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. Accord-
ingly, ‘AOT’ is no more than a misnomer. This report, however,
describes the first recognition of cellular atypia and pleomorphism
in a peripheral oropharyngeal AOT which embraces an amelo-
blastic component. The overall picture was diagnosed, after care-
ful histological and immunohistochemical assessment, as a periph-
eral adenoid ameloblastoma. This finding may promote a new patho-
genetic scenario to the nosology of this debatable lesion.

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Case report
Adenoid ameloblastoma with dentinoid and cellular
atypia: a case report and literature review
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Key words
Adenomatoid odontogenic tumor • Peripheral adenoid ameloblastoma • Odontogenic tumors

Summary
Adenomatoid odontogenic tumor (AOT) is always benign. Given
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genetic 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 be-
havior, 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.

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 incon-
spicuous. Dentinoid materials were surprisingly remark-
able. 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. Immu-
nohistochemically, 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-in-
vasions. The diagnosis was established as a peripheral
adenoid ameloblastoma.

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

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

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

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. 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, 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 odontogenic 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. 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