Odontogenic fibroma like lesions: Clinico-pathological considerations

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Abstract  Radiographically enlarged dental follicle is observed in many cases associated with delayed tooth eruption. Pericoronal radiolucencies are seen in neoplasms (odontogenic fibroma), developmental anomalies (regional odontodysplasia), hamartomatous lesions (odontogenic epithelial hamartoma), opercula of third molars, in follicles associated with unerupted third molars, regional odontodysplasia, in dental follicles around impacted teeth of enamel dysplasia with hypodontia syndrome and amelogenesis imperfecta cases with multiple impactions. Interestingly, operculum and the follicle of these lesions histopathologically are identical to odontogenic fibroma (WHO type). However, the lack of universally accepted clinic-pathological features for such lesions may hinder their recognition. We report a case of regional odontodysplasia wherein (in which) the impacted canine is surrounded by radiolucency measuring about 1.5–2 cm, histopathologic examination of the excised opercula revealed features reminiscent of central odontogenic fibroma (WHO type) with an abundance of odontogenic epithelium and calcifications. The aim of this paper is to discuss various lesions exhibiting histopathological features similar to odontogenic fibroma, as awareness of the clinicopathological features of such lesions is very important to plan proper treatment.

Introduction

The crowns of unerupted teeth are normally surrounded by the dental follicle, a remnant of the enamel forming organ that is lined by reduced enamel epithelium. The follicle space can vary considerably under normal conditions and tends to enlarge during eruption. Guidelines to differentiate between a normal and an abnormal dental follicle space include: pericoronal space exceeding 2.5 mm for teeth other than maxillary canines on periapical radiographs or 3 mm in panoramic radiographs, or follicular radiolucency exceeding 2.5 mm1). Various pathologies are associated with pericoronal radiolucencies including cystic lesions, neoplasms (odontogenic fibroma), developmental anomalies (regional odontodysplasia) and hamartomatous lesions. Normal dental follicles associated with unerupted teeth are frequently misinterpreted histologically as central ossifying fibroma (COF).

Central odontogenic fibroma is a rare benign tumour arising within the jaws. The relatively few reports in the literature suggest that odontogenic fibroma is rare and, in addition, its histogenesis and even its existence as an independent entity are disputed. Interestingly, the term odontogenic epithelial hamartoma was used in the past to describe lesions that were clinically and histopathologically identical to odontogenic fibroma and considered to represent a transitional stage between a developmental anomaly and a true odontogenic tumor2).
opercula of which histopathologically, showed features consistent with odontogenic fibroma (WHO type). Discussion of the origin, histogenesis and clinicopathological features of those lesions which shows features of odontogenic fibroma histopathologically is provided.

**Case Report**

A 13 year old male was referred to the Clinic of Jaipur Dental College with the presenting complaint of mild soft tissue swelling over the right side of the mandibular arch and delayed teeth eruption. Past dental and medical history was not contributory. There was no history of trauma or local damage and no significant family medical history.

Clinical examination revealed the presence of a soft tissue swelling on the right side of the body of the mandible which was $1 \times 1$ cm in size, irregularly shaped, firm in consistency with overlying mucosa normal in colour. Right permanent mandibular central incisors, canine and second premolar were missing while right mandibular lateral incisor showed signs of hypoplasia (Fig. 1). Remaining teeth were

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**Fig. 1** Intra-oral photograph showing swelling in relation to canine region, missing mandibular central incisor, second premolar and hypoplastic lateral incisor

**Fig. 2** Orthopantomogram (OPG) reveals missing central incisor and pericoronal radiolucency surrounding impacted canine and premolar
unaffected. Therefore, the dental index showed

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\begin{array}{cccccccc}
7 & 6 & 5 & 4 & 3 & 2 & 1 & 1 \\
6 & 5 & 4 & 3 & 2 & 1 & 2 & 3 \\
4 & 2 & 1 & 2 & 3 & 4 & 5 & 6 \\
2 & 1 & 2 & 3 & 4 & 5 & 6 & 7
\end{array}
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Orthopantomogram revealed the missing right permanent mandibular central incisor, lateral incisor was hypoplastic (root canal treated), mandibular right canine and premolar were impacted. An unusual finding in relation to impacted right permanent mandibular canine was the presence of enlarged pulp chamber and a radiolucency completely surrounding the crown (Fig. 2). A provisional diagnosis of enamel hypoplasia was made and before planning any treatment, orthodontist opinion regarding orthodontic management of impacted teeth was taken. After thorough examination orthodontist was of the view that 43 was malformed and did not show any signs of eruption though dental age was beyond 12 years. So it was decided to first surgical remove 43 under local anesthesia and then fixed orthodontic therapy was recommended to align 45 into occlusion.

