

Pleomorphic adenoma of the upper lip presenting as multiple swellings: aesthetic and surgical perspectives

Caso Clínico

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Resumo

Pleomorphic adenoma is the most common benign salivary gland neoplasm, yet its occurrence in the upper lip, especially as multiple swellings, is extremely rare. We present a 46-year-old female with progressive, painless multiple upper lip swellings over one year. Clinical examination, fine-needle aspiration cytology, and contrast-enhanced computed tomography imaging supported the diagnosis. Surgical excision via a mucosal approach was performed, preserving anatomical and aesthetic structures. Histopathological analysis confirmed pleomorphic adenoma. The patient had an uneventful recovery with no recurrence at six months. This case highlights the importance of recognizing atypical Pleomorphic Adenoma presentations and ensuring complete, cosmetically sensitive excision.

Keywords: Pleomorphic adenoma, upper lip, minor salivary gland tumor, aesthetics, cosmetic surgery

Introduction

Pleomorphic adenoma (PA) is the most prevalent benign tumour of the salivary glands, accounting for 60–70% of all salivary gland neoplasms^{1,10}. While the majority arise from the major salivary glands, particularly the parotid, a significant proportion—up to 20%—originate from minor salivary glands, most frequently located in the hard palate, followed by the buccal mucosa, lips, and retromolar region^{2,5,7}. The upper lip is an uncommon site for Pleomorphic Adenoma, and fewer than 15 well-documented cases exist in the literature^{3,4}. The clinical presentation of PA is typically a painless, slow-growing, mobile mass. Intraoral lesions may go unnoticed for long durations due to their asymptomatic nature. The diagnosis is often delayed and may be confused with other benign entities like

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mucocoeles, lipomas, fibromas, or sebaceous cysts^{4,8,11}. Imaging e.g., Computed Tomography [CT] or Magnetic resonance imaging [MRI] can be helpful in assessing deep extensions or bony changes. Fine needle aspiration cytology [FNAC] is useful for preoperative evaluation, but histopathology remains the gold standard for confirmation^{2,4,5}. The following case presents a rare instance of multiple pleomorphic adenomas involving both mucosal surfaces of the upper lip, an even rarer manifestation. This report emphasizes clinical, radiological, and histopathological correlation, and discusses surgical considerations in the context of functionality and aesthetics. A 46-year-old female presented to our outpatient department with a progressive swelling of the upper lip, which had been present for the past one year. The patient reported that the swelling initially appeared as a single, small, painless nodule but gradually increased in size and developed into multiple swellings. There was no history of pain, trauma, bleeding, fever, systemic illness, or other ENT-related complaints. The patient had no significant past medical or surgical history, and her family and psychosocial history were unremarkable.

Description of the case

Clinical Findings

On physical examination, three distinct swellings were noted on the upper lip:

- The first swelling measured 1 × 1 cm, located 2 cm inferior to the ala of the nose, medially at the midline, and 1.5 cm medial to the angle of the mouth.
- The second swelling, measuring 3 × 2 cm, involved the mucosal surface of the upper lip, extending superiorly 0.5 cm from the vermilion border, inferiorly 3 cm toward the gingiva, 0.5 cm from the midline medially, and 2 cm lateral to the lateral incisors.
- A third, multilobulated swelling was noted inferior to the second swelling, with associated fullness in the left upper lip, extending medially to the philtrum and laterally to the nasolabial groove.

All swellings were:

- Firm to hard in consistency
 - Mobile, non-tender, non-pulsatile
 - Covered with smooth mucosa
 - The overlying skin was pinchable, with no signs of inflammation or ulceration
- No regional lymphadenopathy was appreciated.

Diagnostic Assessment:

A FNAC was performed and was suggestive of a benign salivary gland neoplasm, with a differential of pleomorphic adenoma or myoepithelioma. A contrast-enhanced CT scan of the face revealed a well-defined soft tissue lesion in the left upper lip, measuring 2.8 × 1.6 × 1.9 cm, with evidence of scalloping and remodeling of the adjacent maxillary bone, but without signs of invasion into surrounding structures.

Therapeutic Intervention:

The patient underwent surgical excision under general anaesthesia. An L-shaped incision was made over the midline mucosal surface of the upper lip, extending along the gingivolabial sulcus. The soft tissues were carefully dissected, maintaining the integrity of the tumour capsule. The mass was noted to extend deep toward the maxilla, with an indentation but no invasion. The tumour was completely excised in toto, and adjacent anatomical structures were preserved. No intraoperative or postoperative complications were noted.

Follow-up and Outcomes

The patient recovered uneventfully. At 6-month follow-up, there was no clinical or radiological [CT] evidence of recurrence, and the cosmetic and functional outcomes were satisfactory. Histopathological examination confirmed the diagnosis of pleomorphic adenoma, revealing epithelial and myoepithelial components in a chondromyxoid stroma, with no signs of malignant transformation.

