Locally advanced salivary duct carcinoma of the parotid gland

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ABSTRACT

Due to the low incidence of salivary duct carcinoma (SDC), there is limited data in regard to the biologic behavior of this tumor, histopathological characteristics and its management. There is diversity in the management of parotid SDC. Various authors manage it with radical parotidectomy with or without neck dissection; others add adjuvant radiotherapy with radical surgery. The objective of the study is to see the biological behavior and management of the three patients with locally advanced SDC and review with the literature.

Key Words: Adjuvant, carcinoma, ductal, radiotherapy, salivary gland neoplasms

INTRODUCTION

Salivary duct carcinoma (SDC) is a rare malignant epithelial tumor and is characterized histologically by multiple neoplastic epithelial nests similar to the ductal carcinoma of the breast. Although first described by Kleinsassar *et al*. in 1968, SDC was not formally recognized in the World Health Organization classification of salivary gland tumors until 1991.[1,2] Salivary duct carcinoma is a high-grade malignant tumor exhibiting aggressive growth with early regional and distant metastasis. SDC mainly affects the older male patients.[1,2] This study reports a case series of three patients of SDC of the parotid gland including one with a sarcomatoid variant, managed by multimodality therapy.

CASE REPORTS

Case 1

A 35-year old female had presented to the department with a painless lump over the left preauricular region since last 7 months. She also complained of trismus and difficulty in closing the left eyelid. Physical examination showed a 6 × 5cm hard mass in the left preauricular region fixed to the underlying tissues and overlying skin. There were palpable upper cervical lymph nodes. Computed Tomography revealed a large necrotic mass involving the left parotid, infiltrating the masseter muscle and the overlying skin [Figure 1]. Fine needle aspiration cytology was suggestive of carcinoma. A total left radical parotidectomy with enbloc excision of the overlying skin, left modified neck dissection, and

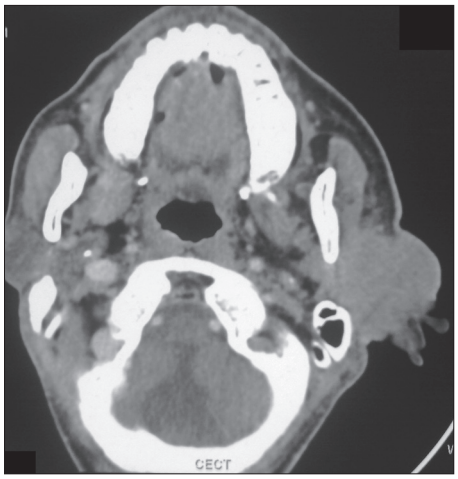

Figure 1: Represents the computed tomography of a patient with salivary duct carcinoma which shows a mass arising from the left parotid gland infiltrating the deep lobe and the masseter muscle

reconstruction of the defect with pectoralis major myocutaneous flap (PMMC) were carried out under general anaesthesia. The intra-operative finding was a hard mass involving both the superficial and deep lobe of parotid gland and infiltrating the masseter muscle and the mandibular condyle periosteum. There were multiple cervical lymph nodes at all levels of the left side of neck. Histopathology was suggestive of salivary duct carcinoma involving the parotid gland. Two out of seven nodes dissected were positive histopathologically for SDC. Patient received 60 grays of external beam radiotherapy [EBRT] in thirty fractions. The patient is on regular follow-up since last 8 years and is disease free with no evidence of recurrence or distant metastasis.

Case 2

A 50-year old male had presented to the department with left facial nerve palsy and a lump over the preauricular region since 8 months. Computed Tomography revealed a large mass involving the superficial and deep lobe of left parotid gland. A total left radical parotidectomy with enbloc excision of the overlying skin, left modified neck dissection, and reconstruction of the defect with PMMC were carried out under general anaesthesia. The intra-operative finding was a hard mass of size 5 × 4 cm involving both the lobes of parotid gland and infiltrating the overlying skin. Histopathology was suggestive of SDC with metastases to 12 of the thirteen nodes dissected [Figure 2]. Immunohistochemistry was positive for cytokeratin and Her2 neu. Patient received 60 grays of adjuvant EBRT to the loco-regional site. He is on regular follow-up and is disease free since last 6 months.

