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Case Report

Salivary Duct Carcinoma of the Minor Salivary Gland: 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. Nearly 85% of the cases occur in the parotid gland followed by submandibular gland. Rarely is it described in the hard palate. 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. It is characterized by aggressive behavior with early metastasis, local recurrence and significant mortality. The tumor has predilection for older men in the 6th to 7th decades of life. In this article; we report a case of a salivary gland carcinoma which was present in the right posterior region of the maxilla of a 50 year old female patient.

INTRODUCTION

Carcinomas of the salivary glands are rare tumors, which account for less than 1% of all head and neck malignancies. A group of malignant salivary gland tumors characterized by ductal formations and central necrosis were first described by Kleinsasser et al. in 1968 [1]. Pathomorphologically, these tumors showed great similarities to ductal carcinomas of the female breast, which is why they described this tumor as "salivary duct carcinoma (SDC)" in their original article. SDC represents a rare tumor with an estimated incidence of 1–3% of all malignant salivary gland tumors [2, 3]. They are characterized by indolent growth and long term survival. SDC is thought to be a distinct malignancy of the major salivary glands, because of its highly aggressive behavior, high rate of recurrence, nodal and distant metastases, and death related to the tumor [4].The clinical aspects and the histopathological patterns of SDCs are well described. The tumor is more frequent in elderly men, occurring predominantly in the parotid gland and occasionally in the submandibular gland. A number of patients experience facial nerve palsy or paralysis and/or pain, and have cervical lymphadenopathy at presentation.

Histologically, it shows a striking resemblance to breast carcinoma of the ductal type, presenting intraductal and invasive components [5].

CASE REPORT

We present a case of 50 year old female patient presented with a painless swelling in the upper right posterior region of the maxilla since five years, with a history of a rapid increase in the size of the swelling since three months (Figure 1). On intra oral examination, there was a swelling which was non tender, dome shaped, ulcerated extending from canine to second molar region. The swelling was approximately 5x3 cm in size, which was hard in consistency on palpation. The overlying mucosa was of the normal color as the adjacent mucosa (Figure 2). There was no evidence of either facial nerve involvement or regional lymphadenopathy. Clinically, there was no evidence to suggest breast carcinoma. The patient was subjected to CT scan to evaluate the total

extension of the tumor and the bone involvement. The scan revealed loss of bone with the maxillary sinus involvement. The tumor extended beyond the boundary of bone and had lead to the loss of fat also. The lesion extended till the skin without breaching it (Figure 3). The provisional diagnosis of a malignant tumor including squamous cell carcinoma and mucoepidermoid carcinoma was given, based on the clinical details. The patient underwent biopsy under local anesthesia and then the tissue was sent for histopathological diagnosis.

Figure 1. Photograph showing female patient with swelling on the right side of the face.

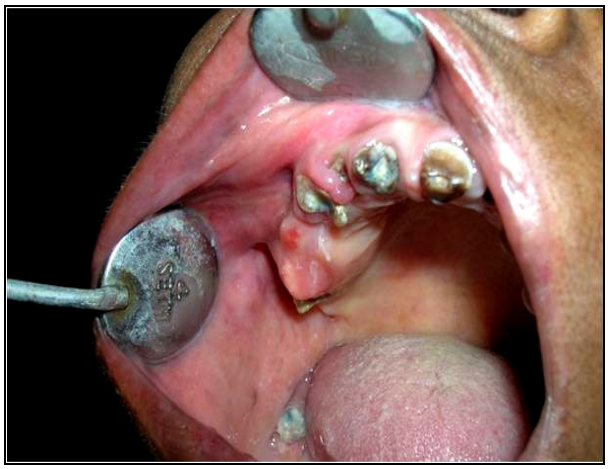

Figure 2. Photograph showing intraoral swelling in the right maxillary posterior region.

