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Case Report

Contralateral maxillo-mandibular presentation of central ossifying fibroma

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Abstract

Central ossifying fibroma is a slow-growing, circumscribed, benign and aggressive fibro-bony tumor, known for its expansive growth, the replacement of bone for a fibrous cellular tissue with a focus on mineralization. This lesion occurs in the third or fourth decades of life, with a predilection for females. It affects the mandible in 70% to 80% of cases. Treatment includes enucleation and curettage or surgical resection for larger lesions. Thus, the aim of this article is to present and evaluate a clinical case of central ossifying fibroma in the maxilla in a female patient aged 34 years with a swelling on the left side of the face from child hood which gradually increased in size, with no nasal stuffiness, or loss of vision, smell associated and a mandibular swelling on the contralateral side. An incisional biopsy was performed and the specimen obtained was subjected to histopathological examination which was conclusive of a benign bone lesion, suggestive of central ossifying fibroma.

Keywords: Ossifying fibroma, Neoplasms, Maxilla, Fibro-osseous lesion.

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1. Introduction

Ossifying fibroma (OF) is defined as a benign fibro-osseous, expansile lesion with a circumscribed margin arising from the cells of the periodontal ligament. It is characterized by the replacement of typical bone architecture with either fibroblasts or collagen fibres containing a variable amount of bone or cementum-like tissue or both.[1] It has been known for its progressive, slow-growing behaviour; however, this characteristic may vary broadly. Some lesions may eventually become massive, causing significant aesthetic problems and functional deformities such as facial asymmetry and tooth displacement. Ossifying fibroma, cemento-ossifying fibroma, and cementifying fibroma are often used to describe these entities due to their bone and cementum-like tissue content.[2]

Ossifying fibroma is frequently diagnosed among the population in their second to fourth decade with a male-to-female ratio of 1:5, seldom occurring in children, adolescents, and the elderly. Facial bones, particularly the posterior region of the mandible, are the repeated site of emergence for 75%.[3-4] Moreover, evidence has described their development in the paranasal sinuses, frontal-ethmoid-sphenoid bone, and orbital floor.

With this background, a case report of a 34-year-old female patient diagnosed with ossifying fibroma of the left maxilla is presented in this article with emphasis on clinical, radiological and histopathological features.

2. Case Presentation

A 34 year old female patient visited the outpatient department of our institution with a chief complaint of swelling over the right lower face since one week and a swelling over the left upper face since almost 30 years. Patient had noticed a swelling at the right lower face 1 week ago and was as the same size since then, not associated with pain or paraesthesia. She did not have any difficulty in chewing or eating. Swelling over the left side of the face from child hood, gradually increased in size, and there was no nasal stuffiness, loss of vision, loss of smell, or chronic pain associated with it. Past

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medical, dental and personal history were assessed and found to be non-contributory.

Clinical examination revealed, A single dome shaped swelling measuring around 2.5×2.5 cm present over the right lower 3rd of the face. Anteriorly it extended from about 1 cm from the corner of the mouth, and posteriorly 4 cm from the angle of the mandible, inferiorly up to the lower border of the mandible and superiorly up to the ala tragal line. Skin over the swelling appeared normal, no sinus discharge, no ulceration, it was firm and non-tender on palpation.

The patient had also noticed another progressively growing swelling on the contralateral side albeit in the lower jaw with an ill-defined margin measuring around 8× 8 cm in size over the left side of the upper face extending anteriorly from the ala of the nose posteriorly 2 cm from the tragus of the ear inferiorly up to the corner of the mouth and superiorly 0.5 cm below the lower eye lid, with obliteration of nasolabial fold was evident. Surface appeared smooth and normal on palpation, the swelling was bony hard and non-tender.(**Figure 1**)

Intra-oral examination revealed, A single swelling obliterating the left buccolabial sulcus extending from the distal aspect of 22 posteriorly up to 28. Palatally, it was extending up to the junction of hard and soft palate. There was no surface ulceration, on palpation the swelling was bony hard in consistency and non-tender.(**Figure 2**)

Further, intraoral examination of the other swelling was around 2.5 x 2.5 cm in size obliterating the right buccal sulcus mesial of tooth 43 to the middle of tooth 46, margins were ill-defined and there was evidence of buccal-lingual cortical expansion, on buccal side it extended from the mesial of tooth 43 till the distal of tooth 46, lingually from distal of tooth 46 till tooth 48. Inferior border of the mandible was not palpable due to the swelling. All other para-oral structures were normal.(**Figure 3**)

2.1. Provisional /differential diagnosis

On the basis of history and clinical examination, a provisional diagnosis of a benign bone lesion probably of fibro-osseous nature was given. The following lesions were considered in the differential diagnoses; Central ossifying fibroma and its variants, Fibrous dysplasia, Paget's disease and other odontogenic tumours.

