Journal of Dental Sciences 20 (2025) 1977–1979

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-jds.com

Correspondence

Adenomatoid odontogenic tumor arising from a dentigerous cyst in an elderly patient: An unusual presentation

KEYWORDS
Adenomatoid odontogenic tumor;
Dentigerous cyst;
Cytokeratin 19;
Vimentin

Adenomatoid odontogenic tumor (AOT) is an epithelial odontogenic tumor that is histologically characterized by duct-like structures.[1] AOT is predominantly diagnosed in younger patients and rarely occurs in association with dentigerous cyst.[2,3] In this report, an unusual case of AOT possibly arising from a dentigerous cyst in an elderly patient was presented.

A 62-year-old male presented with a mandibular radiolucent lesion discovered incidentally on routine dental radiography. A panoramic radiograph showed a well-circumscribed radiolucency surrounding the crown of the unerupted right mandibular second premolar (Fig. 1A). No calcifications were found within the radiolucency. The radiological diagnosis was dentigerous cyst. Surgical enucleation was performed with extraction of the associated premolar. Histologic examination showed a cystic structure containing a small intraluminal mass (Fig. 1B). The cystic lining consisted of nonkeratinized stratified squamous epithelium, as is seen in dentigerous cyst (Fig. 1C). The cystic cavity was partially filled with a pool of blood (Fig. 1B), with the effacement of the adjacent lining epithelium and a mild inflammatory infiltrate in the fibrous cystic wall (Fig. 1D). The intraluminal mass consisted of nodules of cuboidal or columnar epithelial cells with duct-like structures characteristic of AOT (Fig. 1E). Rosette-like structures were occasionally found (Fig. 1F). No calcifications were detected histologically. Immunohistochemically, the lining epithelium was positive for cytokeratin 19 (CK19) (Fig. 1G), which also highlighted the duct-like structures in the intraluminal mass (Fig. 1H). Vimentin was negatively expressed in the lining epithelium (Fig. 1I) and the center of the nodules (Fig. 1J); however, anastomosing strands at the periphery of the nodules were positive for vimentin (Fig. 1J). A diagnosis of AOT was made. The patient was followed up for 3 years after surgery with no evidence of recurrence.

While most AOTs are diagnosed in the second and third decades of life and are notably uncommon in patients older than 30 years,[1,2] the present AOT case occurred in an elderly patient, making it difficult to reach a clinical diagnosis. Radiological features, such as the apical extension of the radiolucency over the cementoenamel junction and fine calcifications within the radiolucency, may help to differentiate AOT from dentigerous cyst;[2] however, no such features were observed in the present case, resulting in a radiological diagnosis of dentigerous cyst. These unusual clinical and radiological manifestations support the assumption that the present AOT case might develop from a pre-existing dentigerous cyst.

The present case histologically showed a cystic structure with an intraluminal mass characterized by duct-like structures. According to a recent study, calcifying odontogenic cyst (COC) can also show duct-like/cribriform structures and intraluminal epithelial proliferation.[4] Therefore, care is needed to distinguish AOT arising from

