Clinicopathological Correlation for Adenomatoid Odontogenic Tumor of Maxilla an Enigma to Maxillofacial Surgeons

Charu Dixit, Shaji Thomas, Vertika Dubey, Pooja Khokhar, Siddharth Dubey, Satyaprakash Nigam

Department of Oral and Maxillofacial Surgery, People's College of Dental Sciences and Research Centre, Bhopal, Madhya Pradesh, India.

Abstract

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic benign lesion with a slowly progressing growth predominantly associated with impacted maxillary cuspids. The majority of the cases are reported to occur in young female patients and the common site of occurrence is in maxilla particularly in anterior region. However, cases of AOT in posterior maxilla have also been reported, though extremely rare. In this paper, we present a case report of one such case where a 12-year-old female presented to our hospital with chief complaint of pain and swelling over right side of the face since 1 1/2 months, which was misdiagnosed as dentigerous cyst, both clinically and radiographically but the histopathological examination revealed it to be an AOT. Site specificity for AOT with the occurrence of an impacted cuspid should point toward the primary diagnosis of AOT over dentigerous cyst in such clinical presentations.

Keywords: Adenomatoid odontogenic tumor, Adenomatoid odontogenic tumor, Dentigerous cyst, Non-neoplastic, Odontogenic tumor.

Introduction

In 1948, Stafne was the first person to recognize this uncommon entity with a report of three cases. He described adenomatoid odontogenic tumor (AOT) as epithelial tumors that are commonly misdiagnosed for ameloblastoma. Unat *et al*. have listed various nomenclature for AOT in their study.[1] Various forms used for this tumor include – adenoameloblastoma, cystic complex composite odontome, tumor of enamel organ epithelium, ameloblasticadenomatoid tumor, epitheliomaadamantinum, and teratomatousodontoma.[2-4] Few authors have reported it to be a benign neoplasm while some claimed that it was a variety of hamartomatous malformation.[5] It is considered odontogenic in origin due to the fact that it is always associated with unerupted or impacted tooth and cytologically it bears resemblance to dental lamina and enamel origin.[6-9] This was mentioned in some of the studies by few authors as "envelopmental theory."[10,11] The mean age of occurrence is 13.2 years with an age range from 3 to 28 years. There is a marked gender predilection as the majority of the cases were reported in females (F: M ratio = 2.3:1). The common site reported is anterior maxilla when compared to posterior maxilla and mandible (Maxilla: Mandible = 2.6: 1). Philipsen and Reichart have reported three clinical variants of AOT. They are follicular, extrafollicular, and peripheral types. The follicular and extrafollicular variants are intraosseous whereas peripheral variant is extraosseous. The majority of the AOTs are follicular variant (71%).[3] We report one such case of a 12-year-old female patient who was provisionally diagnosed to have dentigerous cyst, both clinically and radiographically, but the histopathological examination revealed it to be an AOT of follicular variety. This article also provides an insight into review of the literature on the clinical, histopathological, radiological, and immunohistochemistry features of the lesion.

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Case Report

A 12-year-old female patient came to outpatient department of oral and maxillofacial surgery with a chief complaint of pain and swelling in the right side of the face since 1 1/2 months. Patient gave a history of mobile upper front tooth which was asymptomatic and she tried to pull the tooth out. Swelling gradually increased in size over the right side of face in 15 days. Patient also gave a history of watery eyes since 5 days.

Address for correspondence: Shaji Thomas, Department of Oral and Maxillofacial Surgery,PCDS & RC Bhopal, Madhya Pradesh, India.
E-mail: shajihoss@gmail.com

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On examination extraorally, a well-defined solitary swelling was present over the right side of middle 1/3rd of the face extending 1 cm beneath the right infraorbital rim and extending up to the lateral aspect of the corner of the mouth superioinferiorly. Anteroposteriorly, it extends from lateral aspect of right side of nose to zygomatic buttress region, measuring approximately 3.5*4.5 (Figure1). The skin over the swelling was smooth and shiny in texture. Color over the skin was normal. Obliteration of nasolabial fold was present, with evidence of straw colored fluid draining from the right nostril. On palpation, temperature of the overlying skin was elevated. Swelling was tender and firm in consistency.

