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Treatment of unicameral bone cyst in Pediatric patient with an injectable steroid

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Abstract

Introduction: Unicameral bone cyst is a benign fluid filled cavity which enlarges over time, resulting in thinning of the bone. These cysts can also be multi-loculated. Usually these cysts are reported in the metaphyseal areas of long bones with open phases. Diagnosis is typically based on x-ray imaging features, age, localisation at proximal humerus and femur and the absence of symptoms until pathological fracture development.

Materials and Methods: This study reports a 10 year old boy, reviewed in Sree Mookambika Institute of Medical Sciences, who presented with pain in the left shoulder and difficulty in doing above head activities for a period of 1 month. On examination, bony tenderness was present over proximal humerus and restriction in the range of movement was present. MRI showed large intramedullary cystic lesion in the proximal metaphyseal region of the left humerus. Inj. Methylprednisolone was given on four sittings 3 weeks apart and series of x-rays were taken on the following timeline.

Result: The clinical and radiological examination confirmed the diagnosis of unicameral bone cyst. There was no evidence of recurrence after 6 months of follow up and the bone regeneration was almost complete.

Conclusion: The management of unicameral bone cyst with infiltration of prednisolone injection is an accepted norm. In our case study Inj. Methylprednisolone gave good healing of the unicameral bone cyst and improvement of the symptoms. This procedure is not expensive, mini-invasive, with low surgical risk and short hospitalization. The steroid injections showed to be an excellent treatment for UBC, with complete healing of the lesions.

Keywords: Unicameral bone cysts (UBC), Pediatric patient, proximal humerus

1. Introduction

Unicameral bone cysts (UBC) also known as simple cysts or solitary bone cysts, are the most common benign intraosseous lesions found in children ^[1]. The incidence is about 1 per 10,000 children each year with a male predominance of 2:1 ^[2]. Some lesions remain asymptomatic and resolve spontaneously after skeletal maturity, whereas others will enlarge causing symptoms or fracture secondary to minor trauma. Treatment is typically indicated if pain or a pathologic fracture exists, or when there is risk of an impending fracture because of cyst.

The first treatment method used was curettage and grafting ^[3]. However high recurrence risk after surgery and complications linked to surgery were reported ^[3]. To reduce these complications many methods were described ^[4]. These include injections (steroid, autogenous bone marrow, demineralized bone matrix), multiple drilling and flexible intramedullary nailing ^[5]. The aim of all treatments is to prevent pathologic fractures and shortening and ensure early mobility of the patient ^[6].

2. Review of literature

V. Pavone *et al.* (2013) ^[20] conducted a retrospective study among 23 children with unicameral bone cyst. The patients were followed up at 1, 3, 6 and 12 months and then every year until the adolescence. They concluded that after treatment, in 15 out of 23 patients (65.2%), the humeral cysts were referred, respectively, as Grade 1 and in four as Grade 2. In 4 patients, a refracture occurred. Statistical analysis showed an overall good response in 82.6% of patients at the end of the follow-up. Minor complication including skin discoloration accounted for 13.04%.

Kamal *et al.* (2018) ^[21] conducted a computerized search of operative reports to identify all SBC patients who were treated by PSI 3 times between 2013 and 2016. This study includes 10 patients consisting of 8 males and 2 females, with the mean age of 7.8 years old (range 3-14 years old). The mean follow-up was 12.6 months (range 5-48 months). Successful clinical outcomes with very good MSTS score were observed 90 of 10 (90%) and all cases 100% in 3 and 6 months after 3rd PSI, respectively. Muhammad Azeem Akhund *et al.* (2019) ^[22] conducted a retrospective study of 141 children and concluded that humerus was the most common site of involvement followed by femur. Neer stage I was found in 57(40.4%), stage II was found in 41(29.1%), stage III was found in 28(19.9%) and stage IV was found in 15(10.6%) of the patients. Total 69.5% had good treatment response whereas 43(30.5%) had poor treatment response. There was a significant association of healing with latent cyst and unilocular cyst. Vahdet UÇAN *et al.*, (2020) ^[23] conducted a retrospective study among seventeen patients with unicameral bone cyst and concluded that: Treatment with MPA resulted in complete recovery in ten cysts (55%) and recovery with residual lesions in three cysts (17%). Three cysts (17%) did not respond to steroid treatment and two patients (11%) developed recurrence. The results were

satisfactory (72%) in patients with complete recovery and partial recovery with residual lesions, and unsatisfactory in five patients (28%). No procedure-related complications were encountered. Achmad Fauzi Kamal *et al.*, (2021) ^[18] conducted a case series among 16 children with unicameral bone cyst which showed Musculoskeletal Tumor Society (MSTS) functional score in 15 cases (94%) showed very good results, while 1 (6%) case showed fair result after 3rd injection of steroid. Decompression and percutaneous steroid injection therapy for SBC yielded good outcome. In our study we aimed to evaluate the efficacy of steroid injections at three weeks intervals for unicameral bone cyst of left humerus.

