

CASE REPORT

Nasal hamartomas: a case series and literature review

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ABSTRACT

BACKGROUND. Nasal and nasopharyngeal hamartomas are rare, benign developmental lesions that often mimic other sinonasal pathologies. Misdiagnosis may lead to unnecessary interventions.

OBJECTIVE. To present a case series involving three histological subtypes of nasal hamartomas and to review the current understanding regarding their classification, diagnosis, and management.

MATERIAL AND METHODS. We describe three patients treated at our institution, each with a distinct subtype of nasal hamartoma: respiratory epithelial adenomatoid hamartoma (REAH), seromucinous hamartoma (SH), and nasal chondromesenchymal hamartoma (NCMH). Clinical, radiological, surgical, and histological data were analyzed.

RESULTS. All patients underwent successful endoscopic surgical excision. Postoperative outcomes were favorable with no recurrence during follow-up. Histological analysis confirmed the diagnosis in each case.

CONCLUSION. Nasal hamartomas, though rare, should be included in the differential diagnosis of sinonasal masses. Accurate diagnosis relies on histopathology, and complete surgical excision leads to excellent prognosis.

KEYWORDS: nasal hamartoma, respiratory epithelial adenomatoid hamartoma, seromucinous hamartoma, chondromesenchymal hamartoma, endoscopic surgery, sinonasal tumors.

INTRODUCTION

Hamartomas are non-neoplastic malformations characterized by a disorganized proliferation of native tissue components. In the sinonasal region, these lesions are rare and may be misdiagnosed as more common pathologies such as nasal polyps, papillomas, or malignancies. Accurate recognition is crucial to avoid overtreatment and unnecessary alarm.

Nasal hamartomas are classified based on their distinct histopathological characteristics¹. One subtype, Respiratory Epithelial Adenomatoid Hamartoma (REAH), is defined by the presence of proliferative invaginations of ciliated respiratory epithelium, often accompanied by a distinctive pattern of hyalinization around the glandular structures. Another variant, known as Seromucinous Hamartoma (SH), is composed of benign, irregularly distributed seromucinous glands and ducts embedded within the submucosal tissue. These structures lack atypia and exhibit a disorganized architectural pattern. A third and notably rarer type is the Nasal Chondromesenchymal Hamartoma (NCMH), which is primarily seen in pediatric patients. This form is characterized by the presence of various mesenchymal components, including immature cartilage

and bone, within a myxoid stroma. Importantly, NCMH has been associated with mutations in the *DICER1* gene, suggesting a potential genetic underpinning in its pathogenesis.

This paper presents three cases of nasal hamartomas, each illustrating a different histological subtype, and provides a concise review of the literature.

CASE SERIES

Case 1

A 27-year-old man presented with a long-standing complaint of a blocked left nostril and a slightly reduced sense of smell. He did not report any other symptoms, and his personal and family medical history did not reveal anything significant. During the clinical examination, we observed a pale growth obstructing his left nasal fossa. A CT scan of the nose and paranasal sinuses without contrast confirmed the presence of a soft tissue density lesion in the left nasal cavity, measuring approximately 2 to 3 cm in size (see Figure 1).

The patient underwent surgery using an endoscopic approach, which allowed us to completely remove the mass. The

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Received for publication: July 16, 2025 / **Accepted:** August 29, 2025

