

*Case Report*

Adenomatoid Odontogenic Cyst of Mandible: A Rare Case Report

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ABSTRACT

Adenomatoid odontogenic cyst (AOC) is a cyst that has a hamartomatous intraluminal proliferation of epithelial cells derived from Hertwig epithelial root sheath. Earlier it was classified as a tumour but on close inspection it will reveal its emergence from an epithelial lining. Therefore, the appropriate term is adenomatoid odontogenic cyst rather than adenomatoid odontogenic tumour. AOC accounts for about 1% to 9% of all odontogenic tumours. It is mainly found in young and female patients, located more often in the maxilla in most cases associated with a unerupted permanent tooth. AOC frequently resemble other odontogenic lesions such as ameloblastoma or dentigerous cysts. Surgical enucleation is treatment of choice. Here we present a rare case of an AOC in the mandible.

Key-words: Adenomatoid, Odontogenic cyst, Ameloblastoma, AOT, Adamantinoma.

INTRODUCTION

Adenomatoid odontogenic tumour (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.^[1] However, a variety of terms have been used to describe this tumour. Unal et al,^[2] produced a list containing all nomenclatures for AOT reported in the literatures. Many different names like adenoameloblastoma, Adamantinoma, epithelioma adamantinum or teratomatousodontoma have been used before to define the lesion currently called AOT.^[3] Today we recognise that AOT is not a tumour but a cyst that has a hamartomatous intraluminal proliferation of epithelial cells derived from Hertwig epithelial root sheath. Earlier it was classified as a tumour but on close

inspection it will reveal its emergence from an epithelial lining. Therefore, the appropriate term is adenomatoid odontogenic cyst (AOC)^[4] rather than adenomatoid odontogenic tumour.

Clinical features mainly include a missing tooth. The lesion usually present as an asymptomatic swelling which is slowly growing and often associated with an unerupted tooth.^[5,6] Unerupted permanent canine are the most often involved in AOCs. Radiographically AOC shows resemblance with other odontogenic lesions such as calcifying odontogenic tumours, dentigerous cysts, ameloblastoma, odontogenic keratocysts, calcifying odontogenic cysts, globulo-maxillary cysts and periapical disease.^[7,8]

On the other hand, the follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth, the radiolucency of the extra follicular type is located between, above or superimposed upon the roots of erupted permanent teeth. Cyst expansion may cause displacement of adjacent teeth much more commonly than root resorption. The peripheral lesions may cause erosions of the adjacent cortical bone. [8]

CASE HISTORY

A 15 year old female patient reporting to the Outpatient department of Oral and Maxillofacial surgery with chief complaints of swelling at right mandibular anterior region since two months . Patient was apparently asymptomatic two months back when she develops swelling at right mandibular anterior region, which was non-tender in nature. Swelling was gradually increasing in nature as time passes. After that she reported to some local practitioner and took some medication. But the swelling was still there and was not subsiding. In the meantime, she did not take any medication. After that she reported to us. Beside an uneventful medical history the patient presented no conspicuous intraoral clinical findings except the absence of the tooth 44 with non tender bony hard swelling from 43 to 45, obliterated labial vestibule due to expansion of the buccal cortical plate (Figure 1).The size of the swelling was 4.0 cm x 2 cm in diameter. The shape was ovoid, the colour of the overlying skin of the lesion was normal, the consistency was soft to firm, texture was smooth in nature and there was slight depressibility.

INVESTIGATION

Radiographs

Panoramic radiograph of the patient revealed a well defined circumscribed, corticated unilocular expansile radiolucent lesion extending from 43 to 45 with an impacted 44 which is displaced bucco-inferiorly toward the lower border of the mandible (Figure 2). The adjoining teeth show inclination. The expansion of buccal cortical plate is noted with no discontinuity. The lesion has radiopaque flecks of calcified material. No resorption of the root apices was observed.

Routine examinations

Blood investigation was within normal limits except the count of lymphocytes were increased. Aspiration was negative.