Intraorally, a diffuse swelling was present in the right maxillary buccal vestibular region extending from lateral incisor (12) to first molar (16) region. On palpation, the swelling was tender and firm in consistency with buccopalatal expansion of the right alveolar ridge. Presence of a retained deciduous tooth (53) along with clinically missing 13 was evident.

Radiographically, panoramic radiograph (OPG) revealed, a radiolucency, measuring approximately 4 cm in diameter extending from radiographic apex of 12 to apex of 16 in anteroposterior diameter. A radiopaque tooth like structure was evident at the apex of 15 which morphologically resembled a maxillary canine. Hence, a diagnosis was made favoring toward that dentigerous cyst was given as per the clinical and radiographic data available (Figure 2).

Cone beam computed tomography (CBCT) revealed a single expansile osteolytic lesion present in the right maxilla extending from 12 to 16 mesiodistally and inferiorly from the crest of alveolar bone to the floor of the right maxillary sinus measuring approximately 3.8 × 3.2 cm. Internal structure revealed minor radiopaque flecks with thinning and perforation of buccal cortical plate and displacement of 13 toward the right maxillary sinus was evident (Figure 3). Correlating the CBCT and OPG findings with clinical examination, the lesion appeared to be a dentigerous cyst with differential diagnosis of ameloblastoma and AOT.

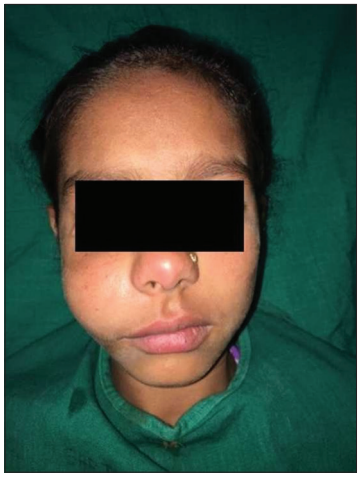

Figure 1: Swelling in the right side middle third of the face

Surgical enucleation of the cyst was planned under local anesthesia (2% Lignocaine with 1:2,00,000 adrenaline) under standard aseptic protocols. A trapezoidal mucoperiosteal was raised after the incision extending from the mesial aspect of 11 to distal aspect of 16. A small bony window was created measuring approximately 4 cm x 3 cm in the labial cortical plate from 12 to 15 region. The complete mass was enucleated with the embedded tooth followed by debridement and copious saline irrigation (Figure 4). The mucoperiosteum was positioned back in its anatomic position and wound closure was done using 3–0 polyglactin sutures, (Figure 5). The enucleated specimen was sent for histopathological examination after tissue fixation with 10% formalin solution, (Figure 6). Patient was administered antibiotics, analgesics and was kept on 0.2% chlorhexidine mouthwash.

The histopathological examination of the H and E stained section of the specimen showed presence of portions of

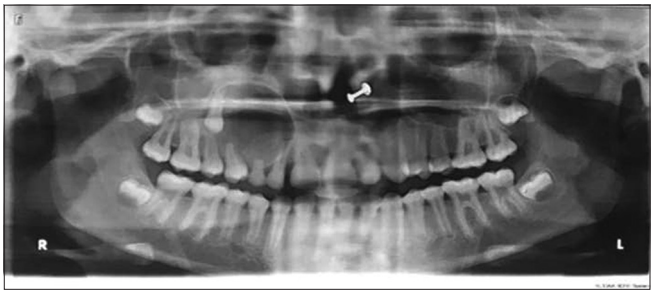

Figure 2: OPG reveals radiolucency involving apex of 12 to mesial aspect of 16 along with a tooth like structure at the apex of 15 suggestive of an impacted/unerupted tooth