3. Investigations/Imaging/Histopathology

Routine blood investigations were found to be normal. Serum calcium, phosphate, and alkaline phosphatase levels were within normal limits. Thermal and electric pulp testing was done for the teeth associated with the swelling. Teeth 21,22,23,41,42,43 showed immediate response, however teeth 24, 25, 26,27, 44, 45, 46,47,48 showed no response.

IOPA radiograph showed periapical mixed radiolucent and radiopaque lesion involving teeth 22, 26. loss of lamina dura, blunting root resorption in teeth 22, 23, 24. Root displacement in teeth 23, 24 25.In relation to teeth 43,44,45 a periapical radiolucency extending from tooth 43 till the distal aspect of tooth 46 was evident, however full extent was not covered on the radiograph , blunting root resorption and loss of lamina dura in relation to teeth 43,44,45 was observed.

Mandibular occlusal radiograph showed buccal and mild lingual cortical expansion. Displacement of teeth 33 and 34 was evident on the mandibular occlusal radiograph. (**Figure 4**)

Orthopantomography (OPG) showed an increased radiopacity over the left side, complete obliteration of the maxillary sinus, displacement of the teeth 23, 24, 25 with root displacement inferiorly .Periapical radiolucency measuring around 2.5 x 2.5 cm in size with a well-defined corticated margin was seen in relation to teeth 44, 45, 46.Slight bowing of the inferior border of the mandible was also evident which is a characteristic radiographic feature of central ossifying fibroma.

Right lateral oblique view showed radiolucency with a corticated margin seen in relation to teeth 44, 45, radiopaque flecks can be seen internally. Loss of lamina dura and blunting root resorption in teeth 45, 46, which are again characteristic of ossifying bone lesions.

*3.1. Paranasal sinus view revealed complete opacification over the left maxilla.(**Figure 5**)*

CT scan showed a mixed hypo, hyper intense mass in the left side of the maxilla, expansile in nature, and internally it showed some calcified flecks. Hypodense area with evidence of thinning of the lower border of the mandible, with mild flecks of calcification was seen internally in relation to the mandibular swelling. (**Figure 6**)

3.2. Skeletal radiographic survey of long bones and pelvis did not reveal any other bony lesion.

However, a further histopathologic investigation showed islands of trabeculae of mature bone, characterized by prominent osteoblastic rims and highly cellular fibrous connective tissue matrix surrounded with oval spindle cells, consistent with the diagnosis of ossifying fibroma.

Figure 1: Extra-oral swelling over the left maxilla.

Figure 2: Intra-oral swelling over the labial and palatal left maxilla

Figure 3: Inta-oral swelling over the right buccal sulcus.

Figure 4: Inta-oral periapical and mandibular occlusal view showing root resorption and tooth displacement.

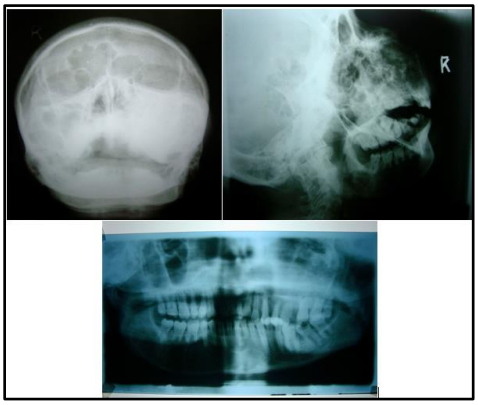

Figure 5: Orthopantomograph and Extra-oral views of skull (Water's and Lateral oblique) showing complete opacification over the left maxilla and a corticated radiolucency with few radiopaque flecks over the right mandible.

