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Case Report

Adenocarcinoma of the sublingual salivary gland – A case report

Sahana Adirajaiah[a,*], Venkatesh Anehosur[b], Sumana[c], K. Gopalakrishnan[b]

ABSTRACT

Background: This report and review of literature aimed to assess a case of adenocarcinoma of sublingual salivary gland.

Method: We present a case of a 52-year-old female with adenocarcinoma of sublingual salivary gland displaying painful swelling in the floor of the mouth, which was affecting her speech and mastication.

Conclusion: Surgery is the treatment of choice for malignant sublingual gland tumors, but the type of surgical intervention depends on the extent of the primary tumor itself. Even though the tumors of the sublingual salivary gland are rare, they are a diagnostic challenge to every head and neck surgeon.

Keywords: *Adenocarcinoma, Sublingual gland*

INTRODUCTION

Minor salivary gland malignancies are uncommon, accounting between 10% and 15% of all salivary tumors.[1] 70–80% of salivary gland tumors originate in the parotid gland; 7–11% in the submandibular gland; less than 1% in the sublingual gland. Generally, the smaller the salivary gland involved, the higher the possibility of the tumor being malignant. Malignant tumors comprise of 15–32% of parotid tumors, 41–45% of submandibular gland tumors, and 70–90% of sublingual gland tumors. Most tumors of the sublingual gland are malignant, with adenoid cystic carcinoma and mucoepidermoid carcinoma being the most frequent. Many other malignant tumor types, such as acinic cell carcinoma, malignant mixed tumor, squamous cell carcinoma and clear cell carcinoma, have also been reported.[2]

Sublingual salivary gland tumors occur most frequently in patients during the sixth decade of life, without gender predilection. Clinically, these tumors present as an asymptomatic mass located in the floor of the mouth, which can be misdiagnosed as intraoral minor salivary gland tumors or as other benign and malignant lesions located in this region. Correlation of the clinical, surgical, radiographic, and microscopic findings is important for the correct diagnosis. The prognosis depends mainly on the histologic type of the tumor and adequate primary surgical treatment.[3]

It should also be considered against a differential diagnosis that includes ranula, mandibular tori, dermoid cyst, squamous cell carcinoma, and other salivary gland tumors. Compared with tumors of the parotid gland, a greater proportion of submandibular and sublingual salivary gland tumors are malignant.[4]

The aim of the present case report is to report a rare case of adenocarcinoma with histopathological variant not otherwise specified, showing aggressive features like perineural spread which responded to surgery and radiotherapy.

CASE REPORT

A 52-year-old female patient visited our department with the complaint of painful swelling in the floor of the mouth,

[a]Assistant Professor, [b]Professor, [c]Oral Surgeon, Department of Oral and Maxillofacial Surgery, SDM College of Dental Sciences and Hospital, Sattur, Dharwad, Karnataka 580009, India.
[*]Corresponding author. *email: drsahana11@gmail.com*
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affecting her speech and mastication. It was present for last 8 months and was increasing in size. Tongue movements were not affected and there was no paresthesia or history of xerostomia (Fig. 1a). The past medical history was non-contributory. Clinical examination showed a solitary firm swelling in the floor of the mouth measuring 3 × 4 cm, extending anteriorly from the midline, posteriorly till distal aspect of 2nd molar region and superiorly above the occlusal level. The borders of the swelling were well defined and overlying surface was smooth, with missing 2nd premolar and 1st molar. On palpation the mass was non-tender, firm in consistency and not fixed to the underlying structures. Examination of right submandibular duct revealed normal salivation. Based on the history and clinical examination, a provisional diagnosis of ranula was made. Differential diagnosis included sialolithiasis, benign and malignant tumor of sublingual gland. There was no radiological evidence of calculus, or associated lymphadenopathy. Orthopantomogram showed no bone resorption near the lesion. USG showed well-defined homogeneously isochoric nodular lesion in the right sublingual region measuring 2.5 × 2.4 cm with no cystic change or calcification. There were few mildly enlarged submandibular lymph nodes measuring less than 1.5 cm. T1-weighted MRI revealed a well-defined, rounded, solid, heterogeneously enhancing soft tissue mass seen arising from right sublingual gland. The margins of mass were causing compression over adjacent structures in sublingual space and muscles of floor of mouth. There was no evidence of invasion into adjacent soft tissues and bone (Fig. 1b).

