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RESEARCH ARTICLE

Polyostotic fibrous dysplasia- a case report

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ABSTRACT

Fibrous dysplasia is a developmental disorder characterized by replacement of fibrous tissue which causes destruction and overgrowth of the affected bone .when this dysplastic process occurs in single bone is called as monostotic and when occurs in multiple bone is called as polyostotic. This report discusses about a case of polyostotic condition that involves right maxilla, left elbow, right knee and the management of occlusion.

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INTRODUCTION

Fibrous dysplasia is a benign fibro osseous lesion in which medullary bone is replaced by fibro osseous tissue which causes distortion and overgrowth of affected bone. The most commonly occurring type is monostotic Fibrous dysplasia (FD) (Sanjay Kumar Bhadada, 2011). Monostotic FD affects craniofacial bones and ribs, other bones involved are clavicle, tibia and femur; maxilla is more involved than mandible. Polyostotic FD includes jaffes type and Albright syndrome. Commonly affected sites are frontal, sphenoidal, maxilla and ethmoidal, less common are occipital and temporal bone. Painless swelling with cranial and facial asymmetry is the frequently occuring signs (Mansi Agarwal *et al*., 2014), Monostotic FD results in lesser complications than polyostotic (Tomoaki Fukui 2013). Polyostotic fibrous dysplasia patients have more severe deformities in early adolescence and often continue to enlarge after skeletal maturity with progressive deformity and end up with pathological fracture (Matthew, 2005). There may be some mal –alignments, tipping or displacement of teeth due to progressive expansile of lesion.

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In this article we present a rare case of non syndromic polyostotic fibrous dysplasia involving right maxilla, left elbow and right knee regions is presented.

Case presentation: A 32 yrs old male reported with chief complaints of swelling on the right side of his face, left hand region and right knee region for past 20years. No history of skin lesion or endocrine abnormalities. The swelling started in left hand, right leg and involved right maxilla. Growth of the swelling was slow and stabilized to present size. On clinical examination, Extraorally a diffused swelling was present on the right maxilla measuring about 4cm superioinferiorly. 6cm mediolaterally in size , swelling was diffused in shape, with ill defined margins, medially extending to the ala of the nose, laterally extending to zygomatic buttress region, superiorly to infraorbital region inferiorly to upper border of mandible (Fig 1) Swelling on the left elbow, measures about 2cm x 2.2cm in size was ovoid in shape, with ill defined margins (Fig. 2). Swelling on right leg region was ovoid in shape measuring about 3.5cm x 4cm in size (fig. 3). All swellings were hard in consistency, skin color over the swelling, and surrounding structures were normal without secondary changes. Intraoral Swelling involved the alveolar mucosa and palatal region of maxilla extending anterioposteriorly 5cmx2.5cm in size mediolaterally from 12 region to 17 region, with irregular

