Peripheral Ameloblastoma of the Buccal Mucosa: Case Report of a Rare Tumor

Abstract

Peripheral ameloblastoma (PA) refers to a rare neoplasm that accounts for approximately 1%–5% of all ameloblastomas. It arises in the soft tissues overlying a tooth-bearing region, and those found in nontooth-bearing locations, such as the buccal mucosa, lips, and palate. This article aims at describing a case of a rare form of extragingival PA in the nontooth-bearing region and challenges in its management in settings with limited resources. A 55-year-old African male presented with a painless swelling on the right cheek for about 6 years, which was clinically diagnosed as pleomorphic adenoma; however, both pre- and postoperative histopathological results reported the swelling to be follicular ameloblastoma. Extragingival PA is a rare variant of ameloblastoma, which needs to be included as one of the differential diagnoses of the buccal mucosal swellings.

Keywords: *Buccal mucosa, extragingival, peripheral ameloblastoma*

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Introduction

Ameloblastomas overwhelmingly occur centrally within the jaws but have the potential to occur in extraosseous locations,[1] and they are termed as peripheral ameloblastoma (PA). PA, which is a rare odontogenic soft-tissue tumor that was first described in the literature by Kuru in 1911, is reported to account for approximately 1%–5% of all ameloblastomas.[2,3] Although its clinical appearance varies, the PA generally presents clinically as a slow-growing, firm, painless mass with a sessile or pedunculated base, varying in size from 0.2 to 4.5 cm in diameter, and usually has no radiographic evidence of bone involvement.[1,2]

Several cases of PA have been reported after Kuru's description, most of which seem to have originated from gingival mucosa or attached oral mucosa adjacent to the mandible or maxilla.[4] Some few reports have documented PA cases occurring in an extragingival region, which are unrelated to tooth germs such as in buccal mucosa and floor of the mouth.[5] A recent case report[3] stated that only six cases of extragingival PA in buccal mucosa had been reported till the year 2015.

We report a case of an extragingival PA, which occurred in the buccal mucosa, basing the discussion on the challenges in its diagnosis in settings with limited diagnostic resources. Moreover, the case report also aims to add another rare case of PA into the existing data pool.

Case Report

A 55-year-old African male reported to the oral and maxillofacial unit clinics of our institute in December 2017 with a complaint of painless swelling in the right cheek for 6 years. The patient reported that he had noticed a small swelling in the region about 6 years before presenting to us. He did not seek medical attention at that time, since it was not bothering him, until about a year ago when he started seeking for medical attention after experiencing several episodes of self-biting on the cheek. The biting resulted into ulceration of the cheek with association of pain and at times pus discharge. Before being referred to our institute, he attended several dental clinics where he was given antibiotics.

On local clinical evaluation, he presented with an obvious facial asymmetry due to a swelling on the right side of the face. Extraorally, the swelling extended from the level of zygomatic arch (superiorly) to the level of angle of the mouth (inferiorly),

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Access this article online

Website: www.ccij-online.org

DOI: 10.4103/ccij.ccij_93_18

Quick Response Code:

How to cite this article: Sohal KS, Owibingire SS, Moshy JR, Kileo BF. Peripheral ameloblastoma of the buccal mucosa: Case report of a rare tumor. Clin Cancer Investig J 2018;7:227-30.

it was oval shaped, with a normal overlying skin. The swelling was firm to palpation, mobile, without raise in local temperature; however, it was mildly tender. Intraorally, the swelling was covered with a slightly hyperemic and ulcerated mucosa, and it measured about 5 cm × 4 cm and was located anteromedial to the mandibular ramus.

Computed tomography scan of the head-and-neck region (with and without contrast) revealed a well-demarcated mass on the right buccal region, causing some degree of bone resorption on the right maxillary tuberosity due to pressure effect. The tumor had mixed radiopaque and radiolucent nature, without infiltration to the surrounding structures [Figure 1]. A clinical provisional diagnosis of pleomorphic adenoma was made with differentials of fibrolipoma and carcinoma ex-pleomorphic adenoma.

An incisional biopsy was taken transorally under local anesthesia and submitted for histopathological analysis. Microscopically, the tissue section showed a neoplastic lesion forming follicles lined by epithelium with palisading cuboidal-to-tall columnar cells and stellate cells [Figure 2], and thus, a diagnosis of follicular ameloblastoma was made. The patient was discussed in the panel of specialists. A request for slide review was made, and the results remained unchanged.

