Annals of Clinical Case Reports

Case Report
Published: 10 Jan, 2025

A Rare Case of an Unusually Large Adenomatoid Odontogenic Tumor in the Mandible

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Abstract

Adenomatoid odontogenic tumor is a benign tumor having an odontogenic origin specifically from the enamel organ, reduced enamel epithelium and dental lamina. Steensland initially identified the adenomatoid odontogenic tumor in 1905. It is a benign epithelial neoplasm that is very uncommon, distinct, slow-growing, and odontogenic.

Keywords: Adenomatoid odontogenic tumor; Tumours; Pseudoadenoameloblastoma

Introduction

Adenomatoid odontogenic tumor is a benign tumor having an odontogenic origin specifically from the enamel organ, reduced enamel epithelium and dental lamina. Steensland initially identified the adenomatoid odontogenic tumor in 1905 [1]. It is a benign epithelial neoplasm that is very uncommon, distinct, slow-growing, and odontogenic [1]. Driblet referred to it as a pseudoadenoameloblastoma in 1907 [2] Stafne referred to it as "an epithelial tumor" in 1948 and regarded it as a separate entity. It was classified as an adenoameloblastoma by Thoma in 1955 [3].

In 1969, Philipsen and Birn introduced the term Adenomatoid Odontogenic Tumor (AOT) claiming that due to its distinct characteristics, it should not be considered a variant of ameloblastoma [4].

The incidence of AOT is 3% of all the other odontogenic tumours [5]. Adenomatoid Odontogenic Tumour (AOT) is an odontogenic tumour with a prevalence of 2.2% to 7.1%. AOT is a benign, non-invasive, and progressive lesion which is also known as “a two third tumour” [6] As the name suggests the tumour occurs in the maxilla in two third of cases. It occurs in young patients in two third of cases and associated with missing or unerupted teeth in two third of cases. Two third cases are associated with the maxillary canine. Age range of AOT is from 3 to 82 years old, the majority of these tumours are diagnosed during the teenage years, with over half of cases occurring between 13 and 19 years of age [7].

AOT is considered as a hamartoma. The limited size of AOT, attributed to its minimal growth, along with lack of recurrence even after incomplete removal, reinforces this perspective [7,8]. Experts consider AOT to be a benign tumour that is nonaggressive and non-invasive. They suggest that generally it is in small size of <3 cm., often identified through routine dental X-rays, leading to prompt removal before the tumour grows large enough to cause noticeable clinical symptoms. Additional support comes from the microscopic features of the lesional tissue that show greater variation from the arrangement of the normal odontogenic apparatus than should be expected in a developmental anomaly [8].

We report an unusually large follicular type of AOT in the mandible in an elderly male highlighting clinical, morphological and biological characteristics.

Case Presentation

A 75-year-old male patient reported to the Department of Oral medicine and Radiology of SDM college of Dental sciences and Hospital with the chief complaint of swelling on the right lower jaw region since 4 years. Initially the swelling was in the size of a peanut and gradually progressed

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Received Date: *08 Dec 2024*
Accepted Date: *31 Dec 2024*
Published Date: *10 Jan 2025*

Citation:
Ann Joshep R, Chopra I, Shekar B, Kumar K, Naikmasur V, Anehosur V. A Rare Case of an Unusually Large Adenomatoid Odontogenic Tumor in the Mandible. Ann Clin Case Rep. 2025; 10: 2711.

ISSN: 2474-1655.

Figure 1: Extraoral appearance of the swelling at the right lower jaw.

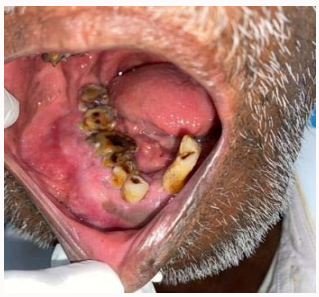

Figure 2: Intraorally the swelling extending from 43 to 48 region.

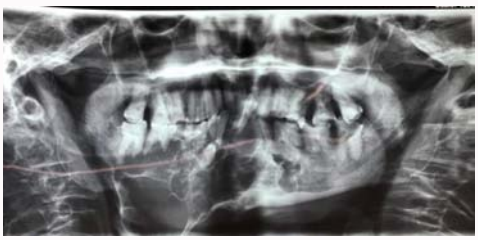

Figure 3: Orthopantomogram depicting the extension of the tumor from 36-48.

to the present size. Patient also complains of numbness in the lower border of jaw since 2 years. The past dental and medical history were unremarkable.

