Adenomatoid Odontogenic Tumor Mimicking A Dentigerous Cyst - A Clinical, Radiological, and Histopathological Dilemma? - A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) is a very well-known and well acknowledged non-malignant, slow growing tumor developed from complex system of dental lamina or its remnants. It is also called as "the master of disguise" because of it different clinical and histopathological appearances. This benign tumor is divided into three variants of which the more common type is follicular type which is often mistaken for dentigerous cyst. AOT mainly presents radiologically as a unilocular cystic lesion surrounding the unerupted tooth, which is frequently misdiagnosed as a dentigerous cyst clinically. Here, we present a successful management of a case of AOT associated with the impacted teeth, mimicking, and misdiagnosed as a dentigerous cyst in the initial examination in a 10-year-old male, by surgical excision.

Keywords: Adenomatoid odontogenic tumor, Dentigerous cyst, Impacted canine, Anterior maxilla, Surgical excision.

Introduction

Adenomatoid odontogenic tumor (AOT) is defined as a benign tumor composed of odontogenic epithelium organized into different patterns of histopathological appearances and embedded in mature connective tissue. Although the 2005 WHO blue book on "Pathology and Genetics of Head and Neck Tumours" classifies AOT under the first category containing "tumors composed of odontogenic epithelium only," the prior WHO classification included the lesion in the second category of "tumors containing both odontogenic epithelium as well as ectomesenchyme with or without dental hard tissues.[1]"

AOT was first described by Ghosh in 1934 as an adamantinoma of the maxilla and was first identified as a separate entity by Stafne in 1948. It is also called as adenoameloblastoma, ameloblastic odontogenic tumor, odontogenic adenomatoid tumor, and cystic complex composite odontoma.

Philipsen *et al*. divided the AOT into three variants referred to as follicular, extrafollicular, and peripheral. The follicular and extrafollicular variants are the most common with 96% of all AOT and among these 71% are follicular variants. The peripheral variant is very rare. Only 18 cases reported so far. The follicular variant is mainly associated with the crown and often part of the root of an impacted tooth. The most commonly involved teeth are maxillary canines, next the permanent molars. Based on the clinical and radiographic examination, the follicular variant is mistaken and misdiagnosed as dentigerous cyst.[2]

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Case Report

A 10-year-old boy presented to the Department of Oral and Maxillofacial Surgery with a swelling on the left side of the face. The history reported by the father revealed that they first noticed the swelling 3 months back, it was gradual in onset. The swelling was progressively increasing in size. On general examination, patient was well built with all the vitals in normal range and stable.

On extra-oral examination, the swelling was located in the maxillary region on the left side lateral to the nose just below

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infra orbital margin, resulting in the facial asymmetry. The swelling was irregular in shape and measured approximately 4 cm × 3 cm, extending superiorly to the infra orbital margin, laterally to the zygomatic bone and inferiorly to the angle of the mouth. On palpation, the swelling was bony hard in consistency, non-tender, and immobile. No palpable lymph nodes in the neck region.

On intra-oral examination (Figure 1), the swelling was present in the buccal vestibule on the left side extending from the mesial surface of the 11 to the mesial surface of the 26, obliterating the buccal vestibule. The swelling was oval in shape measuring 4 cm × 3 cm, the overlying mucosa was inflamed. The deciduous teeth 62, 63, 64, 65, and 26 were present. On palpation, the surface of the swelling was smooth and edges were well defined. On aspiration, no fluid was seen.

On radiographic examination (Figure 2), OPG reveals an ill-defined radiolucent lesion extending from mesial of 11 to mesial of 26, located superiorly just below the infra-orbital margin. The pressure caused by the expansion of the lesion displaced the multiple impacted permanent teeth.

