Available online at http://www.journalcra.com

International Journal of Current Research
Vol. 7, Issue, 11, pp.23208-23214, November, 2015

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

ISSN: 0975-833X

CASE STUDY

A LARGE ANEURYSMAL BONE CYST OF PROXIMAL HUMERUS IN A 4 YEARS MALE CHILD : A CASE REPORT

***,[1]Ujjal Bhakat, [1]Chinmay Biswas and [2]Saikat Sau**

[1]Department of Orthopaedics, B.S.Medical College, Bankura, India
[3]Department of Orthopaedics, IPGMER, Kolkata, India

ARTICLE INFO

Article History:
Received 20th August, 2015
Received in revised form
22nd September, 2015
Accepted 17th October, 2015
Published online 30th November, 2015

Key words:

X ray, Expansible lytic lesion,
Loculated appearance, Cortical break.

ABSTRACT

Aneurysmal bone cysts are rare benign bone tumours that were first described by Jaffe and Lichtenstein in 1942. They account for 1% of all biopsied primary bone tumours and appear as rapidly growing destructive lesions that expand the cortices. ABC can exist either as a primary bone lesion (70%) or as a secondary lesion when a preexisting osseous lesion can be identified (30%). A 4 years old male child present with a progressive swelling over proximal part of right arm for last 10 months. For last 3 months it rapidly increased in size, accosiated with throbbing pain. X ray shows Expansible lytic lesion, loculated appearance, cortical break and metaphysical lesion over proximal humerus. Histopathology report shows ANEURYSMAL BONE CYST. Exicision of whole mass with free fibular graft done under general anesthesia. Union at both ends of the fibular graft and over head hand movement possible at 8 month follow up. Complete re-modelling and reappearing of epiphysis with acceptable range of movement of upper limb seen after one year six month follow up.

***Citation*:** Ujjal Bhakat, Chinmay Biswas and Saikat Sau**,** 2015. "A large Aneurysmal bone cyst of proximal Humerus in a 4 years male child : A case report", *International Journal of Current Research*, 7, (11), 23208-23214.

INTRODUCTION

Aneurismal bone cyst constitutes 1.4% of all primary bone tumors and 15% of all primary spine tumors (Ameli *et al*., 1985; Dahlin and Mcleod, 1982). It is usually an eccentric lytic lesion blowing up the bone. It consists histologically of blood filled spaces surrounded by connective tissue septa containing osteoclast giant cells and newly formed bone trabeculae (Jaffe, 1950; Lichtenstein, 1950). It commonly involves the metaphysis of long bones (proximal humerus, femur, tibia) and the flat bones of the pelvis. In the spine it commonly involves the neural arch (Turker *et al*., 1998). The cyst is occasionally associated with other tumors of the bone, such as osteoblastoma, osteosarcoma, giant cell tumor and fibrous dysplasia (Kleuver *et al*., 1998; Kransdorf and Sweet, 1995) and has occasionally been reported to occur at a site of previous trauma to the bone (Dabezies *et al*., 1982). There is tendency towards rapid growth with local expansion, which can exert pressure on an adjacent structure (Turker *et al*., 1998). Among the various modality of treatment in aneurismal bone cyst, orthopaedic surgeons are really in a dilemma in some aggressive tumors whether limb salvage or amputation is best. One such case which occurred in a 4 years male over proximal humerus is being described.

***Corresponding author: Ujjal Bhakat,**
Department of Orthopaedics, B.S.Medical College, Bankura, India

CASE HISTORY

A 4 years old male child present with a progressive swelling over proximal part of right arm for last 10 months**.** For the last 3 months it rapidly increased in size, associated with throbbing pain over right arm (Figure 1,2,3). There was no history of trauma, fever or similar swelling over the body. The child was otherwise healthy.

On examination there was 10cm/10cm tense ,globular mass with venous prominence over proximal humerus. On palpation the swelling was non tender, well defined, variegated in consistency, surface was smooth. The proximal part of humerus was poorly delineated. The skin was tense but free from underlying structure, arising from metaphysis and shoulder joint was free. There is no neurovascular deficit and no lymphadenopathy. Radiological examination-serial skiagrams starting from 3 month of onset of symptoms shows gradual radiological progression of disease. Latest preoperative skiagram shows Expansible lytic lesion with loculated appearance and Cortical break over metaphysis of proximal humerus. Proximal epiphysis and shoulder joint are remains intact (Figure 4,5,6). Preoperative MRI also support the findings in favor of aneurismal bone cyst (Figure 7).

