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Case Report

Osteofibrous dysplasia of clavicle: a rare case presentation

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ABSTRACT

Osteofibrous dysplasia is a rare entity which occurs in long bones. Tibia is the most common bone to be affected by osteofibrous dysplasia. We report a rare case presentation of osteofibrous dysplasia of clavicle in an 18 year old male student who presented to us with painful swelling over right clavicle region. Radiograph showed expansile lytic lesion in middle third of clavicle with multiple loculations and intervening calcified septae. On excisional biopsy and histopathological examination this lesion found to be osteofibrous dysplasia of clavicle. To the best of our knowledge, it is a first case of osteofibrous dysplasia affecting clavicle to be reported in the literature.

Keywords: Osteofibrous dysplasia, Clavicle, Osteolytic

INTRODUCTION

Ostefibrous dysplasia is a rare, non-neoplastic condition of unknown etiology affecting the long bones.[1] Most lesion of osteofibrous dysplasia affects the anterior cortex of tibia.

After the first description of this condition by Frangenheim in 1921,[2] Numerous cases of osteofibrous dysplasia affecting the Tibia have been reported in literature.[3-5] Sometimes with involvement of ipsilateral fibula also.[4,5] Lesions in radius[6] and in humerus[7] were also described.

After thorough search of literature, we found no case of osteofibrous dysplasia affecting the clavicle reported till now.

CASE REPORT

An 18 year-old male student presented with six month old painful swelling of right clavicle region. Patient had no other significant history suggestive of weight loss or constitutional symptoms.

On clinical examination the swelling was diffuse, 5x5 cm in size, firm in consistency, originating from Clavicle, nonmobile, not adherent to overlying skin. Overlying skin was normal (Figure 1).

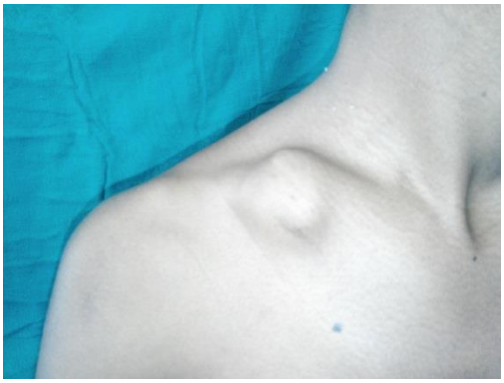

Figure 1: Clinical photograph of patient showing large clavicular swelling.

Range of motion of right shoulder joint was full and painless. There was no significant axillary or cervical lymphadenopathy and no neurovascular deficit was present.

Radiograph right shoulder with clavicle anteroposterior view showed expansile, lytic lesion in the midshaft region of right clavicle with multiple radiolucent areas and intervening calcified septae. Zone of transition was narrow. Cortex was thinned out and ballooning of cortex was present. There was no periosteal reaction and surrounding soft tissue shadow was normal. There were evidence of cortical breaches at 2-3 places (Figure 2).

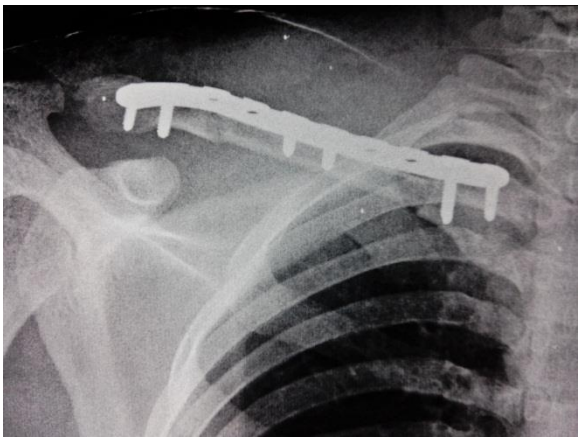

Figure 2: Post-operative radiograph of clavicle AP view showing reconstructed clavicle with fibular graft fixed with plate.

Since the fine-needle aspiration cytology examination was inconclusive and radiographically the lesion seemed to be some active, we excised the lesion and sent it for histopathological examination.

After excision of the lesion we reconstructed the clavicle with the help of fibular strut graft taken from ipsilateral lower limb and internally fixed it with locking clavicle plate and screws (Figure 3 & 4).

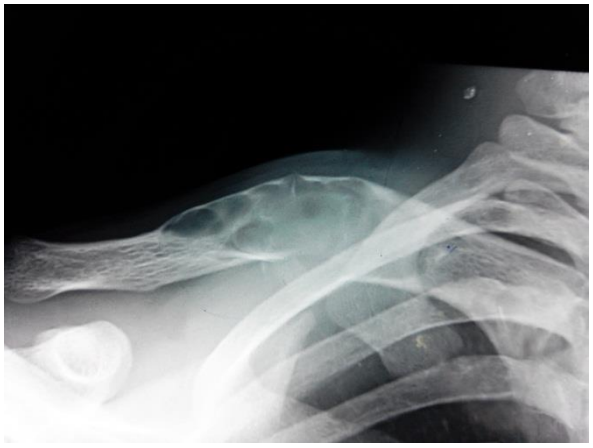

Figure 3: X-ray clavicle AP view showing expansile lytic lesion in mid shaft region of clavicle.

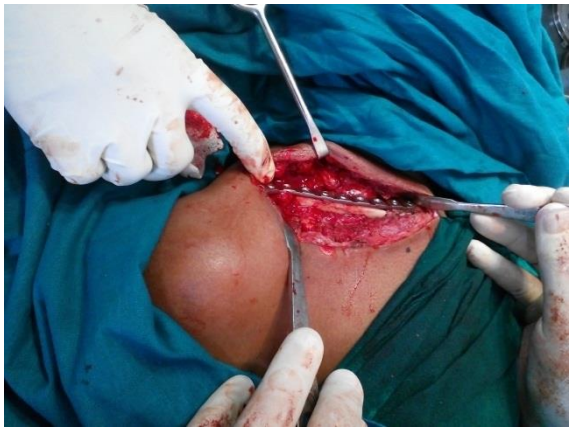

Figure 4: Intraoperative photograph after reconstruction of defect with fibular graft.

