Adenomatoid Odontogenic Tumour– A case report
Ramaswamy P1, Geethika VR2, Sreenivasulu Pattipati3*, Vijay Srinivas G4, Praveen Kumar B5 and Uday Ginjupalli6

ABSTRACT
Adenomatoid odontogenic tumour (AOT) is a benign, non-invasive odontogenic tumour with slow and sustained growth. It is relatively rare and unusual, and mainly affects females in their second decade of life, exhibiting predilection for the anterior region of the maxilla. The lesion is usually associated with the crown of an enclosed tooth, most commonly the maxillary canine. In this paper, we present a case of a 19-year-old female having the classical clinical, radiological and histopathological features, leading to the diagnosis of AOT.

KEYWORDS: Adenomatoid odontogenic tumour, Odontogenic tumour, Odontogenic epithelium, infected dentigerous cyst, Enucleation

INTRODUCTION
Adenomatoid odontogenic tumour (AOT) is considered as an epithelial neoplasm of odontogenic origin, having an incidence rate of 2.2–7.1% [1-4]. It was first described by Steensland in 1905 as epithelioma adamantine [3, 5, 6]. In 1907, it was described by Dreibladt and was called as a pseudoadenoma adamantinoma [7]. In 1915, Harbitz reported a case of cystic adamantinoma in a 15-year-old girl. This tumour was believed to be a variant of ameloblastoma, but in 1948, Stafne described it as a distinct histological entity [8]. Bernie, Aid Tiecke published a case using the name adenoameloblastoma [9]. In 1969, Philipsen and Brin proposed the name AOT, which was widely accepted, and the term was adopted by first edition of the World Health Organization (WHO) in 1971 [10].

The WHO, in 2005, defined the AOT as a tumour composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterised by a slow progressive growth[2,6].

It was referred to as ‘two-thirds tumour’ because it occurs in the maxilla in about 2/3 cases, about 2/3 cases arise in young females, 2/3 cases are associated with an unerupted tooth and 2/3 affected teeth are canines [2, 11]. Here, we describe a classic case of AOT with clinical, radiological and histological features, which can be appropriately termed as two-thirds tumour.

CASE REPORT
A 19-year-old female reported with a complaint of swelling on the left side of face since the last 3 months. The swelling started small and gradually increased in size over a period of 3 months to attain the present size. It was associated with mild, intermittent pain since 1 month. There was no history of trauma or numbness in this region.

Extraoral examination revealed facial asymmetry with obliteration of naso-labial fold. On inspection, a diffuse

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1Professor & Head, 2Post Graduate Student, 3Senior Lecturer, 4Reader, Department of Oral Medicine & Radiology, 5Professor & Head, Department of Oral & Maxillofacial Pathology, St. Joseph Dental College & Hospital, Duggirala, Eluru-534003, Andhra Pradesh, India. Corresponding author email id: *dr.pattipati@gmail.com; ramomdr@gmail.com; 2geethika24@yahoo.com; 4vijaysrinivasg@hotmail.com; 5dr.praveenrao@yahoo.co.in; 6uday2361@gmail.com
swelling was seen on the left middle third of the face measuring approximately 3 x 3 cm² in size, extending anteroposteriorly from the ala of the nose to 2 cm in front of the preauricular region and superioinferiorly from the infraorbital rim up to 0.5 cm above the corner of the mouth (Figure 1). On palpation, the swelling was tender and firm-to-hard in consistency. A local rise in temperature was also noted. Bilaterally, submandibular lymph node was palpable, measuring approximately 1 x 0.5 cm² in size, single, firm, mobile and tender on palpation.

Intraorally, an oval-shaped swelling was seen on the buccal aspect of teeth 22-26, measuring approximately 3 x 2 cm² in size, extending anteroposteriorly from the mesial aspect of tooth 22 to the distal aspect of tooth 26, and superioinferiorly from the marginal gingival to upper buccal vestibule. Obliteration of buccal vestibule was present (Figure 2). Retained deciduous tooth was present with relation to tooth 63, where a missing upper left canine was also noted. Mobility with relation to teeth 63, 24 and 25 was noted. Based on the history and clinical features, an infected dentigerous cyst with relation to tooth 23 was provisionally diagnosed.

Radiological investigations were performed and panoramic radiography showed a large unilocular radiolucency on the left side of the maxilla, extending from tooth 22 to 26. External root resorption was observed in relation to teeth 63 and 26. Impacted tooth 23 was present in the radiolucency (Figure 3). Paranasal sinus view showed increased radiopacity in the left maxillary sinus region with impacted tooth 23 (Figure 4).

On fine-needle aspiration, an amber-coloured fluid was obtained which revealed isolated and clusters of keratinocytes with normal nuclei, which suggested a dentigerous cyst (Figure 5). Therefore, extensive enucleation of the lesion was done with the patient under general anaesthesia, and the specimen was sent for histopathological examination (Figures 6 and 7).

Examination of the haematoxylin and eosin-stained sections showed a multinodular proliferation of spindle, cuboidal, columnar and stellate reticulum-like cells arranged in sheets. Duct-like layers, anastomosing strands of basaloïd cells and focal calcifications were evident in the delicate fibro-vascular stroma (Figures 8 and 9). Based on these findings, a final diagnosis of AOT was considered.

