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Pathological Fracture through Latent Unicameral Bone Cyst of Distal Femur Shaft in a Young Adult – A Rare Case

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Abstract:
Introduction: *The simple or unicameral bone cyst is a lesion of unknown aetiology that is commonly found between the ages of 5 and 15 years. The cysts most commonly occur in the proximal humerus (50% to 60%) or proximal femur (25% to 30%) of a growing child, but other bones may be affected. Unicameral bone cysts often are asymptomatic, unless a pathological fracture has occurred. Hereby we are reporting a case of a pathological fracture in an adult of 22 years through a latent unicameral bone cyst in a distal femur which was treated surgically by curettage, autogenous bone grafting and intramedullary interlocking nailing.*
Case report: *A 22 year old Indian male was referred to our casualty with complain of pain around left knee and inability to stand after a trivial slip from steps at home. On clinical examination, patient had swelling, tenderness, deformity and abnormal movement around left distal femur shaft and primary diagnosis of distal femur shaft fracture was considered. However radiographs of left femur with hip and knee joint of patient revealed displaced femur shaft fracture through a radiolucent cystic lesion with violated integrity of cyst and "Fallen Fragment sign" within cyst. Patient underwent further Magnetic resonance imaging to confirm diagnosis and Bone scan to rule out any malignancy. Patient underwent open curettage , intramedullary interlocking nailing and bone grafting as a primary management . Since there was no literature regarding management of pathological fractures through unicameral bone cyst in adults , we decided to extrapolate management of pathological fractures in children due to unicameral bone cyst to adults and found it helpful.*
Conclusion: *By reporting this case, we hope to alert Orthopaedic surgeons that not only in growing children but also in young adults , one should consider differential diagnosis of Unicameral bone cyst as a cause of pathological fracture in long bones.*
Keywords: *Unicameral bone cyst, Pathological fractures, Curettage and bone grafting.*

I. Introduction

The simple or unicameral bone cyst is a lesion of unknown aetiology that is common in the first two decades of life, primarily between the ages of 5 and 15 years. Boys are affected three times more commonly than girls .The cysts most commonly occur in the proximal humerus (50% to 60%) or proximal femur (25% to 30%) of a growing child, but other bones may be affected .[1] In older children and adults, cysts occur most commonly in the calcaneus or flat bones.[2]

Unicameral bone cysts often are asymptomatic, unless a pathological fracture has occurred. A Unicameral bone cyst is usually the most common cause of pathological fracture in children with a bone lesion. The most common sites of pathological fractures due to unicameral bone cyst are the proximal humerus followed by the proximal femur. The natural history of a unicameral bone cyst is to stabilise in size, and, with the natural growth of the bone, drift away from the physis. Although some can heal spontaneously, the majority will become inactive or latent and persist after 12 years of age.

Hereby we are reporting a case of a pathological fracture in an adult of 22 years through a latent unicameral bone cyst in a distal femur which was treated surgically by curettage, autogenous bone grafting and intramedullary interlocking nailing. Pathological fracture through latent unicameral bone in distal femur shaft of an adult is a rare event and there has been no case reports regarding distal femur unicameral bone cyst complicated by pathological fracture.

II. Case Report

A 22 year old Indian male was referred to our casualty with complain of pain around left knee and inability to stand after a trivial slip from steps at home. On primary clinical examination, patient had mild hypovolemic shock which was managed with wide bore intravenous cannula and crystalloids infusion. On further clinical examination, swelling, tenderness, deformity and abnormal movement were elicited at left distal femur shaft and primary diagnosis of distal femur shaft fracture was considered. Patient had no other musculoskeletal injury on examination. Patient underwent anteroposterior and lateral radiograph of left femur

with hip and knee joint, which revealed displaced femur shaft fracture through a radiolucent cystic lesion with violated integrity of cyst [**Figure 1].** In radiographs, a radiolucent single bone cyst at junction of middle and distal shaft of femur was visualised with thinning of cortex and its long axis aligning along axis of femur. Moreover, a cortical fragment was seen lodged inside a cyst cavity in a dependent manner on lateral radiograph which represents 'Fallen Fragment sign'. At this stage a provisional diagnosis of pathological fracture through unicameral bone cyst was established. For relief of pain and splinting, patient's limb was immobilised in a Thomas splint.

