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# Chondro-Osseous Respiratory Epithelial Adenomatoid Hamartoma (COREAH) in the Sinonasal Tract: Case report and review of the literature

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## Abstract

Chondro-osseous respiratory epithelial adenomatoid hamartoma (COREAH) is a rare benign expanding lesion of the nose and paranasal sinuses. The authors report the case of a 54-year-old female patient who exhibited long-standing symptoms of nasal obstruction and headaches, with chronic abuse of vasoconstrictor sprays. Nasal endoscopic examination showed a yellow fleshy polypoid mass in the left sphenoethmoidal recess. CT imaging demonstrated complete opacification of the left sphenoid sinus and a polypoid formation containing small calcifications in the left superior meatus and sphenoethmoidal recess. The patient initially declined surgical intervention and was managed with topical corticosteroids. Eight years later, she returned to the ENT clinic with persistent symptoms and consented to surgical resection. Histopathological examination revealed respiratory epithelium on the surface with small glands imbricated with bony trabeculae in the stroma, therefore establishing the diagnosis of COREAH. The postoperative course was uneventful. The authors report the case and review the pertinent worldwide literature.

**Keywords**: Chondro-osseous respiratory epithelial adenomatoid hamartoma; COREAH;

Nasal hamartoma; Sinonasal tract; Nasal obstruction

## Introduction

Hamartomas are defined as benign tumors resulting from aberrant differentiation. They are characterized by a disorganized mass of mature specialized cells or tissues that are native to the site where they develop. Hamartomas of the sinonasal tract are very rare lesions, with Chondro-Osseous Respiratory Epithelial Adenomatoid Hamartoma (COREAH) representing a particularly rare subtype first described in 1995. In 2021, Yu Y et al. reported two cases of COREAH. Their review of literature at the time noted sixteen other cases **[1]**. Another two cases were described by Hachemi M et al. and Divya N et al. in 2024 respectively **[2,3]**. To our knowledge, this case presents the 21st documented case of COREAH published in the literature to this date.

## Case Presentation

A 54-year-old female patient consulted for bilateral nasal obstruction and chronic headaches. She used vasoconstrictor sprays daily. Her medical history was uneventful. She had no known allergies. She was a nonsmoker and had no notable history of alcohol misuse. Flexible nasoendoscopic examination revealed a yellow, fleshy polypoid mass in the left sphenoethmoidal recess **(Figure 1)**. Computed Tomography (CT) scan of the paranasal sinuses showed complete opacification of the left sphenoidal sinus that continued as a polypoid formation, with a calcified core, at the posterosuperior part of the left nasal cavity **(Figure 2)**. Endoscopic resection was recommended but the patient declined and was managed with corticosteroid nasal spray. Eight years later, she returned to the ENT clinic with persistent nasal obstruction and headaches. A new computed tomography scan demonstrated progression of the posterior sphenoethmoidal lesion, extending into the left nasal cavity reaching below the lower edge of the inferior turbinate **(Figure 3)**. The patient agreed to undergo functional endoscopic sinus surgery with complete polypectomy.

The patient was followed annually post-operatively for five years without recurrence endoscopically or radiologically **(Figure 4)**. Histopathological examination was notable for a polypoid lesion, superficially lined by respiratory-type epithelium. Within the stroma, small glands and invaginations of the surface respiratory epithelium with intricately interwoven bony trabeculae were noted. These trabeculae were occasionally located directly beneath the epithelial surface or in immediate proximity to the proliferating small glands. There was no evidence of dysplasia or cellular atypia. These features are consistent with COREAH **(Figure 5)**.

