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Long-Standing Pleomorphic Adenoma on a Female's Palate: A Case Report

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ABSTRACT

This case report elucidates the significant oncologic risk associated with long-standing pleomorphic adenoma (PA), wherein the cumulative risk of malignant transformation to carcinoma ex pleomorphic adenoma (CXPA) escalates over time. It details the management of a 46-year-old female with a 25-year history of a palatal mass, which exhibited recent rapid growth—a key clinical warning sign. Diagnosis was confirmed via incisional biopsy following clinical and cone-beam computed tomography assessment, which revealed bony erosion. The patient was successfully treated with complete surgical excision, with final histopathology confirming a benign PA. The case underscores a critical clinical dilemma: delayed intervention for PA inadvertently heightens malignancy risk and complicates management. Consequently, this report strongly advocates for prompt diagnosis and definitive surgical excision upon initial detection of PA to preempt transformation and mitigate the profound prognostic implications of CXPA.

Keywords: Pleomorphic adenoma; Carcinoma ex pleomorphic adenoma; Palatal neoplasms.

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Introduction

Pleomorphic adenoma (PA) is the most prevalent salivary gland neoplasm, accounting for approximately 60% of all benign salivary gland tumors [1]. While the majority arise in the parotid gland, a significant proportion (10-20%) originates from the minor salivary glands, with the palate being the most frequent intraoral site [2]. The tumor demonstrates a distinct epidemiological predilection, occurring more commonly in females, typically presenting in the fourth to sixth decades of life [3]. The pathogenesis is fundamentally driven by cytogenetic alterations, most notably translocations involving the PLAG1 gene or, less commonly, HMGA2, which disrupt normal regulation of cell growth and proliferation, leading to the characteristic biphasic histology of epithelial and myoepithelial cells within a chondromyxoid stroma [4]. Despite its benign classification, PA is notorious for its potential for malignant transformation to carcinoma ex pleomorphic adenoma (CXPA), a formidable malignancy associated with a significantly poorer prognosis. This risk is not trivial; long-standing, neglected PAs, particularly those present for more than a decade, are considered to carry a cumulative risk of transformation estimated 1.5% to 6.3% [5]. The pathogenesis of CXPA is understood as a multi-step process wherein the benign adenoma accumulates additional genetic hits—such as mutations in TP53, PIK3CA, and HRAS—over a prolonged period, culminating in the emergence of a malignant clone within the pre-existing benign tumor [6]. This established risk of carcinogenesis underscores the critical importance of complete surgical excision with clear margins upon initial diagnosis and forms the central clinical dilemma in managing long-standing palatal lesions, where diagnostic delay can inadvertently increase the oncologic risk.

Case Presentation

A 46-year-old female presented to the Department of Oral and Maxillofacial Surgery at Isfahan University of Medical Sciences with a chief complaint of a "slow-growing mass on the roof of the mouth" present for approximately 25 years. The patient reported that the lesion had remained largely asymptomatic and stable in size for the first two decades. However, over the preceding 2-3 years, she had noticed a significant and rapid increase in its volume, causing mild discomfort during mastication and speech, but no pain, paresthesia, or spontaneous hemorrhage. Intraoral examination revealed a well-defined, ovoid swelling on the left hard palate, lateral to the midline. The overlying mucosa was intact and normal in color, with no signs of ulceration or surface vascularity (Figure 1). Palpation confirmed a non-tender, firm-to-rubbery mass with fixed borders. Pre-operative radiographic assessment via cone-beam computed tomography revealed a solitary, well-circumscribed, ovoid soft-tissue mass occupying the left palatal region, extending from the distal aspect of tooth #25 to the maxillary tuberosity. Critical analysis of the bony architecture demonstrated smooth, pressure-induced erosion of the hard palate on the sagittal view and marked thinning of the lateral nasal floor on the coronal view. The lesion measured 20 mm buccolingually and 16 mm mesiodistally. (Figure 2). Given the clinical history of a long-standing lesion with recent rapid growth and radiographic evidence of a well-defined but locally aggressive mass with bony erosion and a potential neurovascular association, the principal differential diagnoses included a benign neoplasm with malignant transformation potential—specifically, a PA at risk for malignant transformation for CXPA—and a low-grade mucoepidermoid carcinoma.