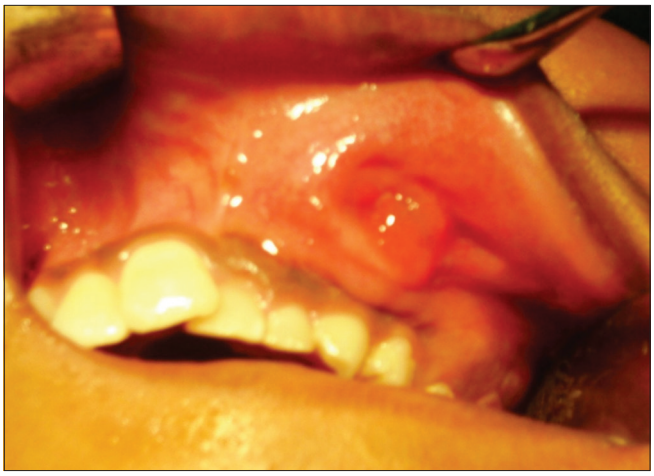

Figure 1: Intra-oral view of the swelling

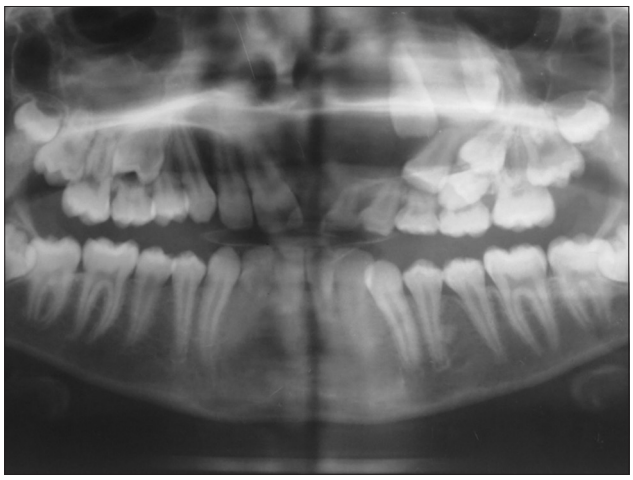

Figure 2: OPG

Based on clinical and radiographic evaluation, a provisional diagnosis of dentigerous cyst was made and conservative approach was planned considering the age of the patient. Swelling was marsupialized under local anesthesia. Part of cystic lining was sent for histopathological examination, it shows squamous epithelial lining in the connective tissue capsule which was suggestive of dentigerous cyst.

In the meanwhile, regular follow-ups were made and the lesion was evaluated periodically. Swelling persisted even after 1 month of the treatment and there was no eruption of the permanent teeth involved within the lesion.

Hence, the case was planned for excision of the lesion in toto under general anesthesia followed by histopathological examination. The lesion was approached intraorally. After reflection of the mucoperiosteal flap, the lesion was seen as the buccal cortical bone was resorbed. The entire lesion was seen as a separate encapsulated mass and removed in toto. The impacted teeth were seen within the mass of the lesion. There was no evidence of oronasal and oroantral communication and the palatal mucosa was intact. The wound was then sutured. The surgical specimen with impacted permanent teeth was smooth, round ball like, and measured approximately 24 mm × 21 mm × 10 mm (Figure 3) submitted for histopathological examination, it shows proliferation of polygonal and spindle shaped cells with connective tissue associated with duct-like and rosette-like structures (Figure 4).

Discussion

Following the initial report, even after a century, the AOT is still a widely researched tumor due to its unique biological nature. Cystic presentation of AOT has been reported way back in 1915 by Harbitz who reported the lesion as "cystic Adamantoma."[1] The bisected lesion sometimes shows mild cystic to complete cystic components. However, in pediatric population, occurrence of AOT developed in association with a dentigerous cyst in very few cases is described.[2]

AOT is also known as the "Two-third's tumor" because of its presence in maxilla in about 2/3rd cases, 2/3rd of its occurrence