https://doi.org/10.1016/j.jds.2025.04.021

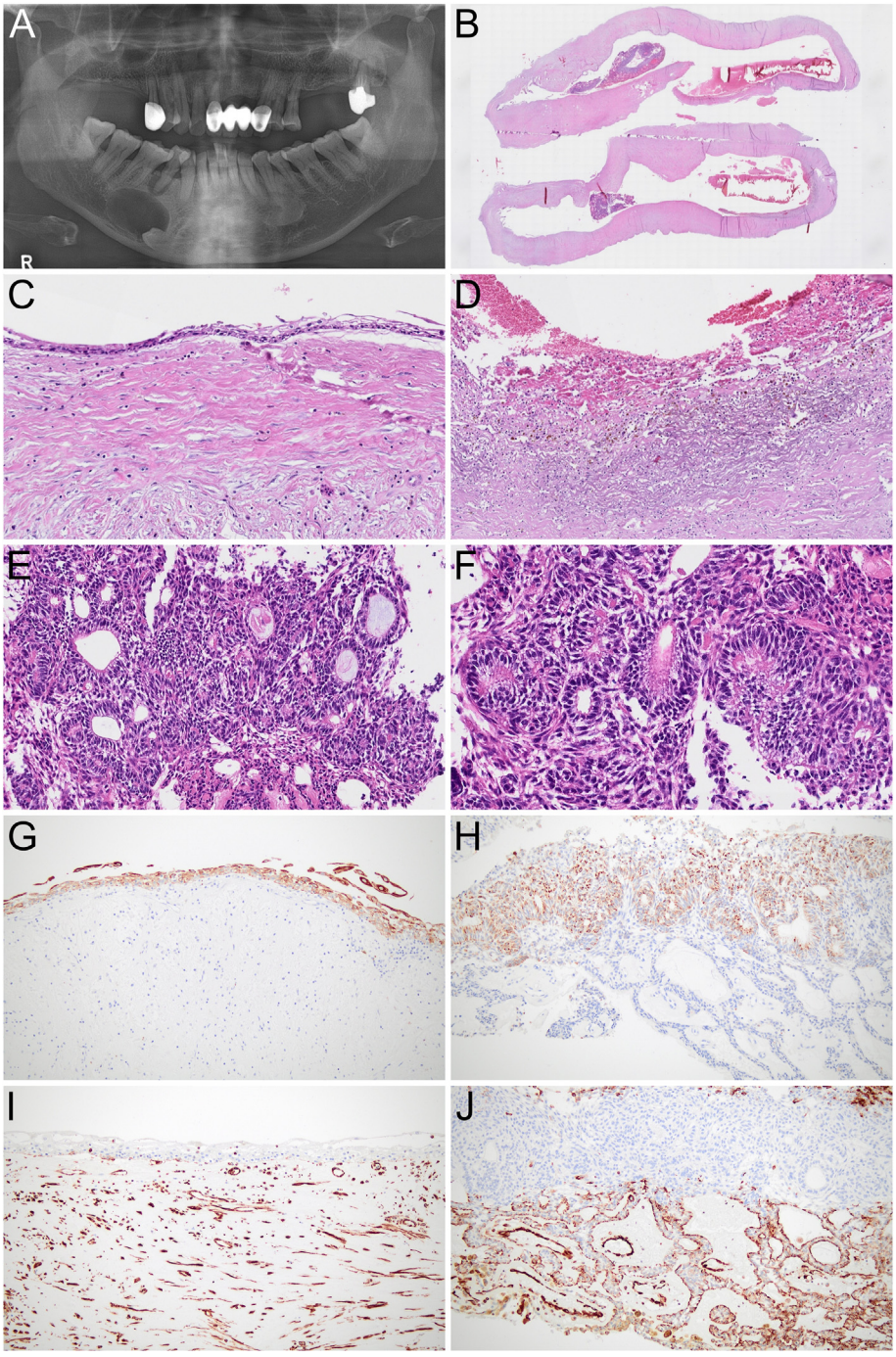

Figure 1 Radiological, histologic, and immunohistochemical findings of an adenomatoid odontogenic tumor arising from a dentigerous cyst. (A) A panoramic radiograph shows a well-defined radiolucent lesion surrounding the crown of the unerupted right mandibular second premolar. (B) A low-power histologic image shows a cystic structure with an intraluminal mass. The cystic cavity is partially filled with a pool of blood. (C) The cystic lining consists of nonkeratinized stratified squamous epithelium, indicative of dentigerous cyst. (D) Adjacent to the blood pool, the lining epithelium is lost, and a mild inflammatory infiltrate is seen in the fibrous cystic wall. The intraluminal mass consisting of nodules of epithelial cells shows duct-like (E) and rosette-like (F) structures, characteristic of adenomatoid odontogenic tumor. Cytokeratin 19 is positive in the cystic lining (G) and highlights the duct-like structures (H). Vimentin is negative in the cystic lining (I) but positive in anastomosing strands at the periphery of the nodules (J).

a cyst with COC, which may be facilitated by identifying the presence of ghost cells or dentinoid—the characteristic features of COC. According to a previous AOT study, CK19 is strongly expressed in columnar ameloblast-like cells, and vimentin is negative in the center of solid nodules but positive at the periphery;[5] these immunohistochemical profiles were identified in the intraluminal mass of the present case.

Declaration of competing interest

The author has no conflicts of interest relevant to this article.

Acknowledgments

The present research was supported by the research fund of Dankook University in 2022.

References

1. WHO classification of tumours editorial board. Head and neck tumours [Internet]. In: *Lyon (France): International Agency for Research on Cancer*. 5th, 9; 2023 [cited Apr 19, 2025]. WHO classification of tumours series. Available from:. https://tumourclassification.iarc.who.int/chapters/52.
2. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and maxillofacial pathology-E-Book*. Elsevier Health Sciences, 2023.
3. Manjunatha BS, Harsh A, Purohit S, Naga MV. Adenomatoid odontogenic tumor associated with a dentigerous cyst. *J Cancer Res Therapeut* 2015;11:649.
4. Oh KY, Kim JH, Yoon HJ. Calcifying odontogenic cyst demonstrates recurrent WNT pathway mutations and so-called adenoid ameloblastoma-like histology: evidence supporting its classification as a neoplasm. *Mod Pathol* 2024;37:100484.
5. Passador-Santos F, de Oliveira CRR, Teixeira LN, et al. Adenomatoid odontogenic tumor: features of ameloblastic-like epithelial cells differentiation, secretion, and the nature of tumor cells products. *J Oral Pathol Med* 2023;52:644—53.

Kyu-Young Oh*
Department of Oral Pathology, College of Dentistry, Dankook University, Cheonan, Republic of Korea
Department of Oral Pathology, Seoul National University Dental Hospital, Seoul, Republic of Korea

*Department of Oral Pathology, College of Dentistry, Dankook University, 119 Dandae-ro, Cheonan, 31116, Republic of Korea.
E-mail address: bigfish@snu.ac.kr

Received 19 April 2025
Final revision received 20 April 2025
Available online 29 April 2025