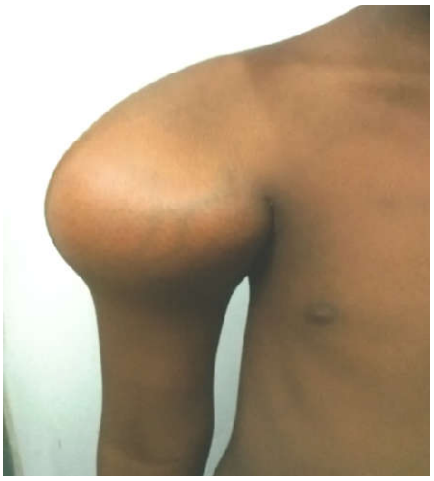

Figure 1

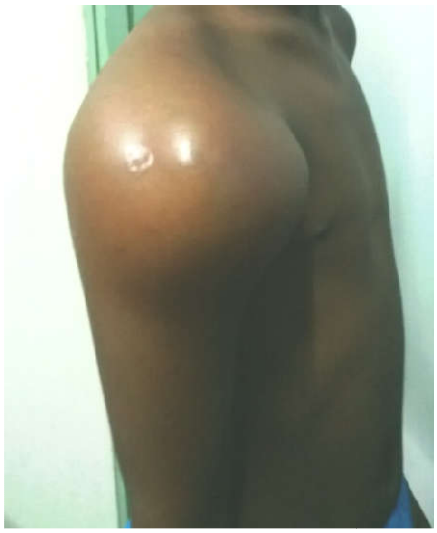

Figure 2

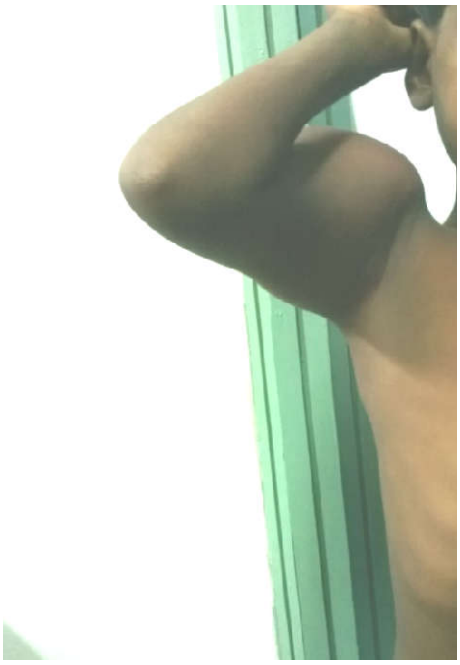

Figure 3. Shoulder joint is free

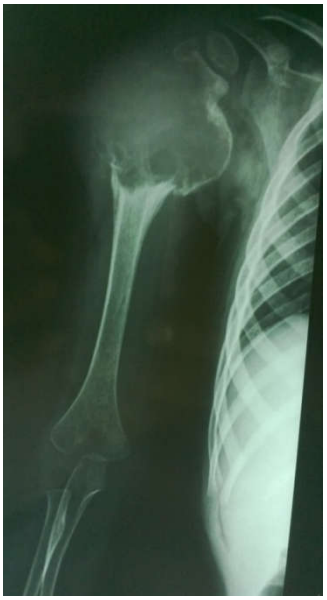

Figure 4. At 3 month

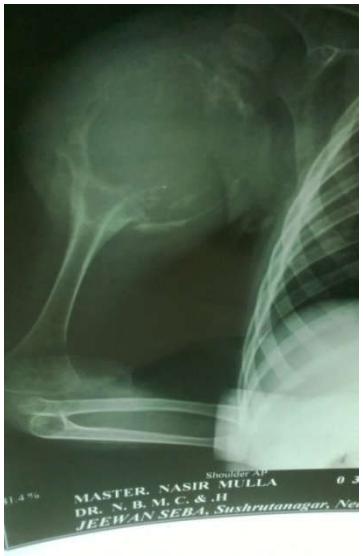

Figure 5. At 6 month

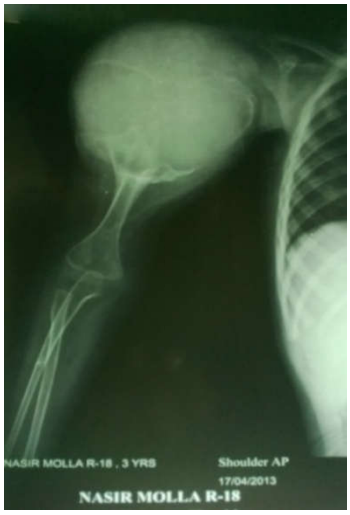

Figure 6. At 10 month

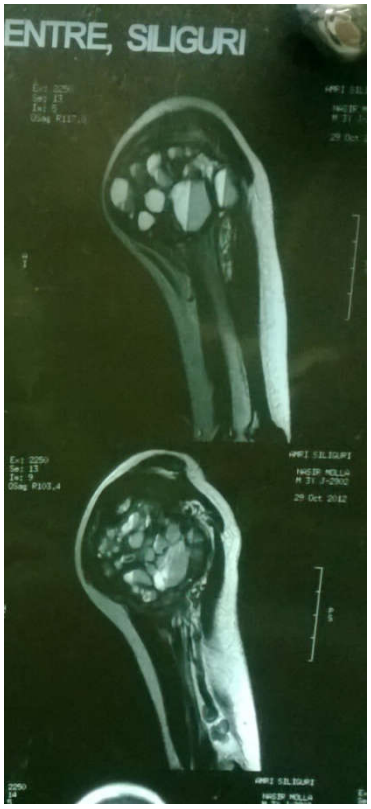

Figure 7. MRI

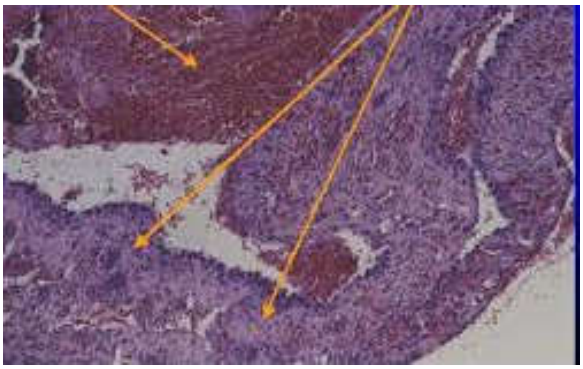

Figure 8. Histological feature

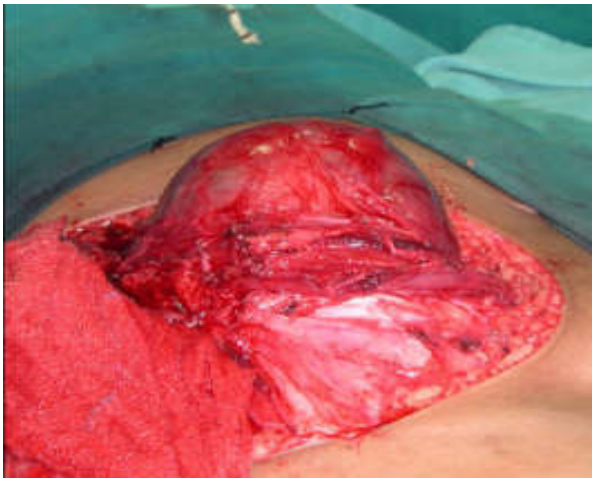

Figure 9. Intraoperative picture

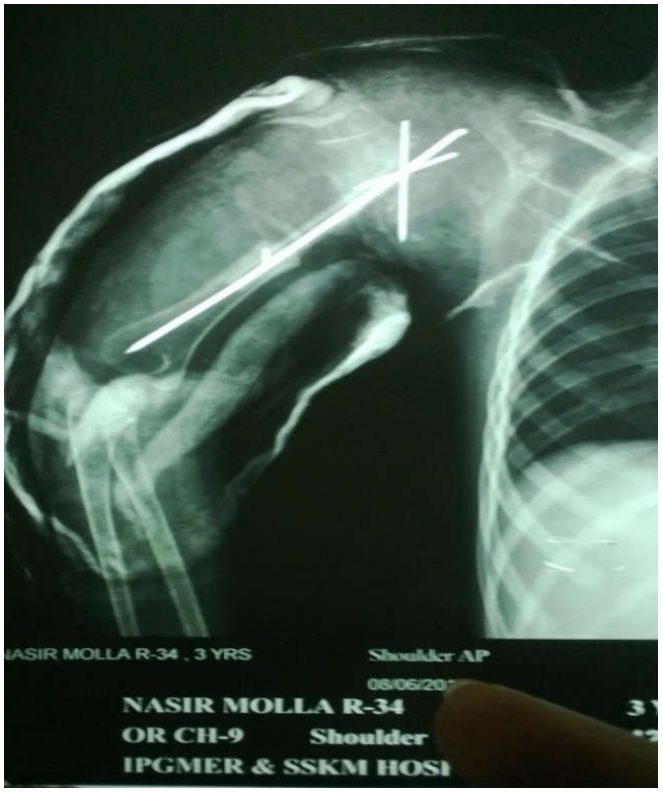

Figure 10. Immediate post operative

