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Case Report

Intestinal type sinonasal adenocarcinomas: a tale of two rare cases

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ABSTRACT

Sinonasal adenocarcinoma is a rare malignancy arising from the glandular epithelium of the nasal cavity and paranasal sinuses, accounting for <1% all malignant sinonasal tumours. It commonly presents with symptoms such as nasal obstruction, epistaxis, and facial pain, often leading to late diagnosis. Here we are presenting cases of two young individuals with left nasal mass extending into cranial cavity and frontal sinuses. CT findings described them are infiltrative lesions with heterogenous contrast enhancement with involvement of cranium. Histopathology and PAS stain revealed atypical cells arranged in papillaroid to compact glandular growth pattern. At places, back-to-back arranged glands were also noted, composed of columnar cells having abundant eosinophilic cytoplasm and basally located nuclei with mild anisonucleosis. Relevant history was taken to exclude the possibility of colorectal metastasis; later CT abdomen was also done. Finally, IHC was performed on both cases which revealed positive staining for CK7, CDX2, SATB2 and negative staining for CK20, supporting the diagnosis. Thus, sinonasal adenocarcinoma should be considered in cases of persistent nasal symptoms. Early detection through imaging and histopathology aids in timely intervention.

Keywords: Intestinal, Adenocarcinoma, Young patients, Nose, Sinuses

INTRODUCTION

There are approximately 0.2 percent to 0.5 percent of malignant tumors in the nasal cavity and the paranasal sinuses of the human body.[1] Malignant tumors of the sinonasal region account for 1 to 4 percent of all head and neck cancers.[2] Following adenoid cystic carcinoma and squamous cell carcinoma, adenocarcinoma is the third most prevalent mucosal cancer in the head and neck area. Adenocarcinomas most commonly originate in salivary glands, but some exhibit histologic patterns similar to those of the colon and are referred to as intestinal-type sinonasal adenocarcinomas (ITACs).[3] They account for 8–25% of all sinonasal carcinomas.[4] In addition to being more prevalent in males, sinonasal adenocarcinomas (SNACs) have a poor overall survival rate because they are often diagnosed at an advanced stage when there are fewer therapeutic options available.[5] These tumors are frequently detected in male employees in the shoe and hardwood industries. They are regarded as profession-related cancers because wood dust exposure raises the risk of adenocarcinoma by 900 times.[4] Early diagnosis is crucial for better management and prognosis. Herein, we discuss two cases of ITAC in a 21-year-old male and a 32-year-old female with left-sided nasal obstruction. This case highlights the importance of histopathology and immunohistochemistry in evaluating these tumors.

CASE REPORT

Case 1

A 21-year-old male presented to the ENT clinic with chief complaints of progressive left nasal obstruction. During

the clinical assessment, a polyp-like growth was observed on the left side, positioned laterally to the middle turbinate. Additionally, a yellowish discharge was noted in the same area. During clinical examination, polypoidal tissue was seen lateral to the medial turbinate on the left side, along with a yellowish discharge. CT scan revealed a large well-defined heterogeneous hypodense mass lesion of size 54×39 mm attached to the nasal septum, extending into the left nasal cavity. The lesion is also extending into the left frontal sinus showing no contrast enhancement.

A histopathological examination was done. H& E-stained sections showed a tumor arranged in irregular glandular pattern, cords, as well as singly scattered cells on an extensive mucin-rich background with mildly pleomorphic cells that show high nucleocytoplasmic ratio and hyperchromatic nuclei with inconspicuous to conspicuous nucleoli at places. Few cells also display mucin vacuoles. Mucinous areas and mucous cells were intensely PAS positive.

On immunohistochemical examination, tumor cells were positive for CK20, CDX2 & SATB2 supporting histopathological diagnosis. Based on characteristic histopathological features and immunohistochemical analysis a diagnosis of sinonasal adenocarcinoma, intestinal type was made.