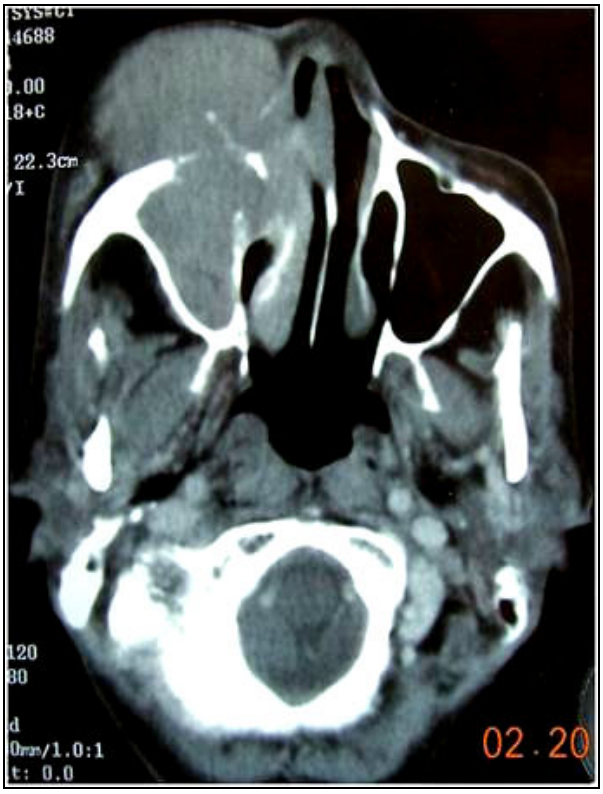

Figure 3. CT scan showing extension of the lesion with bone loss.

Histopathologically, 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". The neoplastic cells were cuboidal epithelial cells forming solid nests, cribriform and comedone patterns (Figure 4 and 5). The cells exhibited cellular pleomorphism, eosinophilic cytoplasm, nuclear hyperchromasia and prominent nucleoli. The papillary epithelial projections into duct-like structures, and densely sclerotic hyalinized stroma were also evident. The tumor was compressing the adjacent normal-looking salivary acinar component. The diagnosis of salivary duct carcinoma was made based on these histopathological features.

After confirming the diagnosis patient underwent surgery in general anesthesia and is still about 6 months follow up.

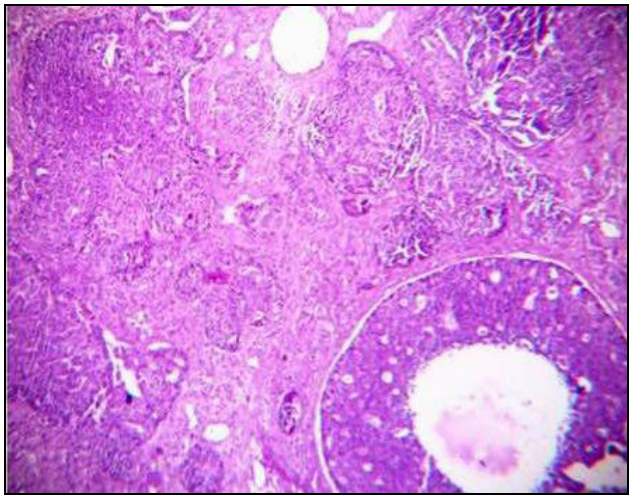

Figure 4. Histopathologically, the lesion reveals ductal pattern with comedonecrosis and solid islands of neoplastic cells (H&E, x200).

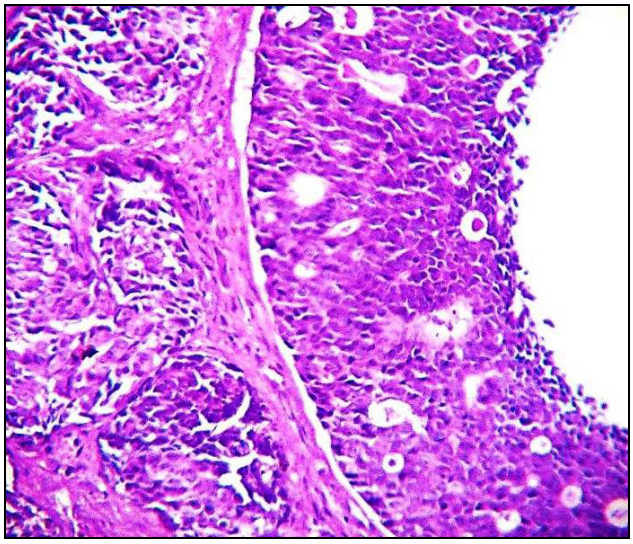

Figure 5. At high magnification, the lesion reveals ductal pattern with sharply define cell margins, pleomorphic nuclei and nucleoli (H&E, x400).

