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HER2/*neu* negative salivary duct carcinoma of parotid: A case with forty months recurrence free follow up

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ABSTRACT

INTRODUCTION: Salivary duct carcinoma (SDC) is very rare, only 1–3% of all salivary gland tumors are reported as SDC. SDC predominantly occurs in elderly males, SCD is characterized by an aggressive clinical course of the disease with less than 60% five years survival from the day of initial diagnosis, lymph node metastasis and facial nerve involvement is common, the current literature lacks protocol regarding management of this entity and the advantage of adjuvant therapy has not been evaluated due to its rare occurrence.
PRESENTATION OF CASE: We report patient with stage IV HER2/*neu* negative SDC successfully treated with surgery followed by adjuvant radiotherapy, patient is followed up for 40 months without evidence of recurrence or metastasis.
DISCUSSION: SDC is reported to be similar to mammary duct carcinoma in clinical and immunohistologic typing, further it shows an association of expression of HER-2/*neu* and *p*53, with early local disease recurrence, distant metastasis and survival, however; current case was adequately followed up and reevaluated after 26 months, MRI did not show evidence of recurrence.
CONCLUSION: SDC is a rare tumor and information on association of HER2/*neu* with survival is useful in further research on this tumor.

1. Introduction

Salivary gland cancers make up 5% of all head and neck cancers, most common of them is mucoepidermoid carcinoma. Incidence of salivary duct carcinoma (SDC) is very rare and reported 1–3% of all salivary gland tumors [1,2].

SDC is uncommon and is characterized by noticeable similarity to mammary duct carcinoma; it occurs predominantly in parotid of elderly males, involvement of facial nerve is common, incidence of cervical lymph nodal metastasis up to 73% [3], the aggressive clinical course associated with high incidence of recurrence and metastasis leads to poor survival in these patients. Similar to mammary duct carcinoma, HER2/*neu* positivity is associated with poor outcome in the patients with SDC, immunohistochemical typing shows a very high proliferation rate of Ki-67 in this tumor [4].

We report patient with HER2/*neu* (human epidermal growth factor receptor 2) negative SDC, successfully treated with surgery and adjuvant radiotherapy, patient is followed up for 40 months without evidence of recurrence or metastasis.

2. Presentation of case

A 51 years old man presented with a hard swelling over right parotid region, on clinical examination; the swelling was adherent to overlying skin, a hard enlarged submandibular lymph nodes were palpable, however; the facial nerve function was intact without evidence of palsy of facial muscles. FNAC of the mass suggested adenocarcinoma, MRI image findings revealed well defined hyper intense nodule at inferior pole of the right parotid gland, measuring 1.7 × 1.4 centimeters. Multiple enlarged lymph nodes were seen at level Ib, level II, level III, level IV and level V (Figs. 1 and 2,). Patient was evaluated for other co morbidities, posterior-anterior view of X-ray chest and MRI abdomen showed normal findings.

Patient was planned for radical parotidectomy, an incision encircling the parotid, around the adherent skin was planned, cervical extension of same incision was modified along neck crease and utilized for neck dissection, intra operative findings showed tumor extending up to the base of skull involving the internal jugular vein,

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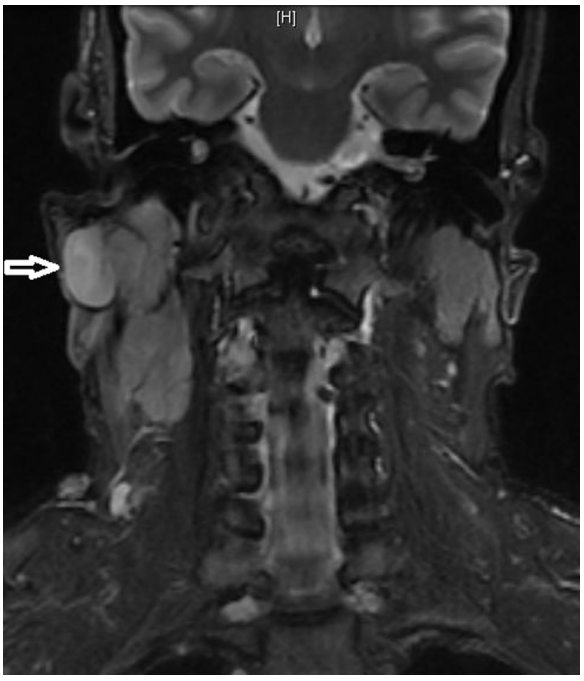

