Adenomatoid Odontogenik Tumor (AOT): A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon neoplasm originating from odontogenic epithelium, accounting for approximately 3–7% of all odontogenic tumors. It is a benign, non-invasive lesion characterized by slow but progressive growth. AOT is more commonly found in the maxilla, particularly in females, and is frequently associated with impacted canines.

We report a case of A 14-year-old female presented with a swelling on the left side of her upper jaw, which had been gradually enlarging over the past year. The lesion increased in size over time. Extraoral examination showed facial asymmetry. A solitary, diffuse, hard mass on the left anterior maxilla. The overlying mucosa appeared normal, and the lesion measured approximately 2.5 cm in diameter while tooth 22, 23, and 24 were unerupted.

The lesion was treated by enucleation, along with removal of the embedded teeth. Histopathological examination confirmed the diagnosis of adenomatoid odontogenic tumor. No recurrence was observed during a 6-month follow-up period.

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Introduction

Adenomatoid odontogenic tumor (AOT) is a rare, benign odontogenic neoplasm primarily affecting young individuals, particularly females, during their second decade of life. Characterized by slow, asymptomatic growth, AOT is often classified into three variants: follicular, extrafollicular, and intraosseous, with the follicular type being the most prevalent. This tumor is typically associated with impacted teeth, especially the maxillary canine, and is considered more of a hamartoma due to its limited growth potential and low recurrence rate[1,2,3]

AOT usually presents as a painless swelling in the anterior maxillary or mandibular regions. It can cause midline deviation and is often discovered incidentally during dental examinations[3,4]. Histologically, AOT consists of odontogenic epithelium arranged in various patterns within a fibrous stroma. The presence of duct-like structures and whorled cell masses is characteristic[2,3]. Surgical enucleation is the standard treatment, with a favorable prognosis and minimal complications reported post-surgery[3]. Long-term follow-up is essential to monitor for any potential recurrence, although this is rare[1].

Despite its benign nature, AOT's varied clinical and histopathological presentations can complicate diagnosis, necessitating careful evaluation to differentiate it from other odontogenic lesions. In this paper, we reported case of adenomatoid odontogenic tumor on a young female with a swelling on the left side of her upper jaw.

Case Report

A 14-year-old female complained of growth in the left of her upper jaw that occur since 1 year ago. it was small initially but then slowly growing attaining the present size. There was no history of trauma, pain or discharge from the lesion. Extraoral examination revealed facial asymmetry. Intraoral examination revealed a solitary diffuse hard mass on anterior the left side of the anterior maxilla. The colour of overlaying

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mucosa was normal. Swelling was roughly oval, measuring approximately 2,5 cm in diameter. It has reported of unerupted teeth 22,23,24 and persistent teeth 63, 64 and 65. The character of palpation on swelling was bony hard, non-tender, and immobile.

Radiographically, a well-defined 4 cm unilocular ovoid radiolucent lesion with sclerotic borders extending anteroposterior from distal aspect of 21 to mesial aspect of 25. The floor of the maxillary sinus was highly displaced, limiting superiorly the lesion. Aspiration biopsy carried out revealed nothing from the lesion. We make provisional diagnosis of dentigerous cyst with differential diagnosis of AOT, Unicystic ameloblastoma, Odontogenic Keratocyst OKC (Keratocystic Odontogenic Tumour KOT).(Fig. 1,2)

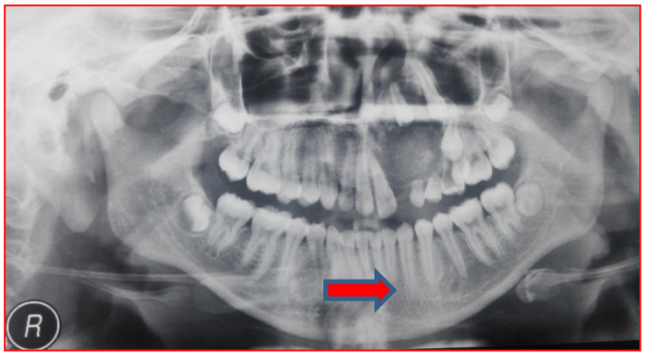

Figure 1. Orthopantomograph (OPG) reveals unilocular radiolucency with impacted 22,23, 24.

Figure 2. Intraoral view demonstrates the labial aspect of the swelling.

