

# Painless Palatal Swelling Revealing Adenoid Cystic Carcinoma: Case Report

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## Abstract

## Case Report

**Background:** Adenoid cystic carcinoma (ACC) is a rare malignant tumor of the salivary glands, most frequently arising from the minor salivary glands of the palate. Despite its slow growth, ACC is characterized by an insidious course, marked perineural invasion, and a high propensity for local recurrence and distant metastasis. Early diagnosis remains challenging, as palatal lesions may initially mimic benign or inflammatory conditions. **Case Presentation:** We report the case of a female patient presenting with a painless palatal swelling initially misdiagnosed as a palatal abscess. Imaging revealed a soft-tissue mass with focal bone lysis and extension toward the maxillary sinus and nasal floor. Histopathological analysis confirmed ACC with predominant cribriform and tubular patterns and focal perineural invasion. The patient underwent radical surgical resection with oncologic margins, followed by reconstruction using a para-lateronasal approach and temporalis fascia flap. **Discussion:** This case illustrates the deceptive clinical presentation of palatal ACC and highlights the importance of advanced imaging in detecting subtle bone and sinus involvement. The histopathological features identified—particularly the cribriform/tubular architecture and perineural invasion—correlate with the known biological behavior of ACC. Radical surgery with postoperative radiotherapy remains the standard of care for resectable lesions, while long-term surveillance is critical due to the high rate of late recurrence and distant metastasis. **Conclusion:** Any persistent palatal swelling under intact mucosa should prompt consideration of ACC, especially when imaging demonstrates bone erosion or sinus extension. Early recognition, appropriate surgical planning, and vigilant follow-up are essential to optimize outcomes given the tumor's aggressive and unpredictable behavior.

**Keywords:** Adenoid cystic carcinoma; Palate; Minor salivary glands; Perineural invasion; Surgical resection; Case report.

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## INTRODUCTION

Minor salivary gland tumors account for about 41.5% of all salivary gland neoplasms, with malignancies comprising nearly 60% of these cases [1]. The palate is the most common site for intraoral minor salivary gland carcinomas, and adenoid cystic carcinoma (ACC) is its most common histologic type. ACC is also one of the most common cancers of the major salivary glands (the parotid, submandibular and sublingual salivary glands). ACC can also involve lacrimal and ceruminous glands as well as other sites in the head and neck.

The very first description of ACC was in 1853 and 1854 by Robin, Lorain and Laboulbene in two different articles reporting on two nasal tumors and one

parotid. [2] They described the cribriform arrangement of tumor cells very characteristic on the microscope. Thus, its first name by Billroth in 1859 using the term Cylindroma. The current name of “adenoid cystic carcinoma” was introduced by Spies in 1930.

ACC was regarded as variant of the benign mixed tumor until Dockerty and Mayo established the malignant nature of this tumor. The exact etiology of this neoplasm remains unknown. However, recent studies have shed light on molecular abnormalities associated with ACC development and progression. The most common genetic abnormalities involve translocations of the myeloblastosis (MYB) gene on chromosome 6q and the nuclear factor 1 B (NF1B) gene on chromosome 9p [3].

The treatment for ACC is typically customized based on the tumor's location and stage. The common treatment for ACC is radical surgical resection, ensuring free margins, and postoperative radiotherapy.

We present a case of adenoid cystic carcinoma of the palate, along with a concise review of the literature focusing on its clinical, histopathological, and therapeutic characteristics.

## CLINICAL FINDINGS

A female patient with no relevant medical history presented with a painless unique lesion on the palatal mucosa growing back to several weeks. The lesion was initially presumed to be a palatal abscess (Figure 1). Despite large spectrum antibiotic therapy, no clinical improvement was observed.



**Figure 1: Palatal mucosal swelling associated with an ulcerative lesion**

Panoramic radiography revealed no bone lysis or dental involvement. Cone-beam computed tomography (CBCT) demonstrated no sinus obstruction or bone destruction, but a slight deformation of the palatal vault was visible on frontal sections.

A biopsy (figure 2; figure 3) of the palate lesion confirmed the diagnosis of adenoid cystic carcinoma

(cylindroma). Histopathological examination revealed a proliferation of epithelial cells arranged in tubular and cribriform patterns, with hyaline material within the lumina. The tumor cells showed enlarged nuclei with fine chromatin and eosinophilic cytoplasm. A focal perineural invasion was noted.



