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CASE STUDY **OPEN ACCESS**

Ductal Cell Carcinoma of Salivary Gland (A Rare Entity) - Report of Two Cases

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ABSTRACT

Ductal Cell Carcinoma Of Salivary Gland or SDC is a very rare, most aggressive natured tumor having very poor prognosis with an evidence of local and regional metastasis and increased mortality rate resembling breast ductal carcinoma. This article deals with two cases of Ductal Cell Carcinoma of Salivary Gland (SDC) arising from duct of major (Parotid) and minor salivary gland. Aggressive resection with selective neck dissection and post operative radiotherapy were reserved as treatment for both cases. The role of neck dissection with adjuvant postoperative radiotherapy aid in disease prognosis owing to a tumour aggressive nature.

Keywords: *Ductal Cell Carcinoma, Salivary Duct Carcinoma, Aggressive tumour , Neck Dissection*

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INTRODUCTION

Kleinsasser et al in 1968 describedDuctal Cell Carcinoma or SDC comprising about 1 to 3 % of malignant salivary gland tumors [1]**.** Later in 1991, SDC was included in the second version of WHO classification of salivary gland tumors [2]. SDC is an aggressive adenocarcinoma which resembles a high-grade mammary duct carcinoma, an epithelial tumor having salivary gland sites. Clinically SDC has a local recurrence with a significantly high mortality rate [3]. Histologically 27% of SDC arose from pre-existing pleomorphic adenoma of the parotid gland (carcinoma ex-pleomorphic adenoma) with perineural invasion and 73% cervical lymph node involvement [4,5]. Three histopathological variants of SDC seen in literature are invasive micro-papillary , low grade and sarcomatoid tumour [6].

CASE PRSENTATION

CASE 1

A 52-year-old female patient reported to our institute, with a complaint of painful swelling on the right side of the face for past 6 months. The swelling measured 8 x 7 cm in diameter with throbbing type of pain. The swelling was hard in consistency, fixed, warm and tender with an ulcerated surface. Facial Nerve functions were intact. Neck examination was positive for multiple lymph node involvement. Contrast-enhanced CT scan revealed large moderately enhancing soft tissue density lesion arising from the right parotid gland with regional lymph node involvement. Owing to big sized swelling incisional biopsy was not feasible hence FNAC opted. FNAC reported was evident of ductal cell carcinoma of the parotid gland.

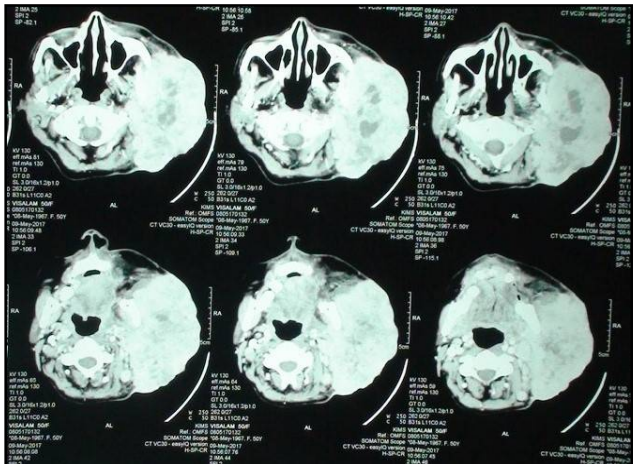

Fig 1 Contrast enhanced CT scan shows evidence of large heterogeneously enhanced lesion with multiple hypodense area noted with epicentre with left deep and superficial lobe of patrotid gland.

CASE II

A 40-year-old male patient reported to our institution with a complaint of mobile tooth in left upper back too the region for past two months. History of presenting illness reveals patient noticed the mobility two months before followed by swelling and loss of sensation in the left side of face. Patient extra oral examination revealed facial asymmetry with round shaped swelling on left side of the face measuring 6.5cm X 5.4cm X 4.3cm palpable left submandibular lymph nodes. Patient intraoral examination revealed painful ulcer over the distal surface of 27 with grade III mobility of the respective tooth. Incisional biopsy report confirmed the diagnosis of ductal cell carcinoma of minor salivary gland. Contrast enhanced CT scan revealed erosion of the left anterior inferolateral wall of left maxilla involving lateral pterygoid plate and left orbital floor with the positive involvement of level B nodes.

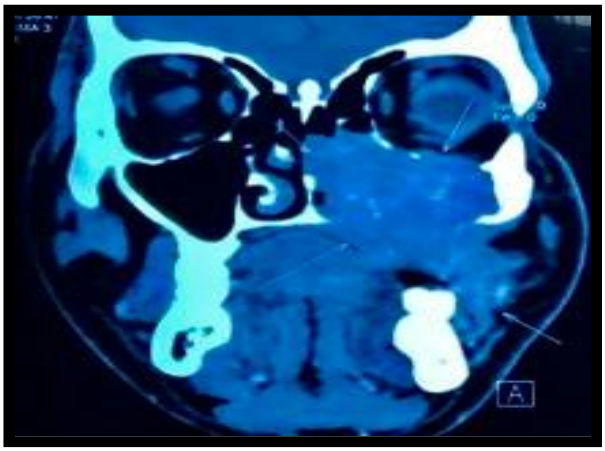

Fig 2 Contrast enhanced CT scan revealed erosion of left anterior infolateral wall of left maxilla involving lateral pterygoid plate and left orbital floor

SURGICAL TECHNIQUE

Treatment comprised of wide local excision (WLE) of a tumour with total parotidectomy including facial nerve for the case 1 and left partial maxillectomy for case II and selective neck dissection (SND I-III) of left side and right side respectively. Reconstruction of defect made with SSG (split skin graft) harvested from the left thigh region for the case I and impression compound with an acrylic plate for case II. Postoperative HPE confirmed the preoperative diagnosis. A month later postoperatively both patients were sent for adjuvant radiotherapy.

