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Developing Complex Odontome–an Enigmatic Entity: A Case Report

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ABSTRACT

A benign mixed odontogenic lesion, with features of ameloblastic fibro odontoma previously allocated in the WHO 2005 classification, and presently removed in the subsequent WHO 2017 and 2022 classifications, shows certain changes that result in the formation of enamel and dentin, suggestive of a developing complex odontoma rather than ameloblastic fibro odontoma. It commonly affects individuals in the first and second decades of life. The present case exhibits histopathological features which show enamel and dentin formation and strands of ameloblast-like cells with an inductive process. The histopathology points to a hallmark diagnosis of a "developing" complex odontoma.

Keywords: WHO; Odontogenic; Inductive changes; Ameloblast.

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Introduction

According to the recent WHO 2022 classification, some of the mixed odontogenic lesions were removed, and only the odontoma was retained. The entities that were removed include: ameloblastic fibrodentinoma and ameloblastic fibro-odontoma. Ameloblastic fibroodontoma (AFO) is a benign, mixed odontogenic lesion [1]. The WHO has defined AFO as a lesion similar to Ameloblastic fibroma (AF) but also shows specific inductive changes that lead to the formation of dentin and enamel. The ambiguity in the earlier literature stated this entity as Ameloblastic odontoma. Later in the year 1967, Hooker et al. drew a clear distinction between these two entities, stating that AFO should be considered as a separate entity. However, according to the 2017 classification provided by the WHO, it was not considered a separate entity but rather a spectrum of developing odontoma. Once dentin and enamel formation occur, it usually represents the first stage in maturation and is more compatible with a developing odontoma [2].

AFO since it exhibits characteristics of both complex odontomas (CO) and AF. Hooker took into account 26 AFO cases. He said that the age range was 0.5–39 years old, with 11.5 years old being the mean3. Additionally, he discovered that the maxilla and mandible were equally predisposed in the patients he examined. AFO exhibits features of both AF and CO, hence the name Ameloblastic fibro-odontoma. Later, Slootweg reviewed another 50 cases, out of which 28 were male and 22 were female. In this series, Slootweg had observed that the mean age was 8.1 years, with 62% of the patients below 10 years. Slootweg also noted that of the total number of lesions in the jaw, 54% were found in the posterior region, with 38% occurring in the maxilla and 62% in the mandible. Both have stated that an unerupted tooth is frequently seen on radiographs and that it frequently occurs as a central variety in conjunction with a swelling. It is sometimes referred to as "TUMOR OF CHILDHOOD and ADOLESCENCE"3 since it primarily affects people in their first two decades of life. The information about a rare entity in a male patient from the South Indian community is presented here.

Case Report

A 28-year-old male patient reported to us in November 2023 with a chief complaint of swelling in the lower right back tooth region for 3 months. The patient reports a history of gradual onset, initially minor in size, and swelling that has now reached its present size (Figure 1). On intraoral examination, a localized diffuse swelling is seen on the alveolar ridge of the lower right mandibular region, concerning 44,45. The patient also reported a history of avulsed teeth in that region, and adjacent teeth showed mobility. The swelling was extending from the 44,45 region, obliterating the buccal vestibule, and the skin over the swelling was normal in comparison to the adjacent area. The lesion measured about 1 cm x 2cm mesio-distally. An orthopantomogram (OPG) was advised and it revealed a well-defined radiolucency without any cortication. The radiolucency showed some mixed radiolucent to radio-dense opacities. The opacities are not as dense as a completely formed odontoma, indicating its formation stage.

The radiograph also showed root resorption in tooth 46 and divergence of the root in tooth 44 (Figure 2). The lesion was incised, and the samples were sent to the Department of Oral Pathology & Microbiology. Upon macroscopic examination, the received specimen was white in color, soft in consistency, and the entire tissue was fixed in 10% neutral buffered formalin for processing (Figure 3). Later, the embedded tissue was sectioned and stained with haematoxylin and eosin, then viewed under a microscope. On microscopic examination, the soft tissue section exhibited connective tissue with dense collagen fibre bundles and fibrocytes. (Figure 4.) The connective tissue also exhibited ameloblastic islands, characterized by tall columnar cells and central stellate reticulum-like cells. These islands were surrounded by an eosinophilic area representative of the induction phenomenon (Figure 5). The tissue also exhibited primitive odontogenic epithelial cells in the form of cords, strands, and rosettes, and a few inflammatory cells (Figure 7). Later, an excised tissue sample of the same patient was received. The excised samples were three in number, white in colour, and of hard consistency. The tissue specimens were labelled as A (buccal), B (lingual), and C (teeth) (Figure 7). A measured about 2 x 1.9 x 1.6cm, B measured about 0.8 x 0.6 x 0.4cm and C is the extracted mesial teeth (canine) which showed mobility. A is cut into three parts labelled as A1, A2, and A3. Hard tissue is taken for slow decalcification in 10% formic acid and 5% nitric acid for four alternate days. After processing, the tissue specimens are embedded and sectioned. Microscopical examination revealed odontogenic islands with tall columnar cells and central stellate reticulum-like cells. Surrounding the islands, there are empty spaces formed due to the retraction of the formed dental hard tissues,