For establishing final diagnosis, patient underwent Magnetic resonance imaging which showed a well defined abnormal area of signal intensity in distal shaft of femur which is expansile and centrally located lesion with narrow zone of transition. The lesion was hypointense on T1W and showed fluid level in T2W with lesion dimensions of 6.5(SI) ×4.6 (AP)× 5.3 (Tr) and " Fallen Fragment sign"[**FIGURE 2**]. Bone scan was also done to rule out any malignancy or cyst elsewhere[**FIGURE 3**]. Since there was no literature regarding management of pathological fractures through unicameral bone cyst in adults , we decided to extrapolate management of pathological fractures in children due to unicameral bone cyst to adults. Thus, open curettage , autogenous bone grafting and intramedullary interlocking nailing was planned. Patient was operated in a lateral decubitus under spinal anaesthesia and curettage of cyst was done by lateral approach to femur. At curettage, it was found that violated cyst was filled with haemorrhagic fluid and lined by thin friable tissue which was scrapped off using ring curettes[**FIGURE 4**]. After curettage , displaced femur fracture was stabilised with intramedullary interlocking nailing (reamed) and cystic cavity was filled with autogenous bone graft. Postoperative radiographs of patient shows acceptable reduction of fracture with cystic cavity filled with bone graft [**FIGURE 5**]. Since postoperative second day, patient has been mobilised non weight bearing with help of a walker and was discharged on fourth day. Histopathological examination of scrapped tissue showed fibrous lining interspersed with few giant cells and no cellular atypia. Patient had complete uneventful recovery from surgery and was followed up at 4 weeks with no pain at fracture site or restriction of motion around left knee.

III. Discussion

The unicameral bone cyst is a benign fluid-filled lesion, located mainly in the metaphyses of the long bones in skeletally immature patients . More a developmental or reactive lesion than a real tumour, it is sometimes classified as a tumor-like lesion. The cavity of the cyst usually contains a small amount of clear yellowish sero-sanguinous fluid. In the case of a pathological fracture through the cyst, the cyst contains blood. The cyst can be in the growing skeleton, i.e. expanding and/or recurring after treatment, particularly if it is in close proximity to the growth plate. These cysts might heal spontaneously in adulthood. They tend to appear in age group of 5-15 years, mostly in boys (2:1). On X-ray, the cyst appears as a well-localized radiolucent lytic lesion in the medullary canal, well differentiated, and without periosteal reaction. When the cyst is complicated by a pathological fracture, periosteal reaction can be seen at the fracture-healing stage. The cyst can expand concentrically but never penetrates bone cortex. The indication for treatment is pain, pathological fracture, or risk of such a fracture, for example, if a large cyst is situated in a weight-bearing area. [6] In 1876, Virchow first described simple/Unicameral bone cyst first time. Since then etiopathogenesis of this entity remained controversial.[6] Proposed pathogenesis of simple bone cyst include increased production inflammatory markers as described by Komiya et al. Increased level of prostaglandin-E2, interleukin-1 and gelatinase induce bone resorption and high intraosseous pressure occurs due to venous obstruction.[5] Some authors classify these cysts as active when they are juxtaposed to the growth plate, and latent when the growth plate has migrated away from the cyst. In epidemiological data of Atlas of Muskuloskeletal tumors and tumor like conditions , P. Ruggieri [8] reported only 43 lesions in distal femur region out of 1481 Unicameral bone cysts registry making our case a rare event.

Fractures through the cysts in the upper extremity can be treated conservatively, contrary to pathological fractures in the femur or tibia where curettage, bone grafting or internal fixation is indicated.[4] Urakawa et al proposed that fracture risks in Unicameral bone cysts were significantly higher in humerus , male sex, with ballooning of bone , thin cortical thickness and multilocular cyst formation. [7] Notably , there are multiple literatures on management of unicameral bone cysts in children but scarcely any literatures have been found showing unicameral bone cysts in distal femur shaft region complicated by pathological fractures and its management. Though there are case reports showing presence of unicameral bone cysts in adults in calcaneus or flat bones but none in distal femur region. Thus, hereby our case report stands as a one of rare event which was treated by curettage, bone grafting and internal fixation.

