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Case Report

Sinonasal teratocarcinosarcoma: a rare case report from a tertiary care centre of North East India

Barasha S. Bharadwaj, Asreen Suhana*, Muktanjalee Deka, Jagannath D. Sharma, Adahra P. Beso, Upasana Kalita, Bibhash C. Goswami

Department of Oncopathology, SCI, GMC, Assam, India

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***Correspondence:**
Dr. Asreen Suhana,
E-mail: asreensuhana@gmail.com

ABSTRACT

Sinonasal teratocarcinosarcoma is an extremely rare malignant tumour arising in the sinonasal tract, that may extend intracranially to complicate the treatment and further worsen its dismal prognosis. Diagnosis is challenging because of its rarity and morphologic heterogeneity. Here, we reported a case of a 55 years old male who presented with complaints of left sided nasal blockage and facial swelling. CECT showed a large sinonasal mass with epicentre in the left nasal cavity extending to post nasal space and nasopharynx, eroding the left medial orbital wall and cranially the cribriform plate. Histopathologically, malignant epithelial component comprising of squamous cell carcinoma, mesenchymal component comprising of fibrosarcoma with focal chondroid differentiation, primitive blastemal component with extensive necrosis was noted. Immunohistochemistry demonstrated positivity for synaptophysin, chromogranin, Pan-CK, EMA, CD99, focal p63 in areas of squamous metaplasia; stromal cells showed Desmin, S100, SOX 10 expression: Ki-67 was 30-40%. Immunohistochemistry confirmed the diagnosis of sinonasal teratocarcinosarcoma. Knowledge about this tumour is important because of its heterogenous morphology which often leads to a misdiagnosis, necessitating repeated biopsies and thorough examination of the surgical specimen.

Keywords: Teratocarcinosarcoma, Sinonasal, Immunohistochemistry

INTRODUCTION

Sinonasal teratocarcinosarcoma is an extremely rare malignant tumour of uncertain histogenesis arising in the sinonasal tract, that is locally aggressive with high rates of recurrence with overall survival of <2 years.[1,2] The probable origin is from a primitive olfactory membrane cell, that produces neuroectodermal features of neuroblastoma, along with the capacity of divergent differentiation.[3] These tumours are often misdiagnosed because of its rarity and morphologic heterogeneity.

CASE REPORT

Here, we reported a case of a 55-year-old male who presented with complaints of left sided nasal blockage and facial swelling for 4 months. CECT PNS showed a large sinonasal mass measuring 9.8×4.3×4.5 cm with epicentre in the left nasal cavity extending to post nasal space and nasopharynx, eroding the left medial orbital wall and cranially the cribriform plate with multiple enhanced lymph nodes. Biopsy was taken and histopathology combined with immunohistochemistry, it was diagnosed as sinonasal teratocarcinosarcoma. The

patient was treated with left medial maxillectomy in December 2021 and then advised adjuvant chemoradiotherapy. The patient received 60 Gy in 30 fractions along with 5 cycles of concurrent carboplatin from March 2022 to July 2022. On follow-up, the CECT PNS showed a residual mass of 4 cm extending to the nasopharynx with bony erosion without intra-orbital or intra-cranial extension and the mass progressed as compared to the previous scan. The patient also developed lung metastases. Due to breathing difficulty, debulking surgery was performed, but the patient eventually succumbed to death.

Fine needle aspiration findings

Cellular smears from left level II lymph nodes showed clusters and scattered highly atypical cells with high nucleocytoplasmic ratio, hyperchromatic nuclei, irregular nuclear membrane and conspicuous nucleoli in a haemorrhagic background. Features suggestive of high grade malignant lesion (Figure 1).

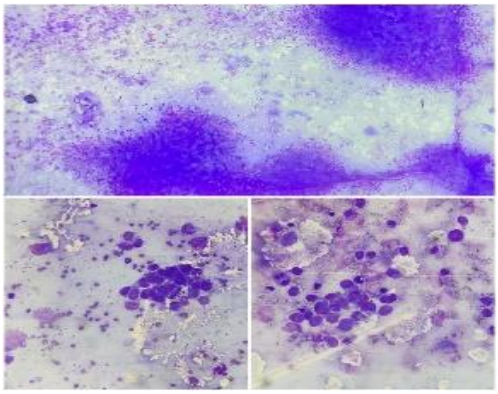

Figure 1: Low and high power views of fine needle aspiration smears.

Gross findings

Following debulking surgery, we received multiple pieces of friable tissue aggregate measuring 6×5×3 cm^3; cut surface was solid, grey white with few haemorrhagic and necrotic areas (Figure 2).