Surgical excision of the mass was performed under general anesthesia. Incision was placed on the lesion just lateral to the Wharton's duct cutting through the mucosa. Blunt dissection was carried out around the capsule of the tumor. The Wharton's duct and lingual nerve were identified and peeled off from the tumor. Entire tumor was completely excised without spillage (Fig. 2a). After achieving complete hemostasis the wound closure was done with vicryl. The specimen measured about 4 × 3 × 3 cm, with creamish brown, irregular surface. Cut surface was creamish white in color, and firm in consistency. Histopathologic sections showed, hyalinized to fibromyxoid stroma with numerous clusters, chords and cribriform pattern of tumor. Areas of isolated tumor cells were also evident. Cells forming clusters showed monomorphic, vesicular nuclei and clear cytoplasm cells. The cells seemed to show neurotropism and perivascular arrangement. Few ductal spaces were lined by cuboidal cells and seen as part of cribriform pattern and in isolation. Lobules of mixed salivary gland elements were also observed (Fig. 2b). The tumor was histologically graded as moderated differentiated (Table 1). Since there was absence of features specific to any of the salivary gland malignancies, a final diagnosis of adenocarcinoma was made. Postoperatively patient was subjected to radiotherapy with a total dose of 65 Gy with daily fractions of 2 Gy over a period of 1 month. 1 year follow-up revealed no evidence of recurrence.

DISCUSSION

When a malignant salivary gland tumor is discovered in the oral floor region, it is important to determine whether it originated in the sublingual gland or in a minor salivary

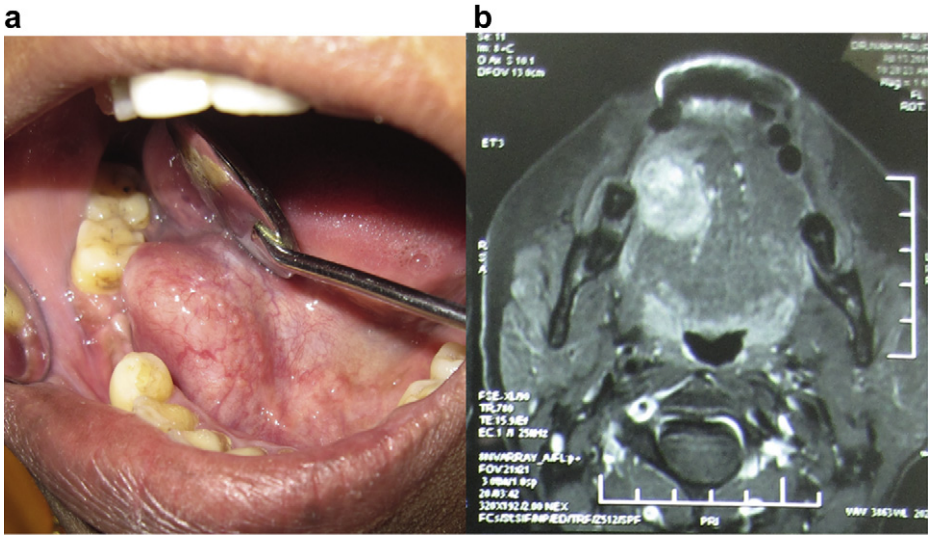

Fig. 1 a: Tumor extension. b: MRI scan of the tumor.

