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Adenoid variant of peripheral ameloblastoma with cellular atypia in the retromolar pad area: A case report

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Abstract

Adenoid ameloblastoma (AA) is a very rare subtype of solid/multicystic ameloblastoma. It demonstrates areas of typical or atypical adenomatoid odontogenic tumor along with the ameloblastic content. To date, there are 17 reported cases of AA. This paper reports, however, the first introduced adenoid variant in a peripheral ameloblastoma. It also depicts an unusual appreciation of cellular atypia and nuclear pleomorphism. This finding may promote a new pathogenetic scenario to the current nosology of this debatable lesion and to the WHO taxonomy of benign ameloblastomas in 2005.

Keywords:
Adenoid ameloblastoma
Retromolar
Peripheral ameloblastoma
Adenomatoid odontogenic tumor

1. Introduction

Philipsen and Birn [1] proposed the designation of adenomatoid odontogenic tumor (AOT) which was, two years later, promoted by the World Health Organization. Adenomatoid odontogenic tumor (AOT), both in nature and in designation, is now questioned. Based on clinical and immunohistochemical findings, AOT was suggested to be a hamartomatous entity emanating from the reduced enamel epithelium [2]. Owing to its benign behavior, slow growth and clear delineation, as well as its low tendency to recur (0.2%), the treatment of choice is conservative surgical enucleation and simple curettage [3]. Later, AOT has eschewed the very usual pathway by appearing in collision with ameloblastic elements [4] and by integrating into the so-called adenoid ameloblastoma (AA) [5]. Moreover, AA trails a new pathway by its representation, as reported in this paper, peripherally in the retromolar pad area.

2. Case presentation

A 38-year-old female had a small swelling at the retromolar pad of the right mandible which annoyed her during mastication. The asymptomatic exophetic swelling measured approximately 1 × 1.5 cm. The overlying mucosa displayed normal color and texture. The lesion was totally asymptomatic and innocuously seemed non-suspicious. The radiological picture, moreover, demonstrated no bony involvement. The lesion was surgically excised 14 months ago. To date, there is no evidence of recurrence.

Histologically, atypical AOT areas with rosettes and duct-like structures were observed in a pool of ameloblastomatous lesion. Both were interspersing the salivary tissue of the retromolar pad. The lesion was unecapsulated and was surrounded by a normal histological architecture of the retromolar pad: fibrous, adipose and salivary tissues (Fig. 1). Both the ameloblast-like cells and duct-like structures manifested tall columnar epithelial cells. Interestingly, the lesion evinced occasional nuclear atypia, even some mitotic figures, and hyperchromatic tumor cells. Less significant, washed-out nuclei were conspicuous (Fig. 2). Although induction materials and cystic changes were evident in the atypical arrangements which recapitulated the appearance of AOT, the classical eosinophilic materials, which are typically seen in AOT, were unremarkable (Fig. 3). Moreover, the tumoral cells were observed, intermittently and sporadically, interfering with the salivary architecture (Fig. 4). No necrosis was obvious. The cellular atypia could not prove to promote a malignancy. The specimen margins were negative for any micro-invasions. Sonographically, all lymph nodes were oval in shape and were preserving the sinus fat. The largest lymph node revealed a reactive enlargement, yet subcentimetric (0.08 × 0.04 in the largest dimensions). Immunohistochemically, the tumoral cells were strongly positive both for Calretinin and for Wt-1 but were negative for Ber-EP4. Therefore, the lesion was signed out as a benign adenoid variant of peripheral ameloblastoma.

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3. Discussion

Ameloblastoma is divided, according to the taxonomy of WHO, into four types: solid/multicystic, extra-osseous/peripheral, desmoplastic, and unicystic ameloblastomas. The solid/multicystic type encompasses several subtypes, which include follicular, plexiform, basaloid, clear cell, acanthomatous, keratoameloblastoma, hemangimatous, mucinous and granular cell patterns [6].

Adenomatoid odontogenic tumor could be traced in association with other pathoses as well as per se. A hybridization of ameloblastoma and AOT was reported [4]. However, AOT was homogeneously observed in cases where it intermingles with native ameloblastic components. This rarity was designated “adenoid” ameloblastoma (AA). Attributing such an “adenomatoid” or “adenoid” designation was mainly meant to specify those tumors which reveal impressive occurrence of AOT-like areas along with the tumoral ameloblastic components. That is why the diagnosis of adenoid ameloblastoma is always challenging [5,7]. Complicating matters, atypical AOT was recently reported in the literature, in a non-ameloblastomatous context, by Kawahara et al. [8]. They reported unusual histopathological features which included lack of duct-like structures, tumor droplets and the characteristic lining columnar epithelial cells, which were detected in approximately 99% of the reported cases of AOT.

Peripheral ameloblastoma (PA) consists histologically of a central mass of loosely connected stellate reticulum-like cells, with similar picture to conventional ameloblastoma, as well as areas surrounded by a layer of tall columnar cells with well-polarized “juxtaposed” nuclei; mimicking basal cell carcinoma. Immunohistochemically, PA is, unlike basal cell carcinoma, negative for Ber-EP4 [9]. Moreover, ameloblastomas show specific immunoreactivity for Calretinin and Wt-1 [10]. The tumoral cells of the reported case were strongly positive for Calretinin and Wt-1 but were negative for Ber-EP4. This confirmed the adenoid ameloblastic nature of the innocently-looking lesion.

Now that several histological features have debuted at the present case, a rapt attention should be paid to the ameloblastomatous rarities. The rarities of the present case, other than being the first reported case of adenoid peripheral ameloblastoma, include, first, the site of the ameloblastic lesion, retromolar pad, which is extremely rare. Second, occasional cellular atypia and mitotic figures were evident. Although they were not abundant enough to support a frank malignancy, mitotic figures count was 2
at the field of high power magnification. Third, the peripheral ameloblastoma, which is the rarest type, has characterized an adenoid pattern. This should, all in all, prompt new speculations about the validity of the contemporary histological taxonomy of the ameloblastic lesions. This holds true especially with the paucity of available information about ameloblastic lesions which develop malignancies.

4. Conclusions

Peripheral ameloblastoma can show an adenoid pattern. Although this is the first reported case in the medical literature, cellular atypia and mitotic figures should warrant longer schedule of close follow-up. Therefore, clinicians and pathologists should reconsider the benignancy of atypical cases. More important, the contemporary nosology and updates about ameloblastomas should develop the current WHO taxonomy of ameloblastoma so that it may correlate the histological phenotypes to the malignancy potential.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent was sent to the Editor-in-Chief of this journal.

Competing interest

None.

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References