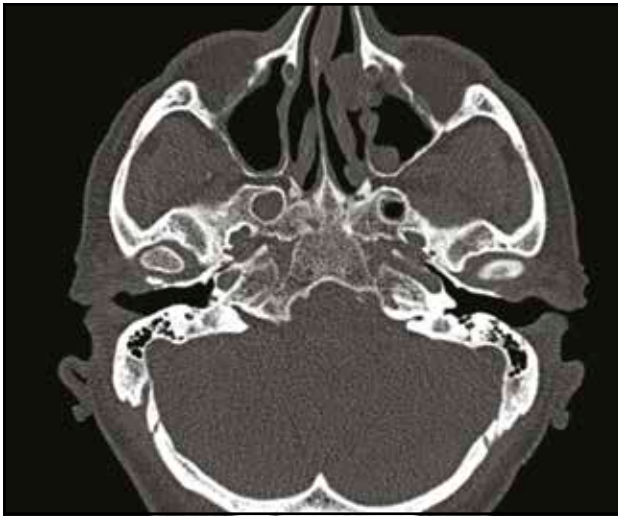


Figure 1. Axial craniofacial CT scan image showing a mass in the left nasal cavity.

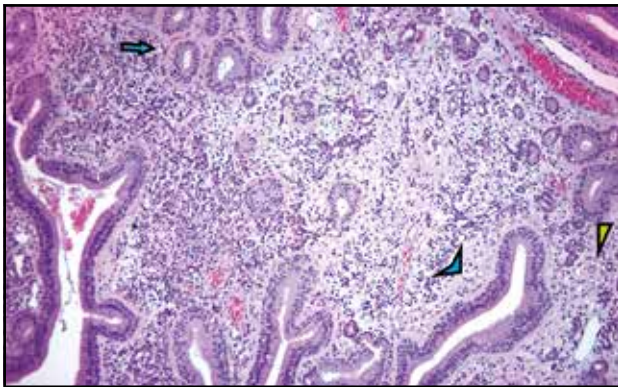


Figure 3. Histological section of REAH (high magnification, Ob10X; Hematoxylin-Eosin staining): round to oval glands, composed of ciliated epithelium, identical to the ciliated surface epithelium (small blue arrow and yellow arrow); submucosal glands enveloped by eosinophilic basement membrane material of variable thickness (hyaline) (long blue arrow).

postoperative course was uneventful.

Histology confirmed respiratory epithelial adenomatoid hamartoma (REAH), showing glandular proliferation lined by respiratory epithelium. Figure 2 presents a low magnification (Ob4X) of the histological section, showing a glandular proliferation in which the glands have a branched pattern of growth and are in direct continuity with the surface epithelium. Normal seromucous glands are found within the glandular proliferation. In Figure 3, at a higher magnification (Ob10X), we can see round to oval glands, composed of ciliated epithelium, identical to the ciliated surface epithelium. The submucosal glands are enveloped by eosinophilic basement membrane material of variable thickness (hyaline). The stroma is edematous, containing a mixed chronic inflammatory cell infiltrate.

Case 2

Our second case involves a 36-year-old man who came for a consultation after a mass was discovered in his sphenothmoidal recess. Interestingly, the mass was found incidentally during an MRI scan

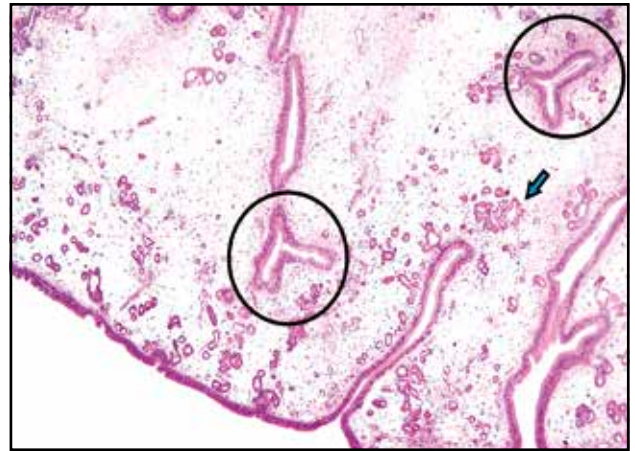


Figure 2. Histological section of REAH (low magnification, Ob4X; Hematoxylin-Eosin staining) showing a glandular proliferation in which the glands have a branched pattern of growth (black circles), with normal seromucous glands within the glandular proliferation (blue arrow).

that had been ordered to evaluate his vertiginous syndrome. The craniofacial MRI shows a well-defined, T2-hyperintense lesion in the left sphenothmoidal recess (see Figure 4). There is evidence of mild displacement of surrounding structures, including medial bowing of adjacent ethmoid walls and narrowing of the posterior nasal airway, though without frank bone destruction.