Case 3

A 42-year old female had presented to the department with a lump over the right preauricular region since 3 months. Physical examination showed a 5 × 4 cm hard lump in the right preauricular region. She had no cervical lymphadenopathy nor any clinical signs of facial nerve palsy. Computed Tomography revealed a lump involving the superficial lobe of right parotid gland with no adjacent structures being involved. She underwent facial nerve preserving radical parotidectomy [Figure 3] with supraomohyoid neck dissection. Histopathology was suggestive of SDC with no nodal metastases. Histopathologically, there was a striking resemblance to ductal carcinoma of the breast with central comedo necrosis and some areas of sarcomatoid morphology. Immunohistochemistry was positive for cytokeratin and negative for SMA, desmin and myogenin in both the areas [Figure 4]. Patient received 60 grays of EBRT and is on regular follow-up. She had no signs of recurrence since seven months of surgery.

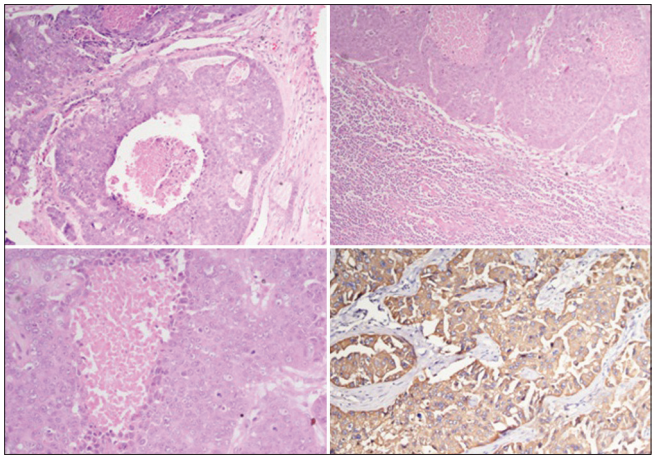

Figure 2: Represents the histopathology suggestive of salivary duct carcinoma

DISCUSSION

Salivary duct carcinoma represents a rare and aggressive tumor with an estimated incidence of 1-3% of all malignant salivary gland tumors.[2,3] SDC commonly arises from the parotid gland and clinically presents as a rapidly growing mass infiltrating the facial nerve. Most treated patients with SDC present with early loco-regional recurrence with high propensity for distant metastases to the lung, bone and liver.[2] Seventy percent of the patients with SDC die of their disease within 3 years of diagnosis due to widespread metastases.[2]

Histopathologically SDC bears a striking similarity to ductal carcinoma of the breast and is composed of intra ductal and invasive components. Similar to the breast carcinoma, the intra ductal component may appear as a cribriform, papillary, or solid growth pattern, often with comedo like central necrosis. The invasive carcinoma consists of irregular glands and cords of cells that frequently elicit a prominent desmoplastic reaction. Salivary duct carcinoma can occur de novo or as the malignant component of carcinoma ex pleomorphic adenoma.[4] Most pathologists stress on the presence of flat sheets of tumor cells with cribriform patterns as characteristic feature of SDC.[4]

Delgado *et al*. based on the degree of intra ductal or infiltrative component have classified SDC into 3 subtypes: 1) Predominantly intra ductal, where 90% of the tumor is intra ductal 2)