Complete enucleation of the lesion was done under general anesthesia with removal of impacted 22,23 and 24. A mucoperiosteal flap from midline to the left premolar region was reflected to expose the labial aspect of the tumor. The labial cortex was very thin. Enucleation was achieved by separation of the lesion from bone with complete removal of the capsule. The area between the roots of involved teeth were curetted well. The cavity was packed with Lyostypt (BBraun, AESCULAP AG ft CO. KG) and the flap was sutured in place. Healing was uneventful, and there was no evidence of recurrence 6 months after surgery.(Fig. 3,4)

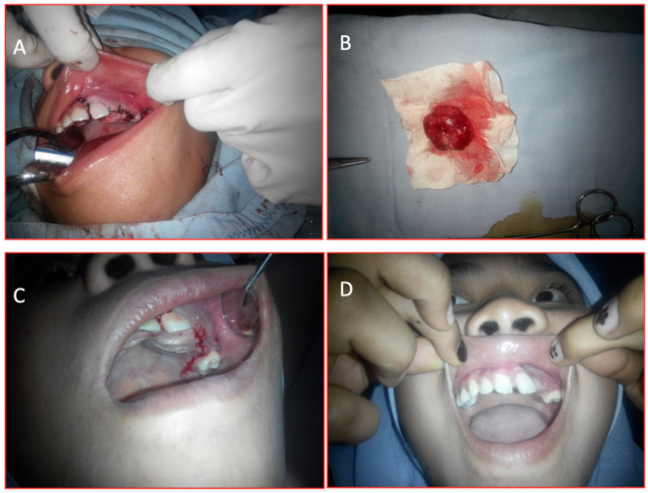

Figure 3. Surgical procedure: A. Removal of the lesion: B. The excised tissue; C. 7 days post operative; D. 6 months post operative.

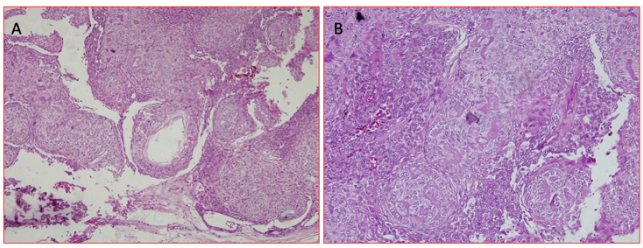

Figure 4.A H&E showed nets and duct like structures lined by low columnar epithelial cells (H&E stain 10x); **4.B.** H&E showed thick collagenous wall on the outermost followed by thick epithelial lining and cystic lumen filled with eosinophilic material (H&E stain 10x).

Histopathological examination (H&E) revealed sheets of polygonal cells throughout the fibrous connective tissue stroma. The ductal lumina were surrounded by columnar epithelial cels and filled in some areas with eosinophilic material. In other places amorphous calcified material was present. The histopathological report confirmed the diagnosis of adenomatoid odontogenic tumor.

Discussion

Adenomatoid odontogenic tumor (AOT) is a benign epithelial tumor often found in adolescents and young adults, with a marked predilection for females and the anterior maxilla[1,2,3,5]. In this case, the patient was a 14-year-old female, consistent with the typical demographic profile. AOT commonly presents as a slow-growing, painless swelling, often discovered incidentally or due to facial asymmetry, as observed in this case[6,7,8,9,10,11,12,13].

There are two main types of AOT: central (intraosseous) and peripheral[6]. The central variant can be either follicular, which is typically linked to unerupted teeth, or extrafollicular, which is not associated with teeth but is usually located between the roots of existing teeth[6,7]. The case described is an intraosseous follicular variant, which has an extremely low likelihood of recurring following surgical enucleation.

Radiographically and clinically, AOT may resemble other odontogenic lesions, such as dentigerous cysts or other benign tumors[3,7]. The presence of unerupted teeth, particularly canines and adjacent teeth (in this case, tooth 22, 23, and 24), is a hallmark feature that often guides diagnostic suspicion toward AOT[8]. Surgical enucleation is the treatment of choice for AOT, given its benign and non-invasive nature[1,2].

A complete excision was carried out, considering the patient's age, the lesion's morphology, and the associated functional impairment[14]. Complete excision usually results in an excellent prognosis with minimal risk of recurrence. If left untreated, the tumor may result in various complications. Complete enucleation along with the extraction of unerupted teeth continues to be the standard treatment approach[15]. In this case, enucleation along with the removal of the unerupted teeth was successfully performed. Histopathological examination confirmed the diagnosis, revealing characteristic features such as duct-like structures, epithelial rosettes, and spindle-shaped epithelial cells.

Follow-up is essential to monitor for any signs of recurrence, although recurrence is extremely rare when the tumor is completely excised. The absence of recurrence in the 6-month follow-up period supports the effectiveness of the surgical approach used. This case underscores the importance of including AOT in the differential diagnosis of jaw swellings in young patients, especially when associated with impacted teeth. Early diagnosis and proper management are key to preventing potential complications and ensuring favorable outcomes.

Conclusions

Adenomatoid odontogenic tumor (AOT) is a rare, benign, and slow-growing lesion that typically affects young females and is commonly associated with impacted teeth in the anterior maxilla. Early diagnosis, supported by clinical, radiographic, and histopathological evaluation, is crucial for effective management. Surgical enucleation remains the treatment of choice, offering excellent prognosis with minimal risk of recurrence. This case highlights the need for clinicians to consider AOT in the differential diagnosis of anterior maxillary swellings in adolescents to ensure timely and appropriate intervention.

Declaration of Interest

The authors report no conflict of interest.

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