Adenomatoid Odontogenic Tumor of Maxillary Sinus - A Diagnostic Dilemma: Case Report and Brief Literature Review

Deepak Passi¹, Sarang Sharma²*, Shubha Ranjan Dutta³ and Dhirendra Srivastava⁴

¹ESIC Dental College and Hospital, Rohini-85, Delhi, India; ²Department of Conservative Dentistry and Endodontics, ESIC Dental College and Hospital, Rohini-89, Delhi, India; ³Department of Oral and Maxillofacial Surgery, MB Kedia Dental College, Birgunj, Nepal and ⁴Department of Oral Surgery, ESIC Dental College and Hospital, Sector 15, Rohini-89, Delhi, India

Abstract: Adenomatoid odontogenic tumor (AOT), constituting approximately 3-7% of all odontogenic tumors is described as a rare benign epithelial tumor of odontogenic origin. It is composed of odontogenic epithelium arranged in a variety of histoarchitectural presentations and closely resembles an ameloblastoma. AOT is mostly asymptomatic and is usually associated with impacted teeth in maxillary anterior region. We present a case of intraosseous follicular AOT existing in the maxillary sinus of a 14-year-old female child involving lateral incisor and canine impacted towards the orbital floor. The cystic tumor filled maxillary sinus and was removed along with the retained tooth. At one year follow up, healing was uneventful and no local recurrence was seen.

Keywords: Adenomatoid odontogenic tumor, Dentigerous cyst, Maxillary sinus, Odontogenic tumour, Enucleation.

1. INTRODUCTION

Adenomatoid odontogenic tumor (AOT), first described by Steensland in 1905, is reported as a relatively infrequent distinct odontogenic neoplasm [1]. Earlier, it has been described in literature by different names like adamantinoma, adenoameloblastoma, epithelioma adamantinum, ameloblastic adenomatoid tumor, or teratomatous odontoma [2]. Philipsen and Birn in 1969 coined the term AOT, emphasizing that it was not a variant of ameloblastoma rather was a separate entity [3]. This term was included in the first classification of odontogenic tumors by WHO in 1971. Pindborg classified it as an odontogenic epithelial tumor with duct like structures, presenting inductive effect on the connective tissue [4].

AOT is also sometimes referred to as the “Two Third’s tumor” because of its reported presence in maxilla in about 2/3rd cases, about 2/3rd of its occurrence is seen in young females, 2/3rd cases are associated with impacted teeth and 2/3rd of the affected teeth are canines [5]. The tumor most commonly is diagnosed in the second decade of life, mostly during teenage years (13 –19 yrs). Women are affected about twice as often as compared to men (Female: Male ratio is 2.3:1). It is more commonly located in the maxilla than mandible (2.6:1) and anterior portions of the jaw are much more affected than posterior portions. Despite the higher incidence seen in maxilla, AOT of maxillary antrum is extremely rare.

AOT exists in 3 clinico-topographic variants (i) central variant: follicular (73%) and extrafollicular (24%) (ii) peripheral variant (3%). Histologically, all variants are remarkably similar and are composed of odontogenic epithelium embedded in connective tissue stroma. Follicular (with embedded tooth) and extrafollicular (without embedded tooth) variants are both intrabony and together account for approximately 96% of all AOTs [6]. The tumor presents as a cyst, or solid masses in some lesions may present as nodules in the wall of a cyst. Here, a case of AOT of maxillary sinus in a young girl, treated by surgical enucleation of tumor with no recurrence at 1 year follow up is presented.

2. CASE REPORT

A 14-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with chief complaint of mild swelling in left cheek and with nasal obstruction since 6 months (Figure 1). Intraoral examination revealed a firm ill defined swelling extending from the upper left central incisor tooth to the second premolar on same side (Figure 2). The swelling was bony hard and non-tender. Deciduous lateral incisor and canine were seen to be retained while their permanent counterparts were clinically missing. There was no evidence of oro-nasal or oro-antral communication. Patient was radiographically investigated with orthopentogram, which revealed a cystic lesion involving maxillary sinus retaining lateral incisor, and permanent canine impacted towards the orbital floor region (Figure 3). A differen-
tial diagnosis of dentigerous cyst, unicystic ameloblastoma and AOT was made. Diagnostic aspiration was negative. The mass was entirely enucleated along with the embedded lateral incisor (Figures 4a and 4b). The specimen was submitted for histopathological examination, which confirmed the diagnosis of AOT (Figure 5). Healing was uneventful. Patient was disease and symptom free during one-year period of follow-up control. However, the patient was lost to further follow up.

