PATHOLOGICA 2017;109:379-381

Case report

Adenoid ameloblastoma with dentinoid and cellular atypia: a case report and literature review

B.AE. O. KHALELE
Department of Diagnostic Pathology, Ministry of Health, Egypt

Key words

Adenomatoid odontogenic tumor • Peripheral adenoid ameloblastoma • Odontogenic tumors

Summary

Adenomatoid odontogenic tumor (AOT) is always benign. Given the very rare recurrence rate and the zero potential of malignant transformation, authors have considered it a hamartoma. Accordingly, 'AOT' is no more than a misnomer. This report, however, describes the first recognition of cellular atypia and plemorphism in a peripheral orophayrngeal AOT which embraces an ameloblastic component. The overall picture was diagnosed, after careful histological and immunohitochemical assessment, as a peripheral adenoid ameloblastoma. This find may promote a new pathogenetic scenario to the nosology of this debatable lesion.

Background

Philipsen and Birn [1] proposed the designation of AOT which was two years later, promoted by the World Health Organization. Adenomatoid odontogenic tumor (AOT), both in nature and designation, is now questioned. Based on clinical and immunohistochemical findings, it was suggested to be hamartomatous with histogenesis 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 to appear in combination with ameloblastic elements and exhibit new features. This paper reports one of the rarest findings in this domain.

Case report

A 38-year-old female manifested a small swelling at the retromolar pad of the right mandibule. The asymptomatic exophetic swelling measured 1 x 1.5 cm. It was incidentally discovered during a routine examination. The overlying mucosa displayed normal color and texture. The radiological picture, moreover, showed no bony involvement. The lesion was surgically excised 8 months ago with no evidence of recurrence so far.

Histologically, atypical AOT areas with rosettes and duct-like structures were intervening the salivary tissue in conjunction with peripheral ameloblastic elements, both in a mass (Fig. 1) and intermittent configurations (Fig. 2). The classical eosinophilic materials were inconspicuous. Dentinoid materials were surprisingly remarkable. Intriguingly, the lesion evinced nuclear atypia, even some mitotic figures, and hyperchromatic tumor cells (Figs. 3-4). No necrosis was obvious. The cellular atypia could not prove to promote a malignancy. Immunohistochemically, the lesion was strongly positive for p53 (Fig. 5).CD-31 and S-100 expression were negative. The specimen margins were negative for any micro-invasions. The diagnosis was established as a peripheral adenoid ameloblastoma.

Discussion

Adenomatoid odontogenic tumor (AOT) is an uncommon, progressively growing, and asymptomatic benign non-invasive lesion, which occurs twice as often in females and usually in the second decade of life. The three variants of AOT are characteristic – a follicular, extrafollicular, and peripheral – endorsing the hamartomatous nature of this lesion, rather than being a true neoplasm. A low neoplasticity of AOT is also proposed. The peripheral variant is, among all, the rarest comprising only

Correspondence
Bacem AE. O. Khalele, Department of Diagnostic Pathology, Ministry of Health, Egypt - E-mail: bacemottoman@gmail.com

Fig. 1. Photomicrograph displaying a mass of AOT-like lesion and follicular ameloblastomatous proliferations intervening the minor salivary glands (H&E stained Original magnification 10x).

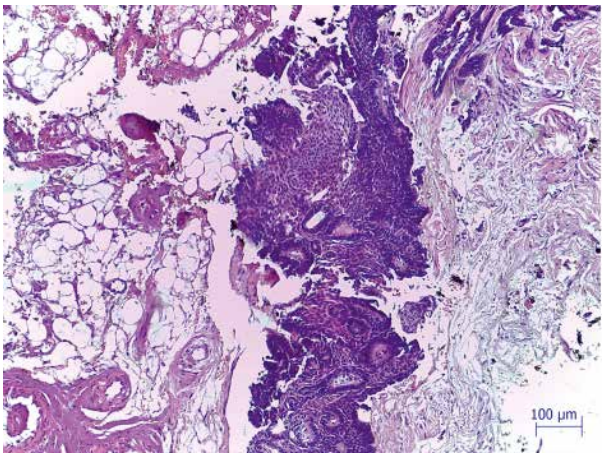


Fig. 2. Photomicrograph revealing sporadic configuration of the tumoral cells and duct-like structures (H&E stained, Oil magnification power).