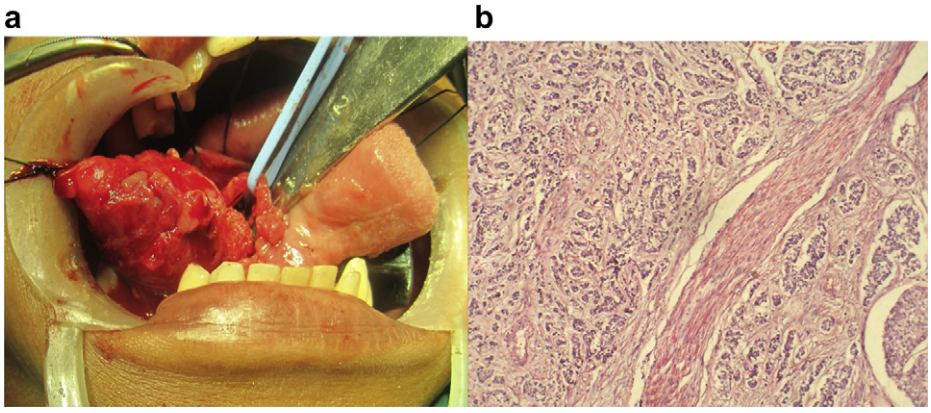

Fig. 2 a: Tumor excision. b: Tumor histopathology.

gland. It is difficult to identify the tumor's origin based solely on its anatomic location. Recently, however, advances in diagnostic imaging technology have enabled better identification of the origin of tumors arising in the oral floor. In this case, diagnostic imaging was an effective tool in deciding the tumor's origin. It was diagnosed as having originated in the sublingual gland based on Omura's criteria[4]: 1) clinically, the tumor was in the sublingual gland position, and 2) histologically, the tumor replaced the entire sublingual gland and showed minimal invasion into the surrounding oral mucosa. Also, a small amount of residual sublingual gland tissue was found surrounding the tumor. Tumors of the sublingual salivary gland occur infrequently, constituting less than 1% of all salivary gland tumors.[3,4] It has been found, however, that 70% of the tumors located in this area are malignant.[5] Sublingual gland malignancies are estimated to represent 0.5–4.7%[6,7] of all major salivary gland neoplasm. Adenocarcinoma; NOS, accounts for about 12% of all sublingual gland malignancies[5] and for 17% of the malignant tumors involving major and minor salivary glands.[8] Additional series confirm that adenocarcinoma NOS is the third or fourth commonest malignant salivary gland tumor.[9,10]

The malignant tumors originating in the sublingual gland show varying degrees of malignancy and present difficult problems in terms of diagnosis, management, and treatment. Approximately 50% of all malignant salivary gland tumors recur after initial treatment. This fact emphasizes the need for improved initial evaluation and more effective primary surgery. The histologic grade of adenocarcinoma, NOS, correlates with the rate of recurrence, the amount of cervical and distant metastases, and ultimately, the rate of survival. The cure rates for grades I, II, and III tumors at 5 years are 69%, 46%, and 8%, respectively, and at 15 years, the rates are 54%, 31%, and 3%, respectively. Surgery remains the primary therapeutic modality for adenocarcinoma, NOS, and both the stage and grade of the tumor should play a role in determining the final decision regarding treatment. However, because of the rarity of sublingual gland tumors, specific suggestions for treatment have not been developed.

Surgery is the treatment of choice for malignant sublingual gland tumors, but the type of surgical intervention depends on the extent of the primary tumor itself.[6] Even though the tumors of the sublingual salivary gland are rare, they are a diagnostic challenge to every head and

Table 1 Salivary adenocarcinomas, NOS: histopathologic grading criteria.

	Well-differentiated	*Moderately differentiated*	*Poorly differentiated*
1. Quality and quantity of glandular formation	>50%	20–50%	<20%
2. Degree of nuclear atypia	Minimal, 1+	Minimal to moderate, 1–2+	Moderate to severe, 2–4+
3. Mitotic activity	3/10 hpf	3/10 hpf	10/hpf
4. Presence of necrosis	Absent	Minimal (no more than occasional single-cell)	Single-cell common (cavitary or comedonecrosis may be present)

From: Matsuba et al [1988].

neck surgeon. They easily get confused with tumors of the floor of the mouth and submandibular salivary gland tumors. The tumors of sublingual salivary gland tumor are generally recognized in advanced stage, mainly because of minimal symptomology.[6]

This case report of adenocarcinoma of sublingual gland is an addition to the cases reported earlier in the literature. The key in the management includes early diagnosis with advanced imaging techniques, surgical excision of the involved gland followed by radiotherapy.

CONFLICTS OF INTEREST

All authors have none to declare.

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