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Case Report

Adenoid Cystic Carcinoma of the Sublingual Gland Mimicking a Benign Tumor: A Case Report and Literature Review

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A B S T R A C T

Background: Adenoid cystic carcinoma (ACC) is an uncommon malignant tumor of the salivary glands characterized by slow progression, perineural invasion, and a high risk of late recurrence and distant metastasis. Sublingual gland involvement is rare and may pose a diagnostic challenge because of its indolent clinical course and nonspecific radiologic features.

Case presentation: We report the case of a 56-year-old patient with no significant medical history who presented with a slowly progressive, painless swelling of the sublingual region evolving over two years. Clinical examination revealed a well-defined sublingual mass without mucosal ulceration or cervical lymphadenopathy. Magnetic resonance imaging demonstrated a well-circumscribed lesion arising from the right sublingual gland, initially suggestive of a benign salivary gland tumor such as pleomorphic adenoma. However, histopathological analysis of an incisional biopsy established the diagnosis of adenoid cystic carcinoma.

Conclusion: This case illustrates the diagnostic challenges associated with sublingual ACC and highlights the importance of early histopathological evaluation. Our findings, supported by a review of the literature, emphasize the need for long-term follow-up given the risk of late recurrence and distant metastasis.

Keywords: Adenoid cystic carcinoma; Sublingual gland; Floor of mouth; Salivary gland neoplasm; Case report

Introduction

Adenoid cystic carcinoma (ACC) is a relatively uncommon malignant tumor of the salivary glands, accounting for approximately 1% of all head and neck malignancies and about 10% of salivary gland tumors. It most frequently arises in minor salivary glands, particularly the palate, whereas involvement of the sublingual gland remains rare[1]. ACC is characterized by slow but infiltrative growth, a marked tendency for perineural invasion, and a high risk of late local recurrence and distant metastasis, especially to the lungs[1,2].

Clinically, ACC typically presents as a slowly enlarging painless mass, which may lead to delayed diagnosis. Radiological findings are often nonspecific, and lesions may appear well circumscribed on imaging, thereby mimicking benign salivary gland tumors such as pleomorphic adenoma[3]. Consequently, histopathological examination remains essential for establishing the definitive diagnosis[4].

Given the rarity of sublingual gland involvement and the potential for misleading clinical and radiological features, reporting additional cases contributes to improved recognition and management of this entity. We report a case of adenoid cystic carcinoma of the sublingual gland presenting as a long-standing painless swelling initially suggestive of a benign salivary gland tumor on imaging.

Case Presentation

A 56-year-old patient with no significant past medical history presented with a progressively enlarging swelling of the floor of the mouth that had evolved over a two-year period. The lesion was painless and not associated with dysphagia, odynophagia, bleeding, paraesthesia, or weight loss.

Intraoral examination revealed a well-circumscribed, firm mass arising from the right sublingual region. The lesion was non-tender and covered by intact mucosa without ulceration **(Figure 1)**. On palpation, it was mobile with no apparent fixation to adjacent structures. No cervical lymphadenopathy was detected, and the remainder of the head and neck examination was unremarkable.

Magnetic resonance imaging (MRI) demonstrated a well-defined lesion arising from the right sublingual gland, appearing hyperintense on T2-weighted images and showing homogeneous enhancement after contrast administration **(Figure 2)**. The lesion was initially suggestive of a benign salivary gland tumour, particularly pleomorphic adenoma. No definite invasion of adjacent structures was identified.

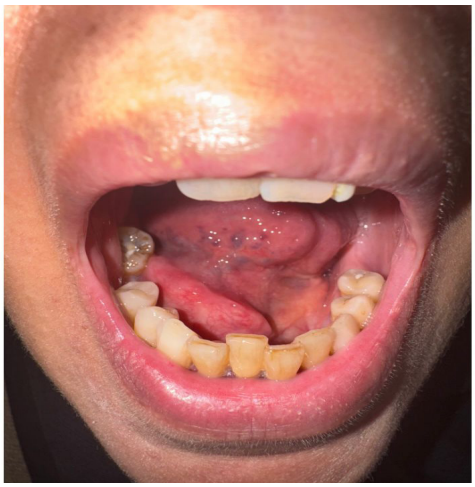

Figure 1: Intraoral clinical photograph showing a well-circumscribed sublingual mass arising from the floor of the mouth, covered by intact mucosa without ulceration.

An incisional biopsy was subsequently performed. Histopathological examination revealed findings consistent with adenoid cystic carcinoma

Discussion

Adenoid cystic carcinoma (ACC) is an uncommon malignant epithelial tumor of the salivary glands characterized by slow but relentless progression, a marked propensity for perineural invasion, and a high risk of late local recurrence and distant metastasis[5-9]. It accounts for approximately 1% of all head and neck malignancies and nearly 10% of salivary gland tumors[5,6]. ACC most frequently arises from minor salivary glands, particularly the palate; however, involvement of the sublingual gland is distinctly uncommon, with relatively few well-documented cases reported in the literature[5,10-12]. Because of its rarity and indolent growth pattern, diagnosis is often delayed.