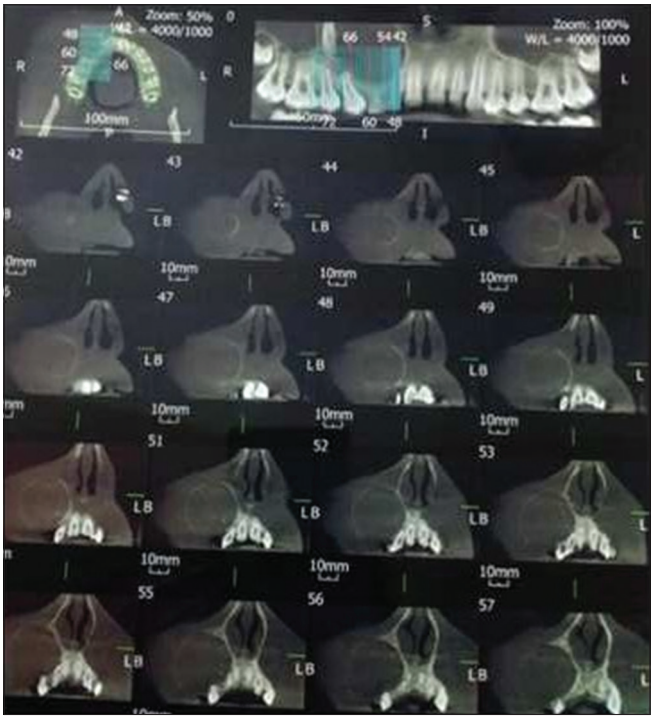

Figure 3: CBCT reveals radiopaque flecks with thinning of the buccal cortical plate with expanded osteolytic lesion and presence of 13 in the vicinity of the right maxillary sinus

incomplete peripheral capsule with the central regions exhibiting cystic as well as solid areas. Biomorphic, spindle, and cuboidal cells are seen with spindle cells arranged in whorled and rosette pattern. Cuboidal cells arranged in a duct like pattern around a central space. Areas with plexiform cords of basaloid appearing cells were also evident. Numerous microcystic spaces with eosinophilic coagulum were observed. Vascular areas and hemorrhage were also present. Histological along with clinical features indicated toward the final diagnosis of AOT (Figures 7 and 8). Patient was on a regular follow-up for 3 months postoperatively. Post-operative period was uneventful.

Discussion

AOT is generally an uncommon odontogenic tumor with diverse histomorphological features which were first described in 1905 by Steensland.[3] Philipson and Birn in 1969 proposed the term AOT considering the fact that it was a different entity from ameloblastoma.[4,10] Philipson and Richard put forward three variants, namely, peripheral, follicular, and extrafollicular type of AOT.[3,11-13] According to them, the follicular type which is also termed as pericoronal type is the one related to unerupted tooth. However, the extrafollicular or extracoronal type is not associated with unerupted teeth.[3] The peripheral counterpart is usually extraosseous and occasionally shows erosion or Saucerization of the involved alveolar bone crest.[7] According to Philipsen *et al*., the peripheral variant is usually painless and occurs as gingival masses in maxilla and a marked female predilection is seen for gingival lesion with female to male ratio of 14:1.[6] As per the literature search, the clinical feature of AOT is often presented as tender or non-tender swelling of jaw associated with unerupted/impacted tooth which is usually slow growing,[3,6,7,10,14] and commonly occurs in anterior maxilla.[10,15] John *et al*. emphasized that AOT often envelops the crown and root portion of the involved tooth unlike a dentigerous cyst. He has highlighted the case reports in the literature where it is believed that AOT develops in the dentigerous cyst's capsule and its association with dentigerous cyst.[10,16,17] Rosa *et al*. in their study have reported a case of follicular AOT associated with calcifying epithelial odontogenic tumor (CEOT).[15] In our reported case, patient had complaint of watery eye which could possibly be due to expansion of the AOT causing obstruction of nasolacrimal duct due to the pressure effect on the duct.

Radiographically, AOT presents with unilocular radiolucency with well-defined border but sometimes a sclerotic margin with or without bony expansion may be evident.[12,13,15] Root divergence and displacement of the involved tooth rather than root resorption are common.[6,10,15] Extrafollicular variants exhibit flocculent pattern showing mixed radiolucency and radiopacity, resembling a radicular cyst.[3,15] Some cases exhibit radiolucency with or without internal calcifications.[15,18] If present, calcifications are snowflake like in appearance which may be a differentiating feature of AOT from dentigerous cyst.[18,19] Other radiographic differential diagnosis that can be considered is keratocystic odontogenic tumor, Ameloblastic fibroma, odontogenic myxoma, unicysticameloblastoma, and dentigerous cyst.[15,19]

