Case Report

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Pathology Section

A Case of Sinonasal Intestinal Type Adenocarcinoma Mimicking CNS Tumour: A Pathologist's Perspective

POOJA BHARTI[1], SANJEEV KUMAR SINGH[2], ARTI AGARWAL[3], RASHMI[4], ANITA OMHARE[5]

ABSTRACT

Sinonasal Intestinal-Type Adenocarcinoma (ITAC) is a rare neoplasm. The overall incidence of ITAC is less than 1% of all neoplasms, but almost 4% in sinonasal malignancies. This tumour tends to be very locally aggressive but rarely metastasises. It is frequently misdiagnosed clinically as a salivary gland tumour or Central Nervous System (CNS) tumour. In this case, a 50-year-old female presented with chief complaints of headache, vomiting and occasional nasal bleed for a few months. Radiological diagnosis (MRI) of CNS tumour was made. The other relevant necessary investigations and clinical management was done as per surgical oncology team. The tumour was removed by bifrontal craniotomy. On histopathological examination, two differential diagnoses of metastatic Gastrointestinal (GI) adenocarcinoma and sinonasal ITAC were made. Tumour cells exhibited strong immunopositivity for CK-20, CDX2, and Epithelial Membrane Antigen (EMA), while being negative for Glial Fibrillary Acidic Protein (GFAP). Thus, the final diagnosis made was well-differentiated sinonasal adenocarcinoma of the intestinal type. This case highlights the importance of histopathology with Immunohistochemistry (IHC) when most probable clinical as well as radiological diagnosis was CNS tumour. Histopathological examination with a combination of IHC had confirmed the diagnosis of sinonasal ITAC.

Keywords: Bifrontal craniotomy, Paranasal sinus, Sinonasal malignancy

CASE REPORT

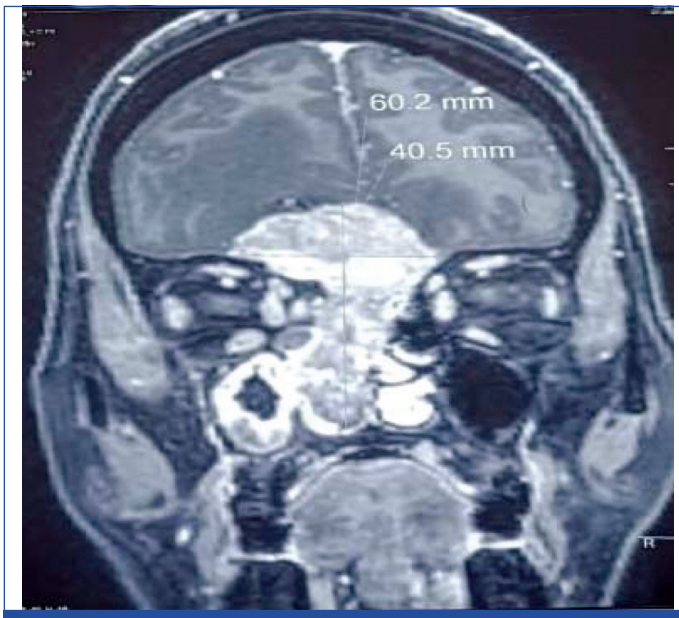
A 50-year-old female presented to the Neurosurgery department with a chief complaint of headache for four months, vomiting for 15 days, and three episodes of bilateral nasal bleed since one week. There was a history of nasal obstruction, snoring during sleep, and occasional blurry vision. There was no other significant medical, surgical or family history. The patient gave no history of smoking or wood dust exposure.

On examination, the patient was conscious, cooperative, and well oriented to time, place and person. Glasgow Coma Scale (GCS) was 15/15, visual assessment was 6/60 and other cranial nerve examinations were unremarkable. Complete Blood Count (CBC), and organ functions tests were within normal limits. No rhinorrhoea or facial asymmetry was identified.

On MRI scan (T1 coronal view), a well-defined soft-tissue Space Occupying Lesion (SOL) measuring 6×4.5 cm, was noted involving the visualised right maxillary sinus, right ethmoid air cells and right sphenoid sinus occupying the entire sinus. The soft-tissue mass was seen extending into the frontal intracranial region with significant perifocal oedema noted, possibility of neoplastic aetiology, involving the paranasal sinus extending into the intracranial region [Table/Fig-1,2]. Evidence of focal bony involvement or destruction was seen.

