Ossifying Fibroma of the Anterior Maxilla: A Case Report

Satya Ranjan Misra[1], Shivani Sahu[2]

[1]Professor & Head, [2]Intern, Department of Oral Medicine & Maxillofacial Radiology, Institute of Dental Sciences, Siksha 'O' Anusandhan (deemed to be University), Bhubaneswar-751003, India

Abstract

'Ossifying fibroma'can be described as a rare benign neoplasm classified under fibro-osseous lesions. Arising from the periodontal ligament's mesenchymal blast cells, it forms bone, cementum as well as osteo-cementum like hard tissue. Though a benign osseous tumour, it exhibits an aggressive behaviour on recurrence. Clinical features vary from swelling, facial asymmetry, mild displacement of teeth, pain or paraesthesia when there is entrapment of nerves. As in other osseous neoplasms, this also has radiologic changes ranging from a mixed radiopaque-radiolucent to a totally radiopaque appearance, but there is no pathognomonic manifestation. Maxillary lesions have to be differentiated from fibrous dysplasia. A case report of ossifying fibroma in the anterior maxilla is reported here.

Keywords: *Benign aggressive tumour, fibro-osseous lesion, mixed radiolucent-radiopaque appearance.*

Introduction

Ossifying fibroma is currently defined by WHO as a benign neoplasm, often presents well-demarcated borders and is composed histologically of fibro-cellular stroma and variable amounts of mineralised materials showing different morphological appearance. Initially WHO had classified Ossifying Fibroma (OF) as a fibro-osseous neoplasm. In 1972, WHO had first classified it into 'ossifying fibroma' and 'cemento-ossifying fibroma'. In 1992, WHO brought it under 'cemento-ossifying fibroma'. In 2005, it was replaced by WHO as 'ossifying fibroma'[(1)].

OF, a benign neoplasm mostly occurs in the tooth bearing areas of the jaws[(2)], commonly seen in mandibular molar and premolar regions[(3)], and do not arise in the long bones. This lesion is predominant in 2nd to 4th decade of life and more common in females, although the central variant is rare among females[(1)]. This lesion is believed to arise from the multipotent periodontal ligament cells which form cementum, alveolar bone and fibrous tissues. Treatment is done conservatively if the lesion is small, otherwise surgical resection. Recurrence rates of OF is low, but follow-up should be done.

Case Report: A female patient of age 27 years, reported to the hospital, complaining of swelling on the left upper front teeth region of the face for the past 3 months. History revealed that the swelling was a slow growing one and was not associated with pain or any discharge. On clinical examination extra orally a single localized, bony hard swelling was visible on the right side of the upper part of anterior maxilla. The swelling was not mobile but the skin over the swelling was freely movable without any secondary changes. Intra orally the bony swelling in the anterior part of the maxilla involved the alveolus of central, lateral and canine region and extended both labially and palatally without any secondary changes. There was displacement of 21 and 22, extrusion of 21 and spacing between 21 and 22.

Since the history revealed that the swelling was a fresh one, slow growing and bony hard in consistency and the swelling was well localized it is a bony swelling.

Corresponding Author:
Dr. Shivani Sahu
Intern, Department of Oral Medicine & Maxillofacial Radiology, Institute of Dental Sciences, Siksha 'O' Anusandhan (Deemed to be University), Bhubaneswar-751003, India
e-mail: shivanis32@gmail.com

As the swelling extends both labially and palatally, it is a benign bony neoplasm. Fine needle aspiration cytology (FNAC) was performed and no aspirate was obtained suggestive of the solid nature of the lesion.

Patient was subjected to radiologic investigations. Periapical radiograph of maxillary left anterior region shows well defined unilocular radiolucency which is ovoid in shape and is present between 21 and 22. Maxillary cross-sectional occlusal radiograph shows unilocular radiolucency ovoid in shape in between 21 and 22 regions with slight displacement of 21 and 22. Panoramic radiograph revealed a well circumscribed unilocular radiolucency involving 21-22 region displacing 21 and 22. Radiolucency was ovoid in shape with a radiopaque border.

