

A Rare Case Of Bilateral Unicameral Bone Cyst In An Adult

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KEYWORD

Unicameral
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tumour,
proximal femur

ABSTRACT:

Introduction: Unicameral bone cysts (UBCs) are benign cavities in immature bones, affecting 3% of bone tumors. They can weaken surrounding bones and increase fracture risk. Management varies, especially in children and adolescents. Several hypotheses exist, but cause remains elusive. **Aims:** The study investigates the outcomes of operative management of a pathological fracture caused by a Unicameral Bone Cyst in the proximal femur, along with the discovery of the same tumor. **Methodology:** A 27-year-old male patient with minor injury was diagnosed with a comminuted pathological fracture in the left intertrochanteric femur, requiring bilateral surgery to prevent future fractures. **Results:** The patient's suture lines healed and suture removal was performed, and after 1.5 months of partial weight bearing, they were allowed full weight bearing walking by 3 months. **Discussion:** A 27-year-old patient with a comminuted femur fracture was found to have a bilateral unicameral bone cyst, a rare occurrence in adults, potentially contributing to structural weakness. **Conclusion:** This case report discusses the clinical manifestations of bilateral unicameral bone cysts, emphasizing the importance of thorough examination, MRI scans, and prophylactic surgery for accurate diagnosis.

Introduction:

Unicameral bone cysts (UBCs), also known as simple bone cysts, are benign fluid-filled cavities that primarily occur in the long bones of skeletally immature individuals, particularly in the proximal humerus and femur. First recognised by Virchow in 1876, UBCs account for nearly 3% of all bone tumours¹. Though generally asymptomatic and often discovered incidentally, they can weaken the surrounding bone, increasing the risk of fractures. This paper aims to review the current evidence-based diagnostic and treatment modalities, addressing controversies and challenges associated with managing these lesions, particularly when they occur in locations such as the proximal femur interestingly bilaterally in this particular case.

While the etiopathogenesis of unicameral bone cysts (UBCs) remains largely unclear, with various theories proposed to explain their development, their clinical presentation and radiographic features are well-established. Despite their common occurrence, particularly in children and adolescents aged 5 to 15 years when skeletal growth is most active, there is considerable variability in the management of unicameral bone cysts (UBCs)¹. These cysts are most commonly found in the proximal humerus and proximal femur, although they can occasionally arise in other long bones or, more rarely, in locations such as the pelvis. In most of the cases the presentation is unilateral with slim chances of the cyst occurring bilaterally. There is a slight male predominance, with boys being affected about twice as often as girls². Several hypotheses have been proposed to explain the occurrence of UBCs, including venous obstruction leading to increased intra osseous pressure, a disturbance in bone growth regulation, or abnormal activity in the growth plate, which may create a localised area of poor vascularity or fluid accumulation. Despite these theories, the precise cause remains elusive, and further research is needed to fully understand the underlying mechanisms that contribute to the formation of UBCs^{1,2}.

Aim :

To study the results of operative management of a pathological fracture due to Unicameral Bone Cyst of proximal femur along with the incidental finding of same tumour at the similar site on opposite side.

Materials And Methods:

A 27-year-old male patient presented to the Orthopaedics OPD of a tertiary care center in Karad with a history of a trivial injury. He complained of pain in his left hip and an inability to bear weight on the left hip joint. He was diagnosed with Comminuted Pathological Fracture Intertrochanteric Femur Left Side after undergoing a series of Xrays.



