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Case Report

Management of a labial pleomorphic adenoma in the odontostomatology department of the Idrissa Pouye general hospital in Dakar

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ABSTRACT

Pleomorphic adenoma of the salivary glands (formerly called mixed tumor) represents 60% of oral salivary tumors. The accessory salivary glands have their preferential location at the palate, the lips of the cheek and the oropharynx. At the level of the lips, this tumor most often affects the right upper lip.

We report a case of pleomorphic adenoma of the right upper lip evolving for more than 4 years in a 70-year-old melanoderm patient who was referred to the odontostomatology department of the Idrissa Pouye General Hospital (HOGIP).

The swelling was large, slightly exceeding the midline of the upper lip. The swelling measured approximately 20 cm from its long axis. It was firm, painless and did not bleed on contact and covered with a healthy-looking mucosa. The management was surgical under local anesthesia and the postoperative course was simple.

Histological analysis specified that it was a well-limited nodular lesion, corresponding to a mixed epithelial and mesenchymal proliferation with a thin capsule in favor of a pleomorphic adenoma. No recurrence was observed 4 months after the operation.

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1. Introduction

Pleomorphic adenoma is a benign tumor of the salivary glands, with a double component, epithelial and mesenchymal. It represents 40 to 70% of benign tumors of the salivary glands.¹ It most often develops at the level of the main salivary glands, especially the parotid gland.²

Tumors arising from the minor salivary glands are rare clinical entities, accounting for 10–25% of all salivary gland tumors. The palate is the most common site among the accessory salivary glands where pleomorphic adenomas occur, but they can also occur in the upper lip,

cheek, floor of the mouth, larynx, and trachea. They are distinguished from the parotid by their clinical, histological and therapeutic characteristics,³ and are distributed between the submandibular gland and the accessory salivary glands (palate, lips, tuberosity and cheek).

This tumor can appear at any age and mainly affects women between the fourth and sixth decades. Clinically, it is a slow-growing lesion, which presents as a well-defined, firm and painless nodule on palpation, pushing back the mucosa without ulcerating it.⁴

However, if pleomorphic adenoma of the parotid gland is common, that of the accessory salivary glands is rarer, especially when it is located at the level of the upper lip, the latter was found in a case reported by Kazikdas et al.⁵

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In Senegal, no study had been conducted concerning labial pleomorphic adenoma. We report a case of pleomorphic adenoma located at the level of the upper lip in a 74-year-old patient.

2. Case Presentation

The case described concerned a 70-year-old melanoderm woman, a housewife, referred to the odontostomatology department of the Idrissa Pouye General Hospital in DAKAR (HOGIP) for a bothersome upper lip swelling that had been developing for approximately 4 years.

The interview only revealed hypotension that was not followed up. The history of the disease went back about 4 years with an onset marked by a small, painless swelling that gradually increased in size until it caused discomfort when chewing and swallowing, and speech disorders. The upper lip location and the size of the tumor mass contributed to the stigmatization and social exclusion of the patient.

Exobuccal examination, upon inspection, facial asymmetry was observed which is a consequence of an upper right labial swelling (figure 1). It occupied the subnarar region and a labial inoclusion occupied the right labial commissure. The covering mucosa is healthy in appearance.



Figure 1: Exoral view

On palpation, the tumor was firm in consistency, cartilaginous in appearance, painless, lifting the levator muscle of the nasal wing. Muscle tone was low, and no adenopathies were noted.

The intraoral examination showed poor oral hygiene and unrehabilitated edentulism in the maxillary and mandibular incisor -canine sectors.

Examination of the upper lip revealed a large swelling on the right side, slightly extending beyond the frenulum. The

swelling, touching the vermillion, measured approximately 35 mm along its long axis and 10 mm in height. The nodule was submucosal, well-demarcated and not fixed on the deep and superficial planes (Figure 1).

It was a slow-growing lesion that had been developing for about four years, causing discomfort when chewing, swallowing and speaking. The diagnostic hypothesis was in favor of a benign tumor of the oral mucosa (fibroma, pleomorphic adenoma).

Due to its slow progression, its cartilaginous appearance on palpation, its painlessness, its normal covering mucosa and its topographical location, the presumed diagnosis was in favor of a pleomorphic adenoma.

Surgical excision, under local anesthesia, was performed. It consisted of a superficial incision of the mucosa 2 cm long along the long axis was made using a number 11 scalpel blade followed by dissection of the fibers of the orbicularis oris muscle (Figure 2).