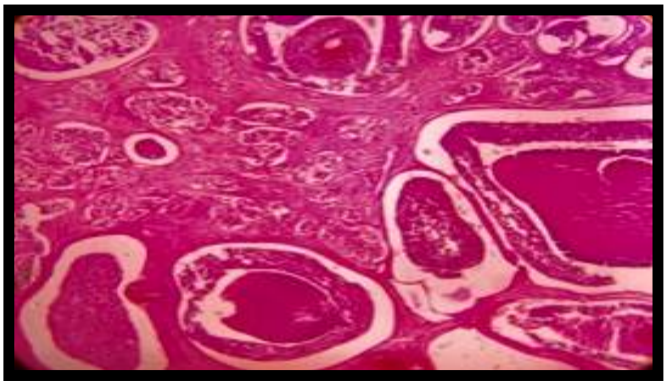

Fig 3 Soild island of Tumor shows Comedo pattern of with central necrosis

IMMUNOHISTOCHEMISTRY

Owing to its rare nature immunohistochemistry proved to be a valuable tool for establishing a definitive diagnosis. Both cases were positive for Her2/neu(c- erbB-2) marker and also for androgen receptor(AR). Examining SDC for this marker have shown expression rates upto 100%. [14]

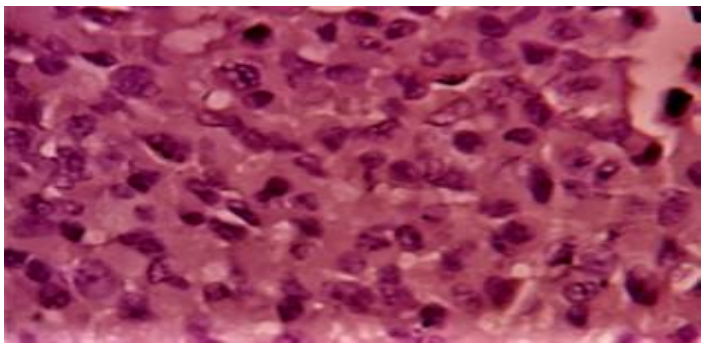

Fig 4 Tumor cell shows immunopositivity for Her2/eu x40 and perineuralinavsion

DISCUSSION

Ductal Cell Carcinoma of Salivary gland is a rare aggressive malignant tumour of salivary glands. SDC of parotid region accounts for 0.2-10% seen in the fifth or sixth decade of life, with a male to female ratio of 2:1 [3]. Clinical data and clinical studies are scanty owing to rare nature of a tumour and certain studies reported more than 50 SDC patients. In English language literature, SDC had been reported that 37 cases originated from minor salivary glands and mostly seen in hard palate (14 cases) [4]. Facial paralysis had been misdiagnosed as Bell's palsy in parotid gland SDC. SDC has a poor prognosis with a dismal rate of 26-66% local recurrence, 52.9-60 % neck metastasis and 50-66% of distant metastasis to lung, bone, liver, back and leg [7-10] The poor prognostic factors in SDC were age, tumour size, and lymph node recurrence rate have noted, if neck dissection has not addressed with radical parotidectomy. In advanced cases of SDC. Paclitaxel is reserved as the chemotherapeutic drug of choice as reported by Kuroda et al [5-8]. The positivity of diagnosis is depended on histopathologically, which is not reliable with CT and USG [9]. In the primitive lesion, SDC calcification is noted radiographically. The differential diagnosis of SDC is Mucoepidermal carcinoma and Mammary adenocarcinoma metastasis [3]. Shreds of evidence suggest that Trastuzumab and anti-androgen therapy may be efficacious in managing either primary and/or metastatic disease [11,12]. Studies have shown abnormalities in the phosphoinositide 3-kinase pathway in some SDCs, creating another parallel with breast carcinoma and additional targets for therapy [13]. Outcomes usually poor among patients with ductal cell carcinoma of salivary gland; local recurrence, regional metastasis, and distant metastasis are frequent, and many patients die of the disease, although exceptions do rarely occur [14].

Giving these poor outcomes our both cases underwent aggressive resection of a tumour along with neck dissection and for postoperative radiotherapy.

CONCLUSION

The prognosis is very poor due to the aggressive behavior and metastatic potential. The survival rate of SDC is only 50 % within 4 to 5 years. Due to large area of necrosis, the diagnosis may be missed on FNAC, CT, and USG. Early diagnosis is mandatory, as SDC is asymptomatic in onset and neck dissection is

mandatory in regional lymph node involvement. Adjuvant chemotherapy and radiotherapy will help in the oncological cure of the disease. Reconstruction should be considered for the defect where aesthetics and functional activities play a major role.

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