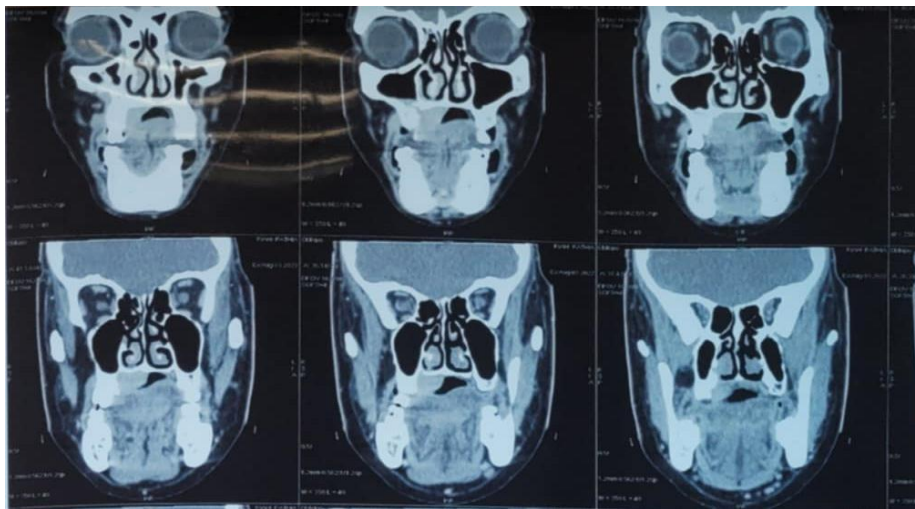
**Figure 2: limits of the palatal biopsy**



**Figure 3: Biopsy sample**

A contrast-enhanced CT scan was demanded to explore the extension of the lesion (figure 4). It showed an oval-shaped soft tissue mass centered on the upper right part of the oral cavity, measuring  $34 \times 19 \times 17$  mm, with homogeneous enhancement. The lesion was in contact with the soft palate and associated with focal

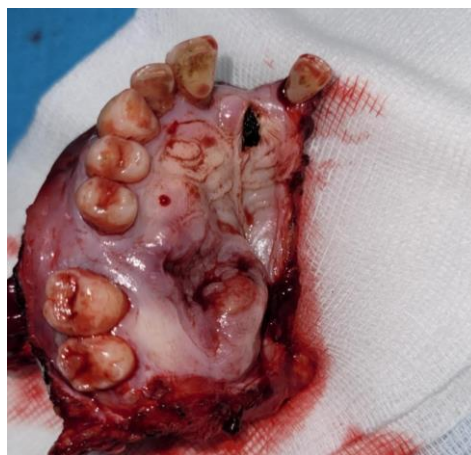
bone lysis of the maxillary bone and the lower wall of the maxillary sinus, accompanied by fine tissue extensions into the sinus cavity and nasal floor, suggesting a locally aggressive and rapidly progressive lesion. No cervical lymphadenopathy was detected, and the remaining craniofacial and cerebral structures appeared normal.



**Figure 4: contrast-enhanced CT scan showing oval-shaped soft tissue mass centered on the upper right part of the oral cavity, measuring  $34 \times 19 \times 17$  mm**

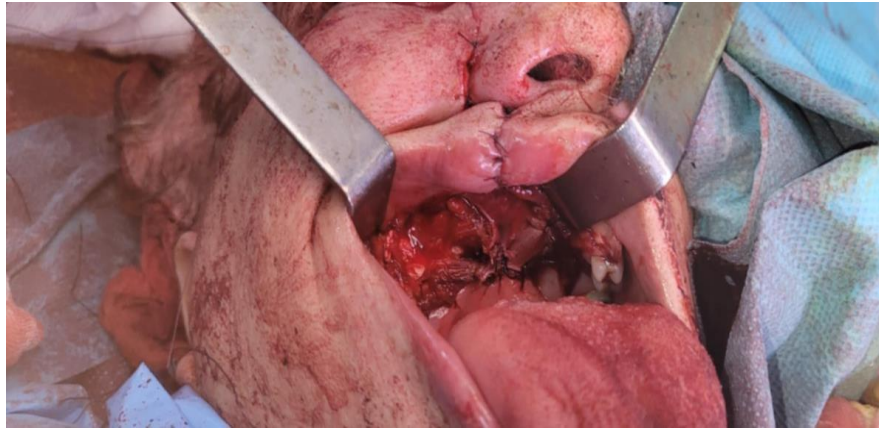
Definitive management consisted of a surgical excision with oncological safety margins. A para-lateronasal flap was used to expose the lesion, and to

ensure functional and aesthetic restoration a temporalis fascia flap was added. (figure5; figure 6; figure7)



**Figure 5: Surgical specimen**





**Figure 6: para-lateronasal flap suturing after tumor resection**



**Figure 7: temporalis fascia flap suturing with drain strip**

## DISCUSSION

Adenoid cystic carcinoma (ACC) is considered a rare malignant tumor of the salivary glands, accounting for approximately 1% of all head and neck malignancies and about 10% of salivary gland tumors [4]. It most frequently arises in the minor salivary glands, particularly in the palate, followed by the parotid and submandibular glands. Despite its slow growth, ACC is characterized by an insidious course, marked perineural invasion, and a high rate of local recurrence and distant metastasis, often to the lungs and bones.

