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NEUROENDOCRINE TUMOR OF NASAL CAVITY: A CASE REPORT

Dr. Bhupendra Singh Chahar*, Dr. Ashok Kumar Arya, Dr. Roopali, Dr. Durgesh

Resident Doctor, S.N Medical College, Agra, Uttar Pradesh, India.

***Corresponding Author: Dr Bhupendra Singh Chahar**
Resident Doctor, S.N Medical College, Agra, Uttar Pradesh, India.

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INTRODUCTION

The sinonasal tract is the site for a large variety of benign and malignant tumors. The diversity is mainly due to the anatomic complexity and widely varying tissues in this compact area. Malignant sinonasal tract tumors comprise less than 1% of all malignancies[1] and primary neuroendocrine carcinomas represent only 5% of malignancies arising in the sinonasal cavity.[2] In the nasal cavity and paranasal sinuses, the most common site of origin of this malignancy is the ethmoid sinus and the nasal cavity. Neuroendocrine cancer of sinonasal cavity show high propensity for recurrences after treatment and owing to its rarity, there is no standard treatment guideline for these tumors making it challenging to decide the optimal treatment for these cases.

CASE PRESENTATION

A seventy three years old male presented with complains of nasal discharge, on and off nasal blockage and a few episodes of epistaxis for 3 months. On examination, there was gross deviation of nasal septum with a polypoidal mass in the left nasal cavity reaching upto the left nostril. There was no clinically palpable cervical lymphadenopathy. No neurological deficit was elicited and ophthalmic assessment was normal.

A CECT scan of PNS was done which revealed a heterogeneous mass of 8.3* 3.8* 3.2 cm involving the left nasal cavity, left ethmoid sinus, left maxillary sinus, bilateral sphenoid sinus and left orbit, with extension into the nasopharynx. No cervical lymphadenopathy was seen. On HPE, there were malignant cells in small to large clusters having high nucleus/cytoplasm ratio, scanty cytoplasm, high mitosis, and some poorly formed rosettes. It was positive for synaptophysin and NSE; focal positive for Pan CK and EMA. Negative for CK5/6, chromogranin, vimentin and calretinin and the impression was that of Neuroendocrine carcinoma grade III. CECT chest and abdomen was normal. So we made a diagnosis of primary neuroendocrine carcinoma of sinonasal cavity with no regional or distant metastasis.

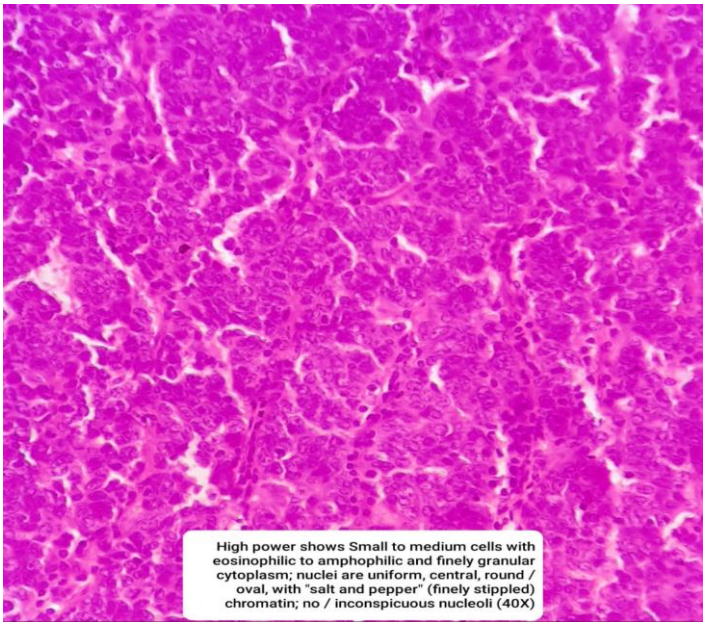

Figure 1: H and E stained slide showing neuroendocrine tumor of nasal cavity.