In Figure 5, we present an image of the lesion as seen on a craniofacial CT scan without contrast in the coronal plane. The CT scan was performed to evaluate potential bony involvement; however, no osseous abnormalities were identified.

The patient had undergone surgery a few years earlier for a sino-nasal polyp on the left side, thus explaining the enlargement of the maxillary sinus ostium on the same side. During the endoscopic examination, we observed a polypoid mass in the left nasal cavity, completely blocking the left sphenothmoidal recess. Figure 6 shows the endoscopic image of a pale pink tumoral mass with a slightly irregular surface, completely occupying the sphenothmoidal recess.

The mass was successfully removed using an endoscopic approach under general anesthesia. Surgical excision was complete.

Histopathology showed small seromucinous glands in clusters and lobules, consistent with seromucinous hamartoma (SH). Figure 7 shows the image of the histopathological section under low magnification, illustrating a submucosal epithelial proliferation of small glands, serous acini, and tubules growing in clusters and lobules, alternating with larger glands and cysts. In Figure 8, the same specimen is presented under high magnification: the glands are lined by low cuboidal or flat epithelium, with round and oval nuclei and a variable amount of eosinophilic cytoplasm; the glands contain eosinophilic secretions. There is periglandular hyalinization and large, thick vessels present in the stroma.

Case 3

A 30-year-old male reported right-sided tinnitus. Endoscopy found a lobulated mass blocking the left choana (see Figure 9). The patient performed a craniofacial CT scan without contrast, which revealed a mixed-density mass in the nasopharynx, occu-

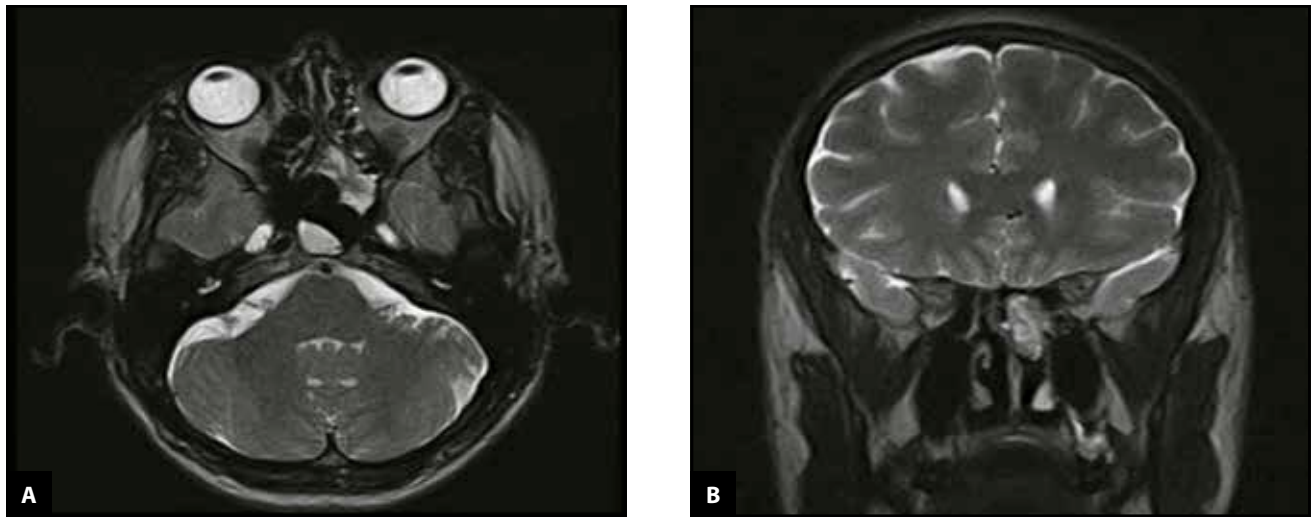


Figure 4. Craniofacial MRI, axial and coronal slices, showing a gadolinium-enhancing lesion in the left sphenothmoidal recess.