Clinically, palatal ACC often presents as a painless, firm swelling under intact mucosa, which can easily be misinterpreted as a benign or inflammatory lesion, as occurred in the present case initially treated as a palatal abscess. Pain or ulceration tends to occur later, usually indicating perineural or bone invasion. [5] This may be a sign of the local aggressiveness of the tumor or associated with advanced- stage disease.

Radiological exams play a key role in evaluating local and distant extent of the tumor. Preoperative diagnostic imaging of ACC includes computed tomography (CT) and/or magnetic resonance imaging (MRI). This allows estimating the anatomical

disease extent, which is crucial for accurate surgical planning. It is well accepted that CT is better at outlining bone invasion, whereas MRI superior for assessing soft tissue extension. In our case, contrast-enhanced CT imaging revealed an oval-shaped soft-tissue mass with focal bone lysis of the maxillary bone and the lower wall of the maxillary sinus, as well as soft tissue extension into the sinus cavity and nasal floor, this indicates a locally invasive pattern.[5] A CT study of patients with ACC showed that in the palate this tumor is typically diagnosed at a more advanced stage than other tumor types, as it tend to infiltrate surrounding structures, extending through bone into the maxillary sinus, nasal cavity, skull base, or even the cavernous sinus.

Histopathological examination remains the gold standard for diagnosis and is particularly essential when the proposed treatment includes radical surgery or potential sacrifice of the facial nerve.

Histologically, ACC displays three classic architectural patterns: cribriform, tubular, and solid. They may occur either separately or together in the same tumor. [6] The cribriform pattern is characterized by islands of scattered pseudocysts of small, hyperchromatic myoepithelial neoplastic cells. The

tubular pattern consists of pseudoducts and tubules, which are lined by one or several layers of myoepithelial cells, often surrounded by a hyaline stroma. The solid type is formed by islands of small, hyperchromatic neoplastic cells.

The cribriform pattern is the most common and often corresponds to intermediate prognosis, while the solid pattern is associated with worse outcomes [7]. In our case the biopsy revealed a predominant cribriform/tubular pattern with focal perineural invasion. Perineural invasion is particularly significant, as it correlates with higher local recurrence and distant metastasis rates. In fact, it can extend through the nerves at a considerable distance from the primary tumor. However, perineural invasion as a prognostic factor is still considered ambiguous.

Treatment of ACC is influenced by tumor location, stage at diagnosis and its biologic behavior which is determined by the histologic grade. [6] In the Spiro system, tumors are classified as grade I (occasionally solid tubular or cribriform patterns), grade II (substantial solid > 50%), and grade III (solid only).

The “gold-standard” treatment for ACCs, that is considered as potentially resectable, is radical surgical resection, ensuring free margins, and postoperative radiotherapy [8]. In the literature review, the highest radiation doses administered were 70 Gy for tumors with positive margins. The treatment was delivered at a daily dose of 1.8 to 2 Gy over a period of 7 to 8 weeks [7]. ACC is considered chemoresistant. Thus, chemotherapy is limited only to palliative cases. The main drugs of choice were cisplatin and 5-fluorouracil [9]. It is noted that there is no role for adjuvant chemotherapy after locoregional treatment of ACC.

Overall, the prognosis of adenoid cystic carcinoma of the head and neck remains poor, as many clinicians consider complete cure unlikely. [10] Although favorable 5-year survival rates are occasionally reported, long-term outcomes are generally limited due to the high incidence of distant metastases—most commonly to the lungs. [11] Given the tumor’s indolent but persistent nature, long-term follow-up is essential, as survival extending beyond 15 to 20 years is possible, and metastatic disease may develop more than five years after initial treatment.

## CONCLUSION

In summary, this case highlights several key lessons. Any palatal swelling with intact mucosa—even if painless—should raise suspicion for ACC mainly when imaging shows bone erosion or sinus extension. Imaging findings of bone destruction or maxillary sinus or nasal involvement indicates preoperative planning for aggressive resection. Functional reconstruction is important in the palate to preserve speech and

swallowing. Vigilant long-term surveillance is critical given the tumor’s poor prognosis and high recurrence.

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