CASE REPORT

PERIPHERAL AMELOBLASTOMA: DIAGNOSTIC DILEMMA- A CASE REPORT

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ABSTRACT

Ameloblastomas are benign epithelial odontogenic neoplasms which are locally aggressive. Based on the clinical, radiographical, histopathological, behavioral and prognosis, four variants of ameloblastoma are present. They include solid/multicystic ameloblastoma, unicystic ameloblastoma (UA), peripheral ameloblastoma (PA) and desmoplastic ameloblastoma. Peripheral ameloblastoma is a rare, benign, extraosseous odontogenic soft tissue tumor that accounts for 2-10% of all ameloblastomas. It usually occurs in the gingiva or alveolar mucosa. The PA represents the same histological characteristics of intraosseous ameloblastoma, although it is less aggressive than this classical subtype. Here, a case of peripheral ameloblastoma in a 48-year-old-male affecting the lingual alveolar mucosa of the mandibular premolars is presented which was clinically diagnosed as pyogenic granuloma and also described about the importance of histological examination to the diagnosis.

KEY WORDS

Peripheral ameloblastoma, odontogenic neoplasm

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INTRODUCTION

Ameloblastomas are a group of epithelial odontogenic tumors which are benign, locally aggressive in nature and exhibit diverse clinicoradiological and histological features. Ameloblastoma accounts for 30% of all odontogenic tumor [1]. Tumors may originate from the rests of dental lamina, enamel organ, lining or walls of nonneoplastic odontogenic cyst and also from the basal layer of oral epithelium[2]. Peripheral ameloblastoma (PA) is a rare, benign, extraosseous odontogenic soft tissue tumor that exhibits the same histological characteristics as solid/multicystic ameloblastoma. The peripheral ameloblastoma (PA) was first reported in the literature by Stanley and Krogh in 1959 [3].The clinical appearance of the PA may vary, but most of time it represents clinically as a painless, exophytic, slow-growing, firm, painless, sessile or pedunculated based mass with a smooth to granular surface, pink to dark red in colour, pebbly or warty in appearance and is localized to the soft tissues overlying the tooth-bearing areas of the jaws. The overall average age is 52.1 years, slightly higher for males (52.9 years) than for females (50.6 years).The male/female ratio is 1.9:1. According to the location of PA, the maxilla/mandible ratio is 1: 2.6. Gingiva of the mandibular premolar region is the most commonly affected site [4, 5]. It has also been reported at unusual site like buccal mucosa[6], base of tongue[7], and floor of the mouth[8], regardless common site being mandibular lingual gingival. Radiographically, it may exhibit a "cupping" effect due to the pressure resorption[9-13]. Here, a case of peripheral ameloblastoma in a 48-year-old male is presented that occurred in the right lower lingual alveolar mucosa of the mandible.

CASE REPORT

A 48-year-old male was referred to the Department of Oral Pathology at the Dr. R. Ahmed Dental College and Hospital (Kolkata-14) with three months history of a painless, slowly growing mass in the right lingual aspect of mandible. There was no past medical history.

