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RESEARCH ARTICLE

JUVENILE OSSIFYING FIBROMA: NOT SO JEJUNE

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Abstract

Juvenile ossifying fibroma (JOF) is a rare benign fibroosseous tumor which involves maxilla more than mandible. It usually occurs in children below 15 years of age. It is locally aggressive with significant growth potential which can raise fears of malignant pathology. This article reports a case of JOF occurring in the maxilla followed by surgical treatment.

Introduction:-

According to new edition of classification of the World Health Organization [1], ossifying fibromas which appear between 5 and 15 years of age, fast growing, radiographically well bordered and histologically consistent with ossifying fibromas are referred as Juvenile Ossifying Fibroma.

Juvenile ossifying fibroma (JOF) is a fibro-osseous, non-odontogenic tumor. It is a rare tumor made up of fibrous tissue and bone and/or cemental elements in various degrees [2,3]. They mainly involve maxilla and very rarely mandible. It is locally aggressive giving suspicion of malignancy. Histopathologically two types have been identified namely psammomatoid and trabecular types [4]. Authors herein presents a case of JOF (psammomatoid type) of the maxilla which caused expansion and destruction of right orbital floor.

Case Report

A 19-year-old female patient reported to our institute with a chief complain of swelling on right mid facial region. While taking history patient reported a rapid and progressive increase in size over a 6 months period, with aesthetic issue but without pain. There was no other relevant medical history nor was any evidence of lymphadenopathy.

On extraoral examination there was visible facial asymmetry on right side of face extending from ala of nose to tragus of ear and from infraorbital margin to upper vermillion border of lip. Intraorally, there was palatal swelling on right, which extended from teeth 11-15 and measured approximately 2 cm in dimensions (Figure 1). On palpation, the mass was uniformly hard in consistency without fluctuation or tenderness. The teeth were healthy and immobile and mucosal lining was normal in appearance. Clinically, due to rapid growth and size of the lesion raised the suspicion of malignancy despite the lack of pain. The differential diagnosis was Fibrous Dysplasia, JOF and Malignant Bone Tumor.

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A CBCT represented heterogenous radiodense/radiolucent mass expanding the right orbital floor (Figure 2). Considering the patient's aesthetic issue and benign radiological features, surgical excision was decided. Therefore, surgical intervention under general anaesthesia was scheduled for lesion excision followed by histological diagnosis.

On gross examination, received partial right-side maxilla with the lesion which was firm in consistency and measuring approximately 06x05x05 cm in dimensions in the Department of Oral and Maxillofacial Pathology & Oral Microbiology. On microscopic examination, the hematoxylin &eosin stained sections revealed extremely fibrocellular connective tissue stroma showing predominately plump fibroblasts & few collagen fibers. Numerous eosinophilic ossifications are seen within the stroma. Concentric lamellated calcified structures with brush borders resembling psammoma bodies and few bony trabeculae were also seen (Figure 3). Therefore, based on histopathological features it was diagnosed as psammatoid form of JOF.

Discussion:-

The most common fibro-osseous lesion of the oral and maxillofacial region is Ossifying fibroma (OF). It is benign neoplasm having progressive growth potential, bony expansions that can result in asymmetry, compromised facial aesthetics and malocclusion [5]. In 2017 WHO classified OF into three types Cemento-ossifying fibroma, Juvenile Trabecular Ossifying Fibroma (TJOF) & Juvenile Psammomatoid Ossifying Fibroma (PJOF) [6].

The histologic variants TJOF & PJOF were first described by El-Mofty [7]. Approximately 75% of PJOF commonly seen within ethmoidal sinuses followed by frontal, maxillary and sphenoidal sinuses whereas only 25% of all cases involves maxilla or mandible [8,9]. In a review of literature PJOF reported case are more as compared to TJOF. Both the types showed predilection for males. In our reported case involved site is maxilla and patient is female [7]. Radiographically JOF exhibit radiolucent and radiodense areas depending upon on degree of calcification [10]. The clinical presentation of JOF comprises of asymptomatic bony hard swelling, rapid painless expansion of affected bone. Complication may arise due to impingement of on neighboring structures due to continuous growth. Intracranial extension has been discovered in neoplasms arising adjacent to the cribriform plates [11].

