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CASE REPORT

Adenomatoid dentinoma or adenomatoid odontogenic hamartoma: what is the better term to denominate this uncommon odontogenic lesion?

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We report two cases of an uncommon odontogenic lesion, previously described as adenomatoid dentinoma. They were well-circumscribed unilocular radiolucent lesions exhibiting discrete radiopacities, located in the left mandibular third molar region. Microscopically they were composed of odontogenic hard and soft tissues, similar to a dental germ. Dental papilla and dentin were easily identified. Odontogenic epithelium formed adenomatoid-like structures, and by scanning electron microscopy a layer of enamel was seen in contact with the dentin. Based on these clinical, radiographic, histological and electron microscopical features we proposed the diagnosis of adenomatoid odontogenic hamartoma. Treatment consisted of surgical removal, and no recurrence was observed. In our opinion all similar cases previously reported pertain to the same spectrum of this lesion and thus should be named as suggested above. Moreover, ultrastructural observations using 5 μm sections can be useful to better characterize the presence of hard tissues.
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Keywords: adenomatoid dentinoma; adenomatoid odontogenic hamartoma; mandible; odontogenic tumors

Introduction

Allen *et al* (1998) described four interesting cases of radiolucent well-circumscribed lesions located in the posterior mandibular region that were called adenomatoid dentinoma. They suggested this term because microscopically the lesion showed adenomatoid epithelial structures associated with dentin. According to the authors no enamel matrix nor dental papilla were observed in these cases. Other authors reported similar cases in the English-language literature but suggested different names as adenoameloblastic odontoma (Dunlap and Fritzlen, 1972), plexiform ameloblastoma with dentinogenesis (Orlowski *et al*, 1991) and adenomatoid odontogenic tumor arising in an odontogenic cyst (Tajima *et al*, 1992).

The aim of this paper is to describe two new cases with identical clinical and radiographic features of adenomatoid dentinoma and based on histology and scanning electron microscopy to suggest a new name to designate this unusual lesion.

Case reports

Case 1

A 16-year-old white boy presented an asymptomatic, well-delimitated, 1×1 cm unilocular radiolucency in the left side of the posterior area of the mandible, just distal to the mandibular left second molar (Figure 1a,b). The patient showed agenesis of the mandibular left and right third molar. On clinical and radiographic examinations no expansion of the cortical bones was detected. After surgical removal, the material was sent for histopathological study. An uninflamed connective tissue capsule surrounded a ring-like deposition of dentin, and a thin layer of calcified material suggestive of enamel was observed (Figure 2). The dental papilla was evident and the inner and outer surface of the dentin had a smooth contour. In the internal surface of the dentin there was a layer of predentine, and cuboidal cells compatible with odontoblasts (Figure 3a). The external area of the calcified material was lined by columnar and/or cuboidal cells that were morphologically consistent with ameloblasts. The ameloblast-like cells were confluent with sheets of stellate reticulum (Figure 3b). Duct-like structures lined by cuboidal and/or columnar epithelial cells were focally seen close to the stellate reticulum area (Figure 4). We performed a scanning

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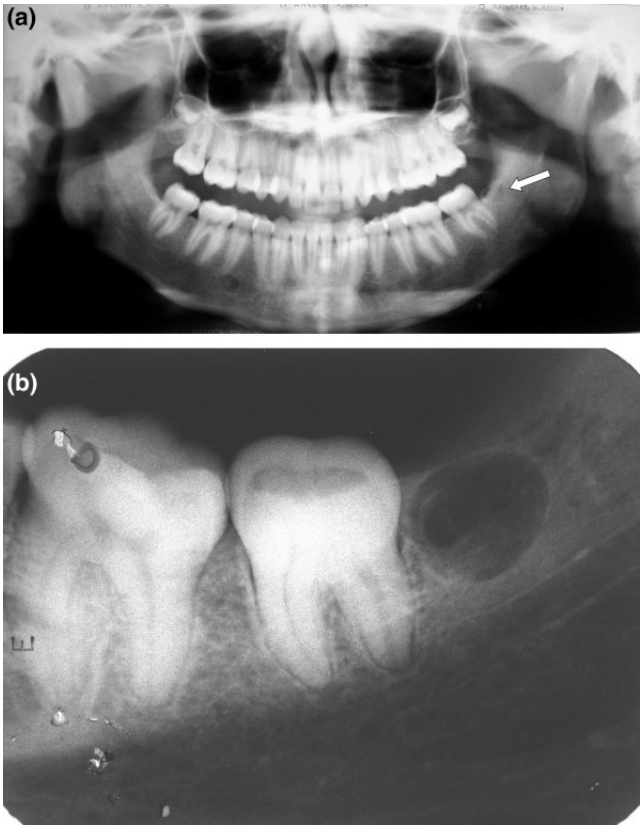

