Salivary duct carcinoma of the minor salivary glands: A rare case report

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ABSTRACT

Salivary duct carcinoma (SDC) is a rare invasive malignancy arising in the ductal epithelium of the salivary glands. These are an uncommon but distinct group of highly malignant salivary gland tumours. These are usually seen in the major salivary glands, especially in the parotid gland. This malignancy was established as a distinct clinicopathological entity and was delineated from 'adenocarcinoma (NOS)" in 1991 by the World Health Organization. Salivary duct carcinomas affecting the minor salivary glands have been reported in only 4% of the SDC cases and constitute 2% of all the salivary gland malignant neoplasms. Its biological behaviour is highly aggressive; the metastatic and the tumour-related death rates were 75% and 73%, respectively. In this article, wereport a case of a salivary gland carcinoma which was present in the palate of a 55-year-old male patient.

Key Words : Salivary Duct Carcinoma, Minor salivary glands

CASE REPORT

A 55-year-old male patient was referred with a complaint of growth in the left upper posterior region of the maxilla for the past 4 months. He was apparently healthy and no abnormalities were detected on extra oral examination. On intra-oral examination, a non tender, dome shaped, ulcerated swelling was seen extending from the upper left second premolar on the buccal side to the first molar region. The lesion was 3 x 3 cm in size, erythematous and with irregular margins. On the lingual side, a hard mass was present since 30 years. [Table/Fig-1]

On palpation, the lesion was found to be tender and soft to hard in consistency. No lymph nodes were detected. The orthopantamogram revealed a wide area of bone loss in the upper left molar region. [Table/Fig-2] A provisional diagnosis of peripheral giant cell granuloma and squamous cell carcinoma/ mucoepidermoid carcinoma was given, based on the clinical details. An incisional biopsy was performed under local anaesthesia and the specimen was sent for histopathological evaluation. The histopathological features of the lesion revealed numerous infiltrating islands and cords of neoplastic glandular epithelium in a fibrovascular connective tissue. Most of the neoplastic islands exhibited central "comedonecrosis [Table/Fig-3].

The neoplastic cells were cuboidal to polygonal in shape and exhibited cellular pleomorphism, eosinophilic cytoplasm, nuclear hyperchromtism and prominent nucleoli [Table/Fig-4]. Numerous blood vessels and mitoses were present. The lesion was covered by a parakeratinized, stratified squamous surface epithelium. Based on the histopathological features, a diagnosis of salivary duct carcinoma was made.

DISCUSSION

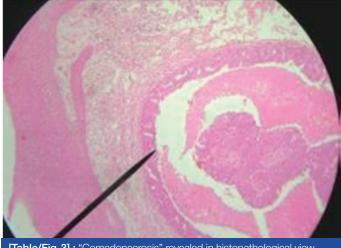
SDC was first described by Kleinsasser in 1968 [1] and was further defined by several authors. It was only recently recognized as a distinct clinic-pathological entity. The origin of the neoplasm from the salivary ducts was initially suggested by its morphological resemblance to mammary duct carcinoma and



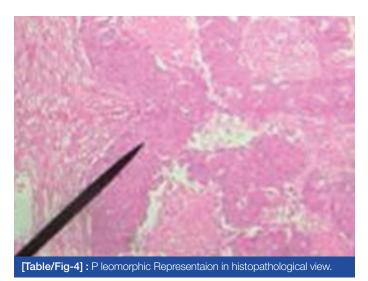
[Table/Fig-1]: Clinical view of the mass observed on the Lingual side



[Table/Fig-2] : Radiographic representation of the Lesion.



[Table/Fig-3] : "Comedonecrosis" revealed in histopathological view.



it was later supported by the identification of preinvasive disease, ductal dysplasia and carcinoma in situ in the intralobular and the extralobular ducts.[2] [3]

Salivary ductal carcinoma occurs predominantly in males, with a male to female ratio of 2:1. The ages of these patients ranged from 23-80 years, with more cases being reported in the fifth and sixth decades of life. These are usually seen in the major salivary glands, especially in the parotid gland. [4] 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. Intra-orally, the common sites of occurrence are the palate, followed by the buccal mucosa / vestibule, the upper lip, the maxilla and the mandible. [5]

Salivary duct carcinoma, which was recently recognized as a high grade, aggressive malignancy of the major salivary glands, is characterized histologically by a striking resemblance to ductal carcinoma of the breast. The terms, 'cribriform salivary carcinoma of the excretory duct' and 'infiltrating salivary duct carcinoind' 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 because it is used in the WHO classification of tumours." The separation of SDC from the category, "adenocarcinoma NOS (not otherwise specified)", is warranted by its predictably aggressive behaviour. [4] This tumour may arise within a pleomorphic adenoma as a result of the malignant transformation of the ductal epithelial cells. In addition, a multifocal origin from the major excretory ducts surrounding a pleomorphic adenoma was observed in one of the cases. SDC exhibits a wide range of histological appearances. [3] The histopathological features of SDC consist of atypical cuboidal or polygonal cells which are arranged in papillary cribriform and solid growth patterns along with duct like structures. The tumour cells are polygonal in shape, with granular eosinophilic cytoplasm, enlarged hyperchormatic, pleomorphic nuclei and prominent nucleoli. Pseudocyst formation and central comedonecrosis are seen in the neoplastic islands. ('Comedonecrosis' means a type of necrosis occurring in the glands, in which there is central luminal inflammation with devitalized cells, which usually occurs in the breast in intraductal carcinoma). Vascular invasion and perineural infiltration have been reported in some cases. Atypical mitotic figures are seen in most of the lesions. Dystrophic calcifications are seen in some cases.

The differential diagnosis of SDC [6], [7], spans a range of low-grade and high-grade salivary gland malignant neoplasms, the most common ones of which include papillary cystadenocarcinoma, papillary cystic acinic cell carcinoma, metastatic adenocarcinoma, etc.

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. However, multiple metastases and a high mortality rate of 60-75% are associated with the minor salivary gland SDCs. A recurrence rate of 33-35% has been reported from a study on SDC patients.[8],[9] [10].

The treatment of this lesion involves the radical surgical excision of the lesional tissue and its associated structures, with concomitant neck dissection, followed by post-operative radiation therapy.

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