IV. Conclusion

The simple or unicameral bone cyst (UBC) is a lesion of unknown aetiology that is common in the first two decades of life, primarily between the ages of 5 and 15 years commonly situated at proximal humerus or proximal femur region. Unicameral bone cysts often are asymptomatic, unless a pathological fracture has occurred.

This article reports an occurrence of pathological fracture through a unicameral bone cyst in a young adult through distal femur diaphysis . By reporting this case , it is our hope to alert surgeons that pathological fractures in young adults could be because of latent unicameral bone cyst and possibility of such occurrence should be kept in mind while managing such cases.

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Clinical Message

Unicameral bone cysts and its management is a unsolved riddle yet. Unicameral bone cyst complicated by pathological fracture requires approach to manage fracture as well as healing of cyst. Rare occurrence of unicameral bone cyst and pathological fractures in adults should not surprise surgeon and strategy to treat these entities should be similar to those of children.

Abbreviations

UBC – Unicameral bone cyst , SI – Superoinferior , AP- Anteroposterior , Tr – Transverse

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

UGW- Searching Literature. Writing case report and discussion sections. Collecting radiological investigations, Assistant surgeon in surgery. **AK**- Main Surgeon. Searching Literature. **SR** - Contributor in writing case report and collecting images. Editing case report. Searching Literature. **PR** – Editing Case report and discussion . All authors read and approved the final manuscript.

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Conflict of Interest

Nil.

Illustrations and Figures

Figure 1 - Anteroposterior and Lateral view of distal femur showing pathological fracture Through unicameral bone cyst with "fallen fragment sign"

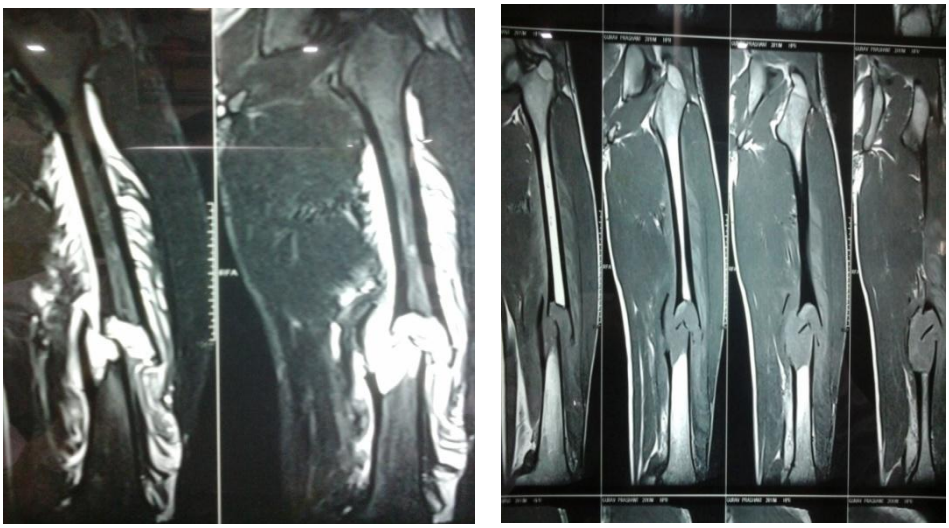

Figure 2 – T1W and T2W Magnetic resonance imaging images of unicameral bone cyst with Fracture showing fluid level within cyst and fractured cortical fragment.

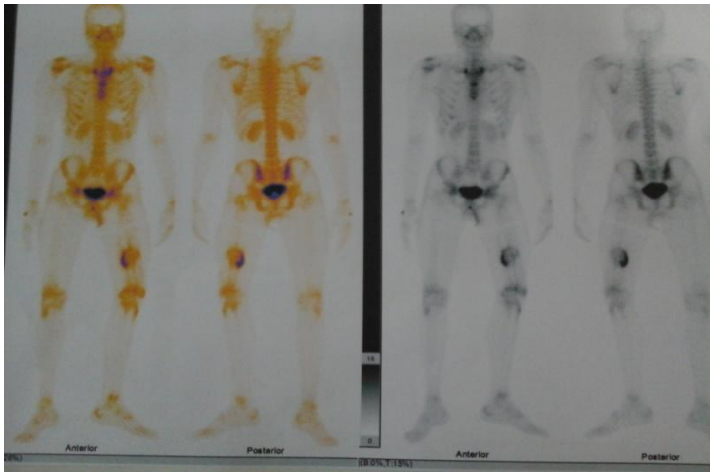

Figure 3 – Bone scintigraphy showing increased uptake at fracture site with ballooning of cyst.