Figure 1: Figure showing swelling in the left cheek.

Figure 2: Figure showing ill-defined swelling extending from the upper left central incisor to the second premolar of same side.

3. DISCUSSION

Adenomatoid odontogenic tumor (AOT) belongs to a group of rare benign odontogenic lesions and constitutes approximately 3–7% of all odontogenic tumors [7-9]. It affects young patients and is usually associated with an impacted tooth, generally the canine. AOT involves both bone and soft tissues in anatomic configuration [10]. There are reports showing AOT affecting mandible [11] molar areas [12], maxillary sinus [13], embedded primary teeth [14], infants [15] and also individuals in eighth decade of life [16].

Figure 3: OPG revealed a cystic lesion involving maxillary sinus with retained lateral incisor and canine impacted towards orbital floor region.

Figure 4: (a) Enucleated mass (b) Space left after surgical removal of mass.
Clinical features generally focus on complaints regarding a missing tooth. The lesion usually presents as an asymptomatic swelling, which is slow growing, non invasive and is often associated with an unerupted tooth. Larger lesions are known to cause expansion of cortical plates [15], displacement of teeth [14], root resorption [12] or hypaesthesia [15]. Reports have also shown its association with dilacerated tooth and supernumerary teeth [17]. Garg et al. reported a case of unencapsulated AOT that was fast growing in nature and caused root resorption [5]. There are reports in literature that have shown AOT to involve anomalous tooth forms and acquire sizes of as large as 12 cm [16]. In one report, it was observed that the lesion existed for a period of as long as 37 years [10].

The tooth most often associated with AOT is seen to be an unerupted permanent canine. Maxillary permanent canines constitute 41.7% and all four canines constitute approximately 60.1% of AOT-associated embedded teeth [14]. However, in our case lateral incisor was involved in the lesion and canine was impacted/displaced superiorly towards the orbital floor.

Histologically, AOT mostly is surrounded by a well-developed connective tissue capsule. The presence of an intact capsule reinforces its benign nature. Its presentation may however vary, ranging from a solid mass to a single large cystic space (unilocular), or numerous small cystic spaces (multilocular). All variants of AOT appear remarkably similar histologically and are composed of odontogenic epithelium embedded in connective tissue stroma. The presentation is usually seen as spindle or polygonal shaped cells arranged in sheets and whorled masses in a scant connective tissue stroma. Between epithelial cells as well as in the centre of rosette-like structures is an amorphous eosinophilic material. Duct-like structures in the lesion are seen to be lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen [18]. The reason for characteristic ductal architecture is still hypothetical. Some believe it to be due to a cystic change in the follicles of tumor islands, or probably an attempt to form glandular tissue, since the origin is from the basal cells of the oral epithelium that have multiple differentiation capacity [19].

Dystrophic calcifications, in varying amounts and forms such as irregular masses, leisegang rings, spheroidal masses or globular forms, are usually encountered in 78% of AOTs, within the lumen of duct-like structures, scattered among epithelial masses, or in the stroma [18]. Calcifying epithelial odontogenic tumor (CEOT) shows larger and more numerous calcifying spherules within the eosinophilic cytoplasm of larger cells along with smaller cells having hyperchromatic nuclei [20].

The histogenesis of AOT is still uncertain. Whether it is a hamartomatous malformation or a neoplasm continues to be debatable. Because of the relatively small size of lesion and lack of recurrences in most cases, findings to a large extent, support the fact that it is a hamartoma. On the other hand, few authors suggest that early detection is the reason for small size of the lesion. Increased variation and aggressive features in few reported cases is indicative of its neoplastic origin [19].

Radiographically, extraosseous and peripheral variants of AOT are rarely visible on radiographs, with only slight erosion of the adjacent alveolar bony cortex seen. Intraosseous follicular variety presents as a unilocular radiolucency but multilocular variants are also reported. Follicular variant of AOT has a central lesion, is associated with an embedded tooth and is usually confused with a dentigerous or follicular cyst initially, where as extrafollicular variant has a central lesion but is not associated with a tooth and presents as a unilocular radiolucency between, above or superimposed over roots of erupted teeth. Its four different manifestations on radiographs can be regarded as $E_1 - E_4$: $E_1$, without relation to tooth structures either erupted or un-erupted; $E_2$, inter-radicular, adjacent roots diverge apically as a result of tumour expansion (mimicking a globulo-maxillary cyst); $E_3$, superimposed on the root at the apex level (mimicking a radicular cyst) and $E_4$, superimposed at the mid-root level [21]. Extrafollicular AOT is often confused with globulo-maxillary or lateral periodontal cyst. The rare peripheral
variant occurs primarily in the gingival tissue, either palatally or lingually of the involved tooth and is not seen on the radiograph [22,23]. Discrete foci of calcifications in an AOT are visible as radiopacities, much better on a periapical radiograph than a panoramic radiograph.