This patient did not want any surgical procedure so we have treated him with neoadjuvant chemotherapy followed by radiotherapy. In neoadjuvant chemotherapy, he received 4 cycles of Cisplatin and Etoposide based chemotherapy. After this the patient was evaluated with clinical and radiological examination and no mass was seen and only soft tissue density in left maxillary and ethmoidal sinus was seen on CT scan. Further, the patient was planned for EBRT using cobalt -60 to a total dose of 60 Gy in 30 fractions[using one wedged anterior and one left lateral fields]. After recieving 40 Gy/20 fractions treatment, he defaulted due to covid-19 pandemic. He again visited us after a gap of 4 months, and was evaluated with imaging. The CECT PNS scan shows complete tumor response with no evidence of any abnormal lesion in the nasal cavity or paranasal sinus. In view of complete response and long gap the patient was kept on close followup only.

During treatment, the patient did not have any severe side effects, but due to large treatment field and 2-D planning he had burning sensation and redness in left eye. On repeat ophthalmic assessment his vision was found normal.

Now it has been 9 months post treatment and he does not have any symptoms or signs of disease recurrence.

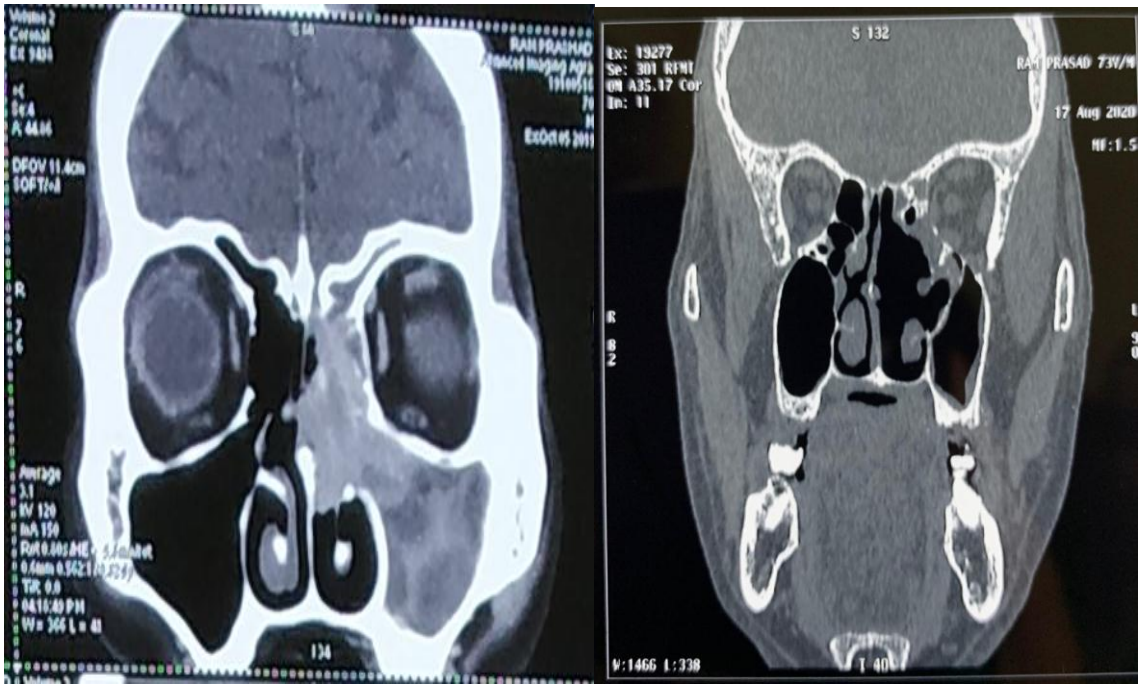

Figure 2: Pre-treatment vs post-treatment CECT PNS coronal section.