Fig 1 - On Admission Xray Pathological fracture with Fallen leaf sign

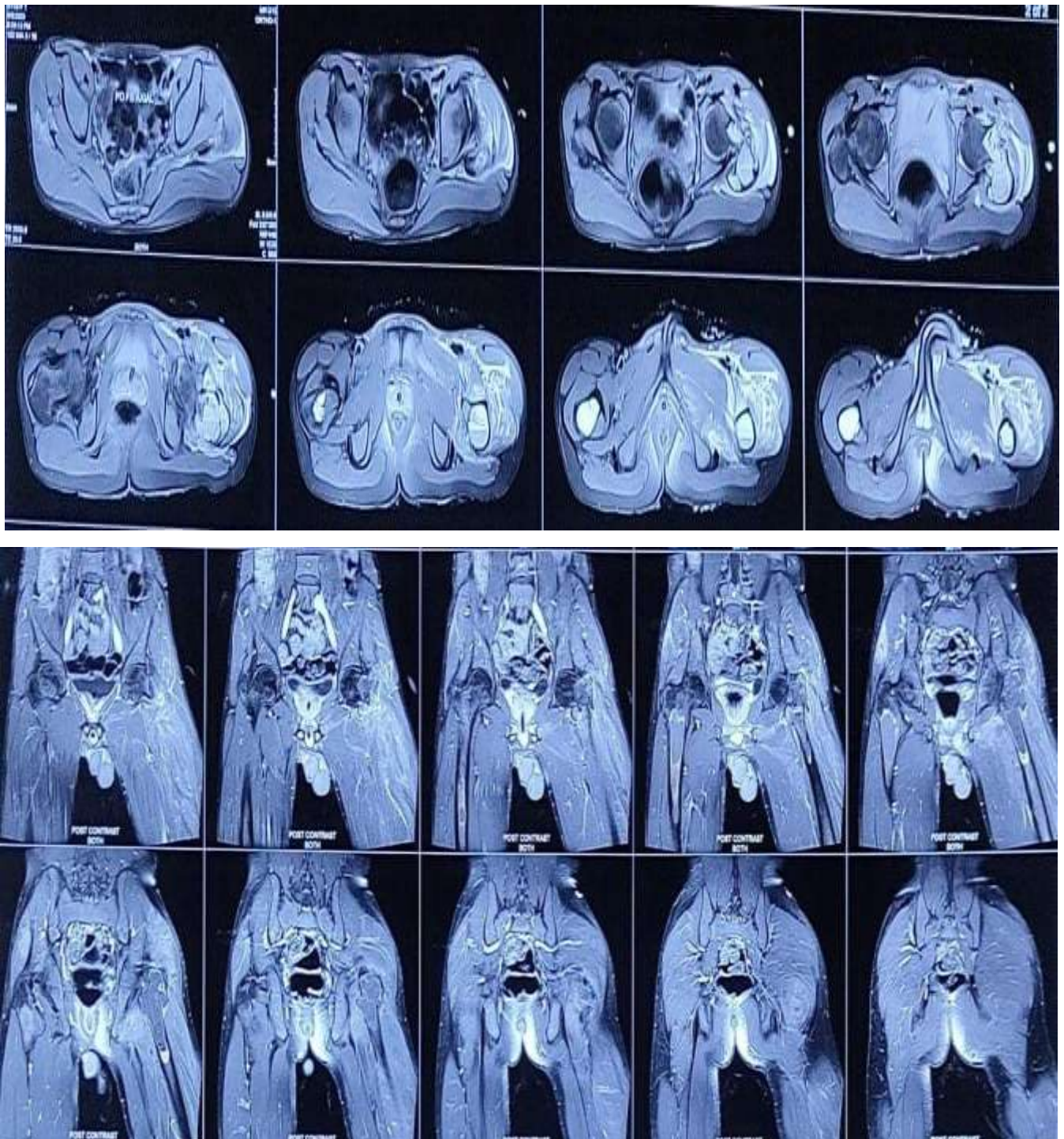
Patient was thinly built and fragile and on further history taking it was found that the fracture had occurred as a result of low velocity trauma. Possible differentials for the case included hyperthyroidism , bone tumours , osteoporosis , aneurysmal bone cyst , etc

