Scientific Research Publishing

Open Journal of Stomatology, 2025, 15(5), 69-75
https://www.scirp.org/journal/ojst
ISSN Online: 2160-8717
ISSN Print: 2160-8709

Adenomatoid Odontogenic Tumor Occurring on the Mandible

—Review of the Literature and Report of a Rare Case

Tom Osundwa[1,2], Elizabeth Dimba[1], Maryam Idris[1]

[1]The University of Nairobi Dental Hospital, Nairobi, Kenya
[2]Gertrude's Children's Hospital, Nairobi, Kenya
Email: osundwa69@gmail.com, elizabeth.dimba@uonbi.ac.ke

How to cite this paper: Osundwa, T., Dimba, E. and Idris, M. (2025) Adenomatoid Odontogenic Tumor Occurring on the Mandible. *Open Journal of Stomatology*, **15**, 69-75.
https://doi.org/10.4236/ojst.2025.155006

Received: March 29, 2025
Accepted: May 13, 2025
Published: May 16, 2025

Abstract

Adenomatoid Odontogenic Tumor (AOT) is a rare benign odontogenic tumor of the jaw. It occurs more frequently in females around the second decade, in the anterior maxilla, and with an impacted canine. We report a case of AOT in a 16-year-old female in the mandible associated with the premolar on the right side. The tumor was spherical, depicting its centrifugal growth pattern and extrafollicular based on its relation with the affected first premolar. The differential diagnosis of AOT includes Odontogenic Keratocyst, Ameloblastoma and Dentigerous cysts. Clinical-radiologic and histologic data, when integrated, remain the best diagnostic tool. The need to report more on this entity cannot be overemphasized, as this will lead to a better understanding of its biological nature.

Keywords

Adenomatoid Odontogenic Tumor, Tumor, Mandible

1. Introduction

Adenomatoid odontogenic tumor (AOT) is a rare tumor of the jaws accounting for about 2% - 7% of all odontogenic tumors [1]. In the past, other names like Adenoameloblastoma, Adamantinoma, Pseudoadenomatous ameloblastoma, Teratomatous odontoma and Odontogenic adenomatoid tumor have been used to describe the same entity. Adenomatoid odontogenic tumor has been called the two-thirds tumor because two-thirds of this tumor occurs in the maxilla, two-thirds in young females, two-thirds are associated with unerupted teeth, and two-thirds of the teeth involved are canines [1]-[4]. Mandibular tumors and the asso-

ciation with teeth apart from the canine are features seldom reported [5]. Our case report is that of an anterior mandibular AOT tumor associated with the first premolar, making it a rarity.

2. Etiology

Adenomatoid odontogenic tumor is a benign epithelial odontogenic tumor arising from the remnants of the dental lamina. 70% - 75% of AOT lesions are associated with KRAS mutations, resulting in activation of MAPK/ERK pathway, which is key to the processes of both cellular proliferation and survival [6] [7]. AOT has also been linked to Wnt/β-catenin and the Hedgehog signaling pathways that are associated with cellular proliferation. The association of AOT with impacted teeth raises more questions than answers. Is it possible that the impaction may result in the stimulation of dormant cell rests into proliferation?

3. Epidemiology

Adenomatoid Odontogenic Tumor has often been referred to as the "Tumor of 2/3" because 2/3 of this tumor is associated with the following: occurrence in females, occurrence in the maxilla, occurrence in the anterior portion of the jaws and finally occurrence with an impacted tooth, usually the canine [2]. There are three variants of AOT, namely follicular, extrafollicular, and peripheral. The difference between the follicular and extrafollicular is the association the follicular has with the crown of the tooth, mimicking a dentigerous cyst, albeit its attachment goes beyond the cemento-enamel junction. The peripheral type is extrabony, although it may be associated with saucerization of the crest of the bone [5].

4. Case Presentation

PMM is a 16-year-old female who presented to the University of Nairobi Dental Hospital on 11th October 2024, as a referral from Mama Lucy Hospital. She complained of an eight-month-old swelling of the right lower jaw. The swelling had an insidious onset, gradually increasing to its present size. There was no accompanying pain, no bleeding, no discharge, or paresthesia in the affected area. There were no constitutional symptoms related to the swelling within the intervening period. PMM had good oral hygiene habits except for tongue thrusting and nail biting. The index visit was the second encounter with an oral health specialist, the initial visit being the one to Mama Lucy Hospital.