Figure 1 (a) Panoramic X-ray displaying agenesia of the mandibular third molars and a radiolucent lesion in the posterior left mandible (white arrow). (b) Radiograph showing well-circumscribed radiolucency distal to the mandibular left second molar. A very subtle radiopacity can be seen within the lesion

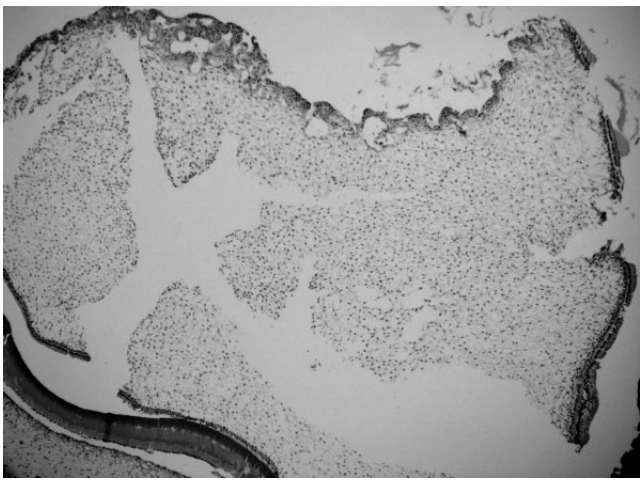

Figure 2 Low-power photomicrograph exhibiting a well-delimitated lesion composed by soft and hard odontogenic tissues. (H&E, original magnification, ×40)

electron microscopic (Jeol JSM 5600 LV, Dental School of Piracicaba-UNICAMP, Brazil) examination using a 5 μm paraffin section as previously described by Tay *et al* (1995). We found an unequivocal enamel matrix

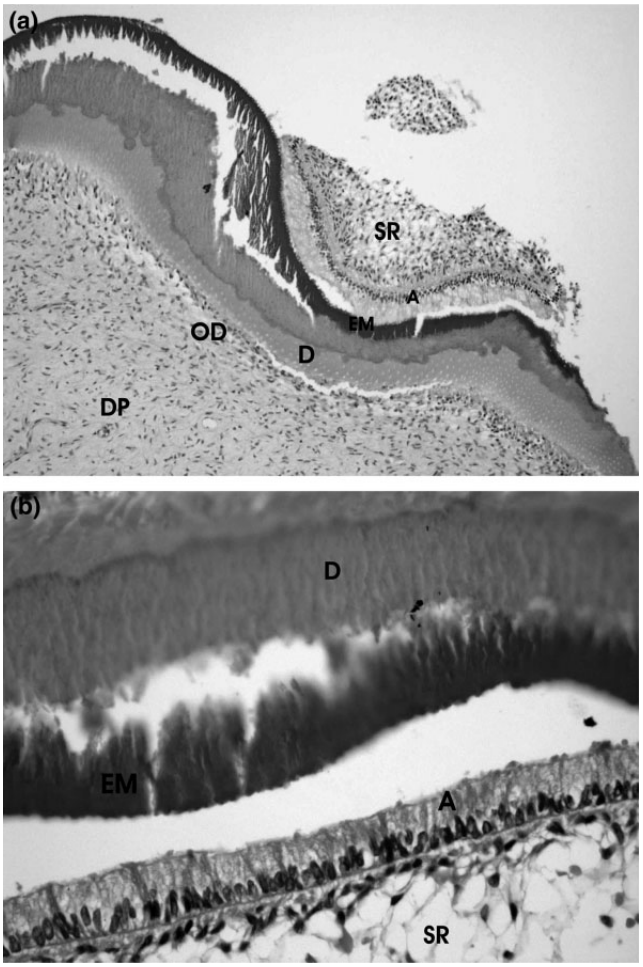

Figure 3 (a) Medium-power view showing dental papilla (DP), odontoblasts (OD), tubular dentin (D), enamel-like matrix (EM), ameloblasts (A) confluent with stellate reticulum (SR) (H&E, original magnification, ×100). (b) High-power view of Figure 3a displaying stellate reticulum (SR), ameloblasts (A), enamel matrix (EM) and dentin (D) (H&E, original magnification, ×200)