Computed tomography scan of the swelling in axial, coronal and sagittal sections shows a space occupying lesion in the left maxillary alveolar segment. The lesion gives areas of both hypodense and hyperdense. The Hounsefield value was 750 suggestive of a benign bony neoplasm. 3DCT shows extension of the lesion till the floor of the nasal cavity on the left side without breach in the nasal floor.

Under local anaesthesia, incisional biopsy was done in 21-22 region and specimen stored in formalin and sent for histopathological examination. Histology revealed cellular fibrous tissue containing mineralized material. Connective tissue stroma with cementum like areas within the connective tissue; with spindle and stellate shaped cells. Histopathologic features suggestive of ossifying fibroma. The patient underwent complete surgical excision of the lesion for treatment.

Discussion

'Ossifying Fibroma' term has been used since 1927, and grouped together as 'cementum containing tumours' since 1968[4]. According to Eversole et al, Central Ossifying Fibroma (COF) can be described as:a well-demarcated benign fibrous neoplasm which is aptly designated as an 'fibro-osseous lesion'. Menzel described cemento-ossifying fibroma in 1872, while Montgomery coined it in 1927. Until 1948, COF and Fibrous Dysplasia were considered as variants of same entity, but OF has been considered a distinct lesion after Sherman and Sternberg described it in detail[5].

This lesion shows female preponderance, initially ratio being 3:1, while currently it has been recorded as 1.8:1[6]. In a report done in 2017 by Tapas et al suggests of female predominance in the range of 2:17 to 5:111[2]. Odontogenic fibroma (OF) is a 'benign odontogenic tumourwhich is characterized by various amounts of odontogenic epithelium in a mature fibrous stroma. Two types of the lesion are: an intraosseous or central OF (COF) and an extraosseous or peripheral. The intraosseous variant is an extremely rare tumour with clinical, radiographic, and histopathologic variable findings. A thorough review of the English literature revealed 78 cases of COF so far. Thus, we report an additional case of COF occurring in the maxilla of a 27-year-old woman. In addition, we performed a brief description and discussion of the cases reported in the maxilla and mandible. Matos et al has noted that this lesion was exclusive to mandible, however epidemiologic profile has changed with an equal distribution between anterior maxilla [54.4%] and posterior mandible [45.6%][6]. Eversole has reported in his studies that most frequent lesion occurred in mandibular molar [52%], then premolar [25%] as the second most common site in mandible, then incisor [12%]as third and lastly cuspid [11%] regions, the least commonly affected site of mandible[3]. OF has been reported to be prevalent in 2nd to 4th decades of life. In cases of maxilla, it is commonly seen in canine fossa and zygomatic arch area. OF clinically appears as a slow-growing expansile asymptomatic infra-bony mass presenting with facial asymmetry. Displacement of teeth may be an early clinical feature, while it may be present for some years. Pain and paraesthesia may be uncommon, yet still reports of presence have been recorded.

The cause of origin of OF is mostly considered to be the pluripotent mesenchymal cells of Periodontal Ligament. Pathogenesis of tumour can be induced by trauma to PDL from extraction, chronic inflammation due to pericoronitis, fibrosis due to periapical infection, or injury, etc. Brademann et al presented a hypothesis claiming mesenchymal cells present in bones other than maxilla and mandible can differentiate into PDL and a trauma may induce the lesion. Recently it has been discovered that a gene mutation of HRPT2, a tumour suppressor gene, can cause this lesion[1].

Fibro-osseous lesions are considered as there placement of bony constituent by fibrous tissue that later mineralize into woven or lamellar bone or cementum, presenting with various clinical features and microscopic appearances[3]. Radiographically the lesion appears variably due to difference in degree of mineralisation.

Amount of calcification differentiates a unilocular or multilocular radio-opaque image, or a radiolucent image with mixed density of opacified material. Root resorption and mobility can be appreciated in few cases[7]. Rind sign is a characteristic radiographical feature which is seen as a radiolucent line around the lesion's periphery, suggesting a capsule like nature[5]. MacDonald-Jankowski claims that OF is well demarcated and round or ovoid in shape as compared to Fibrous Dysplasia (FD), whereas OF presents an irregular shape, especially if occurs rapidly. If it grows along the body of the jaw, showcases an aggressive local growth. As compared to OF, fibrous dysplasia has the classical ground glass appearance radiographically. Radiotherapy is contra-indicated as a definitive measure of management.