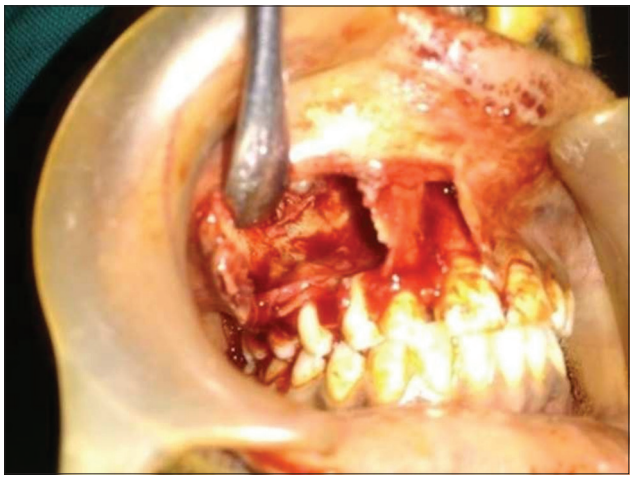

Figure 4: Bony window was created surgically followed by elevation of mucoperiosteal flap, the cystic contents were enucleated followed by irrigation and debridement

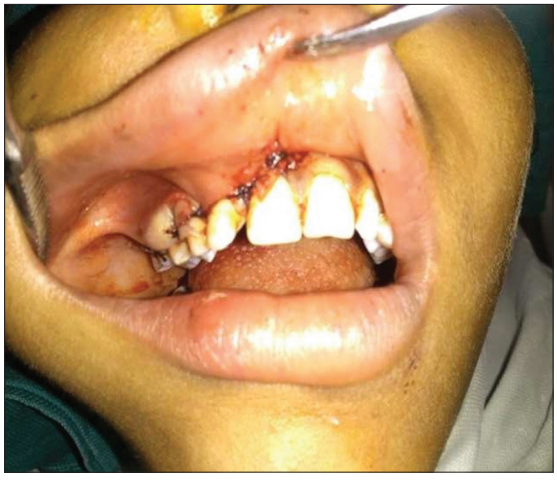

Figure 5: Wound closure done with 3–0 Polyglactin suture

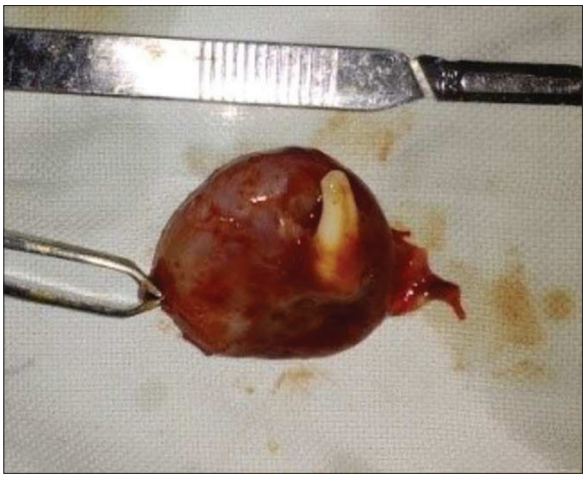

Figure 6: The enucleated specimen along with the tooth was sent for histopathological examination

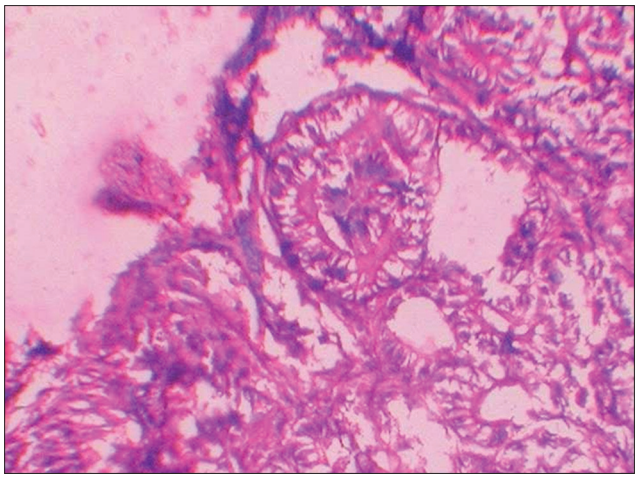

Figure 7: Histopathological section shows spindle cells arranged in whorled and rosette like pattern. The presence of plexiform cord with basaloid cells can also be seen