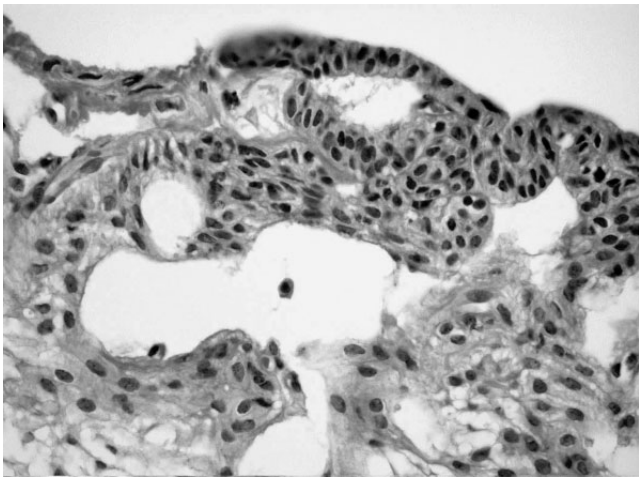

Figure 4 High-power photomicrograph displaying duct-like structures lined by cuboidal or columnar cells in the stellate reticulum area (H&E, original magnification, ×400)

overlying the dentin (Figure 5). Furthermore, we carried out an immunohistochemistry study for cytokeratins 14 (clone: LL002, dilution 1:200; Novocastra, Newcastle-upon-Tyne, UK) and 19 (clone: RCK108, dilution 1:400; Dako, Glostrup, Denmark) to confirm the presence of ameloblasts. Ck 14 was positive in stellate reticulum, ameloblasts and adenomatoid structures, while Ck 19 was only immunoreactive in the ameloblasts (Figure 6). Due to the presence of enamel overlying dentin we preferred the term adenomatoid odontogenic hamartoma (AOH), instead of adenomatoid dentinoma. After 6 months of follow-up, no recurrence has been observed.

Case 2

A 14-year-old healthy white girl was sent for diagnosis and treatment of a well defined, 1.2×1.0 cm radiolucency located distally to the left lower second molar. This lesion was detected as an incidental finding in a

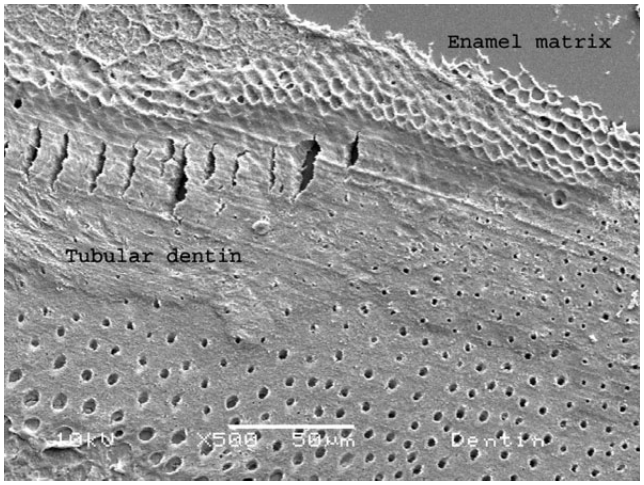

Figure 5 Scanning electron microscopical image showing enamel matrix overlying the tubular dentin (×500)

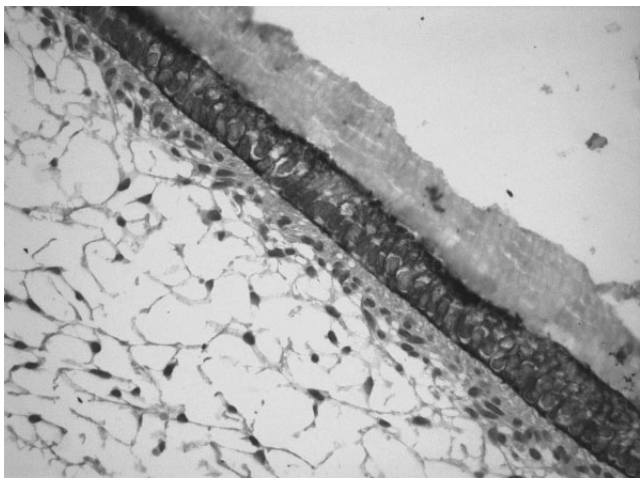

Figure 6 Ameloblasts showing intense immunoreactivity for cytokeratin 19 antibody (immunohistochemistry, original magnification, ×400)