3. Case

10-year-old boy, presented to us with complaints of pain in the left shoulder and arm. And the patient had a history of difficulty in doing overhead activities for a period of 1 month.

At initial physical examination, the patient appeared moderately built and nourished. On inspection there is no swelling, bony tenderness over proximal left humerus was present on palpation. The range of motion (ROM) was 90 degrees of active abduction and passive up to 110 degrees.



Fig 1: Clinical picture showing decreased range of motion-Pre steroid status

There was an internal rotation of 50 and 75 degrees in active and passive movements respectively. Further movements were painful and restricted. Flexion, extension, adduction and internal rotation were pain free. There is no distal neurovascular deficit.

On investigation, initial radiographs showed a well-defined, central, lytic lesion with cortical thinning on the metaphyseal region of the left humerus. There was no

periosteal reaction, cortical disruption, scalloping or other signs of aggressiveness. Lesion was Grade IA using the classification of Lodwick ^[12].

MRI of the left humerus shows large expansile intramedullary altered signal intensity lesion with few inferior internal septations and sharp geographical margin of size 10 x 3 cm in the metadiaphyseal region of the left humerus. Possibly suggestive of Unicameral Bone cyst.

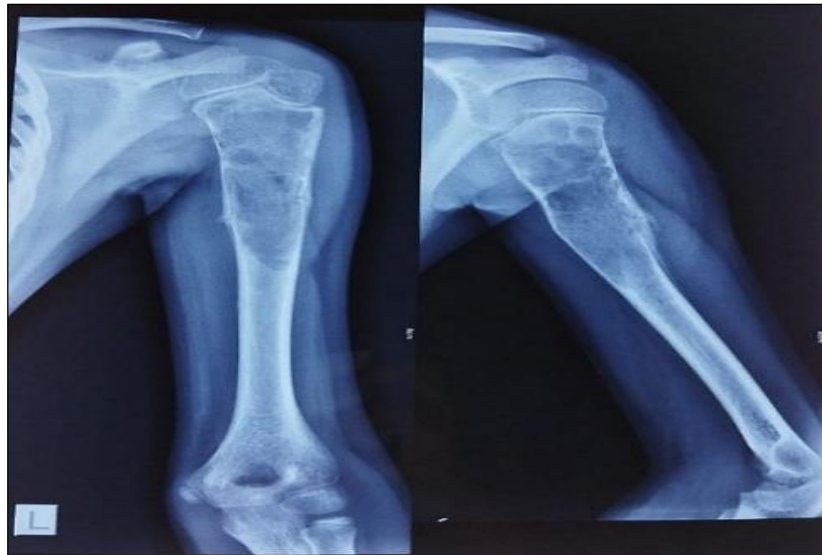


Fig 2: 10 year old boy with left humerus unicameral cyst- AP & Lateral radiographs show central cystic cavitation, well demarcated with cortical thinning and mild expansion

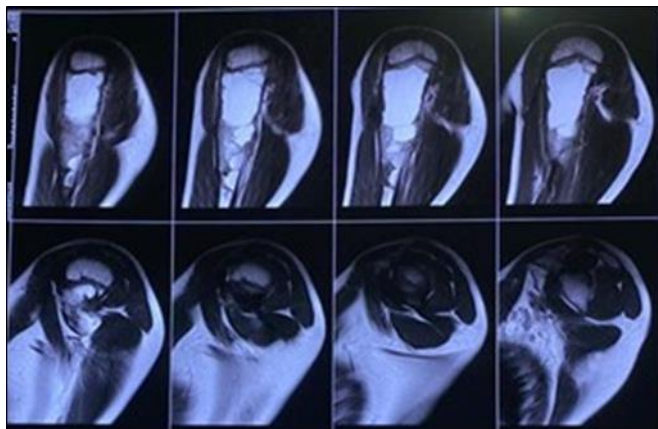


Fig 3: MRI of left humerus

CT showed large central expansile intramedullary abnormal soft tissue intensity lesion of size 3x3x8.3cm in the metaphyso-diaphyseal region of the left humerus. No evidence of cortical break-Benign bone cyst-Unicameral bone cyst. Impacted stress metaphysis fracture of proximal shaft of the left humerus.