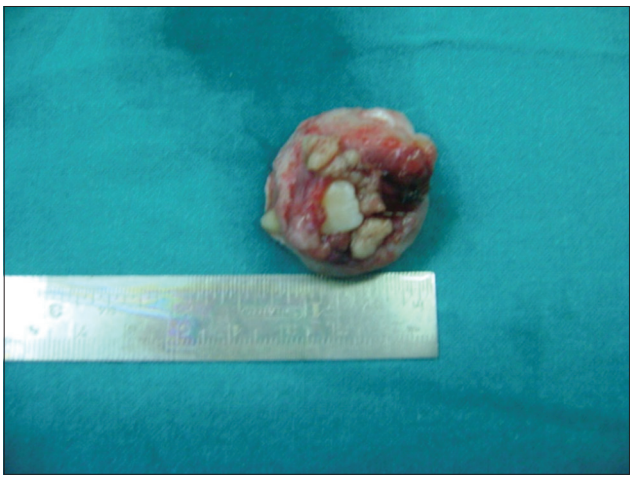

Figure 3: Excision of lesion

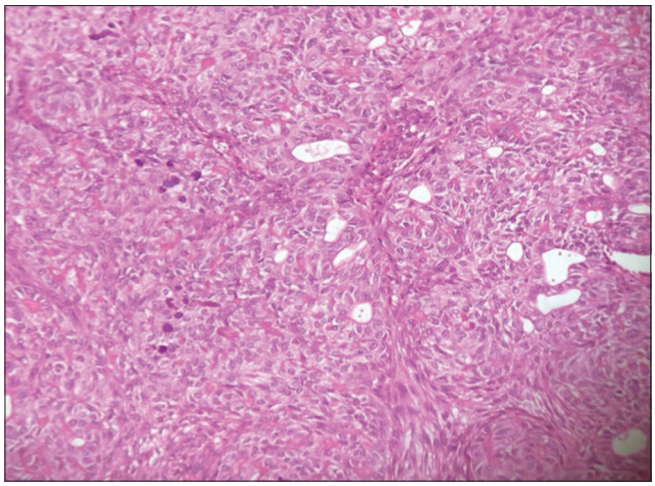

Figure 4: Excisional biopsy histopathological slide

is seen in young females, 2/3rd cases are associated with impacted teeth, and 2/3rd of the affected teeth are canines. Differential diagnosis includes dentigerous cysts, globulo-maxillary cysts, ameloblastomas, odontogenic keratocysts, calcifying odontogenic cysts, calcifying odontogenic tumors, and periapical disease.[3]

AOT is very well known for its different histopathological appearances and patterns.[4] It is a "perfect imitator of dentigerous cyst."[5] Occasionally, AOT develops as a mural growth in a dentigerous cyst. Santos *et al*. reported a case of AOT developed in the fibrous capsule of the dentigerous cyst.[6] Garcia-Pola *et al*. described the prolipheration of an adenomatoid odontogenic cyst in the epithelial border of a dentigerous cyst.[7] The systematic review of the literature of AOTs associated with or originating from an odontogenic cyst has been conducted by Gadewar *et al*. The cystic component of AOT has been variedly termed as dentigerous cyst, calcifying odontogenic cyst, or unilocular ameloblastoma.

The interest and relevance of the present case are the difficulty to diagnose accurately based on the radiograph and histopathology. The initial histopathological report in the present case stated findings suggestive of dentigerous cyst and later report suggested findings corresponding to those of AOT. Whether it was dentigerous cyst transforming to adenomatoid tumor or a cystic variant of AOT in the present case could not be stated as initially to preserve associated tooth only part of cystic lining was removed for histopathological evaluation.

As dentigerous cyst and AOT both are benign, slow growing, hamartomatous, non-invasive, and encapsulated lesions, conservative managements such as surgical enucleation or curettage are the treatment of choice.[8] From the literature, it is evident that none of the cases showed recurrence, suggesting that the tumor can be treated conservatively. We should be careful to follow-up cases after surgery.

Conclusion

In this present case report, AOT is misdiagnosed as dentigerous cyst clinically, radiologically, and histopathologically and in that scenario, even in pediatric population, few case reports of AOT arising from or associated with dentigerous cyst have been reported. Clinician should have detailed knowledge about the lesions with similar presentation. Expert clinical, radiological, and histopathological observations are required to achieve an accurate diagnosis. However, the present case highlights the importance of the fact that in cases of unilocular lesion surrounding the impacted tooth in the anterior maxillary region the treatment as per AOT should be considered.

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