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RESEARCH ARTICLE

A CASE SERIES OF 2/3 RDTUMOR -ADENOMATOID ODONTOGENIC TUMOR

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ABSTRACT

Adenomatoidodontogenic tumor (AOT) is a relatively rare, benign, hamartomatous, and cystic odontogenic neoplasm that was first described more than a century ago. The lesion still continues to intrigue experts with its varied histomorphology and controversies regarding its development. The present article describes threecases of AOT with an unusual locationin maxillary sinus and the other two cases associated with an impacted canine.The rarity of AOT, association ofthis lesion with regards to maxillary sinus in one ofthe cases, the exaggerated size at presentation, uneventful healing of the bony defect makes this cases unique.

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905. AOT is an uncommon, hamartomatous, benign, epithelial lesion of odontogenic origin which was first described by Driebaldt in 1907, as a pseudo-adenoameloblastoma. In 1969 Philipson and Birn proposed the name adenomatoid odontogenic tumor (Philipsen, 1969). Later the AOT was adopted in the initial edition of World Health Organization (WHO)'s histological typing of odontogenic tumor, jaw cysts, and allied lesion in 1971(Pindborg, 1971) and retained in the 2nd edition of WHO in 1992.Adenomatoidodontogenic tumor is also called 'two-thirds tumor,' because 2/3rd of adenomatoid tumors occur in the maxilla, 2/3rd occur in young females, two-thirds of the cases are associated with un-erupted teeth, and two-thirds of the affected teeth are canines (Marx, 2003; Philipsen, 2007). In a review of 272 AOTs by Becker *et al*., (1971) the patients' ages at the time of diagnosis ranged from 3 to 82 years (mean, 18.4 years).

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The maxilla-to-mandible ratio was 1.7:1. In 77% of the lesions, small opacities were present, and most were associated with expansion of the cortical bone (Pindborg, 1971). There are three pathologic types of AOT, intraosseous follicular, intraosseous extrafollicular, and peripheral, all of which have the same histological identity. The follicular type is a central intraosseous lesion associated with an impacted tooth, whereas intraosseous extrafollicular AOT is similar to the follicular type but has no relation with an unerupted tooth. It usually develops around or is superimposed onto adjacent teeth but in our case it was found in maxillary sinus of right side. The peripheral type usually looks like a gingival fibroma or epulis (Reichart, 1998). Radiographically, AOT is usually unilocular, although a few multilocular cases have been reported. In addition to AOT, the differential diagnosis should include a dentigerous cyst. Radiographically, the pericoronal radiolucency of a dentigerous cyst occurs most frequently in the jaws, and does not extend over the cement-enamel-junction of the tooth. However, an AOT often envelops the crown as well as the root past the cemento-enamel-junction, which distinguishes AOTs from dentigerous cysts. AOTs have numerous, variable-shaped radiopaque foci, which also distinguish them from dentigerous cysts; 78% of AOTs have these foci.

Tumor expansion causes displacement of the adjacent teeth, and tooth displacement is more common than root resorption (Philipsen, 1991).

CASE REPORT

CASE 1: A 29 year-old female patient referred to our department from endodontist due to radiolucent lesion seen with respect to 14 and 15. Patient gives history of swelling which had gradually increased in size and slight pain associated with the involved teeth. Extra-oral, physical examination revealed a single diffuse swelling in the right anterior maxillary region measuring about 3 cm × 4 cmin size. On palpation, the swelling was hard and tender. On intra-oral examination, a single, well-circumscribed swelling with a smooth surface was present in the buccal aspect with respect to 53, 14 and 15region. CBCT was adviced to know the exact location ofthe suspected lesion.

CBCT revealed well-defined cystic lesions extending from 53, 14 and 15 and vertically impacted 13 (Fig 1).Treatment planned was enucleationand chemical cauterization ofthe cyst with extraction of deciduous canine and retaining the permanent canine, to bring in occlusion by orthodontic treatment. The procedure was performed under LA. The cyst was removed in to with the cystic lining in situ (Fig 2). Over retained deciduous canine was extracted. Then the cavity was irrigated thoroughly using betadine and saline solution. Closure was done using 3.0 silk.

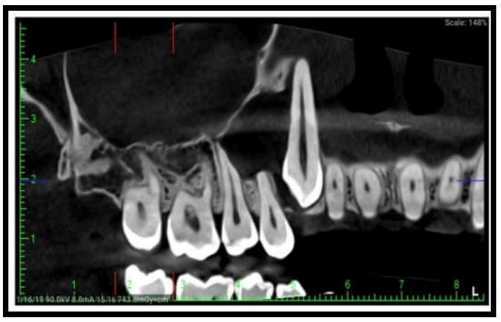

Fig 1. Ct scan showing the radiolucent cystic lesion

CASE 2

The patient, 22-year-old male was referred to the dental surgeon with the chief complaint of swelling in the right side of maxilla. The lesion was asymptomatic and clinical examination revealed facial asymmetry. Intraorally, expansion of the vestibular on buccal side of the maxilla was observed, covered with normal oral mucosa and without any signs of inflammation. Radiographically a unilocular and well-defined radiolucency, in the left side of the maxilla was noted, with 6 months of evolution.

