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Case Report

Mural Adenomatoid Odontogenic Tumor in the Mandible - A Rare Case

Komal Khot, Pavitra A Vibhakar

Abstract

Adenomatoid odontogenic tumor is a uncommon benign hamartomatous lesion of odontogenic origin, which affects young individuals, with a female predilection and mainly occurs in the second decade. It is considered as a hamartoma because of its limited size, due to minimal growth potential, and lack of recurrence. The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm-like ameloblastoma or Adenomatoid odontogenic tumor. Such combined lesions are rare and must be carefully diagnosed by an oral pathologist so that optimum treatment needs of the patient can be met. We are here reporting a case of cyst associated with Adenomatoid odontogenic tumor in a 17 year old female patient in the mandible, which was clinically diagnosed as dentigerous cyst or unilocular ameloblastoma. This report of a cyst in the mandible associated with an Adenomatoid odontogenic tumor is extremely rare. Combined lesions of such variety are uncommon and hence the importance of grossing and histopathology in such cases should not be undermined.

Keywords: Adenomatoid Odontogenic Tumor; Adenoameloblastoma; Tooth; Impacted; Bone Neoplasms; Adamantinoma

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Introduction

Adenomatoid odontogenic tumor (AOT), an uncommon benign epithelial lesion of odontogenic origin, was first described by Dreibaldt in 1907 as a Pseudoadenoameloblastoma.[1] Harbitz in 1915 reported it as ‘‘Adamantoma’’ and, Bernier and Tiecke were the first to publish a case using the name ‘‘Adenoameloblastoma.’’ Finally the term ‘‘Adenomatoid Odontogenic Tumor’’ was proposed in 1969 by Philipsen and Birn.[2]

According to the second edition of the World Health Organization ‘‘Histological Typing of Odontogenic Tumors’’, AOT is defined as ‘‘A tumor of odontogenic epithelium with duct like structures and varying degrees of inductive change in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst.[3]

The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm-like ameloblastoma or AOT. There have been many reports of odontogenic cysts associated with odontogenic tumors.[4,5, 6] The purpose of this paper is to present a case of AOT that originated in the wall of a pre-existing cyst of the mandible, review the literature and stress that some AOTs can arise as a secondary phenomenon within the pre-existing cysts. 35.7% of all AOT variants occur in the maxilla of which 35.3% are intraosseously located. The total number of mandibular AOT cases reported is 213 in the Chinese and Japanese literature[12]. However a total of 9 cases of AOT arising from a dentigerous cyst have been reported in the available English literature[9].

Case report

A 17 year old female patient reported with a chief complaint of pain in the right back region of the lower jaw since the past 2 years. The associated symptoms included discomfort during chewing of food, swelling and pus discharge. The swelling was slow growing which had now increased to the present size. Extra orally, the swelling extended from the midline of the mandible on the right to the angle of the mandible (Fig 1) and inferiorly to the hyoid bone region. On palpation the swelling was firm with enlarged and palpable right and left submandibular lymph nodes.

Intraoral examination revealed a buccal and lingual expansion of the mandible extending from the left mandibular canine to the right ramus of the mandible. The swelling was approximately 7x15 cm in size. An over retained deciduous right mandibular canine

and its missing permanent successor was noted. The overlying mucosa was normal. On palpation the swelling was bony hard.

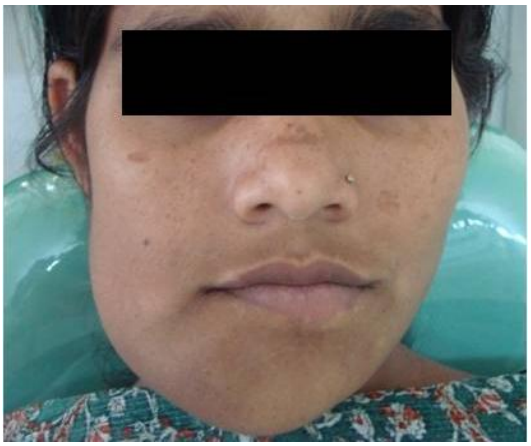

Figure 1: Extra orally the resultant disfigurement due to the intraosseous lesion can be appreciated on the right side of the lower jaw.