Histopathologically PJOF is characterized by presence of spherical ossicles known as psammoma bodies in fibroblastic stroma [12]. The unique spherical structures "psammoma like bodies" were termed by Gögl [13]. The psammoma like bodies possesses dark rim of crystals from which needle-like crystalloids project towards the periphery [13] giving it an appearance like brush borders. Sometimes in hyalinized background JPOF shows deeply basophilic concentric lamellated particles [9].

Treatment of PJOF is simple excision with surrounding marginal bone. Larger lesions need more aggressive surgical management. With incomplete surgical excision there is possibility of recurrence hence long term follow up is required [14]. Due to high recurrence rate (30-50%) and aggressive nature of lesion, JOF should be treated as locally aggressive neoplasm [15].

Conclusion:-

PJOF is rare and aggressive neoplasm of young age. Its diagnosis depends upon clinical, radiological and histopathological features. This neoplasm needs to be considered in differential diagnosis of fibro-osseous lesion of head and neck region in young patients. Complete surgical resection is treatment of choice followed by long term follow up due to its high risk of local recurrence.

Figure 1:- Swelling present onright side of palate.

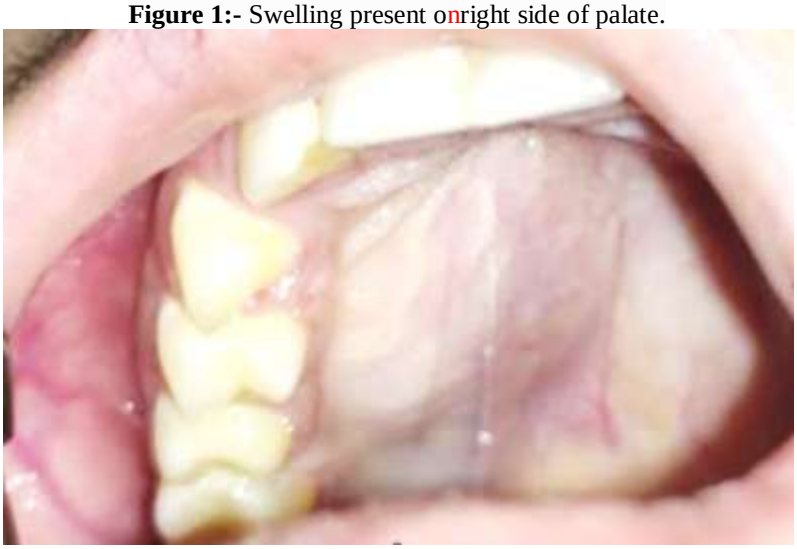
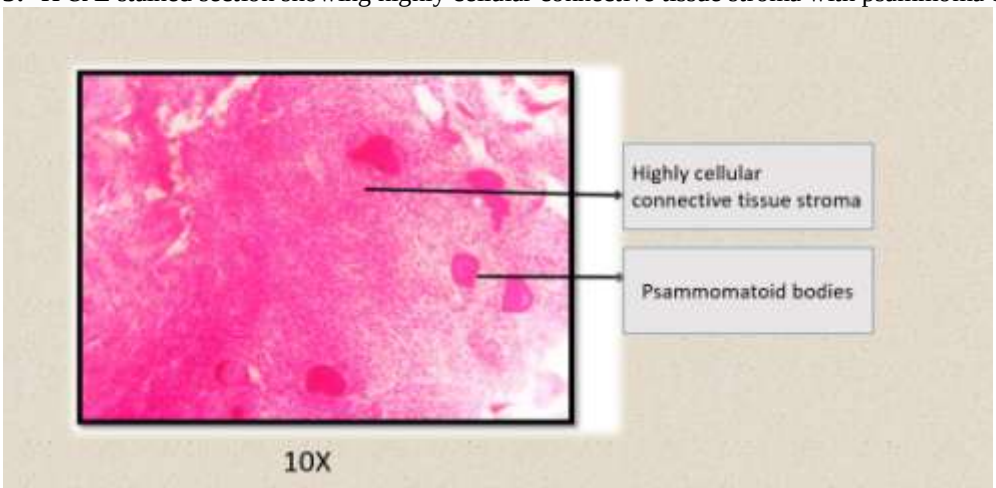


