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Pleomorphic Adenoma of the upper lip: A diagnostic quandary due to coinciding traumatic history – A rare case report

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Abstract

Though pleomorphic adenoma (PA) is the most common tumor of the salivary glands, its occurrence on the upper lip is infrequent and has been rarely documented in literature. In this article, we present a case of a 43-year-old female, with a history of trauma who reported with a swelling in the upper labial mucosa since two months which was subsequently diagnosed as pleomorphic adenoma.

Keywords: Common, subsequently, diagnosed

Introduction

Pleomorphic adenoma (PA) or mixed tumor is a benign salivary gland tumor most commonly presenting itself in the parotid gland ^[1]. In the intra-oral sites, the palate is considered the most common site (42.8 – 68.8%) followed by the upper lip (10.1%) and cheek (5.5%). Other rare sites include throat (2.5%), retromolar region (0.7%), floor of the mouth and alveolar mucosa ^[2]. Histologically, it is characterized by a large variety of tissues consisting of epithelial cells arranged in a cord like pattern, together with areas of squamous differentiation or with plasmacytoid appearance. Myoepithelial cells are responsible for the production of abundant extra-cellular matrix with chondroid, collagenous, mucoïd and osseous stroma ^[3]. The present case study unfolds the diagnostic journey commencing from a painless mass with a short duration of history of trauma which was traced back to a rare case of PA of the upper lip on Histopathological Examination (HPE).

Case presentation

A 43-year-old female reported to the Unit of Oral and Maxillofacial Surgery complaining of a painless mass in the upper anterior labial mucosa past two months. The patient gave a positive history of trauma two months back. Medical history was non-contributory. No specific findings were noted on extra-oral examination, while intra-oral examination revealed a well-circumscribed mass measuring (1 x 1.5 cm) approximately. The mass was firm on palpation with normal overlying mucosa. We performed an excisional biopsy of the mass via intra-oral vestibular incision with safe margins. With traumatic fibroma as clinical diagnosis the specimen was sent for histopathological examination to help in determining a conclusive final diagnosis. (Figure 1A and Figure 1B)

Histopathology: Microscopic examination with hematoxylin and eosin-stained section revealed a well-circumscribed encapsulated tumor having multiple large solid nests, trabeculae, cords of spindle cells and epithelioid cells having uniform nuclei and eosinophilic cytoplasm in myxoid stroma with a definitive diagnosis of “pleomorphic minor salivary gland adenoma showing predominantly myoepithelial element”. Microphotograph of pleomorphic adenoma in magnifications of 20x and 40x showing seeds and cords of benign epithelial cells, myoepithelial cells and occasional cystically dilated spaces with focal hyalinization were seen. (Figure 2A and Figure 2B)

Discussions

PA is rarely found in the minor salivary glands, but a few cases have been found that most commonly involve the palate followed by upper lip and buccal mucosa [4]. Literature suggests that the average age of presentation is between 40 – 60 years with a slight female predilection which was consistent with our case study [5]. They usually present as painless and well-defined masses which are usually mobile. The present case shows PA of the upper labial mucosa, clinically misdiagnosed as traumatic fibroma concealed by patient's traumatic history.

Apart from its association with the PLGA gene, very little is known regarding the aetiology of PA. Its aetiology has been linked to other variables including radiation exposure and oncogenic viruses [6]. Although there is a dearth of research to support the idea that trauma causes PA, a meta-analysis conducted by Schieber et al. found that chronic trauma is associated with carcinogenesis, DNA modification, and the production of oxidative stress, all of which are related to tumour growth [7]. But surprisingly our case was associated with a short duration of history of trauma which led to the formation of PA. The possible pathogenesis could be mechanical stress-induced increase in the release of inflammatory cytokines interleukin-6 and interleukin-8 thus inducing the production of Fibroblast Growth Factor (FGF-2) and Epidermal Growth Factor (EGF) as means of cellular defence mechanism [6].

The ratio of occurrence of PA in upper lip to lower lip is 6:1 [8]. These tumors usually go unnoticed as they are painless and slow growing rarely showing ulcerations, pain or bleeding. The most common symptom is a swelling arising inside the mouth which is a well-defined, round mass usually encapsulated and firm in consistency. A mobile lesion that is not fixed to the underlying skin is usually indicative of a benign nature, but a biopsy is always recommended for definitive diagnosis [9]. HPE has always proven to be the gold standard for the confirmatory diagnosis of befuddling lesions of the upper lip. Pleomorphic adenoma can be classified histologically as per classification given by Foote and Frazell in 1954 [10]. Usually PA of the minor salivary glands are type 3 and 4. Our case belonged to the type 3 variant.

Though recurrence rates are lesser as compared to PA in major salivary glands, the ideal treatment is a wide surgical excision [11]. An inadequate resection or rupture of tumor capsule leading to tumor spillage during excision can lead to local recurrence [12]. PA of the lip is a rare neoplasm, the diagnosis of which requires a high level of suspicion. Long term follow-up is a must as there can be recurrence as well as malignant transformation even after many years [13]. The present case did not show any recurrence at one year follow up.



Fig 1A: Shows tumor marking and vestibular incision to expose surface of tumor.



Fig 1B: Shows encapsulated mass excised with safe margins.

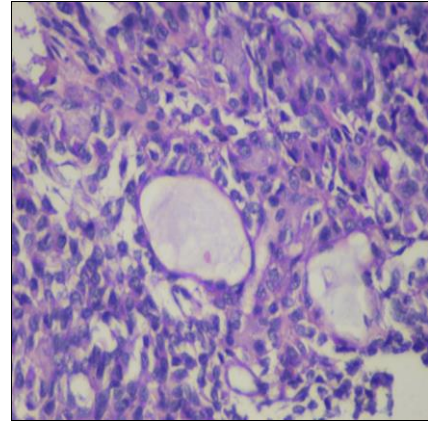


Fig 2A: H&E-stained section in 20x magnification.

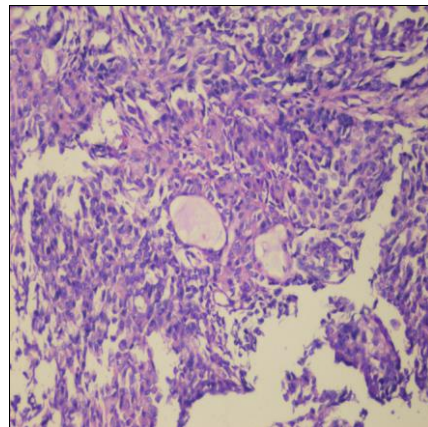


Fig 2B: H&E-stained section in 40x magnification.

Conclusion

This case report emphasizes the role of histopathological evaluation and regular follow-up as the gold standard regime even for simple intra-oral swellings and is an addition to the existing literature of this rare entity. All lesions should be sent for HPE and should not be solely relied on clinical diagnosis as it may lead to dire consequences if misdiagnosed because one never knows the molehill might actually be the mountain.

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