Extraosseous Ameloblastoma of Maxillary Gingiva- A Case Report

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Abstract Ameloblastoma is a benign neoplasm of the jaw bones that originate from the odontogenic epithelium. They are more common on the mandible than the maxilla. Rarely such tumours arise outside these bones, when they are termed extraosseous or peripheral ameloblastoma. We report a case of extraosseous ameloblastoma in a 30 year old woman, who presented with a painless lesion on the upper gingiva. The lesion was excised completely and the histopathology was suggestive of extraosseous ameloblastoma.

Keywords Extraosseous · Ameloblastoma · Upper gingiva · Excision

Introduction

Ameloblastoma are neoplasms of odontogenic epithelial origin, which arise within the jaw bones. They in general are benign neoplasms, malignant ameloblastoma being extremely rare [1]. The peripheral or extraosseous ameloblastoma is rare, occurring on the gingiva, buccal mucosa or the floor of mouth [2, 3]. These lesions appear to arise from either remnants of the dental lamina within the gingiva or from the surface epithelium [4, 5].

Extraosseous ameloblastoma is less invasive than its intraosseous counterpart. Surgical excision with adequate disease free margin is the treatment of choice. Unlike the intraosseous ameloblastoma, recurrence after surgical

excision is not common. [6] We report a case of an extraosseous ameloblastoma of the upper gingiva in a 30 year old woman.

Case Report

A 30 year old woman presented with a painless swelling on the upper gingiva, behind the third molar tooth of right side. She noticed the swelling incidentally before three months of presentation, when the swelling was very small in size, and ignored it. Later the swelling started increasing in size, but was never painful. She did not have any history of trauma, tooth ache or undergoing any dental treatment. On examination, her dental hygiene was good. There was a firm swelling with granular surface measuring about 3 cm X 2 cm over the upper gingiva, posterior to the right upper third molar tooth. (Fig. 1) The lesion was extending on to the hard palate, but it was attached with a narrow base to the gingiva. There was no bleeding on palpation. Clinically we diagnosed it as a pyogenic granuloma or epulis. The mass was excised with a half centimeter margin with diathermy under general anesthesia. (Fig. 2) Postoperative period was uneventful. Patient was allowed oral food same day. She was discharged from the hospital next day. Histopathological examination of the mass showed islands of proliferating odontogenic epithelial cells embedded in a fibrous stroma. The islands were containing, centrally a loose meshwork of cells resembling stellate reticulum and peripheral palisading of basal cells. Features were suggestive of ameloblastoma. (Fig. 3) We did not do any further surgical procedure, as we had excised the mass completely with a small margin of normal tissue. The patient was followed for one year and she had no signs of recurrence.

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Fig 1 Picture showing the lesion on the maxillary gingiva posterior to molar teeth

Discussion

Ameloblastoma is an epithelial odontogenic tumour of the jaw bones. Falkson in 1879, described this lesion in detail, but the term ‘ameloblastoma’ was coined by Churchill in 1933 [7]. It represents approximately one per cent of all oral tumours, with nearly 80 % of ameloblastomas occurring in the mandible. About 20 % of them occur in the maxilla. They develop from the odontogenic epithelium and its derivatives or remnants [7]. Extraosseous or peripheral ameloblastoma accounts for 2–10 % of all ameloblastomas. The first report of peripheral ameloblastoma was by Kuri in 1911. In 1959, Stanley and Krogh defined the clinical and histopathologic characteristics [5]. They grow in the soft tissue overlying the tooth bearing areas of the jaws. The commonest site of peripheral ameloblastoma is mandibular gingiva; mandibular premolar region accounting for 32.6 % of all sites [8]. But occasionally it can be found even in the non tooth bearing areas of gingiva, buccal mucosa, base of the tongue or floor of mouth [5]. However Philipsen et al. are of opinion that the extralingival ameloblastoma are of salivary gland origin [8]. In the case we presented here, the lesion was on the non tooth bearing area of maxillary gingiva, posterior to the upper third molar.

Fig 2 Post of photograph

Fig 3 H& E staining showing islands of odontogenic epithelial cells, with central stellate reticulum and peripheral palisading

The etiology of extraosseous ameloblastoma is unclear. The tumor can be derived from the extraosseous epithelial remnants of the dental lamina or from the basal cell layer of the oral mucosa, which is believed to have odontogenic potential [5]. The clinical presentation of extraosseous ameloblastoma is in the form of a painless slow growing mass. In most of the reported cases, the mass had a smooth surface [8]. However, in our case, the surface is rather granular. According to Pekiner et al., during mastication the mass gets traumatized so that the original smooth surface may look ulcerated or keratotic [10]. There will not be involvement of the underlying bones radiologically, but a superficial bone erosion, known as cupping or saucerization may be detected during surgery [8].

Extraosseous ameloblastoma is rarely a clinical diagnosis, the diagnosis is made only after histopathological examination. They shows histologic patterns found in the intraosseous ameloblastoma but has a tendency to be acanthomatous [4]. The peripheral ameloblastoma shows histology of solid or multicystic type of ameloblastoma, as per World Health Organization classification [1]. The differential diagnosis of extraosseous ameloblastoma include pyogenic granuloma, peripheral giant cell granuloma, peripheral odontogenic fibroma, peripheral ossifying fibroma, papilloma, and epulis [5].

Surgical excision with a small margin of normal tissue is the recommended treatment of extraosseous ameloblastoma. The extent of safety margin is not clearly defined. However most of the authors had done an excision of 5 mm when the diagnosis is known, and the underlying periosteum was included in the resected mass [9]. But in our case, as ameloblastoma was not the clinical diagnosis, the surgical free margin was probably less than 5 mm. But there is no recurrence at the site of excision in one year. This shows that the extraosseous ameloblastoma are less aggressive than the classical ameloblastoma. Gardener DG in his article proposed that the dense fibrous tissue of the gingiva and the peristeam of alveolar process is an effective barrier for the spread of tumour [10]. However this theory does not hold good in lesion of buccal mucosa, tongue base or floor of mouth where there is no barrier for the spread of tumour. The extension or resection in these sites needs further study, as the incidence of tumour at these sites is very rare.

Malignant ameloblastoma are extremely rare [7]. Edmondson et al. was the first to report a case of extraosseous ameloblastoma with malignant transformation [11]. Song-Chyr et al. documented a malignant peripheral ameloblastoma that metastasized to the supra-clavicular lymph nodes [12]. Ide et al. reported a case of malignant ameloblastoma with metastasis, and they opined that a large size of the tumour is a predictor of malignant transformation [13]. Our case even though was measuring 3 cm × 2 cm, the histopathology was typically benign, and the patient is disease free one year after excision. Treated cases of Peripheral ameloblastoma need to be followed for longer period for early detection of recurrence or malignant transformation [14].

Conclusion

Ameloblastoma, which is a tumour common in the mandible, can also be seen in a non-tooth bearing region of the gingiva. In the literature there is evidence of extraosseous ameloblastoma occurring in the buccal mucosa, lips, palate, base of tongue, floor of the mouth. Most of the times, the diagnosis is not done clinically, but after histopathological examination. Hence extraosseous ameloblastoma need to be considered in the differential diagnosis of gingival lesions resembling epulis or pyogenic granuloma, and histopathological examination of resected tissue to be performed in such cases.

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