Histological image suggests of ossified islands of bone and cementum within the hypercellular fibrous stroma. The trabeculae are variable in sizes, from lamella to woven patterns[8]. The characteristic sign of OF is interspersion of bony trabeculae and cementum like materials in a connective fibrous tissue stroma, along with presence of osteoblastic rimming[5]. CT and CBCT aid in radiographical differential diagnosis of OF[1].

Table 1 the comparison between OF and FD needed for their differential diagnosis, as they present similarities in their features.

Table 1: Differences between COF and FD

	Central Ossifying Fibroma	**Fibrous Dysplasia**
1. Category	Benign neoplasm	Bone disease
2. Clinical features	Painless hard swelling, facial asymmetry, tooth dislocation	Painless hard swelling, facial asymmetry, compression (paraesthesia, deafness)
3. Age prevalence	2^{nd}-4^{th} decade	1^{st}-2^{nd} decade
4. Sex predilection	Female	Both genders are equally affected
5. Variants	Juvenile and Normal	Mono-ostotic and Polyostotic
6. Etiology	PDL (induced by trauma), HRPT2 gene mutation	Gene mutation of GNAS1 gene
7. Hormone involvement	Not hormone related	Hormone related
8. Invasive	Continuous growth	Self-limited
9. Radiographically	Well demarcated bony margins, 'rind' appearance	Ill-defined margins, ground glass appearance, radiolucent lytic lesion
10. Histopathology	Fibrous capsule, osteoblastic rimming, deposits of osteoid	Chinese letter pattern, woven bone, loosely arranged fibrous stroma
11. Treatment	Surgical enucleation	Surgical recontouring
12. Radiotherapy	Contra-indicated	
13. Prognosis	Good, though there are chances of recurrence for larger lesions	Good

Taking into consideration the size, location and aggressiveness of the lesion, there are varied ways for management of the tumour, e.g. conservative observation, curettage, resection. In smaller size lesions, management is approached conservatively. In such cases long term follow-up is maintained. Recurrences in those conditions are treated by resection. Although resection of a massive portion of the jaw even though the lesion is benign in nature, is discouraged by many dentists. Some authors contradict radiotherapy for the management of this neoplasm, although roentgen therapy can be advocated to inhibit further growth of remnants left after operation. Aggressive lesion can be managed by en bloc resection as a definitive therapy[5].

Figure 1a: A single localized, bony hard swelling was visible on the right side of the upper part of anterior maxilla; 1b: A well circumscribed swelling in the left anterior maxillary lateral incisor region with bicortical expansion. 1c: A well circumscribed swelling in the left maxillary lateral incisor and canine, with displacement of 21, obliterating the labial sulcus; 1d: Diffuse swelling in the left anterior hard palate in relation to maxillary lateral incisor and canine.

