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ANEURYSMAL BONE CYST DEVELOPING IN MONOSTOTIC FIBROUS DYSPLASIA OF PROXIMAL RADIUS

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ABSTRACT

Fibrous dysplasia in the proximal radius is an unusual presentation. The development of aneurysmal bone cyst (ABC) against the back ground of fibrous dysplasia has not been reported in this location. A twenty four old male presented with pain, swelling and tenderness over the proximal radius on the left side. It was diagnosed as secondary ABC developing in fibrous dysplasia. It was treated by segmental excision and fibular grafting. This pattern of presentation at this location has not been reported in the literature.

INTRODUCTION

Fibrous dysplasia is reported in 0.2-0.8 % of bone tumours. Secondary ABC developing against a background of fibrous dysplasia is a rare presentation. Literature search revealed less than 50 cases, mostly in the axial skeleton [1-4]. We could find only 5 cases where this tumour developed in the extremities (femur-2,tibia-1, humerus-1, ulna-1) [5]. In this case report we wish to highlight that lytic lesions of bone present diagnostic challenges and histo-pathology alone will provide a definitive diagnosis.

CASE PRESENTATION

A twenty four year old, male, house painter, presented with pain in the left forearm for the past one month. He sustained injury to the left elbow two years ago. He could not give us details of the treatment received. Examination of the left upper limb revealed tenderness over the front of the elbow. The range of movement was, flexion 0-130°(degrees) and supination - pronation were restricted with tenderness over the proximal radius. evaluation (X-rays and MRI) showed an expansile lytic lesion in the proximal radius with breach in the cortex on the medial aspect(Figure 1). Based on clinical and radiological examination a differential diagnosis of giant cell tumour (GCT) / ABC was considered. Fine needle aspiration cytology (FNAC) revealed multinucleated giant cells and spindle shaped cells against a hemorrhagic background and a provisional diagnosis of GCT was made. The segment of bone was excised and fibular interposition graft placed in the defect. Fixation was achieved with an intramedullary fixation device.

The excised specimen measured 6×4×2 centimeter. The cut section showed thinned out cortices with multiple honey combed channels filled with blood. Histopathological diagnosis was fibrous dysplasia with secondary ABC formation (Figure 2 and 3).

Post operatively,patient was given a protective back slab for a period of six weeks. Physiotherapy was started and patient allowed gradual activity. Follow-up x-ray, 6months after surgery shows incorporation of the fibular graft and bone union (Figure 4). There was no radiological evidence of recurrence. Patient returned to his pre- operative vocation as a painter.

Fig 1. X-ray showing expansile lytic lesion in proximal radius

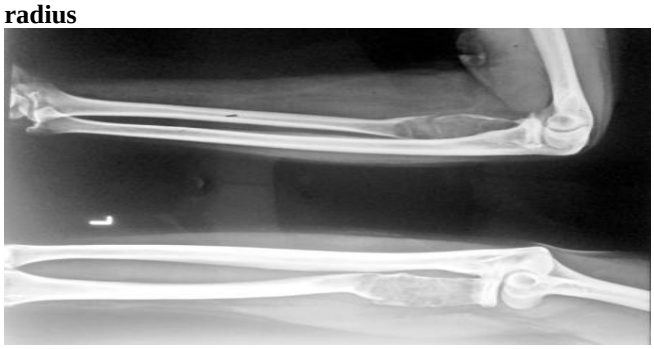


Fig 2. Photomicrograph showing begnin fibroblastic tissue with irregular trabeculae of woven bone without osteoblastic rimming. Blood filled aneurysmal space without endothelial lining.(H&E x 400)

