

Oncocytic Papillary Cystadenoma of Minor Salivary Glands: A Case Report

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ABSTRACT

Oncocytic papillary cystadenoma is an extremely rare benign tumor that can affect salivary glands. Few cases of this entity have been reported in medical literature, making the clinical diagnosis very challenging.

This paper reports the case of an 84-year-old female patient who was referred for a nodular lesion in the right side of the upper lip. Over the span of 8 months, the patient's nodule increased in size, making it impossible to insert the maxillary prosthesis.

Clinical examination revealed partial labial eversion, caused by the presence of a painless bluish nodular lesion in the right side of the upper lip.

A complete excision of the lesion was performed under local anesthesia. Histological examination led to the diagnosis of an oncocytic papillary cystadenoma, and the postoperative course was straightforward with no complications.

This case aims to highlight the clinical manifestations, histological characteristics and the possible differential diagnoses of this rare type of salivary gland tumors.

Keywords: Benign tumor, minor salivary glands, oncocytic papillary cystadenoma.

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1. INTRODUCTION

Salivary gland tumors comprise 2% to 6% of all head and neck neoplasms, 80% of these tumors are benign [1]. The parotid gland is the predilection site, accounting 60% to 75% of all salivary gland tumors, followed by the sub-mandibular gland (10% to 15%) [2]. Minor salivary glands are rarely involved; they account for approximately 14,7% of all salivary gland tumors [3].

We report a case of an oncocytic papillary cystadenoma located in the upper lip, which reveals another clinical manifestation of this rare type of salivary gland tumors. This type of tumor exhibits a wide range of clinical characteristics, raising many diagnosis possibilities, which make histopathological examination necessary for the final diagnosis.

2. CASE REPORT

Patient information: An 84-year-old female patient was referred to the Department of Oral Medicine and Oral Surgery at Ibn Roch University Hospital (Casablanca, Morocco) regarding an oral nodular mucosal lesion, located on the upper lip. The mass had gradually increased

in size over the past 8 months. Anamnestic investigation revealed no significant medical or surgical history. There was no history of antecedent trauma, and the patient had been edentulous for over 13 years due to dental caries.

Clinical findings: Extra-oral examination revealed a slight face asymmetry, due to a mild swelling of the right upper lip (Fig.1).

Clinical intra-oral examination revealed three well-defined nodules, round to oval, located in the buccal mucosa of the upper lip. The overlying mucosa appeared bluish-purple, smooth, translucent, and showed no signs of inflammation. On palpation, the mass was firm in consistency, mobile and painless (Fig. 2).

Orthopantomography did not reveal any pathology.

Diagnostic assessment: Based on these findings, a variety of diagnoses were suggested; benign tumor such as a lipoma, or cystic lesion such as a mucocele.

A complete surgical excision of the lesion followed by histological examination were decided for both diagnostic and therapeutic purposes.

Therapeutic intervention and histopathological examination: Under local anesthesia, a median incision, limited to the superficial plane of the inner labial mucosa, was performed along the length of the lesion. The nodule





Fig. 1. Extra-oral examination revealing upper right lip swelling.



Fig. 2. Clinical maxillary occlusal view showing three well-defined nodules on the buccal mucosa of upper lip; the overlying mucosa appeared bluish-purple and translucent.



Fig. 3. Intra-operative image after incision revealing the well-circumscribed nature of the lesion.

was removed gently through a careful dissection using a dissecting scissor (Fig. 3).

After excision, the surgical specimen measured $2.5 \times 1.5 \times 1.3$ cm (Fig. 4).

The excess mucosa was excised (Fig. 5), and wound edges were closed using simple interrupted sutures.

Histopathological examination described an encapsulated nodular lesion exhibiting a cystic architecture in certain areas; partially lined by a regular cylindrical epithelium; and the presence of papillary tumor proliferation.



Fig. 4. Macroscopic appearance of the Oncocytic Papillary Cystadenoma, the specimen measuring: $2,5 \times 1,5 \times 1,3$ cm.



Fig. 5. Intra-operative image showing the excision of the overlying mucosa in excess after removing the nodules.