Table 1 Adenomatoid odontogenic hamartomas of the jaws reported in the literature with other nomenclature. Nine cases were reported in five articles

Report	*Odontogenic lesion*	*Gender*	*Age (years)*	*Site*	*Size (cm)*	*Treatment*	*Follow-up*
Dunlap and Fritzlen (1972)	Adenoameloblastic odontoma	F	5	Posterior left mandible	NA	Enucleation	NA
Orlowski *et al* (1991)	Plexiform ameloblastoma with dentinogenesis	F	42	Right mandible	1.5×1.4	Enucleation	NA
Tajima *et al* (1992)	AOT arising in an odontogenic cyst	M	15	Left maxillary sinus	4.0×4.0	Enucleation	FOD 5 years
Allen *et al* (1998)	Adenomatoid dentinoma	M	37	Left mandibular third molar region	1.5×2.0	Enucleation	FOD 2 years
		M	29	Left mandibular third molar region	0.75×0.75	Enucleation	FOD 8 years
		M	35	Left mandibular third molar region	1×0.8	Curettage	FOD 3 years
		F	29	Right mandibular third molar region	2×2.75	Enucleation	FOD 8 years
Vargas *et al* (this study)	Adenomatoid odontogenic hamartoma	M	16	Left mandibular third molar region	1×1	Curettage	FOD 6 months
	Adenomatoid odontogenic hamartoma	F	14	Left mandibular third molar region	1.2×1	Curettage	FOD 7 years

NA, not available; FOD, free of disease; AOT, adenomatoid odontogenic tumor.

panoramic radiograph taken for orthodontic evaluation. On close examination, a subtle linear radiopacity was seen within the lesion, but there was no root resorption or any evidence of inflammatory changes on adjacent periodontal or mucosal structures. There was anodontia of all four third molars. The lesion was enucleated and no recurrence has been detected 7 years postoperatively. Macroscopic examination disclosed a white-yellowish spheroidal soft tissue specimen that measured $1.2 \times 1.0 \times 1.0$ cm and was soft in consistency. The light and scanning electron microscopy identified the same features already reported in Case 1. Based on these features we diagnosed this case as AOH.

Discussion

Table 1 shows that the mean age of patients with AOH in the jaws is nearly 25 years (range 5–42 years), therefore affecting mainly young adult patients. There is a slight male predilection. The main site is the mandibular third molar region (eight cases), followed by only one case involving the left maxillary sinus. Clinically, all the reported lesions were asymptomatic. Three of nine AOH reported cases showed clinical history of previous surgical removal of the mandibular third molar of the affected region and three other cases presented agenesia of the mandibular third molar. AOH size ranged from 0.75 to 4.0 cm in diameter, with an average of 1.6 cm. Surgical enucleation has been the treatment of choice and no recurrences have been described heretofore.

All lesions reported in the literature with similar clinical and radiographic features did not show evident enamel matrix in H&E examination, and we are convinced this is the main reason for different names for the same lesion. Allen *et al* (1998) used the term adenomatoid dentinoma because enamel and ameloblasts were not found. However, in our both cases enamel matrix was unequivocally seen by scanning electron microscopical examination, and this finding associated with the clinical, radiographic and microscopical characteristics prompt the diagnosis of AOH. These lesions resemble a tooth in development, favoring the term AOH instead of adenomatoid dentinoma. Moreover, the small size of AOH (1.6 cm) reflects its limited growth potential and non-neoplastic nature. In our opinion a better definition for AOH would be a non-neoplastic hamartomatous proliferation of mature hard and soft dental tissues interspersed with remnants of odontogenic epithelium forming duct-like structures, which tend to occur mainly in the mandibular third molar region.

In conclusion, the current cases are similar to what has been described as adenomatoid dentinoma, however, in our opinion the better term to denominate this lesion is AOH. All similar lesions should be submitted to scanning electron microscopical analysis to better identify the hard tissues. Based on the clinical, radiographic and microscopical features, it seems that adenomatoid dentinoma and other similar lesions called by different terms represent a spectrum of the same odontogenic lesion that we propose to name AOH.

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