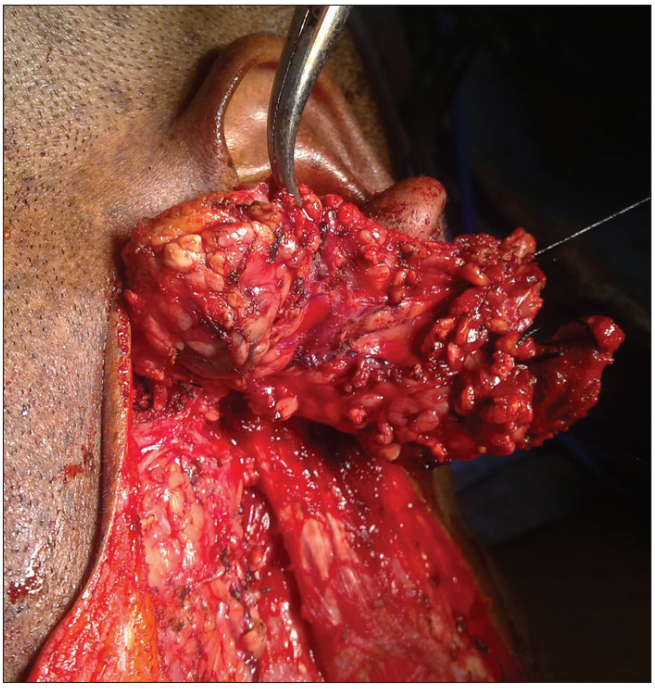

Figure 3: Represents the intraoperative finding showing the mass arising from the superficial lobe of parotid gland

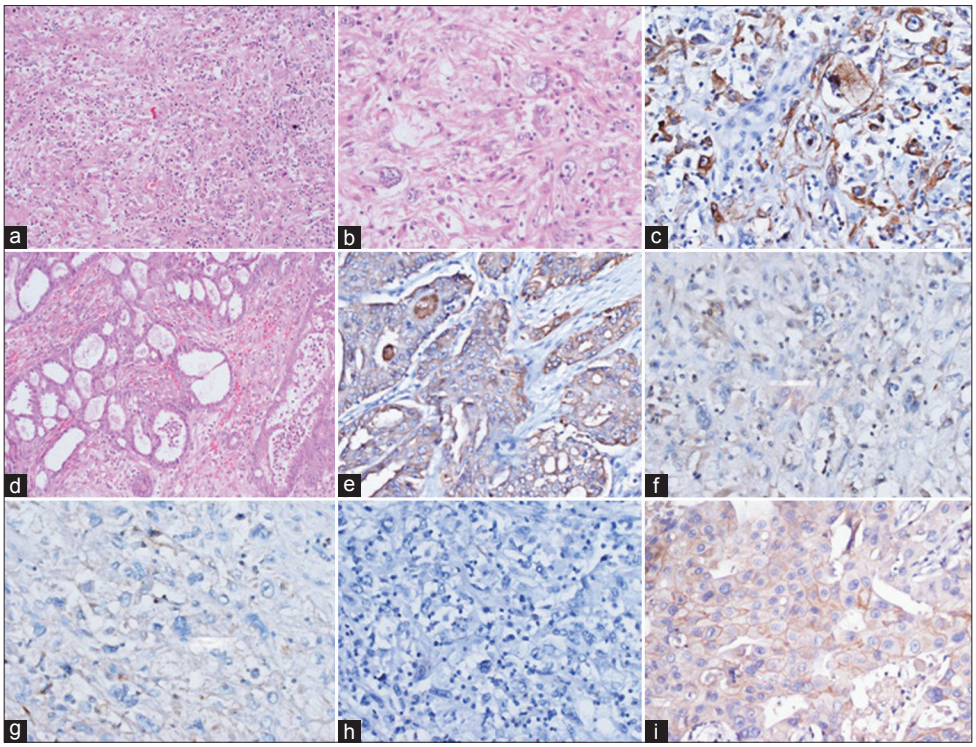

Figure 4: a] Tumor cells with spindle shape arranged in illdefined fascicles and solid sheet(x100), b] Tumor nuclei showing marked pleomorphism in sarcomatoid area, c] Cytokeratin positive in sarcomatoid area, d] Apocrine snouts in tumor cells towards luminal side, e] Cytokeratin positive in the comedo area, f] Desmin negative, g] Myogenin negative, h] Smooth muscle actin negative, i] Tumor cells showing Her 2 neu positivity

