Case Report
DOI: 10.22037/jds.v42i4.45878 ISSN 2645-4351

Adenoid Ameloblastoma as a Trap for Inexperienced Pathologists: A Case Report

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(Submitted: 28 July 2024 – Revised version received: 1 September 2024 – Accepted: 7 September 2024– Published online: Autumn 2024)

Abstract

Objectives: Ameloblastoma is the second most frequent type of odontogenic tumors. They are thought to be formed by epithelium of ectodermal origin, Histopathologically, ameloblastoma has plexiform, follicular, acanthomatous, desmoplastic, granular cell, and basal cell subtypes. Adenoid ameloblastoma, also called dentinal adenoid ameloblastoma, is a rare odontogenic tumor. About 40 cases have been reported, with the highest incidence in the fourteenth year of life (age range:25-52 years), indicating a small female population and a similar population for ameloblastoma. The differential diagnosis includes odontogenic lesions such as calcifying odontogenic cyst, calcified epithelial odontogenic tumor, ameloblastoma and adenomatoid odontogenic tumor (AOT). Histopathological results show that odontogenic epithelial tumors consist of nests, islands, cords, anastomotic chains and large areas of epithelium, ameloblast-like cells in the periphery, stellate reticulum-like cells in the space and cystic/microcystic patterns, acanthomatous changes and ductal patterns similar to AOT.
Case: The aim of this report was to present a unique case of adenoid ameloblastoma affecting the right side of mandible in a 47 y.o woman which had a previous pathology diagnosis of adenoid cystic carcinoma for this lesion in her medical history.
Conclusion: Such rare cases of adenoid ameloblastoma can pose challenges in the initial diagnosis and treatment.
Keywords: Ameloblastoma; Odontogenic tumor; Diagnosis; Differentiation; Adenoid

How to cite:
Mashhadiabbas F, Gholami Toghchi S, Kashefi Baher R, Arefi Fard H. Adenoid Ameloblastoma as a Trap for Inexperienced Pathologists: A Case Report. J Dent Sch 2024;42(4):184-187.

Introduction

Ameloblastomas account for approximately 1% of all tumors in the jaw, although they are the second most frequent type of odontogenic tumors and thought to originate from the ectoderm. This means that they start from the cells around or close to the root of the tooth; the ectodermal layer. They usually occur in the mandible rather than the maxilla and are more common in the posterior area than the anterior area. Although ameloblastomas are benign, they can exhibit aggressive behavior and rarely develop or are associated with malignancy (malignant ameloblastoma or ameloblastoma). Because ameloblastoma is associated with the teeth and related structures, it is rare in other sites outside the maxilla and mandible.[1] In 1930, Ivey and Churchill rechristened this lesion as "ameloblastoma".[2] In 2017, the World Health Organization classified ameloblastoma as an epithelial odontogenic tumor.[3] Many ameloblastomas show MAPK pathway-associated gene mutations; the most common of which is the BRAF-V600E mutation. [3] Histopathologically, ameloblastoma has plexiform, follicular, acanthomatous, granular cell, desmoplastic, and basal cell subtypes. Granular cell subtype is rare, accounting for only 5% of all ameloblastomas.[4]
Adenoid ameloblastoma (AAME), also known as dentinal adenoid ameloblastoma, is a rare epithelial odontogenic tumor. AAME is locally invasive, with aggressive clinical behavior, and a high recurrence rate (approximately 70%) after enucleation.[5-7] Approximately 40 cases have been reported, and the highest incidence is in the 40s (25-52 years), slightly more prevalent in women, with a population similar to ameloblastoma.[5,7] It appears to mostly affect the lower jaw (64.7%), usually presenting with a painless swelling.[7]
From an imaging perspective, the majority of AAMEs (~82%) appear as radiolucent lesions on examination; occasionally, radiopaque lesions are demarcated and have cortical perforations. Histologically, AAME shows odontogenic epithelium as duct-like structures, epithelial rings, and cribriform pattern, similar to conventional ameloblastoma.[5-7,8] Some of these features resemble ameloblastoma, while the adenoidal component resembles adenomatoid odontogenic tumors.[5]
Dentinogenic ghost cell tumor, another histopathological differential diagnosis of AAME, may show clinical features overlapping with dentinoid odontogenic carcinoma, but definitive diagnostic criteria have not been established.[5,7] Dentinogenic cell tumors [9,10] and odontogenic carcinoma with dentinoid [11] have CTNNB1 (beta-catenin) exon 3 mutations, similar to other cell-rich tumors such as calcifying odontogenic cysts.[12]

Case Report

The patient was a 47-year-old female referred to the department of Oral and Maxillofacial Pathology at

Faculty of Dentistry, Shahid Beheshti University of Medical Sciences (SBMU), in August 2023 for a pathological consultation. Initially, she was referred to an oral surgeon in December 2022 with a chief complaint of pain and swelling on the right side of her mandible. After the primary evaluation and workup, an excisional biopsy had been done, and the specimen submitted to a general pathology center, where the pathologist had diagnosed adenoid cystic carcinoma (ADCC).