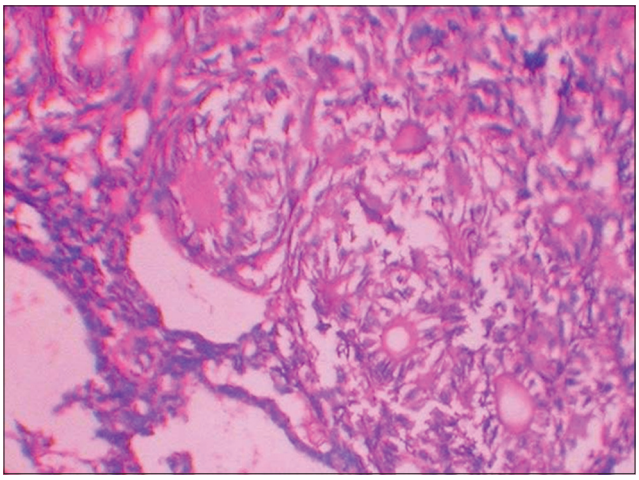

Figure 8: Numerous microcystic spaces with eosinophilic coagulum are observed

Certain areas of the tumor exhibit similar histomorphological features of CEOT. The pathogenesis of this tumor is not exactly known. The most common source of tumor development is residue from dental lamina and odontogenic epithelium adjacent to reduced enamel epithelium. A well-defined capsule surrounds the proliferating epithelium of the tumor with spindle, polyhedral, and cuboidal cells. The epithelium is in the form of whorled masses of spindle cells as well as sheets of plexiform strands. The duct like appearance is formed by rings of coronal cells. Duct like cystic spaces is also evident along with new blood vessels lined by columnar epithelium and the nuclei are polarized away from the lumen.[4,6, 15,20,21] Tumor droplets are also evident which are homogenous matrix of eosinophilic amorphous material.[2,21,12] The nodules present are composed of polyhedral and eosinophilic epithelial cells of squamous appearance with prominent intercellular bridge. Some authors have advocated the presence of dentinoid like material or osteodentin. Rarely presence of dentinal tubules may be evident.[8,15,21] Calcifications may occur in varying degrees.[10]

Immunohistological studies on AOT have been put forth by several researchers in recent years emphasizing that AOTs are identified by a cytokeratin (CK) profile which is similar to that of gingival epithelium positive for CK 5, 17, and 19. It has been also reported that AOT is negative for CK 4, 10, 13, and 18. A study conducted by Crivelini detected the presence of CK 14 and inferred that this expresses its origin in the reduced dental epithelium as they are positive for staining with CK 14 antibodies.[3,13,22] The studies suggest that AOT is also positive for Ki67 antigen which suggests that it marks for low cell proliferation, thus endorsing for slow growth of the tumor.[10]

Clinically for AOT, differential diagnosis can be dentigerous cyst, ameloblastoma, and calcifying odontogenic cyst.[23] Surgical enucleation with or without reconstruction is the treatment of choice. Choice of reconstruction, if required, depends on the size of the defect.

Conclusion

Several authors had mentioned in their study that AOT is also referred to as "Two-third's tumor" as 2/3rd of the tumor site is maxilla, 2/3rd cases are diagnosed in young females patients, 2/3rd cases were all associated with an impacted/unerupted tooth, and in 2/3rd of the cases, the tooth impacted/unerupted is canine.[9,13,19,20] During examination of an asymptomatic, painless, and slowly progressing swelling in the jaw, AOT should be considered as one of the differential diagnosis. As it is a benign well capsulated tumor, conservative surgical enucleation with or without reconstruction of defect has shown complete cure. The treatment was planned specifically for the case. This article highlights the occurrence of AOT in a young female patient with an impacted canine and the lesion involving maxilla contrary to the usual clinical presentation which was misdiagnosed as dentigerous cyst. Site specificity for AOT with the occurrence of an impacted cuspid should point toward the primary diagnosis of AOT over dentigerous cyst in such clinical presentations.

Ethical Approval

All the procedures performed in this case were in accordance with the ethical standards of the Institutional and/or National Research committee and with the Helsinki declaration and it's later amendments or with any other comparable ethical standards.

Informed Consent

Informed consent was obtained from the patient involved in this report.

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