After the patient had consented for surgery, the tumor was widely excised under general anesthesia through an intraoral approach, and resulting defect was repaired. The tumor was then submitted for histological analysis. Microscopically, the section of postoperative tissues showed a capsulated tumor, with cystic dilatation beneath the capsule. The tumor had a follicular pattern with varying size of interconnected islands of odontogenic epithelium which were lined with stratified epithelium composed of palisading columnar cells [Figure 3]. Diagnosis of follicular ameloblastoma was made. The postoperative period was uneventful, and the patient was discharged home, with first follow-up made 1 month postoperatively, followed by follow-up every after 3 months.

Discussion

The credit of reporting the first case of PA is given to Kuru by some authors.[2,3] However, what Kuru described as peripheral amelobastoma was infact an intraosseous ameloblastoma that had penetrated through the alveolar bone and fused with the oral epithelium, thereby presenting itself clinically as a "peripheral lesion."[6] The first completely documented case of a PA must be attributed to Stanley and Krogh.[6,7]

PA refers to neoplasm arising in the soft tissues overlying a tooth-bearing region, together with those found in nontooth-bearing locations, such as the buccal mucosa, lips, palate, and other parts of oral mucosa.[8] PA is a relatively uncommon pathology accounting for approximately

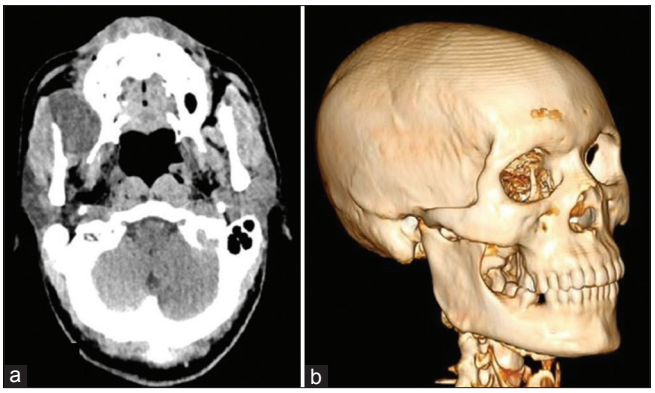

Figure 1: An axial view a welldemarcated, hypodense cystic lesion occupying the buccal space causing thinning of the part of the ascending ramus of the mandible and posterior part of the maxillary buccal cortex (a). The threedimensional reconstruction image shows the remodeled posterior segment of the right maxillary bone without any signs of bone destruction (b)

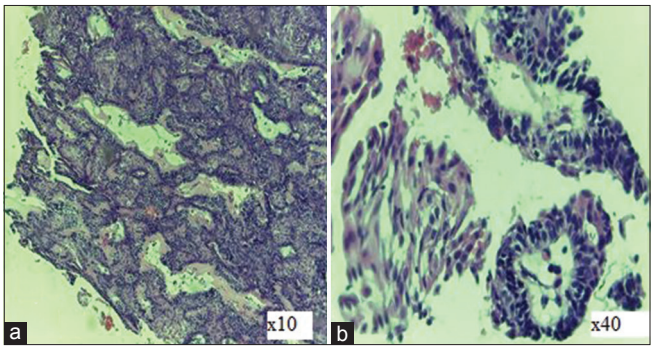

Figure 2: The preoperative incisional biopsy histopathological images (H and E) showing tumor islands in follicular pattern, interconnected to each other. The tumor is lined with epithelium which is composed of palisading, stratified cuboidal cells. Stellate reticulum is found at the center of the tumor. (a) (magnification, × 10) and (b) (magnification, × 40)

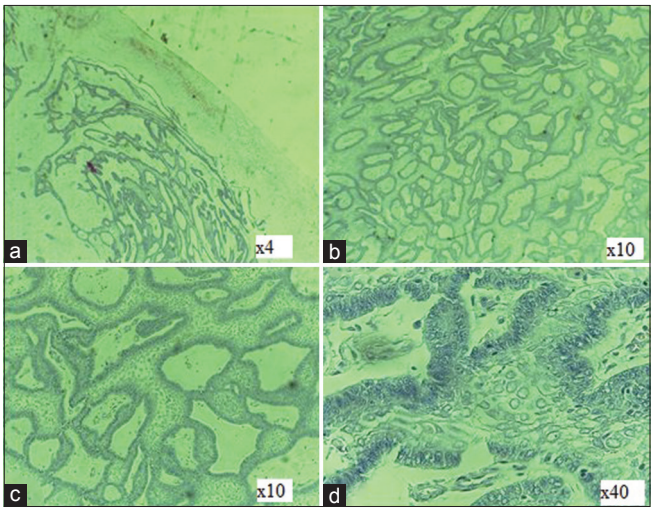

Figure 3: Postoperative histopathological images (H and E): A fibrous capsule surrounding a well-demarcated tumor (a). There is cystic dilated tumor with interconnected islands lined with epithelium (b). The epithelium has cuboidal cells which are palisading and are stratified (c). The epithelium has cuboidal cells that are being placed at the base with a stellate reticulum at the center of tumor that appears to have a follicular pattern (d)

1%–5% of all ameloblastomas.[2,3] PAs of the extragingival areas are even more extremely rare.[9] Literature search of

occurrence of extragingival PA in buccal mucosa from the 1980s to date showed <10 reported cases to the best of our knowledge.