Figure 2: Superficial incision and dissection of the mucosa to avoid damage to the noble anatomical organs

2.1. Yellowish, oval, encapsulated mass was revealed after exobuccal digital pressure

Hemostasis of the surgical site was achieved by manual compression and simple sutures on two levels, deep and superficial, using absorbable threads for the deep level and non-absorbable threads on the superficial level.

The surgical specimen was cut in the middle to allow the formalin to diffuse inside the cells so as not to lose its properties. It also allows the encapsulated appearance of the tumor to be clearly seen (Figure 3 A, B).

The postoperative prescription was based on:

1. Amoxicillin and clavulanic acid at a rate of 1g/125mg, 3 times a day,
2. Paracetamol at a rate of 3g per day,
3. Corticosteroid at a rate of 1mg/kg/day in a single dose and without exceeding 4 days of treatment,

4. Chlorhexidine-based oral antiseptic, rinsed 3 times a day.

Checks were carried out on D3 and D10 after the operation, the follow-up was simple (Figure 4).

Two weeks after the intervention we observed complete healing of the surgical wound without fibrosis with restored aesthetics (Figure 4).

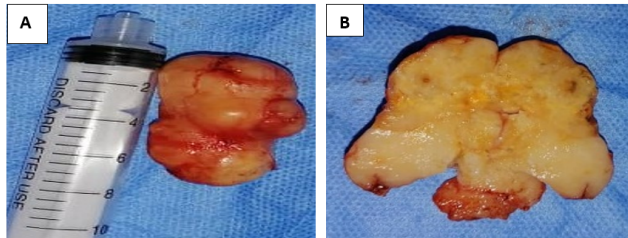


Figure 3:



Figure 4: Exoral view at D10

Extemporaneous macroscopic examination showed a surgical specimen measuring 4 x 2.5 x 2 cm and weighing 10 grams, yellowish-white in color, with an irregular and bumpy surface. When cut, the section slices are tissue-like, nodular with calcific changes.

Histological analysis of the sections performed showed a benign tumor proliferation with a dual epithelial and connective tissue component. It consisted of irregular tubular epithelial structures formed of cylindrocubic cells with more or less abundant cytoplasm, containing an ovoid nucleus with fine chromatin. It was ultimately a pleomorphic adenoma (Figure 5).

3. Discussion

Salivary gland tumors are rare. They represent less than 4% of all head and neck tumors. Pleomorphic adenoma is the

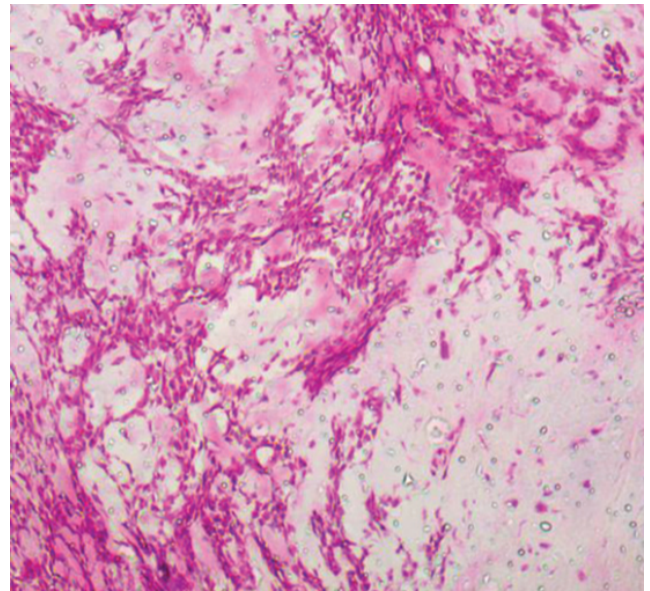


Figure 5: Histological section of the tumor: serous acini (dark red), mucous membrane (light red) and excretory duct (HEX100)

most common among these and represents approximately 70%. These tumors occur more frequently in women according to the cases reported in the literature.^{1,6,7} Thus, our clinical observation concerned a seventy-year-old melanoderm woman. Nevertheless, the work of Wu Y et al. and Youstra et al. showed a prevalence of labial pleomorphic adenoma of 52% in men.^{1,8}

The age group of patients most often affected is between the fourth and sixth decades.⁷ However, the work of Wu Y et al, showed that these lesions occurred in patients whose age was between 20 and 70 years.⁸

Pleomorphic adenoma is a slow-growing benign tumor. Consultation time is often late because it develops in the form of an isolated labial swelling often discovered after a few years of development.⁹

Thus, in our patient, the consultation period was 4 years; this could be explained by the lack of means, the use of traditional medicine and the inaccessibility of specialized structures. Furthermore, similar studies by Salha et al.¹⁰ and Kalwaniya et al.,⁷ also noted an average consultation period of 4 years with extremes ranging from 1 month to 13 years.

