Case Report

Adenomatoid Odontogenic Tumor: An Uncommon Location

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Abstract: *Adenomatoid odontogenic tumor is an uncommon tumor of odontogenic origin. It is usually located in the anterior region of the maxilla associated with impacted canines. The purpose of this paper was to present a case of adenomatoid odontogenic tumor in a 14-year-old girl with unusual clinical manifestations involving an impacted mandibular lateral incisor as well as its treatment follow-up. (Pediatr Dent 2011;33:437-9) Received March 2, 2010 / Last Revision May 6, 2010 / Accepted May 7, 2010*

KEYWORDS: TUMOR, ADENOMATOID ODONTOGENIC TUMOR, MANDIBLE, ENUCLEATION

The tumor that meets today's diagnostic criteria of an adenomatoid odontogenic tumor (**AOT**) has been known for more than 10 decades. AOT is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.[1] Unal et al., provided a list containing all nomenclatures for AOT reported in the literature, including adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma admantinum, and teratomatous odontoma.[2] Philipsen and Birn introduced the term AOT, to be adopted by the World Health Organization in their "Histologic Typing of Odontogenic Tumors, Jaw Cysts, and Allied Lesions", and it is now the generally accepted nomenclature.[3]

The purpose of this paper was to describe a case of a girl diagnosed with adenomatoid odontogenic tumor in the anterior mandible that was associated with an impacted left lateral incisor as well as its management.

Case report

A medically healthy, 14-year-old girl presented to the Department of Pediatric Dentistry, SDM School of Dental Sciences, Dharwad, Karnataka, India. Her dental history revealed a painful swelling experienced over the past 12 months at the chin area. It was approximately pea-sized initially and nontender but gradually increased to the present size of 4 x 6 cm. An extraoral examination revealed a solitary swelling extending laterally approximately 2 cm below the angle of the mouth on either side of the chin and inferiorly approximately 3.5 cm from the mandible's lower border. This resulted in facial asymmetry (Figure 1).

On palpation, the swelling was bony hard in consistency, immobile, and tender. The swelling surface was smooth with well-defined edges. An intraoral examination revealed a well-defined swelling extending from the permanent mandibular right second premolar to the permanent mandibular left second premolar in the labial vestibule and from the permanent

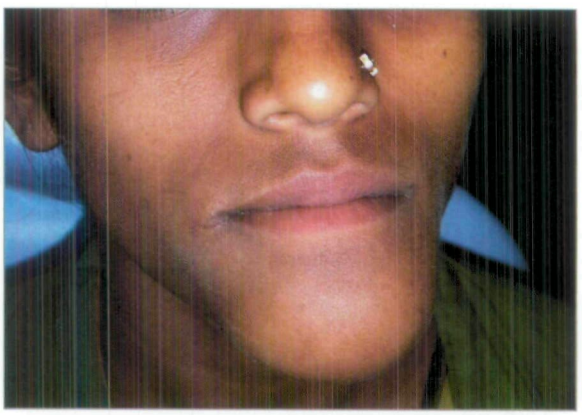

Figure 1. Extraoral photograph taken before treatment.

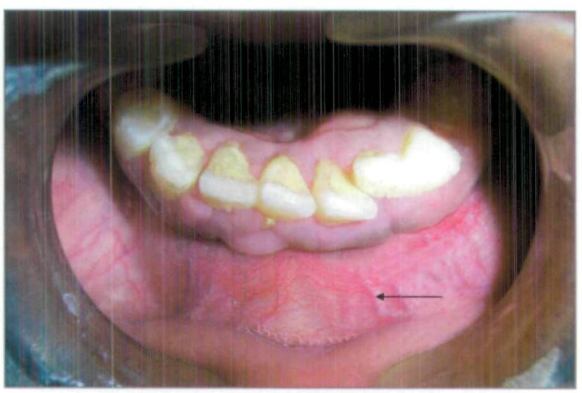

Figure 2. Intraoral photograph taken before treatment.