Figure 5. Non-contrast craniofacial CT scan in coronal plane showing an opacification of the left sphenothmoidal recess.



Figure 6. Endoscopic image of the tumoral mass occupying the left sphenothmoidal recess.

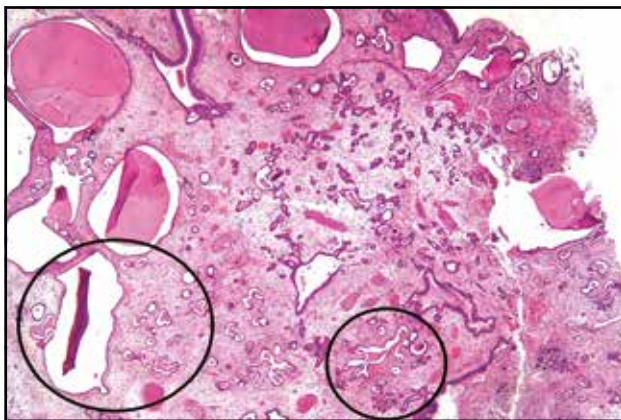


Figure 7. Histopathological image of seromucinous hamartoma (low magnification Ob4X, Hematoxylin-Eosin staining): submucosal epithelial proliferation of small glands, serous acini, and tubules growing in clusters and lobules, alternating with larger glands and cysts (black circles).

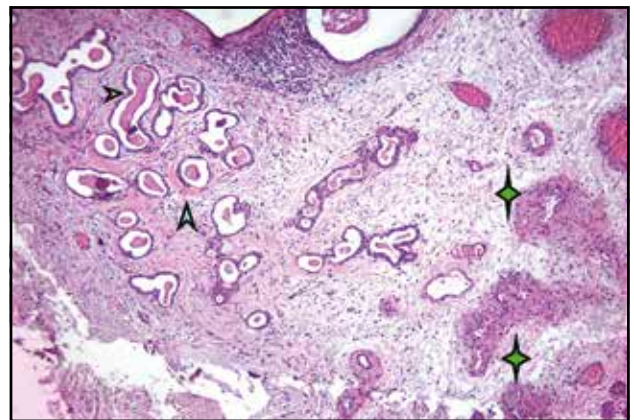


Figure 8. Histopathological image of seromucinous hamartoma (high magnification Ob10X, H&E): glands lined by low cuboidal or flat epithelium, containing eosinophilic secretions (blue arrows). There is periglandular hyalinization (blue arrows) and large, thick vessels present in the stroma (green stars).

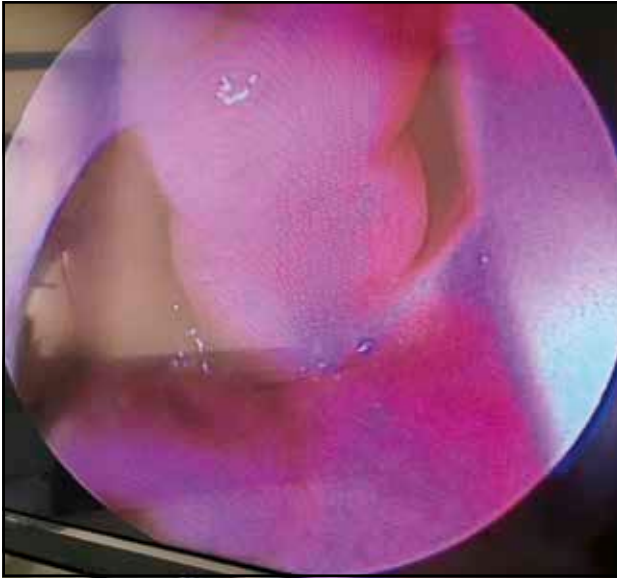


Figure 9. Endoscopic image of the rhinopharyngeal lesion, a lobulated mass occupying the left choana.