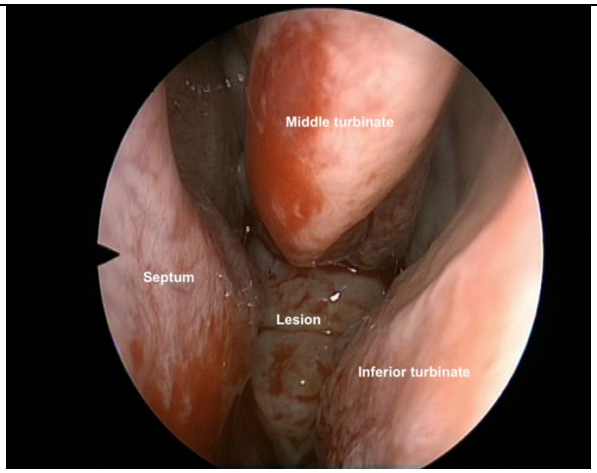


**Figure 1:** Nasal endoscopy of the left nasal fossa showing the yellow polypoid mass.

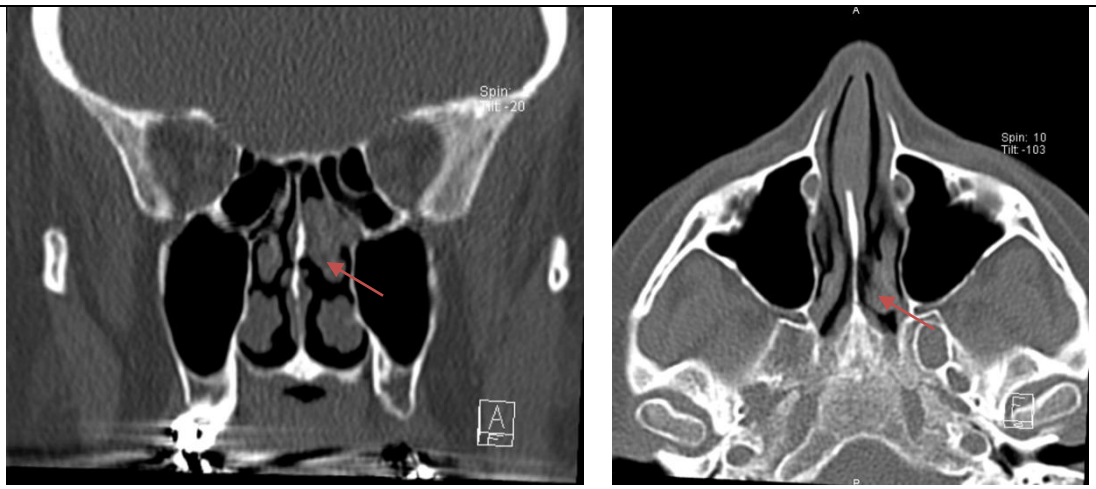


**Figure 2:** Initial coronal and axial CT (bone window) showing the presence of the spheno ethmoidal polyp (arrow).

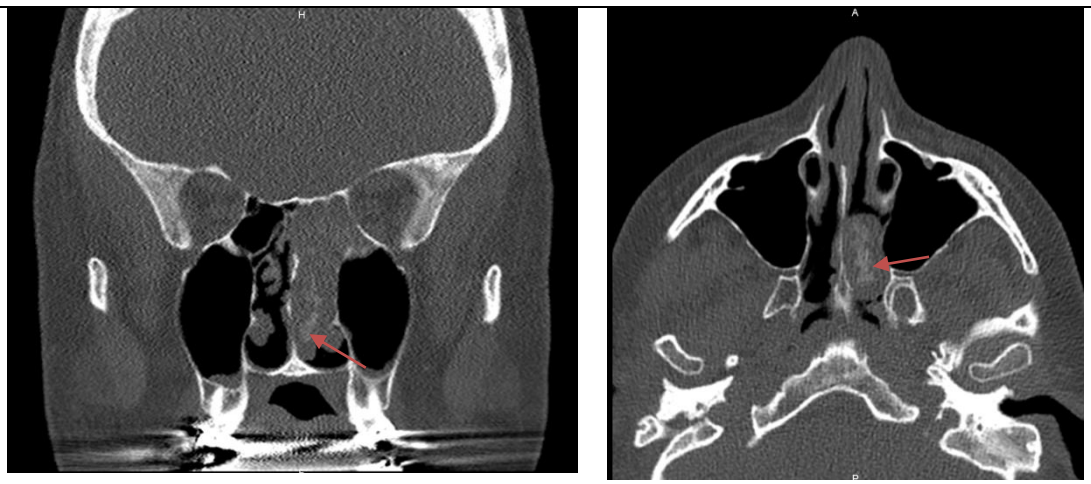

**Figure 3:** 8 years later coronal and axial CT (bone window) showing the progression of the mass reaching below the lower edge of the inferior turbinate. Note the presence of a calcified core in the mass (arrow).