OPG showed hypodense lesion in relation with tooth 21–24 and impacted canine (Figs. 3). The lesion was completely enucleated (Fig 4). According to the clinical, radiographic and microscopic features, the final diagnosis was of an adenomatoid odontogenic tumor. During the 6 months follow-up period, there were no signs of recurrence, and new formed bone around the tooth region was observed.

CASE 3

A 62-years-old female reported to the maxillofacial surgery department with the complaint of a painless mass in the right buccal area of the maxilla. Furthermore, the patient was edentulous since 3 years.

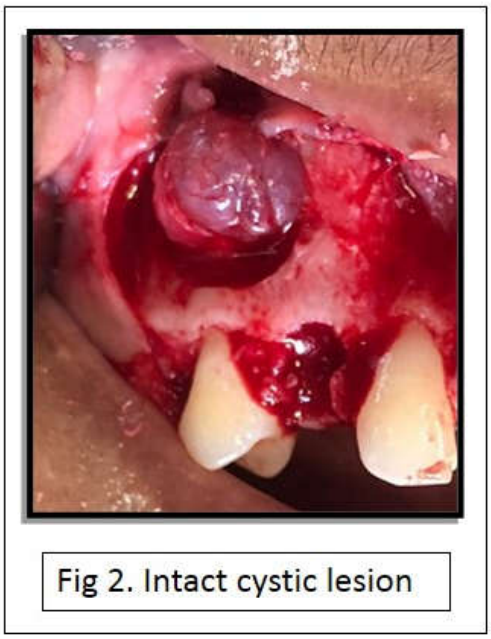

Fig 2. Intact cystic lesion

On extraoral examination, there was a swelling on the right side of the face causing obvious asymmetry with obliteration of nasolabial sulcus. Intraorally, there was an expansion in all dimensions measuring about 5 cm × 5 cm in anteroposterior and superior inferior direction. On palpation, the swelling was hard, nonlobulated; nontender, not fixed to the overlying skin and local temperature was not raised (Fig 4). . The CT scan revealed (Fig 5) solid mass, round in shape with regular margin. Yellow fluid on aspiration gave the provisional diagnosis of okc but histopathological diagnosis revealed the final diagnosis of AOT. Patient was planned for surgical removal of the cystic mass under GA. An adequate window was created and the tumor mass was enucleated along with the sinus lining (Fig 6). The lesion was partially solid with partial cystic degeneration, and a gritty sensation could be elicited on examination. The remaining cavity was found to be clean without any tissue tags after chemical and mechanical curettage. The wound was sutured with 3–0 vicryl. Patient was followed for next 4 months which showed uneventful healing. Histopathology revealed cuboidal to columnar cells arranged in the form ofnests and rosettes. Solid areas, duct-like pattern, whorled arrangement of cells, and tubular appearance is evident. Convoluted structures were noted and at the periphery of the lesion tumor cells are arranged in a strand-like configuration. Few cells were also arranged in a plexiform pattern and cribriform areas are also seen. Latticework pattern is seen closer to the connective tissue capsule and foci ofdense extravasated red blood cells were also seen in few areas.

DISCUSSION

AOT usually occurs within the tooth bearing areas ofjaws and often found in association with impacted teeth. The origin of

AOT is controversial, but many author believe in odontogenic source. AOT has cytological features similar to various components of enamel organ, dental lamina, reduced enamel epithelium, and its remnants (Sato, 2004). It is a slow growing lesion, constituting about 3% of all odontogenic tumors followed by odontoma, periapicalcemental dysplasia (Cementoma), myxoma, and ameloblastoma (Garg, 2009).