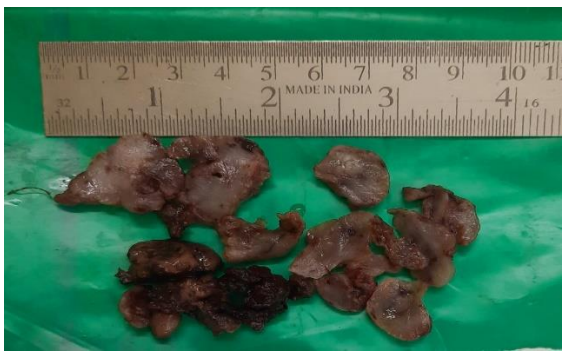

Figure 2: Gross picture of the debulking surgery specimen.

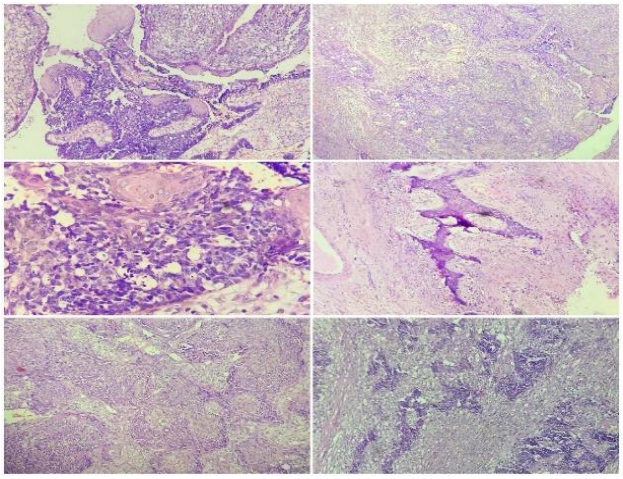

Figure 3: Low and high power views of HPE findings of teratocarcinosarcoma.

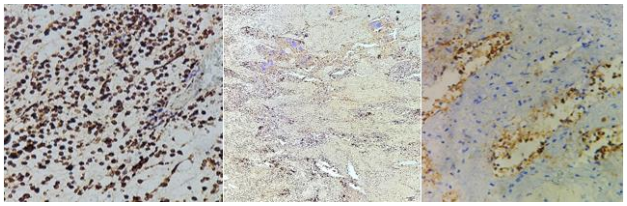

Figure 4: IHC images showing Pan CK, desmin and synaptophysin positivity.

Histopathological examination

Microscopy showed tissue lined by ciliated pseudostratified squamous epithelium with malignant epithelial component comprising of squamous cell carcinoma, mesenchymal component comprising of fibrosarcoma with focal chondroid differentiation, primitive blastemal component with extensive necrosis (Figure 3).

Immunohistochemistry findings

The neuroepithelial component showed positivity for synaptophysin, chromogranin, EMA, CD99 and retained BRG-1 and INI-1 (Figure 4). The glands expressed Pan-CK, focal p63 in areas of squamous metaplasia; stromal cells showed Desmin, S100, SOX 10 expression. Ki-67 was 30-40%. Immunohistochemistry confirmed the diagnosis of sinonasal teratocarcinosarcoma (Figure 4).

DISCUSSION

Sinonasal teratocarcinoma is a rare tumour with about 100 cases reported in world literature and was first described by Heffner et al in 1984.[1] It is mainly a tumour of adults (18-79 years) with male predominance (7:1 to 8:1).[1,4,5] It arises primarily in the ethmoid sinus and maxillary antrum, cases had been reported in the oral cavity, orbit and nasopharynx.[6] Grossly, these tumours are bulky, friable to firm, red-brown masses. On

microscopy, teratocarcinosarcomas are characterized by a combination of epithelial and mesenchymal tissues with variable growth patterns. The epithelial components include glandular or ductal structures lined by benign-appearing, partly ciliated columnar epithelium with transitional areas of nonkeratinizing squamous epithelium. In addition, areas of squamous carcinoma and adenocarcinoma are present. The mesenchymal components may include fibroblasts or myofibroblasts of benign and malignant appearance, benign cartilage with an immature appearance and chondrosarcoma or osteogenic tissue.[7,8] Fetal-appearing squamous epithelium with clear cell change and immature neuroepithelium are important histopathologic clues.[9] Local spread by causing bony destruction as well as distant spread to lymph nodes, lungs was seen. Surgery with adjuvant chemoradiation was the treatment of choice. Role of neoadjuvant therapy was still to be proven. Recurrence was the most common cause of treatment failure, so aggressive follow-up is very important. The average survival is <2 years with 60% surviving less than 3 years.[10]

CONCLUSION

The varied components of this rare tumour require histopathological and immunohistochemical correlation. Biopsies from multiple sites and thorough examination of the resected specimen are crucial for diagnosis. Aggressive follow-up and management with surgery and adjuvant chemoradiation is a must to prevent local as well as distant spread and to detect early recurrences.

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