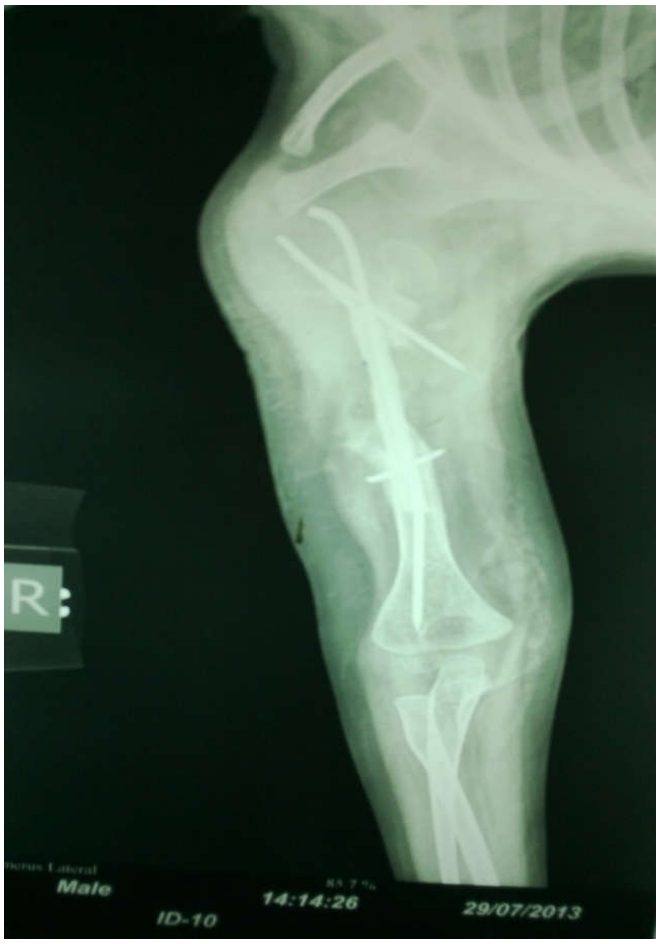

Figure 11. 2month post op

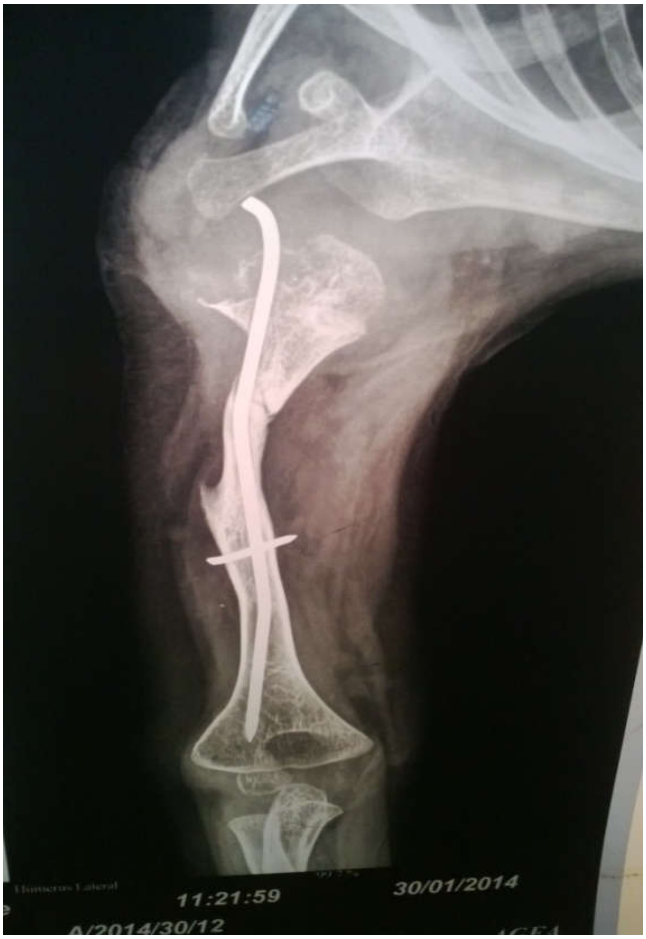

Figure 12. 5month post op

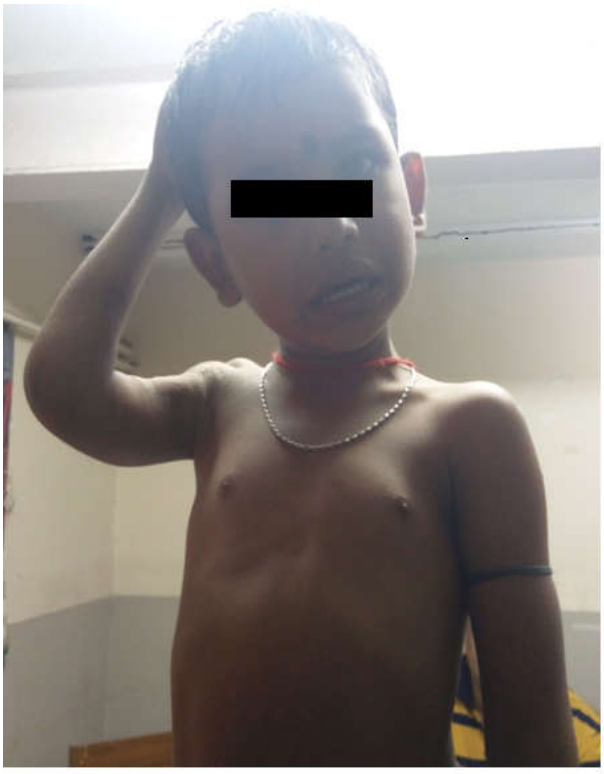

Figure 13. 5month post op

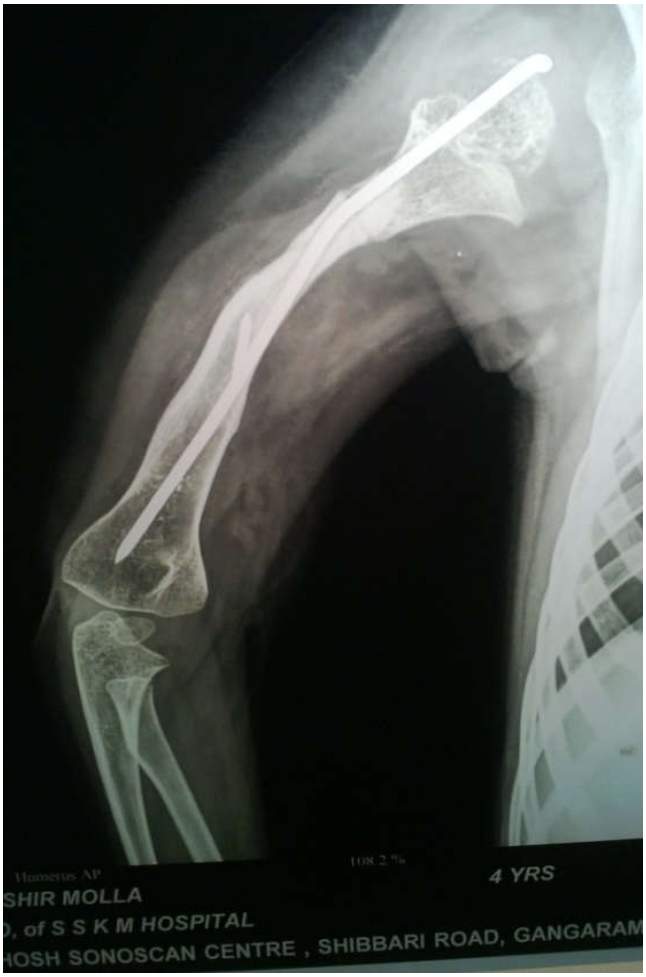

Figure14. 8 month follow up

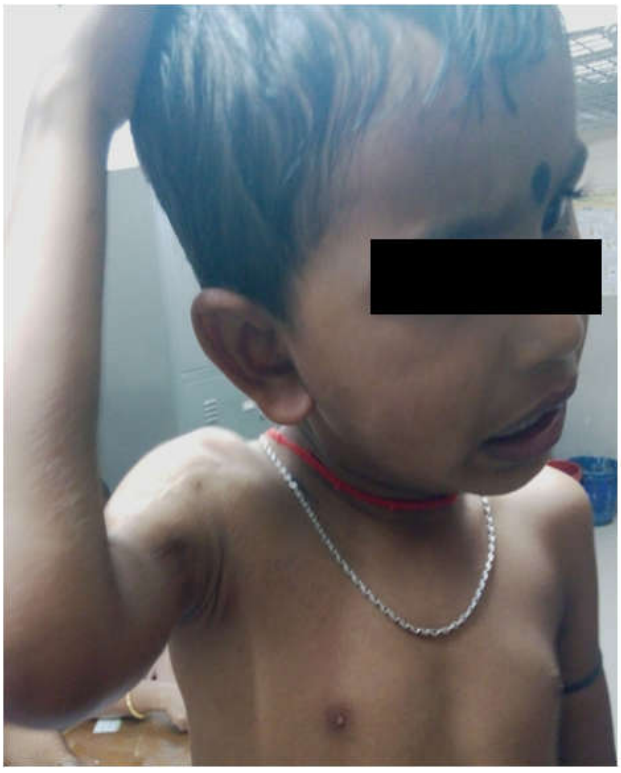

Figure 15. 8 month follow up

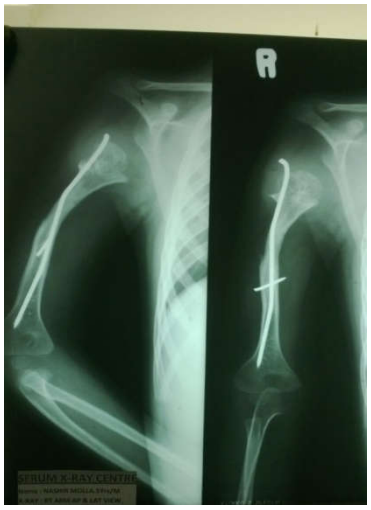

Figure 15. 8 month follow up

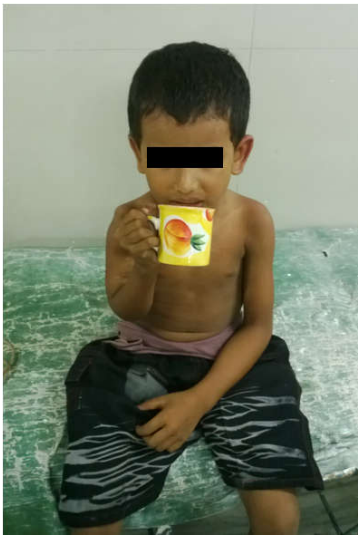

Figure 18. one & half year follow up

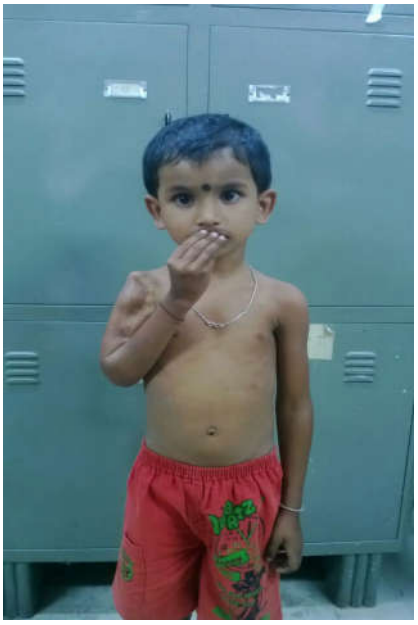

Figure 19.one year six month follow up

On aspiration from the swelling fresh blood comes out. Incisional biopsy was done and histopathological report shows