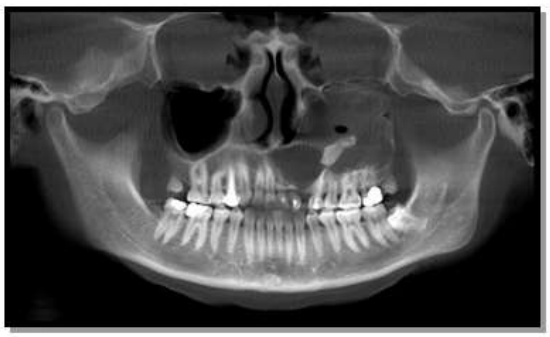

Fig 3. OPG showing cystic lesion with impacted canine

Fig 4. Intraoperative removal cystic lesion

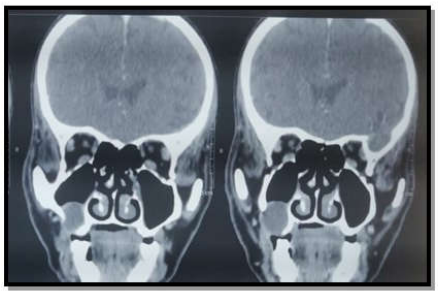

Fig 5 . Ct scan coronal view showing cystic lesion on right side.

Most ofthe cases reported in the literature were in the maxilla affecting the anterior segment and were associated with the canine tooth (Rick, 2004) The origin of AOT is believed to be from an odontogenic source; the cytologic features are similar to those of the enamel organ, dental lamina, reduced enamel epithelium, and / or their remnants (Rajendran, 2009). AOT shows centrifugal expansion (uniform expansion in all directions). It has been hypothesized that at an early stage AOT may expand the cortical plates, which within the cancellous bone spread linearly and then later may affect the cortical plates by expansion / resorption (Sato *et al*., 2004) Bicortical expansion was seen in all our patients.AOT can occur both intraosseously and extraosseously (Rajendran, 2009).

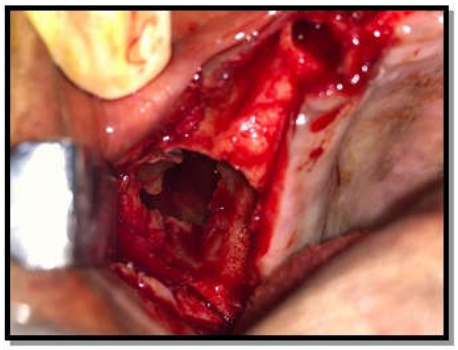

Fig 6. Enucleation of cystic lesion

All the cases reported by us were of the intraosseous type. Intraosseous AOTs are characterised by a well-defined unilocular radiolucency surrounding the crown, which is often part ofthe root ofthe unerupted tooth, and follows a follicular pattern. Intraosseous type accounts for about 73% ofall AOTs. The extrafollicular variant accounts for about 24% ofall AOTs and presents as a unilocular radiolucency found between, above, or superimposed on the roots of erupted teeth (Garg, 2009). All our cases were of the intraosseous type, of which the first and second cases represents the follicular variant and the third case, the extrafollicular variant.

However, radiological findings of AOT simulate many other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts or tumor, ameloblastoma, keratocysticodontogenic tumor or periapical disease (Bernier, 1959). The above-mentioned lesions along with the nasiolabial cyst, nasopalatine duct cyst, and odontogenickeratocyst can be considered in the differential diagnosis of lesions occurring in the anterior maxilla. Our first and second cases resembled a dentigerous cyst and the third one, an odontogenickeratocyst.
The histological features of AOT show a tumor of the odontogenic epithelium, with duct-like structures, and with varying degrees of inductive changes in the connective tissue (Philipsen, 1969). The most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures, with minimal stromal connective tissue (Pindborg, 1971).

The tumor may contain pools of amyloid-like material and globular masses of calcified material (Philipsen, 1969) Our case is consistent with the common histological features that are reported in the literature. The origin of the follicular variant can occur before or after cystic expansion (Cystic expansion in the jaw bone refers to the nature of expansion of the cyst through the buccal and lingual/palatal cortical plates). If it occurred after cystic expansion, then it effectively meant that the origin was from a dentigerous cyst and several such cases have been reported (Rick, 2004;

Reichart, 1998) If it occurred before cystic expansion, then the tumor tissue would fill the follicular space and the AOT would present as a solid tumor (Reichart, 1998). In case 1, the cystic lining was present, with deeper areas showing tumor islands, suggesting that the tumor could have occurred after cystic expansion.

Conclusion

The cases discussed emphasize the importance of recognizing neoplasms arising in the odontogenic tissues. AOT has unique clinical, radiographic, and histopathological features. However, the clinical and radiographic features may often present similarity to those of an odontogenic cyst. Persistence of deciduous teeth for a longer duration and unerupted succeeding permanent teeth, when associated with a swelling, always need to be investigated for odontogenic lesions. The term adonomatoidodontogenic cyst as suggested by Marx and Stern is controversial. But in our case presented, the presence ofunilocular cystic lesion, fluid on aspiration, in third case and cystic cavity on transection has to some extent support the terminology adenomatoidodontogenic cyst (AOC) as termed by Marx and Stern (Marx, 2003)

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