Clinical and Radiographic Assessment after Tumour Enucleation
Clinical and radiographic assessment was done at 3 months after enucleation. During follow-up, there were no signs of inflammation, infection or recurrence of the tumour; clinically and radiographically, there was a large radiolucent area with degree of decrease in radiolucency (Figures 10 and 11).

DISCUSSION
Odontogenic tumours are a group of heterogeneous lesions, features of which have been catalogued for several decades. AOT is a relatively rare and distinct odontogenic tumour that is exclusively odontogenic epithelium in origin. Various synonyms have been used for this tumour. Unal et al. listed the catalogue for AOT that is reported in the literatures. These are adenoameloblastoma, ameloblastic adenomatoid tumour, adamantinoma, epithelioma adamantinum or teratomatous odontoma [3]. They mostly occur in young people, peak incidence is in the second decade of life, uncommon in patients above 30 years of age. The incidence of female to male ratio is 1.8:1. It is predominant in the anterior region of the jaws. They are frequently encountered in the maxilla than in the mandible, with a ratio of 2:1 and are most commonly associated with impacted teeth. Canines are the most common tooth associated with AOT [12].

AOT is divided into two variants: central and periphery. Further, the central variant is divided into two subtypes: follicular and extra-follicular. The central variant occurs at rate of 97.2%, out of which, 73.0% are follicular. The follicular variant is three times more common than the extra-follicular type. The follicular variant is
diagnosed earlier in life (mean age 17 years) than the extra-follicular type (mean age 24 years); 53.1% of all variants occur within the teen years (13-19 years). A study showed that follicular AOT was associated with one embedded tooth in 93.2% of the cases. Maxillary permanent canines account for 41.7% and all four canines for 60.1% of AOT-associated embedded teeth [13].

The follicular variant is associated with unerupted tooth surrounding the crowns and is attached to the necks of the tooth [14]. The extra-follicular variant is not associated with the impacted tooth, superimposed over the roots of the teeth. A peripheral variant occurs over the gingiva and looks as a gingival growth [14, 15]. Histologic and immune-histochemical evidences are present for the follicular variant showing that they originate from the reduced enamel epithelium of the dental follicle [16]. Origin of the extra-follicular variant is less clear. Philipsen et al. [17] suggest that all the AOT variants show identical histology, have a common origin and implicate the dental lamina or its remnants. However, the hypotheses for the pathogenesis of AOT are still uncertain [17]. It was proposed that it may arise from the enamel organ, the epithelial lining of dentigerous cyst, epithelial rests of Malassez of the deciduous or permanent tooth or the remnants of the dental lamina [18], though none of these theories had ample support [17].

Clinically, AOT is a slow growing, asymptomatic swelling associated with unerupted tooth. It is usually discovered during routine radiographic examination [19]. The radiographic features of AOT frequently resemble other odontogenic lesions, such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumours, globulomaxillary cysts, ameloblastomas, odontogenic keratoctysts and periapical disease [20]. The follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and, often, with a part of the root of an unerupted tooth, and sometimes it is associated with radiopaque calcifications. This feature of sparse pebble-like calcifications and the attachment of radioluency more apical to the cemento-enamel junction can distinguish the lesion from a dentigerous cyst [21]. The radiolucency of the extra-follicular type is located between, above or superimposed upon the roots of the erupted permanent teeth. Displacement of neighbouring teeth due to tumour expansion is much more common than root resorption. The peripheral lesions may show some erosions of the adjacent cortical bone [13].

Macroscopically, AOT is surrounded by a connective tissue capsule. When dissected, it may appear as a solid mass, as seen in our case, or may have varying degrees of cystic changes [2].

The AOT has a distinctive histopathologic appearance, i.e. spindle-shaped epithelial cells are arranged in the form of sheets, strands or whorled masses surrounded by connective tissue stroma. These epithelial cells may form rosette-like structures with a central space, which may be empty or contain an eosinophilic amyloid-like material. The characteristic feature of AOT is the presence of duct-like structures that are surrounded by columnar cells with nuclei oriented away from the lumen. These structures are not true ducts, and no glandular elements are present in the tumour. Small foci of calcifications are scattered throughout the tumour. It may also present in the lumen of the structures [1, 2, 17 and 19].

Conservative surgical enucleation is the treatment modality of choice. Recurrence of AOT is rare [115 and 17]. Only three cases in Japanese patients are reported in which the recurrence of this tumour occurred; therefore, the prognosis is excellent when the lesion is completely removed [22].
Figure 1: Extra oral swelling noted on left middle third of face.

Figure 2: Intraoral swelling and vestibular obliteration noted on left upper buccal vestibule.

Figure 3: Panoramic radiography demonstrates a large unilocular radiolucency on the left side of the maxilla.

Figure 4: Paranasal sinus view demonstrates increased radiopacity in the left maxillary sinus with impacted canine.

Figure 5: Fine needle aspiration an amber colored fluid was obtained

Figure 6: Showing enucleation of the lesion.
CONCLUSION

The present case is a rare report of an adenomatoid odontogenic tumor presented in maxilla of 19 year old female involving impacted left upper canine and showing sheets of epithelium and focal calcifications in histopathological picture and it was treated by surgical enucleation.

ACKNOWLEDGEMENT

We would like to acknowledge Dr. Anuradha A. for reporting and helping us in the histopathology.

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