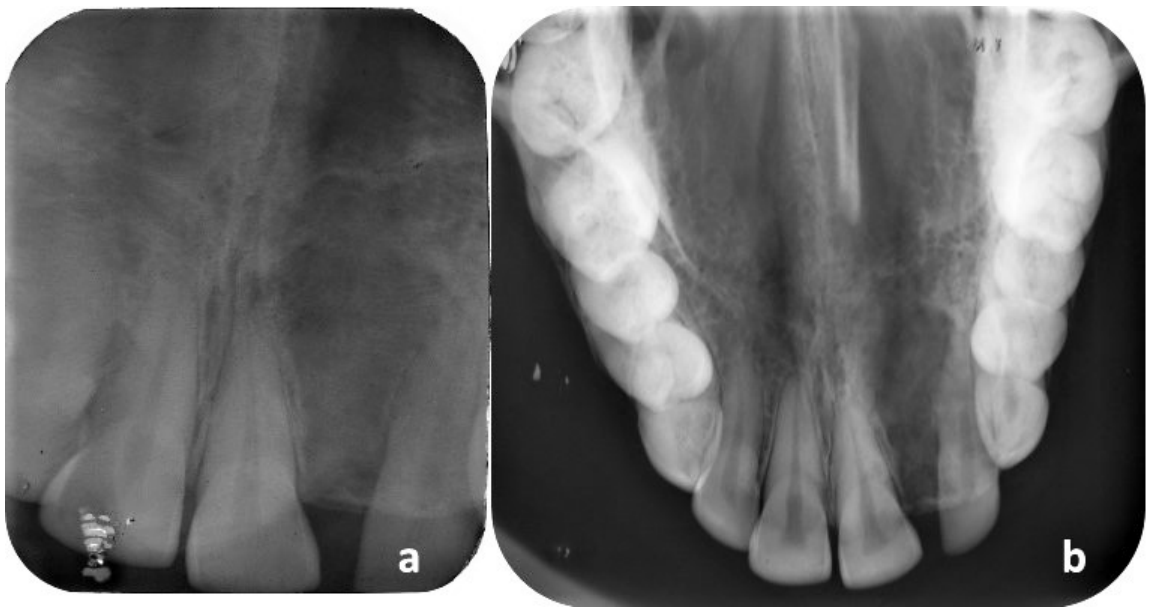

Figure 2a: Intra-oral periapical radiograph showing well defined unilocular radiolucency which is ovoid in shape and is present between 21 and 22. 2b: Maxillary cross-sectional occlusal radiograph shows unilocular radiolucency ovoid in shape in between 21 and 22 regions with slight displacement of 21 and 22.

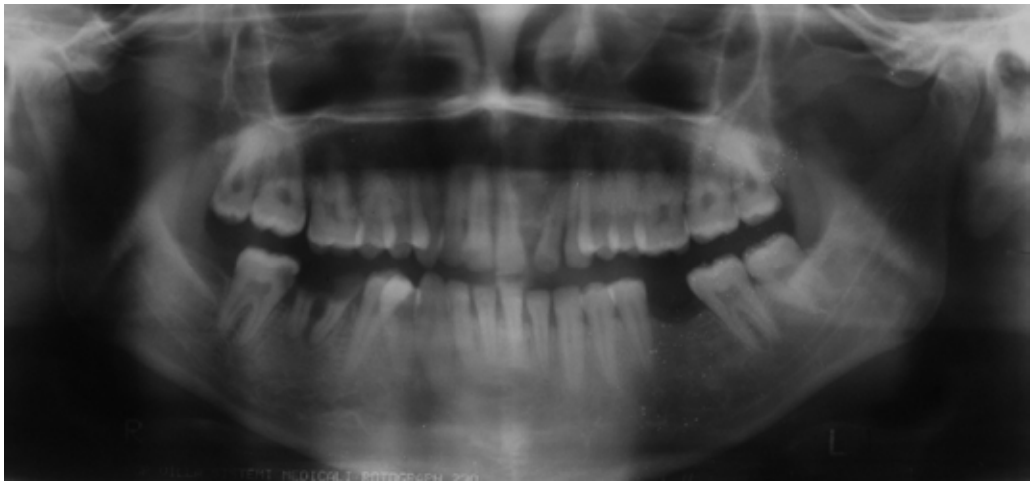

Figure 3: Panoramic radiograph revealed a well circumscribed unilocular radiolucency involving 21-22 region displacing 21 and 22. Radiolucency was ovoid in shape with a radiopaque border.