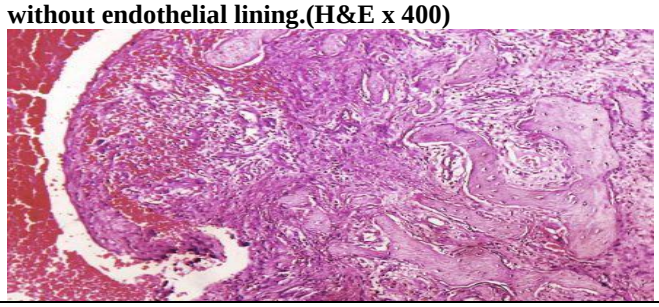


Fig 3. High power view

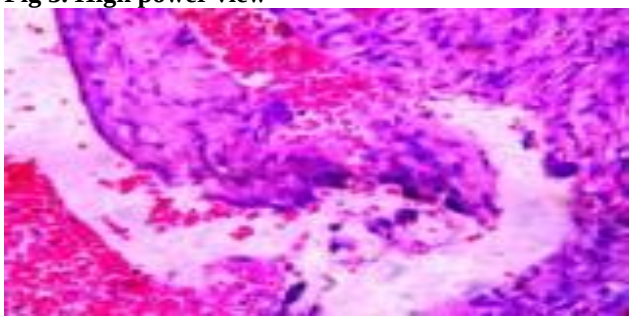
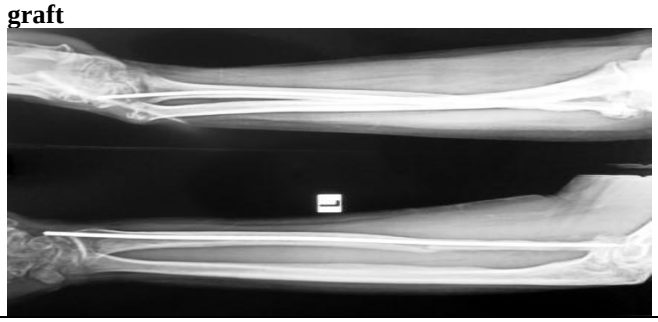


Fig 4. X-ray showing incorporation and union of fibular graft



DISCUSSION

Fibrous dysplasia is a developmental condition often presenting in long bones, cranium and pelvis. It can present as single (monostotic) or multiple (polyostotic) forms. The behavior and microscopic appearance of both forms is similar. Left alone fibrous dysplasia can continue asymptomatic for many years. Most often it is an incidental finding in x-rays. Most common sequel of fibrous dysplasia is pathological fracture.

Development of ABC in fibrous dysplasia is uncommon and most reports are in the flat bones and axial skeleton. Initial reports suspected the development of ABC in fibrous dysplasia to be malignant change. The short duration in which the bone expansion occurred and the computed tomography (CT) evidence of cyst formation and fluid levels suggest a diagnosis of ABC. Literature shows that ABC has a tendency to arise in preexisting lesion by forming arterio-venous fistulae. The resultant increase in intraosseous pressure produces bone expansion [6, 7]. Due to circumfrential involvement of the bone, segmental excision of the lesion was performed to avoid spillage of cells. The defect was reconstructed with diaphyseal segment of the fibula which has dimensions similar to the excised bone. Stabilization was performed with intramedullary nail. Patient was allowed flexion-extension after six weeks and supination-pronation after 12 weeks. At 16 weeks post operative x-rays showed incorporation of graft and painless range of movement. Patient regained full pronation but only 20° of supination. He had no radiological evidence of recurrence six months after surgery.

CONCLUSION

In our opinion, this is the first presentation of secondary ABC in fibrous dysplasia of proximal radius. Total excision and fibular grafting resulted in good functional outcome. It is difficult to differentiate ABC from GCT in small bones such as radius. Lytic lesions of the bone present diagnostic challenges, which can only be confirmed by histo-pathology.

REFERENCES

1. Marieke N Snieders, Folkert J Van Kemenade, Barend J Van Royen. (2009). Monostotic Fibrous dysplasia of a lumbar vertebral body with secondary aneurysmal bone cyst formation: a case report. *J medical case reports*, 3, 1-5.
2. Haddad GF, Hambali F, Mufarrij A, Nassar A, Haddad FS. (1998). Concomitant fibrous dysplasia and aneurysmal bone cyst of the skull base. Case report and review of the literature. *PediatrNeurosurg*, 28(3), 147-53.
3. Malik R, Pandya VK, Awasthi P. (2006). Secondary Aneurysmal bone cyst in association with fibrous dysplasia. *Ind jRadiolimag*, 16(1), 27-8.
4. Lin WC, Wu HT, Wei CJ, Chang CY. (2004). Aneurysmal bone cyst arising from fibrous dysplasia of the frontal bone. *EurRadiol*, 14(5), 930-2.

5. Maurizio Montalti, Macro Alberghini, PietroRuggieri. (2009). Secondary aneurysmal bone cyst in fibrous dysplasia of the proximal femur. *Orthopaedics*, 32(5), 501-10.
6. Dickers D , Sauter AM, Mallens WMC. (1986). Aneurysmal bone cyst in association with fibrous dysplasia-a case report. *J bone j surg*, 68(b), 144-6.
7. Buraczewski J, DaBska M. (1998). Pathogenesis of aneurysmal bone cyst .relationship between the aneurysmal bone cyst and fibrious dysplasia of bone. *Cancer*, 28, 597-604.