like the dentin and enamel. (Figure 8). And the section showed thin odontogenic cells arranged in the form of cords or strands having drumstick appearance, which is characteristic of ameloblastic fibro-odontoma.4 The section also showed bone with osteocytes within the lacunae (Figure 9) and extravasated red blood cells. The patient was followed up for one year and did not show any recurrence, indicating the lesion was well excised and did not show any difficulty during its removal. Based on the overall clinical, radiographical, and their correlation with the histopathological features, the final diagnosis was suggestive of "Developing Complex Odontome" as ameloblastic fibro-odontoma has been excluded in the recent classification.

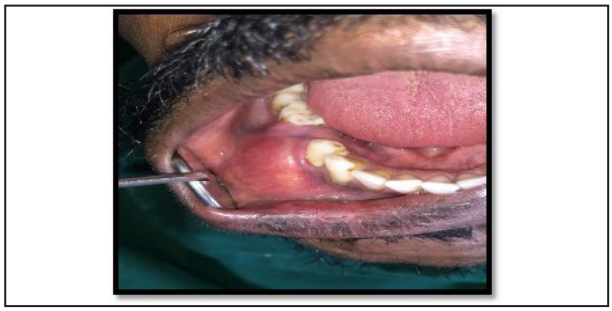

Figure 1. Diffuse swelling is seen on the 44, 45 region, obliterating the buccal vestibule. The skin over the swelling showed no changes compared to the adjacent area.

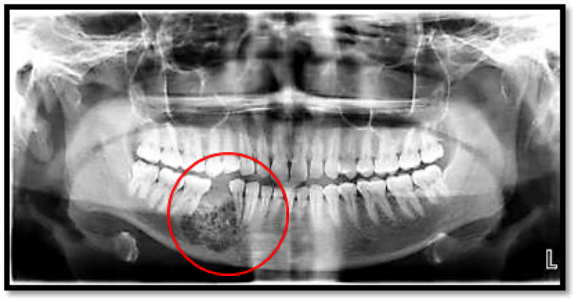

Figure 2. Well-defined radiolucency with radiopacities within the lesion. Teeth distal to the lesion showed root resorption, and the mesial teeth showed a root divergence.

Figure 3. Macroscopic examination of the incised tissue specimen reveals a white colour, soft consistency, and measures approximately 1cm x 0.6cm x 0.6cm in size.

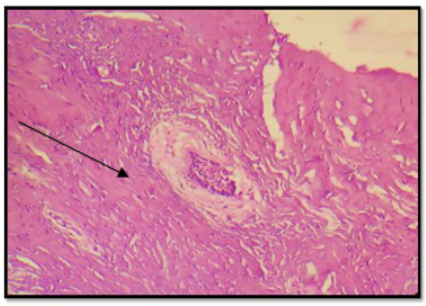

Figure 4. (10x) Histopathological features exhibit a follicle with an eosinophilic area surrounding it and dense collagen fiber bundles with fibrocytes.

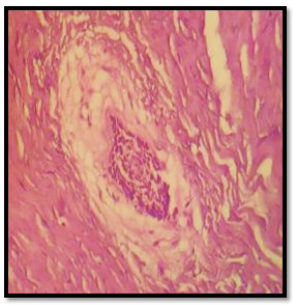

Figure 5. (20x) The section reveals ameloblastic follicles with stellate reticulum-like cells within. The follicle is surrounded by an eosinophilic material, which is believed to be the induction process.

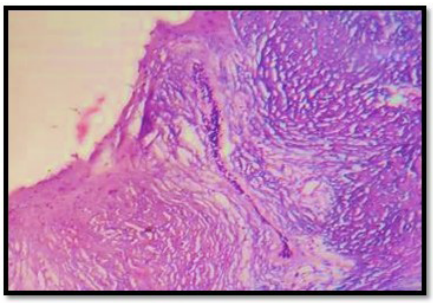

Figure 6. (20x) The connective tissue also exhibits primitive odontogenic epithelial cells arranged in the form of cords and strands.