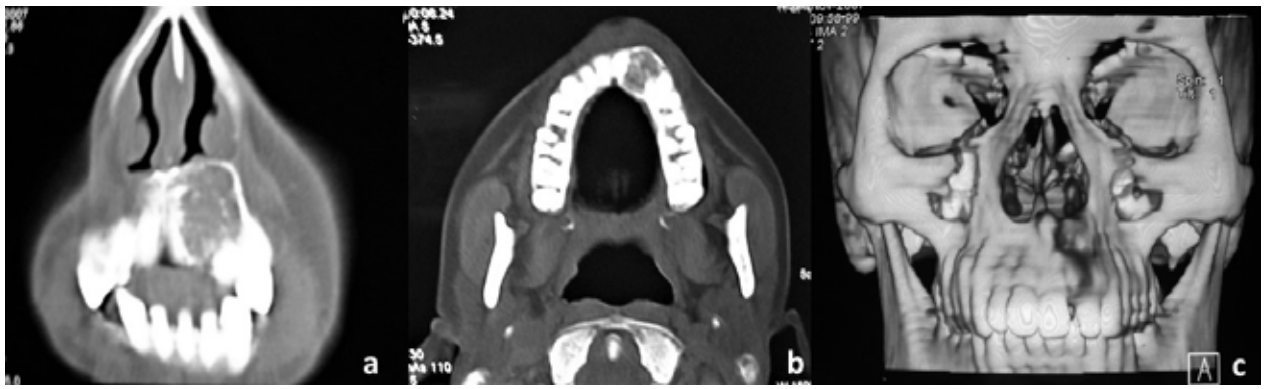

Figure 4a: Computed tomography scan of the swelling in coronal sections shows a space occupying lesion in the left maxillary alveolar segment with heterodense internal structure; 4b: Computed tomography scan of the swelling in axial sections shows a space occupying lesion in the left maxillary alveolar segment with areas of both hypodense and hyperdense. 4c: Three-dimensional reconstructed Computed Tomography scan shows extension of the lesion till the floor of the nasal cavity on the left side without breach in the nasal floor.

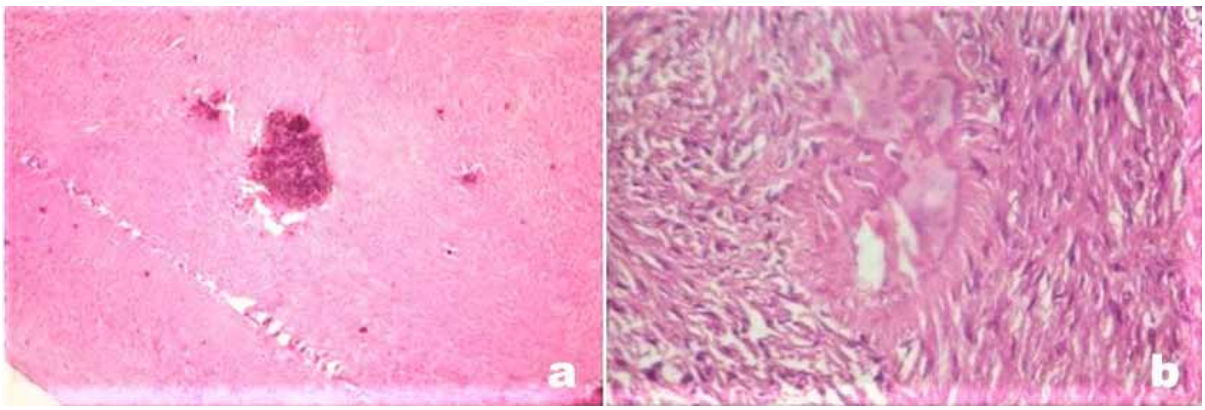

Figure 5a: Histopathologic examination (original magnification 10X) revealed cellular fibrous tissue containing mineralized material. Connective tissue stroma with cementum like areas within the connective tissue; with spindle and stellate shaped cells, suggestive of ossifying fibroma; 5b: Histopathologic examination (original magnification 40X) revealed cellular fibrous tissue containing mineralized material. Connective tissue stroma with cementum like areas within the connective tissue; with spindle and stellate shaped cells, suggestive of ossifying fibroma.

Conclusion

Amongst the many bony swellings encountered in the oro-facial region, odontogenic tumours and cysts are amongst the most common lesions. However, fibro-osseous neoplasms like the ossifying fibroma should be considered in the differential diagnosis and maxillary lesions especially have to be differentiated from fibrous dysplasia because of the aggressive nature of this benign neoplasm, the treatment has to be quite radical.

Conflict of Interests: None

Ethical Permission: Approved

Funding: Nil

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