Table 1: Salivary duct carcinoma of the parotid gland reported in the English-language literature

Author, year of publication	Age/gender	Stage	Treatment	Locoregional recurrence	Systemic metastasis	Follow-up
Butterworth *et al.*[8] (1992)	59/M*	T2N0M0	Parotidectomy–ND–RT‡	No	No	NED§ 3 months
Minamiguchi *et al.*[9] (1996)	60/M*	T2N0M0	Parotidectomy–ND–RT‡	No	Yes	DOD¦ 9 months
	66/M*	T2N0M0	Parotidectomy–ND–RT‡	No	No	NED§ 27 months
	47/F†	T3N1M0	Parotidectomy–ND–RT‡	No	Yes	Alive with disease on 7 months of follow-up
Kruslin *et al.*[10] (1996)	47/F†	T3N1M1	Parotidectomy–ND–RT‡	No	Yes	DOD¦ 5 months
Ikeda *et al.*[11] (1997)	65/M*	T2N1M1	Parotidectomy–ND–RT‡	No	No	NED§ 25 months
Madrigal *et al.*[12] (1999)	78/M*	T2N0M0	Parotidectomy–ND–RT‡	No	No	NED§ 11 months
Bhalla *et al.*[13] (2006)	53/M*	T3N1M0	Parotidectomy–ND–mastoidectomy—RT‡	Yes	Yes	Alive with disease on 10 months of follow-up
Alsharif *et al.*[7] (2008)	22/M*	T1N0M0	Parotidectomy–ND–RT–nerve transplantation‡	No	No	NED§ 10 years
Current series	35/F†	T4N2M0	Parotidectomy–ND–RT‡	No	No	NED§ 96 months
	50/M*	T4N2M0	Parotidectomy–ND–RT‡	No	No	NED§ 6 months
	40/F†	T3N0M0	Parotidectomy–ND–RT‡	No	No	NED§ 7 months

*M - Male, †F - Female, ‡ND - Neck dissection, RT - Radiotherapy, §NED - No evidence of disease, ¦DOD - Died of disease

Predominantly infiltrative, when less than 20% of the tumor is intra ductal or 3) Infiltrative, when the tumor is entirely infiltrative.[5] The significance of this classification is not known but it has been seen that the predominantly infiltrative tumor has dismal prognosis.[5]

Several variants of SDC such as sarcomatoid variant, low grade or mucin rich SDC have been described.[5-7] Literature reviews on the sarcomatous variant of SDC have been reported in only sixteen patients till date. Sarcomatoid SDC is characterized histologically by a biphasic neoplasm with carcinomatous and sarcomatoid elements. Nagao *et al*. in his review of patients with sarcomatous SDC, reported 25% patients dying of disease within 2 years of completion of treatment.[6] He concluded that sarcomatoid SDC is a highly aggressive tumor, similar to conventional SDC.

Due to the rarity of this malignancy, no consistent therapeutic concept and protocol exists for this tumor entity. Majority treat patients with SDC with a radical surgical approach and adjuvant external radiotherapy [Table 1][7-13] whereas others recommend adjuvant therapy based on the pathological stage, grade, margin and perineural invasion.[7] Recurrence was seen in all the patients reported in literature with parotid SDC who have undergone parotidectomy and did not undergo lymph nodal neck dissection.[7] In the present series, all the three patients have been treated with a radical approach that includes radical parotidectomy, neck dissection and adjuvant radiotherapy. In this case series, one of the three locally advanced SDC patients has survived disease free even 96 months after completion of treatment. There by we stress the fact that oncological cure in patients with SDC can still be achieved with the radical surgical approach and adjuvant radiotherapy.

CONCLUSION

Salivary duct carcinoma is characterized by aggressive growth, a high incidence of early lymph node metastasis, and frequent local recurrences after surgical excision. Combined modality that includes radical surgery with neck dissection and adjuvant radiotherapy may result in the oncological cure of the disease.

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