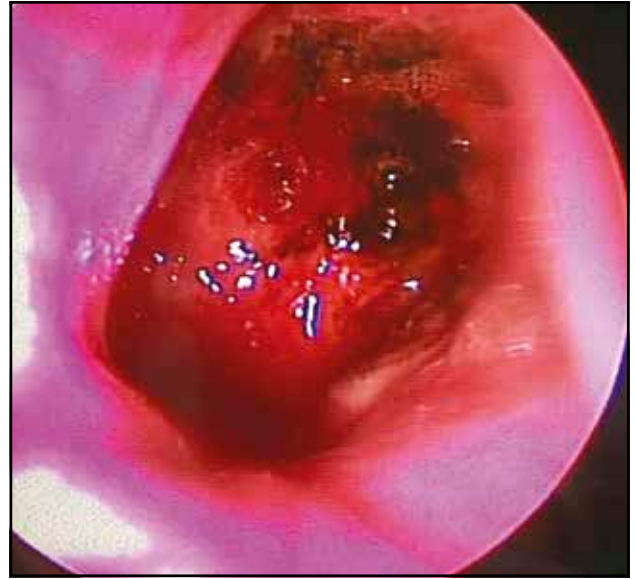


Figure 10. Endoscopic image of the rhinopharynx after removal of the lesion.

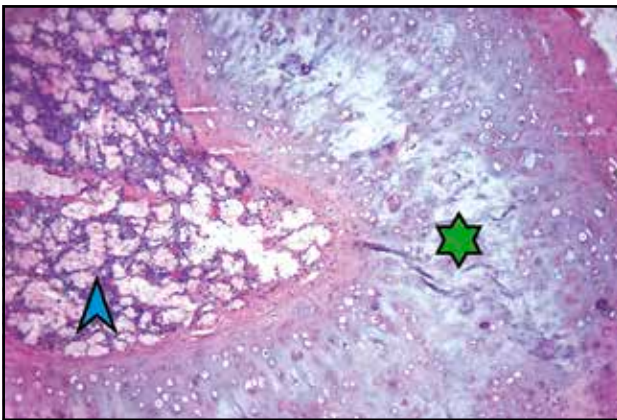


Figure 11. Histologic section showing mature cartilaginous tissue (green star) with admixed mature seromucous glands (blue arrow).

pying the left side of the nasopharynx.

Endoscopic removal showed a tumor with cystic and cartilaginous components. An image of the rhinopharynx after the endoscopic removal of the tumor is shown in Figure 10. Notice the broad-base attachment on the posterior and superior walls of the rhinopharynx.

Microscopy (Figure 11) confirmed mature cartilage and seromucinous glands, the aspect being pathognomonic for the diagnosis of nasal chondromesenchymal hamartoma (NCMH).

DISCUSSIONS

Respiratory epithelial adenomatoid hamartomas (REAHs) are benign acquired non-neoplastic overgrowths of indigenous glands of the nasal cavity, paranasal sinuses, and nasopharynx, arising from the surface epithelium and devoid of ectodermal, neuroectodermal