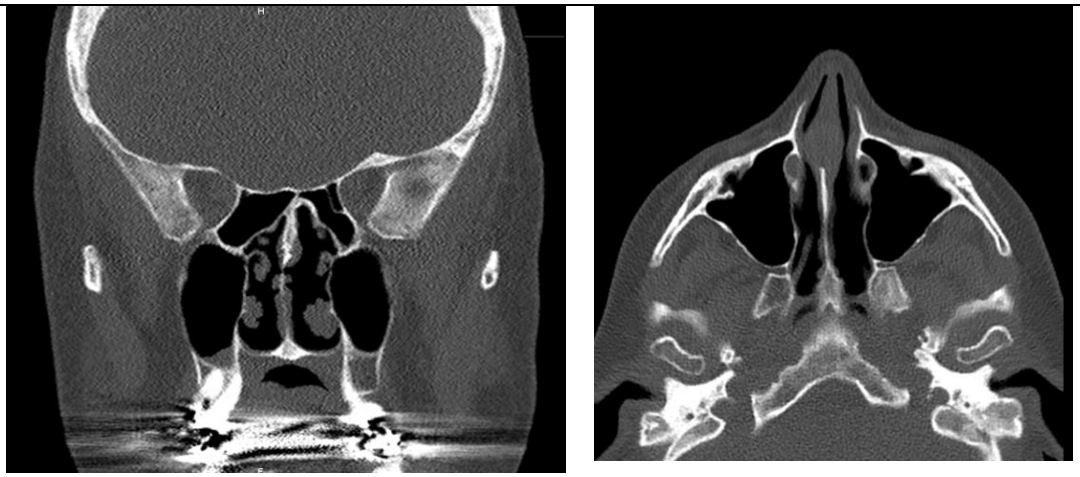

**Figure 4:** 5 year post operative coronal and axial CT (bone window) confirming the absence of recurrence of the hamartoma.

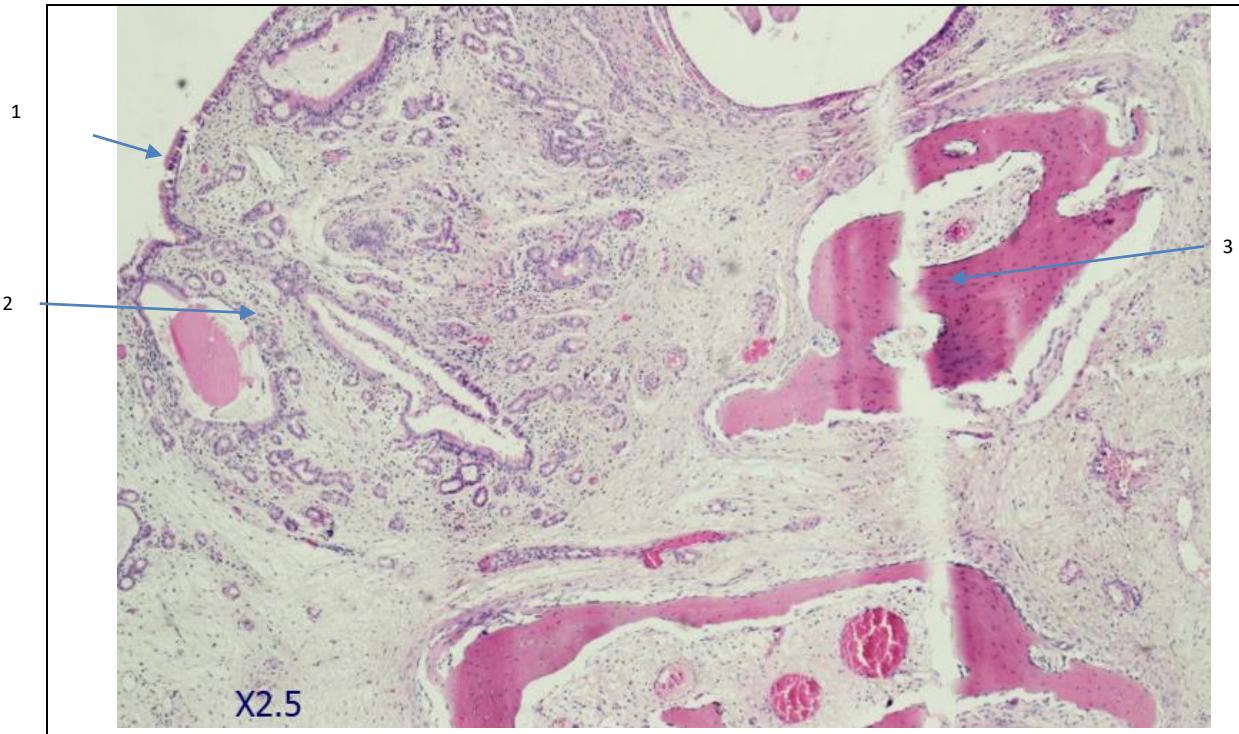


**Figure 5:** The lesion is lined superficially by a respiratory-type epithelium (1). The stroma contains small occasionally pseudo pyloric glands and invaginations of the surface respiratory epithelium (2), which were intricately interwoven with bony trabeculae (3). *Hematoxylin and eosin stain, magnification 2.5x*.

