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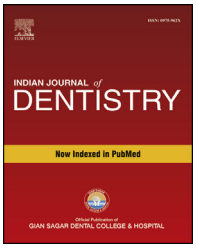

Case Report

Adenomatoid odontogenic tumor associated with a dentigerous cyst: A case report

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is an uncommon benign lesion of epithelial origin, which mainly affects young women and is usually found in the anterior maxilla. AOT is a slow-growing asymptomatic tumor which is commonly associated with an impacted tooth, often the canine. Treatment consists of conservative surgical enucleation and recurrence is rare. This paper reports a case of AOT associated with a dentigerous cyst (DC) located in the left maxilla of a 17-year-old girl.

1. Introduction

Odontogenic tumors comprise a heterogeneous group of lesions that develop in the gnathic bones and possess different histopathological characteristics and clinical manifestations.[1,2] The last classification of odontogenic tumors by the World Health Organization (2005) describes adenomatoid odontogenic tumor (AOT) as a benign neoplasia originating from the odontogenic epithelium, with a mature fibrous stroma and no ectomesenchymal component.[1–7]

AOTs account for 1–7% of all odontogenic tumors. The tumor is slow-growing and is usually associated with an impacted tooth.[1–16] There are three variants of AOT: follicular, extrafollicular, and peripheral. The follicular variant is the most common. Since this variant is associated with the crown

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of a tooth, it is often confused with a dentigerous cyst (DC). The extrafollicular variant develops without association with particular teeth and may resemble a lateral periodontal cyst. The peripheral variant is rare and generally appears as a swelling similar to gingival fibroma.[1,3,4,6,7,10,14–18]

The association of AOT with other odontogenic lesions, such as DC, has been described in the literature.[5,6,8,10,16,18–20] This study reports a case of AOT associated with a DC and describes its clinical, radiological and histological features.

2. Case report

A 17-year-old girl was seen at the Oral and Maxillofacial Surgery Service of the School of Dentistry, Federal University of Bahia, with an 8-month history of swelling over the upper left canine, which caused elevation of the nose wing (Fig. 1). The patient reported no pain or other associated symptoms and had no history of diseases.

Intraoral examination showed a firm swelling suggestive of expansion of the buccal cortical plate, in addition to clinical absence of the left maxillary canine (2.3) (Fig. 2). Panoramic radiography revealed a unilocular radiolucency in the left maxilla associated with the crown of an impacted canine, as well as the displacement of adjacent teeth (Fig. 3). The patient was submitted to local anesthesia and the lesion was completely enucleated. The impacted tooth was removed together with the lesion (Fig. 4).

The surgically removed fragment was sent to the Laboratory of Surgical Pathology for histopathological examination. Gross pathology revealed a globular cystic structure of dark brown color with a smooth surface, measuring 2.5 × 2.5 × 2.0 cm (Fig. 5). Histomorphological examination confirmed the cystic structure of the lesion consisting of a fibrous wall lined by squamous epithelium with few cell layers, as well as nodules of polyhedral cells amid small tubules of short columnar cells containing eosinophilic and mineralized material (Figs. 6 and 7). The histopathological diagnosis was AOT associated with a DC. After 17 months, the patient shows no clinical or imaging signs of recurrence.

Fig. 1 – Swelling in the region of the upper left canine and elevation of the nose wing.

Fig. 2 – Intraoral view showing swelling and absence of the left canine.

3. Discussion

Few studies focus on the association between DC and AOT.[5,6,8,10,16,18–20] According to Ide et al,[11] AOT can originate from the enamel organ, epithelial rests of Malassez, or remnants of the dental lamina. Furthermore, some authors proposed that this tumor arises from the epithelial lining of a cystic lesion, with the tumor mass protruding towards the lumen.[5,6,10,19] We report a case report of a follicular variant of AOT associated with a DC.

AOT is a noninvasive, slow-growing, progressive and asymptomatic epithelial odontogenic tumor, which occurs about twice as often in women as in men and normally in the second decade of life. This follicular variant is often diagnosed as DC because of clinical and radiological similarities.[2,4,10,18] These data are similar to the present study.

In a review of the English literature, we found six cases with a final diagnosis of AOT associated with a DC. Most cases were women and patient age ranged from 12 to 39 years, with a mean of 22.5 years. The most common location was the anterior maxilla, and the canine was involved in most cases. Tumor size ranged from 1.5 to 6.0 cm and conservative surgical enucleation was the treatment proposed in all cases (Table 1).

Fig. 3 – Panoramic radiograph showing a unilocular radiolucent image in the left maxilla.

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