Upon first visit to the Oral Pathology OPD, a

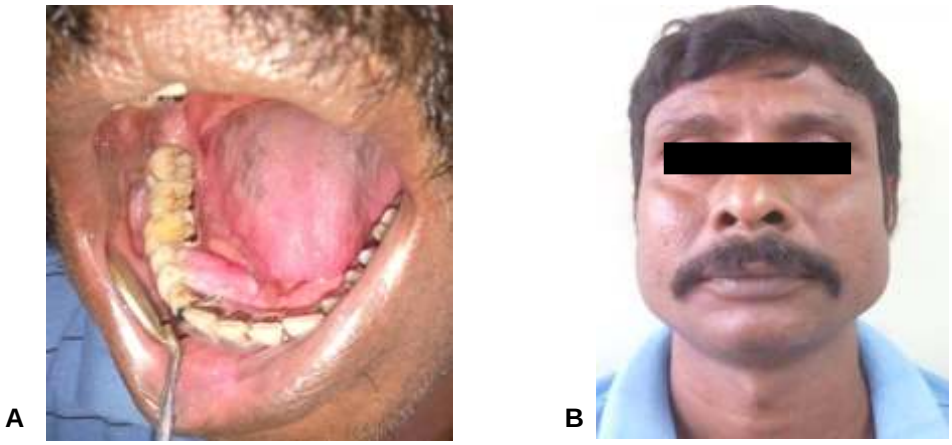

Fig 01: A. showing intraoral B. extraoral clinical picture

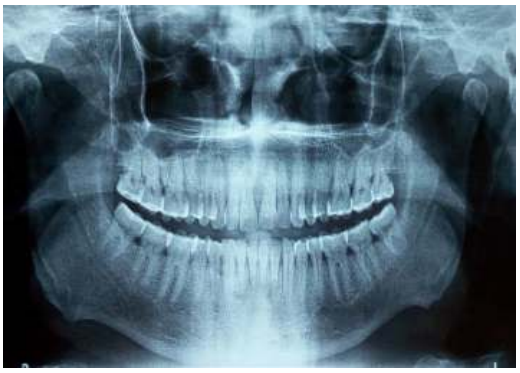

Fig 02: OPG showing an ill defined radiolucency in relation to lower right premolars

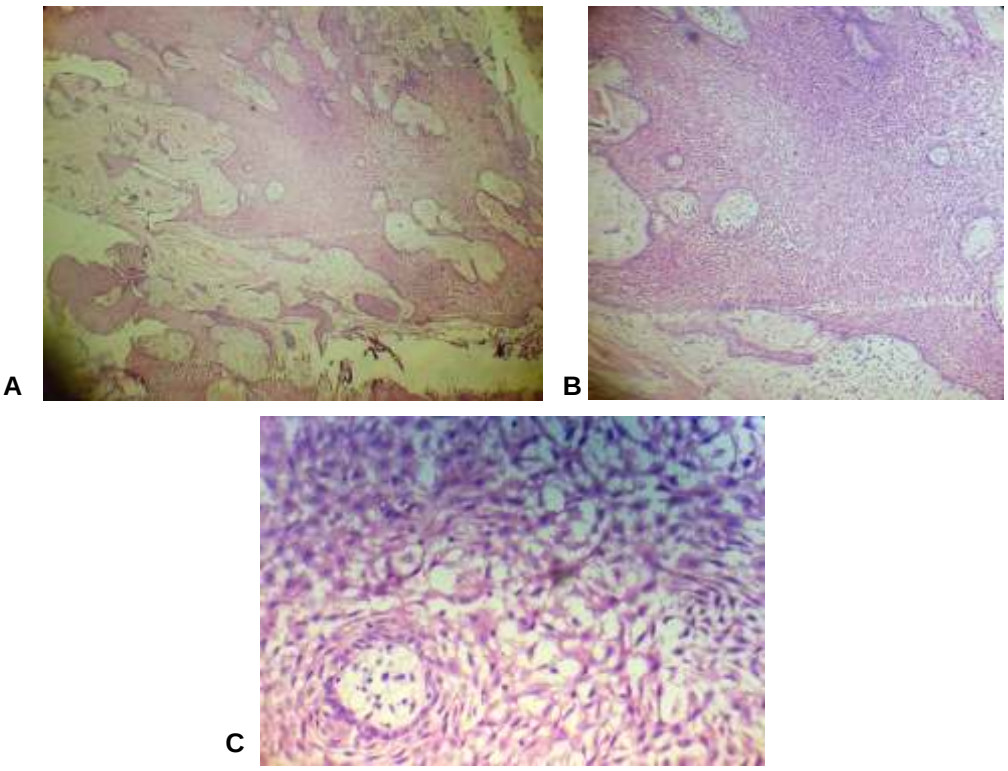

Fig 03: showing H & E stained section of peripheral ameloblastoma
A. low power view B. medium power view C. high power view

thorough clinical examination revealed a sessile based, pebbly surfaced, normal mucosa color, exophytic growth in the lingual aspect of right lower premolars that was firm in consistency (Fig 01A). It was approximately 1.5 cm x1 cm in diameter. An extraoral examination revealed no significant clinical findings (Fig 01B).