## Discussion

Head and neck hamartomas, including the sinonasal tract, are exceptionally rare. Nevertheless, their clinical relevance was recognized by the World Health Organization (WHO), which categorized them as a distinct entity in the revised 2017 classification of head and neck tumors [4]. In 1974, Baillie and Batsakis described the first seromucinous hamartoma (SH) [5]. In 1995, Wenig and Heffner were the first to describe respiratory epithelial adenomatoid hamartoma (REAH) [6]. A year later, Adair, Thompson,

Wenig, and Heffner presented a series of 13 REAHs, distinguished by the presence of both chondro-osseous and respiratory epithelial components. They introduced the term chondro-osseous respiratory epithelial adenomatoid hamartoma (COREAH) [7]. Finally, in 1998, McDermott et al. described Nasal chondromesenchymal hamartoma (NCMH) in a case series of 7 patients [8]. Consequently, there are four different subtypes of hamartomas: respiratory epithelial adenomatoid hamartoma (REAH), seromucinous hamartoma (SH), nasal chondromesenchymal hamartoma (NCMH), and chondro-osseous respiratory epithelial adenomatoid hamartoma (COREAH). The different characteristics of the four main subtypes of hamartomas are summarized in the following table **(Figure 6)**. COREAHs were initially thought to be more common in women. As more cases are described, sex predilection seems less likely. COREAH cases have been observed in patients from 3 to 83 years old. COREAHs are most commonly located in the lateral nasal wall. Unlike NCMHs, COREAHs do not exhibit aggressive pathology such as intracranial or intraorbital extension. Similarly to REAH, patients usually present with nasal obstruction, epistaxis, and chronic or recurrent rhinosinusitis.

COREAHs and REAHs share similar histopathological characteristics including glands lined with ciliated respiratory epithelium and goblet cells originating from surface epithelial invaginations. The stroma exhibits features typical of sinonasal inflammatory polyps, such as edema, vascular and fibroblastic proliferation, and chronic inflammation. However, COREAH can be differentiated from REAH by the presence of cartilaginous and/or osseous trabeculae which are often seen as calcifications on CT scans. The pathophysiology of COREAH remains unclear. There are two main hypotheses. One hypothesis is that COREAH, like other hamartomatous lesions, may be congenital arising from an inherent developmental error. The second hypothesis is that COREAH is due to an inflammatory process perhaps explaining the association between hamartomatous lesions and chronic rhinosinusitis. The onset of symptoms in our patient's 40s would suggest against a congenital etiology. COREAH is a benign expanding tumor, making it crucial to establish a differential diagnosis with neoplasic lesions such as inverted papilloma or low-grade adenocarcinoma. Inverted papillomas arise from the Schneiderian membrane and are characterized by papillary projections with a fibrovascular core, covered by several layers of squamous or transitional epithelium **(Figure 7)**. In contrast, COREAHs are adenomatoid structures lined by a single layer of ciliated epithelium. Neutrophils can also be seen migrating through epithelium. Moreover, unlike COREAHs, inverted papillomas are known to be locally aggressive tumors whose clinical progression involve bone lysis, local invasion, and malignant transformation potential [17].

| | COREAH [9,10] | REAH [11] | SH [12] | NCMH [13-16] |
|---|---|---|---|---|
| **Age** | 3-83 years old | 30-90 years-old (peak at 50-60) | 60 years old | Children and infants under the age of 1 year old |
| **Association** | Associated to chronic rhinosinusitis<br>No genetic mutation identified | Associated to chronic rhinosinusitis.<br>No genetic mutation identified | No genetic mutation identified | Associated to DICER-1 mutation, which is liked to pleuropulmonary blastoma and ovarian sex cord-stromal tumor |
| **Sex preference** | No sex preference | Male | Female | Male |
| **Frequent localisation** | Lateral nasal wall | Posterior nasal septum | Posterior nasal cavity or rhinopharynx | Ethmoid sinus |
| **Symptoms** | Nasal obstruction, epistaxis (rarely), and chronic or recurrent rhinosinusitis | Nasal obstruction, epistaxis (rarely), and chronic or recurrent rhinosinusitis | Nasal congestion, rhinorrhea and epistaxis | Nasal obstruction, face and dental pain, vision impairment |
| **Malignant transformation potential** | None | None | None | Yes: 2 cases documented.<br>Bone erosion and intracranial extension possible |
| **Anatomopathological findings** | Like REAH with the addition of cartilaginous and/or osseous trabeculae | Respiratory epithelium lined with ciliated and goblet cells, with stromal changes like sinonasal polyps | Respiratory epithelium and submucosal seromucinous glands in a lobular arrangement, with a lymphoplasmacytic infiltrate and eosinophilic granules | Chondroid tissue islands such as hyaline cartilage, foci of calcification, and elements of mesenchymal cells like myxoid stroma and spindle cell |