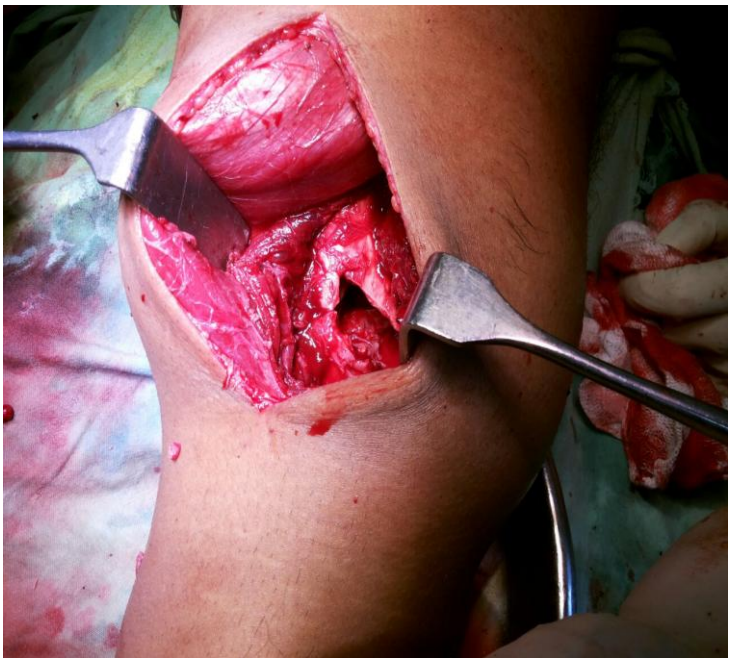

Figure 4 - Pathological fracture through unicameral bone cyst showing haematogenous fluid with fallen fragment of fracture within.

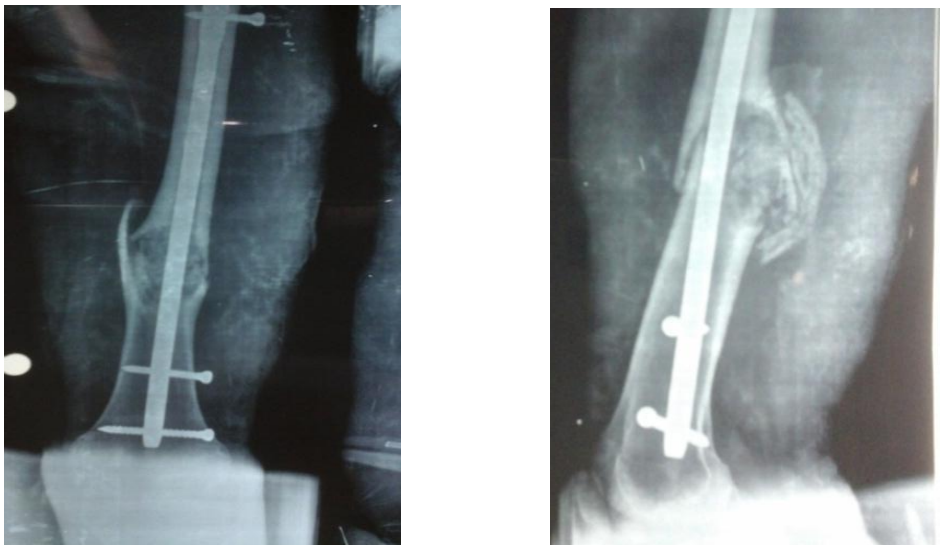

Figure 3 - Anteroposterior and Lateral view of femur after curettage of cyst , open reduction and Internal fixation with intramedullary interlocking nail and bone grafting.