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Dr. Zeine El Abidine Baba El Hassene
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Dr. Mahassine Ines
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Dr. Oussalem Amine
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Dani Bouchra
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Boulaadas Malik
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Corresponding Author:
Dr. Zeine El Abidine Baba El Hassene
Departement of Maxillo-Facial Surgery and Stomotology University Hospital of Ibn Sina, Rabat, Morocco

Adenoid cystic carcinoma of the floor of the mouth: A case report

Zeine El Abidine Baba El Hassene, Mahassine Ines, Oussalem Amine, Dani Bouchra and Boulaadas Malik

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Abstract
Adenoid cystic carcinoma (ACC) is a rare malignant tumor primarily affecting the salivary glands, most commonly arising in the minor salivary glands of the hard palate. Its occurrence in less common sites such as the floor of the mouth is rare, representing a small percentage of intraoral minor salivary gland tumors. ACC is characterized by slow growth but aggressive biological behavior, including a high risk of perineural invasion, local recurrence, and late distant metastases, which complicate long-term prognosis. The mainstay of treatment is radical surgical excision with clear margins, usually followed by postoperative radiotherapy, while chemotherapy shows limited effectiveness.
This is a case presentation of a 55-year-old woman with a painless, slowly enlarging mass on the floor of the mouth, present for two years. Imaging and clinical findings indicated a well-defined lesion confined to the right hemilingual region without nodal involvement. Complete surgical excision was performed, and histopathology confirmed cribriform subtype ACC with negative margins and no perineural invasion. Adjuvant radiotherapy was administered due to the tumor's location and known risk for recurrence. At six months follow-up, the patient remains disease-free. This case underscores the diagnostic challenge of ACC in uncommon intraoral sites and highlights the importance of a multidisciplinary approach, complete resection, and vigilant long-term surveillance.

Keywords: Adenoid cystic carcinoma, floor of the mouth, minor salivary glands, head and neck tumors, case report, radiotherapy

1. Introduction
Adenoid cystic carcinoma (ACC) is a rare malignant tumor, representing approximately 1% of all cancers in the oral and maxillofacial region [1]. It accounts for about 22% of all salivary gland malignancies and is considered the fourth most common malignant neoplasm affecting the minor salivary and seromucinous glands [2, 3]. ACC typically arises in the head and neck region, with nearly 50% of cases occurring intraorally, most frequently in the hard palate [4, 5]. Other, less common intraoral sites include the lower lip, retromolar area, sublingual gland, buccal mucosa, and the floor of the mouth [4]. The tumor is known for its slow-growing but aggressive nature. Despite its often benign histologic appearance, ACC demonstrates a high tendency for perineural invasion, local recurrence, and distant metastasis [6]. Cervical lymph node metastases are relatively uncommon, occurring in approximately 8-13% of cases, while distant metastases—particularly to the lungs and bones—are reported in up to 50% of patients[4]. Notably, ACCs originating from minor salivary glands generally have a poorer prognosis compared to those arising from the major salivary glands [4].

2. Case Presentation
We report the case of a 55-year-old female patient with no significant past medical history, who presented with a slowly enlarging mass of the oral floor, first noticed approximately two years prior to consultation. The lesion had progressively increased in size without associated symptoms such as pain, dysphagia, odynophagia, paresthesia, weight loss or cervical swelling.
On clinical examination, a firm, painless, bilobed mass was palpated on the floor of the mouth, primarily on the right side but extending beyond the midline toward the left. The mass measured approximately 4 cm in its greatest dimension, was mobile relative to the deeper planes, and extended posteriorly to the region adjacent to the mandibular right third molar (tooth 48), and anteriorly to the lingual surface of the lower incisors. The overlying mucosa was intact, with no ulceration or discoloration.