Figure 1
Upper lip swelling



Figure 2
Swelling in the mucosal surface of the upper lip



Figure 3a
Intraoperative picture of the tumour



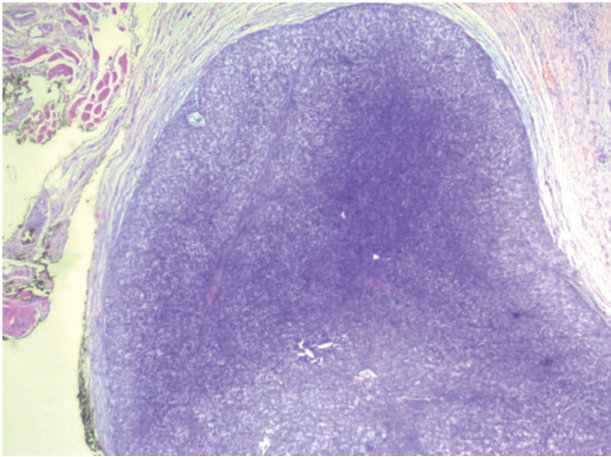
Figure 3b
Intraoperative picture of the tumour



Figure 4
Intraoperative picture showing closure in layers



Figure 5
histopathology consistent with findings of
pleomorphic adenoma



Discussion

Minor salivary gland tumours comprise a heterogeneous group of neoplasms, accounting for about 10–25% of all salivary gland tumours^{2,5,10}. Among these, Pleomorphic adenoma is the most common benign entity, particularly affecting the palate, cheek, and upper lip¹³. A large-scale review by Eveson and Cawson¹ of over 2,400 salivary tumors found that only 16% involved minor salivary glands, and among those, upper lip involvement was rare. Similarly, Waldron et al.⁵ and Toida et al.⁷ found that Pleomorphic Adenoma was the predominant histologic subtype in minor salivary gland tumors but noted the limited number of upper lip cases.

Histologically, Pleomorphic Adenoma exhibits a mixture of epithelial, myoepithelial, and stromal elements, often making the diagnosis straightforward on microscopic evaluation^{4,8,11}. FNAC is valuable in preoperative planning, though sampling error can occur, particularly in lesions with a prominent myxoid stroma^{2,4}. Wang et al.², in a study of 195 minor gland PAs, found that complete excision with negative margins significantly reduces recurrence. Spiro¹⁰ similarly highlighted the importance of early surgical intervention to avoid malignant transformation.

In our case, the distinct feature was the presence of multiple swellings involving both mucosal surfaces of the upper lip—a

presentation scarcely reported in literature. The presence of a groove over the lesion suggested compartmentalization or pressure effects, possibly related to the slow expansion within confined tissue planes. CT imaging was instrumental in delineating the lesion's extent and identifying bony remodeling, which is an uncommon finding but has been documented in rare cases.

Surgical excision with adequate margins remains the treatment of choice^{6,9,10}. However, as noted by de Visscher et al.⁶, surgical approaches in the upper lip must balance complete tumor removal with aesthetic and functional preservation—a challenge in such a cosmetically sensitive area. In our patient, this was particularly critical due to the bilateral mucosal involvement, raising concerns of tissue distortion. Despite the rarity of the presentation, clear surgical margins were achieved, and the patient remained disease-free at six months post-operatively.

The literature consistently emphasizes that incomplete excision increases the risk of recurrence, which may occur even years later^{5,9,11}. Moreover, although rare, malignant transformation into carcinoma ex pleomorphic adenoma has been described, especially in long-standing or recurrent lesions^{2,4,10}. Therefore, long-term follow-up is warranted even in benign cases.

Paediatric cases of upper lip Pleomorphic Adenoma are exceedingly rare and management in such populations requires additional caution regarding growth and psychological impact. While our patient was not in the paediatric age group, the principle of conservative and complete excision remains the same.

Conclusion

Pleomorphic adenoma of the upper lip is a rare presentation of a common benign neoplasm of minor salivary glands. Our case underscores a unique variant involving multiple swellings across mucosal planes, with CT-demonstrated bony remodeling, emphasizing the need for thorough evaluation and individualized

surgical planning. Histopathology confirmed the diagnosis, and complete excision with preservation of aesthetics and function was achieved. The rarity of such presentations highlights the importance of reporting atypical cases, both to enhance clinical suspicion and to refine surgical approaches. Early diagnosis, careful radiologic and histologic evaluation, and long-term follow-up are critical to ensuring optimal outcomes and minimizing recurrence risk.

Conflicts of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Data Confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

Privacy policy, informed consent and Ethics Committee Authorization

The authors declare that they have written consent for the use of photographs of patients in this article.

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Availability of scientific data

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Declaration of Generative AI and AI-Assisted Technologies

During the preparation of this manuscript, the authors used perplexity AI to assist with language refinement, sentence restructuring, and improvement of clarity in certain sections

of the manuscript. The scientific content, clinical details, interpretation of findings, and conclusions were entirely generated by the authors.

After using this tool, the authors carefully reviewed, edited, and validated the content and take full responsibility for the accuracy, originality, and integrity of the manuscript.

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