

Asymptomatic Lower Lip Swelling...Not Always Benign

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Abstract

Case Report

Although adenoid cystic carcinoma (ACC) is the most common tumor of the minor salivary glands, it's relatively rare to affect the lower lip. Its non-specific clinical presentation may pose a diagnostic dilemma. Therefore, biopsy and histological examination are of extreme importance to establish the final diagnosis. In this paper, we report a case of ACC of the lower lip occurring in 35-year old patient mimicking a benign lesion.

Keywords: Adenoid cystic carcinoma, minor salivary glands, salivary gland neoplasms, lower lip, histopathology.

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INTRODUCTION

With a reported incidence rate of about 1% of all head and neck malignancies, adenoid cystic carcinoma (ACC) is an uncommon malignant tumor. It constitutes about 10% of all tumors of the salivary glands and considered as the most frequent cancer of the minor salivary glands (MSG) [1, 2].

Among ACC of the MSG, 50% arises on the palate. Other less common intra-oral sites are the floor of the mouth, the buccal mucosa and the lip [3, 5].

ACC occurs at all ages with a peak incidence after the 5th decade and without any gender predilection. No factors including smoking are known to have an effect on the incidence [1-3].

CASE REPORT

A 35-year old female patient was referred to the Oral Medicine Oral Surgery Department, University Clinic of Dental Medicine, and Monastir, Tunisia in September 2019 for extraction of the left upper third molar. Patient had no significant medical history.

Clinical examination revealed a 1cm × 1 cm, dome shaped, palpable, soft-to-firm, well-demarcated mass on the left side of lower lip with normal overlying mucosa (Figure 1). The patient had only noticed the swelling few weeks before consultation. Digital pressure did not evoke pain. No signs of dysesthesia were noted and no lymph nodes were palpable on the neck area.



Fig-1: Clinical image of a mass of the lower lip

Based on the clinical features, the provisional diagnosis was in favor of a benign lesion: fibroma and mucocele were considered. Therefore, the decision to postpone the extraction was made and the excision of the lesion was carried out under local anesthesia. To expose the lesion, a linear incision was made using a number 15 scalpel blade. A blunt dissection was then used to facilitate the resection of the lesion from its base. Intraoperatively, the mass was noted to have poorly-defined limits with an unclear demarcation between the lesion and the surrounding tissue (Figure 2). An unexpected moderate bleeding was successfully controlled by applying local pressure and ligation. Intermittent sutures were then placed to assure primary closure. The postoperative period was uneventful. The surgical specimen was sent for histological examination.

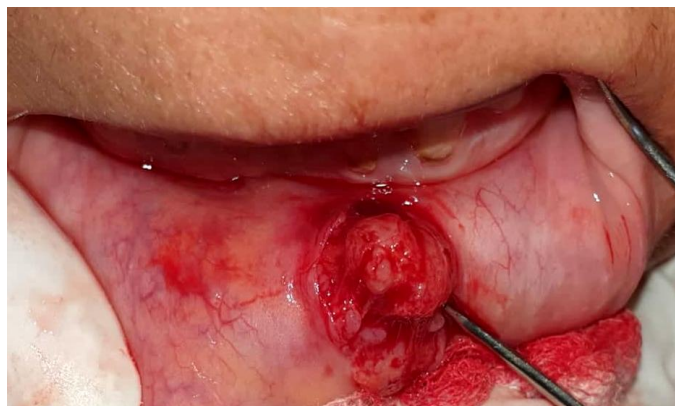


Fig-2: Intraoperative view of the lesion

Histopathological diagnosis was consistent with ACC of the MSG. The surgical margins were positive. Hence, the patient was referred to an Oral and

maxillofacial Surgery Department. She underwent a large surgical resection of the tumor. The microscopic analysis confirmed that the margins were clear.

