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Case Report
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Pleomorphic Adenoma in Large Size Upper Lip: Report of Case

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Abstract

Pleomorphic Adenoma (PA) is a benign tumor of the salivary glands that can affect both sexes and usually presents as a slow-growing, painless solitary nodule. We report a case of a large pleomorphic adenoma in the upper lip in a 42-year-old man with a 6-month evolution, which due to its size could be mistaken for a malignant lesion. Biopsy is essential to close the diagnosis in these cases and to exclude the hypothesis of a malignant lesion; the treatment was performed with surgical excision.

Keywords: Pleomorphic adenoma; Salivary gland tumor; Benign neoplasm

Introduction

Pleomorphic adenoma is the most common benign mixed tumor of the salivary glands. May occur in both major and minor salivary glands [1,2]. This tumor usually presents as a solitary, painless, slow-growing nodular mass, with prevalence in females and with a higher occurrence between the third and sixth decade of life. Histologically, the lesion may present variable epithelial, myoepithelial and mesenchymal elements, usually presenting a well-circumscribed capsule. Epithelial cells can form ducts, islands, or cords of cells [3,4]. Differential diagnoses include mucoepidermoid carcinoma, salivary gland tumors, mucocele, neurofibroma, schwannoma and secondary infections. Treatment consists of surgical excision of the lesion with a safety margin. Incomplete removal cases can cause recurrences. Post-surgical follow-up is essential to assess the possibility of malignant transformation [3,5,6].

Case Presentation

Male patient, 42 years old, sailor, was referred to our department for presenting a painless swelling in the upper lip region. The patient reported evolution of the lesion of six months. During the anamnesis, the patient had a good general condition and reported being hypertensive controlled by medication. In the intra-oral clinical examination, the patient presented a large, solitary, painless, hardened, normocolored, pedunculated, smooth-surfaced tumor lesion in the region of the upper left labial mucosa, with an evolution of six months (Figure 1). Patient had a urine test result, performed six days before our appointment, without the presence of abnormalities.

Given the different diagnostic hypotheses, an excisional biopsy of the lesion was requested to clarify the diagnosis. The macroscopic material sent for analysis consisted of a soft tissue fragment, with a smooth surface, irregular shape, firm consistency, brown in color, measuring 20 mm × 10 mm × 10 mm (Figure 1). Histological sections revealed a fragment of glandular neoplasia. Neoplastic cells form ducts with two or seven layers of cells containing hyaline material in their interior. In some fields, neoplastic cells displayed eccentric plasmacytoid-like nuclei. In other fields, the presence of deposition of hyaline material and myxoid material was observed. The neoplasm was delimited by dense fibrous connective tissue and showed, in some fields, neoplastic cells with a polyhedral shape that, sometimes, presented eccentric nuclei with a plasmacytoid shape. Completing the histopathological picture, a discrete mononuclear inflammatory infiltrate, extravasation of red blood cells and ectatic blood vessels of varying sizes were present in the specimen (Figure 2). The final diagnosis was compatible with pleomorphic adenoma.

The treatment was performed by surgical excision of the lesion and was uneventful (Figure 2). Antibiotic, anti-inflammatory, and analgesic were prescribed to control postoperative symptoms. The patient had a satisfactory recovery and within the expected parameters.

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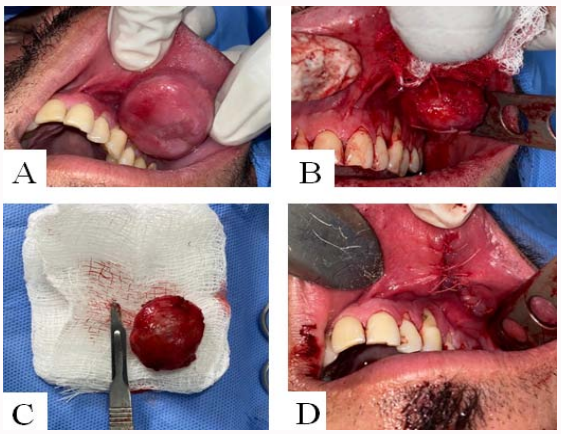

Figure 1: A) Initial appearance of the lesion. B) Surgical process. C) Surgical piece measuring 20 mm × 10 mm × 10 mm. D) Immediate postoperative final appearance.