Clinical as well as radiological diagnosis was a CNS tumour. The other relevant necessary investigations and clinical management was done as per surgical oncology team. The SOL was totally removed by bifrontal craniotomy surgery and followed up by the onco-surgery department.

Intraoperatively tumour was large around 6×4.5 cm, irregular, greyish white in colour, and moderately vascular nature. A sample was sent for frozen section examination. Frozen section showed tissue chiefly comprising variable-sized glands lined by columnar cells, lying in loose stroma, which was infiltrated by mixed inflammatory infiltrate. Some cells also showed nuclear pleomorphism and hyperchromasia, most likely atypia. Though these features were suggestive of a high-grade tumour, but no definite diagnosis could be made.

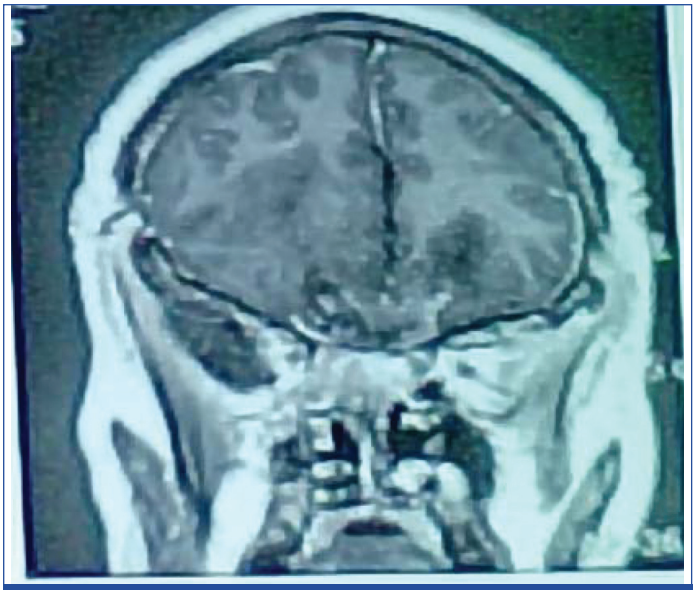


[Table/Fig-1]: MRI Scan image: Pre-operative coronal view showing a well-defined soft-tissue Space Occupying Lesion (SOL) involving the right maxillary sinus, right ethmoid air cells and right sphenoid sinus. The soft-tissue mass extended into the frontal intracranial region with significant perifocal oedema.

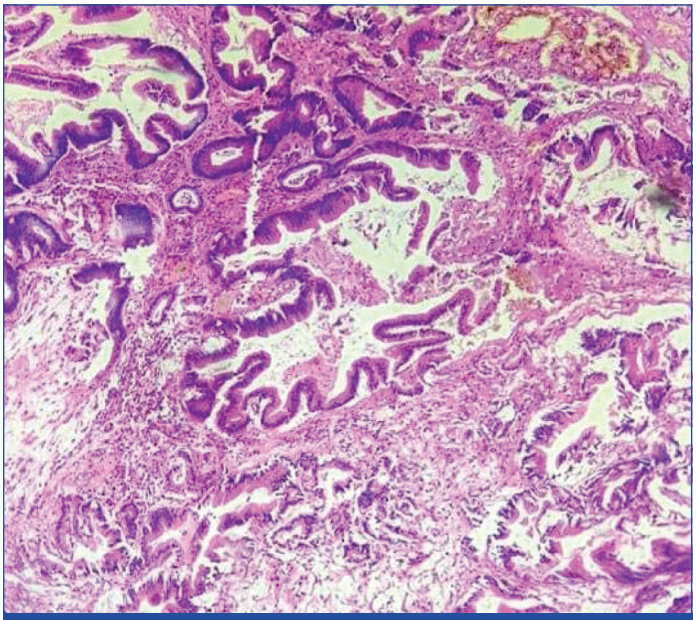
After that the surgically excised tissue was sent for routine histopathological examination. Grossly, multiple irregular greyish brown soft-tissue pieces were received, measuring altogether 6×5.5×1.0 cm.

Microscopy revealed a tumour with well-differentiated tubulopapillary architecture. These tumour cells showed atypical features e.g., nuclear pleomorphism, hyperchromasia, and irregular nuclear membrane. A dense acute inflammatory infiltrate was also seen. Two differential diagnoses were made. These were Metastatic GI adenocarcinoma and sinonasal low-grade ITAC [Table/Fig-3,4].