PMM's past medical history was unremarkable, with no history of chronic illness, the only hospital admission being in 2018 for tonsillectomy following recurrent tonsillar infections. There was no positive history of food or drug allergies. PMM is the second of three siblings with no family history of a similar affliction or any other chronic illnesses. PMM is social and an average performer at school. A systematic review of all systems was unremarkable.

PMM's chronologic age was commensurate with her physique. Her vital signs were within the normal range. She had slight facial asymmetry due to a right

mandibular para-symphyseal swelling originating from the underlying bone but free from the overlying skin that had normal color and texture. The swelling was more apparent intra-orally, extending from the 43 to the 45 and inferiorly into the associated vestibule with buccal-lingual expansion. The 44 appeared to have a mesial tilt with the anterior aspect of its crown partially submerged in the swelling (**Figure 1**). The overlying mucosa was normal in texture and color except for an erythematous halo around the partially erupted 44. Upon palpation, the swelling was non-tender and firm, measuring about 2.8 × 2.5 × 2.0 cm.

An orthopantomogram was taken to determine the effect of the swelling on the mandible, and a well-defined circular radiolucency was observed in the fourth quadrat of the mandible. The lesion had a sclerotic border and had displaced 43 and 44, with part of the root of the 44 engulfed (**Figure 2**).

Histopathology: An excisional biopsy via an intra-oral approach was performed on the 11th October 2024. The histopathology report was as follows.

Macroscopy: The gross specimen consisted of a dark brown soft tissue mass with 44 attached and measuring 2.5 × 2.3 × 2.0 cm (**Figure 3**).

Microscopy: The lesion was well encapsulated and consisted of odontogenic cells organized in benign rosettes and duct-like structures resembling an adenoma, with interposed occasional eosinophilic material. The cells exhibit minimal cellular atypia with a low mitosis rate. The appearance is suggestive of an Adenomatoid odontogenic tumor (**Figure 4**).

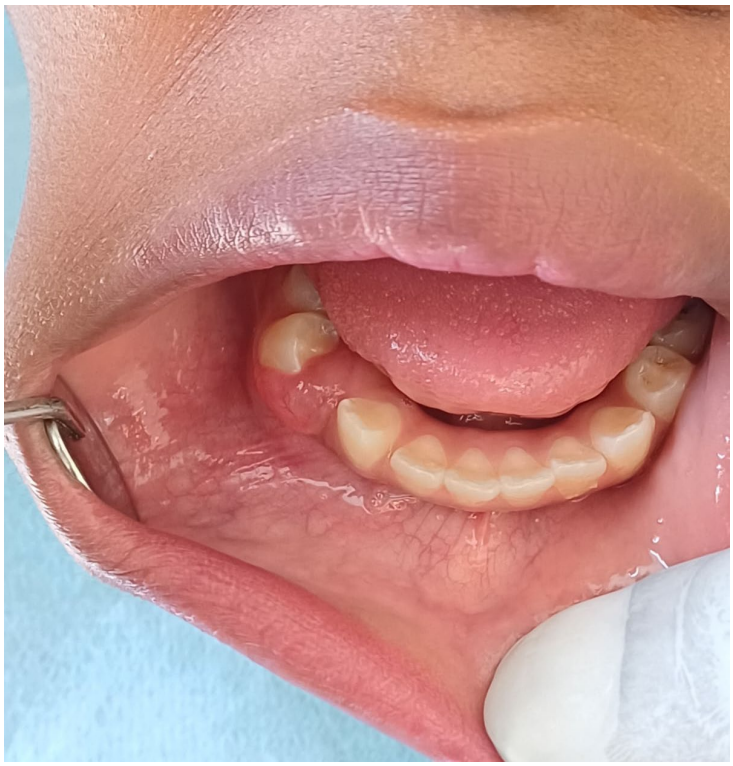

Figure 1. Intraoral picture of PMM taken showing the swelling extending from the 43 to the 45 with evidence of buccal-lingual expansion.