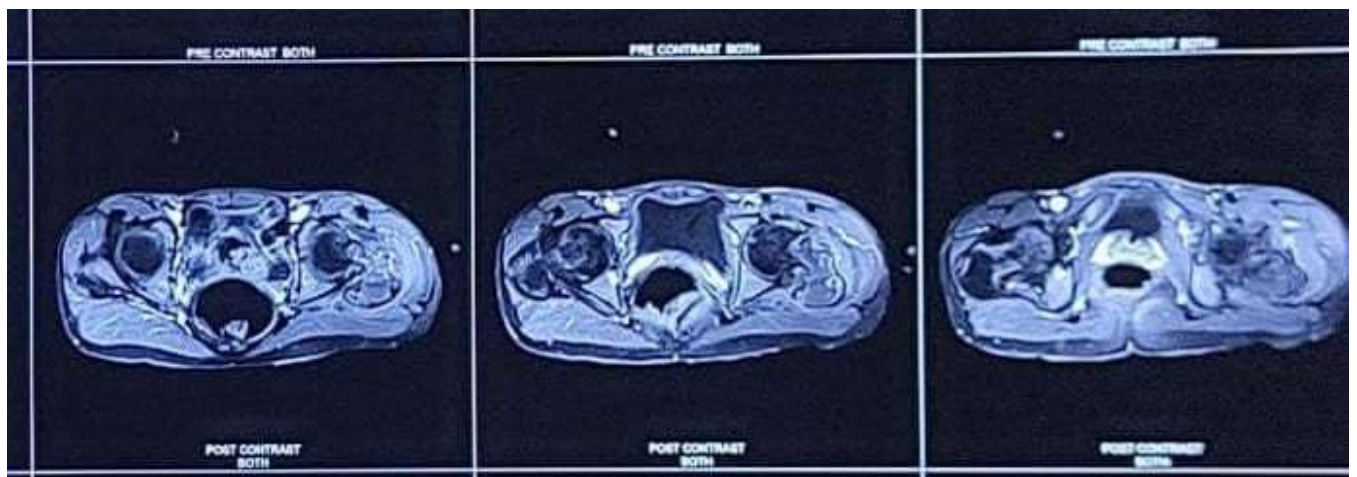
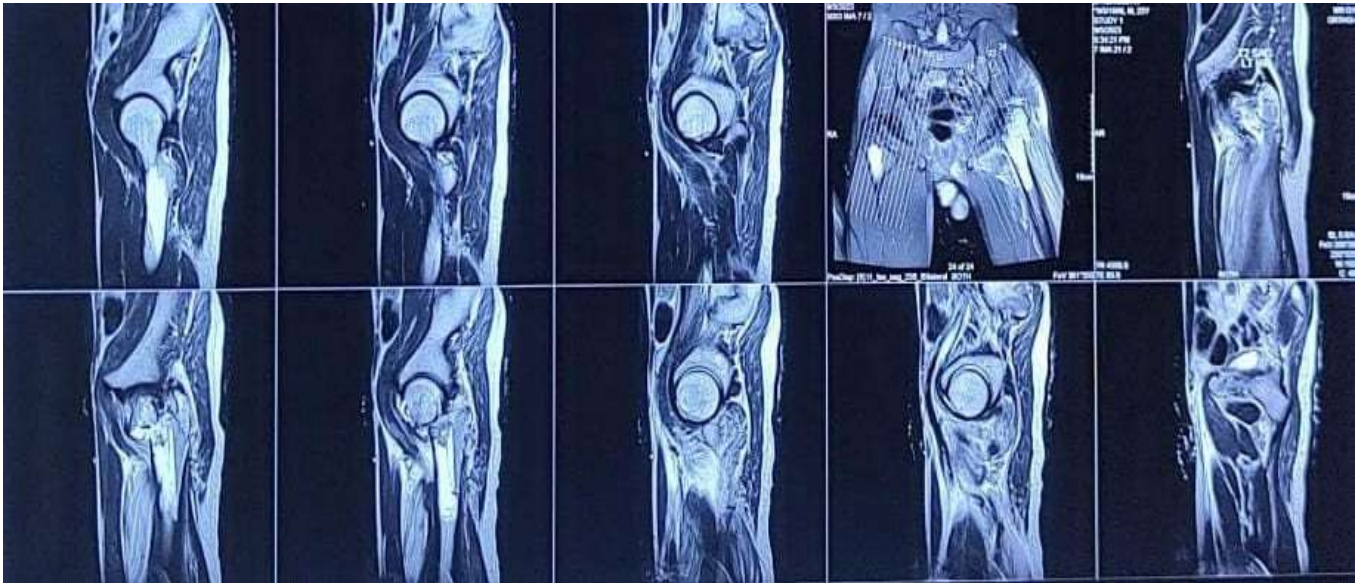
In order to reach a clear diagnosis an array of investigations were sent viz. blood work to rule out metabolic conditions , USG local part neck to rule out any changes in the thyroid or hypoparathyroid glands, Ct scan of pelvis to study the fracture and extent of the lesion, MRI to see if any metastatic or invasive modality was present and biopsy to confirm the diagnosis

