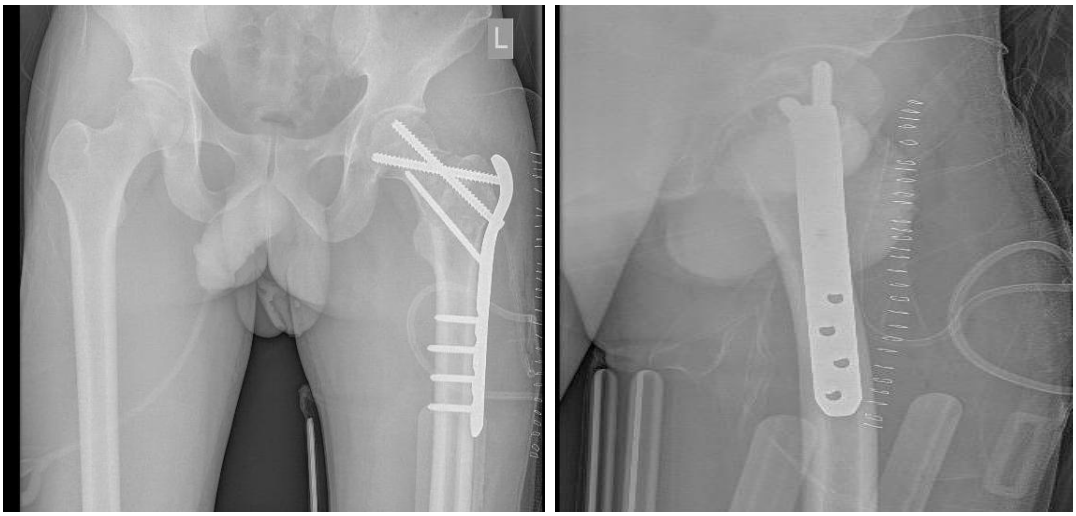

Fig. No 7: 4 months follow-up Hip and Pelvis X-ray (AP and Lateral)

Fig. No. 8: Left hip range of movements after 4 months with a healed surgical scar over the lateral aspect of the thigh

DISCUSSION

The exact etiology and pathogenesis of UBC remain unclear. The most popular pathogenesis theory is a blockage in the intramedullary venous drainage postulated by Cohen [20,21]. In brief, an insult to the bone causes venous obstruction, which in turn leads to fluid accumulation, increased pressure, and bone resorption [20-22]. Several therapeutic approaches have been established based on this mechanism, including decompression or fenestration of the cyst

Although UBCs may occur at any site, the metaphysis and diaphysis of long bones are the most common locations. More than half of the lesions are diagnosed in the proximal humerus, followed by the proximal femur and proximal tibia [10]. The calcaneus is also a relatively common site for UBC, which tends to occur in the slightly older population.

Advanced imaging may be valuable in differentiating a UBC from similar lesions in equivocal cases. A computed tomography (CT) scan shows cortical thinning, fluid density inside the lesion, and occasionally the fallen fragment sign [23]. On magnetic resonance imaging (MRI), UBC is characterized by a hypointense signal on T1- weighted images and a hyperintense signal on T2-weighted and fluid-sensitive sequences. Septations, if present, are also visible on MRI. On post-contrast images, peripheral enhancement is appreciated. When a fracture is present, T1-weighted images will be hypersensitive due to blood, while marrow edema, periosteal reaction, and surrounding edema are hyperintense on T2-weighted and fluid-sensitive sequences. Fluid-fluid levels may be present, especially in the presence of a pathologic fracture. MRI is particularly useful for the evaluation of a suspected insufficiency fracture and physeal involvement/ disruption by the cyst [23].

Over the years, several management strategies and surgical techniques have been introduced for UBC. Neer et al. reported surgical treatment of 129 cases of UBC by curettage (ILE) and bone graft. They evaluated 24 out of 31 cases located in the proximal femur and reported recurrence in four cases (17%). The results of allograft were compared to autograft used to fill the defect after curettage of 93 cysts located in the proximal femur and humerus. In 35 cases treated with autograft, 21 cases (60%) were excellent; six cases (17%) showed residual defects; and eight cases (23%) required re-operation. In 58 cases treated with allograft; 28 cases (48%) were excellent, 12 cases (48%) showed residual defects, and 18 (31%) required re-operation.

Thus, autograft was slightly better [4] and therefore indicated in recurrence when there is a sufficient quantity of bone that can be conveniently taken to fill the defect [2, 4]. Allograft is indicated for the pediatric age group with large cysts [2]. Campanacci et al. did not find any relation between type of bone graft and rate of recurrence and pointed out the importance of packing the cyst, as residual empty spaces might be a source for recurrence [3].

Campanacci et al. compared 178 cases of UBC treated by curettage and bone graft to 141 cases treated by methylprednisolone injection. The recurrence rates were 33 and 15%, respectively. A pathological fracture developed in two cases during injection treatment and in another 11 cases after the recurrence of the cyst. AVN of the femoral head was observed in one case treated by injection therapy [3]. In the present case report, surgical treatment of a pathological neck of the femur fracture following a unicameral bone cyst managed with internal fixation with a proximal femoral plate and bone grafting showed a good clinical and radiological outcome.

CONCLUSION

UBC of the proximal femur needs special consideration as it exhibits unique characteristics and complications. Each case should be evaluated individually.

The conclusive findings from this case report are:

- Intralesional curettage and bone grafting with internal fixation of fracture neck of femur using proximal femur locking plate leads to good clinic radiologic outcome.
- Surgical stabilization of the cyst is recommended to avoid mal-union and to facilitate post-operative rehabilitation and earlier return to normal activities.
- Surgical stabilization also lessens the risk of additional surgeries.

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