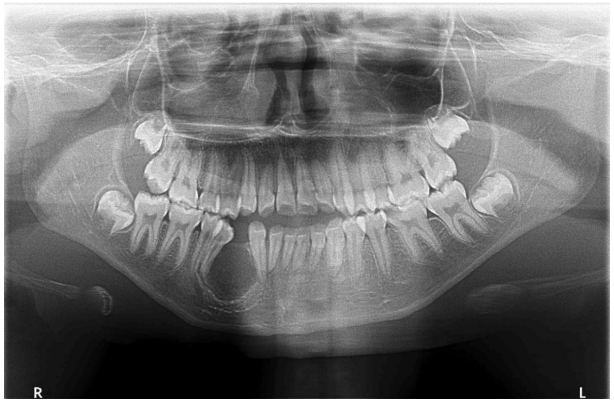

Figure 2. Orthopantomogram of PMM showing a well-defined circular radiolucency in the fourth quadrat of the mandible. The lesion has a sclerotic border and is engulfing the root of 44.

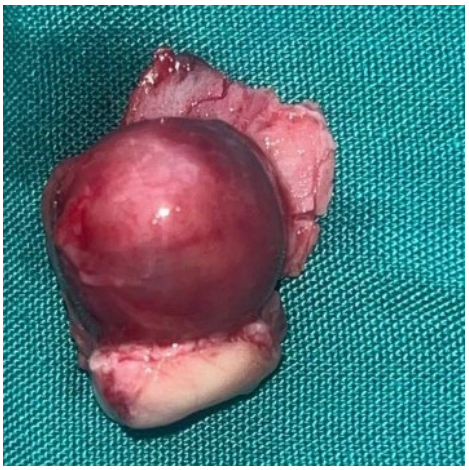

Figure 3. Gross specimen of the excised lesion with the entire capsule intact. The close association/attachment with the 44 is evident.

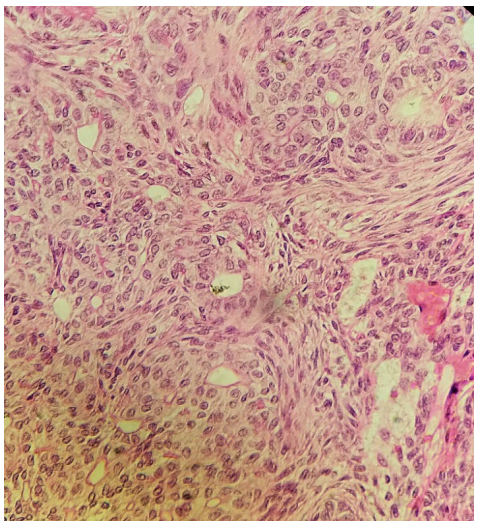

Figure 4. Spindle and polygonal-shaped epithelial cells arranged in duct-like sheets and rosette patterns surrounded by a fibrous stroma.

The differential diagnosis of Odontogenic Keratocyst, Ameloblastoma, Dentigerous cyst and Calcifying epithelial odontogenic tumor was considered and ruled out based on clinical-radiologic and histologic analysis of the case.

Rehabilitation: PMM had an uneventful recovery. An interim partial denture prosthesis was made two months from the date of surgery (**Figure 5**). She is currently well and on track with her education, with no signs of recurrence 5 months later. PMM is on periodic follow-up for an extended period during which, apart from examinations, serial radiographs will be taken to rule out any bone changes. Ultimately, with cessation of active growth, the space occasioned by the loss of 44 will be filled with an appropriate fixed prosthesis.

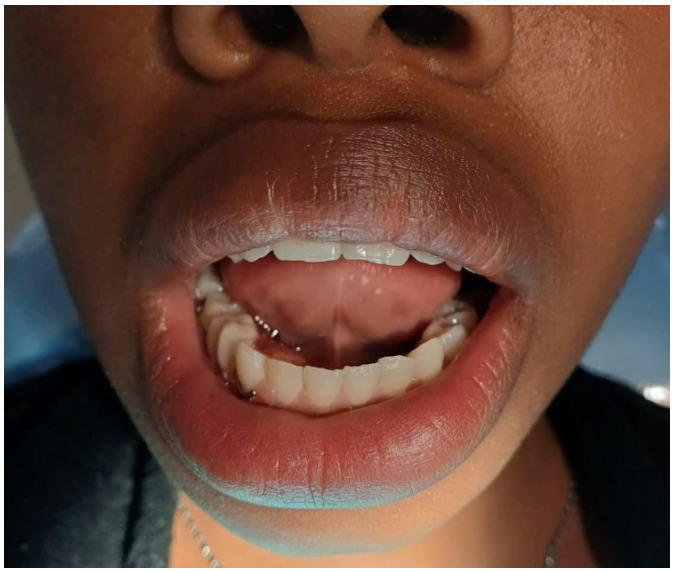

Figure 5. PMM three months after surgery with an interim partial denture *in situ*.

