Adenomatoid Odontogenic Tumor of the Mandible

Kailasam Subramaniam, Manoj Kumar Sachidhanandan, Subha Manoharan, Malarvizhi Elangovan

ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a benign odontogenic tumor, which presents as a noninvasive lesion of the jaws with slow but progressive growth. AOT is often misdiagnosed as an odontogenic cyst. It accounts for about 2.2 to 7.1% of all odontogenic tumors. It is predominantly seen in young females, in the anterior maxilla, associated with unerupted canines. This report describes a rare case of AOT in the mandible. This paper highlights the controversies regarding its origin and management in the light of recent findings.

Keywords: Adenomatoid odontogenic tumor, Adenoameloblastoma, Ameloblastic adenomatoid tumor, Adamantinoma.


Source of support: Nil
Conflict of interest: None declared

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905.1 Unal et al2 produced a list of nomenclature for AOT reported in literature, they are adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium or teratomatous odontoma and is currently called as AOT. Dreiblingtin in 1907 described it as a pseudoameloblastoma.3 Harbitz in 1915,4 reported it as cystic adamantoma. Ghosh in 1934 described it as adamantinoma and was recognized by Stapne in 1948 as a distinct pathological entity. Finally in 1969 Philipsen and Birn5 proposed the term AOT and was adopted by WHO in 1971.5

According to the second edition of the World Health Organization journal ‘Histological Typing of Odontogenic Tumors’, AOT is defined as ‘A tumor of odontogenic epithelium with duct-like structures and varying degrees of inductive change in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst.6

It is an uncommon odontogenic tumor with frequency 2.2 to 7.1%.7 It appears as an intraoral and extraoral swelling. It is referred as ‘Two-third tumor’ because it occurs in maxilla in about 2/3 cases, about 2/3 in young females, 2/3 cases are associated with unerupted tooth and 2/3 affected teeth are canines.5

This report describes an AOT in the mandible, illustrate the clinical, radiological and microscopic features.

CASE REPORT

A 22-year-old female patient came to the Ragas Dental College and Hospital with the chief complaint of a swelling in the lower front teeth region since 2 months (Fig. 1). She noticed a swelling in the lower chin region 2 months back and it’s the same since then. She has no relevant medical or dental history except for pain on pressure over the swelling.

On examination, there was a single swelling in the chin region, measuring about 3 × 4 cm, ovoid in shape with smooth surface, extending from the midline 3 to 4 cm posteriorly and 5 mm from the corner of the mouth till 1 cm beyond the inferior border of the mandible supero-inferiorly with mild facial asymmetry due to the swelling in the right side chin region. On palpation, it was bony hard, well defined and tender.

Intraorally, 32 was missing and there was a swelling involving the buccal and lingual cortical plates extending from 33 to 43 region buccally and 33 to 42 region lingually. On palpation, it was bony hard except in relation to 43 region where it was yielding, well defined and tender on palpation (Fig. 2). All the teeth in the region of the lesion were vital. Aspiration was negative.

Considering the clinical features, the lesion was provisionally diagnosed as a dentigerous cyst and probable differential diagnosis can be any benign odontogenic tumor. Intraoral periapical radiograph revealed horizontally impacted 32, surrounded by a well-defined radiolucency and floccular radiopacities around the crown of the impacted 32 (Figs 3 and 4), displacement and divergence of roots of central incisor and canine. Occlusal radiograph reveals a radiolucency extending from 33 to 44 region along with an
Adenomatoid Odontogenic Tumor of the Mandible

irregular lingual bone expansion (Fig. 5), it shows region crossing midline and cyst do not. Lingual bone is thinned. Orthopantomogram revealed an single unilocular, well defined radiolucency on the midline of the mandible with hyperostotic border, measuring 5 × 3 cm, oval in shape, extending mediolaterally from apical region of 44 to 33 and superoinferiorly 0.5 cm from crest of the alveolar bone till above the inferior border of the mandible. The radiolucency completely encloses the tooth including root. The internal structure revealed presence of radiopaque structure morphologically similar to that of a tooth extending from center of radiolucency obliquely toward the border, presence of a floccular radiopacity around the crown of the tooth structure. There is external resorpsion in relation to 44, 43, 42, 41, 31 with distal displacement of the roots (Fig. 6).

