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CASE REPORT

A Rare Case of Salivary Duct Carcinoma of the Hypopharynx

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ABSTRACT

Salivary duct carcinoma (SDC) is a rare invasive malignancy arising from the ductal epithelium of the major salivary glands, especially the parotid gland. These are an uncommon but distinct group of highly malignant salivary gland tumors. This malignancy is well known for its aggressive behavior, metastasis to distant sites and high mortality rate. Few cases of SDC arising from minor salivary glands have been reported in literature. Till now no case has been reported to be occurring in the minor salivary glands of the hypopharynx. In this article, we present a case of salivary duct carcinoma of a 44-year-old male patient occurring in the minor salivary glands of the hypopharynx. The relevant literature and its treatment aspects are discussed.

Keywords: Salivary duct carcinoma (SDC), Minor salivary glands, AE fold.

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INTRODUCTION

Salivary duct carcinoma (SDC) is a rare invasive malignancy arising in the ductal epithelium of salivary glands.[1] Salivary ductal carcinoma occurs predominantly in males, with a male to female ratio of 2:1. The age of these patients range from 20 to 80 years, with more cases seen during the fifth and sixth decades of life. These are usually seen in the major salivary glands, mainly in the parotid gland.[2] Salivary duct carcinoma affecting the minor salivary glands has been reported in only 4% of the SDC cases and constitutes 2% of all the salivary gland malignant neoplasms.[3] Intraorally, the common sites of occurrence are the minor salivary glands of the palate, followed by the buccal mucosa/vestibule, the upper lip, the maxilla and the mandible.[4] Occurrence of this lesion below the level of the oral cavity and oropharynx has not been reported in the literature. We are reporting here a very rare occurrence of SDC arising from minor salivary glands of the hypopharynx.

CASE REPORT

A 66-year-old man presented with dysphagia for solid foods since 2 months and dysphonia since 1 month duration. Patient was not having any complaints of dyspnoea or throat pain. Patient was a smoker since 25 years but had left smoking since the appearance of the symptoms. There was no history of any medical comorbidities or illnesses.

On indirect laryngoscopic examination, there was an exophytic irregular pedunculated tumor of about 3×3 cm arising from lateral side of left aryepiglottic (AE) fold blocking the airway partially on respiration. Bilateral vocal cords were mobile. Rest of the head and neck examination was normal and no significant cervical lymphadenopathy was detected. Computed tomography (CT) scan of the neck showed homogenously enhancing mass arising from lateral wall of left AE fold filling rest of the lumen of the pyriform sinus (Fig. 1). No cervical lymphadenopathy was detected. His routine blood investigations, chest and cardiac evaluation were within normal limits.

Because of the exophytic pedunculated nature, patient was planned for microlaryngeal examination and excision biopsy of the lesion with prior tracheostomy for intubation. The pedunculated lesion with its peduncle attached to the left AE fold was confirmed. Medial wall of the AE fold was normal without any extension (Fig. 2). Microlaryngeal excision of the mass was done. Arytenoid cartilage was not involved and it was not disturbed. Tracheostomy tube was retained for 3 days and decannulated later. Patient was on