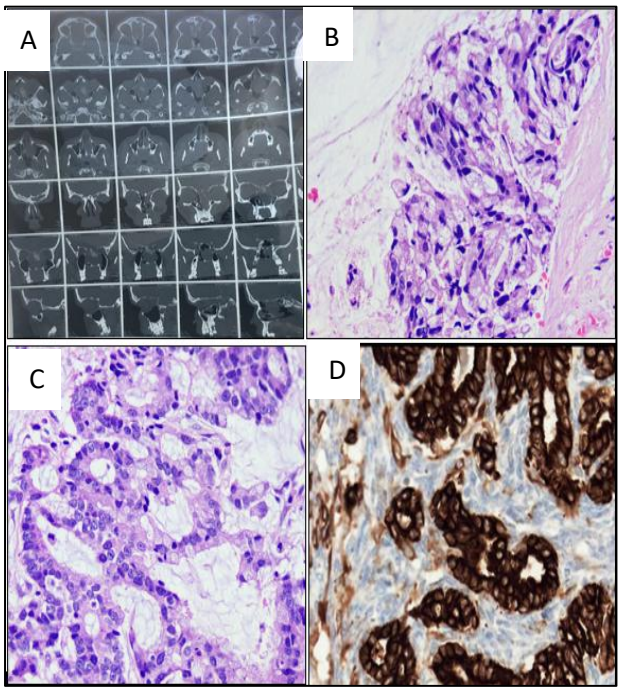

Figure 1: (A) Contrast-enhanced CT showed a large well-defined heterogeneous hypodense mass lesion of size 54×39 mm attached to the nasal septum, extending into the left nasal cavity along with frontal sinus extension. (B and C) H& E, 20X stained sections from case 1 show atypical cells arranged in compact glandular growth pattern with variable mucoid areas. (D) Tumour cells are displaying diffusely membranous positive for CK7 on 40X.

Case 2

A 32-year-old female presented to the ENT clinic with complaints of progressive left sided nasal obstruction, intermittent epistaxis, and facial discomfort for 6 months. Nasal endoscopy revealed a mass occupying the left nasal cavity. Contrast-enhanced CT and MRI showed an ill-defined mass with heterogeneous enhancement involving the nasal cavity with intracranial extension. The patient was advised for further histopathological correlation. H & E stain sections of the biopsy composed of atypical cells arranged in papillaroid to compact glandular growth pattern. At places, back-to-back glands were also noted, composed of columnar cells having abundant eosinophilic cytoplasm and basally located nuclei with mild anisonucleosis. Immunohistochemistry (IHC) revealed positive staining for CK 20 and CDX2, supporting the diagnosis. Based on characteristic histopathological morphology and immunohistochemistry, a final diagnosis of sinonasal adenocarcinoma, intestinal type, was made.

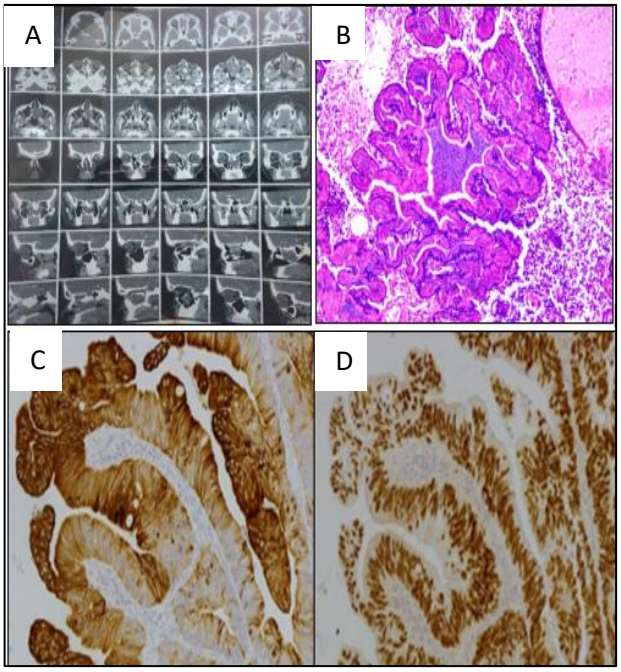

Figure 2: (A) Ill-defined mass with heterogeneous enhancement involving the nasal cavity with intracranial extension. (B) H& E, 20X stained sections from case 2 show atypical cells arranged in papillaroid to compact glandular growth pattern with mucin filled cysts. (C) The tumour cells are displaying membranous positivity for CK 7 on 40X. (D) The tumour cells display nuclear positivity for CDX2 on 40X.