Differential diagnosis radiographically includes dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globo-maxillary cysts, ameloblastomas, odontogenic keratocysts and periapical disease. As AOT imitates a dentigerous cyst in large number of cases, differentiation from a dentigerous cyst is important. The later frequently appears as a pericoronal radiolucency in the jaws i.e. enclosing only the coronal portion of the impacted tooth whereas AOT shows radiolucency usually enclosing both the coronal and radicular portions of the tooth [20]. Mostly, follicular AOT closely resembles a follicular cyst, extrafollicular AOT a residual or "globulo-maxillary" cyst, and the peripheral variant a gingival fibroma.

Considering the benign behaviour of AOT, its slow growth, clear delimitation, and low recurrence, the treatment of choice is conservative enucleation and simple curettage. In exceptional cases where tumor size is large or when risk of bone fracture exists, partial en bloc resection of the mandible or maxilla is indicated. Where surgical extirpation has left a large exposed osseous cavity, additional use of lyophilized bone and guided tissue regeneration is recommended [24]. Recurrence of AOT is exceptionally rare. Only three cases have been reported till now in Japanese patients where recurrence of this tumour has been observed [25], therefore it can be said that the prognosis is excellent when completely removed in toto.

3. LITERATURE REVIEW

Historically, AOT got its description almost about 100 years back but lot of confusion existed as regards its distinct entity for decades. This was due to the various terminologies that were used for cases appearing similar to AOT in literature. James and Forbes from England in 1909 reported a case of epithelial odontome, which was similar to AOT [26]. Harbitz of Norway in 1915 reported a case of cystic adamantoma [26]. Wohl of Omaha, 1916 reported a case similar to AOT as a tooth germ cyst of the jaw [26]. Bernier and Tiecke were the first to publish a case using the name adenoameloblastoma [27]. Abrams et al. in 1968 proposed the term odontogenic adenomatoid tumor while Philipsen and Birn in 1969 suggested the name adenomatoid odontogenic tumor [3]. Later in 1971, the term AOT was finally adopted in the initial edition of World Health Organization (WHO)'s histological typing of odontogenic tumor, jaw cysts, and allied lesions [28] and continued to exist in the 2nd edition of WHO in 1992. Philipsen et al. in 1997, presented an update on biologic profile of AOTs based on data published by authors between 1990-1996 [25].

4. CONCLUSION

Adenomatoid odontogenic tumor of maxillary sinus is a rare benign epithelial odontogenic tumor, closely resembling an odontogenic cyst. Identification of AOT is possible using standard diagnostic modalities like radiographs, examining clinical and histopathologic presentations, and having detailed knowledge about lesions with similar presentations. Computed tomographic scans supplement diagnostic information by showing the extent of lesion and any infiltrations, hence should be advised, whenever possible. Fortunately, all variants of AOTs have similar biologic behaviour and can be conservatively treated using simple enucleation and curettage with excellent outcomes. Though recurrences reported after excisions are extremely low, careful follow up is still mandated. To conclude, expert morphological, radiological and histological observations are required to establish an accurate diagnosis for all enucleated cysts/tumors in this region, thus contributing to an increase in our literature bank.

REFERENCES

http://dx.doi.org/10.1016/0030-4220(79)90051-3
http://dx.doi.org/10.1186/1746-160X-1-3
http://dx.doi.org/10.1111/j.1600-0714.1991.tb00912.x
http://dx.doi.org/10.1016/0278-2391(92)90327-V
http://dx.doi.org/10.1053/joms.2000.9581
http://dx.doi.org/10.4103/0976-237X.96837
http://dx.doi.org/10.4103/0973-097X.92987
http://dx.doi.org/10.4103/0976-237X.95100
http://dx.doi.org/10.1259/dmfr.20120410
http://dx.doi.org/10.1016/0030-4220(87)90372-0
http://dx.doi.org/10.1902/jop.1996.67.1.46
http://dx.doi.org/10.3353/omp.2.55
http://dx.doi.org/10.2209/tdcpublication.45.223
http://dx.doi.org/10.1016/0030-4220(56)90232-8

© 2015 Passi et al.; Licensee Revotech Press. This is an open access article licensed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licenses/by-nc/3.0/), which permits unrestricted, non-commercial use, distribution and reproduction in any medium, provided the work is properly cited.