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Pleomorphic Adenoma of Upper Lip mimicking A Nasolabial Cyst: A Rare Case Report

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1. Abstract

Pleomorphic adenoma of the upper lip is a rare tumor. Among the minor salivary gland tumors, it is most often seen in the palate, followed by the lip and buccal mucosa. We are reporting a case report of a pleomorphic adenoma of the upper lip in an adult male patient that, clinically, was mimicking as a naso-labial cyst, and was successfully excised.

2. Introduction

'The term "pleomorphic adenoma" (PA) was first suggested by Willis and it signifies a unique histological pattern of mixed glandular epithelium, myoepithelial cells, and connective tissue elements. PA is the most common benign minor salivary neoplasm, accounting for 40–70% of all benign tumors [1, 2]. The parotid gland is the most common extra-oral site for the PA; intra-orally, it is most commonly seen in the palate, followed by the upper lip and buccal mucosa [3].The exact etiology of PA is not known, but cytogenetics and molecular studies point towards a chromosomal aberration at 8 q12 and 12 q15 as the causative mechanism for the development of PA [4]. Clinically, it presents as a slow-growing, painless swelling that usually tends to be single and mobile. It can rarely cause mucosal ulceration and paresthesia secondary to neural involvement [5]. It is slightly more common in females, with a male-to-female ratio of 1:1.5, and the mean age of presentation is 33 years [6].

The nasolabial cyst (NC) is also a rare, non-odontogenic lesion that usually presents as swelling in the upper lip and nasal vestibule [7]. Due to its common location, it can be clinically mistaken for a sebaceous cyst or salivary gland tumor[8]. We present a case report of PA of the upper lip in an adult male patient, mimicking a NC, its diagnosis, and surgical management.

3. Case Report

A 23-year-old male presented to the outpatient department with a nine months history of swelling on the right side of the upper lip. The swelling was painless, slowly progressive, and he denied any history of trauma or infection at the site. On clinical examination, there was a firm, nodular, non-tender swelling on the right side of the upper lip measuring around 2×2.3 cm. It was not crossing the midline, pushing the vermilion border of the upper lip outward, and superiorly extending towards the lower part of the right nasolabial fold (Figure 1). The overlying skin appeared to be adherent to swelling at some places, and the underlying mucosa was normal. There was no cervical lymphadenopathy, and the clinical possibility of a NC was kept. However, other differential diagnoses, i.e. pleomorphic adenoma, fibroma and neurofibroma were also kept. A contrast enhanced computed tomography demonstrated a 2.06×1.44 cm well-defined soft tissue lesion in upper part of the lip with no internal communication, and with maintained soft tissue plane (Figure 2). The fine needle aspiration cytology (FNAC) of the swelling was done, a small clear fluid and some cheesy material was aspirated, however on cytological evaluation only few epithelial cells were seen. In view of the inconclusive FNAC report, a plan for the excisional biopsy was made. A detailed, informed consent was obtained from the patient. Surgery was planned under local anesthesia. Initially, parts were cleaned and draped, and 2% lignocaine with 1:100,000 adrenaline hydrochloride was injected around the lesion. A mucosal incision was given to avoid visible skin scar, and the mass was separated from the surrounding structure by sharp dissection, however, a part of skin was adherent to the swelling, so it was removed along with the mass. The resulting skin defect was of size 0.5×0.5 cm, which was primarily closed. On gross examination, the tumor was well encapsulated except at

skin adherent level, and it was sent for histopathology (Figure 3). Histopathology revealed a well-encapsulated soft tissue mass with epithelial and myoepithelial cell proliferation lining the ducts and chondromyxoid stroma, consistent with features of the PA, and the excised skin was not involved. There was no sign of recurrence during the patient's year-long follow-up at three monthly visits (Figure 4).

A.

B.

Figure 1: Preoperativeclinical photo (A)frontal and (B) Lateral view showing a swelling of right upper lip extending superiorly towards nasolabial fold (black arrow).

Figure 2: Preoperative CT scan axial cut showing 2.06 x 1.44 cm enhancing lession (Black Arrow) upper lip.