On histopathological examination of slides prepared from lesion revealed areas of bland spindle cell proliferation and dispersed bony trabeculae lined by benign osteoblast. No nuclear pleomorphism or mitosis seen in spindled cells (Figure 5).

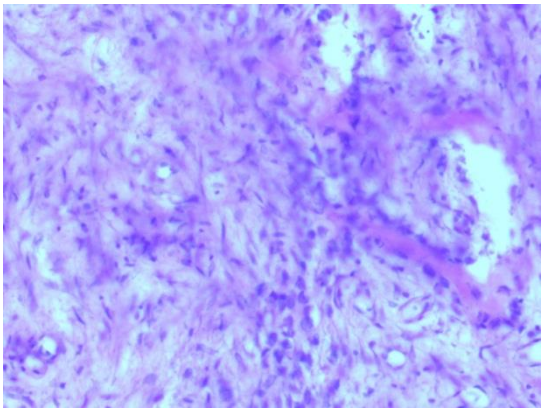

Figure 5: H&E section (400x) showing bland fibrous proliferation with foci of bony trabeculae and osteoblastic rimming.

On follow-up of patient, we removed the suture at two weeks and kept his shoulder immobilized for six weeks. After six weeks we removed immobilizer and started all range of motion exercise of shoulder and other joint. Now At three months, patient is doing his all routine activities without any complaints.

DISCUSSION

Osteofibrous dysplasia is a rare non-neoplastic condition. Our patient was an 18 year old male. It is usually diagnosed in children under 10 years with peak incidence in age group 1-5 years. Though adult patients diagnosed with de novo osteofibrous dysplasia also have been reported. The oldest patient with osteofibrous dysplasia reported in literature was of 39 years.[8]

There is no sex prediliction for osteofibrous dysplasia.

Etiology of osteofibrous dysplasia is not known. It is thought to arise from fibrovascular defect. Based on the common causative factor i.e. fibrovascular defect, Johnson[9] proposed the relationship between osteofibrous dysplasia and admantinoma. According to his theory osteofibrous dysplasia is result of abnormality of Haversian canal on the contrary to admantinoma which is the result of abnormality of intramedullary vasculature.

The natural history of osteofibrous dysplasia is unpredictable. It may grow rapidly or may regress spontaneously on skeletal maturity with time.

We marginally resected the lesion and reconstructed tha defect with fibular strut graft. Non-operative treatment is usually recommended till skeletal maturity by some author but surgical extraperiosteal marginal resection has also been recommended in the literature,[10] as the high recurrence rate is characteristic feature of this lesion following curettage and bone grafting only.

Osteofibrous dysplasia usually affects the tibia. There are many articles present in the literature reporting the lesion in fibula.[4,5] Wang et al. reported this lesion in rradius[6] and Schlitter el humerus.[7] To the best of our knowledge of literature, till now there is no reported case of lesion affecting the clavicle.

CONCLUSION

Osteofibrous dysplasia is a rare non-neoplastic entity which affect tibia most commonly. Clavicle is a rare site for primary lesions. Osteofibrous dysplasia of clavicle has not been reported till now in literature. It is the first case of osteofibrous dysplasia affecting the clavicle.

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REFERENCES

1. McCaffrey M, Letts M, Carpenter B, Kabir A, Davidson D, Seip J. Osteofibrous dysplasia: a review of the literature and presentation of an additional 3 cases. Am J Orthop. 2003 Oct;32(10):479-86.
2. Frangenheim P. Angeborene ostitis fibrosa als ursache einer intrauterinen unterschenkelfraktur. Arch Klin Chir. 1921;117:22-9.
3. Sweet DE, Vinh TN, Devaney K. Cortical Osteofibrous dysplasia of long bone and its relationship to adamantinoma. A clinicopathologic study of 30 cases. Am J Surg Pathol. 1992 Mar;16(3):282-90.
4. Campanacci M, Laus M. Osteofibrous dysplasia of the tibia and fibula. J Bone Joint Surg [Am]. 1981 Mar;63(3):367-75.
5. Ishida T, Iijima T, Kikuchi F. A clinicopathological and immunohistochemical study of osteofibrous dysplasia, differentiated adamantinoma, and adamantinoma of long bones. Skeletal Radiol. 1992;21(8):493-502.
6. Wang JW, Shih CH, Chen WJ. Osteofibrous dysplasia (ossifying fibroma of long bones). A report of four cases and review of the literature. Clin Orthop. 1992 May;(278):235-43.
7. Schlitter HE. Benign bone tumors with unusual localization & spontaneous fracture. Fortschr Geb Rontgenstr Nuklearmed. 1958 Feb;88(2):195-200.
8. Sweet DE, Vinh TN, Devaney K. Cortical osteofibrous dysplasia of long bone and its relationship to adamantinoma. A clinicopathologic study of 30 cases. Am J Surg Pathol. 1992 Mar;16(3):282-90.
9. Johnson LC. Congenital pseudarthrosis, adamantinoma of long bone and intracortical fibrous dysplasia of the tibia. J Bone Joint Surg. 1972;54-A:1355.
10. Lee RS, Weitzel S, Eastwood EW, Monsell F, Pringle J, Cannon SR, et al. Osteofibrous dysplasia of tibia: is there a need for radical surgical approach? J Bone Joint Surg [Br]. 2006;88-B:658-64.

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