In her past medical history, it was observed that a surgical resection on the right side of mandible had been performed in February 2004, with a pathologic diagnosis of plexiform type ameloblastoma.

Radiographic findings showed a mesh-type reconstruction plate on the right side of the mandible, extending from the ascending ramus to beyond the symphysis pubis. The plate had been placed as part of the reconstructive treatment following the surgical resection in 2004 (Figure 1).

The new histopathologic findings revealed an epithelial odontogenic tumor composed of nests, islands, cords, anastomosing strands, and large sheets of odontogenic epithelium. The epithelium featured ameloblast-like cells with reverse polarity at the periphery and stellate reticulum-like cells in the central portion, along with cystic/microcytic formation, acanthomatous changes, and a ductal pattern resembling an adenomatoid odontogenic tumor (AOT). The tumoral cells invaded the muscle fibers. In the fibrous connective tissue, scattered chronic inflammatory cell infiltration, nerve bundles, and adipose tissue were also evident. However, there was no evidence of malignancy (Figure 2). Finally, the definitive diagnosis was Ameloblastoma, adenoid type. Informed consent was obtained from the patient before publishing her radiograph and histopathological view anonymously.

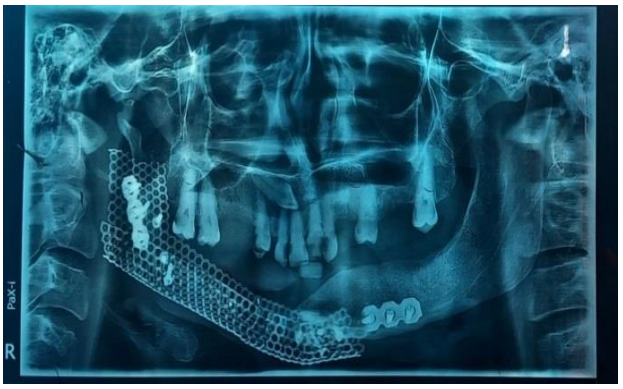

Figure 1: Panoramic view showing a mesh-type reconstruction plate area at right side of the mandible from the ascending ramus to beyond the symphysis pubis. It illustrated the reconstructive treatment following surgical resection in 2004.