Orthopantamograph showed well-defined, unilocular corticated radiolucency extending from the left mandibular canine extending to the right into the ramus of the mandible showing expansion of the cortical plates. An impacted permanent right mandibular canine was noted. The involved teeth showed root resorption (Fig 2). Computerized Tomography scan shows well defined, unilocular, corticated radiolucent lesion with buccal and lingual cortical expansion crossing the midline of the mandible (Fig 3). A Clinical diagnosis of dentigerous cyst or unilocular ameloblastoma was made.

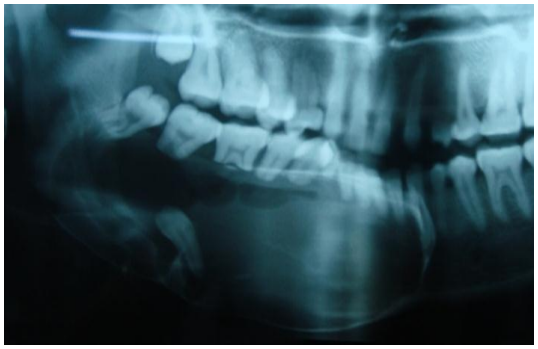

Figure 2: Orthopantamogram showing well demarcated unilocular radiolucency with corticated borders the impacted canine and resorption of roots of the involved teeth.

The patient underwent surgical curettage under general anesthesia. The excised tissue specimens were received in formalin. They were yellow to tan brown in color, irregular in shape, firm in consistency and the largest specimen showed nodular elevation in the luminal surface. The attachment of the cystic lining in the gross pathology could not be well appreciated as the tissue specimens were received separate from the impacted tooth. However, a very small part of the cystic lining attachment running over the root surface was appreciated (Fig 4).

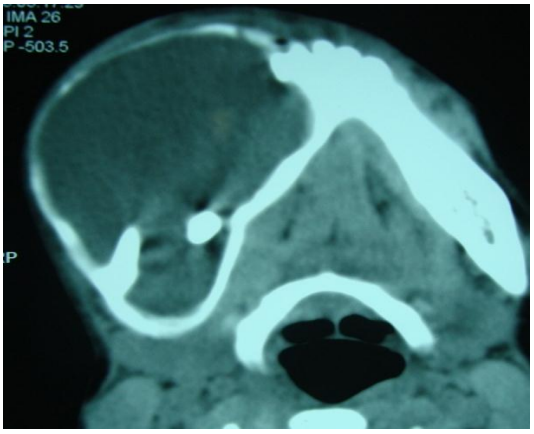

Figure 3: Computerized Tomography scan shows a well-defined, unilocular, corticated radiolucent lesion.

Histological picture showed a cystic lining of nonkeratinized stratified squamous epithelium with proliferation into the underlying connective tissue. The lesional tissue contained islands of odontogenic epithelium with formation of rosette like structures (Fig 5). The tumor islands predominantly consisted of stellate reticulum like spindle shaped cells, and tall columnar cells at places (Fig 6). Some of the rosette-like structures centrally showed clear cell changes. Connective tissue was fibro-cellular with areas of hyalinization, moderate chronic inflammatory cell infiltrate and marked vascularity. Spicules of dystrophic calcification were evident within the epithelial islands as well as the connective tissue. Histopathological diagnosis of a mural AOT in a pre-existing cyst was made.

Discussion

Adenomatoid odontogenic tumor is a slow growing lesion, constituting only 3% of all odontogenic tumors with a predilection for the anterior maxilla (ratio 2:1) relative to mandible usually associated with impacted canine, of young females in the second decade of life.[7] In our case the lesion occurred in the posterior mandible which is unusual.

