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Adenomatoid odontogenic tumor: Clinical and radiological diagnostic challenges

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ARTICLE INFO

Keywords:
Adenoameloblastoma
Odontogenic tumors
Dentigerous cyst
Mandible
Follicular type

ABSTRACT

The adenomatoid odontogenic tumor (AOT) is a relatively uncommon odontogenic tumor with a relative frequency of 3–7%. The tumor is more common in females in their second decade of life. It exhibits a predilection for the maxillary anterior region. In this paper, we present a case of AOT in the mandible of a 27-year-old female. Histopathological examination of the tissue obtained from the initial incisional biopsy showed mostly an epithelial-lined cyst, which included a small nodule with the characteristic histological features of an AOT. Subsequent enucleation was done that confirmed the AOT diagnosis. The uncommon location of the lesion and the predominantly cystic configuration posed a clinical diagnostic challenge.

1. Introduction

The adenomatoid odontogenic tumor (AOT) is a benign slow growing odontogenic lesion. The World Health Organization (WHO) stated that the lesion arises from the odontogenic apparatus odontogenic epithelium with mature fibrous stroma without ectomesenchyme.[1] It represents 3–7% of all odontogenic tumors.[1] Previously, the AOT was considered to be a variant of the ameloblastoma.[2] Histologically, due to the presence of duct-like structures, it is also considered as an adenoameloblastoma.[3]

The literature review was done on documented cases of adenomatoid odontogenic tumor in the mandible. PubMed database was used to search published research reports using the terms ''Adenomatoid Odontogenic Tumor'' AND ''Mandible''. A total of 58 publications were identified from the search report (see Fig. 1). However, 23 publications were suitable for data collection based on inclusion and exclusion criteria. The inclusion criteria for selecting research papers were the following: full-text paper availability, written in English language, adequate and clear content on location, and diagnostic details of adenomatoid odontogenic tumor. The exclusion criteria were the following: incorrect web-link, title and/or abstract duplication, and general review papers. The data were analyzed for specific location in mandible (anterior or posterior), type of tumor (extrafollicular, follicular, or peripheral), information on unerupted/impacted tooth, clinical and radiological diagnosis, and concurrent odontogenic pathologies reported. Thirty-five papers were excluded due to no access to full-text of the research papers, and further two papers were excluded as the content did not match our search criteria (Table 1).

Twenty-one published reports constituted a total of 145 cases of mandibular adenomatoid odontogenic tumor; there were 89 cases of anterior mandible[4–16], 48 cases of posterior mandible[4,17–23], and 8 cases of mandibular adenomatoid odontogenic tumor that did not specify the exact location.[24]

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http://dx.doi.org/10.1016/j.jpfa.2017.06.001

Please cite this article in press as: Rajendra Santosh AB, et al. Adenomatoid odontogenic tumor: Clinical and radiological diagnostic challenges, *J Pierre Fauchard Acad (India Sect)*. (2017), http://dx.doi.org/10.1016/j.jpfa.2017.06.001

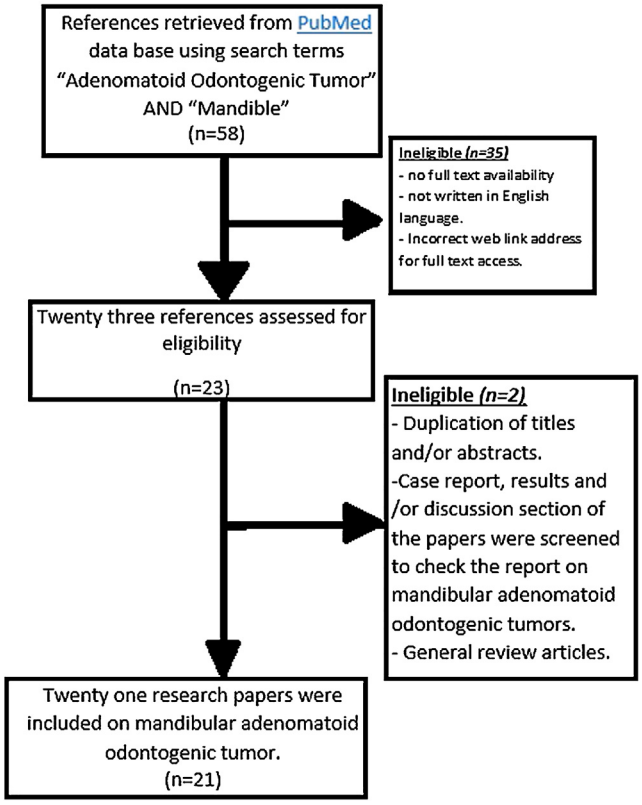

Fig. 1 – Flow chart showing the selection procedure of research papers.