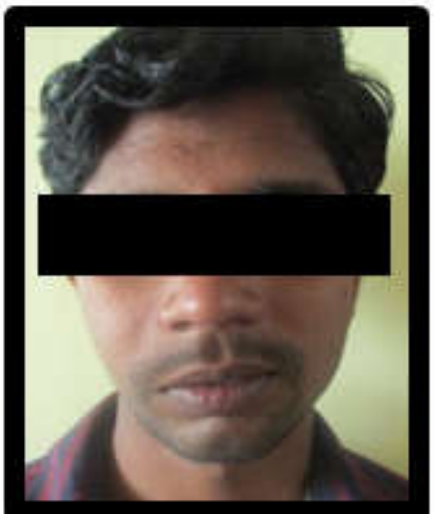

Fig 1. Extra oral

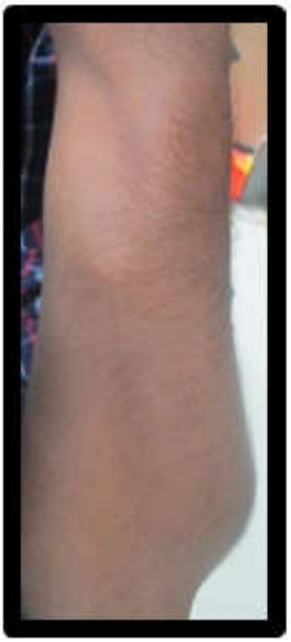

Fig 2. Left elbow

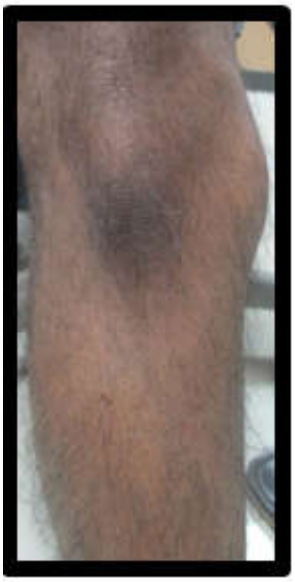

Fig 3. Right knee

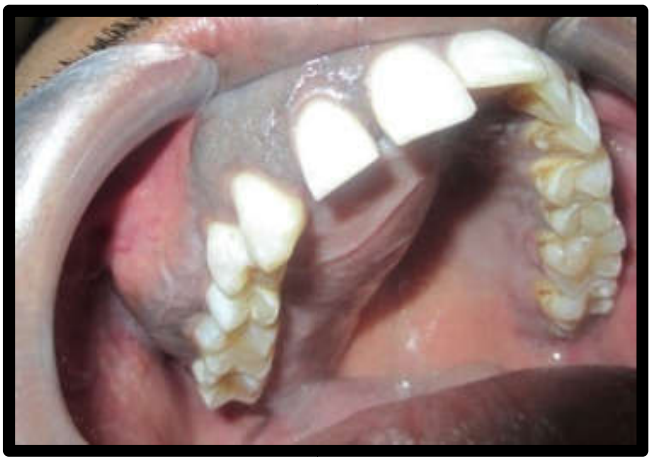

Fig. 4. intra oral

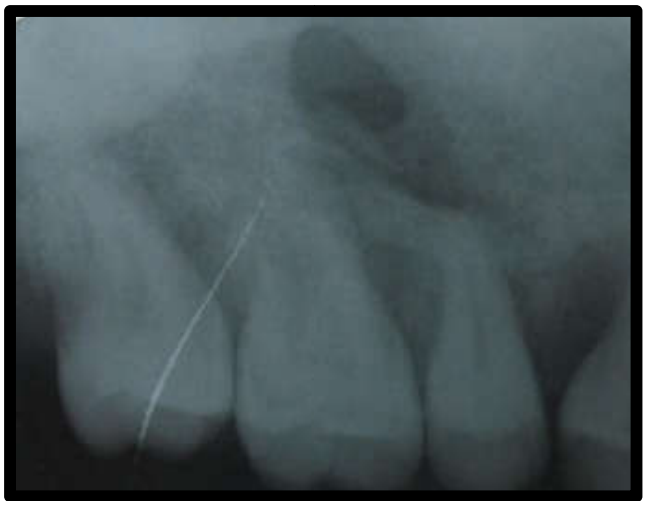

Fig. 5-IOPA 14,15 16,17 region

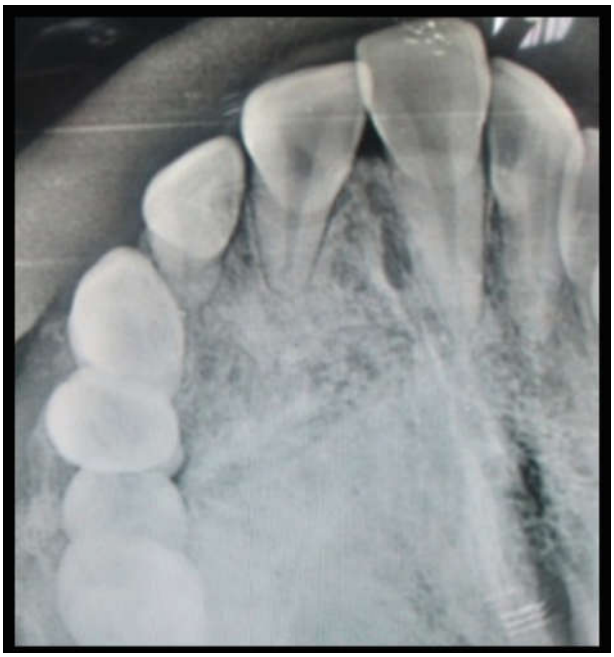

Fig. 6. Occlusal view

shape, ill defined margins, mucosa over the swelling surrounding structures appeared to be normal (fig.4). On palpation it was non tender, bony, hard in consistency without secondary changes.