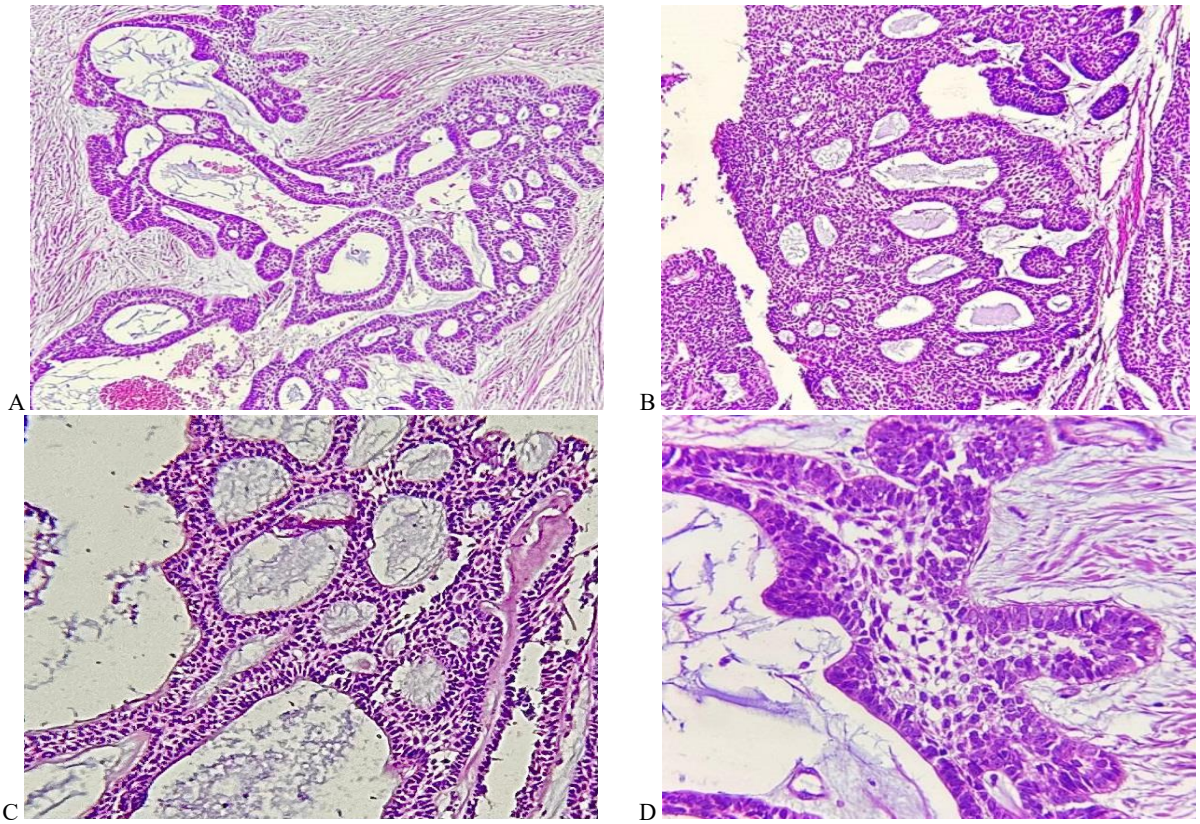

Figure 2: Histopathologic views of the lesion. A, B) Sections at 4x magnification showing a tubular and cribriform pattern of epithelial cells that mimics salivary lesions such as ADCC and may confuse an inexperienced pathologist. C, D) Sections at 10x and 40x magnification showing epithelial cords and islands with columnar ameloblast-like cells at the periphery portion and stellate-like cells in the center portion of the islands, which differ from the basaloid cells in ADCC.

Discussion

Adenoid ameloblastoma is a poorly differentiated tumor with a variety of histopathological features that is rarely reported; 38 cases were reported worldwide in a review of the English literature from 1959 to 2018.[5,13] A new case was presented in this study from August 2023.

Previous case reports revealed no gender propensity, and the age of patients varied from 4 to 82 years.[13] The patient in this case report was female, and her age (47 years) fell within the reported range.

Effiom et al. reported that AAME often occurs at the posterior area of the jaws (especially the mandible, which included 69.4% of affected sites).[14] The lesion location in the present case (posterior mandible) was in line with the common location reported by Effiom et al.[14].

The most frequent clinical sign was reported to be swelling (more than half of the eight cases in Farias' study)[7]; similarly, pain and swelling were the patient's chief complaints in the present study.

Due to de Arruda's review of literature [13], the radiographic appearance of AAME can be unilocular and well-defined or diffuse and multilocular. Radiographic differential diagnosis includes odontogenic lesions such as calcifying odontogenic cyst (COC)[15], calcifying epithelial odontogenic tumor (CEOT), ameloblastoma, and AOT.[16]

Previous studies have reported that AAME is an epithelial odontogenic tumor that can present a pseudoductal structure consisting of columnar cells arranged on palisades, consisting of multiple tumors, histologically composed of nests, islands, cords, and anastomotic chains.[5,7,17] The same histopathological features were observed in the present case.

Dentin deposition has been reported in some reports (three out of five cases in the study by Loyola et al.[5] and two out of eight cases in the study by Adorno-Farias et al.[7]), but it was not found in our microscopic slides.

The histological differential diagnoses include odontogenic lesions exhibiting pseudoductal or cribriform features, such as AOT, COC, and in some cases CEOT.[7,18] In this case AOTs were included in the differential diagnosis due to their ductal pattern.

Careful monitoring of AAME is important, since to date the correct treatment strategy has not been fully developed. The treatment strategy for the present case was resection surgery in the mandible.

Conclusion

In conclusion, we presented a rare case of AAME. The clinical signs, radiographic, and histological findings of AAME can pose challenges in the initial diagnosis, resembling other odontogenic cysts and tumors, as seen in our case report. Therefore, caution is advised in AAME diagnosis to establish a timely and accurate diagnosis and initiate appropriate treatment.

Acknowledgements:
This research was supported by Shahid Beheshti University of medical sciences.

Author Contributions: F.M.A: conception and design of study, H.R.F: conception and design of study, acquisition of data and final approval of manuscript, R.K.B: conception and design of study and acquisition of data, S.G.T: conception and design of study, acquisition of data and critical revision.

Funding: No funding was received for this research.

Ethical Approval: There is no ethical approval code for this case report.

Informed Consent Statement: Informed consent was obtained from the patient before publishing her radiograph and histopathological views anonymously.

Data Availability Statement: For any future correspondence please contact hamidrezaarefi1375@yahoo.com

Conflict of Interest: No Conflict of Interest Declared. ■

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