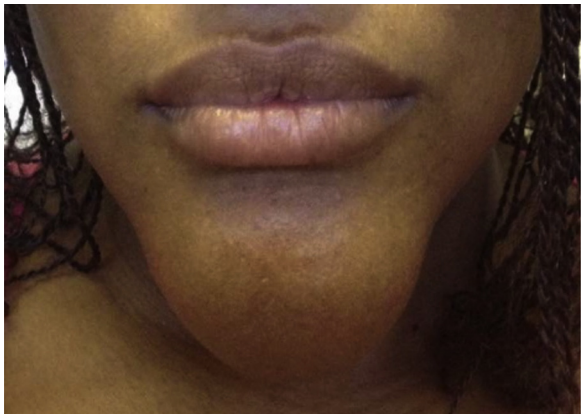

Fig. 2 – Swelling in the mandibular anterior region.

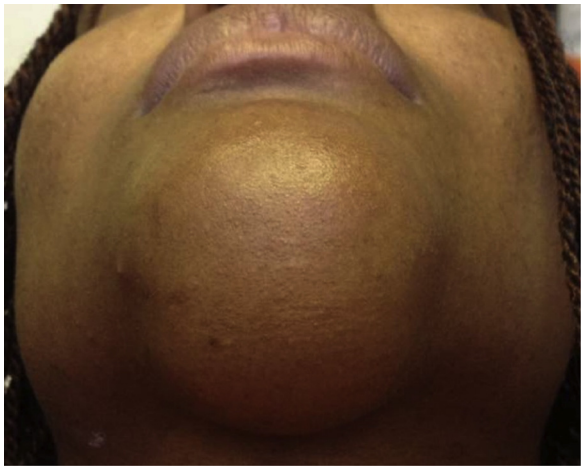

Fig. 3 – Extent of swelling in anterior mandibular region (submental view).

Seventy-four cases were follicular type,[4,5,8–10,15–19,22,23,25] 49 cases were extrafollicular[4,6,7,14,17,18,20,21], 3 cases were peripheral,[11,18] 11 cases were reported as intraosseous[13], and 8 cases did not provide details on type.[24] Four published reports mentioned an unerupted or impacted tooth (lateral incisor, canine, or premolar).[5,6,22,23] Fifteen published reports mentioned clinical and/or radiological diagnosis. The most common clinical and/or radiological diagnosis made was dentigerous cyst (5 cases).[5,9,15,17,23] Other diagnoses included adenomatoid odontogenic tumor (4 cases),[10,13,18,19] ameloblastoma (2 cases),[8,14] residual cyst (1 case),[21] odontogenic keratocyst (1 case),[7] gingival epulis (1 case),[11] and odontogenic cyst (1 case).[16] Three published papers reported concurrent odontogenic pathologies and that included dentigerous cyst (1 case),[22] odontoma (1 case),[20] and cemento-ossifying fibroma (1 case).[9]

In this paper, we present a case of AOT in an uncommon location and which histologically showed the presence of a significant amount of dentigerous cystic epithelium.

2. Case report

A 27-year-old female Jamaican patient of African descent visited a dental office with a chief complaint of swelling in the anterior mandibular region (Figs. 2 and 3) for the past 9 months. The swelling was asymptomatic and her oral hygiene was good. The intraoral examination revealed a nontender labial and lingual expansion of the mandibular anterior cortical plates, which were covered by normal appearing mucosa. The patient had no nerve deficit or adenopathy in the facial or cervical region. An orthopantomograph revealed the presence of a significantly large unilocular radiolucency with ill-defined borders involving the mandibular anterior region and encircling the unerupted left mandibular canine that had radiopaque areas of varying sizes and shapes that resemble calcified deposits. The unilocular radiolucent area was observed to extend from the left first premolar to the right first premolar area. Displacement of the left and right central, lateral incisors as well as retained left deciduous canine was seen. Root resorption of the mesial aspect of the apical root of the left first premolar was also noted (Fig. 4). Based on the clinical and radiological findings, the lesion was diagnosed as a dentigerous cyst with a differential diagnosis of ameloblastoma.

An initial incisional biopsy of the lesion was performed. Microscopically, the hematoxylin- and eosin-stained tissue of the initial biopsy demonstrated mostly a cyst, with a well-defined fibrous capsule and a lining mostly of thin non-keratinized stratified epithelium. A small lesion comprised of

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