

Rare Variety of Nasal Mass: Case Report

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ABSTRACT

Seromucinous hamartoma (SMH) is a rare benign glandular lesion of the sinonasal tract that can clinically and histologically mimic more aggressive neoplasms. We report a case of SMH in a 32-year-old male presenting with unilateral nasal obstruction and snoring, that was initially misdiagnosed as antrochoanal polyp. The lesion was excised endoscopically, and histopathology confirmed the diagnosis. There has been no recurrence at 3 months. This report highlights the importance of considering seromucinous hamartoma in the differential diagnosis of unilateral nasal masses.

Keywords: Hamartoma, Nasal Cavity, Nasal Obstruction, Endoscopy, Snoring, Antrochoanal Polyp

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INTRODUCTION

Hamartoma has been classified as a neoplasm comprised of disorganized tissue native to a particular anatomical location. It may occur sporadically or occasionally as part of a syndrome. Most hamartomas are usually benign, but malignant transformation may occur. They have been typically described in the lungs, hypothalamus, breast, and colon, although they may grow almost anywhere else.¹ In the sinonasal tract, they are classified into four distinct histological types: respiratory epithelial adenomatoid hamartoma (REAH), seromucinous hamartoma (SMH), chondro-osseous and respiratory hamartoma (CORE), and nasal chondromesenchymal hamartoma (NCH).² SMH is characterized by proliferation of seromucinous glands and ducts. Although it was first described by Baillie and Batsakis in 1974, fewer than 60 cases have been reported in the literature.³ There is only one case reported from India.⁴ Due to its rarity and overlapping features with low-grade sinonasal adenocarcinoma and respiratory epithelial adenomatoid hamartoma, accurate histopathological diagnosis is essential. Clinical features are typical as that of any nasal mass. Diagnostic nasal endoscopy (DNE) and imaging would

be the pre-requisites; the former being useful to locate the area of origin of the growth.

The present case was misdiagnosed elsewhere as an antrochoanal polyp on imaging without a DNE. We report its correct management and follow-up as an addition to the literature on this rare entity.

CASE REPORT

A 32-year-old male presented with a one-year history of right-sided nasal obstruction, which had worsened over the past two months. He also reported snoring with associated apnoeic episodes during sleep and intermittent symptoms suggestive of allergic rhinitis. There was no history of epistaxis, facial pain, anosmia, or prior nasal surgery. He was a known case of type 2 diabetes mellitus with blood sugar levels well controlled on medication. For the symptoms, he consulted at an ENT facility, where he was advised to undergo imaging. (computed tomography of the paranasal sinuses). The images showed mucosal thickening of bilateral ethmoid sinuses with a right-sided nasal cavity mass without evidence of bony erosion. (**Figure 1**) This was reported as “antrochoanal polyp”, and referred to our



Figure 1: Selected images from coronal sections of CT scan of nose and PNS of the patient

centre for surgical management.

Here, an initial anterior rhinoscopy revealed a deviated nasal septum to the right and a polypoidal mass in the right nasal cavity. Diagnostic nasal endoscopy was done. This revealed a polypoidal mass arising from the right inferior meatus extending up to the choana (**Figure 2**). This finding raised doubts about the diagnosis of an antrochoanal polyp. He was administered the Epworth Sleepiness Scale (ESS), on which he scored 5.

The patient underwent endoscopic excision of the mass under general anesthesia. Procedure was uneventful. The mass was noted arising from the junction of the anterior one-third and posterior two-thirds of the floor of nose and removed in toto with cauterization of base, and sent for histopathologic examination. (**Figure 3 and 4**) The postoperative period was uneventful. After an immediate (1 week) post-operative follow-up, the patient could review with us only after 2 months due to his job schedule. At this time, he reported relief of

nasal obstruction. ESS was administered; score had improved to 2. The repeat DNE showed healing at the site of origin. (**Figure 6**) He is free of recurrence at 3 months post-op.

Histopathological Findings

Sections revealed a polyp lined by pseudostratified columnar epithelium with areas of squamous metaplasia. The subepithelial stroma showed sero-mucinous glands and glands lined by respiratory epithelium in a fibrotic stroma with congested blood vessels and moderate lymphoplasmacytic infiltration, with large cystic spaces lined by cuboidal epithelium. There was no granuloma / cellular atypia / organism. Microscopic examination revealed a polypoid lesion lined by respiratory epithelium. The subepithelium showed lobular proliferation of small seromucinous glands and ducts lined by bland cuboidal cells with pale cytoplasm, embedded in a fibrous stroma (**Figure 5**). No cellular atypia, mitotic activity, or invasive

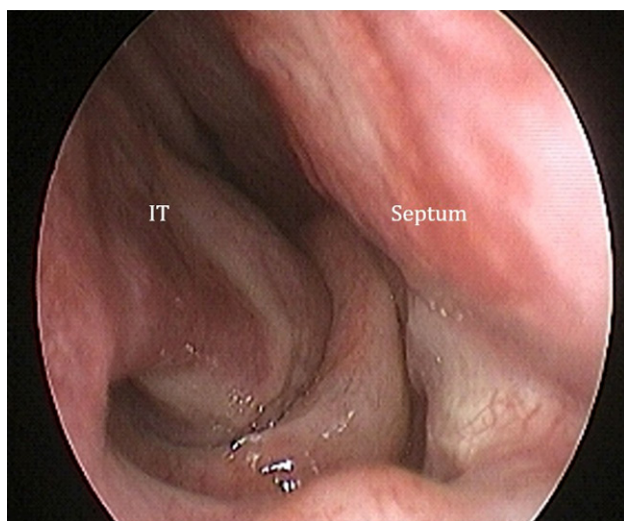


Figure 2: Screen capture image from the video of initial DNE of the patient



Figure 3: Gross appearance of the excised mass



Figure 4: Gross appearance of the specimen after histopathologic fixing

growth was noted. These findings were consistent with seromucinous hamartoma.

