

A CASE REPORT OF JUVENILE CEMENTO-OSSIFYING FIBROMA OF MAXILLA WITH TWO YEARS FOLLOW UP

Mathan Mohan A., Vinod Krishna.K*, Bala Guhan B., Karthikeyan G. R., Thilagam S and Taheera Sultana S

Oral cancer foundation, Head & Neck Services, Karpaga Vinayaga Institute of Medical & Dental Sciences, Madhurantakam, Tamilnadu

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ABSTRACT

Cemento-ossifying fibroma is classified as a benign fibro-osseous lesion of the jaws. It commonly presents as a progressively growing lesion that can attain an enormous size with resultant deformity if left untreated. They commonly affect adult females between the third and fourth decade of life and it is odontogenic in origin. Radiographically, they appear as well-defined unilocular or multilocular intraosseous masses. This lesion is invariably encapsulated and of mixed radiolucent densities. This article reports a case of an 16-year-old female who came to us with the history of recurrent painless swelling at the right maxillary region. The clinical, radiographic and histological features as well as surgical findings are presented.

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INTRODUCTION

A fibro-osseous (FO) lesion which arises from the periodontal ligament is known as Cemento-ossifying fibroma (COF)¹. Juvenile COF arises in children which is more aggressive². COF was first described by Menzel in 1872 as one of a variant of ossifying fibroma³. FO lesions involving jaws and other craniofacial bones are mostly idiopathic, which belongs to heterogeneous group of benign lesions. In this condition, the normal bone is replaced by newly formed mineralized product of fibrous tissue⁴. FO lesions have evolved more commonly with fibrous dysplasia and osseous fibroma rather than florid osseous dysplasia, periapical dysplasia and focal sclerosing-osteomyelitis⁵. COF is a rare, benign, non odontogenic neoplasm of the jaw bone, which commonly occurs in 3rd and 4th decades of life with the female predilection ratio of 2:1 over male. Usually it is asymptomatic, unless a swelling or mild deformity is noted where mandible is more commonly involved than maxilla. Most common site in mandible is pre molar region with 62% to 89 % of incidence, whereas in maxilla it is in molar region with 11% to 26%⁶. Radiographically, COF appears as radiolucent lesion without any internal radio-opacities in early stage when it matures it starts as a radiolucent area which appears as flecked with opacities due to increasing calcification and finally appears as an extremely radio-opaque mass with displacement of adjacent teeth.

As this lesion grows in all direction by equal expansion, it is considered as a centrifugal growth pattern, which is an additional diagnostic criterion. Histopathologically COF consists of proliferating fibroblast and cementoblast with many delicate interlacing collagen fibres arranged in discrete bundles. Increase in islands of cementum will be evolved in matured condition⁶. Authors herein presented a case of juvenile cemento ossifying fibroma of the maxilla which caused maxillary expansion.

CASE REPORT

A 16-year-old female child reported to the outpatient department of oral and maxillofacial surgery, karpaga vinayaga institute of medical and dental sciences, Tamil nadu, India, with a complaint of painless swelling in the right side of the face for the past one year (Fig. 1).



FIG 1 : pre – operative extra oral photograph

On history evaluation, patient had undergone surgery for the same complaint with extraction of right maxillary first pre molar at her native. Further she developed same complaint with a painless swelling and reported to our institute for further management. The patient was apparently asymptomatic 12 months back when she developed a small swelling at right

*Corresponding author: **Vinod Krishna.K**
B30, Kavitha Flats, SBI staff colony 3rd Street, Arumbakkam, Chennai

maxillary anterior labial region, which was gradually increasing in nature

On examination

There was a diffuse swelling present in the right side of the face which measures approximately about 3 x 3 cm which extends superiorly from the right side of the infra orbital rim, inferiorly from 2cm above the lower border of mandible, anteriorly from the ala of the nose and posteriorly it limits at 2.5 cm in front of the tragus of the ear. Intra orally, there was missing of right maxillary first pre-molar, evidence of swelling present in palatal mucosa in relation to right maxillary canine and up to right maxillary first molar which is not crossing to the midline (FIG 2).



FIG 2 : pre – operative intra oral photograph showing there was missing of right maxillary first pre-molar, evidence of swelling present in palatal mucosa in relation to right maxillary canine and up to right maxillary first molar which is not crossing to the midline

Investigation

Routine hematological examinations were carried out and all the blood parameters are within normal limits. Computed tomographic scan of facial bones showed a mixed density of mass with diffuse calcifications involving right maxillary alveolar ridge and expanding the maxillary sinus with well circumscribed outline (FIG.3).

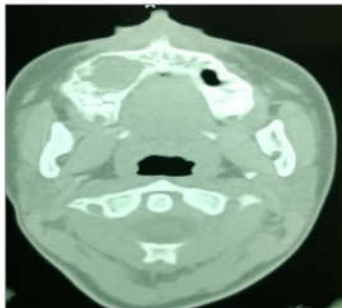


FIG 3: pre – operative CT SCAN of facial bones showing showed a mixed density of mass with diffuse calcifications involving right maxillary alveolar ridge and expanding the maxillary sinus with well circumscribed outline

An Incisional biopsy was done intra orally under local anaesthesia to confirm the type of lesion which was recurrent juvenile cemento-ossifying fibroma (FIG. 4).