An incisional biopsy was performed under local anesthesia. The pathology laboratory received the specimen in formalin, consisting of an ovoid, well-encapsulated, firm mass measuring 3.1 x 1.1 x 0.9 cm. Microscopic examination demonstrated the quintessential biphasic pattern of ductal epithelium and myoepithelial cells—including those with plasmacytoid morphology—within a myxoid and hyalinized stromal background, confirming the diagnosis of pleomorphic adenoma. (Figure 3). Complete surgical excision was performed under general anesthesia. A circumferential mucosal incision was made with an adequate safety margin beyond the palpable borders of the tumor. A full-thickness mucoperiosteal flap was then elevated, meticulously dissecting the lesion from the underlying bony palate. The dissection was carried out with careful attention to the integrity of the tumor capsule to avoid rupture and minimize the risk of recurrence. The mass was delivered en bloc, and the surgical bed was thoroughly inspected to ensure complete removal. (Figure 4). Hemostasis was achieved, and the wound was primarily closed, ensuring a tension-free closure of the mucoperiosteal flaps. The excised specimen was submitted in its entirety to confirm the diagnosis and verify the adequacy of the surgical margins. Histopathologic assessment revealed no signs of malignancy and the patient was free of any symptoms after a 12-month follow-up. (Figure 1).

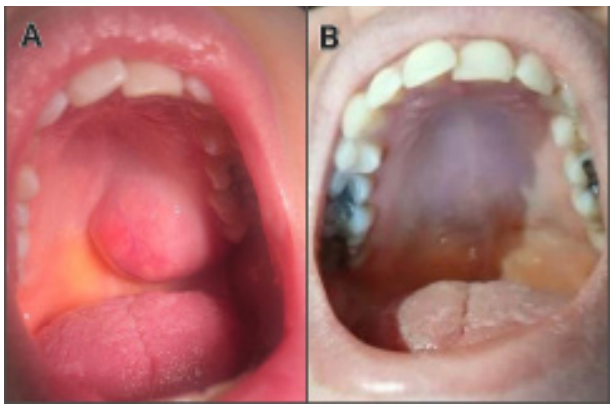

Figure 1. A) Pre-operative Intraoral view, revealing a well-defined, ovoid swelling on the left hard palate. B) Post-operative Intraoral view after 12 months of follow-up.

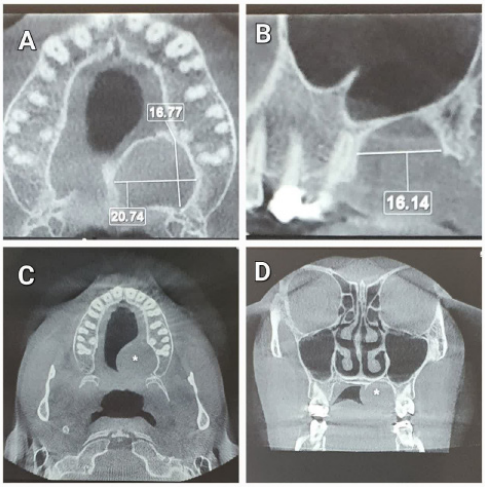

Figure 2. CBCT radiograph in A) Reconstructed axial view showing a lesion measured 20 mm buccolingually and 16 mm mesiodistally, B) Reconstructed sagittal view, C) Axial View revealing a solitary, well-circumscribed, ovoid soft-tissue mass occupying the left palatal region, D) Coronal View.

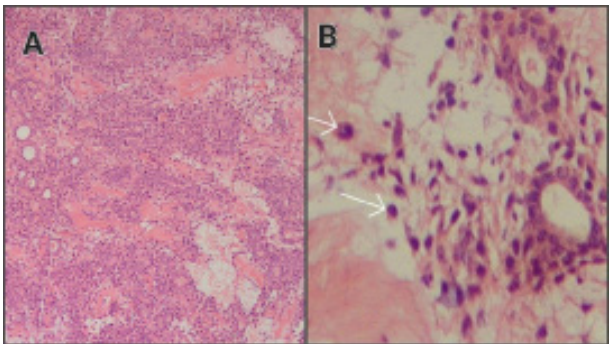

Figure 3. Microscopic view of H&E staining revealing: A) (X40) biphasic pattern of ductal epithelium and myoepithelial cells within a myxoid and hyalinized stromal background, B) (X100) white arrows pointing to myoepithelial cells with plasmacytoid morphology.