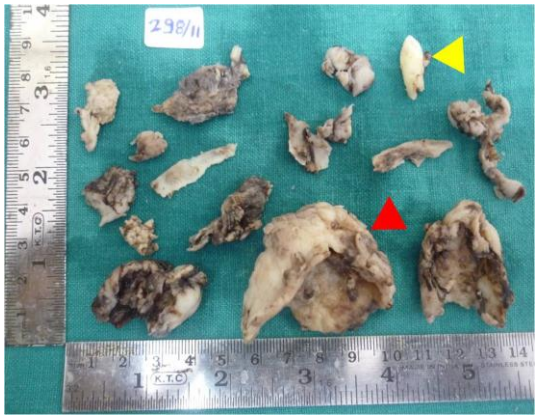

Figure 4: Macroscopic examination shows several bits of tissue including the impacted canine (yellow arrowhead) with a small bit of soft tissue attached on its root surface and the largest specimen showed the nodular growth into the cystic lumen (red arrowhead).

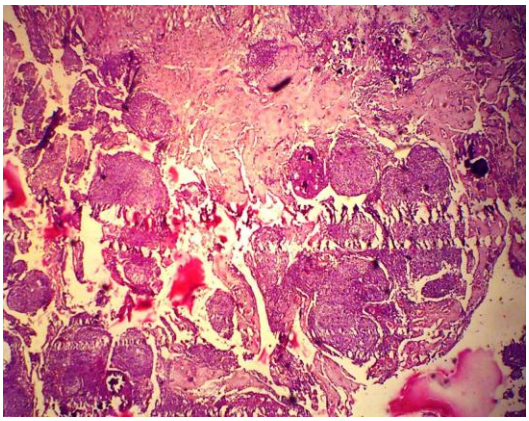

Figure 5: Photomicrograph (40X) showing odontogenic islands in the form of rosette like structures.

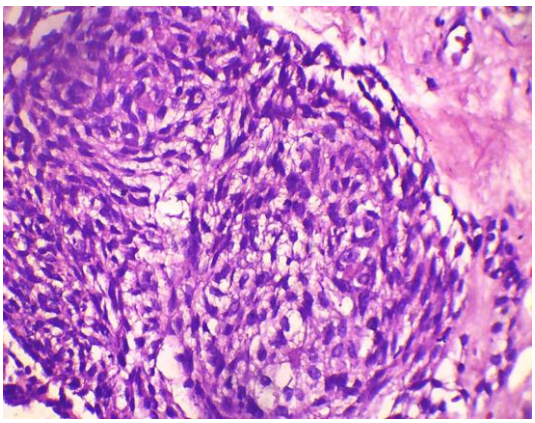

Figure 6: Photomicrograph (400X) showing Rosettes predominantly composed of spindle shaped cells and stellate reticulum like cells.

Rick et al have reviewed cases of AOT in association with dentigerous cysts and stated that although most central AOTs occur in a pericoronal relationship with an associated tooth, there is no way to be certain whether the lining of an associated cyst represents a true dentigerous cyst or a secondary cystic change within the AOT[10]. In this case, AOT and dentigerous cyst are found in the same lesion.

When we consider the etiopathogenesis of such a tumor, the origin of AOT is controversial. Some believe they originate from the odontogenic epithelium of a dentigerous cyst. Santos et al reported a case of AOT being developed in the fibrous capsule of the dentigerous cyst, while Garcia Pola et al described the proliferation of an AOT in the epithelial border of a dentigerous cyst[7,8,9]. All the cases of AOT associated dentigerous cyst reported by J. Baby John et al., Valderrama, Warter et al., Takahashi et al., Brave et al., Chen et al., Simarpreet V. S., Garcia Pola et al., have in common a unilocular radiolucency except the one reported by Tajima et al., presents as a radiopaque mass. All of these have occurred in the maxilla and two associated are associated with the maxillary sinus. Of these, all except two occur in the posterior region of the jaw and all are associated with impacted teeth of which the most common tooth involved is the canine. All but three have occurred in men. The age of all cases reviewed by Simarpreet VS. lies between 8-25 years and the case reported by J. Baby John was aged 39[7,11].