multiple giant cells with hemorrhagic tissue with cavernous spaces separated by cellular stroma, there was no malignant cells and features suggestive of aneurysmal bone cyst (Figure 8).

intraoperatively the swelling was below the deltoid like a membranous pouch (Figure 9). The whole mass was dissected out and the bony gap replaced by free fibular graft and fixed with multiple k wires (Figure 10).

At 2 month follow up there was no sign of infection and there was signs of union over proximal and distal end of fibular graft (Figure 11). After 5 month of operative procedure we remove 2 k wires from the proximal end (Figure12,13). After 8 month there was complete union at both the proximal and distal end of fibular graft and over head movement was possible (Figure14,15).

After one year his dominant hand work normally and x-ray shows complete union with remodeling (Figure16,17). After one year and 6 month follow up complete remodeling was seen on x-ray and all functional range of movement was possible and there was no sign of recurrence (Figure18,19).

DISCUSSION

Aneurysmal bone cyst was first described by Jaffe and Lichtenstein in 1942 and was subsequently further defined by both of these authors and became known as Jaffe-Lichtenstein disease (Jaffe, 1950; Jaffe and Lichtenstein, 1942; Lichtenstein, 1957). The term aneurysmal seems to relate to the blow out distension and the word cyst relates to the fact that the tumor often presents as a blood filled cavity (Lichtenstein, 1957). An ABC is an uncommon expansile osteolytic lesion of bone consisting of a proliferation of vascular tissue that forms a lining around a blood filled cystic lesion. It is clinically important because it is easily mistaken for a malignancy pathologically in its early to mid-phase. In 20 to 30 % of cases, an aneurysmal bone cyst may be associated with an underlying tumor i.e. osteosarcoma, giant cell tumor, chondrosarcoma, fibrous dysplasia etc (Kleuver *et al*., 1998; Kransdorf and Sweet, 1995; Martinez and Sissons, 1988).