The age range for PA generally has been documented to be between 9 and 92 years with a mean age of 52.1 years and a male-to-female ratio of 1.9:1.[7] For the case of extragingival PA, the mean age of occurrence has been reported to be similar to that of gingival PA. With regard to gender of the patients, extragingival buccal mucosa PAs have been reported in five men and two women to date.[3,8-10] In the current case, the patient's age falls within the given mean age.

There are two main theories regarding origin of PAs: the first proclaims PAs to originate from the extraosseous epithelial remnants of the dental lamina and its organ derivatives within the underlying connective tissue and the second states the origin from the basal cell layer of the oral mucosa, which is believed to have odontogenic potential.[2] It has been reported that the potential sources of extragingival PA include odontogenic remnants of vestibular lamina, pluripotent cells in the basal cell layer of the mucosal epithelium, and pluripotent cells of minor salivary glands.[9] Based on the information available from the literature, it is reasonable to consider that buccal PA may be linked to the developmentally included enamel organ or its remnants as parts of the buccal mucosa during the embryogenesis of the vestibular lamina.[8]

Similar to the majority of case reports of buccal mucosa PA,[6] extragingival PA was not the initial diagnosis in the current case. Clinical diagnosis is frequently reached by a combination of detailed clinical history and physical examination aided by radiological findings. Physical examination of a pathological lesion gives details on nature of overlying skin/mucosa, location, size, and consistency. Basing on tumor location, diagnosis is made in consideration of the knowledge of cells and tissues found in that region, aided by reports from different literature on common condition in a given part of the body. Taking that into account, ameloblastoma is rarely encountered in buccal mucosa, thus never the initial clinical diagnosis. Therefore, histological diagnosis must be relied upon. In the case reported, the panel of specialists in oral and maxillofacial surgery and oral medicine queried the histological diagnosis, as the location was not favoring the diagnosis nor was the epidemiology, and thus, a review of slide was requested from a panel of different pathologists, who reported similar results, and later to be further confirmed by postoperative analysis of the whole tumor.

Histological examination of PA is often difficult, as PA and basal cell carcinoma (BCC) exhibit similar growth patterns and share some histological features. There are some authors who believe that PA and BCC represent the same neoplasm[3] while others report them as different entities. It has been reported that because of histopathological features, it is possible to mistake PA for BCC.[6] Some authors believed that the PA exhibits central polarization of the nuclei of the peripheral cells, while the true intraoral BCC shows a sharp demarcation between the peripheral cells and the central reticular cells.[8] In one case study, the authors examined the genetic features of the tumor that was diagnosed as extragingival PA using microarray analysis to obtain a conclusive diagnosis.[3] They concluded that microarray analysis allowed a more efficient method of analysis, enabling a better diagnosis. The use of microarray analysis in settings with limited resources is very challenging in terms of cost and technical know-how. Moreover, in developing countries, there is a huge burden of other diseases that require to be taken care off, and thus, little emphasis is given to rare diseases.

Challenges in clinical diagnosis and lack of diagnostic tools such as frozen section led to the team of surgeon to be aggressive in treatment of the lesion and handled the lesion as a malignant condition, and hence, this increased morbidity to the patient. Had there been better diagnostic tools then the treatment could have been more conservative than what was offered. Although the patient is doing well, a long-term follow up is mandatory.

Conclusion

Extragingival PA is a very rare variant of ameloblastoma, whose clinical knowledge is of interest to the dentists, pathologists, and maxillofacial surgeons. Extragingival ameloblastoma should be one of the differential diagnoses of the buccal swellings. The histopathological results are not the only best approach for a proper diagnosis, but also, microarray analysis can be helpful and thus should be considered.

Acknowledgment

The authors would like to sincerely thank the patient for accepting his case to be presented, and also the residents and specialists in the department of Pathology for reviewing the pre and post-operative slides of tissue sample and providing us with the histopathological images.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. The patient understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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