ISSN: 0976-3031

Available Online at http://www.recentscientific.com

CODEN: IJRSFP (USA)

International Journal of Recent Scientific Research
Vol. 9, Issue, 1(D), pp. 23157-23159, January, 2018

International Journal of Recent Scientific Research

DOI: 10.24327/IJRSR

Case Report

SALIVARY DUCT CARCINOMA OF PAROTID GLAND IS A RARE ENTITY: CASE REPORT

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DOI: http://dx.doi.org/10.24327/ijrsr.2018.0901.1408

ARTICLE INFO

Article History:

Received 17th October, 2017
Received in revised form 21st November, 2017
Accepted 05th December, 2017
Published online 28th January, 2018

Key Words:

Salivary Gland, Ductal Carcinoma, Parotid Gland, Neck Dissection

ABSTRACT

A 52-year-old female patient presented to our hospital with complaint of painful swelling in the left side of the face for past 6 months. Patient's history reveals the swelling was initially small and attained the present size of 8 x 7 cm, associated with a throbbing type of pain. The swelling was hard in consistency, fixed, warm and tender with ulceration with bleeding spots on the surface, with no other secondary changes. The mouth opening was within acceptable limits with no facial nerve involvement. On general examination, the patient was poorly built and nourished with no recent weight loss. Lymph node examination revealed multiple regional lymph node involvement. Contrast enhanced CT scan revealed large moderately enhancing soft tissue density lesion arising from left parotid gland with regional lymph node involvement. Pre-operative aspirated cytology shows evidence of ductal carcinoma of left parotid gland. Treatment co0mprised of wide local excision (left segmental mandibulectomy + total parotidectomy) with compromising facial nerve, selective neck dissection (I-III), and reconstruction made with SSG (split skin graft) harvested from right thigh region. The excised specimen and lymph nodes were sent for histopathological evaluation which reveals that sections studied from the excised mass show a malignant neoplasm arranged in sheets nests and comedo pattern with central necrosis. The circumferential margins and sections from segmental mandibulectomy are free of tumor, involvement of skin, perineural invasion and vascular invasion of tumor is noted. Sections from the lymph node specimen shows secondary carcinomatous deposits. Patient had been sent for adjuvant radiotherapy post operatively.

INTRODUCTION

In 1968 Kleinsasser *et al* described salivary duct carcinoma (SDC) which is comprising about 1 to 3 % of malignant salivary gland tumors[1]. However it was described in 1968, it was not included in WHO classification till 1991, later it was included in second version of WHO classification of salivary gland tumors[2]. SDC resembles a high grade mammary duct carcinoma which is an epithelial tumor having salivary gland sites as a rarest and it is also an aggressive adenocarcinoma. Clinically SDC is characterized by aggressive, early metastasis and local recurrence with significant mortality[3]. SDC hadresulted in to distant metastasis which includes lungs, liver and bones[4,5] and mostly within 4 to 5 years 50 % of patients will die due to this disease[6,7]. Histologically 27% of SDC arose from pre-existing pleomorphic adenoma of parotid gland with perineural invasion and 73% findings of cervical lymph node involvement[8]. Histopathological variants of SDC are invasive micropapillary, low grade and sarcomatoid variant. In English language literature there are more than 250 cases of SDC which had been reported[9, 10]. We report a case of SDC arising from parotid gland.

DISCUSSION

Kleinsasser was the first person to describe SDC in 1968 and it is a rare entity, which has been included in the WHO's second edition of classification of salivary gland tumors. In the year 2005 WHO classification defines SDC "*an aggressive adenocarcinoma which resembles high grade breast carcinoma*" [(11)]. Cribriform salivary carcinoma of excretory ducts and infiltrating SDC are the other names for SDC [(12, 13)]. Among all parotid tumors, SDC accounts 0.2% to 2% and 6 % to 10% of parotid cancers. Commonly seen in fifth or sixth decade with a mean age of 60 years, which usually affects men, with a male to female ratio of 2:1[(3)]. SDC had been reported to arise from long standing obstructive sialadenitis and it can be established from pleomorphic adenoma (carcinoma ex plemorphic adenoma)[(9)]. In English language literature, SDC had been reported that 37 cases originated from minor salivary

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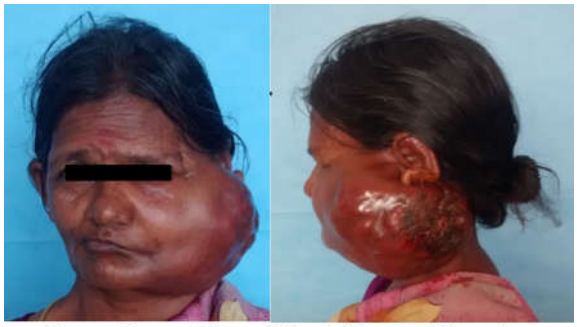

Figure 1. swelling of the left parotid region

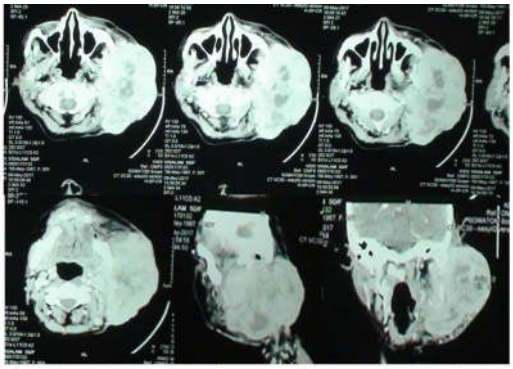

Figure 2. contrast enhanced computed tomography scan showing a soft tissue density lesion arising from left parotid gland with no clear margin

