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Sinonasal Intestinal Type Adenocarcinoma Involving the Paranasal Sinuses: A Rare Case Report

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Abstract

Sinonasal intestinal-type adenocarcinomas (ITAC) are very uncommon, with an overall incidence of <1 case per 1 million person-years and prevalent among people who work for prolonged periods in wood or leather industries. Here, we present a rare case of sinonasal intestinal-type adenocarcinoma in a 38yr old male.

Keywords: Sinonasal intestinal-type adenocarcinoma (ITAC), nasal cavity

Introduction

Sinonasal intestinal type adenocarcinomas (ITAC) are very rare, where men are most likely to develop ITAC in sixth to seventh decades of life. The preferential site of origin is commonly identified in the nasoethmoidal complex. The incidence of these tumours varies and is prevalent among people who work for prolonged periods in wood or leather working industries. The exposure to the wood dust increases the risk of adenocarcinoma by 900 times. Adenocarcinoma in sinonasal tract include salivary and non salivary type which is further subclassified into intestinal and non-intestinal type.

Sinonasal intestinal-type adenocarcinoma (ITAC) is morphologically similar to adenocarcinomas of intestinal origin and is characterized by the immunohistochemical expression of intestinal markers, such as cytokeratin (CK) 20, CDX2, and villin. We present a case of sinonasal intestinal type adenocarcinoma presented with foul smelling bloody discharge and nasal obstruction in a young male.

Case Report

A 38-year-male patient presented with bloody and foul smelling left nasal discharge, associated with nasal obstruction since 02 months. On examination a polypoidal mass was seen in left nasal cavity arising from middle meatus which bleeds on touch. Tenderness was present in the Ethmoidal and paranasal sinus. All the relevant haematological, biochemical investigations and chest X-ray

were within normal limits.

CT scan Paranasal sinus: Isodense soft tissue density noted in the left frontal, maxillary, ethmoid, sphenoid sinuses extending into the nasal cavity on left side. Adjacent bony thinning noted [Fig.1]. A provisional clinical diagnosis of sinonasal polyposis was made and transmural biopsy was done.

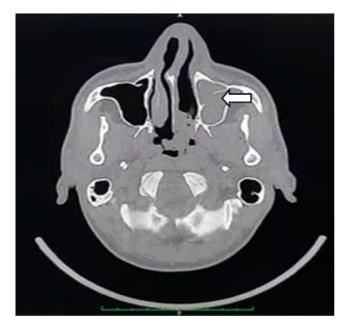


Figure 1: CT Paranasal sinus image showing isodense soft tissue density (arrow) extending into the nasal cavity on left side with adjacent bony thinning.

Grossly specimen consisted of single gray white soft-tissue

bit measuring 1.5x1x0.5 cm. Microscopy showed tubulopapillary structure lined by cuboidal to low columnar cells with stratification having apical mucin and basally placed large pleomorphic hyperchromatic nuclei [Fig.2]. The intervening stroma showed pools of mucoid areas in which numerous signet ring cells were seen [Fig.3]. On Immunohistochemistry, CK7 [Fig.4] and MUC2 [Fig.5] markers were done for confirmation and a final diagnosis of Intestinal Type Sinonasal Adenocarcinoma (ITAC) was rendered. Patient refused for any treatment, got discharged against medical advice and lost for follow-up.

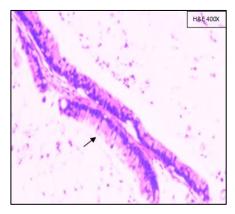


Figure 2: Microphotograph: showing cuboidal to low columnar cells with stratification having apical mucin and basally placed large pleomorphic hyperchromatic nuclei (arrow)

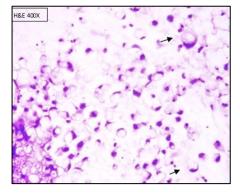


Figure 3: Microphotograph: The intervening stroma shows pools of mucoid areas in which are seen numerous signet ring cells (arrow)

Discussion

Sinonasal malignant tumour is an unusual tumour and account for 3% of all malignant tumours of head and neck which primarily occurs in men aged 55-60years of age.[1] The large majority of ITAC (88%) can be attributed to the occupational exposure, the most important being the exposure to the wood dust followed by products in the textile industry. The time between the first occupational

exposure to wood dust and the development of adenocarcinoma of the sinonasal tract averages 40 years. [1-2] Tumours related to occupational exposure affect men in 85-95% of cases, and show a strong tendency to arise in the ethmoid sinuses. The gross appearance of these tumours is similar to that of colonic Adenocarcinoma. The adenocarcinoma histologic types are tubulopapillary and intestinal. Squamous cell carcinoma is the most common type and most of them are high grade lesions Adenocarcinomas account for 10-20% of all malignancies

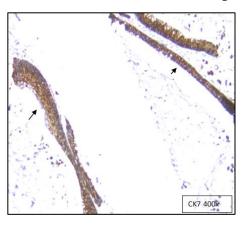


Figure 4: CK 7 marker showing strong cytoplasmic positivity for the glands.

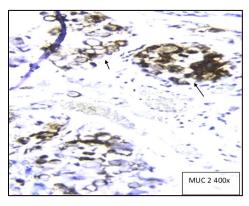


Figure 5: MUC2 marker showing strong Positive for mucin in signet ring cells.

of nasal cavity and paranasal sinuses. Adenocarcinomas are locally aggressive tumours with propensity to local recurrence despite well differentiated in nature. Lymph node metastasis is rare. [1-3]

The sites for ITAC are ethmoid sinus (40%), nasal cavity (25%), maxillary antrum (23%) and indeterminate (9%). Histological variants of ITAC are papillary, colonic, solid, mucinous and mixed. Papillary type has best prognosis as it behaves as a locally destructive lesion with limited tendency for metastasis. Solid and mucinous tumours are the most

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aggressive histological subtypes. Intracranial invasion has the worst prognosis and was found to be more frequent in mucinous subtype tumours. [1-3] The optimal treatment for sinonasal ITAC is complete resection with adjuvant radiation therapy to the region of the tumour. Regardless degree of differentiation, all forms of ITAC should be considered locally aggressive. [5-6]

Current advanced imaging techniques may be useful for evaluating the extent of the tumour and differential diagnosis preoperatively. The differential diagnosis of ITAC includes metastatic gastrointestinal carcinoma and sinonasal low grade non intestinal adenocarcinoma. Both ITACs and colorectal carcinomas express CK20, CDX-2, Villin and MUC2, but the expression of CK7 in a tumour may be suggestive of ITAC.[5]

Here, we present a case of sinonasal ITAC which was confirmed on immunohistochemical markers MUC2 and CK.

Conclusion

Sinonasal ITAC is a rare tumor with variable biologic behavior in relation to the histologic subtype. The case presented here is for its rarity and one should be very cautious of diagnosing a nasal polyp clinically especially in elderly male with occupational exposure of wood, nickel, and leather dust. Histopathological diagnosis is always gold standard.

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