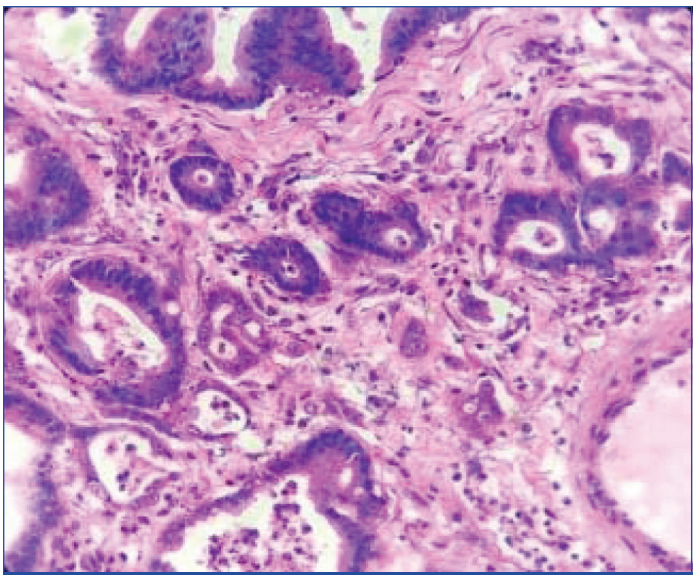
For confirmation, IHC for CK20, CDX2, EMA and GFAP were applied: Tumour cells showed Strong positive immunoreactivity to CK-20, CDX2, EMA, while negative for GFAP. So primary glial



[Table/Fig-2]: MRI scan image: Postoperative coronal view.



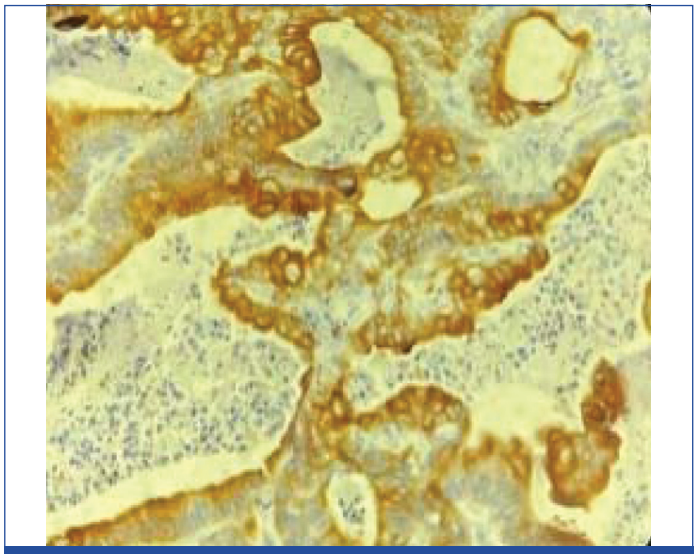
[Table/Fig-3]: H&E stained section (100x magnification)- tumour with well-differentiated tubulopapillary architecture.



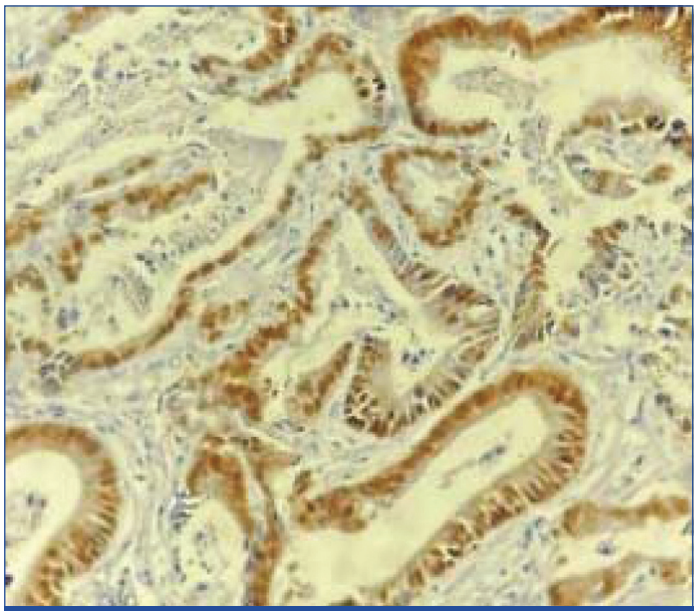
[Table/Fig-4]: H&E stained section (400x magnification)- high magnification image showed tumour cells with nuclear pleomorphism, hyperchromasia, and irregular nuclear membrane.

tumour was ruled out. Final diagnosis of well-differentiated sinonasal adenocarcinoma intestinal type had been made [Table/Fig-5-8].