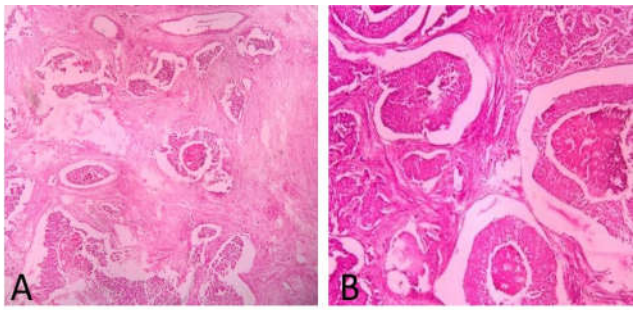

Figure 3. Histopathologic findings for salivary duct carcinoma. H&E-staining, original magnification of 50x (A) shows the individual cells are round to oval with eosinophilic cytoplasm and hyperchromatic nuclei, most of which are seen surrounding the central necrosis. (B) magnification of 100x shows malignant neoplasm arranged in sheets, nests and comedo pattern with central necrosis.

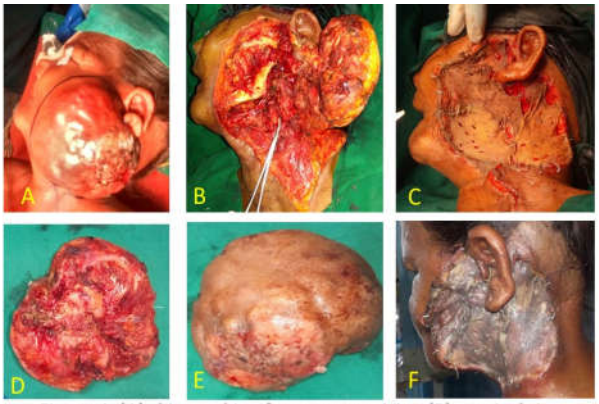

Figure 4. (A) skin marking for tumor excision (B) surgical site during tumor excision and SND (C) Surgical site closure with SSG (D & E) Macroscopic appearance of the excised tumor (F) Post operative view of surgical site after 1 month

glands and mostly seen in hard palate (14 cases)[14]. Facial paralysis had been misdiagnosed as Bell's palsy in parotid gland SDC. This tumor has Prognosis with a very dismal rate of 26 to 66 % local recurrence, 52.9 to 60 % neck metastasis and 50 to 66% distant metastasis to lung, bone, liver, back and leg. Due to wide spread of metastasis 70% of patients with SDC die of the disease within three years. Radical parotidectomy is mandatory in SDC patients presenting with facial nerve paralysis. SDC of parotid recurrence rate was reported in patients those who undergone parotidectomy and did not undergo lymph node dissection. Paclitaxel is reserved as a chemotherapeutic drug of choice in advanced salivary duct carcinoma as reported by Kuroda *et al*. [9, 15].Histopathologic findings dependence shows the positivity in the diagnosis of SDC when it is compared to FNAC, which is not reliable with CT and USG [16]. In the primitive lesion SDC calcification are noted radiographically. MRI findings shows a uninodular or multinodulartumor in T1 isosignal or T2 signal strongly enhanced by godolinium. The differential diagnosis of SDC are mucoepidermal carcinoma and mammary adenocarcinoma metastasis [3].

SDC has similarity with ductal carcinoma of breast histopathologically and it appear as cribriform, papillary or solid growth pattern often with central necrosis in intraductal component. In prominent desmoplastic pattern, SDC comprises of irregular bands and cords of cells. Based on the degree of intraductal or infiltrative component, Delgado *et al* have classified into three subtypes as follows

1. Predominantly intraductal
2. Predominantly infiltrative
3. Infiltrative[17]

CK 7 gives valuable evidence of immunohistochemistry staining for diagnosis of primary salivary gland carcinomas, metastatic carcinomas and in the diagnosis of salivary gland carcinomas. In initial diagnosis of SDC, analysis of p53 and HERR-2/ neu expression predicts the risk for development of local recurrence and metastasis. The presence of increased cellular activity and aggressive cli nical behaviour are depicted with high level of Ki-67 labelling index [11]. In present case, patient has been treated with a total left parotidectomy along with left segmental mandibulectomy, with excision of the adjacent facial nerve and surgical site reconstruction was done with split skin graft which was harvested from right thigh region. Patient had been sent for post operative radiotherapy as this case had extra parotid extensions with skin and regional lymph node involvement.

CONCLUSION

The prognosis is very poor due to the aggressive behavior and metastatic potential of SDC. 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 and is mandatory in advanced cases with regional lymph node involvement. Adjuvant chemotherapy and radiotherapy will help in oncological cure of the disease. After the excision of SDC involving the skin or adjacent structure, reconstruction should be considered for the defect where esthetics and functional activities plays a major role.

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How to cite this article:

A. Mathan Mohan *et al*.2018, Salivary Duct Carcinoma of Parotid Gland is a Rare Entity: Case Report. *Int J Recent Sci Res.* 9(1), pp. 23157-23159. DOI: http://dx.doi.org/10.24327/ijrsr.2018.0901.1408

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