The surgery was planned with consent of parents. The Inferior alveolar nerve block was given on right side. The incision extending from mesial of 42 to 44 was made and the envelop flap was raised and the lesion was exposed. Then 43 was surgically extracted along pericoronal tissue. The defect was curetted followed by repositioning of flap and interrupted sutures were given. Then both hard tissue and soft tissue specimen was sent to Department of Oral and Maxillofacial Pathology, Jaipur Dental College for histopathological examination. Patient was referred to Orthodontic Department for fixed appliance therapy, where his treatment is in process (Fig. 3).

Soft tissue specimen measured 1.5 \times 1 \text{ cm} was firm in consistency with smooth surface and was found to be gritty on cutting. The hard tissue (tooth in two pieces) was sent for decalcification (Fig. 4). Microscopic examination revealed the presence of cellular connective tissue stroma made up of dense bundles of collagen fibers arranged in whorled pattern and the lesional tissue varies from fibrous to myxoid type with islands of odontogenic epithelium and calcifications scattered all over the connective tissue stroma (Fig. 5a). Some of the islands showed
ameloblasts like cells and stellate reticulum cells while some are solid lacking typical features of odontogenic epithelium (Fig. 5b). Numerous calcifications in the form of dentinoid and cementum were seen (Fig. 5c). Dysplastic dentin with numerous areas of inter-globular dentin surrounding the enamel space and normal cementum was seen in decalcified sections (Fig. 5d).

Discussion

Odontogenic fibromas are defined by the World Health Organization (WHO) as benign fibroblastic odontogenic neoplasms containing varying amounts of apparently inactive odontogenic epithelium and foci of dystrophic calcifications. The WHO-type central odontogenic fibroma has characteristic histopathologic features including a mature cellular fibroblastic proliferation in association with abundant odontogenic epithelial strands with varying amounts of calcified material resembling dysplastic dentine. Calcifications associated with odontogenic epithelial cells can also be found in pericoronal third molars, in follicles associated with unerupted third
molars\textsuperscript{5}, with regional odontodysplasia\textsuperscript{6}, in dental follicles of multiple impacted teeth\textsuperscript{7}, in amelogenesis imperfecta cases with multiple impactions\textsuperscript{8} in Enamel Dysplasia with hypodontia and central odontogenic fibroma like tumors\textsuperscript{9}.

Philipsen et al.\textsuperscript{10} studied 74 non-erupted molars and found 54.1\% of the specimens could be diagnosed as odontogenic tumors or hamartomas. Twenty-nine point seven percent showed a hitherto unrecognized odontogenic lesion of hamartomatous character, termed odontogenic giant cell fibromatosis (OGCF). Normal dental follicles associated with unerupted teeth are frequently misinterpreted histologically as COF\textsuperscript{11}. According to Gardner, lesions surrounding the crown of an impacted tooth and consisting of fibrous connective tissue with variable amounts of odontogenic epithelium and calcifications should be classified as hyperplastic dental follicles (HDF), despite their superficial resemblance to COF\textsuperscript{12}.

Picosirius red polarization method can be used as a diagnostic tool to differentiate between COF and HDF. The polarization colors of thick fibers (1.6–2 mm) of COF will be significantly more green and greenish-yellow as compared with those HDF. Greenish to yellow polarizing fibers of COFs suggest that the collagen in COFs is loosely packed and might be composed of procollagens, intermediates, or other pathologic collagens rather than the tightly packed fibers seen in hyperplastic dental follicles\textsuperscript{13}. Present case histopathologically and radiographically was consistent with HDF but the presence of underdeveloped tooth needed further investigation to confirm the association of developmental disturbances.