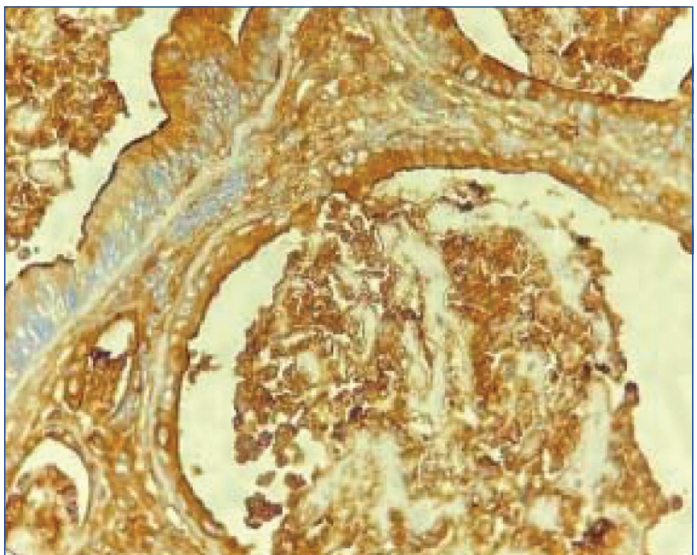
On histopathology, two differential diagnoses were given. Only after the IHC, a confirmatory diagnosis of sinonasal ITAC was made. Surgery and postoperative stay were uneventful. However, patient was lost to follow-up subsequently.



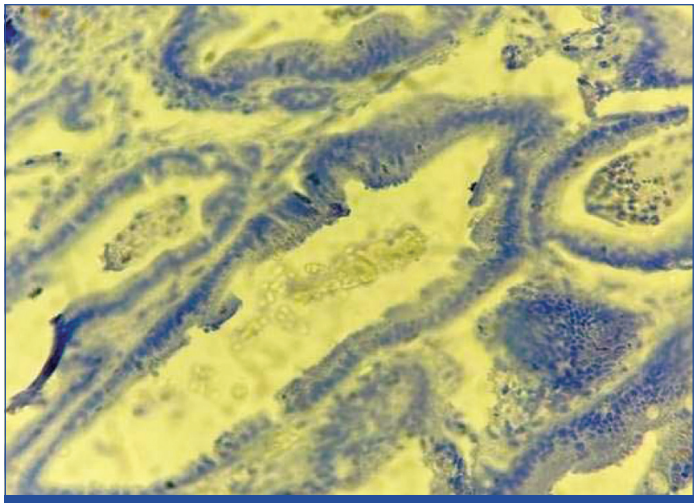
[Table/Fig-5]: IHC section- showed positive immunoreactivity of tumour cells for CK20.



[Table/Fig-6]: IHC section- showed positive immunoreactivity of tumour cells for CDX2.



[Table/Fig-7]: IHC section- showed positive immunoreactivity of tumour cells for EMA.



[Table/Fig-8]: IHC section- showed negative immunoreactivity of tumour cells for GFAP.

DISCUSSION

Most of the adenocarcinomas that occur in the head and neck region originate from minor or major salivary glands. Sinonasal adenocarcinomas, however, are in the category of non-salivary tumours that originate from the surface epithelium of the nasal and paranasal sinuses. They are divided into intestinal or non-intestinal-type, depending on the resemblance to GI mucosa. The intestinal type is histologically composed of columnar, goblet and mucus-producing cells which are usually indistinguishable from those of typical adenocarcinomas of the GI tract [1].

Adenocarcinomas account for 10-20% of all primary malignant tumour of the nasal cavity and paranasal sinuses. Sinonasal ITAC is a rare neoplasm (almost 4%). The overall incidence of ITAC is less than 1% of all neoplasms, however ITAC constitutes almost 4% of malignancies in the sinonasal area [2,3].

The ITAC occur primarily in the fifth to sixth decades of life, with a slight male preponderance and a strong association with occupational and environmental carcinogens such as wood and leather dust. Exposure to wood dust has been reported to increase the risk of adenocarcinoma 900-fold [4,5]. The common presenting symptoms of sinonasal ITAC are nasal obstruction and epistaxis. The tumour tends to be very locally aggressive but rarely metastasizes. Frozen section in this case showed occasional nuclear pleomorphism and hyperchromasia, but not confirmative evidence, because of similar artefacts microscopically. Diagnosis with histological confirmation is important because it is frequently misdiagnosed as a salivary gland tumour [6].