Investigations-

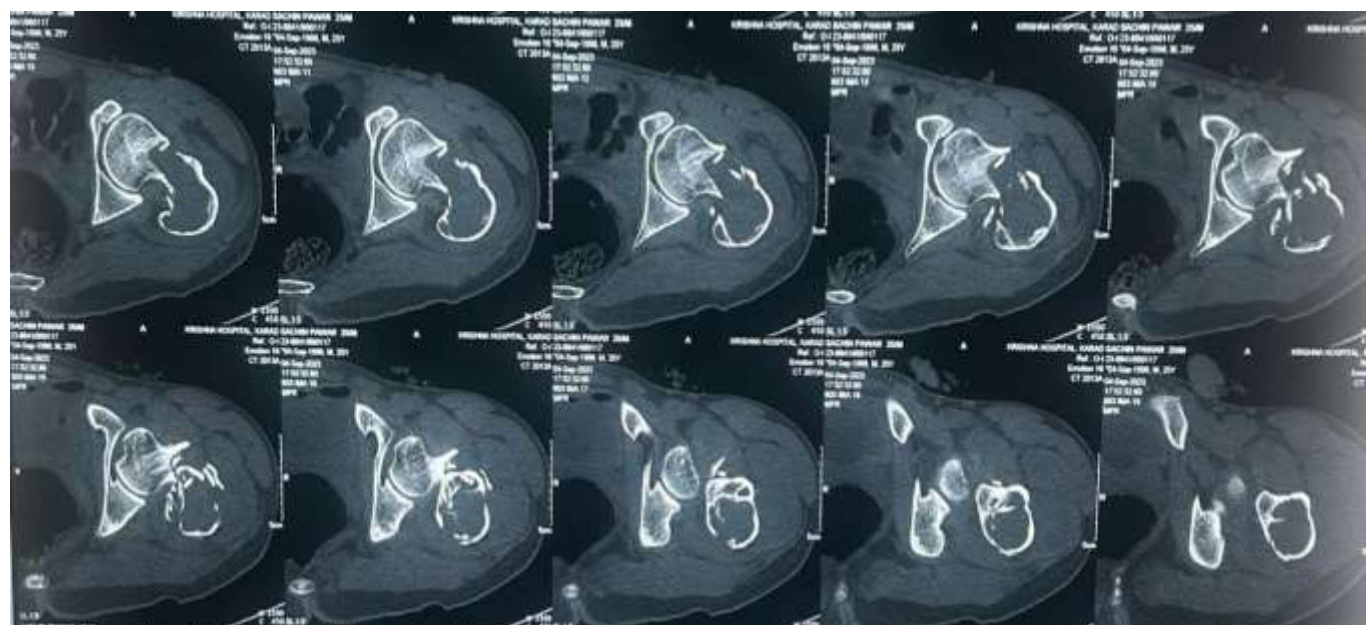
- 1) Blood Investigations yielded the following results:
 - PT-17 [Normal- 13]
 - INR 1.4 [Normal -1.1]
 - T3-212 [79-158]
 - T4-12.5 [4.9-11]
 - TSH- below 0.01 [0.38-4.31]
 - TB-1.8, DB-0.5, IB-1.3 =slight increase
 - Acid phosphatase-75 [normal-0-5.4]
 - Serum calcium-10.9 [upto 10.5].
- 2) PTH levels were low indicating a hypoparathyroid state
- 2) USG neck was suggestive of thyroiditis with spongiform generation
- 3) PSA - 6.56 (<4)
- 4) CT PBH showed 'Multifragmentary displaced fracture of neck and intertrochanteric region of left femur with few fracture fragments displaced superiorly and laterally'
- 5) HPE findings were as follows:- Section studied shows bone marrow tissue with very few bony trabeculae and predominantly hematopoietic element. Trilineage hematopoiesis with predominance of myeloid lineage is evident. Erythroid and megakaryocytic lineage appears unremarkable. Simple bone cyst with thin fibrous lining containing fibrous tissue, giant cells, and hemosiderin pigment seen, chronic inflammatory cells found in small numbers.