Tongue mobility was preserved, and there was no evidence of regional lymphadenopathy. Sensory examination of the lingual and inferior alveolar nerve territories was normal. (Fig.1)

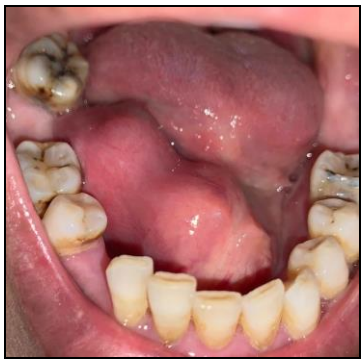

Fig 1: Preoperative examination showing a bilobed mass on the floor of the mouth

Magnetic resonance imaging (MRI) revealed a well-defined lesion centered on the floor of the right hemilingual region, with no extension to the contralateral side or invasion of adjacent bony or muscular structures. (Fig. 2, 3)

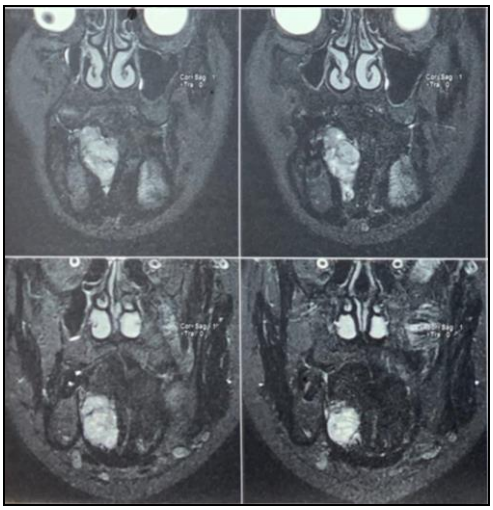

Fig 2: Coronal MRI showing a well-defined, heterogeneous mass of the floor of the mouth, extending towards the submandibular space, consistent with adenoid cystic carcinoma.

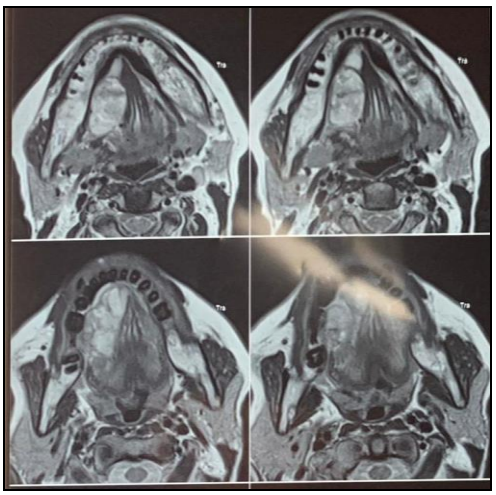

Fig 3: Axial MRI demonstrating the tumor's extent in the floor of the mouth with close relation to adjacent muscles and submandibular glands, without obvious bony invasion.

The patient underwent complete surgical excision of the mass via an intraoral approach under general anesthesia. Intraoperatively, the tumor was well circumscribed and dissected with preservation of surrounding anatomical structures. No lymph node dissection was performed due to the absence of clinical and radiological evidence of nodal involvement. (Fig.4)

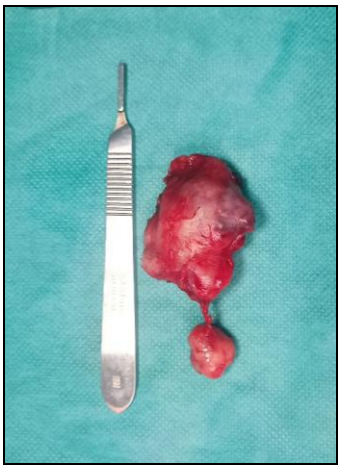

Fig 4: Excised lesion

Histopathological examination confirmed the diagnosis of adenoid cystic carcinoma of the cribriform subtype with clear surgical margins. Given the high risk of local recurrence and the known perineural tropism of ACC, the case was discussed in a multidisciplinary tumor board. Despite the absence of histological perineural invasion, adjuvant radiotherapy was recommended and administered, owing to the lesion's anatomical location and the tumor's known propensity for delayed local recurrence.

The postoperative course was uneventful, and the patient recovered without complications. She remains under regular clinical and radiological follow-up, with no signs of recurrence at the six-month mark.

3. Discussion

Adenoid cystic carcinoma is an uncommon malignancy of the salivary glands, with a marked predilection for the minor salivary glands of the hard palate. In contrast, its occurrence in less typical locations such as the floor of the mouth, tongue, lower lip, or retromolar region is significantly less frequent, collectively representing only about 5% of intraoral minor salivary gland tumors. In a cohort of 40 histologically confirmed ACC cases, merely 7 were located in the floor of the mouth, underscoring the rarity of this presentation [7]. In our case, the tumor's location in the floor of the mouth aligns with these findings and highlights the diagnostic challenge posed by such unusual anatomical presentations.

