CASE REPORT

Peripheral ameloblastoma of gingiva with cytokeratin 19 analysis

Shivjot Chhina,¹ Ajit Singh Rathore²

¹Department of Periodontics, ITS Dental College, Hospital & Research Centre, Greater Noida, Uttar Pradesh, India

²Department of Oral Pathology, ITS Dental College, Ghaziabad, Uttar Pradesh, India

Correspondence to Professor Shivjot Chhina, drshivjot74@gmail.com

Accepted 17 May 2015

SUMMARY

A 42-year-old woman presented with a gingival mass in the lingual vestibule of the mandibular incisor premolar region. On intraoral examination, the swelling was non-tender and firm. Surgical excision was carried out and subsequent histopathological examination revealed areas resembling ameloblastoma-like and basaloid areas with atypical features suggestive of basal cell carcinoma. Immunohistochemical analysis was carried out to ascertain the origin of the lesion.

BACKGROUND

These lesions rarely occur in the gingiva. Owing to the overlapping histological features with basal cell carcinoma (BCC) and ameloblastic carcinoma, diagnosis is challenging. A proper assessment of the case by conferring with a periodontist and an oral pathologist is warranted, so as delineate a proper treatment plan.

CASE PRESENTATION

A 42-year-old woman presented with a painless gingival swelling. The lesion presented on the lingual aspect extending from the permanent mandibular right incisor up to the mesial of the right mandibular first molar. The patient reported of difficulty in speech, as the lesion was interfering with her tongue. On intraoral examination, the swelling was non-tender and firm, and measured 2×1.5 cm (approximately) ([figure 1](#)). General examination was unremarkable and no associated lymphadenopathy was detected.

INVESTIGATIONS

Haematological and biochemical values were in the normal range. Orthopantomogram and intraoral periapical X-ray did not reveal any bony involvement.

DIFFERENTIAL DIAGNOSIS

The preoperative differential diagnosis included: gingival tumours, such as peripheral ossifying fibroma, peripheral odontogenic fibroma and inflammatory hyperplastic lesions such as pyogenic granuloma or peripheral giant cell granuloma.¹

Peripheral ossifying and odontogenic fibromas show varied clinical appearances that may closely resemble those of peripheral ameloblastoma (PA). Therefore, clinical features of these lesions are not pathognomonic, and histopathological examination is essential for the definitive diagnosis. On the other hand, peripheral fibromas usually lead to a

separation between adjacent teeth and radiopaque foci, which can be appreciated radiographically.²

Inflammatory gingival hyperplasias are a second group of conditions that need to be considered in the differential diagnosis. Commonly, these are associated with chronic irritants, such as calculus or overhanging margins of dental restorations, which on elimination usually result in regression. Pyogenic granuloma is an asymptomatic mass that appears red and bleeds readily and presents a rough, ulcerated necrotic surface. Peripheral giant cell granuloma appears red, pink or bluish in colour, is most often nodular or polypoid in shape, and may be soft to hard in consistency.²

PA presents histological characteristics that can be similar to BCC, basaloid variant of squamous cell carcinoma and ameloblastic carcinoma (dedifferentiated),³ as these tumours exhibit similar basal cell proliferation forming epithelial strands. PAs usually arise from the gingival epithelium,^{4 5} whereas oral BCCs rarely, if ever, originates from gingival epithelium.^{6 7} BCC is a neoplasm occurring on the skin, with a variable morphology and a wide age range of incidence. Therefore, previously reported cases of so-called intraoral BCCs (of the gingiva) may, in retrospect, be considered to be ameloblastic carcinoma of the secondary type (dedifferentiated).⁸ Ameloblastic carcinoma, however, can be differentiated due to presence of mitotic figures, areas of necrosis and varied degree of anaplasia.

TREATMENT

Complete surgical excision of the lesion followed by curettage was carried out.

Figure 1 Preoperative view showing swelling of about 2×1.5 cm in lingual aspect of left mandibular central incisor to distal of second premolar.

CrossMark

To cite: Chhina S, Rathore AS. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-210227

Figure 2 Photomicrograph showing overlying epithelium and sheets of tumour cells forming a follicular and basaloid pattern (H&E, ×40).

OUTCOME AND FOLLOW-UP

Microscopic evaluation revealed parakeratinised stratified squamous epithelium with basal cell layer showing proliferation at multiple sites into the underlying connective tissue as strands, nests and islands of tumour cells. Follicular islands showed peripheral palisading, with cells in the centre resembling stellate reticulum. Other areas displayed a basaloid appearance with a few atypical cells (figure 2). Cytokeratin (CK) 19 analyses revealed expression in the basal cells, stellate reticulum cells and a few peripheral cells (figure 3). These features are considered characteristic of PA.

The patient has been on regular, periodic follow-up for more than 2 years and has not presented with any recurrence of the lesion.

DISCUSSION

PA is a painless, sessile, firm and exophytic growth with no radiographic involvement, as also seen in the present case. However, a few of the reported cases have shown radiographically superficial erosion, depression or saucerisation of the involved bone.⁵ The mandible is a more common site than the maxilla, with the ratio being 2.5:1. In the mandible, the lingual aspect of the premolar region, with 32.6% incidence, is the most common site for PA, as described in our case, followed by the anterior mandibular region, with 20.7% incidence. In the maxilla, the soft palatal tissue of tuberosity accounts for 11.1% of cases. The male to female ratio is 1.9:1 compared with 1.2:1

Figure 3 Photomicrograph showing positive cytokeratin 19 expression in basal cell layer of epithelium and the proliferating strands and tumour islands (H&E, ×40).

seen in solid multicystic ameloblastoma (SMA).⁹ Mean age of occurrence is 52.1 years with mean age of men 52.9 years, slightly higher than that of women, for whom it is 50.6 years; however, in the present case, the age of occurrence was 42 years.

