A case of peripheral ameloblastoma of retromolar trigone: Histopathological and immunohistochemical profile

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Abstract

Peripheral ameloblastoma (PA) is a rare and unusual entity with histological characteristics similar to those of the common intraosseous ameloblastoma. In this paper, we present a case of PA in a 44-year-old male affecting the right retromolar trigone area along with its immunohistochemical profile using CK19 and Ber-EP4 markers.

Keywords: Ameloblastoma, immunohistochemistry, peripheral, retromolar trigone

Introduction

The peripheral ameloblastoma (PA) is generally described as an exceedingly rare lesion, which accounts for 1–5% of all ameloblastomas.[1] The PA is also known as the extra-osseous ameloblastoma, soft tissue ameloblastoma, ameloblastoma of mucosal origin, or ameloblastoma of the gingiva. PA shows several histologic characteristics of an intra-osseous, infiltrating ameloblastoma (IA) but it occurs in soft tissues overlying and does not invade the underlying bone.[2]

Case Report

A 44-year-old male patient presented to the Department of Oral Pathology and Microbiology with a firm soft-tissue lesion behind the maxillary right third molar. The patient has been aware of the lesion since last 1-year and the lesion was gradually increasing in size.

There was no extraoral swelling on the right side of face or associated lymph node enlargement. On intraoral examination, there was no obvious lesion noted. Overlying mucosa was normal in color without any obvious pathology. On palpation, a firm submucosal mass was revealed posterior to maxillary right third molar of approximately 2 cm × 2 cm in size. Lesion was spherical in shape and not fixed to the underlying structures [Figure 1].

Intraoral periapical showed no evidence of any bone involvement. Magnetic resonance images report revealed a well-defined lesion of 1.8 cm × 1.6 cm in the right retromolar trigone with no obvious erosion of bony surface. It appears as hypointense signal T1-weighted image [Figure 2a], and hyperintense signal on short tau inversion recovery image [Figure 2b]. On suspecting a benign soft tissue tumor of salivary gland, a peripheral odontogenic tumor or a tumor like-growth, incisional biopsy was performed.

H and E stained sections showed dense connective tissue stroma containing numerous islands and cords of odontogenic epithelium [Figure 3]. Peripheral cell were tall columnar/cuboidal, palisaded, and polarized with stellate-like cells present in the center of the islands showing squamous metaplasia.

The histologic findings were suggestive of peripheral acanthomatous ameloblastoma. To further confirm the diagnosis, immunohistochemical analysis was done using CK19 and Ber-EP4 markers [Figure 4]. Lesional areas showed strong positive expression for CK19 while negative expression for Ber-EP4.

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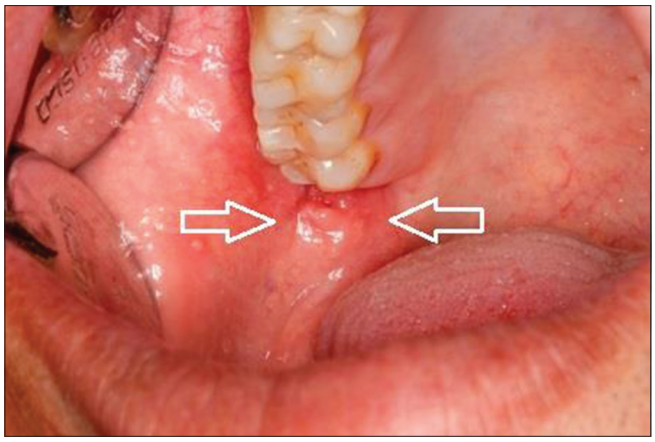

Figure 1: Intraoral view of 2 x 2 cm lesion in right retromolar trigone area

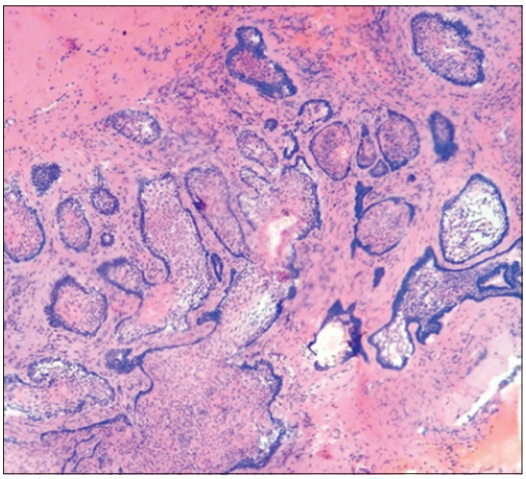

Figure 3: Islands of odontogenic epithelium showing microcysts and central stellate reticulum-like cells with squamous metaplasia (H and E stain, ×10)

Discussion

PA can be confused clinically with an epulis, fibroma, gingival tumor, or carcinoma including intraoral basal cell carcinoma (BCC) and hence difficult to diagnose based only on clinical findings. PA is frequently diagnosed only after a histological examination.[3] Similarly in our case, we were not able to suspect PA based only on clinical findings and only after histopathological examination, the final diagnosis was achieved.

Bone involvement is noted in only few cases of PA, which has been referred to as cupping or saucerization which results from the pressure of the tumor on bone, while most of the cases of PA are superficial to cortical bone with no sign of bone involvement.[3] In our case, no bone resorption was seen.

PA occurs at a significantly higher age and is more common in males as compared to IA[2,4] [Table 1].