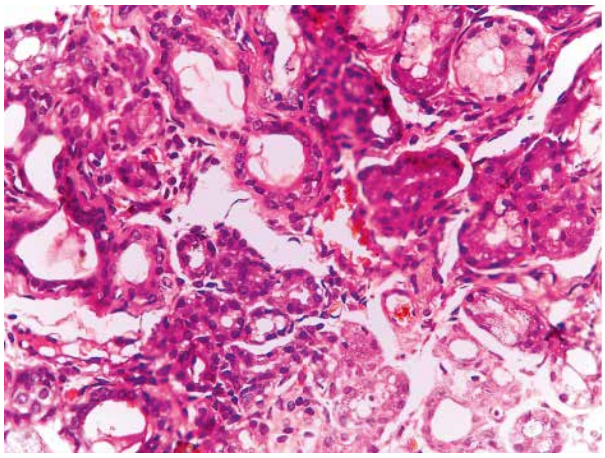


Fig. 3. Photomicrograph characterizing typical cellular atypia, nuclear hyperchromatism as well as dentinoid materials. (H&E stained, Original magnification: 40x).

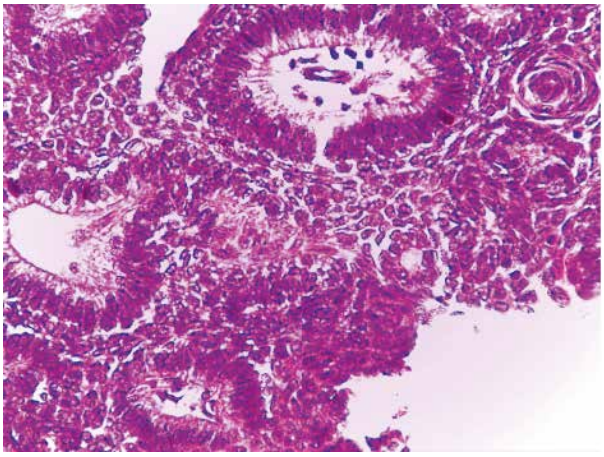


Fig. 4. Photomicrograph showing dentinoid materials interspersed in the tumoral nest (H&E stained, Original magnification: 40x).

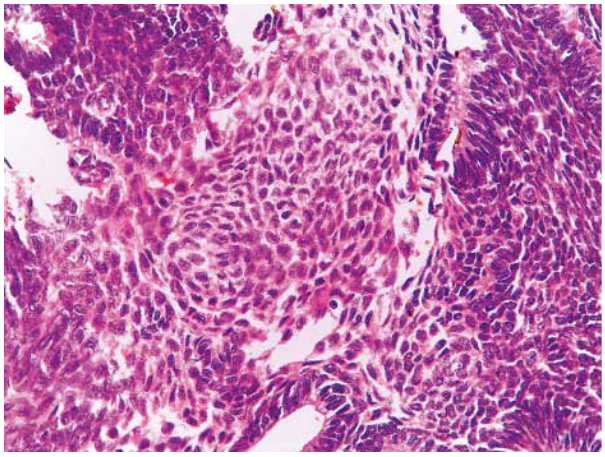
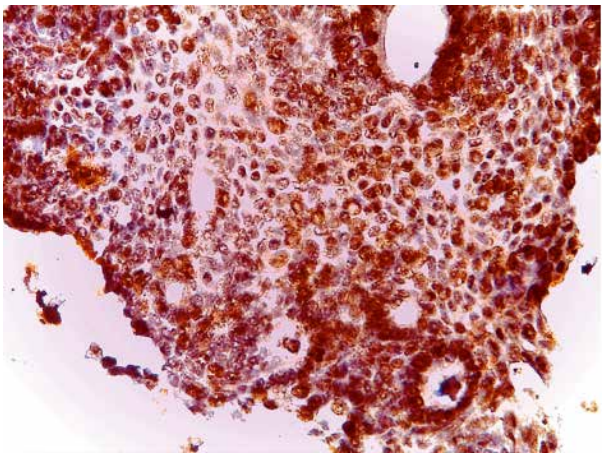


Fig. 5. Photomicrograph showing strong positive expression for p53 (Original magnification: 40x).