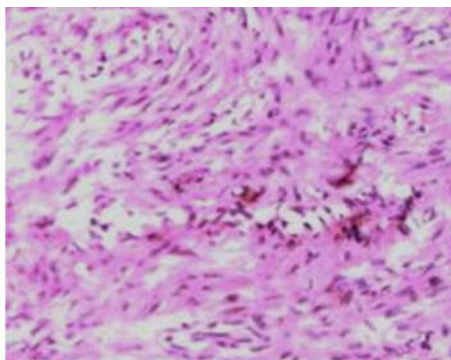


FIG 4: histopathological finding shows trabeculae of immature bone of irregular shapes and sizes and lined by plump osteoblasts and few osteoclasts in a highly cellular and vascular connective tissue. The periphery of the lesional tissue is bordered by many linear, thick and immature bony trabeculae, which reveals recurrent cemento ossifying fibroma.

Management

Pre operatively intra – oral maxillary impression were taken, maxillary acrylic obturator fabricated, informed consent obtained and complete general anaesthetic evaluation done in prior to surgery. Right maxillary weber – ferguson skin incision marking placed and incision made, muco–periosteal flap raised, right partial maxillectomy performed which involves from right maxillary canine anteriorly, medially up to mid palate, posteriorly up to pterygoid plates and superiorly up to infra orbital rim and closure made after placing the obturator in the operated site (FIG. 5).



Fig 5: A – skin incision marking, B – incision and flap raised, C – osteotomy in relation to right maxillary lateral incisor and canine region, D – defect before closure

The excised specimen sent for histopathological examination as excisional biopsy. After a month post-operative, permanent obturator were made and inserted (FIG. 6).



FIG 6: A and B – acrylic obturator, C and D – two weeks post-operative extra – oral photographs

Patient had been called up for periodical follow up once in 15 days for first two month, followed it once in one month for next 4 months and once in two month for next 6 months to observe any changes like recurrence. After two years of follow up, patient adviced to take computed tomography of face which reveals no evidence of recurrence (FIG. 7).

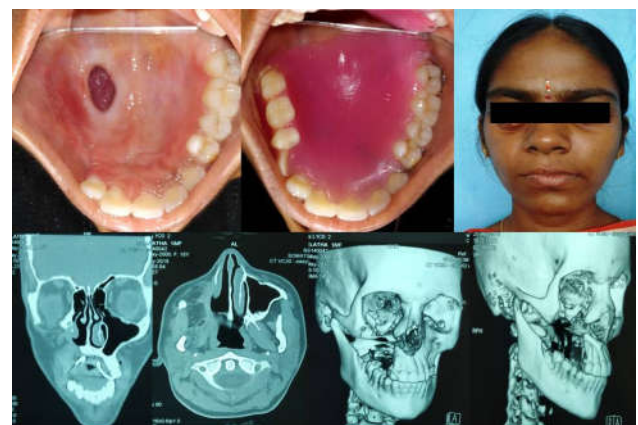


Fig. 7: two years post-operative follow up, clinical and computed tomography of facial bones photographs

Since patient completed her growth spurts, we planned secondary reconstruction for the orbital floor & maxilla with autogenous bone grafts.

DISCUSSION

Waldron and Kramer *et al.* have classified the fibro-osseous lesions involving the jaws³. Occurrence of COF in maxilla is uncommon. Usually FO lesions are seen with the features of slow growing and asymptomatic, but at the age less than 15 years the lesion usually represents as destructive and more aggressive⁸. Classification of Odontogenic neoplasms was given in recent WHO (2005), stating that the COF was replaced by ossifying fibroma⁹. In reference to the literature, COF arose due to developmental origin from the multi-potential cells of periodontal membrane, having ability to produce tumors from cementum, fibrous tissue or lamellar bone¹⁰. COF lesions are seen as spherical, oval or multilocular which are separated by the osteolytic borders and surrounding bone clearly¹¹.

Histopathologically COF reveals proliferation of irregular shaped calcifications within hyper cellular fibrous connective tissue stroma and cementum deposition is noted, appearing as psammoma bodies embedded in a benign fibrous stroma. Radiographically COF reveals as a well circumscribed solitary radiolucency with scattered radiopaque foci. Initially, it doesn't have internal radiopacities. Later as the tumour matures, radiolucent area becomes flecked with opacities, until it appears as radiopaque mass. It has centrifugal growth pattern, with cortical bone expansion and without any perforation. COF resembles spherical in nature and aids in displacement of adjacent teeth. The only treatment modality is surgery but the growth of facial skeleton must be taken into consideration especially in pediatric cases. Extensive tumours are invasive and difficult to manage. For the fear of recurrence, enucleation and curettage are avoided. Extended caldwell luc sublabial approaches are suitable for maxillary COF without affecting esthetics. Lateral rhinotomy is preferred in cases of sinonasal cavity. Endoscopy can be used for assistance for better precision^{7,12}. COF transformation into malignancy is very rare and the prognosis is good¹³.

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