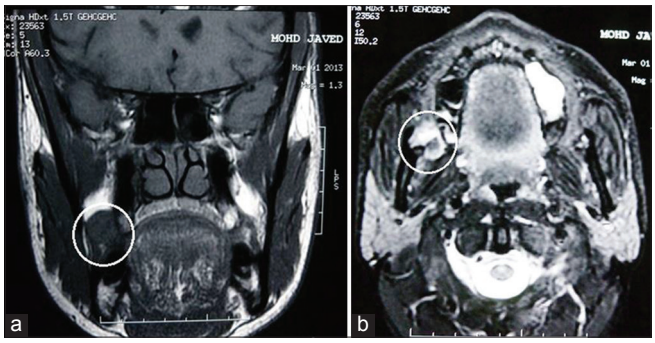

Figure 2: (a) MRI shows a well-defined lesion of 1.8 x 1.6 cm in the right retromolar trigone. T1 weighted image showing hypointense signal. (b) lesion appear as hyperintense signal on STIR (short TI inversion recovery) image

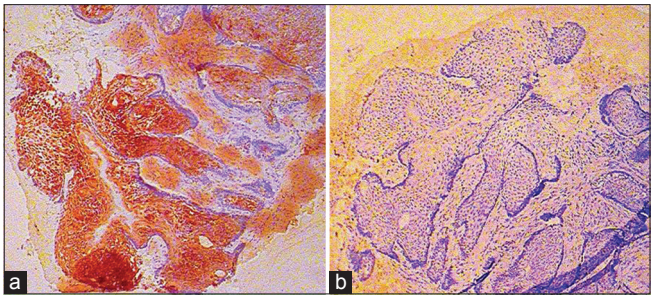

Figure 4: (a) Lesional tissue showing strongly positive expression for CK19 (×10) (b) lesional tissue showing negative expression for Ber-EP4 (×10)

The most common site for PA is mandibular premolar region (32.6%) followed by anterior mandibular region (20.7%) and maxillary soft palatal tissue of the tuberosity area (11.1%). In our case, location of lesion was right retromolar trigone.

Similar to IA, PA may exhibit various histological patterns. According to Gardner,[5] PA has a marked tendency to be acanthomatous as seen in our case.

Regarding the cellular origin of PA, two major sources are usually considered. First, lesions which are located entirely within the connective tissue of the gingiva, showing no continuity with the surface epithelium, most likely arise from remnants of the dental lamina located in the soft tissues overlying the tooth-bearing areas of the jaw bones.[2] Second, lesions may arise from the surface epithelium, in some cases at one or a few sites and in others multifocally.[6] In the present case, as there was no continuity of the lesional tissue with the surface epithelium, hence it appears to arise from the remnants of the dental lamina.

Immunohistochemical profile of peripheral ameloblastoma

Kato *et al*.[3] performed immunohistochemical (IHC) studies to further evaluate PA. Their findings are summarized in Table 2.

Previous studies have shown that PA and IA are positive for CK19 and negative for Ber-EP4, whereas the opposite

Table 1: Comparison between PA and IA

Parameter	PA (Philipsen *et al*.[2])	IA (Reichart *et al*.[4])
Average age (years)	52.1	37.4
Male/female ratio	1.9:1	1.2:1
Maxilla/mandible ratio	1:2.4	1:4
Recurrence rate (%)	19	33

PA: Peripheral ameloblastoma; IA: Intraosseous ameloblastoma

Table 2: Immunohistochemical profile of PA

Marker	Result	Remarks
CK14	++	Major cytoskeletal polypeptide in ameloblastomas
CK19	+	Marker of odontogenic origin
EMA	-	Labels normal and neoplastic epithelium
p53	-	Tumor suppressor protein
p63	+	Role in epithelial development
Ber-EP4	-	Labels most epithelial cells and is expressed in neoplastic basal cells
Ki-67	LI: 2.22%	For IAs it is 1.37%, this indicates low growth potential of PA, supporting the benign biological behavior of these lesions

PA: Peripheral ameloblastoma; IAs: Intraosseous ameloblastomas; LI: Labeling index

is true for BCC. In the study done by Kato *et al*., all cases were negative for Ber-EP4, suggesting that PA and IA are tumors with a common origin, and that PA is derived from odontogenic epithelial remnants, rather than from basal cells of the oral epithelium.[3]

Thus IHC markers such as CK19 and Ber-EP4 can be used to differentiate PA from intraoral BCC. In our case, CK19 [Figure 4a] was expressed strongly positive while Ber-EP4 [Figure 4a] showed negative expression as shown in previous reports. These findings further confirm our diagnosis of PA.

Treatment and prognosis

Gardner (1977)[5] stated that term PA is potentially dangerous in that this diagnosis may lead to unnecessarily aggressive treatment. The current treatment of choice is conservative supraperiosteal surgical excision with adequate disease-free margins.[2] In the present case, the patient denied any form of surgical treatment and hence is kept on regular follow-ups.

Because of the common perception that PA does not exhibit the persistent growth and invasiveness of IA, it is usually assumed that lesion will not progress after excision. However, it has been seen that some tumors indeed have significant invasive capacity with high recurrence potential. According to Philipsen *et al*. (2001),[2] a total of six cases of a very rare malignant variety of PA have been published.

Ide *et al*. (2004)[7] reported the first completely documented example of malignant PA with metastasis and they suggested that large size (over 2 cm in diameter) is a powerful predictor of aggressive behavior, no matter how apparently innocuous.

Conclusion

PA is a rare tumor that is histologically identical to IA usually treated by conservative supra-periosteal excision. Long-term follow-up is advised to detect late local recurrences and metastatic transformation.

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Conflicts of interest

There are no conflicts of interest.

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