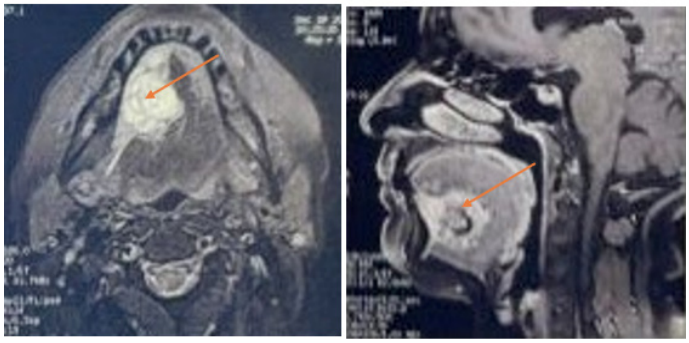

Figure 2: Magnetic resonance imaging of the floor of the mouth (A) Axial T2-weighted image showing a well-defined hyperintense lesion arising from the right sublingual gland (arrow).
(B) Sagittal contrast-enhanced T1-weighted image demonstrating a well-circumscribed enhancing mass without obvious invasion of adjacent structures (arrow).

Clinically, ACC typically presents as a slowly enlarging painless mass, often resulting in prolonged symptom duration prior to consultation[6-8]. Pain or paresthesia, when present, is usually related to perineural invasion rather than tumor size[7,9]. In our case, the lesion evolved over two years without associated symptoms, consistent with prior reports describing a prolonged asymptomatic course in sublingual ACC[10-13]. Several studies have emphasized that tumors of the sublingual gland may remain clinically silent until they reach a considerable size, which contributes to diagnostic delay and may adversely affect prognosis[10,11].

Radiologic evaluation is essential for assessing tumor extent and planning surgical management; however, imaging findings remain nonspecific. On MRI, ACC typically appears iso- to hypointense on T1-weighted sequences and hyperintense on T2-weighted sequences with variable contrast enhancement[14-16]. These imaging features frequently overlap with those of benign salivary gland tumors such as pleomorphic adenoma, representing a well-recognized diagnostic pitfall[14]. Furthermore, although MRI is considered superior for detecting perineural spread, subtle neural invasion may still be underestimated[15,16]. In the present case, imaging findings initially suggested a benign lesion, underscoring the limitations of radiologic assessment alone and highlighting the importance of histopathological confirmation for persistent sublingual masses.

Histopathologically, ACC demonstrates three principal architectural patterns-tubular, cribriform, and solid-with the solid subtype associated with poorer prognosis and higher metastatic potential[5,17]. Perineural invasion is a hallmark feature observed in a large proportion of cases and is strongly associated with local recurrence and worse long-term outcomes[7,9,18]. Prognosis depends on multiple factors including tumor stage,

histologic subtype, margin status, and presence of perineural or vascular invasion[5,7,18]. Some studies have also suggested that tumor location and size at presentation may influence survival outcomes[10,11].

Surgical resection with negative margins remains the cornerstone of treatment for ACC [6–8]. However, achieving adequate margins may be particularly challenging in the floor of the mouth because of proximity to neurovascular structures and functional considerations[10]. Postoperative radiotherapy is commonly recommended in patients with high-risk features, including advanced-stage disease, close or positive margins, perineural invasion, or high-grade histology[6,8,19]. Despite multimodal treatment, ACC is associated with a significant risk of late recurrence and distant metastasis, most commonly involving the lungs[5,9,20]. Long-term follow-up is therefore mandatory, as metastases may occur many years after initial treatment[20,21].

A review of previously reported cases of sublingual ACC reveals several consistent clinicopathologic features. Most patients present in the fifth to sixth decades of life, with no clear sex predominance reported across published series[9-13]. The most common clinical presentation is a slowly enlarging painless sublingual mass, often evolving over several months or years prior to diagnosis[10-12]. Imaging findings are frequently nonspecific, and several reports describe lesions initially suspected to be benign salivary gland tumors[11,12]. Histologically, the cribriform pattern appears most frequently reported, while the solid subtype is associated with poorer outcomes[5,17]. Treatment strategies consistently emphasize complete surgical excision, frequently combined with postoperative radiotherapy in selected cases[6,19]. Despite treatment, long-term outcomes remain guarded because of the risk of late recurrence and distant metastasis[20,21].

Compared with previously reported cases, our patient demonstrated similar clinical and radiologic features, particularly the indolent course and misleading imaging findings, further supporting the diagnostic challenges associated with sublingual ACC.

A summary of previously reported cases of sublingual ACC is presented in **(Table 1)**.

Table 1: Summary of previously reported cases of sublingual adenoid cystic carcinoma.

Author (year)	Age/Sex	Duration of symptoms	Tumor size	Imaging findings	Histologic subtype	Treatment	Outcome
Kokemueller et al. (2004)	52/F	18 months	NR	Well-defined lesion	Cribriform	Surgery + RT	No recurrence
Fordice et al. (1999)	60/M	12 months	NR	Suggestive of salivary tumor	NR	Surgery	NR
van der Wal et al. (2002)	55/F	24 months	NR	Nonspecific	Cribriform	Surgery + RT	Lung metastasis
Rapidis et al. (2005)	58/M	NR	NR	NR	Mixed	Surgery	NR
Ellington et al. (2012)	NR	NR	NR	NR	NR	Multimodal	NR
Present case	56/F	24 months	NR	Suggestive of benign tumor	NR	NR	NR

Conclusion

Sublingual adenoid cystic carcinoma is a rare malignant tumor that may present as a slow-growing, painless mass with misleading radiologic features suggestive of a benign lesion. This case highlights the diagnostic challenges associated with its indolent clinical course and nonspecific imaging findings. Early histopathological evaluation of persistent sublingual masses is essential to avoid delayed diagnosis and ensure appropriate management. Long-term follow-up remains crucial given the risk of late recurrence and distant metastasis.

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