and mesodermal elements. The morphological characteristics are defined by a prominent proliferation and accumulation of glandular structures within the submucosa. These glands are consistently lined by pseudostratified ciliated columnar epithelial cells, resembling normal respiratory epithelium. Notably, the epithelial lining exhibits no signs of cytological atypia, dysplasia, or metaplastic alteration, supporting the benign nature of the process. The overall architecture remains orderly, and there is no disruption of the surrounding tissue planes². Differential diagnosis is essential and it should consider excluding inverted papillomas, nasal polyps, and adenomas³. Sinonasal papillomas of the inverted type are usually composed of significantly thickened epithelium (squamous compared to normal respiratory epithelium), with associated microcysts, scattered mucocytes and inflammatory cells. In addition, inflammatory polyps are characterized by a polypoid configuration with marked stromal edema, abundant fibroblastic proliferation, and prominent vascular congestion or neovascularization. These histopathological features help distinguish inflammatory polyps from hamartomatous lesions, which typically exhibit a more organized glandular architecture, lack significant edema, and do not demonstrate the same degree of stromal cellularity or vascular proliferation. The most important differential diagnosis is sinonasal adenocarcinoma. The characteristic ciliated respiratory epithelial proliferative component helps distinguish REAH from a malignant tumor. Nonetheless, careful histopathological examination is required of the whole specimen, as there are studies that show that this kind of lesion could be a precursor to a sinonasal adenocarcinoma⁴.

Seromucinous hamartomas (SHs) are benign acquired non-neoplastic overgrowths of indigenous glands of the sinonasal tract, and rarely of the nasopharynx, arising from submucosally situated seromucinous glands. SH was initially described as a polypoid mass of respiratory-epithelium-lined tissue with submucosal seromucinous glands in a lobular arrangement and a fibrous background⁵. Later reports highlighted additional features, such as invaginated

respiratory epithelium-lined gland-like structures in a haphazard arrangement; squamous metaplasia; dense lymphoplasmacytic infiltrate; and dense cytoplasmic eosinophilic granules. The stroma is edematous to fibrotic, with patchy chronic inflammation. No atypia, no mitotic activity, and no epithelial proliferation are seen. Mucinous cells are not a common feature despite the name “seromucinous hamartoma”. The nuclei are small and there is no evident mitotic activity or necrosis. Differential diagnosis⁶ is essential, and the most significant entity is sinonasal adenocarcinoma, low-grade, non-intestinal, non-salivary gland type. This kind of malignancy shows a complex glandular growth characterized by cribriform glands, composed of medium-sized cells with nuclear pleomorphism, mitotic activity and tumor necrosis.

Nasopharyngeal hamartomas are rare entities, and they are usually studied together with nasal hamartomas, REAHs and SHs. They usually contain mesenchymal elements such as cartilage or bone^{7,8}. Chondro-osseous and respiratory epithelial (CORE) hamartomas are related to REAHs, but they have additional features of chondroid tissue. They usually present as polypoid lesions, most commonly affecting the nasal cavity, but the nasopharynx, ethmoid sinus and sphenoid sinus may be a presenting location. Microscopic presence of respiratory adenomatoid components tends to be reduced. It is important not to mistake immature-appearing mesenchyme or cartilage for sarcoma. Complete resection is curative, although there are rare recurrences^{9,10}.

Though uncommon, nasal hamartomas should be part of the differential when encountering unilateral nasal masses, especially when typical inflammatory features are absent. Clinical and imaging findings are non-specific, necessitating histopathological confirmation for a definitive diagnosis.

Histologically, REAHs and SHs may mimic low-grade adenocarcinomas or inflammatory polyps, while NCMH can be mistaken for sarcomas due to their cellular heterogeneity and mesenchymal components. Endoscopic surgery remains the gold standard for treatment.

There is no known risk of malignant transformation, and recurrence is rare if excision is complete. Awareness among otolaryngologists and pathologists is key to avoiding overtreatment.

CONCLUSIONS

Nasal hamartomas, though rare and often misdiagnosed, are important benign lesions that can be effectively managed with minimally invasive surgery. A multidisciplinary approach, incorporating endoscopy, imaging, and pathology, ensures optimal outcomes.

Contribution of authors: All authors equally contributed to this article.

Conflicts of interest: The author declares no conflicts of interest.

Funding: No external funding was received for this study.

Financial disclosure: None.

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