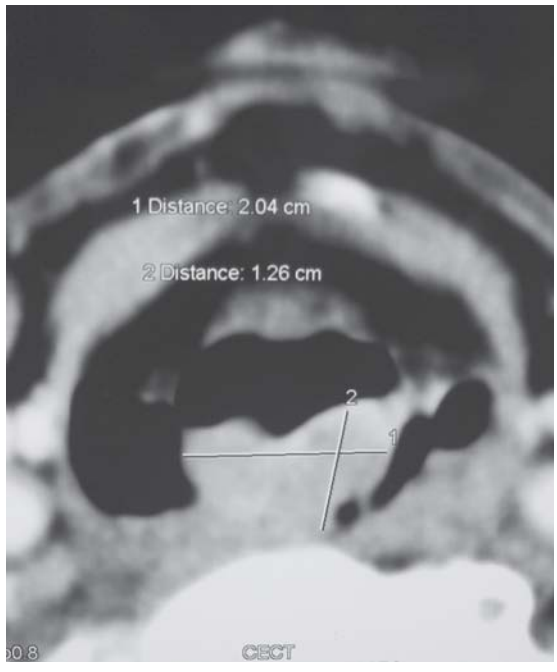

Fig. 1: CT scan of the larynx showing the tumor

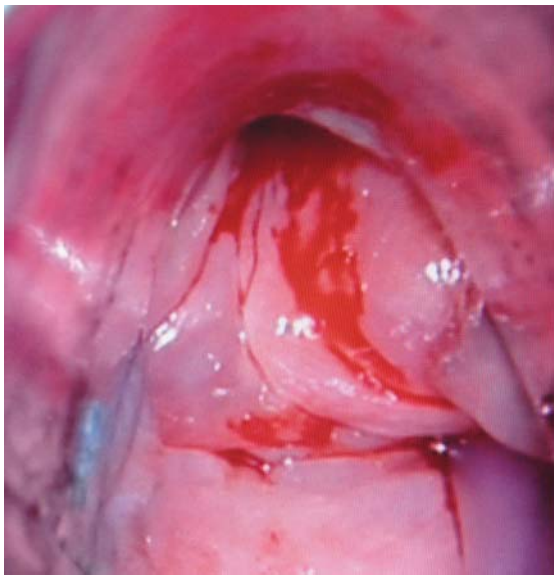

Fig. 2: Intraoperative finding of the pedunculated tumor

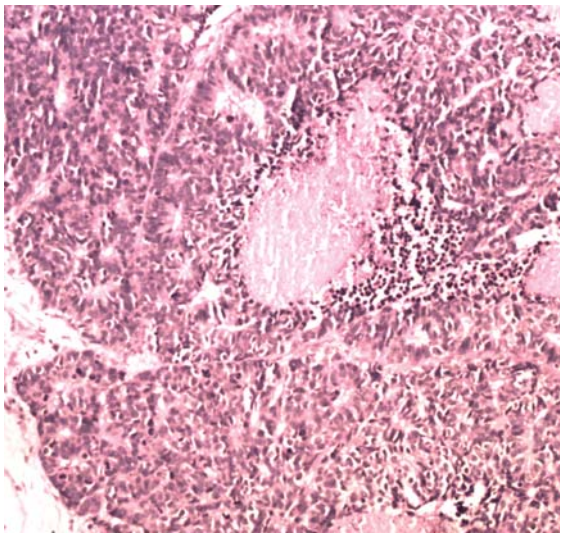

Fig. 3: Histopathological section 10× showing characteristic comedonecrosis

nasogastric tube feeding initially, which was removed after 4 days and swallowing was normal without any aspiration.

At the time of discharge, patient's speech, swallowing and indirect laryngoscopic findings were normal. Histopathologic examination revealed an ulcerative neoplasm composed of malignant epithelial cells with vesicular nucleus and prominent nucleoli exhibiting ductal component with comedonecrosis (Fig. 3). Solid areas and cribriform growth pattern were also seen. Diagnosis of salivary duct carcinoma of the minor salivary glands was made. Patient was advised for postoperative radiotherapy in view of compromised excision from AE fold. He had received 46 Gy in 23 # of external beam radiotherapy to the primary as well as neck. His radiotherapy period went uneventful. After 6 months follow-up examination, there was no evidence of recurrent disease with a good functional larynx. He was advised to be on regular follow-up in view of the aggressive histopathology.

DISCUSSION

Salivary duct carcinoma (SDC) was first described by Kleinsasser et al in I968, who reported a histopathologic similarity to ductal carcinoma of breast.[5] This malignancy was established as a distinct clinicopathological entity and was delineated from 'adenocarcinoma (NOS)' in 1991 by the World Health Organization. Other terms used for SDC are cribriform salivary carcinoma of excretory ducts and infiltrating salivary duct carcinoma.[4,6] Salivary duct carcinoma is an uncommon malignant tumor which occurs predominantly in men and most often in the parotid gland. This neoplasm is aggressive and occurs most often in the middle-aged and older males. Lymph node metastasis is common. Though neoplasms of this type are infrequent in minor salivary glands, they are less aggressive than in the major salivary glands and may lead to early diagnosis before distant metastases could occur.[7]

SDC exhibits a wide range of histological appearances.[8] It consists of atypical cuboidal or polygonal cells arranged in papillary cribriform and solid growth patterns alone with duct like structures.[1] Tumor cells are polygonal in shape with granular eosinophilic cytoplasm. Enlarged hyperchromatic pleomorphic nuclei and prominent nucleoli are seen.[1,9] Pseudocyst formation and central comedonecrosis are seen in the neoplastic islands.[10] Vascular invasion anal perineural infiltration has been reported in some cases.[1,11] Atypical mitotic figures are seen in most of the lesions.[12] Dystrophic calcifications are seen in some cases.

Histologically, it is characterized by a striking resemblance to ductal carcinoma of the breast. The terms, 'cribriform salivary carcinoma of the excretory duct' and 'infiltrating salivary duct carcinoma' have been recommended for SDC, to distinguish it from other salivary carcinomas, many of which may also be 'ductal' in origin. However, the designation, 'salivary duct carcinoma' has gained acceptance. The differential diagnoses for SDC are squamous carcinoma, oncocytic carcinoma and adenocarcinoma (not otherwise specified).[13] SDC can be distinguished from the above lesions by characteristic histopathologic features like comedonecrosis, eosinophilic cytoplasm of atypical cells, cellular pleomorphism and infiltrating cribriform pattern.[9,12]

The presenting symptoms depend upon the site of origin. In major salivary glands, it usually presents as a painful or painless, firm mass of rapid growth. Facial nerve palsy may be seen in advanced cases. Lesions of the minor salivary glands may present as a slow growing intraoral mass or with dysphagia. As it is an aggressive tumor, a thorough search for occult lymph node enlargement should be made with imaging whenever there are no palpable lymph nodes. Incidence of lymph node metastasis is less for minor salivary

gland origin when compared to major salivary glands. Lymph node metastases have been reported in 22% of the SDC cases in the minor salivary glands, as compared to 83% in the SDC of the major salivary glands.[3]

Treatment involves radical surgical excision of the tumor and associated structures with concomitant neck dissection, followed by postoperative radiation therapy.[9,11,12] In our case, patient presented with exophytic pedunculated mobile tumor from the lateral surface of left aryepiglottic fold. Rest of the larynx and hypopharynx were within normal limits. Neck examination revealed no significant lymphadenopathy confirmed by CT imaging. Because of the above presentation, patient was planned for transoral excision followed by postoperative radiotherapy with the intention of organ preservation and better quality of life. The mortality rate for this tumor is also low, although reported local recurrence rates are high. A recurrence rate of 33 to 35% has been reported from a study on SDC patients.[4]

SDC is considered to be a high-grade malignancy capable of distant metastases with a poorer outcome in major salivary glands.[6,14] Metastasis of SDC to lungs, bones and liver has been reported with a high mortality rate.[15] In contrast SDC of minor salivary gland origin is relatively less aggressive and carries a better prognosis.[5,16] Information on the efficacy of adjuvant therapy is limited due to the infrequency of this tumor. Hormonal therapy using an anti-androgen like goserelin has been tried.[2]

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