ACC tends to affect women more often and is generally diagnosed in the fifth to sixth decades of life, with cases occurring in individuals under 20 years of age being exceptional [8]. Clinically, the tumor often appears as a firm, slowly enlarging, unilobular mass. A hallmark feature is pain, which can precede visible swelling and may present as a persistent, dull ache that gradually worsens over time [9]. This early pain, sometimes attributed to perineural invasion, may be one of the earliest diagnostic clues.

Although diagnosis confirmation of ACC is obtained through analysis of cytomorphologic features in fine-needle

aspiration, imaging remains a pillar of preoperative diagnosis. CT is the first choice examination for evaluating bone invasion, while MRI remains the gold standard for identifying soft-tissue tumors and studying their nature, borders and infiltration [10].

Despite its indolent clinical progression, ACC is biologically aggressive and notorious for late metastases and local recurrences. It has been described as one of the most destructive and unpredictable tumors of the head and neck region [11, 12]. Long-term follow-up data demonstrate the silent yet fatal nature of this malignancy: one study among 160 patients reported a 5-year disease-specific survival rate of 89%, dropping drastically to 40% at 15 years due to delayed distant metastases [13, 14]. This dichotomy, between apparent clinical dormancy and eventual lethal dissemination, necessitates vigilant, long-term surveillance even after curative treatment.

Therapeutically, ACC is approached as a high-grade malignancy. The consensus in the literature supports radical surgical excision with the aim of achieving clear margins, followed almost universally by postoperative radiotherapy [15].

However, the role of elective neck dissection (END) in patients without clinically evident lymph node involvement (N0) remains contentious. ASCO guidelines suggest considering END in cases with high T-stage tumors or high-grade histology, but data from the REFCOR cohort showed no significant survival advantage from this approach in cN0 patients [16, 17]. Moreover, while one meta-analysis encompassing nearly 3000 patients suggested a higher rate of cervical nodal metastases in minor salivary gland ACCs compared to their major counterparts, the actual rate of occult metastasis may still be too low to justify routine END [18, 19]. Thus, a more tailored, case-by-case evaluation remains prudent.

Post-operative radiotherapy (PORT) is widely advocated in ACC due to the tumor's tendency for microscopic perineural and local extension. Historical perspectives, including Osborn's early observations in 1977, emphasized combining surgery with radiation [20]. This combination remains the cornerstone of management for non-metastatic disease [21].

Complete resection with negative margins significantly improves local control, while radiotherapy augments disease-free survival, especially in cases with adverse features [22].

Mendenhall *et al*. later confirmed the routine use of PORT, even in cases with clear margins, unless the tumor was small (T1), low-grade, and completely excised [23]. Additionally, PORT is especially indicated for tumors that are T3-T4, involve bone or nerves, or show positive or close margins [24].

The utility of chemotherapy remains limited in ACC due to the tumor's slow growth, which hampers the measurable efficacy of both cytotoxic and targeted agents [25]. Concurrent chemoradiotherapy has not demonstrated added benefit and is not currently recommended [26]. However, in advanced stages where metastases are present and other palliative measures are exhausted, some patients may benefit from palliative chemotherapy, as reported by Laurie *et al*. [27].

In light of our patient's tumor location in the floor of the mouth—a rare and diagnostically challenging site—the above findings reinforce the importance of a high index of suspicion in atypical salivary gland tumors. The histopathological confirmation of ACC justified the radical surgical approach adopted, and the decision to follow up with adjuvant radiotherapy aligns with established best practices. Given the known risk of delayed metastasis, long-term follow-up will be essential in the ongoing care plan.

4. Conclusion

Adenoid cystic carcinoma is a rare malignant tumor of the minor salivary glands, with a known tendency for slow but aggressive progression. Its occurrence in the floor of the mouth is extremely uncommon, which may lead to diagnostic difficulties and delayed management. Despite its indolent clinical appearance, ACC has a high risk of perineural invasion, local recurrence, and late distant metastasis, making long-term follow-up essential. Surgery with clear margins remains the treatment of choice, usually followed by radiotherapy to improve local control.

Conflict of Interest

Not available

Financial Support

Not available

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