On extraoral examination, a large swelling measuring 7 cm × 8 cm was noted on the right lower 1/3rd of the face extending superior inferiorly – from the ala tragus line to the lower border of the mandible and beyond. Antero posteriorly from right Para symphysis to angle of the mandible (Figure 1).

Boders were well defined with overlining skin intact. On palpation it was non tender and bony hard in consistency.

On intraoral inspection, there was generalized severe attrition. A diffuse swelling was noted extending antero-posteriorly from 43-48 region and superior-inferiorly from the free gingival margin to the lower buccal vestibule (Figure 2). On palpation the swelling was non tender and hard in consistency. Orthopantomogram revealed a well-defined multilocular radiolucent lesion extending from the distal aspect of 48 to 36 region crossing the midline. External root resorption was noted with 44,45,47 and 48 (Figure 3) 31,32,34,41 were missing. The lesion was predominantly radiolucent with large locules separated by thin septations. There was impacted 43. Supra eruption and drifting were noted w.r.t 42. There was thinning and

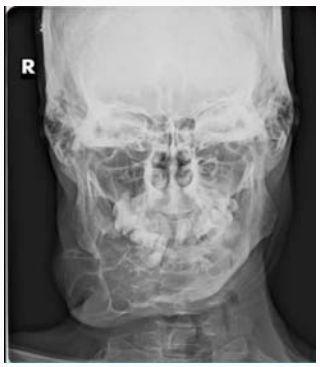

Figure 4: Posterior Anterior view of Mandible.

expansion of lower border of mandible.

Postero-anterior view of mandible revealed a well-defined multilocular radiolucency extending from the left para symphysis to the right retromolar region crossing the midline. Expansion of buccal cortical plate was noted. Thinning of inferior cortex noted with no pathological fracture (Figure 4). With these clinical and radiologic findings, a provisional diagnosis of metacystic ameloblastoma was given and differential diagnoses of odontogenic myxoma and Calcifying Epithelial Odontogenic tumor were considered.

Fine needle aspiration cytology was performed and the results were inconclusive. Incisional biopsy was done and microscopic examination revealed cuboidal and columnar cells with minimal differentiation of stellate reticulum. Tumor also exhibits areas of glandular like differentiation with pseudocyst formation lined by cuboidal cells. The histopathological diagnosis was given as Ameloblastoma with glandular differentiation.

Based on the biopsy report, the tumour resection was planned and procedures were as follows; the tumour extension was marked with 1cm clearance of bony margin. Vestibular incision was made intraorally extending up to last molar bilaterally and to avoid the lip split. The tumour and lower border of mandible was exposed up to right angle of mandible bilaterally. Tumour was resected along with resection of mandible extending from the 47 to 37 regions along with right submandibular gland and associated lymph nodes and sent for histopathological analysis. The lesion measured 7 cm × 10 cm in dimensions ad weighed 250 gms.

Histopathology

Hematoxylin Eosin-stained sections showed tumor composed of odontogenic epithelium and connective tissue areas covered by well-defined thick fibrous capsule. The epithelial cells were polygonal or spindle or cuboidal shaped forming nests, interconnecting cords,

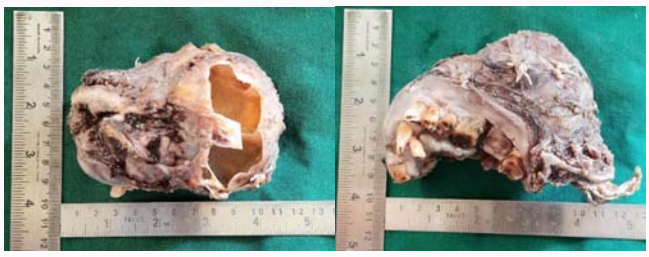

Figure 5: Specimen received after surgical resection measuring 9 cm × 10 cm in diameter with embedded right mandibular canine.

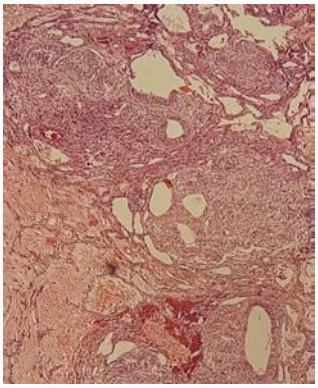

Figure 6: The tumor composed of odontogenic epithelial cells forming nests, interconnecting cords, whorls, rosette-like structures and characteristic duct-like structures (H & E stain, 5X magnification).