5. Discussion

Adenomatoid Odontogenic Tumor is a rare benign odontogenic tumor accounting for about 3% of odontogenic tumors [3]. The rarity seen in AOT may be attributed to its strong predilection for the female gender, its indolent nature making some cases escape undiagnosed, and its reduced propensity to recur, thereby reducing the number of reported cases. The tumor of 2/3, as it is commonly referred to, is more common in the maxilla [2]. In our case, the tumor was in the para-symphyseal region of the mandible. Mandibular AOT with a predilection for the anterior region is a rare occurrence that has been reported [4] [5]. Most cases of Adenomatoid Odontogenic Tumors present radiographically as unilocular radiolucency with a well-defined border. The radiographic presentation for PMM was that of a unilocular radiolucency, a feature consistent with the majority of reported cases [3]. The unilocular radiolucent and spherical nature of the tumor in PMM helped set it apart from Odontogenic Keratocysts, which show longitudinal growth. Ameloblastoma may mimic clinical-radiologic findings associated with the tumor in PMM, and histological studies would be the only way to set

them apart. Although the Central Giant Cell Granuloma was an important differential diagnosis based on the clinical-radiologic findings, the presence of a well encapsulated lesion at surgery ruled it out, as this tumor is never encapsulated. The relationship of the tumor to the 44 was not a dentigerous one and helped rule out the cyst as a possible differential. About 2/3 of the intra-osseous lesions of AOT have been associated with small radiopaque foci. However, this was not the case with PMM. The absence of radiopaque foci has also been reported in a number of studies [4] [8]. The tumor associated with PMM was well encapsulated (**Figure 3**), with cells arranged in rosette patterns in some areas and in others having duct-like spaces. The presence of eosinophilic amorphous material was noted in PMM (**Figure 4**). All these are features consistent with the histologic patterns of AOT as reported in the literature [4]. Histopathologic overlap with AOT-like areas seen in odontomas and calcifying epithelial odontogenic-like areas in some cases of AOT emphasizes the need to integrate clinic-radiologic findings in establishing a final diagnosis [6].

Most cases of AOT are associated with maxillary canines. However, other teeth in the neighborhood have also been implicated [3]. In the case of PMM, the 44 was involved with the disease as evidenced by its attachment (**Figure 3**). The rounded nature of the lesion, coupled with clinical evidence of buccal-lingual, is evidence of the centrifugal expansion growth pattern in AOT and was a feature seen with PMM (**Figures 1-3**). This phenomenon is also consistent with other AOT cases seen elsewhere [4].

Most cases of AOT are easily enucleated without recurrence. However, large tumors necessitating resection and reconstruction have been reported [5]. PMM had a moderately sized tumor in a readily accessible area, making it preferable to enucleate and follow up. Indeed, based on the size of the tumor in the case of PMM, there was no need for reconstruction of the mandible. The loss of 44, resulting in the potential danger of migration of 45 into the space coupled with functional and esthetic considerations, informed our decision to fabricate an interim partial denture prosthesis for PMM. The ultimate permanent fixed prosthesis to replace the missing 44 would be considered at the cessation of active growth, presumably at around 18 years of age.

6. Conclusion

AOT remains an important differential diagnosis of jaw swellings, making it imperative to learn its biologic nature, including ways of presentation. Its rarity justifies the use of case reporting as one of the means in improving existing knowledge. The interrogation of its molecular pathology is equally important and should be emphasized as an area for research.

Consent

Written informed consent for the publication was obtained from the parents of PMM; a copy of this is available for review by the editorial team of this journal.

Acknowledgements

We thank Alice Limo for the laboratory work in preparation of the specimen for histopathology.

Conflicts of Interest

The authors declare no conflict of interest with regard to the publication of this paper.

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