With the evidences of the radiographs, the provisional diagnosis can be modified to an adenomatoid odontogenic tumor or a calcifying epithelial odontogenic cyst. Hematological, biochemical and urine analysis were done to rule out any systemic causes and all were normal.

Computed tomography revealed presence of a destructive lesion in the anterior mandible showing bicortical expansion with perforation of bone in both labial and lingual surface with an impacted tooth within the lesion along with numerous radiopaque flecks (Fig. 7).

Histopathological examination revealed polygonal cells with focal areas of eosinophilic coagulum, hematoxyphilic globular calcifications and hematoxyphilic irregular calcifications. The polygonal cells show tightly packed whorled arrangement. The fibrous connective tissue exhibits cords, islands and sheets of polygonal cells (Fig. 8).

After all the required investigations were done an intentional root canal therapy for the associated teeth was planned followed by curettage and enucleation of the lesion (Figs 9 and 10). Histopathological examination of the enucleated lesion confirmed the diagnosis of AOT.

DISCUSSION

Adenomatoid odontogenic tumor is a slow-growing lesion usually occurring in the anterior maxilla, rarely reported in the mandible. The lesion has a female predilection. Usually do not exceed 1 to 3 cm, in the greatest diameter but can be larger also. The lesion is asymptomatic but may result in
pain due to cortical expansion which is contributory to the diagnosis. Usually, AOT is associated with an impacted tooth which is enclosed by the lesion with displacement of adjacent teeth which is similar to the case discussed above. Root resorption is rare.\textsuperscript{8,9}

A diagnosis of adenomatoid odontogenic tumor should be considered 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 (Figs 3 and 4) than in a orthopantomograph (Fig. 6). However, an AOT envelops crown as well as the root whereas the dentigerous cyst envelops only the crown and is attached to it. Microscopically the tumor is well-circumscribed, encapsulated mass. It is epithelial in nature with rosette of cells. It has eosinophilic coagulum with calcified amorphous material called tumor droplets. Varying amounts of calcifications can also occur.\textsuperscript{10-13} These features are also contributory to the diagnosis of present case.

The origin of adenomatoid odontogenic tumor is controversial. The dental laminar 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, the lesion grows next to or in to a nearby dental follicle leading to envelopmental theory.\textsuperscript{14} In the case reported here the lesion surrounded a fully formed lateral incisor, suggesting envelopmental pathogenesis.

Immunohistochemistry of AOT expresses keratin and vimentin in the tumor cells at the periphery of ductal and whorled structure. Amelogenin and enamelin in small mineralized foci are found in tumor cells and in hyaline deposits.\textsuperscript{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.

For this patient intentional root canal therapy is done in relation to 33 to 44 followed by surgical curettage and enucleation. Postoperative follow-up after 2 months revealed no signs of recurrence (Figs 11 and 12).

**CONCLUSION**

AOT in mandible is very rare, characteristic features of this case helps in diagnosing. Careful diagnosis and adequate interpretation of the clinical and radiographical findings may be helpful in arriving at a correct diagnosis.

ABOUT THE AUTHORS

Kailasam Subramaniam (Corresponding Author)
Professor and Head, Department of Oral Medicine, Diagnosis and Radiology, Ragas Dental College, Chennai, Tamil Nadu, India
e-mail: dr_kailasam@hotmail.com

Manoj Kumar Sachidhanandan
Professor, Department of Oral Medicine, Diagnosis and Radiology, Ragas Dental College, Chennai, Tamil Nadu, India

Subha Manoharan
Senior Lecturer, Department of Oral Medicine, Diagnosis and Radiology, Ragas Dental College, Chennai, Tamil Nadu, India

Malarvizhi Elangovan
Postgraduate Student, Department of Oral Medicine, Diagnosis and Radiology, Ragas Dental College, Chennai, Tamil Nadu, India

REFERENCES