Palatal localization is more frequent and represents approximately 69% of cases, labial localization is rare. Indeed, a study conducted by Almeslet et al. showed a predominance at the palate level (between 42.8-68.8% of cases) followed by the upper labial localization.¹¹

In our study, a bulky one swelling was observed at the level of the upper lip on the right side slightly extending beyond the frenulum and touching the vermilion.

According to the work of Bhatia et al., Umemori et al.,^{12,13} pleomorphic adenomas were located in 10% of cases at the upper lip level and alone 2% of cases

concerned the lower lip. Bidigital palpation allows for a better assessment of the characteristics of the lesion (its size, location, consistency and possible adhesion to the skin or deep plane).¹⁴

In our patient, physical examination found a superior sublabial mass measuring 35 mm in its long axis, of firm consistency, painless on palpation, mobile in both planes (deep and superficial), and lifting the levator muscle of the nasal wing. The overlying mucosa was healthy, the study of Sood et al¹⁵ showed the presence of a pink, non-ulcerated mucosa of normal appearance, except in cases of severe trauma.

Tumors of the salivary glands of the lips are characterized by the discretion of the functional signs. In some cases, labial pleomorphic adenomas can manifest themselves by discomfort when chewing, speech disorder and signs of nerve compression.² In our study, no signs of malignancy were found (pain, nerve damage, fixity). The risk of ulceration is rare and can be explained by trauma from a bite or iatrogenic trauma, following a biopsy.

The differential diagnosis of pleomorphic adenoma of the lips is made with several conditions:

1. Mucoïd cyst presents a translucent, often bluish neoformation containing viscous mucoïd fluid and fluctuating consistency,
2. The Lipoma is sessile, yellowish in color and soft in consistency, sometimes giving the impression of a false fluctuation,
3. Canalicular adenoma is preferentially located in the upper lip and is characterized by the absence of chondroid or myxoid matrix.¹⁶

To aid in the diagnosis of pleomorphic adenoma, some authors mention the usefulness of ultrasound for the salivary glands, magnetic resonance imaging (better identification of the internal characteristics of the tumor) and computed tomography (for calcifications).¹³

Regarding magnetic resonance imaging, pleomorphic adenoma most often presents with the typical appearance of a well-limited, encapsulated tumor, with a characteristic lobulated appearance, in T1 hyposignal, enhancing homogeneously after injection of gadolinium.¹⁷

Treatment is surgical by excision under local or general anesthesia, depending on the size and difficulty of access.^{2,18} The increase in tumor volume often linked to late consultation increases the risks associated with excision during incision and dissection. In our clinical observation, excision under local anesthesia was performed due to the location of the tumor and its size. Any direct incision with lifting of a mucosal flap, in order to allow suturing at the end of the procedure, must be avoided, given the risk of leaving a few tumor islands in the thickness of the mucosa.¹⁹

Therefore, tumor excision requires deliberate sacrifice of the mucosal lining to avoid recurrence.¹⁹ The literature

reports a recurrence rate of 6% of affected patients.^{1,6}

The anatomopathological study of the excised fragments shows a benign tumor proliferation with a dual epithelial and connective component. It consists of irregular tubular epithelial structures formed of cylindrocubic cells with more or less abundant cytoplasm, containing an ovoid nucleus with fine chromatin. This corroborates the study of Rochefort.²

Follow-up of patients with benign tumor of accessory salivary glands should be periodic because of the possibility of late recurrences if excision is not complete. In our clinical observation, no recurrence is reported after one year.

The risk of malignant degeneration has been reported in rare cases, after a long period of evolution.¹⁹

The 5-year and 10-year overall survival rates are 80% and 72%, respectively.²⁰

4. Conclusion

Pleomorphic adenoma of the accessory salivary glands accounts for 60% of all oral salivary tumors.

Clinically, it presents as a firm, painless, slow-growing swelling that does not cause ulceration of the overlying mucosa but may manifest as discomfort when chewing and speech disorders. Clinical and paraclinical data guide the diagnosis, the confirmation of which is exclusively histological. The treatment remains surgical excision. Postoperative monitoring of these tumors is important due to the risk of recurrence.

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None.

6. Conflict of Interest

None.

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