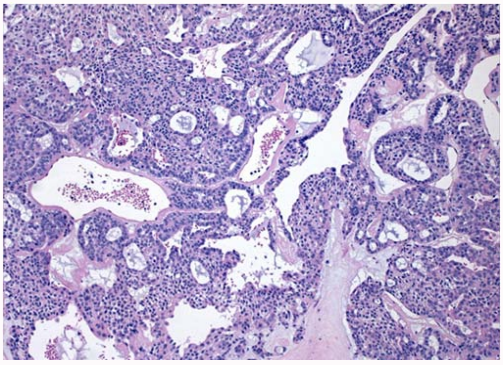

Figure 2: Neoplastic cells forming duct of cells containing hyaline material in their interior (H&E; 100x).

Discussion

Salivary gland tumors are complex lesions that are differentiated by the clinical-histological pattern. These lesions are rare and represent about 3% to 10% of all head and neck neoplasms [7]. Most represent benign tumors, about 75% to 85%; females are the gender most affected by these tumors, the parotid gland represents the most affected location, it can occur in any age group, however, the highest reports occur between the third and sixth decade of life [7-9].

The AP represents the most common neoplasm of the salivary gland. Most of these tumors arise in the parotid gland. Intraorally, the most common sites are the palate, upper lip and buccal mucosa, areas that have many minor salivary glands. The name of this lesion refers to its varied histology, composed of epithelial, myoepithelial and mesenchymal structures [1,2,9]. Clinically, the lesion is usually present as a unilateral, asymptomatic, slow-growing, well-circumscribed and most often encapsulated tumor, the overlying mucosa most often does not present ulceration [1,8].

Minor salivary gland tumors are uncommon and among them the pleomorphic adenoma is the most frequent. In a study of 82 cases of tumors in minor salivary glands carried out by Toida et al. [10], pleomorphic adenoma was the most common tumor intraorally, with the palate being the most common site (44 cases) and females the most affected, with a male/female ratio of 1:3.2 [10]. The occurrence of pleomorphic adenoma in the lip represents about 20% to 40% of all intraoral pleomorphic adenomas [11]. In the study by Toida et al. [10], of the 54 cases of intraoral pleomorphic adenoma, only 3 were in the upper lip [10]. The upper lip is more affected than the lower lip in a ratio of 6:1, the exact cause of this discrepancy is not known, but it is believed that it is due to differences in the embryonic development of the upper and lower lips, another theory is that the cells tumors would be destroyed by the continuous presence of inflammatory cells in the lower lip induced by frequent traumatic events [11-13]. Adenomas that occur on the lips appear to occur at a younger age than those that occur elsewhere, with a peak incidence between the third and fourth decade of life, with a mean of 33.2 years [2,6]. According to a bibliographic survey of 38 cases of AP in the lip performed by Suka, 2021, the upper lip was the most affected, with a predominance of the left side, males (21 cases) were more affected than females (17 cases), the mean age was 43.3 years and the mean diameter of the tumors was 2.6 cm [12]. This information corroborates our case, as it is a 42-year-old male patient with a lesion in the upper lip on the left side.

Due to its nonspecific clinical features, the differential diagnosis of AP in the upper lip includes a wide variety of pathologies: benign and malignant neoplasms of the salivary glands, sialolithiasis, leiomyoma, hemangioma, neurofibroma, schwannoma or infection with a foreign body [2,6]. The diagnosis of pleomorphic adenoma is made by histopathological analysis: It usually presents as a proliferation of encapsulated epithelial and myoepithelial cells with the presence of stroma, which can be myxoid, chondroid or hyalinized. The epithelium may form ducts and cystic structures or may appear as islands or nests of cells [2,4]. Imaging tests, such as magnetic resonance imaging and computed tomography, can aid in the diagnosis, as they allow a better visualization of the location and size of the lesion, in addition to being useful for differentiating between a benign or malignant lesion [3,12].

The treatment of choice for AP in the lip is tumor excision with a safety margin, but some authors suggest performing aspiration biopsy before definitive removal of the tumor, the prognosis is good. Recurrence cases may be associated with incomplete tumor excision or capsule rupture. Malignant transformation presents about 2% to 7% of cases [5,14]. Some authors report that the occurrence of AP in the upper lip is more related to benign tumors, while the lower lip is more related to malignant lesions [2,11,15].

Conclusion

Finally, AP should be investigated and considered as a differential diagnosis in cases of painless tumor masses of the lips. Due to its non-specific clinical characteristics, diagnosis may be difficult, and for this, biopsy is indispensable, imaging tests can also be useful. Surgical excision is the treatment of choice for this lesion and periodic follow-up should be performed to reduce the chances of recurrence and malignant transformation.

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