It has been suggested that ABC is not a true neoplasm but rather a vascular malformation. These cysts are generally thought of as a secondary vascular phenomenon superimposed on a preexisting lesion, which presumably initiates a periosteal or intraosseous arteriovenous malformation (Ameli *et al*., 1985; Buraczewski and Dabska, 1971; Edling, 1965). Aneurysmal bone cyst is an uncommon lesion which has been found to occur mostly in long bones of the skeleton, and the spine (Oliver, 1973; Struthers and Shear, 1984; Kumar *et al*., 2009). The bone cyst produces firm swellings which is rarely associated with pain (Kransdorf and Sweet, 1995; Struthers and Shear, 1984).

Radiographically these lesions are usually unilocular, (Kransdorf and Sweet, 1995; Gardre and Zubairy, 2000) but long standing lesions may show a "soap-bubble" appearance

and may become progressively calcified. Radiographic, differentiation from other expansile lesions may be difficult. CT scan and MRI may not provide clear diagnostic criteria but only outline the extent of the lesion (Asaumi *et al*., 2003; Abuhassan and Shannak, 2010). This is because of the similarity of radiographic picture of these lesions to eosinophilic granuloma, giant cell tumor, non ossifying fibroma, Ewing's tumor, and in older patient metastatic carcinoma or myeloma.

A biopsy is often helpful, but needle biopsy is sometimes a problem because of the material obtained may consists of mostly blood elements. An incisional biopsy and frozen section are more reliable (López-Arcas Calleja *et al*., 2007).

There is no uniform strategy on treatment and management of these lesions due to its varied nature (Abuhassan and Shannak, 2010). The usual treatment of choice is curettage as it is a benign lesion. But there is high recurrence rate (20-30%) reported in various studies (Gardre and Zubairy, 2000; Abuhassan and Shannak, 2010; Möller *et al*., 2011). Other modalities of treatment have been tried, curettage followed by implantation with allograft chips, polymethyl methacrylate, autografts, cryotherapy, local injections of steroids, radiation therapy (Mega voltage, 26-30 Gy), injecting fibrosing agents, demineralized bone powder mixed with bone marrow aspirate introduced into the cyst to halt the expansion phase and to allow the cyst to ossify, segmental resection with use of rhBMP2 along with the rib graft has also given good results (Balaji, 2010). Segmental resection with immediate bone grafting with vascularised bone graft is suggested for extensive lesions such as the present case where a vascularised fibula flap was used to reconstruct the defect (Abuhassan and Shannak, 2010; Möller *et al*., 2011)

Treatment of ABC with simple curettage can result in obliteration of the cyst (Turker *et al*., 1998). Curettage and bone graft has a 20 % recurrence rate, which can be managed with more aggressive curettage or excision (Vergel *et al*., 1992). If postoperative deformity develops surgical stabilization is indicated (Turker *et al*., 1998). The clinical course of aneurismal bone cysts is sometimes unpredictable and local recurrences have been described after various types of treatments (Boriani *et al*., 2001).

Radiotherapy can also be used effectively in patients who are resistant to surgical treatment or are at high risk for surgery (Kamikonya *et al*., 1991). However, possibility of complications such as sarcomatous change, myelopathy, deformation of vertebrae make this mode of treatment less desirable (Frassica *et al*., 1993). Selective embolisation of the tumor is possible in large tumors that have high risk of bleeding and in places where curettage would be difficult (Derosa *et al*., 1990). Intra-lesional injection of sclerosing agents is also an effective method for treatment of ABC. Overall cure rates of 87% have been achieved (Guibaud *et al*., 1998). The prognosis of ABC in children is good although there is a high recurrence rate mainly in the 1st year after operation and in patients who undergo partial resection of the tumor (Ameli *et al*., 1985; Boriani *et al*., 2001). Therefore these children require careful follow-up and might need additional treatment.

REFERENCES

Abuhassan FO, Shannak A. Non-vascularized fibular graft reconstruction after resection of giant aneurysmal bone cyst (ABC). *Strategies Trauma Limb Reconstr*., 2010;5:149-54.

Ameli No, Abbassioun K, Saleh H, *et al*. Aneurysmal bone cyst of the spine. Report of 17 cases. J Neurosurg., 1985; 63:685-690.