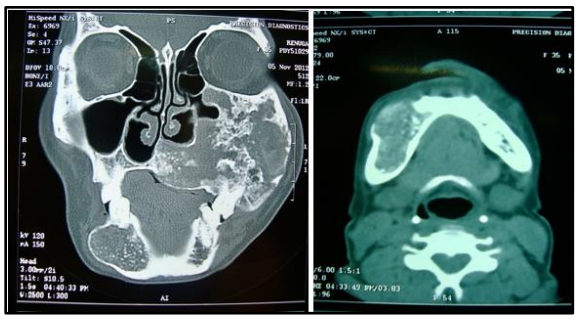

Figure 6: Non-contrast coronal and axial view of maxilla and mandible showing mixed hypo-hyper intense expansile mass with evidence of calcified flecks.

4. Discussion

The first World Health Organization (WHO) in 1971, classified four types of cementum containing lesions: fibrous dysplasia, ossifying fibroma, cementifying fibroma and cemento-ossifying fibroma.[5-6] According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial regions were divided into two categories, osteogenic neoplasms and non-neoplastic bone lesions; cementifying ossifying fibroma belonged to the former category. However, the term ‘‘cementifying ossifying fibroma’’ was reduced to ossifying fibroma in the new WHO classification in 2005.[7-8] To better reflect the fact that these lesions develop in the tooth-bearing regions of the jaws and are benign mesenchymal odontogenic tumors that likely originate from the periodontal ligament, the WHO consensus panel agreed to restore the term "cemento-ossifying fibroma" (COF) in 2017.[9]

Ossifying fibroma has a female predilection and occurs in patients of wide age range between (10- 59yrs, mean 32yrs) with peak incidence in the third or fourth decades of life. Most common site of occurrence is the mandibular premolar- molar region, and about 30% of cases occur in the maxilla. They are slow-growing lesions, hence, the cortical

plates of the bone and the overlying mucosa or skin are invariably intact. This patient had several common characteristics of ossifying fibroma; expansile, smooth surface,hard and non-tender however, this lesion was located in the anterior region of left maxilla and right mandible causing a clinical dilemma of whether it could possibly be fibrous dysplasia. Root divergence, displacement of teeth in the tooth- bearing region, root resorption, mandibular lower border bowing were all associated features of the tumour.

On the basis of its histopathological features, which help differentiate it from other fibro-osseous lesions and provide insights into its behaviour. The two main histopathological variants of COF are described as:

Central Ossifying Fibroma (Cementum-like Type): This variant is characterised by the presence of cementum- like material within the fibrous stroma. Histologically, it shows the formation of acellular cementum-like tissue, which resembles normal cementum found on the root surface of teeth. This type of COF is less cellular and has a more well-defined appearance, exhibits slow growth and a lower recurrence rate.

Central Ossifying Fibroma (Osteoblastoma-like Type): This variant is characterised by the presence of woven bone trabeculae within the fibrous stroma. Histologically, it shows a highly cellular fibroblastic stroma with prominent osteoblastic activity, leading to the formation of woven bone trabeculae. This type is more cellular and exhibits a more aggressive growth, with a greater tendency for recurrence.

In addition to these two main variants, it may also present as lesions exhibiting features of both types, thus showing a combination of cementum-like material and woven bone like pattern.

However there does exist a diagnostic dilemma for fibro-osseous jaw lesions between ossifying fibroma, fibrous dysplasia and focal osseous dysplasia. However, ossifying fibromas are known to be well circumscribed, aggressive lesions characterized by expansion, tooth displacement, and root resorption. Recurrence rate of central ossifying fibroma in maxilla was found to be comparatively higher than that in mandible, due to large size and inaccessibility.[10]

5. Conclusion

Ossifying fibromas ordinarily occur as solitary lesions, but in rare instances they are seen at multiple sites. The present case is one such rare presentation involving contralateral jaw sites. Ipsilateral jaw sites and involvement of multiple sites have been reported in the literature. However, to the best of our knowledge of the available literature, this is the first such case with contralateral site involvement in an adult female patient and hence warrants its understanding to the clinician, pathologist and the surgeon which is the main objective of this case report. The patient underwent enucleation and surgical curettage, under general anaesthesia, and was followed-up for 2 years, with no signs of recurrence.

6. Source of Funding

None.

7. Conflict of Interest

None.

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