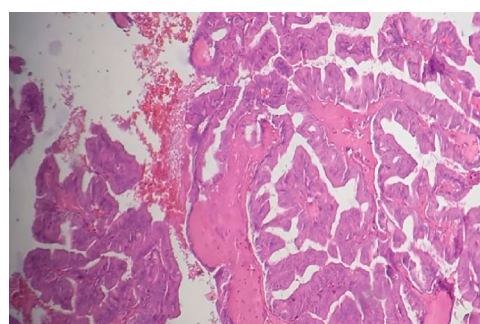


Fig. 6. Histopathologic features revealing papillary projections lined by oncocytic cells with no nuclear atypia.

The papillae were short with a vascular core and covered by a stratified oncocytic epithelium. There was an absence of nuclear atypia (Fig. 6). These findings were consistent with a diagnosis of an oncocytic papillary cystadenoma of the minor salivary glands.

Additional immunohistochemical staining was not required in this case, given the clearly identifiable morphologic characteristics of the cells under microscopic examination.

Follow-up and outcome of intervention: The patient experienced an uneventful postoperative recovery and

expressed satisfaction with the outcome. No sign of recurrence has been observed at the one-year follow-up. A periodic follow-up is still programmed to ensure no recurrence.

Informed consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

3. DISCUSSION

Oncocytic papillary cystadenoma (OPC) is defined as a benign epithelial tumor that can involve both major and minor salivary glands. The incidence of this histological entity is extremely rare. For instance, Santana *et al.* reported only one case of papillary cystadenoma among 238 cases of benign salivary gland neoplasms [4]. Similarly, a retrospective study evaluating the frequency and distribution patterns of minor salivary gland tumors found that only 1.1% of the 485 patients concerned presented a cystadenoma [5]. Epidemiological data further suggest that cystadenomas most commonly affect women over the age of 40 [6].

OPC is characterized by multilocular, slow-growing cysts that preferentially arise in the minor salivary glands. Despite this preference, its incidence remains very low, accounting for only 0.9% to 2% of all minor salivary gland tumors [7].

Minor salivary glands of the lip are the most frequently affected by oncocytic papillary cystadenoma (OPC), although cases have also been reported where OPC occurs on the buccal mucosa, palate, and ectopic locations such as the larynx [8], [9].

Concerning OPC's size, Tjioe *et al.* reported that, among the 11 cases of Papillary Cystadenoma of Minor Salivary Glands they described, the size of the tumors was usually small and had an average of 1.7 cm. In our case, the lesion measured 2.5 cm in greatest dimension, which is higher than the values usually reported in the literature [10]. According to the same study, most of the reported cases involved well-circumscribed lesions, which align with our case.

Histologically, Cystadenomas can be observed in two main forms: papillary and mucinous. The papillary variant is characterized by projections into the cyst lumen and can appear as unilocular or multilocular structures [11].

OPC is identified by the coexistence of oncocytic cells and a papillary configuration, in the absence of lymphoid cells or cellular atypia [2], [10]. It is suggested that these oncocytic cells are the result of the transformation of normal ductal or acinic cells which is probably associated with an inflammatory process or age-related physiological changes [12].

The differential diagnosis for OPC includes mucocele, especially when the lesion occurs on the lips [11]. Warthin tumor (WT) can also be confused with OPC due to histological similarities, but the absence of lymphoid stroma in OPC rules out a diagnosis of WT [6], [7]. Also, the absence of nuclear atypia or mitotic activity excludes a diagnosis of cystadenocarcinoma.

Complete surgical excision is the recommended treatment for OPC, with postoperative courses usually being

simple and uneventful. Recurrence is exceptionally rare owing to the well-circumscribed nature of OPC [2], [8]. However, recurrences can occur due to incomplete excision or misdiagnosis of cystadenocarcinoma, making follow-up highly recommended [13]. Additionally, no case of malignant transformation of OPC has been reported in the literature [10], [14].

Despite the insights provided by this case about OPC of minor salivary glands, further documentation of similar cases and larger case series with long-term surveillance are recommended to assess the risk of recurrence or malignant transformation.

4. CONCLUSION

In summary, OPC is an extremely rare benign salivary gland tumor characterized by a slowly growing painless mass that is well-circumscribed, and which can reach remarkable size in some cases.

Neoplastic lesions of minor salivary glands as OPC should be considered in the differential diagnosis when evaluating nodular lesions of the lips.

Diagnosis is confirmed only by histopathological examination. The management of OPC involves total surgical excision, with simple postoperative courses and very rare risk of recurrence when the lesion is totally excised.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

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