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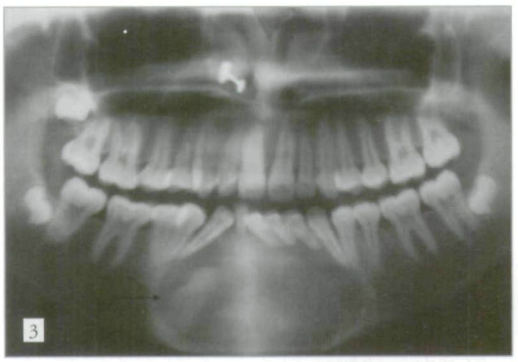

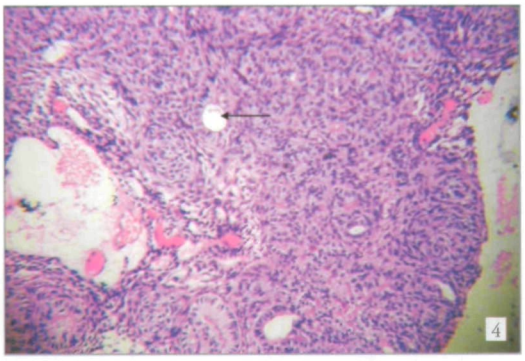

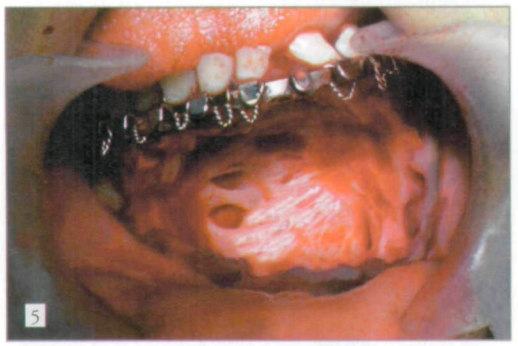

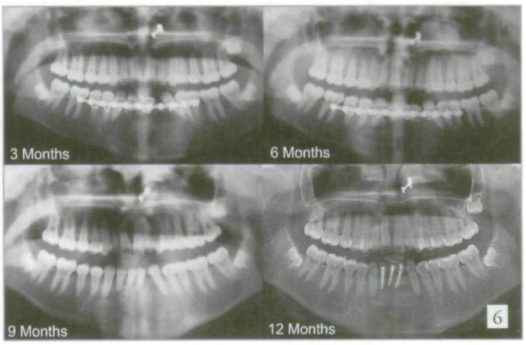

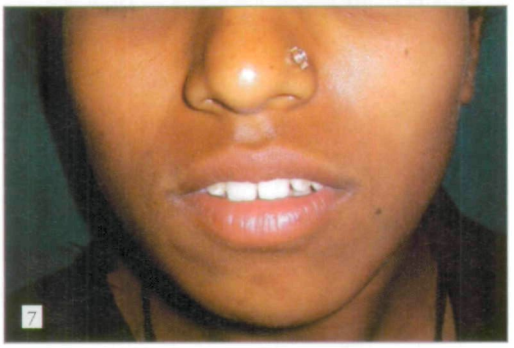

Figure 3. A panoramic radiograph taken before treatment.

Figure 4. Characteristic duct-like structures lined by a single row of columnar epithelial cells, with nuclei polarized away from the empty central lumen (40X).

Figure 5. Resection of the tumor.

Figure 6. Series of orthopantomographs showing satisfactory healing at 3-, 6-, 9-, and 12-month intervals.

Figure 7. Extraoral photograph taken after treatment.

mandibular left second premolar to the permanent mandibular right lateral incisor in the lingual vestibule. There was a missing permanent mandibular left lateral incisor and an over-retained primary mandibular left lateral incisor (Figure 2). All mandibular anterior teeth showed grade II mobility. All teeth involved showed a positive response to electric pulp testing. Based on the clinical features, a provisional diagnosis of dentigerous cyst, AOT, and ameloblastoma was made.

An orthopantomograph (**OPG**) revealed a well-defined, unilocular radiolucency associated with an impacted permanent mandibular left lateral incisor. There was root resorption in relation to the retained primary mandibular left lateral incisor. Root displacement was seen in the mandibular anterior teeth (Figure 3).

An incisional lesion biopsy of approximately 1 cm x 1 cm was taken for histopathology, which revealed a well-developed, fibrous connective tissue capsule and duct-like spaces lined by a single row of low columnar epithelial cells, the nuclei of which were polarized away from the luminal surface (Figure 4), which is distinctive of AOT.