Management

Surgical excision of the lesion was done under general anaesthesia .A trapezoidal incision was given around the defect starting from distal to right mandibular canine up to the mandibular right second premolar. Blunt dissection was done, then lesion was exposed and enucleated with the help of periosteal elevator along with extraction of mandibular second premolar after that sling sutures was given for clavicular incision and interrupted sutures were given for releasing incision (Figure 4)(Figure 5) . Specimen was sent to the department of pathology for histopathological examination (Figure 6). Sutures were removed on 5th post op day and following periods were uneventful (Figure 7).

Histopathological examination shows there were scanty of polyhedral ' spindle shaped epithelial cells of columnar and cuboidal variety arranged in duct like fashion, the epithelial cell lining of the duct resemble the cells of the inner enamel epithelium that may indicate that these cells may develop through the preameloblast stage of the stellate reticulum and the stratum intermedium (Figure 3).

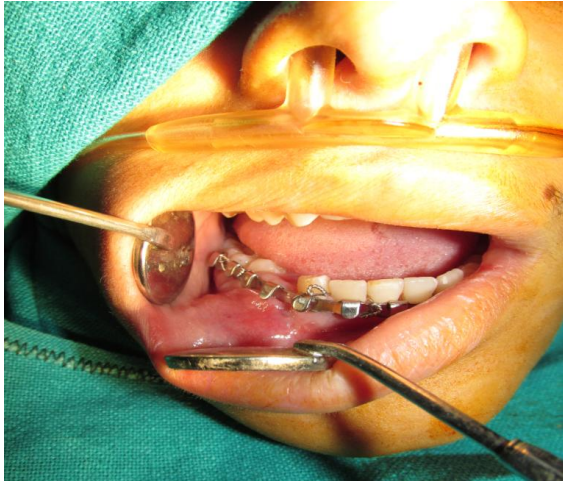


Figure 1 . Intraoral view shows there is uniform swelling with inflammation of the buccal mucosa

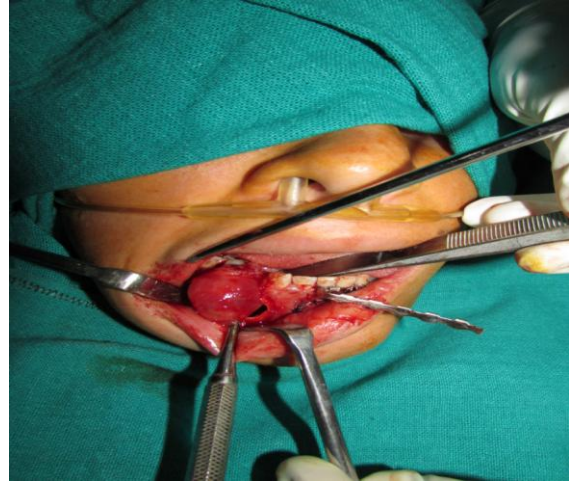


Figure 4. Enucleation of the lesion



Figure 2. Orthopantomogram shows well defined circumscribed, unilocular expansile lesion