DISCUSSION

Seromucinous hamartoma (SMH) typically presents as a unilateral nasal mass causing obstruction or epistaxis. In the present case, snoring was also a feature, which was the reason for initial presentation to the ENT doctor. An article by C. Georgalas has detailed the association.⁵ Another recent study cites the bilateral relationship between nasal symptoms and snoring.⁶ Khan et al has reported a couple of cases of incidental diagnosis of SMH, following imaging for other indications.⁷

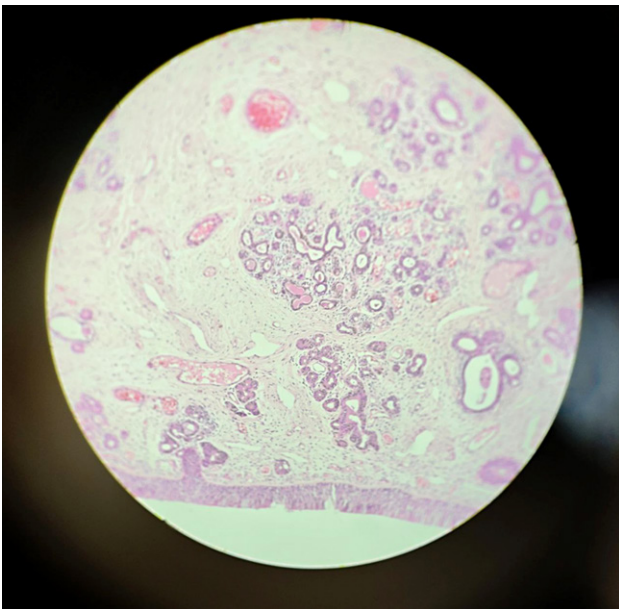


Figure 5: Image from the histopathological slides using hematoxylin-eosin (H&E) with 4x magnification

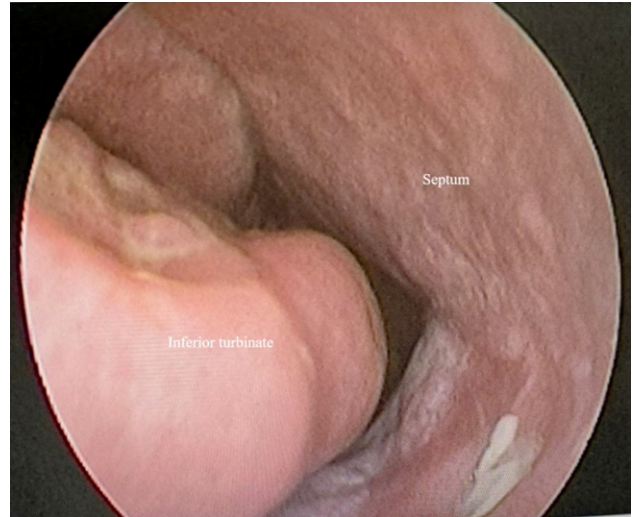


Figure 6: Screen capture image from the video of post-op. DNE of the patient

Diagnostic nasal endoscopy (DNE) is a “gold standard” modality to examine the entire nasal cavity and nasopharynx, especially in the case of a mass lesion. In the latter case, it aids in visual impression of the type of mass, locating its attachment, and if needed, taking a punch biopsy. As such, it is generally advisable to perform this before imaging. Several studies have reported the importance of DNE, including the specific instances where it would be more relevant than imaging.⁸⁻¹⁰ In the present case, imaging (CT PNS) being done prior to DNE, had led to an incorrect diagnosis.

Most lesions have been reported to arise from the posterior nasal cavity. An article reporting from a single centre series of six cases has cited location on the nasal septum, inferior turbinate and sphenoid sinus, with one being bilateral.¹¹ Yet another article has reported a single case of bilateral involvement.¹² In our case, the mass was noted arising from the junction of the anterior one-third and posterior two-thirds of the floor of right nasal cavity. All reports conclude that simple surgical excision (endoscopic) is curative, with recurrence being very rare. One amongst the case series reportedly required medial maxillectomy due to difficulty distinguishing the lesion from adenocarcinoma.¹¹ Histologically, SMH lacks myoepithelial cells and may show focal overlap with respiratory epithelial adenomatoid hamartoma. In their clinicopathological study, Weinreb et al. analyzed the features of seven seromucinous hamartomas. The key pathological finding was that this sinonasal glandular

lesion is characterized by a lack of myoepithelial cells.¹³ The present case displayed similar characteristics, confirming the diagnosis of this rare entity.

CONCLUSION

This case report adds to the existing literature of a rare nasal neoplasm, and shows the importance of diagnostic nasal endoscopy as a primary mode of evaluation of nasal masses.

END NOTE

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Conflict of Interest: None declared

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