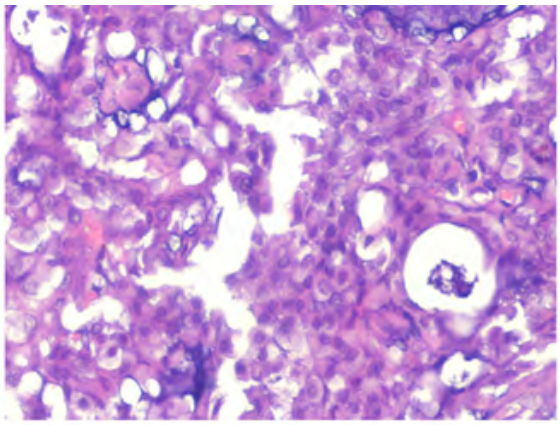

Figure 3: Microphotograph showing glandular formation and plasmatoid and cubiodal cells (400X H& E stain)

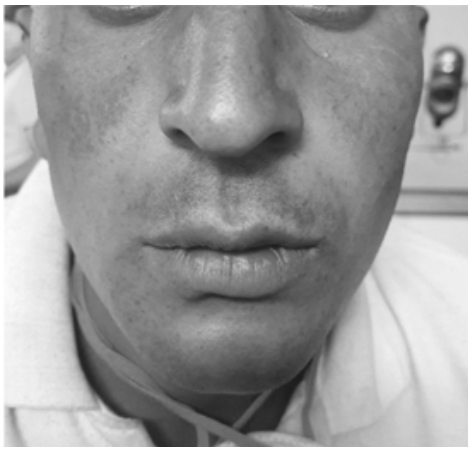

Figure 4: Postoperative frontal view of patient after 3 months with no swelling or scar

4. Discussion

PA constitutes 6.4% of all minor salivary gland tumors [9]. It usually presents as an asymptomatic nodule (82%), rarely it can cause pain(18%) and paresthesia [10]. It is most commonly found in the hard and soft palate, followed by lip and buccal mucosa [11]. In the lip area, it is more common in the upper lip, with a ratio of 6:1[9]. This may be explained on an embryological basis, as the upper lip develops from the fusion of three processes, i.e., maxillary, medial, and lateral nasal, while the lower lip forms from the fusion of bilateral mandibular processes, hence chances of entrapment of embryonic cells is high in upper lip [10]. Whereas the average age of presentation is 33, and it is more common in women, our case was a man in his second decade of life. Hence, PA should be kept in mind as a differential diagnosis even in younger guys with painless upper lip swelling. Though FNAC is diagnostic in cases of PA of the upper lip [11], however, in our case it was non-diagnostic. This could be due to the fact that there are large cystic spaces within the tumor, and it may contain large mucinous material, resulting in non-diagnostic yield on FNAC as in this case.

The NC is thought to be the remnant of an epithelial cyst of the lower part of nasolacrimal duct, it usually presents in adult life as painless swelling in the upper lip and vestibule area. On palpation,

it seems to be cystic, and it may become stiff and painful due to infection or a rapid bleeding. Hence, PA lip can clinically resemble an NC due to a comparable location and presentation. However, Marcoviceanu has described three cardinal symptoms of NC as, partial or complete nasal obstruction, well circumscribed swelling, and localized pain [8]. These symptoms were not present in our patient. So, these three features should be looked for while making a clinical diagnosis of NC.

The treatment for both is surgical excision, which can be performed under local anesthesia with excellent results. The overlying skin and mucosa are normal in most of the cases of PA of the upper lip; however, both can undergo ulceration, possibly because of pressure effect, and not always point to a malignant pathology. Incomplete removal, rupture of capsule and spillage of tumor during surgery can lead to local recurrence [12].

In a recent review article in 2022 by Umemori et al, only 33 cases of PA of the upper lip have been described in the literature since 2000 [13]. Interestingly, none of them mimicked a NC clinically, as the tumor has a more distinct growth toward the mucosal surface. However, in our case, the bulk of the tumor was toward the skin and hence mimicked a NC.

5. Declaration of Patient Consent

The author certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal .The patients understand that their names and initials will not be published and due efforts will not be made to conceal their identity, but anonymity cannot be guaranteed .

6. Conflict of Interest

There are no conflict of interest.

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