DISCUSSION

Salivary duct carcinoma is very rare neoplasm of the salivary glands and was first described by Kleinsasser and coworkers in 1968. They are seen in the parotid in some 80% cases while only 5% are known to occur in intra-oral minor salivary glands. SDC are most frequently seen in the 6th and 7th decades of life with a 3.8:1, male preponderance [1].

SDC is believed to arise from the excretory ducts or as a result of malignant transformation of ductal cells in pleomorphic adenoma. These patients usually present with a swelling which is occasionally painful and may have facial nerve involvement. At times these tumors are clinically misjudged as an adenocarcinoma, adenoid cystic carcinoma, basal cells carcinoma, bone tumors like osteosarcoma and sebaceous cell carcinoma. Grossly, these tumors are small, mottled yellow white with minutely cystic spaces and measure 0.7-4 cm in greatest diameter [6]. Histologically, these tumors are composed of ducts filled and distended with cells having micro papillary, tufted, cribriform and solid pattern with central comedonecrosis. Cells are large and have powdery, eosinophilic cytoplasm with a sharply defined cell margins, pleomorphic nuclei and prominent nucleoli. Roman bridging and cellular atypia are observed. These tumors have a proliferating component, which may be poorly differentiated [7]. Literature search revealed that cribriform growth pattern and comedonecrosis are more frequently encountered in high-grade than in low-grade SDC. Progression is evidenced by the acquisition of higher cytologic grades such as monomorphous cellular composition, well defined cytoplasmic borders and rigid cellular arrangement, Infiltration is associated with hyalinized stroma. Vascular, perineural invasion and osseous metaplasia are also seen sometimes. Histologically, these tumors can be confused with metastatic deposits from breast carcinoma, sweat duct carcinoma and prostatic carcinoma [8]. To make a definitive diagnosis of SDC, Garland *et al*, require circumscribed epithelial nest with central comedonecrosis. Interestingly, in most cases of carcinosarcoma, a rare biphasic tumor consisting of carcinomatous and sarcomatous features the malignant epithelial component is that of SDC [9].

Ultrastructurally, these tumors show intracytoplasmic lumina with microvilli. In addition, irregular cocoon shaped collection of tubular or membrane material, desmosomes, tight junction and basal lamina are seen [4].

The outcome of SDC is unfavorable. Most of the cases reported in literature have an aggressive nature, but a few reports with low grade potential have also been documented. SDC is considered to be a high-grade carcinoma with dismal short term prognosis death eventually occurring within the first 3 years after diagnosis [10]. In contrast, others believe that there exist different sub types, one of which is characteristically aggressive, whereas the other less invasive purely intraductal, perhaps representing pre-invasive phase of high-grade SDC. There is local regional spread as well as distant metastasis to brain, liver, adrenals lung, bone, skin and thyroid. Lymph node metastasis is seen in 60% cases. Recurrence is found in 33.3%. These tumors are usually treated with extended excision of the primary site along with removal of the regional lymph node followed by

radiation therapy [11].

In conclusion, although these terminologies for closely related neoplasm in terms of prognosis may generate academic interest, it is difficult to predict any difference in their biologic behavior. It is important that more cases of this neoplasm should be reported to understand any significant biologic differences that would help to establish a definite conclusion about their nature. It is widely believed that both SDC and SDC in carcinosarcoma are rare high-grade carcinomas capable of distant metastases with a poorer outcome in major salivary glands. In contrast SDC of minor salivary gland origin are relatively less aggressive and has a better prognosis.

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