Figure 2:- CBCT image showing destruction caused by lesion.

Figure 3:- H & E stained section showing highly cellular connective tissue stroma with psammoma bodies.



References:-

1. Reichart PA, Philipsen HP, Sciubba JJ. The new classification of Head and Neck Tumours (WHO)--any changes? Oral Oncol. 2006 Sep;42(8):757-8. Epub 2006 May 6.
2. S. Mohanty, S. Gupta, P. Kumar, K. Sriram, U. Gulati. Retrospective analysis of ossifying fibroma of jaw bones over a period of 10 Years with literature review. J Maxillofac Oral Surg, 13 (4) (déc 2014), pp. 560-567.
3. V. Thierbach, S. Quarcoo, A.I. Orlian.Atypical peripheral ossifying fibroma: a case report.N Y State Dent J, 66 (8) (2000), p. 26.
4. Khanna J, Ramaswami R. Juvenile Ossifying Fibroma in the Mandible. Ann Maxillofac Surg. 2018 Jan-Jun;8(1):147-150.
5. Collins, L.H.C., Zegalie, N.F.T., Sassoon, I. et al. A Clinical, Radiological and Histopathological Review of 74 Ossifying Fibromas. Head and Neck Pathol **17**, 433–446 (2023).
6. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ (2017) WHO classification of head and neck tumours chapter 8, 4th edn. IARC, Lyon, pp 204–260
7. El-Mofty S. Psammomatoid and trabecular juvenile ossifying fibroma of the craniofacial skeleton: Two distinct clinicopathologic entities. Oral Surg Oral Med Oral Pathol Oral RadiolEndod. 2002;93:296–304.
8. Foss RD, Fielding CG. Juvenile psammomatoid ossifying fibroma. Head Neck Pathol. 2007;1:33–4.
9. Malathi N, Radhika T, Thamizhchelvan H, Ravindran C, Ramkumar S, Giri G, Gopal D. Psammomatoid juvenile ossifying fibroma of the jaws. J Oral MaxillofacPathol. 2011 Sep;15(3):326-9.
10. Rao S, Nandeesh BN, Arivazhagan A, Moiyadi AV, Yasha TC. Psammomatoid Juvenile Ossifying Fibroma: Report of Three Cases with a Review of Literature. J PediatrNeurosci. 2017 Oct-Dec;12(4):363-366.
11. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 3rd ed. India: Elsevier Publishers; 2009. p. 648.
12. Barnes L, Eveson WJ, Reichart P, Sidransky D. WHO Classification of Tumours - Pathology and Genetics - Head and Neck tumours. France: IARC press; 2005.
13. Solomon MC, Khandelwal SAR, Carnelio S. Psammomatoid juvenile ossifying fibroma of the mandible – A histochemical insight! Internet J Dent Sci. 2009;7.
14. Kalliath, L., Karthikeyan, D., Pillai, N. et al. Juvenile psammomatoid ossifying fibroma with fluid–fluid levels: an unusual presentation. Egypt J RadiolNucl Med **52**, 189 (2021).
15. Reddy AV, Reddy KR, Prakash AR (2014) Juvenile ossifying fibroma with aneurysamal bone cyst: a case report. J Clin Diagn Res JCDR 8(10):ZD01.