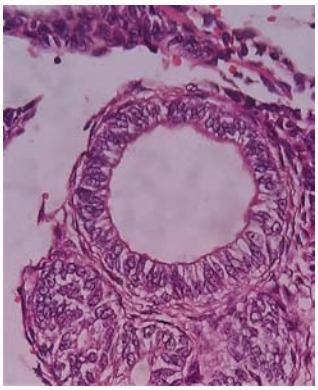

Figure 7: Characteristic duct-like structures lined by single layer of cuboidal to columnar cells similar to promyeloblasts with nuclei placed away from the lumen. The inner surface of the lumen was lined by homogeneous eosinophilic material (Arrow) (hyaline ring,) (H & E stain, 40X magnification).

whorls, rosette-like structures with focal areas eosinophilic hyaline material or empty duct-like structures (Figures 5,6). Characteristic duct-like structures were seen lined by single layer of cuboidal to columnar cells similar to promyeloblasts with nuclei placed away from the lumen. The inner surface of the lumen was lined by homogeneous eosinophilic material (hyaline ring) (Figure 7). The epithelial cells have hyperchromatic nucleus & rare mitosis. The Connective tissue was fibro cellular with areas of hyalinization and myxoid tissue. Moderate chronic inflammatory cell infiltration mainly lymphocytes and marked vascularity seen. The lymph nodes and submandibular salivary gland were negative for tumor invasion and immunohistochemical staining showed membranous and cytoplasmic staining of odontogenic epithelial cells with pan cytokeratin (CKAE1/AE3) (Figure 8). Ki67 immunostaining showed low proliferative index.

Based on the histopathological and immunohistochemistry, a final diagnosis of Adenomatoid odontogenic tumor was given. Patient was followed up post operatively healing was satisfactory (Figure 9). No dehiscence was noted during first and third month follow up. Patient was kept under close observation to monitor for any signs of recurrence.

Discussion

AOT is the 5th common odontogenic tumor, making up between

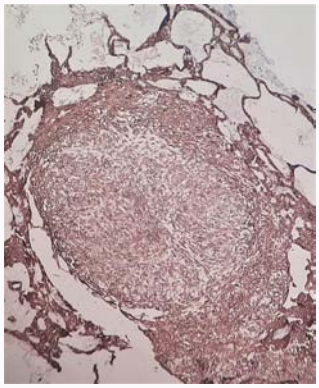

Figure 8: Immunohistochemical staining showing membranous and cytoplasmic staining of odontogenic epithelial cells with pancytokeratin (CKAE1/AE3) (IHC stain, 40X magnification).

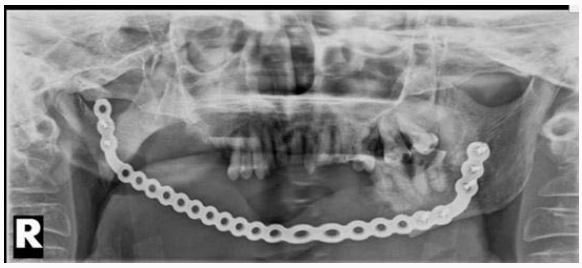

Figure 9: Postoperative orthopantamograph.

2.2% and 7.1% of all odontogenic tumors. AOT comes in three different forms: the central lesion of the follicular type (73% of cases), which is connected to an impacted tooth; the central lesion of the extrafollicular type (24% of cases), which is unrelated to an impacted tooth; and the peripheral variety seen in extraosseous sites (3% of instances) [9]. A corticated radiolucency is produced by both varieties of central intraosseous tumors, occasionally with radiopaque specks. The extrafollicular type presents as a unilocular, well-defined radiolucency found between, above, or superimposed on the roots of erupted teeth and often resembling a residual, radicular or lateral periodontal cyst. The peripheral type usually presents as a gingival swelling, located palatially or lingually relative to the involved tooth [10]. Hence this case is classified as the follicular type of AOT [10].