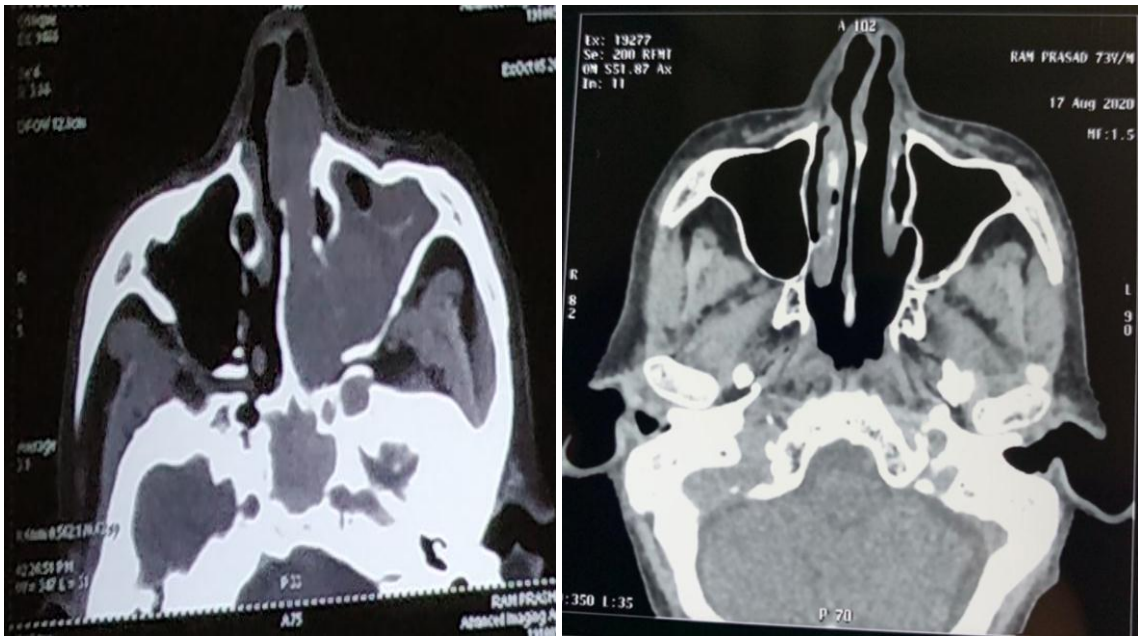

Figure 3: Pre vs post treatment CECT PNS axial section.

DISCUSSION

Primary sinonasal neuroendocrine carcinoma was first described by Silva et al in 1982, before which it was not considered as an independent entity.[3] The mean age of presentation was found to be around 50 years having same male to female predisposition.[4] No specific etiological factors have been described as of now.[5],[6] For staging purpose, the Kadish staging for esthesioneuroblastoma is often employed for neuroendocrine carcinomas of nasal cavity.[7] Also, in addition to this, the AJCC staging system of nasal cavity and paranasal sinuses can also be used.[8] According to the Kadish system as well as the AJCC system our patient belonged to Group C. These tumors are rare, aggressive and have a very high potential for reccurrence.

The standard treatment for sinonasal neuroendocrine tumors remains controversial because of lack of sufficient data, because of the small number of cases owing to its rarity. Although a multimodality treatment approach including surgery, chemotherapy and radiotherapy in advocated due to the aggressive nature of the disease, no consensus regarding the adequate management of the patient is there. In their studies, Bhattacharya et al[9], Fitzek et al[10] and Babin et al[11] proposed chemotherapy with cisplatin and etoposide based regimen followed by radiotherapy and surgery reserved for non-responders as the treatment protocol for these tumors. At the same time, a few recent studies by Chang et al[12], Qian et al[13], and Likhacheva et al[14] advocate in favour of surgery based treatment and have concluded that combined modality treatment including surgery has significantly better disease free and overall survival as compared to treatment without surgery. Van der etal in a meta-analysis of 701 cases of sinonasal neuroendocrine carcinoma concluded that proper histologic diagnosis with grade of differentiation is an important prognostic factor for these cancers. Also, surgery is the cornerstone for treating these malignancies, although radiotherapy plays an important role specially in poorly differentiated cancers.[15]

In our case, the patient was not willing to go for surgical treatment, but the disease showed a very good response to initial chemotherapy. Also the patient received radiotherapy to a total dose of 40Gy, which is lower than the dose given to other tumors of sinonasal cavity and still a complete tumor response was achieved and the patient is disease free 9 months after completion of the treatment. According to the available literature, even with multimodality treatment, risk of local and distant disease recurrence is very high for these tumors, so as seen we can go for a combined treatment modality including chemotherapy followed by radiotherapy given at a lower dose, keeping surgery and further radiotherapy in reserve for any recurrence in the future.

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