DISCUSSION

SNAC (sinonasal adenocarcinoma) is a rare neoplasm in the sinonasal region. In the latest 5th edition of the WHO classification of head and neck tumors, two types of SNAC are distinguished: intestinal-type and non-intestinal-type, the latter subdivided into high- and low-grade tumors.[6]

Several ITACs have been reported worldwide, occurring sporadically or as an occupational risk for wood dust workers. The furniture, leather, and textile industries in European countries exhibit the highest incidence rates.[7] It is believed that about 20% of sinonasal intestinal adenocarcinomas are idiopathic i.e., they are not caused by industrial dust exposure.[1,2] Other factors considered etiologically responsible are Epstein-Barr virus infection and smoking.[8] The condition is thought to arise from the intestinal metaplasia of the ciliated respiratory cells constituting the Schneiderian membrane.[9-11] We had two patients of younger age, neither of whom had any known risk factors.

Clinical symptoms such as nasal obstruction, epistaxis, and rhinorrhoea can be misdiagnosed as chronic sinusitis or rhinitis, which may prolong the diagnosis process.[9,10] These tumors are often missed because their early symptoms are non-specific or asymptomatic. The lesion is characterized by an irregular polypoid pink or white mass that is expanding in the nasal cavity or paranasal sinus. It is frequently necrotic and friable.[9,12] In our case, both patients presented with a polypoid tissue-filled nasal cavity.

Among ITAC origins, 40% related to ethmoid sinuses; 28% to the nasal cavity; 23% to maxillary antrum; and 9% to undetermined sites.[3] In both current cases, the lesion was found to be in the nasal cavity. Nasal endoscopy aids in determining the tumor's location and size. A CT scan and an MRI can identify a disease's exact location and extent. According to immunohistochemistry studies, ITACs are consistently positive for CK20, epithelial membrane antigen, CDX-2, villin, MUC2, and STAB2, but variably positivity for CK7 and CEA is noted.[13] In both cases, tumor cells showed positivity for CD20 and CDX2.

Differential diagnosis of ITAC can include both sinonasal low-grade non-intestinal adenocarcinoma and colonic adenocarcinoma metastasis.[14,15] The differentiation between primary ITAC and colonic adenocarcinoma metastases is based on clinical features and CT abdomen/Colonoscopic findings of a patient. Immunohistochemistry for CK20, CDX-2, villin, and SATB-2 that stains only ITACs helps to exclude sinonasal non-intestinal adenocarcinomas.[16] In the case of non-extensive, resectable ITAC, transnasal endoscopic surgery may be the best treatment option.[11] Advanced stages and high-grade lesions, however, may require adjuvant radiotherapy plus chemotherapy in combination with aggressive surgical resection.[11,17]

Sinonasal AC is a potentially curable disease despite being an aggressive malignancy with a poor natural history. The involvement of key structures such as the anterior skull base (especially the dura mater and the brain), the orbital apex, the cavernous sinus, and the infratemporal fossa is recognized in the literature to be a factor influencing survival. Standard treatment/primary approach is extensive surgery aiming to remove as much of the tumor as possible and for advanced and recurrent cases, a combination of surgical resection and adjuvant radiotherapy along with chemotherapy is administered. Five-year OS rates vary between 21-78% in the international literature.[18]

CONCLUSION

Sinonasal adenocarcinoma should be considered in cases of persistent nasal symptoms. The discussed cases ascertain the pivotal role of histopathology and IHC in the diagnosis of sinonasal adenocarcinomas. Early detection through imaging and histopathological evaluation with IHC characterization aids in timely intervention. Surgical excision remains the gold standard, with radiotherapy playing a key role in preventing recurrence. These rare malignancies have the potential for improved outcomes with earlier diagnosis and better management

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