The most common site of AOT is anterior maxilla, it can be also seen in mandible but it is an infrequent site and commonly seen in second decade of life. Adenomatoid Odontogenic Tumour (AOT) is typically found in the maxilla in about two-thirds of cases, predominantly affects younger patients in two-third of cases, and is often associated with missing or unerupted teeth in two third of cases, hence termed as a "two-thirds tumour. “Our case differs significantly from this criterion. In reports by Jorg et al. S. P. S. Sodhi et al. and Neeraj sham et al., the tumours were located in the mandible and associated with impacted teeth; however, all of these cases involved younger patients and smaller tumour sized. Whereas in the present case an unusually large AOT was identified in the mandible and in an older age group [11-14].

AOTs are typically small, usually measuring between 1 and 3 cm in diameter. However, in a case report by Nguyen Quang Duc et al. a presentation of unusually large AOT in lower 3rd of right face was reported in 24-year-old female patient. The size of the swelling

was approximately 22 cm × 25 cm × 17 cm which is larger than the present case [15]. In a case series by Chandramani et al all the four cases had large tumour size of more than 3 cm. These reports illustrate a tendency for AOT to have unusually large diameter.as noted in our case [16].

Radiologically, AOT typically appears as a unilocular osteolytic lesion with well-defined borders, often the lesion is usually associated with an unerupted tooth and is seen as predominantly radiolucent with small radio-opacities, which can serve as a valuable diagnostic indicator. but cases are reported without radio-opacities as in our case [15]. In the case reported by Veena S et al. the lesion had some unusual manifestations, which included its large size, multilocularity, lingual cortical expansion, root resorption, and cortical perforation which is similar to the present case [16]. Dense septate were prominently observed in the tumour, representing an atypical characteristic in an AOT. A case report by Bhatke et al. details a multilocular radiolucency linked to AOT in a 14-year-old female patient which aligns with our case [17]. Literature provides evidence that AOTs) can present as multilocular spectated lesions, accompanied by root resorption and thinning of the cortical plate, as seen in the current case.

The present case showed all classical histological features of AOT. As all variants of AOT usually shows consistent histological features like solid nests of polyhedral or columnar to cuboidal shaped epithelial cells or rosette-like structures with minimal connective tissue stroma. The amorphous eosinophilic material in between the nests and rosette are considered to be the tumour deposits or some form of enamel matrix (Figure 6A, 6B) [18,19]. As WHO has described the distinct histopathological characteristic of duct-like structures were also present in our case (Figure 7). Another feature of dysplastic dentinoid material or osteodentin as a questionable inductive phenomenon of AOTs is generally seen was not present in our case. The previous immunohistochemical investigations have showed positive staining for CK5, CK17, and CK19 similar to cytokeratin profile of follicular cyst, or oral or gingival epithelium [20]. Crivelini and co-workers have detected AOT positivity for CK14 indicating REE (reduced dental epithelium) is the tumour origin [21]. Few investigators have showed co expression of cytokeratin, vimentin and smooth muscle action pointing out the neoplastic nature of AOT [22-24]. The proliferation rate of AOT, in terms of Ki-67-positive tumour cells, is low in general as reported by few authors [25-27]. The present case showed positivity for pan cytokeratin and low Ki67 staining suggestive of odontogenic epithelial tumour of benign character (low growth potential). All variants of AOT histologically show benign features and almost all are encapsulated having non aggressive biological behaviour. In our case, the histological features were consistent with the other cases reported except the size, mandibular location and age. However, there was no evidence of high growth potential leading to large size histologically or immunohistochemically. Large size of the present case may be due to the long duration [28].

AOTs are frequently misdiagnosed as ameloblastomas due to histologic similarities, as was the case with our incisional biopsy report. Because of the small size of the tissue in an incisional biopsy, the quantity of one histopathological component overshadows the quantity of another, such as the areas of loosely arranged spindle-shaped cells in the psedorosette that appeared as stellate reticulum and duct-like areas that seemed as glandular differentiation Keshwar S, et al. In odontogenic tumors, it is always recommended to confirm the incisional biopsy findings with an excisional biopsy.

Conclusion

- This case suggests that an Adenomatoid Odontogenic Tumour (AOT) can mimic an Ameloblastoma with regard to its location, size, and clinical characteristics.

- This case report highlights the necessity of considering the follicular type of Adenomatoid Odontogenic Tumour (AOT) in the differential diagnosis when faced with a large, aggressively expanding lesion in the mandibular posterior region.

Accurate diagnosis of such rapidly growing lesions can be significantly aided by this consideration.

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