14 reported cases in the medical literature [4]. Nevertheless, AOT can be traced in association with other pathoses as well as *per se*. A hybridization of ameloblastoma and AOT was reported, evident [5]. Complicating matters, AOT was homogenously observed in cases where it intermingles with native ameloblastic components. This rarity was designated "adenoid" ameloblastoma (AA) specifies those tumors which reveal impressive occurrence of AOT-like areas [6].

Histologically, AOT is a multi-nodular proliferation of spindle, cuboidal, and columnar cells in a variety of patterns comprising of scattered duct-like structures. Characteristically, eosinophilic materials are observed along with dystrophic calcifications in several forms; delimited by a fibrous capsule of varying thickness [7]. Pertinently, between the epithelial cells of the nodules and in the center of the rosette-like configuration, pools of amorphous amyloid-like material, hyaline, dysplastic

ones, and even, in very rare cases, dentin-like material may exist in both lesional tissue and stromal cells. Given the rare cases of unequivocal recurrent AOT [8], a malignant AOT is unlikely to be expected. Accordingly, the rarity of this previously unreported may open a strong debate regarding the potential transformation.

Immunohistochemically, AOT is strongly positive for amelogenin, ameoblastin and amelotin which can explain the milder aggression, comparable to other odonotgenic tumors. AOT is also positive for podoplanin; accounting for the proliferative activity which is, again, the mildest. However, AOT do not usually stains positively for p53 [4].

In our reported case, there appeared, for the first time, some clear-cut tumoral features which could prompt deeper speculations about the nature of this confusing disease. Cellular atypia was not abundant enough to support a frank malignancy. The strong expression of p53 was another striking caveat which warranted close follow up for atypical peripheral AOT.

Conclusion

Adenomatoid odontogenic tumor can represent more than a hamartomatous nature. Clinicians and pathologists need to reconsider the benign nature of atypical cases.

References

[1] Philipsen HP, Birn H. *The adenomatoid odontogenic tumour. Ameloblastic adenomatoid tumour or adenoameloblastoma*. Acta Pathol Microbiol Scand 1969;75:375-98.

[2] Crivelini MM, Soubhia AM, Felipini RC. *Study on the origin and nature of the adenomatoid odontogenic tumor by immunohistochemistry*. J Appl Oral Sci 2005;13:406-12.

[3] Nomura M, Tanimoto K, Takata T, et al. *Mandibular adenomatoid odontogenic tumor with unusual clinicopathologic features*. J Oral Maxillofac Surg 1992;50:282-5.

[4] Lee SK, Kim YS. *Current concepts and occurrence of epithelial odontogenic tumors: I. Ameloblastoma and Adenomatoid odontogenic tumor*. Korean J Pathol 2013;47:191-202.

[5] Yamazaki M, Maruyama S, Abé T, et al. *Hybrid ameloblastoma and adenomatoid odontogenic tumor: report of a case and review of hybrid variations in the literature*. Oral Surg Oral Med Oral Pathol Radiol 2014;118:e12-8.

[6] Loyola A, Cardoso S, de Faria P, et al. *Adenoid ameloblastoma: clinicopathologic description of five cases and systematic review of the current knowledge*. Oral Surg Oral Med Oral Pathol Oral Radiol 2015;120:368-77.

[7] Philipsen HP, Reichart PA. *Adenomatoid odontogenic tumour: facts and figures*. Oral Oncol 1999;35:125-31.

[8] Rick GM. A*denomatoid odontogenic tumor.* Oral Maxillofacial Surg Clin N Am 2004;16:333-54.