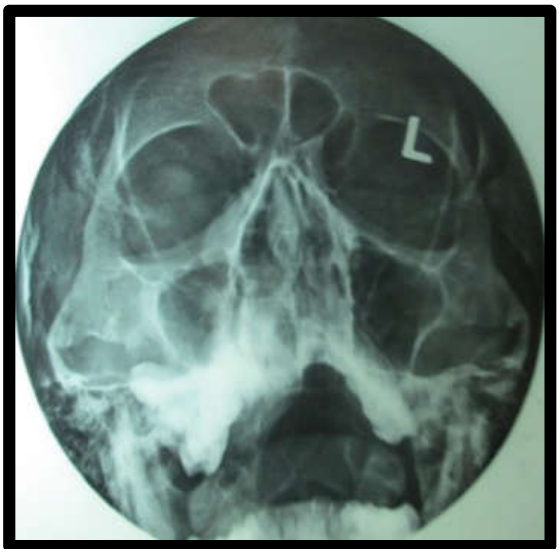

Fig 7. PNS view

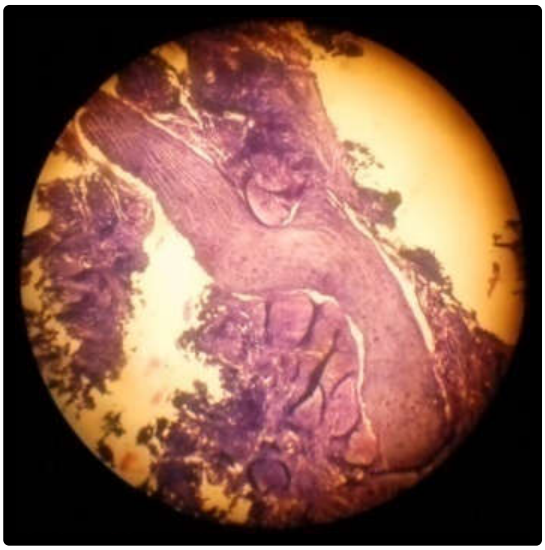

Fig. 8. Histopathology section

Routine hemogram and biochemical were normal. IOPA in relation to 14,16,17 region revealed the presecence of a radiopaque mass present in relation to 14,15,16,17 region with diffuse in nature, flaring of roots with ground glass appearance (fig.5). Diffused radiopacity seen with buccal and palatal expansion extending from 13 to 18 region (Fig 6) PNS view revealed diffuse radiopacity present on right maxilla extending from premolar to maxillary tuberosity and involved the floor of maxillary antrum (Fig 7). On the basis of clinical findings and radiographs the case was provisionally diagnosed as a beningn fibro osseous lesion with differential diagnosis of Paget's disease, and fibrous dysplasia were suspected. Histological diagnosis was fibrous dysplasia (Fig 8). Biopsy performed in right maxilla confirmed the diagnosis of FD. The patient was adviced surgical correction of the lesion; however he was lost follow up.

DISCUSSION

Fibrous dysplasia is a benign bone tumour causing failure in remodeling of immature bone to mature lamellar bone, due to destruction in bone maturation and leaves a mass of immature bone (woven bone) in dysplastic fibrous tissue (Tomoaki Fukui *et al*., 2013).

Trauma is one of etiology for fibrous dysplasia according to various studies due to nonspecific disturbances in bone (White, 2009), however this case is not associated with any history of trauma but with multiple bone involvement since childhood. Radiographic features of fibrous dysplasia shows more radiolucency in earlier stage than in mature lesions with multilocular appearance and may have various radiopaque pattern such as ground glass appearance resembling small fragments of shattered winsheild, that resembles surface of an orange peau d' e orange, a wispy cotton wool arrangement or amorphous dense pattern (Enneking, 1998). In this case, radiographs revealed radiopaque mass present in relation to (Fig.6, 7) 14,15,16,17 region with difussed shape, flaring of roots resembling ground glass appearance hence with radiological findings differential diagnosis of FD, pagets disease and ossifying fibroma Fibrous dysplasia are often present with pathological fractures ,so it's necessary to examine the case completely and single or multiple bone involvement along with radiological findings (Tomoaki *et al*., 2013). Histologic section (Fig. 8) shows dense trabeculae of bone containing osteocytes with minimal marrow spaces .There is lack of osteoblastic rimming surrounding the dysplastic trabeculae suggestive as FD

Conclusion

FD has been reported to respond well to bisphosphonates. Earlier medical therapies with glucocorticoids have been unsuccessful (Anitha, 2015). In this case occlusal discrepancy was present this could be managed by lefort 1 partial osteotomy on the involved side with reduction in vertical height of the maxilla (Denadai *et al*., 2015; Eachempati *et al*., 2015). This approach will correct the growth as well as the occlusion. Prosthetic rehabilitation is followed to correct minor discrepancies by crowns. FD has good prognosis with low rates of malignant transformation.

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