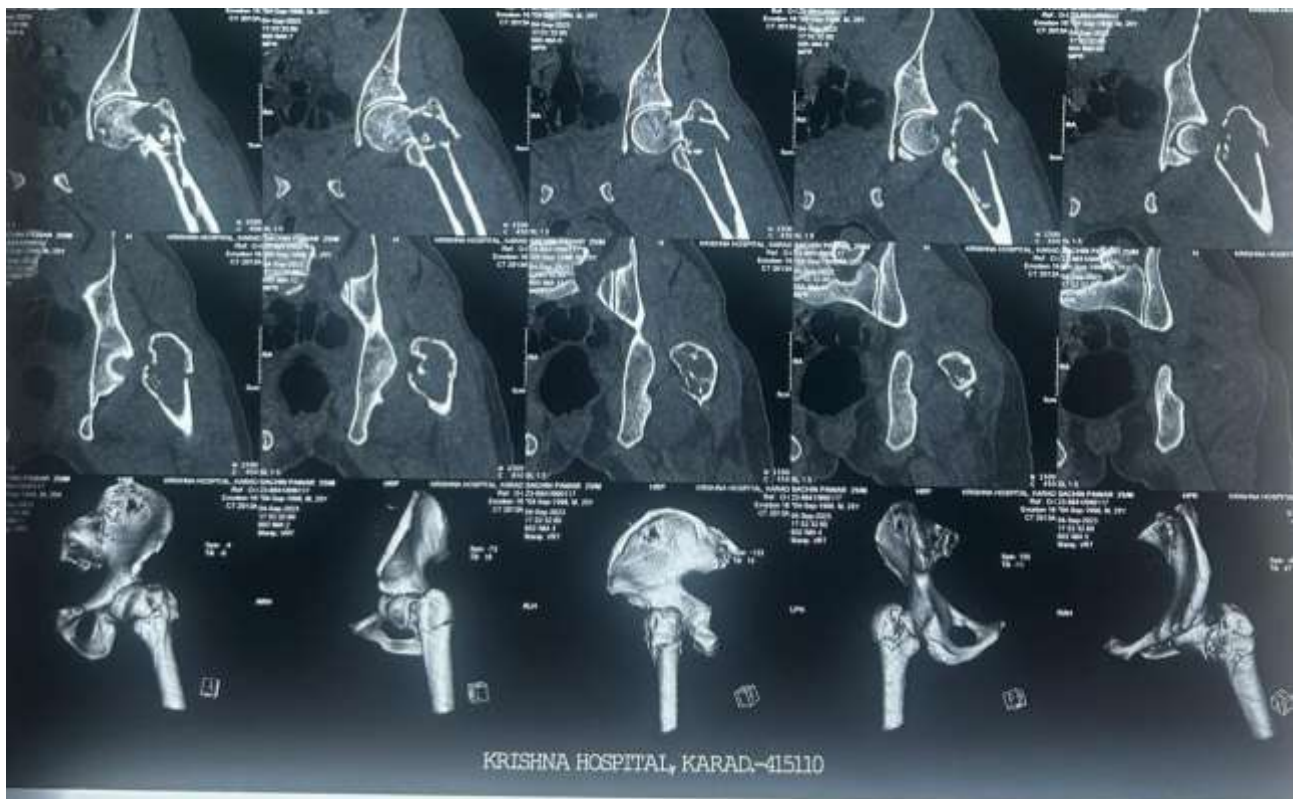
MRI report - Expansile multi lobulated lytic lesion in neck and intertrochanteric region of left femur with morphology and signal characteristics as described - likely aneurysmal bone cyst with pathological intertrochanteric fracture. There was incidental finding of long segment expansile aneurysmal/simple bone cyst noted in proximal meta-diaphysis of right femur for cranio - caudal length of ~ 12 cm similar to left





MRI FILMS





CT FILMS

On the basis of all these findings the diagnosis of unicameral bone cyst left side was decided. Surprisingly the MRI report showed the presence of a similar cyst present in the proximal femur of the right side. Taking into consideration all these factors it was decided to operate the patient bilaterally - prophylactically operating the right non fractured limb to prevent it from getting fractured in the future owing to cortical thinning caused by the cyst.

After one week of conservative management and medical fitness done in ward , patient was operated with ORIF with Long PFN and TSP on the left hip and with Long PFN on the right hip.