Based on the clinical, radiographic, and histopathological findings, a confirming diagnosis of AOT was made. The decision to enucleate the lesion under general anesthesia, the complete procedure, and any probable complications were explained to the patient. Arch bar fixation was done to immobilize the mandibular anterior teeth. The tumor's borders were identified, dissection along the plane was performed, and the tumor was resected (Figure 5). The postoperative course was uneventful. The specimen was sent to the histopathology laboratory to confirm the diagnosis. The patient was seen every 3 months for 1 year, during which endodontic treatment was performed for the mandibular incisors due to the extent of the surgery. A series of OPGs were taken, which showed healing without recurrence (Figures 6 and 7).

Discussion

According to Sonu Nigam et al.,[4] an AOT is a rare tumor that comprises only 0.1% of tumors and cysts of the jaw and 3% of all odontogenic tumors. The mean age of occurrence of AOT is 13.2-years-old (range=3-28 years old); the female to male ratio is 2.3:1; and tumors are predominantly found in the

maxilla (maxilla to mandible ratio=2.6:1).[5] Interesting differences appear between Asians and non-Asians (ie, Asian AOT cases show a female to male ratio of 2.3:1 vs 1.4:1 for non-Asian cases).[3] The tumor is usually associated with unerupted teeth—frequently canines. All 4 canines account for 59%, and maxillary canines account for 40% of cases.[3] The site distribution according to teeth involved in descending order are canines, maxillary incisors, premolars, and molars.[6]

There are 3 clinicopathologic variants of AOT—namely intraosseous follicular, intraosseous extrafollicular, and peripheral, all with identical histologies. The follicular type is a central intraosseous lesion associated with an impacted tooth, while extrafollicular intraosseous AOT has no relation with an unerupted tooth. It is often located between, above, or superimposed upon the roots of adjacent erupted teeth. The peripheral variant appears as a gingival fibroma or epulis attached to the labial gingiva.[3] The follicular and extrafollicular variants account for 96% of all AOT cases.

Radiographically, the intraosseous follicular AOT is seen as a well-demarcated, unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth, thus mimicking a dentigerous or follicular cyst.[3] It should be differentiated from dentigerous cyst, which most frequently occurs as a pericoronal radiolucency in the jaws. Dentigerous cyst encloses only the coronal portion of the impacted tooth, whereas AOT shows radiolucency usually surrounding both the coronal and radicular aspects of the involved tooth.[4] In approximately two thirds of the intrabony variants, the radiolucency shows discrete foci having a flocculent pattern of scattered radiopacities.

AOT is usually surrounded by a well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space, or as numerous small cystic spaces. The characteristic duct-like structures are lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. The lumen may be empty or contain amorphous eosinophilic material.[3] Dystrophic calcification in varying amounts and in different forms is encountered, scattered among epithelial masses or in the stroma, in most AOT structures.

According to Philipsen et al., immunohistochemical and ultrastructural findings have shown that eosinophilic deposits probably represent some form of enamel matrix. AOT's histogenesis is still uncertain, although recent findings strongly indicate that AOT is derived from a complex system of dental laminae or its remnants.[7]

Given AOT's benign behavior, slow growth, clear delimitation, and low tendency to recur, the treatment of choice is enucleation and simple curettage. In exceptional cases of large tumors or, when there is risk of bone fracture, partial en bloc resection of the anterior mandible or maxilla has been indicated.[8]

In the present AOT case, the age, sex, and histopathology are in accordance with the literature. However, the site, side and the tooth involved is a rare occurrence. AOT is more common in the maxilla, while in our case the anterior mandible was involved. Canines are the teeth most often involved, while in this case a lateral incisor was impacted. Swasdison[9] showed that in a Thai population, 67 AOT cases were reported, none of which involved a mandibular lateral incisor. In a survey by Toida,[10] only 7 of 98 cases involved mandibular lateral incisors.

Odontogenic tumors are not uncommon causes of morbidity in children. As pediatric dentists, we should be aware of such rare varieties of the tumor so that early diagnosis, proper investigations, and prompt treatment are made available.

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