After thorough clinical examination, patient was advised for complete blood count, blood sugar estimation both Fasting and PP, bleeding and clotting time, ESR, antibodies for HIV, Hepatitis B and C. Radiological investigation (orthopantomogram) was done (Fig 02).

The investigation reports showed CBC, blood sugar both F and PP, BT, CT, ESR were within normal limit. Viral markers for hepatitis B, C and HIV were negative. The orthopantomogram revealed a cup shaped, ill-defined, radiolucent area in relation to lower right premolars. No root resorption is seen with the associated teeth.

The lesion was provisionally diagnosed as pyogenic granuloma. Then the patient was advised for incisional biopsy for conform the diagnosis.

Section stained with hematoxylin and eosin reveals the presence of hyperplastic, hyperparakeratinized stratified squamous epithelium with broad bulbous rete-pegs. The underlying connective tissue stroma is paucicellular, mature collagenous in nature containing ameloblastic epithelium growing in plexiform pattern. The ameloblastic epithelim contains peripherally tall columnar ameloblast like cells and centrally stellate reticulum like cells. Thus, the overall histopathological findings were suggestive of peripheral ameloblastoma (Fig 03A-03C).

After histopathological diagnosis, the patient was sent to the department of Oral and Maxillofacial Surgery for complete excision of the lesion. After a 3-month follow-up of the patient, no complication or recurrence was reported.

The patient remains well.

DISCUSSION

PA is usually a rare benign, slow-growing tumor with no invasive potential. It accounts for 2%–10% of all ameloblastomas[14]. The pathogenesis of PA has been described as the remnants of dental lamina, the so-called "glands of Serres," odontogenic remnants of the vestibular lamina, pluripotent cells in the basal cell layer of the mucosal epithelium or pluripotent cells from minor salivary glands[6]. A recent investigation demonstrated that alterations of the ameloblastin gene forms the genetic basis for ameloblastoma[10]. PA should be differentiated from peripheral reactive lesions such as pyogenic granuloma, epulis, papilloma, fibroma, peripheral giant-cell granuloma, peripheral odontogenic fibroma, peripheral-ossifying fibroma, Baden's odontogenic gingival epithelial hamartoma, and basal cell carcinoma[15]. Most of the time,presence of stellate reticulum like cells and ameloblast like cells with reverse polarity may suggest a diagnosis of peripheral ameloblastoma. Hence, a careful histopathological evaluation is necessary to differentiate PA from all possible cases. In the present case, its clinical and radiological findings were confused with pyogenic granuloma. Biopsy of the lesion confirmed the diagnosis of a PA. Microscopically, the lesion showed multifocal extensions of surface epithelium that maintained continuity with the ameloblastomatous tissue giving rise to a trabecular or plexiform pattern. Though malignant transformation is rare, metastasis has also been reported[10]. The current treatment of choice is conservative supra periosteal surgical excision with adequate disease-free margins[16]. Continuous follow up is necessary as late recurrence also reported by Later, Buchner and Sciubba in 9% of cases[17]. In this case, complete surgical excision was done, and patient is on continuous follow-up since last 3 months with no recurrence noted so far.

CONCLUSION

The clinical and radiographic findings of PA are varied and often not pathognomonic, concurrent histopathological findings allow for the definitive diagnosis. This type of lesion should be diagnosed carefully because of their clinical, radiological, histopathological overlapping features with some benign and malignant lesions, but their prognosis and treatment protocols are completely different. Hence, a careful clinical and radiological evaluation along with a precise histopathological examination must be carried out.