Fig. 1. Coronal MRI view illustrating the extant of tumor.

facial nerve and adherent to mastoid process and styloid process, right side neck dissection from level I to V was done sacrificing right facial nerve and part of internal jugular vein, three dimensional adequate resection margin was achieved through drilling of mastoid and styloid processes, reconstruction was done with pectoralis major myocutaneous flap.

Gross examination of resection specimen showed skin covered mass measuring 15 × 12.5 × 8 cm with surface nodularity, cut section showed salivary gland measuring 7.5 × 6.5 × 5.5 cm, a minimum 10 mm over all margin around tumor was present.

The histological examination suggested Salivary Duct Carcinoma with discerned lymphovascular emboli, nine of 45 dissected cervical lymph nodes revealed metastasis with no perinodal spread or perineural spread. The largest node measured 3.8 × 1.8 × 1.5 cm. The diagnosis for SDC was followed with HER2/*neu*, the patient was tested negative in this case.

Post operative healing was uneventful; adjuvant radiotherapy of 60 gray was delivered through intensity modulated radiotherapy (IMRT) starting on twenty seventh post operative day.

Patient had been followed for 40 months and did not show any evidence of recurrence, MRI done after 26 months showed small volume oval shaped node measuring 8 mm in diameter at left level III, rest of the scan showed normal findings, the FNAC for this node was negative for malignancy. Fig. 3 illustrates the operative field in MRI post 26 months.

3. Discussion

SDC largely affects males (66%), with a mean age of presentation at 62.5 years, it occurs primarily in the parotid gland (78%; submandibular gland, 12%; Minor salivary glands, 10%) [4]. The diagnosis is usually established through H&E staining, however; the immunohistochemical evaluation has a therapeutic importance.

The carcinogenesis of SDC disease is unknown, however; the recent data suggests a critical role of oncogenic *PIK3CA* in early stages of its development. The incidence of cervical lymph node metastasis is greater in SDC to other parotid malignancies, approximately two thirds of SDC patients present at T3 or T4 stage [4]. The prognosis of SDC is poor with less than 60% five year survival [6,7], the factors considerable regarding prognosis are advanced clinical presentation, resection margin positivity [8] and HER2/*neu* positivity [4]. The current case reported at an advanced stage with multiple cervical lymph nodal metastases, lymphovascular emboli, extension to skull base and internal jugular vein, however, absence of perineural spread, pre operative intact facial nerve function, adequate tumor free resection margins and negative status of HER2/*neu* were favorable indicators.

SDC is reported to be similar to mammary duct carcinoma in clinical and immunohistologic typing, Michael Jaehne et al. [4], in their report of 50 cases with clinical and immunohistologic typing of SDC showed an association of expression of HER-2/*neu* and *p53,* with early local disease recurrence, distant metastasis and survival (*p* value 0.05). They observed that 48% of patients

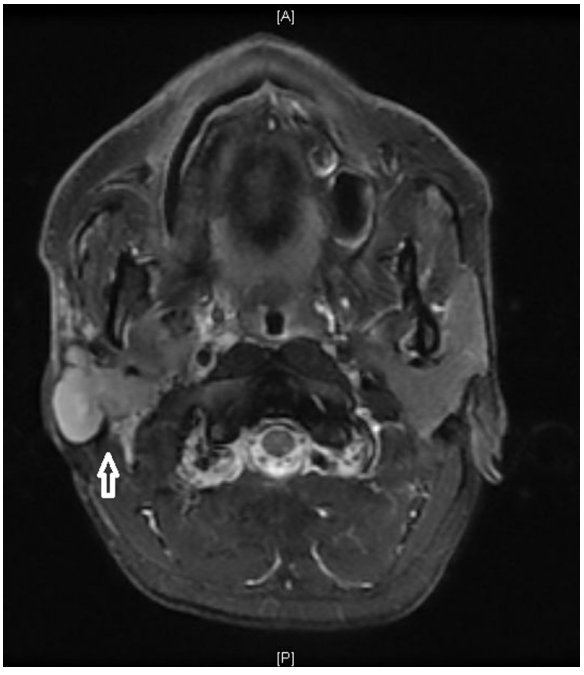

Fig. 2. Axial MRI view illustrating the extant of tumor.