Feller et al.\textsuperscript{14} presented a case with a rare syndrome characterized by enamel dysplasia and multiple unerupted teeth with large solid fibrous pericoronial lesions manifesting with odontogenic fibroma-like features. Though present case showed impacted teeth and enamel hypoplasia, major criteria for the syndrome are enamel dysplasia with generalized amelogenesis imperfecta-like features and atypical hyperplastic dental follicles with microscopic features of central odontogenic fibroma WHO-type (follicle analogue) attached to the crowns of multiple impacted teeth. Minor features of some cases are anterior open-bite malocclusion, supernumerary teeth, pulpal calcification, aberrant roots with hypercementosis, and hypodontia. Calcified psammomatous laminar deposits, frequently associated with inactive odontogenic epithelium are amyloid positive, a feature not found in COF and could be regarded as an important microscopic feature in distinguishing the COF-like tumors in the case reported from true WHO-type COFs\textsuperscript{9}. Amelogenesis imperfecta like features were absent in this case, so diagnosis of enamel dysplasia with hamartomatous atypical follicular hyperplasia (EDHFH) syndrome can be excluded.

Central odontogenic accounts for less than 0.1\% of all odontogenic tumors and the lesion is central in the jaws and has a slow persistent growth. Exceedingly rare is the association of COF with an impacted tooth and very few cases are reported in the literature\textsuperscript{15}. The unusual location, the presence of impacted hypoplastic teeth does not favour the diagnosis of COF.

Regional odontodysplasia (RO) is an unusual nonhereditary developmental anomaly of tooth formation, involving both epithelial and mesenchymal-derived dental tissues characteristically affecting one or several adjacent teeth of the same quadrant\textsuperscript{16}. Usually, it affects a single maxillary quadrant but other instances may be found, such as two different affected quadrants, or involving anterior teeth with midline crossing\textsuperscript{17}.

In the present case, along with the affected right lower anterior teeth, the right mandibular central incisor was missing. Also the second premolar was impacted but radiographic appearance was normal. Radiolucency was noted surrounding canine, both the enamel and the dentin layers of the crown appeared abnormal, and the pulp chamber was enlarged, the root was well formed. The erupted lower right mandibular lateral incisor shows a malformed, undercalcified crown, with enlarged pulp chamber, short root, and a periapical radiolucent lesion. Histopathologically, abnormal enamel matrix and dentin are identified. Adjacent soft tissue contains scattered islands of odontogenic epithelium and enamel-like calcifications surrounded by whorls of odontogenic mesenchyme. Calcifications in the dental follicle are similar to those found in the central odontogenic fibroma WHO type. The COF-like tumors in this case cannot be regarded as hyperplastic follicles as the associated teeth showed enamel and dentinal dysplasia. After correlating the clinical, radiographic and histopathological features, the case was diagnosed as regional odontodysplasia. These features were consistent with follicle of regional odontodysplasia and we conclude that they resemble the hyperplastic follicles of regional
Therapeutic consideration of such cases depends upon the assessment of the defect. Treatment options include extraction of the affected tooth with replacement (fixed or removable prosthesis, implants etc.) for severely malformed teeth. Exposure of the affected tooth with orthodontic traction is treatment of choice for those teeth in which the defect is not in the supporting apparatus of the tooth. Therefore, the prognosis of teeth in which the defect does not involve the supporting apparatus of the tooth is good, but severely malformed teeth are difficult to preserve.

After evaluation of the present case, the eruption of the affected tooth seemed questionable, therefore it was decided to extract the tooth and replace it with prosthesis. Exposure accompanied by orthodontic traction was decided as the treatment option for impacted premolar and the hypoplastic incisor was retained so that it could serve as abutments for restorative care. Hence, a multidisciplinary approach with consultation between pediatric, prosthodontic and orthodontic departments is necessary for each case of regional odontodysplasia. In conclusion it is necessary to discriminate histologically-similar odontogenic tumors, in particular odontogenic fibroma because of its pathogenesis and recurrence potential.

References