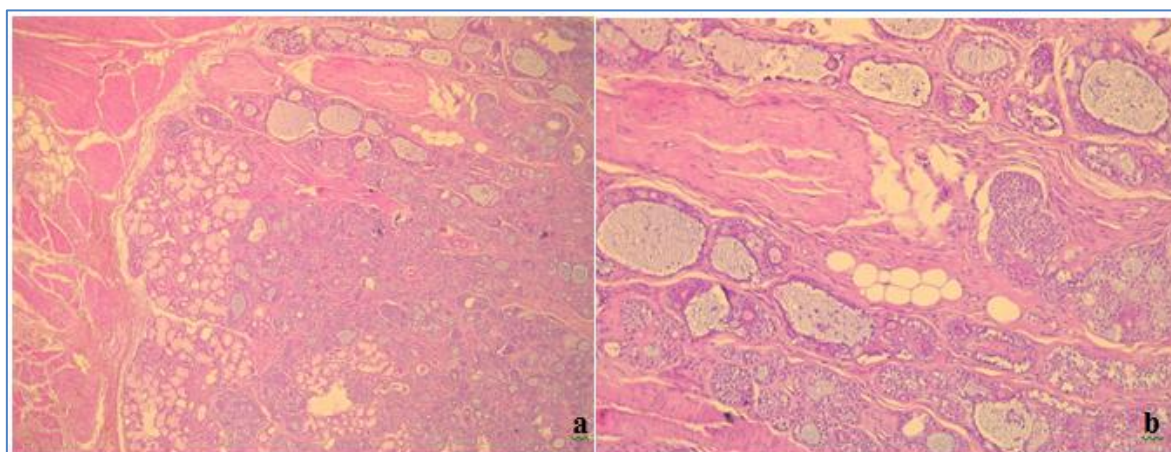


Fig-3 (a): adenoid cystic carcinoma formed by tubular and cribriform structures filled with lightly basophilic myxoid ground substance, HEx40 (b) the tumor shows a large nerve perineural invasion HEX100

DISCUSSION

Originally referred to as “Cylindroma” the nomenclature was later reconsidered and the current name ACC was introduced [1, 3]. ACC has a unique clinical behavior characterized by slow but infiltrative growth pattern, frequent local recurrences, perineural invasion and late distant metastasis [4, 5]. It has been described as an aggressive and unpredictable malignancy of the head and neck [1].

Initially, ACC of the lower lip is commonly presented as a painless slow firm growing mass with ill-defined margins [1, 3, 5]. The initial lack of symptoms may lead to a late diagnosis of the tumor at a more advanced stage [6]. Pain and dysesthesia are the second frequent symptoms and reflect its tendency for perineural invasion. As observed in this case, its benign appearance, slow evolution and absence of neurological deficit contribute to pass unnoticed even by the patient [5].

Regional Lymph nodes are rare and are reported in 5 to 25% of all cases [3]. In the contrary,

distant metastasis occurring via the hematogenous route, is more common ranging from 35 to 50% and usually involve the lungs, bone and liver [1].

ACC is a malignant epithelial tumour composed of ductal and myoepithelial cells arranged in various combinations. Three distinct histologic subtypes have been described: the cribriform variant considered as the most common (46.3%), followed by the solid (38.3%) and the tubular patterns (14.9%) [3, 8].

A frequent histopathological feature is propagation of the malignant cells within the perineural spaces or in the perineural sheaths [7]. Since ACC is considered a high-grade tumor, complete surgical resection with 1-2 cm margins is the current “gold-standard” treatment and is practically followed in all cases by adjuvant radiotherapy due to the high rates of its local recurrences for an optimal result [1, 3, 4, 6]. Although, postoperative radiotherapy is turning out to be the rule rather than the exception, its role is still a subject of controversy and its indications vary within and between the institutions [3,4]. Despite some are in favor of postoperative radiation therapy as an essential

modality for all patients presenting ACC, others limit its use to patients with adverse features such as positive surgical margins and perineural or lymphovascular invasion.

Generally, ACC of the head and neck has a poor prognosis which explains why this neoplasm is regarded as “clinically high-grade” [1]. The 5 year survival rate reported by Van Weert et al. was 68% but the 10 year survival rate dropped to 52% and only 28% of ACC patients were alive at 20 years [9]. The low long-term survival rate correlates with the presence of distant metastasis [1, 3, 4].

The features associated with poorer outcomes include solid histological patterns, over-expression of ki-67 and p53, perineural invasion, lymphovascular invasion, positive surgical margins and advanced stages. High ki-67 and p53 expression is most notable among the solid subtype [4,10].

The diagnosis of asymptomatic swelling of the oral cavity may turn out to be a challenging task. The differential diagnosis includes: traumatic fibroma, mucocele, schwannoma, salivary gland neoplasm and even hemangioma. The clinician should not hesitate to perform a biopsy when the clinical findings and the imaging tests are insufficient to establish the diagnosis. It's the simplest, speediest and most efficient way to obtain the accurate diagnosis and prevent unnecessary delays that may affect the patient's welfare. Histopathological study is regarded as the golden standard exam to detect and diagnose oral cavity malignancies [11-13].

In the case of ACC of the MSG, the effective management of this malignant tumor is favored by its detection at an early stage leading to start the treatment in the briefest time possible.

CONCLUSION

ACC of the MSG is a relatively rare neoplasm characterized by a paradoxical clinical behavior. It's commonly a slow growing malignancy with insidious evolution, making the diagnosis more complicated especially in the cases with a total absence of symptoms.

The inability to control distant disease is often associated with poor survival rate. Thus, a long-term follow-up of ACC patients is of utmost importance for the detection of recurrence as early as possible.

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