Below are the follow up Xrays for the patient -



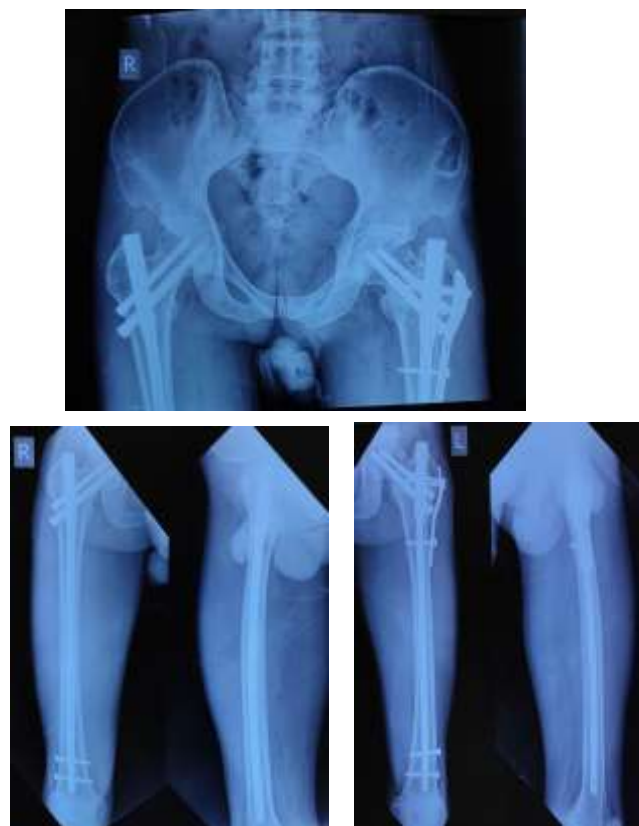
Immediate post operative



One month post operative



6 Months post op



1 Year post op

Results:

Both suture lines healed as expected and suture removal was done on POD 15. Ankle movements, Static quadricep exercises were started on POD 2. Dangling was started by POD 2

At 1.5 months follow up patient was advised partial weight bearing with walker support. On later follow ups the X-rays showed excellent callous formation and with stable reduction over both the limbs and thus the patient was finally allowed full weight bearing walking by POD 3 months Below we have added few pictures following the recovery phase of the patient

Discussion:

In this case of a 27-year-old patient with a comminuted intertrochanteric femur fracture, the incidental finding of bilateral unicameral bone cyst (UBC) is both unusual and significant. While UBCs commonly occur in children and adolescents, their presence in an adult is rare, as these cysts typically resolve or stabilise by skeletal maturity. The incidental discovery of a UBC in an adult with a traumatic femoral fracture suggests that the cyst may have persisted into adulthood, possibly contributing to the structural weakness of the bone and predisposing it to fracture under stress ³.

Another factor what makes this case rare that a similar cyst was found to be present on the opposite side - bilateral presentation of unicameral bone cyst is a rare phenomenon in its own accord. In cases of comminuted intertrochanteric fractures, which involve multiple bone fragments and require complex management, the presence of a UBC can complicate treatment planning. Surgeons must consider whether the cyst contributed to the fracture and assess the need for additional measures, such as curettage, bone grafting, or internal fixation, to stabilise the cystic bone and prevent further complications. This case highlights the importance of recognising UBCs as a potential underlying factor in adult fractures, even though they are more commonly seen in paediatric patients ³.

The MRI report done likens the cyst to be a aneurysmal bone cyst but on closer examination of the radiographs we can clearly see that the cyst does not extend beyond the transverse diameter of the epiphysis and diaphysis of the bone which is one of the factors favouring the diagnosis of an unicameral bone cyst. Instead the cyst seems to be extending into the shaft of the bone resulting in the thinning of the cortices of the bone as a result of which the patient suffered a intertrochanteric femur fracture at a young age resulting from a low velocity injury. As the shaft was being weakened by the cyst we operated with chose to operate with Long PFN as our implant of choice along with trochanteric support plate leading to stable fixation of the proximal femur and strengthening the weakened diaphysis of the affected femur. In the same report a similar cyst is said to be present one the opposite side femur but with no resultant fracture. So in the best interest of the patient we operated on the opposite hip with the same rationale to prevent any fracture occurring on that side in the future



Fig 7- Patient performing full weight bearing walking



Fig 8 - Complete knee flexion with excellent range of motion at hips

Conclusion:

This case report explores patients' clinical manifestations of a bilateral unicameral bone cyst. Though bilateral occurrence is rare, in patients with such a cyst present it is always beneficial to do a thorough examination including screening the contra lateral side. Bilateral occurrence should be on the surgeons's differential.

A thorough clinical examination complete with a MRI scan is a huge help in diagnosis. Keeping a open mind with a broader outlook helps to cinch the diagnosis. Willingness to operate prophylactically keeping the future risks in mind proved to be beneficial for the patient.

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