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Case Report

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Recurrent sinonasal teratocarcinosarcoma treated with IMRT: a case report

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ABSTRACT

Sinonasal teratocarcinosarcoma is a rare malignant tumour displaying aggressive growth .It is known to have poor prognosis with locoregional recurrence. So far, the most widely accepted therapeutic plan for its management has been surgery followed by adjuvant radiation. This study reports a case of 35 year old male diagnosed with sinonasal undifferentiated carcinoma who presented with neck node recurrence after 15 months of definitive radiotherapy. A 35 year old patient presented with nasal obstruction and epistaxis. CECT PNS revealed mass in ethmoid sinus with adjacent bony destruction and infiltration into maxillary sinus and nasal cavity. Biopsy revealed poorly differentiated carcinoma. Patient underwent neoadjuvant chemotherapy followed by definitive radiotherapy by IMRT. On follow up at 15 months, the patient presented with nodal recurrence. Biopsy from neck node revealed metastatic sinonasal teratocarcinosarcoma. The patient underwent neck dissection followed by adjuvant chemoradiation. The patient is currently on follow up and is disease free till date. Surgery alongwith adjuvant chemoradiation is a reasonable therapeutic approach of management for sinonasal teratocarcinosarcoma.

Keywords: Sinonasal carcinoma, Teratocarcinosarcoma, IMRT

INTRODUCTION

Sinonasal teratocarcinosarcoma ia a rare aggressive malignant tumour of the sinonasal region. This clinical entity was first described by Shanmugaratnam in 1983 as teratoid carcinosarcoma.[1] Heffner and Hyams coined the term "teratocarcinosarcoma" based on a clinicopathologic study of 20 cases with sinonasal tract neoplasms.[2]

These lesions are potentially aggressive in nature and commonly recurrent, with an average survival of less than 2 years.[3]

We present here a case of recurrent sinonasal teratocarcinosarcoma managed by surgery, radiotherapy and chemotherapy.

CASE REPORT

A 35 year old male presented at opd with nasal obstruction and ipsilateral epistaxis. On examination, an extensive mass in the nasal cavity involving the choana was noted. There was visible proptosis of right eye.

CECT PNS revealed ill defined heterogenous mass of size 4×3.7 cm with contrast enhancement in right ethmoidal sinus with adjacent bony destruction and infiltration into left ethmoidal sinus, right maxillary, frontal and ethmoid sinus. No cervical lymphadenopathy was noted on physical examination and imaging. The tumour was staged as T4N0M0. Biopsy from the nasal mass revealed poorly differentiated carcinoma. IHC stained positive for synaptophysin, CK and CGA.

In view of advanced disease, the patient was given 2 cycles of neoadjuvant chemotherapy with paclitaxel and cisplatin for debulking. The tumour regressed in size and then the patient was planned for radiation.

The patient was positioned supine in a standard head rest and immobilized with a thermoplastic mask. CT scan was

performed from skull vertex to sternum using iv contrast. 3mm slices were obtained. GTV included the gross primary tumour visible on planning CT images. CTV was drawn with an expansion of 1 cm to GTV. 0.5 cm was further added to CTV to create PTV. OARs contoured were bilateral orbits, lenses and optic nerves and brainstem and parotid gland. Dose constraints were prescribed according to QUANTEC model.[4,5] 7-field IMRT planning was done in Monaco treatment planning system. The patient underwent IMRT to primary site using 6MV photons to deliver 60 Gray in 30 fractions. The patient tolerated the treatment well. Patient was kept on regular follow up.

At 15 months of follow up, the patient presented with right submandibular swelling. PET-CT showed FDG uptake in right level Ib and II neck nodes. There was no uptake elsewhere. FNAC from the submandibular node showed poorly differentiated carcinoma. IHC stained positive for AE1/AE3 and synaptophysin. The patient underwent modified neck dissection. HPE revealed metastatic sinonasal teratocarcinosarcoma. The patient recived adjuvant IMRT 60Gy in 30 fractions to right neck with 6 weekly cycles of concurrent cisplatin. CTV included level I,II and III neck nodes. A 0.5 cm expansion to CTV was added for PTV. The patient is on regular follow up and is disease free till date i.e., 1 year form recurrence.

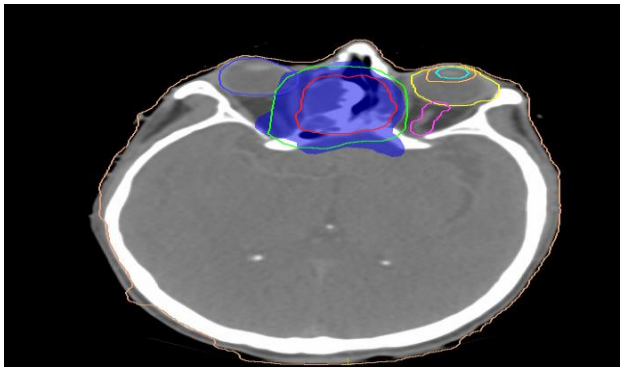

Figure 1: Imrt for primary sinonasal tumour.

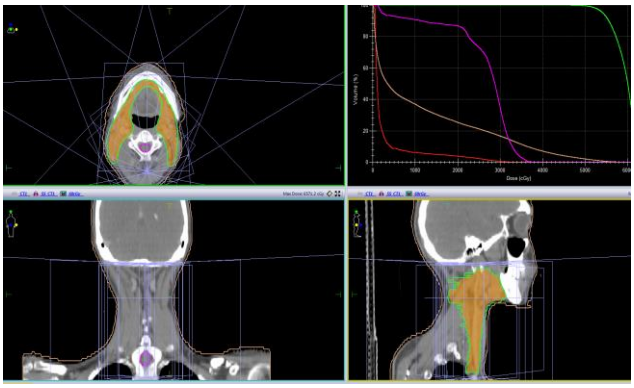

Figure 2: Imrt dose prescription for neck nodal recurrence.

DISCUSSION

The origin of sinonasal teratocarcinoma (SNTCS) remains unknown. SNTCS microscopically consisits of primitive neuroepithelial elements with various malignant epithelial and mesenchymal components probably originating from stem or pluripotent progenitor cells with multidirectional differentiation.[6,7]

The gold standard diagnostic tool for this tumour is immunohistochemical staining. Epithelial components are cytokeratin and EMA positive. Neuroepithelial components are NSE,CD99,GFAP,synaptophysin and S100 positive.[8] As seen in our case, IHC was positive for synaptophysin, CK, S100 and CGA.

Budrukkar et al conducted a study on 22 cases, 14 of them received surgery followed by radiation and 11 cases received chemotherapy as well. At 34 months of follow up, only 5 cases had disease under control. Recurrence was noted in 11 patients. This study advocated that a multimodality approach with surgery, chemotherapy and radiation is the most effective method of mangement.[9]

IMRT has the capability to give concave and rapid dose fall-off distributions leading to higher dose to target volume while reducing dose to organs at risk to preserve organ function and improve quality of life. This technique can be used safely and effectively for nasal cavity and paranasal sinus malignancies.[10-12] IMRT helped us to acieve adequate dose coverage in our case. Locoregional recurrence is commonly noted in SNTCS which was clearly evident in our case. Distant metastasis is however rarely reported.[13]

CONCLUSION

SNTCS is a rare tumour and hence optimal management with surgery, radiotherapy and chemotherapy should be standardized.

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