Microscopic examination revealed a tumour consisting of proliferating odontogenic epithelium that exhibited the same histomorphologic cell types and patterns seen in SMA. In some cases, a basaloid lesion without classic follicular component is seen, which shows a similarity to BCC.

Two main theories regarding the histogenesis of PA are conjectured. Some tumours are located completely within the gingival connective tissue derived from the dental lamina.^{5 10 11} On the other hand, many cases of PA present in a very close relationship with the surface epithelium, suggestive of an origin from the surface epithelial layer.¹²

Immunohistochemical analysis using CK, mainly CK 19, has been carried out to ascertain the odontogenic origin as opposed to origin from the basal cell of gingival epithelium. CK 19 expression is seen in basal cells, suprabasal cells and stellate reticulum-like cells of the tumour,^{10 13} as also discerned in the present case. A study by Hakamawa gave insight into CK 19 expression in poorly differentiated carcinoma of the oral cavity, suggesting that tumour cells retain an immature phenotype of their differentiation.¹⁴

The current treatment of choice for these lesions is a conservative subperiosteal surgical excision with adequate disease-free margins. Recurrence rate is 16–19%, which is lower than that of SMA.^{15 16} Most commonly, recurrence is attributed to incomplete removal. Long-term follow-up is necessary, as cases of benign PA have been reported to recur and present as ameloblastic carcinoma.^{17 18}

Learning points

- ▶ Although uncommonly, peripheral ameloblastoma does present in the gingiva and should be considered as a part of the differential diagnosis.
- ▶ Atypical areas with mitotic activity need to be analysed, as a very thin line differentiates peripheral ameloblastoma from ameloblastic carcinoma.
- ▶ Follow-up is mandated as a few of the reported cases have recurred as ameloblastic carcinoma.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Mintz S, Anavi Y, Sabes WR. Peripheral ameloblastoma of gingiva: a case report. *J Periodontol* 1990;61:649–52.
- 2 Wood NK, Goaz P. *Differential diagnosis of oral lesions*. 3rd edn. St. Louis: The CV Mosby Company, 1985:148–58.
- 3 Niccoli-Filho W, Gomes Mda G, Raldi FV, et al. Peripheral ameloblastoma. *J Nihon Univ Sch Dent* 1997;39:34–7.
- 4 Gardner DG. Peripheral ameloblastoma: a study of 21 cases, including 5 reported as basal cell carcinoma of the gingiva. *Cancer* 1977;39:1625–33.
- 5 Philipsen HP, Reichart PA, Nikai H, et al. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. *Oral Oncol* 2001;37:17–27.
- 6 Koutlas IG, Koch CA, Vickers RA, et al. An unusual ostensible example of intraoral basal cell carcinoma. *J Cutan Pathol* 2009;36:464–70.

- 7 Nishimura N, Sakurai K, Noguchi K, *et al.* Keratotic basal cell carcinoma of the upper gingiva with cervical lymph node metastasis: report of a case. *J Oral Maxillofac Surg* 2001;59:677–80.
- 8 Philipsen HP, Reichart PA, Slootweg PJ, *et al.* eds. *World Health Organization classification of tumors. Pathology and genetics head and neck tumors*. Lyon, France: IARC Press, 2005.
- 9 Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol* 1995;31B:86–99.
- 10 Kishino M, Murakami S, Yuki M, *et al.* An immunohistochemical study of the peripheral ameloblastoma. *Oral Dis* 2007;13:575–80.
- 11 Vanoven BJ, Parker NP, Petruzzelli GJ. Peripheral ameloblastoma of the maxilla: a case report and literature review. *Am J Otolaryngol* 2008;29:357–60.
- 12 Ide F, Mishima K, Miyazaki Y, *et al.* Peripheral ameloblastoma in-situ: an evidential fact of surface epithelium origin. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;108:763–7.
- 13 Kato H, Ota Y, Sasaki M, *et al.* Peripheral ameloblastoma of the lower molar gingiva: a case report and immunohistochemical study. *Exp Clin Med* 2012;37:30–4.
- 14 Tajima Y, Kuroda-Kawasaki M, Ohno J, *et al.* Peripheral ameloblastoma with potentially malignant features: a report of case with special regard to keratin profile. *J Oral Pathol Med* 2001;30:494–8.
- 15 Redman RS, Keegan BP, Spector CJ, *et al.* Peripheral ameloblastoma with unusual mitotic activity and conflicting evidence regarding histogenesis. *J Oral Maxillofac Surg* 1994;52:192–7.
- 16 Buchner A, Sciubba JJ. Peripheral epithelial odontogenic tumors: a review. *Oral Surg Oral Med Oral Pathol* 1987;63:688–97.
- 17 Baden E, Doyle JL, Petriella V. Malignant transformation of peripheral ameloblastoma. *Oral Surg Oral Med Oral Pathol* 1993;75:214–19.
- 18 Fujita S, Anami M, Satoh N, *et al.* Cytopathologic features of secondary peripheral ameloblastic carcinoma: a case report. *Diagn Cytopathol* 2011;39:354–8.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow