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Adenoid ameloblastoma in the maxilla



KEYWORDS

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Ameloblastoma is a benign but locally aggressive odontogenic epithelial neoplasm. Traditionally, it has been classified into three subtypes: conventional, unicystic, and peripheral. In 2022, the World Health Organization (WHO) revised its classification of head and neck tumors, introducing a novel entity termed adenoid ameloblastoma (AdAM). Histologically, AdAM exhibits epithelial characteristics akin to conventional ameloblastoma but is distinguished by the presence of duct-like structures, epithelial whorls, and a cribriform architecture.¹ Here, we reported a case of AdAM in the maxilla.

A 28-year-old Taiwanese female with no significant medical history presented to the Department of Oral and Maxillofacial Surgery at Kaohsiung Medical University Hospital, reporting gingival swelling and a sensation of fullness in the left upper jaw persisting for over a month. Clinical examination revealed expansion of the left upper alveolus in the molar region with tenderness on palpation. A panoramic radiograph demonstrated a well-defined radiolucency in the posterior maxilla, associated with an impacted third molar and root resorption of the second molar (Fig. 1A). Computed tomography identified a hyperdense mass extending into the maxillary sinus (Fig. 1B and C). An incisional biopsy was performed, and histopathological analysis confirmed the diagnosis of AdAM. The lesion was subsequently surgically excised and submitted to the Department of Oral Pathology for further examination. Microscopic evaluation of the specimen revealed a neoplasm with focal cystic components lined by odontogenic epithelium. The peripheral cells exhibited a single layer of ameloblast-like cells with reversed polarity, arranged in a plexiform pattern (Fig. 1D). Additionally,

multifocal duct-like structures and epithelial whorls were observed (Fig. 1E and F). Immunohistochemical analysis showed positivity for CK19 and BRAF in tumor cells arranged in a plexiform pattern (Fig. 1G and H), as well as positive nuclear β -catenin staining in areas exhibiting adenoid features (Fig. 1I). No evidence of recurrence was observed during a nearly 48-month postoperative follow-up.

Odontogenic tumors constitute a rare group of neoplasms derived from the tooth-forming apparatus and its remnants, reflecting various aspects of odontogenesis. Advances in molecular technologies have enhanced our understanding of the molecular pathogenesis of odontogenic neoplasms. Three key signaling pathways implicated in their pathogenesis include the mitogen-activated protein kinase (MAPK) pathway, the sonic hedgehog (SHH) pathway, and the Wnt signaling pathway.² Ameloblastomas are characterized by mutations in the MAPK and SHH pathways, with the *BRAF V600E* mutation being the most prevalent, identified in 87.3 % of Taiwanese ameloblastomas.³ A previous study has demonstrated that AdAM is *BRAF* wild-type and harbors *CTNNB1* (β -catenin gene) mutations, suggesting that it is a distinct entity rather than a subtype of conventional ameloblastoma.⁴ The key histopathological features of AdAM include ameloblastoma-like epithelium, characteristic duct-like structures, cribriform architecture, and cellular condensations known as morules. To date, only a limited number of cases have been reported. A recent systematic review analyzing 30 cases of AdAM concluded that the majority of cases occurred in males, with a slight predilection for the mandible. Surgical resection was the most commonly employed treatment, with a recurrence rate of 30 % reported in the reviewed cases.⁵

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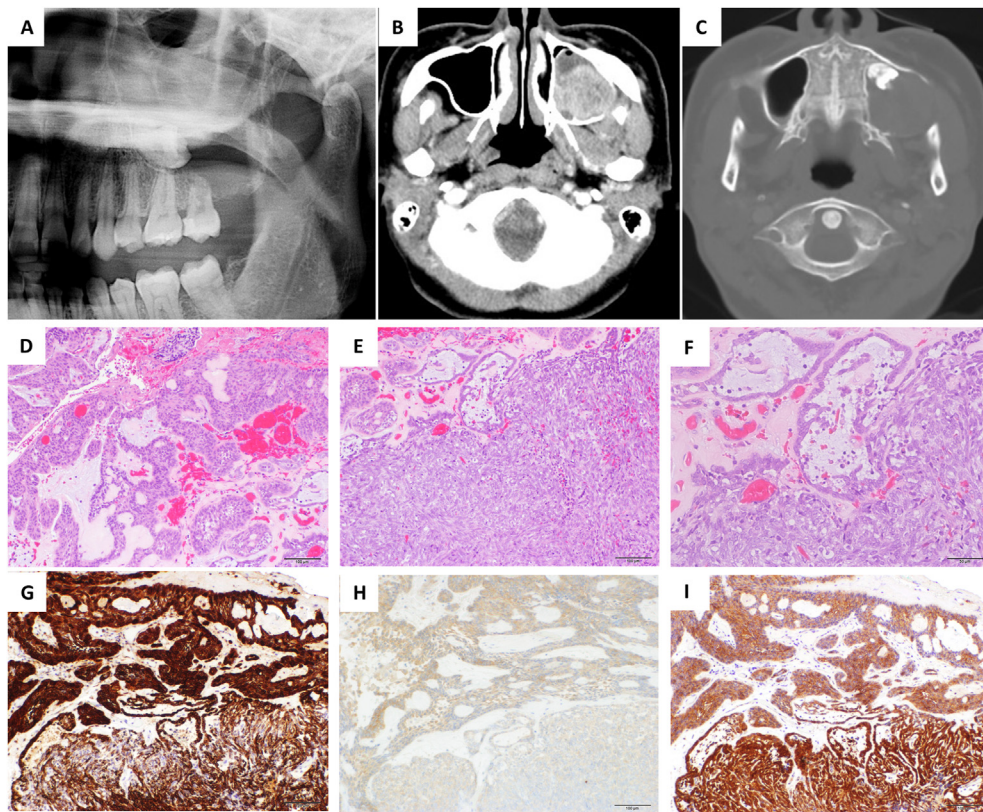


Figure 1 Clinical, radiographic, and microscopic photographs of a case of adenoid ameloblastoma. A well-defined radiolucency in the posterior left maxilla, associated with an impacted third molar and root resorption of the second molar (A). Computed tomography identified a hyperdense mass extending into the maxillary sinus with erosion of lateral and posterior walls of left maxillary sinus (B and C). The histopathological examination showed an odontogenic epithelial neoplasm. The peripheral cells exhibited a single layer of ameloblast-like cells with reversed polarity, arranged in a plexiform pattern (D). Multifocal duct-like structures and epithelial whorls were observed (E and F). (Hematoxylin and eosin stain; original magnification; D, 10×; E, 10 × ; F, 20 ×). Immunohistochemically, the tumor cells arranged in a plexiform pattern showed positivity for CK19 and BRAF (G and H). Positive nuclear β-catenin staining was noted in areas exhibiting adenoid features (I). (original magnification; G, 10×; H, 10 × ; I, 10 ×).

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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