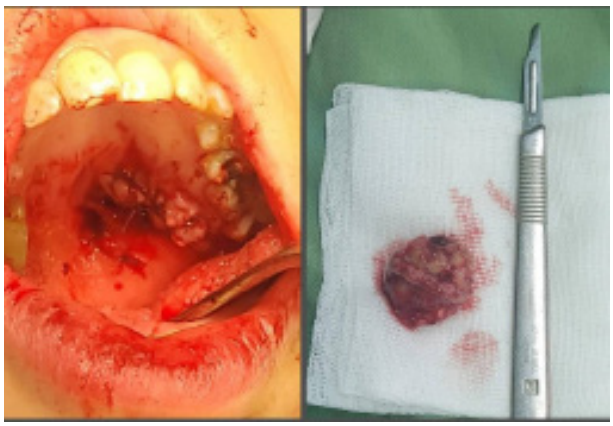

Figure 4. Complete excision of the palatal mass with dissecting the lesion from the underlying bony palate with careful attention to the integrity of the tumor capsule to avoid rupture.

Discussion

The present case of a 46-year-old female with a palatal PA present for approximately 25 years, exhibiting a recent phase of rapid growth, serves as a poignant clinical illustration of the well-documented yet often underappreciated oncologic risks associated with neglected benign salivary gland tumors. While the histopathological diagnosis remained benign, the clinical trajectory and management challenges underscore critical tenets in oral and maxillofacial surgery. The patient's presentation aligns perfectly with the epidemiological profile of minor salivary gland PA, demonstrating a predilection for middle-aged females and the palate as the primary site [3].

However, the 25-year duration of the lesion places it squarely within a high-risk category for malignant transformation. The cumulative risk for CXPA is not linear but escalates significantly with time, with estimates suggesting a risk of 1.5% for the first five years, rising to nearly 9.5% after 15 years [5]. The recent rapid growth reported by the patient is a particularly alarming clinical sign, as a sudden increase in size in a previously stable PA is the most frequently reported symptom heralding malignant change, often due to intralesional hemorrhage or the expansion of a dominant malignant clone [6]. Furthermore, the radiographic findings of bony erosion, though smooth and pressure-induced in this case, blur the line between a benign, slow-growing process and a potentially invasive neoplasm, complicating the preoperative differential diagnosis and heightening surgical urgency. The pathogenesis of CXPA is a paradigm of multi-step carcinogenesis, wherein the initial genetic driver—typically a PLAG1 or HMGA2 translocation that establishes

the benign adenoma—provides a fertile ground for the accumulation of additional molecular insults over time [4]. Recent genomic studies have elucidated that these subsequent "hits" frequently involve mutations in tumor suppressor genes like TP53 and PTEN, as well as activation of oncogenic pathways such as PI3K/AKT and RAS/MAPK, which collectively dismantle cellular senescence and apoptotic mechanisms, unleashing uncontrolled proliferation and invasive potential [7,8]. This case, therefore, embodies a "race against time," where the prolonged dwell time of the adenoma provided a protracted window for such catastrophic genetic events to occur. The surgical imperative for complete, en bloc excision with an intact capsule and clear margins, as performed in this instance, is non-negotiable. Inadequate resection or capsular violation not only increases the risk of local recurrence of the benign component but, more critically, seeds the potential for recurrent tumor to harbor or progress to CXPA, a scenario associated with a markedly worse prognosis [9].

Conclusion

This case powerfully reinforces the doctrine of early and definitive surgical intervention upon diagnosis of PA, regardless of its asymptomatic nature. Managing long-standing lesions transforms a relatively straightforward procedure into a complex oncological resection, emphasizing that in the context of pleomorphic adenoma, procrastination inherently elevates the patient's long-term risk for a devastating malignancy.

Conflict of Interest

There is no conflict of interest to declare.

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