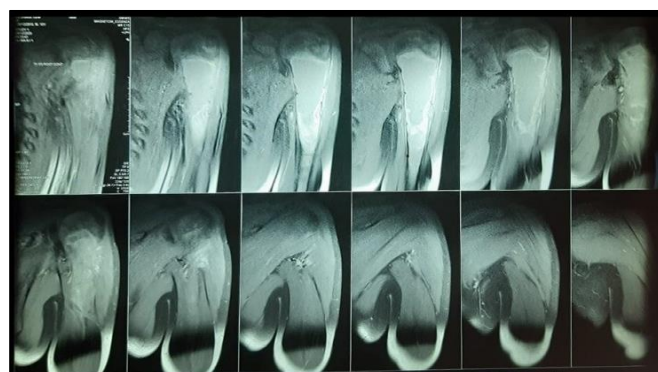


Fig 4: CT of left humerus

Owing to the size and location of the left humeral lesion we recommended percutaneous methylprednisolone injection. With the patient under local anaesthesia, we visualise the lesion with c-arm guidance.

A 11 gauge Jamshidi bone marrow needle was inserted percutaneously to aspirate the contents of cyst and sent for histopathological examination. 160 mg of methylprednisolone was inserted into the cyst. Patient was given an Arm sling and gentle range of motion exercises were initiated and the patient was discharged on the same day. This procedure was performed four times with an interval of three weeks between every injection. He was totally asymptomatic and x-ray showed osteosclerotic changes cortex as well as the medulla of proximal humerus.

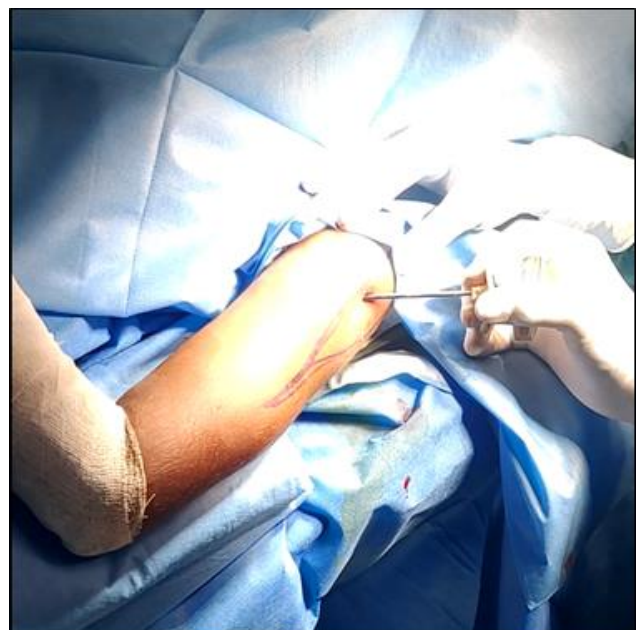


Fig 5: Application of Jamshidi needle to unicameral bone cyst in proximal humerus

The histopathology of the cyst lining revealed fragments of trabecular bone and marrow elements with adjacent fibromyxoid tissue containing chronic inflammation, granulation tissues, hemosiderin laden macrophages, rare foamy histocytes and multinucleated giant cells. No definite cyst lining was identified. This features are consistent with benign bone cyst or unicameral bone cyst.

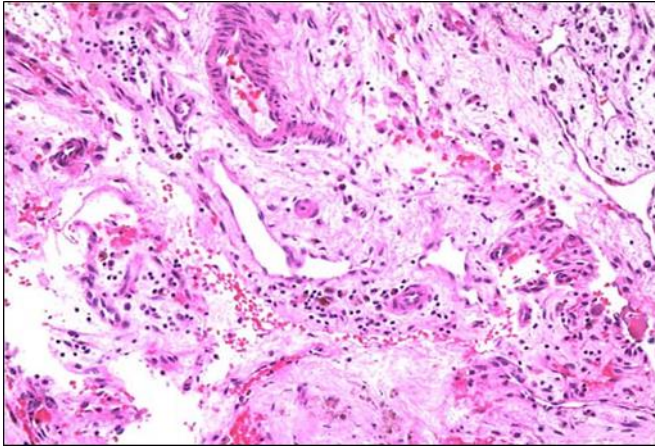


Fig 6: Fragments of trabecular bone and marrow elements with adjacent fibromyxoid tissue containing chronic inflammation, granulation tissues, hemosiderin laden macrophages, rare foamy histocytes and multinucleated giant cells

4. Discussion

Ajiantaro *et al.*, in his study stated that UBC is more common in males than in females with comparison 3:1 and it particularly occurred in the first and second decades of life. The most common predilection of UBC is in proximal humerus (59%) followed by proximal femur (25%) and calcaneus (6%) [13]. Similarly in our case study patient was male with age in the first decade and having solitary bone cyst in the proximal humerus.

It is believed that due to the absence of a significant mechanical load, which usually leads to significant pain, UBC in the humerus remains undetected longer than femur. Thus, UBCs in the humerus often grow until they reveal themselves through a pathological fracture usually arising in the context of a minimal trauma [14]. In our study, patient

with solitary bone cyst had complaints of left arm pain due to pathological fracture at lesion site (metaphysis of left proximal humerus).