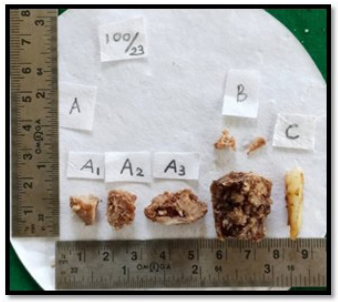

Figure 7. Macroscopic examination of an excised tissue specimen reveals it to be white in colour and hard in consistency.

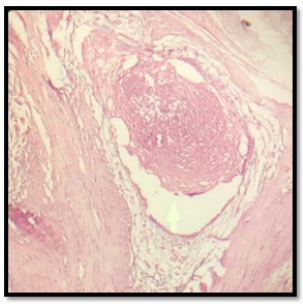

Figure 8. (20x) Ameloblastic epithelial islands showing empty retraction spaces of the enamel and dentin.

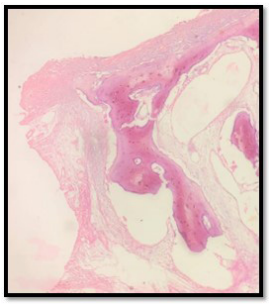

Figure 9. (20x) showing bone with osteocytes within lacunae.

Discussion

The histopathological features of ameloblastic fibro-odontoma are similar to those of ameloblastic fibroma. The features reveal an irregularly arranged enamel, dentinoid, cementum and pulp-like ectomesenchymal tissue. It also exhibits cords, strands, or rosettes of ameloblast-like cells arranged in the form of follicles. The follicles are surrounded by enamel and dentin. The dentin may either be present as dentinoid structure or as a tubular dentin. The ectomesenchyme gradually decreases as the ameloblasts begin to form enamel and dentin. These odontogenic nests can also contain melanin deposition and large aggregates of melanophages [3]. We have included a differential diagnosis of Desmoplastic ameloblastoma, ameloblastic fibroma, and an odontoma.

Desmoplastic ameloblastoma occurs with a mean age of 42.8 years, with the maxilla being predominant over the mandible. Clinically, the lesion is not invasive nor aggressive. The collagenous stroma does not compress the ameloblastic islands. The ameloblastic islands in the section did not fulfill the Vickers Gorlin criteria, despite showing abundant collagen and inductive changes. The radiographic features were also unsatisfactory, as there are well-defined borders in the present case, whereas according to the literature, only 7% of the cases of desmoplastic ameloblastoma showed well-defined borders [3]. The ameloblastic fibroma was ruled out because the histology did not show any primitive ameloblastic epithelium, and the primary ameloblastic differentiation was absent. The prominence is a significant finding in the typical ameloblastic fibroma [3]. Furthermore, the radiograph did not show dense radiopaque entities in the centre of the lesion, typically seen in odontome [4]. The macroscopic features did not reveal any tooth-like structures as well [3]. As the process of diagnosing begins right from the grossing, we have taken into account the clinical considerations, macroscopic features, radiographic details, and finally correlating with its histopathological features to come to a diagnosis of a "developing" complex odontoma. The exact histogenesis of mixed odontogenic lesions is unknown. Cahn and Blum et al stated that ameloblastic fibroma transforms into ameloblastic fibroodontoma and finally into complex odontoma.

For AFO, surgical curettage is the recommended treatment for this lesion, along with the removal of any unerupted teeth. The approach is conservative, as it does not invade the bone and shows a low tendency to recur. There have been cases of recurrence reported in the pediatric age group, according to Tsagaris et al [7]. But for a complex odontoma, the treatment of choice is excisional surgery [8]. The prognosis for developing odontoma is good compared to other AFs, Such as Ameloblastic fibrosarcoma [9]. It is relatively rare, with the prevalence among oral biopsies being about 1% 10 and its frequency among odontogenic tumours being reported at 1% to 3% as stated in the literature [11].

Conclusion

Based on its histopathological features, a developing odontoma is distinguished from other odontogenic tumors due to its characteristic features. In the upcoming classifications, these entities will be incorporated again, as there is an overlap of features and differences in treatment modalities. More case reports should be discussed to aid in classifying such lesions.

Conflict of Interest

There is no conflict of interest to declare.

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