Our case too presents as a unilocular radiolucency in the anterior region of the jaw and associated with impacted canine and the patient's age 14 also corresponds to the above mentioned cases. However, our patient is a female and most of the above lesions have occurred in male and it completely differs as none of the above have manifested in the mandible, making this case truly rare.

The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm-like ameloblastoma or AOT.[4,5,6] A few cases have been observed of early adenomatoid odontogenic tumour that have appeared radiologically and in the gross specimen as dentigerous cysts. The presence of adenomatoid odontogenic tumour may be suspected in the gross specimen by the observation of small white

or yellow nodules on the luminal surfaces of the cyst walls[10]. Few reports have also shown AOT in conjunction to dentigerous cyst. However, all cases were limited to the maxilla and at maximum involved the sinus thus making our mandibular lesion a rare entity.[7,11]

Rick et al have reported AOT to occur with many types of cysts and neoplasms including dentigerous cyst, calcifying odontogenic cyst, odontoma, and ameloblastoma etc. In relation with a dentigerous cyst the AOT may demonstrate, grossly and microscopically, one or more associated cystic cavities. Some these cysts are lined by nonkeratinized stratified squamous epithelium which is similar to the lining of the dentigerous cyst or lined by less structured membrane that may demonstrate bud like extensions into the connective tissue[2]. In our case a moderate amount of the inflammatory component was evident in the sections, which could cause the cystic epithelium to lose its characteristic features and hence restrict the typing to an odontogenic cyst alone.

Odontogenesis is a complex process wherein neoplastic or hamartomatous lesions can occur at any stage of odontogenesis. The secondary development of an ameloblastic proliferation, whether hyperplastic or neoplastic is well known, but remains controversial. In the present case, the multifocal cellular proliferation had the structure of an AOT, while its mural development in a dentigerous cyst is not uncommon.[11]

Whether origin of the follicular variant occurs before or after cystic expansion has taken place is open to conjecture. If it occurs after cystic expansion, then this effectively means origin from a dentigerous cyst, and several such case reports have been published. If it occurs before cystic expansion, then the tumor tissue will fill the follicular space and the AOT will present as a solid tumor. It is reasonable to assume that, given enough time, even those originating from a cyst may grow and fill the lumen completely. It cannot be ruled out that the dentigerous cyst with an impacted canine developed first followed by development of AOT in the cyst wall.[11]

There is an uncertainty whether the lining of an associated cyst represents a true dentigerous cyst, cystic change within an AOT or may represent a distinct entity. Also, it is unclear whether this entity has a more aggressive potential. In such combined lesions it is unlikely that minor foci will have any significant effect on the clinical behaviour of a different associated odontogenic tumor that makes up the majority of the lesion. Until proven otherwise it seems to be safe to treat an affected patient in accordance with the majority lesion or the worst acting lesion if lesions of disparate behaviour are present.[2]

The AOT and dentigerous cyst are both benign, encapsulated lesions and conservative surgical enucleation or curettage is the treatment of choice. The prognosis for a dentigerous cyst is good and recurrences are very rare after complete removal of the lesion.[11] However, in Japanese literature a recurrent case of an AOT with intracranial extension has been reported but has been argued by Rick et al to be an ameloblastoma.[2] Hence, enucleation or curettage should suffice.

Conclusion

This report of a cyst in the mandible associated with an AOT is extremely rare. We believe that the present case represents an odontogenic cyst with neoplastic development, containing both epithelial and mesenchymal components. Meticulous histopathological evaluation is thus required of all enucleated cysts, which could contribute to the diagnosis of similar cases as reported in the present study.

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