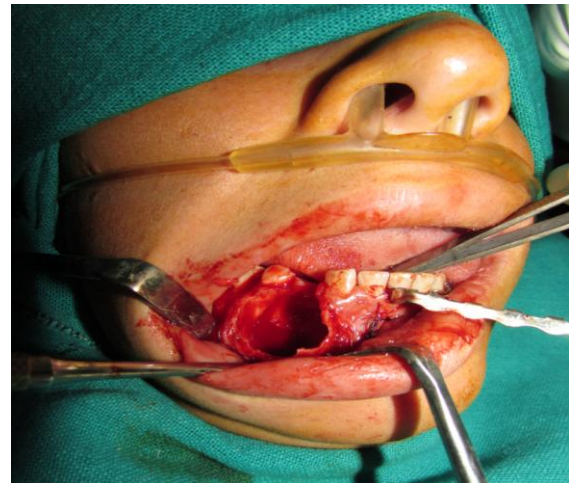


Figure 5. Host bed immediate after the Enucleation

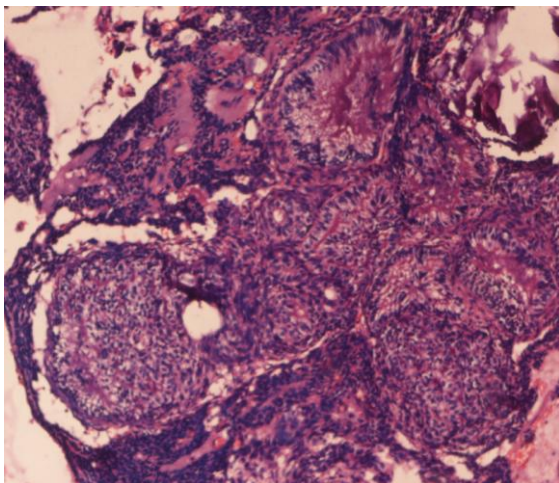


Figure 3. Shows scanty of polyhedral spindle shaped epithelial cells



Figure 6. Resected specimen



Figure 7. Healed lesion after 3 weeks

DISCUSSION

In 1999 Philipsen and Reichart,^[9] presented a review based on reports published until 1997 which showed some interesting aspects regarding epidemiological figures of this tumour. Since then numerous case reports of AOT have been published. From the early 1990s onwards 65 single cases of AOT (excluding case series of more than 10 cases) have been published. The mean age was 13.2 years (range 3 until 28 years) and the female: male ratio was 2.3: 1. The AOT was predominantly found in the upper jaw (maxilla: mandible = 2.6: 1). Regarding the various case series published in the literature and comparing these data with the single case reports mentioned above, it has to be reasoned that the AOT has a prevalence of odontogenic tumours between 1.2% in Caucasian^[10] and 9% in black African patients.^[11] The tumour is most often diagnosed in the second decade of life and women are about twice as many affected than men. The AOT is over two times more located in the maxilla than in the mandible and the anterior jaw is more affected than the posterior area. According to Philipsen and Reichart,^[9] the AOT appears in three clinico-topographic variants: follicular, extra follicular and peripheral. The follicular and

extra follicular variants are both intra bony and account for approximately 96% of all AOTs of which 71% are of follicular type. Dare et al,^[12] found that intraoral periapical radio graphs allow perception of the radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcifies deposits while panoramic often do not. Those calcified deposits are seen in approximately 78% of AOT. In addition, in one recently reported case MRI was useful to distinguish AOC from other lesions, even if it is difficult on periapical ordinal radiographies.^[8,13]

AOC is a slow-growing lesion usually occurring in the anterior maxilla, rarely reported in the mandible. It has a female predilection. Usually do not exceed 1 to 3 cm, in the greatest diameter. The lesion is asymptomatic but may cause pain due to cortical expansion which may be contributory to the diagnosis. AOC is usually associated with an impacted tooth which is enclosed by the lesion with displacement of adjacent teeth similar to the case discussed above. Root resorption is rare.

AOC should be included in the differential diagnosis of corticated radiolucency with small radiopaque foci, especially in teenagers and young adults associated with an impacted tooth. If there are no flecks of radiopacities then it may be a dentigerous cyst. These radiopaque foci are clearly visible in an intraoral periapical radiograph than in an Orthopantomograph. However, an AOC envelops crown as well as the root whereas the dentigerous cyst envelops only the crown and is attached to it. Microscopically the cyst is well-circumscribed, encapsulated mass. It is epithelial in nature with rosette of cells. It has eosinophilic coagulum with calcified amorphous material called tumour droplets. Varying amounts of calcifications can also occur. These features are also contributory to the diagnosis of present case. The origin

of AOC is controversial. The dental lamina remnants are likely to represent the progenitor cells as it not only arises from anterior maxilla but also in the angle and anterior mandible. According to the hypothesis, it grows next to or in to a nearby dental follicle leading to envelopmental theory. In the case reported here the lesion surrounding a fully formed first premolar, suggesting envelopmental pathogenesis. [14,15] The lesion is encapsulated and hence recurrence is very rare and simple curettage with enucleation is sufficient to treat the case. But, regular follow-up is necessary. Only three cases in Japanese patients are reported in which the recurrence of this cyst occurred. [16] Therefore, the prognosis is excellent.

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