We hereby report a case of sinonasal ITAC which was clinically diagnosed as CNS tumour. Adenocarcinoma is a very rare tumour of the nasal cavity, often related to professional exposure to wood dust. This is a tumour with histological features resembling colorectal adenocarcinoma. Epistaxis, unilateral nasal obstruction and rhinorrhoea are the most common presenting symptoms of ITAC [7].

The prognosis for ITAC is poor. Recurrences and subsequent deeply invasive local growth are frequent; however, lymph node and distant metastases are rare [7]. According to Cardesa A and Slootweg PJ the most common location is ethmoid region [8].

Sinonasal tract adenocarcinomas originate from respiratory surface epithelium or underlying seromucinous glands. These adenocarcinomas are divided into salivary type and non-salivary type. Non-salivary type adenocarcinomas are further divided into intestinal type (ITAC) or non-intestinal type adenocarcinomas [9].

In the present case, tumour was spread to maxillary sinus, right ethmoidal, right sphenoidal sinus extending to intracranial region in the frontal lobe. Differential diagnosis of ITAC includes low grade sinonasal papillary adenocarcinoma, nasopharynx adenocarcinoma, and metastatic intestinal adenocarcinomas [10]. To diagnose these tumours, IHC plays an important role. In this case, the glial tumour was excluded by negative immunoreactivity of GFAP. While ITACs are usually positive for CK20, CDX2, Villin and MUC [4,9,10].

In this case strong positivity to CK-20, CDX-2, EMA favoured sinonasal adenocarcinoma intestinal type. Metastatic adenocarcinoma was excluded by clinical and radiological examination of intestines, because usually both are immunopositive for CK20 and CDX2 [11]. It is preferred to do CK-7 marker in this case to rule out metastatic GI/colorectal adenocarcinoma, however, as it was not available at that time, it was not performed.

Complete surgical resection with wide margins with radiotherapy is the treatment of choice. This case report, when compared with other similar case reports [3,12,13], showed similar tumour extension to frontal regions and presented with similar symptoms to this case [Table/Fig-9].

Reference	Age/Sex	Origin	Extension to the brain	Symptoms	Imaging	Surgery
Current case	50/F	Right maxillary sinus	Extend upto the frontal region with perifocal edema	Headache, vomiting and bilateral nasal bleeding	Well defined mass involving maxillary, ethmoid. sinus extending upto frontal intracranial region	Bifrontal craniotomy
Sklar EM, and Pizarro JA [3]	68/M	Ethmoid sinuses	High signal frontal mass	Confusion and weight loss for 3 months	Frontal lesion hyper intense on magnetic resonance imaging	Bifrontal craniotomy, orbital osteotomy, tumour resection
Espitalier F et al., [12]	55/M	Skull base	None	Carpenter with rapid left visual loss	Skull base and meningeal invasion	Resection was performed on a transfacial approach
Robles C and Cooper EM [13]	68/M	Right paranasal sinuses	Extension to both frontal lobes with perilesional oedema	Memory loss, confusion and headache for 6 months	Large mass involving the right nasal cavity, paranasal sinuses with brain extension	Not specified

[Table/Fig-9]: Comparison of previous sinonasal intestinal type with current case study [3,12,13].

CONCLUSION(S)

This tumour is frequently misdiagnosed clinically as a salivary gland tumour or CNS tumour. IHC is important to rule out metastasis. Diagnosis of this case highlights the importance of histopathology with IHC, which confirmed the diagnosis of sinonasal ITAC.

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PARTICULARS OF CONTRIBUTORS:
1. Senior Resident, Department of Pathology, Uttar Pradesh University of Medical Sciences Saifai, Etawah, Uttar Pradesh, India.
2. Professor, Department of Pathology, Uttar Pradesh University of Medical Sciences Saifai, Etawah, Uttar Pradesh, India.
3. Assistant Professor, Department of Pathology, Uttar Pradesh University of Medical Sciences Saifai, Etawah, Uttar Pradesh, India.
4. Associate Professor, Department of Pathology, Uttar Pradesh University of Medical Sciences Saifai, Etawah, Uttar Pradesh, India.
5. Associate Professor, Department of Pathology, Government Medical College, Kannauj, Uttar Pradesh, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Sanjeev Kumar Singh,
Professor, Department of Pathology, UPUMS, Saifai, Etawah-206130,
Uttar Pradesh, India.
E-mail: drsanjeev.rml@gmail.com

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