Asaumi J, Konouchi H, Hisatomi M, Matsuzaki H, Shigehara H, Honda Y, *et al*. MR features of aneurysmal bone cyst of the mandible and characteristics distinguishing it from other lesions. *Eur J Radiol*., 2003;45:108-12.

Balaji SM. Mandibular cystic defect. A composite approach with rhBMP-2 and rib graft. *J Maxillofac Oral surg.,* 2010;8:27-30.

Boriani S, DE Iure F, Campanacci L, *et al*: Aneurysmal bone cyst of the mobile spine: report on 41 cases. *Spine*, 2001;26:27-35.

Buraczewski J, Dabska M: Pathogenesis of aneurysmal bone cyst. Relationship between the aneurysmal bone cyst and fibrous dysplasia of bone. *Cancer*, 1971; 28:597-604.

Dabezies EJ, D'ambrosia RD, Chuinard RG, *et al*. Aneurysmal bone cyst after fracture. A report of three cases. *J Bone Joint Surg Am*., 1982;64:617-621.

Dahlin DC, Mcleod RA: Aneurysmal bone cyst and other nonneoplastic conditions. *Skeletal Radiol*., 1982;8:243-250

DE Kleuver M, Van Der Heul RO, Veraart BE: Aneurysmal bonecyst of the spine: 31 cases and the importance of the surgical approach. *J Pediatr Orthop*., 1998;B 7;286-292

Derosa GP, Graziano GP, Scott J. Arterial embolization of aneurismal bone cyst of the lumbar spine. A report of two cases. *J Bone Joint Surg Am*., 1990;72:777-780.

Edling NP: Is the aneurysmal bone cyst a true pathologic entity? *Cancer*, 1965;18:1127-1130.

Frassica FJ, Frassica DA, Wold LE, *et al*. Postradiation sarcoma of bone. *Orthopedics*, 1993;16:105-106.

Gardre KS, Zubairy RA. Aneurysmal bone cyst of the mandibular condyle: Report of a case. *J Oral Maxillofac Surg*., 2000;58:439-43.

Guibaud L, Herbreteau D, Dubois J, *et al*. Aneurysmal bone cysts: percutaneous embolization with an alcoholic solution of zein-series of 18 cases. *Radiology*, 1998;208:369-373.

Jaffe H 1950. Aneurysmal bone cyst. Bull Hosp Jt Dis 11:313

Jaffe HL, Lichtenstein L. Solitary unicamaral bone cyst with emphasis on the Roentgen picture. The pathologic appearance and pathogenesis. *Arch Surg*., 1942;44:1004-25.

Kamikonya N, Hishikawa Y, Kurisu K, *et al*: Aneurysmal bone cyst treated by high-energy, low-dose radiation therapy: a case report. *Radiat Med*., 1991;9:54-56.

Kransdorf MJ, Sweet DE: Aneurysmal bone cyst: concept, controversy, clinical presentation, and imaging. AJR. 1995;164:573-580.

Kumar VV, Malik NA, Kumar DB. Treatment of large recurrent aneurysmal bone cysts of mandible. Transosseous intralesional embolization as an adjunct to resection. *Int J Oral Maxillofac Surg.,* 2009;38:671-6.

Lichtenstein L 1950. Aneurysmal bone cyst. Cancer 3:279–289

Lichtenstein L. Aneurysmal bone cyst: Observations of fifty cases. *J Bone Joint Surg Am*., 1957;39:873-82.

López-Arcas Calleja JM, Cebrián Carretero JL, González Martín J, Burgueño M. Aneurysmal bone cyst of the mandible: Case presentation and review of the literature. *Med Oral Pathol Cir Bucal*., 2007;12:E401-3.

Martinez V, Sissons HA: Aneurysmal bone cyst. A review of 123 cases including primary lesions and those secondary to other bone pathology. *Cancer*, 1988;61:2291-2304.

Möller B, Claviez A, Moritz JD, Leuschner I, Wiltfang J. Extensive aneurysmal bone cyst of the mandible. *J Craniofac Surg*., 2011;22:841-4.

Oliver LP. Aneurysmal bone cyst. Report of a case. *Oral Surg Oral Med Oral Pathol*., 1973;35:67-76.

Struthers PJ, Shear M. Aneurysmal bone cyst of the jaws. Clinicopathological features and pathogenesis. *Int J Oral Surg*., 1984;30:88-100.

Turker RJ, Mardjetko S, Lubicky J: Aneurysmal bone cysts of the spine: excision and stabilization. *J Pediatr Orthop*., 1998;18:209-213.

Vergel DE Dios AM, Bond JR, Shives TC, *et al*: Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer*, 1992;69:2921-2931.

****