**Figure 6:** Comparison table of the different features of the four main subtypes of hamartomas located in the nasosinusal tract.

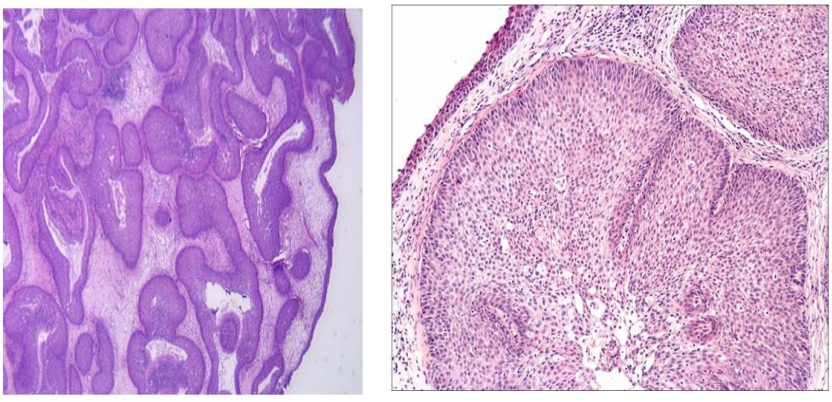

**Figure 7:** Histopathological findings of an inverted papilloma. Thickened squamous epithelium proliferating inward into the underlying stroma. *Hematoxylin and eosin stain, magnification x4 (left) and magnification x20 (right).*

Low-grade adenocarcinomas are less common and less invasive with no known sex or racial predilection. There is no link to wood dust exposure, and they do not tend to metastasize systemically. However, they may have the potential for local invasion and tissue destruction. They can be differentiated from COREAH through histological features such as dysplasia, nuclear stratification, and increased mitotic rate **(Figure 8) [18]**. Extensive surgical intervention is recommended, often in combination with radiotherapy for select cases.

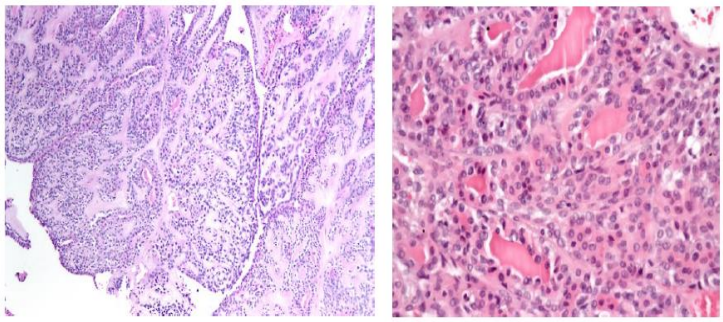

**Figure 8:** Histopathological findings of a low-grade adenocarcinoma. The respiratory mucosa is infiltrated by a neoplasic process forming small glands and acini within a moderately fibrotic stroma. The tumor consists of cuboidal cells with enlarged, irregular nuclei and evident nuclear atypia, with some mitotic activity. *Hematoxylin and eosin stain, magnification x20 (left) and x200 (right).*

## Conclusion

Chondro-osseous respiratory epithelial adenomatoid hamartomas (COREAH) are the least common subtype of hamartoma found in the sinonasal tract. They share similarities with respiratory epithelial adenomatoid hamartoma (REAH) but are characterized by the presence of a mesenchymal component: cartilage and/or osseous trabeculae, leading to the detection of calcification on imaging. As a benign slow-growing tumor, it is crucial to differentiate COREAH from other serious nasal masses to ensure the most appropriate management. The treatment of choice is the complete surgical resection of the mass. Regular long-term follow up is recommended to monitor for recurrence.

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