Mylle *et al.*, mentioned in their study that steroid is a chosen method for treatment of UBC due to its simplicity, effectiveness and safety [15]. Scaglietti *et al.* reported that methylprednisolone injections into the cyst resulted in a 96% treatment success rate [7]. In our study, methylprednisolone injection resulted in good response and no recurrence during follow up.

Campanacci *et al.*, reported healings in 68% of cysts treated with methylprednisolone. Each patient was submitted to 3-4 injections of methylprednisolone [16]. Our patient was treated with 4 times of inj. methylprednisolone with three weeks of interval for each injection.

A study from Scaglietti *et al.*, noticed that quantity of methylprednisolone depends upon the size of the cyst and the age of the patient: 40 to 80 milligrams for smaller cysts and younger patients, but up to 200 milligrams (5 millilitres of Depo-Medrol) for larger cysts [17]. In our study inj. methylprednisolone 160mg was given on four settings.

Kamal *et al.*, reported very good Musculoskeletal tumour society (MSTS) scores observed in 14 of 16 cases (87%) and all cases 100% in 3 and 6 months after 3rd decompression and inj. methylprednisolone [18]. According to MSTS system, our patient had a score of very good after 2 weeks of fourth injection and it shows good functional outcome.

Lakhwani OP *et al.*, using Chang's criteria, reported in their study that clinical and radiological improvement was as high as 90% [19]. In our study, the patient has good radiological outcomes (complete healing) in follow-up, after 4th injection. (cyst filled by the formation of new bone with or without small static, radiolucent area(s) < 1cm in size).

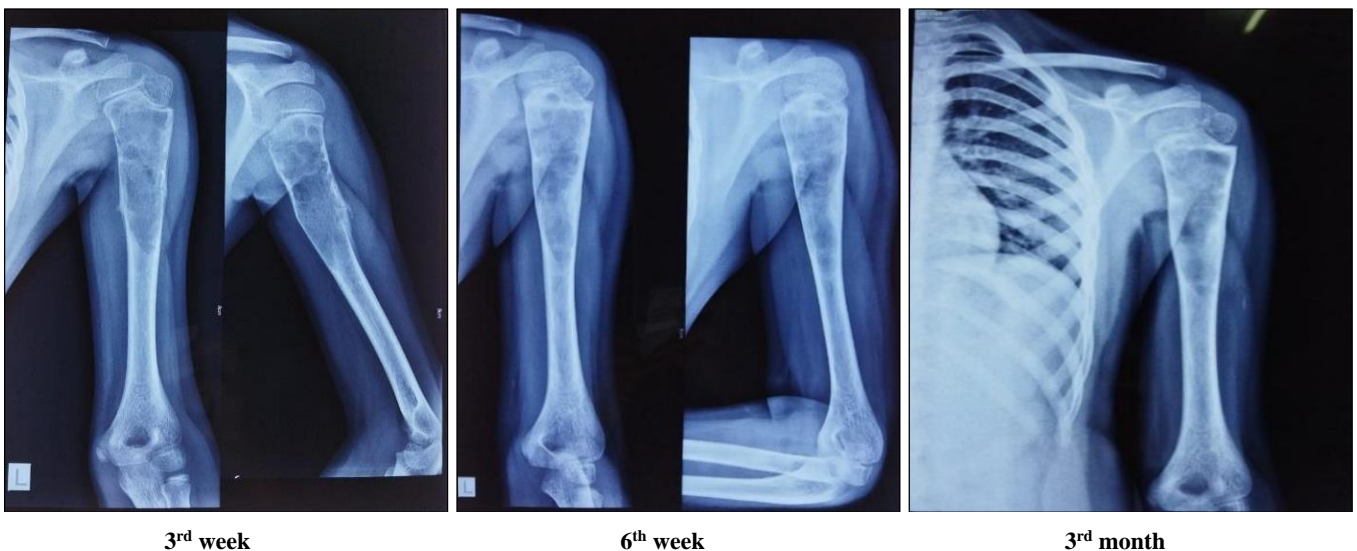


Fig 7: Follow up plain radiographs of left humerus after steroid injection



Fig 8: Clinical picture showing full range of motion at three months-Post steroid status

5. Conclusion

The management of unicameral bone cyst with infiltration of prednisolone injection is an accepted norm. In our case study Inj. methylprednisolone gave good healing of the unicameral bone cyst at left humerus and improvement of the symptoms. This procedure is not expensive, minimally-invasive, with low surgical risk and short hospitalization. The steroid injections showed to be an excellent treatment for Unicameral Bone Cyst, with complete healing of the lesions.

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