Throughout, microscopic examination of the specimens are also needed to ensure that the margins are clear of the tumor. Treated cases of PA should be followed up for a longer duration to detect the late local recurrence.

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REFERENCE

1. A. V. Jones and C. D. Franklin, "An analysis of oral and maxillofacial pathology found in adults over a 30-year period," Journal of Oral Pathology and Medicine, vol. 35, no. 7, pp. 392– 401, 2006.

2. Neville BW, Damm DD, Allen CM, Bouquot JE.

Odontogenic cysts and tumors. In: Oral and Maxillofacial Pathology. 2nd ed. St. Louis: W.B. Saunders Company; 2002. p.610-8.

3. Stanley HR Jr. , Krogh HW. Peripheral ameloblastoma of the gingiva. Report of a case. Oral Surg Oral Med Oral Pathol.1959; 12: 760-5.

4. el-Mofty SK, Gerard NO, Farish SE, Rodu B. Peripheral ameloblastoma: A clinical and histologic study of 11 cases. J Oral Maxillofac Surg 1991;49:970-4.

5. Bertossi D, Favero V, Albanese M, De-Santis D, Martano M, Padovano-di-Leva A, et al. Peripheral ameloblastoma of the upper gingiva: Report of a case and literature review. J Clin Exp Dent 2014;6:e180-4.

6. E. T. Isomura, M. Okura, S. Ishimoto et al., "Case report of extragingival peripheral ameloblastoma in buccal mucosa," Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontology, vol. 108, no. 4, pp. 577–579, 2009.

7. B. C. Rajesh, A. R. Vinayakumar, J. Mathai, S. Valsaladevi, and P. Prathapan Nair, "Peripheral ameloblastoma involving tongue - A rare case," Indian Journal of Otolaryngology and Head and Neck Surgery, vol. 50, no. 4, pp. 387–389, 1998.

8. K. Ramnarayan, R. G. Nayak, and A. G. Kavalam, "Peripheral ameloblastoma," International Journal of Oral Surgery, vol. 14,no. 3, pp. 300–301, 1985.

9. Mintz S, Anavi Y, Sabes WR. Peripheral ameloblastoma of the gengiva. A case report. J Periodontol. 1990; 61: 649-52.

10. Philipsen HP, Reichart PA, Nikai H, Takata T,Kudo Y. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. Oral Oncol. 2001; 37: 17-27.

11. Nauta JM, Panders AK, Schoots CJF, Verney A, Roodenburg JLN. Peripheral ameloblastoma. A case report and review of the literature. Int J Oral Maxillofac Surg. 1992; 21: 40-4.

12. Gardner DG. Some current concepts on the pathology of ameloblastomas. Oral Surg Oral Med Oral Pathol. 1996; 82: 660-9.

13. Wettan HI, Patella PA, Freedman PD. Peripheral ameloblastoma: review of the literature and report of recurrence as severe dysplasia. J Oral Maxillofac Surg. 2001; 59: 811-5.

14. Reichart PA, Philipsen HP. Odontogenic Tumors and Allied Lesions. London: Quintessence Publishing Inc.; 2004. p. 77-86.

15. Perdigao PF, Gomez RS, Pimenta FJ, De Marco L. Ameloblastin gene (AMBN) mutations associated with epithelial odontogenic tumors. Oral Oncol. 2004; 40: 841-6.

16. S. C. Lin, C. M. Lieu, L. J. Hahn, and H.W. Kwan, "Peripheral ameloblastoma with metastasis," International Journal of Oral and Maxillofacial Surgery, vol. 16, no. 2, pp. 202–204, 1987.

17. A. Buchner and J. J. Sciubba, "Peripheral epithelial odontogenic tumors: a review," Oral Surgery, Oral Medicine, Oral Pathology, vol. 63, no. 6, pp. 688–697, 1987.