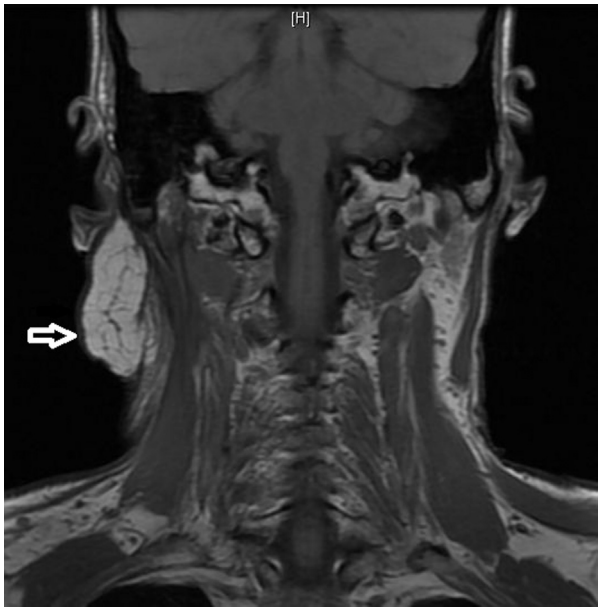

Fig. 3. Coronal MRI view after 26 months follow up.

developed metastasis within an average period of 29 months post surgery.

The reported patient was adequately followed up and reevaluated after 26 months, FNAC of suspected node on MRI and rest of the MRI did not show evidence of recurrence.

The association of HER2/*neu* positivity prompted research on the possibility of delivering chemotherapeutic agents such as trastuzumab, such therapy has been reported with better survival and response in palliative cases in the review by Nabili et al. [9], these authors have proposed routine examination of HER2/*neu* status in all patients with SDCs [9].

Although HER2/*neu* positivity of these tumors showed favorable response to chemotherapeutic agents, ERβ down-regulation was associated with adverse clinical features, this prompted attempts to treat SDC through hormonal therapy, however; the results were extremely variable, absence of uniform androgen or estrogen receptors in all the cases was the major factor determining outcome [10], though morphologically SDC was similar to ductal cell carcinoma of breast, there was significant variation in receptors distribution, a detailed review is presented by Simpson et al. [10], proposing reclassification of this entity.

Currently, the primary therapy for SDC is surgical resection with adequate negative margins, aggressive approach of surgery is stressed, patients often require radical parotidectomy even in early stages, perineural spread is common and facial nerve is often sacrificed to obtain oncologic clearance, regardless of location of primary tumor ipsilateral neck dissection is mandatory, bilateral neck dissection is indicated in patients with SDC of palate. Evaluation of adjuvant radiotherapy was not possible in literature due to paucity of cases, multi centric studies and reviews are required to evaluate response of this tumor to various therapeutic options; however; authors find it justified to subject the patient in stage IV disease to adjuvant radiotherapy as in this case.

4. Conclusion

SDC is a rare neoplasm with aggressive behavior, according to literature prognosis is not good even in initial stages, aggressive approach to surgery is justified and current case is successfully followed up for a period of forty months without evidence of recurrence. HER2/*neu* negativity might have some influence in this regard; however; further research is needed to study SDC.

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Conflicts of interest

None.

Funding

None.

Ethical Approval

None.

Author contribution

1. Sandhya Gokavarapu: study concept or design, data collection, data analysis or interpretation, writing the paper and surgery.
2. Daphne Fonseca: pathology reporting and analysis.
3. Sreenivasa Puthamakula: surgery and recording intra operative findings.
4. B. Sridhar Reddy: literature review.
5. Bal Reddy P: literature review.
6. Sudha Murthy: pathology reporting and analysis.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Guarantor

Sandhya Gokavarapu.

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