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Case Report

Sinonasal respiratory epithelial adenomatoid hamartoma: a rare clinical entity masked as nasal polyposis

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ABSTRACT

A hamartoma is a benign malformation or congenital error of tissue development indigenous to a specific part of the body, common in the lung, kidney, liver, spleen, and intestine but rare in the head and neck region, particularly nasal cavity and paranasal sinuses. A 50-year-old female presented with complaints of decreased smell sensation for 7 years; snoring, bilateral nasal obstruction, serous nasal discharge which is bloodstained occasionally and mouth breathing for 2 years. On Diagnostic nasal endoscopy, on right side, a single, pedunculated, globular mass originating from roof extending up to the floor of nasal cavity, seen between septum and inferior turbinate. Bilateral full house functional endoscopic sinus surgery with posterior septectomy was done and excised mass was sent for biopsy. Histopathological examination shows fragments of polypoidal tissue lined predominantly by pseudostratified ciliated columnar epithelium with intervening goblet-like cells at some areas, underlying stroma is loose, edematous. Irregularly branched, dilated glands lined by respiratory epithelium with thickened basement membrane, abundant intraluminal mucinous secretory material along with foamy macrophages and intervening stroma appears hyalinised with mixed inflammatory infiltrate with no evidence of malignancy and diagnosis was confirmed as REAH with inflammatory nasal polyposis. REAH is very rare and self-limiting lesions which do not regress spontaneously with majority of cases presenting as polypoidal mass causing nasal obstruction, nasal stuffiness, and hyposmia, resembling chronic rhinosinusitis.

Keywords: Case report, Hamartoma, REAH, Polyposis, Nasal mass

INTRODUCTION

Hamartoma is a benign malformation or congenital error of tissue development indigenous to a specific part of the body. It is commonly found in the lung, kidney, liver, spleen, and intestine, but is rare in the head and neck region, particularly the nasal cavity and paranasal sinuses. Respiratory epithelial adenomatoid hamartoma (REAH) is the most common hamartoma of the sinonasal tract, first recognized by Wenig and Heffner in 1995.[1] According to the World Health Organization, REAH is defined histologically by glands lined with multilayered ciliated respiratory epithelium containing goblet cells, a thick eosinophilic basement membrane, and no evidence of atypia or metaplasia.[1] It predominantly affects males between the ages of 30 and 90 years, with a male-to-female incidence of 3:2.[2] Clinically, REAH typically presents as unilateral or bilateral nasal polyposis, exhibiting no tendency toward uncontrolled growth and lacking malignant potential. However, it does not resolve spontaneously.[3,4] Histopathological analysis remains the gold standard for diagnosis. Complete surgical excision, including removal of the lesion's pedicle, is curative, with no reported recurrences following resection.[4,5]

CASE REPORT

A 50-year-old female presented with complaints of decreased smell sensation for 7 years; snoring, bilateral nasal obstruction, serous nasal discharge which is bloodstained occasionally and mouth breathing for 2 years. There is an episode of right nasal bleed, a year ago.

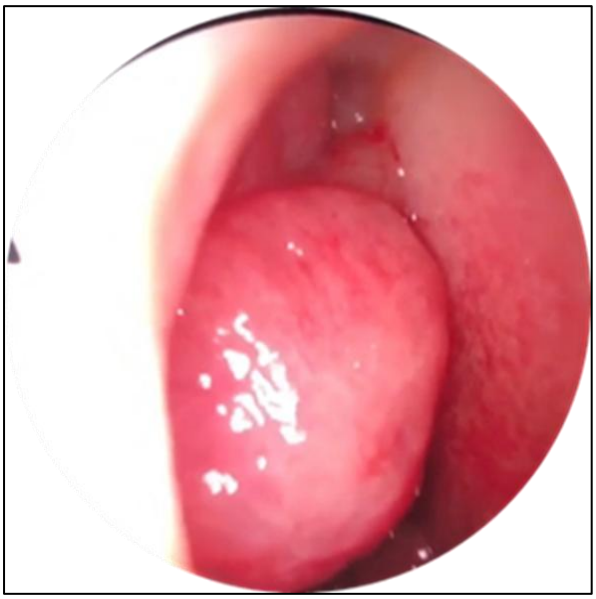

Figure 1: Endoscopic image of right nasal cavity.

History of exposure to cotton dust for the last 5 years. On anterior rhinoscopy, right nasal cavity single, reddish mass between septum and inferior turbinate with smooth surface, bilateral nasal polyps with high deviated nasal septum. Nasal mucosa is congested bilaterally.

Diagnostic nasal endoscopy findings

Right side

A single, pedunculated, globular mass originating from roof extending upto the floor of nasal cavity, seen between septum and inferior turbinate with high deviated nasal septum to right. On probe test mass is firm in consistency, sensitive to touch, probed all around except superiorly and does not bleed on touch.

Left side

Bony spur posteriorly impinging on inferior turbinate. Bilateral polypoidal changes in middle meatus, nasal cavities and spheno-ethmoidal recesses.

Computed tomography (CT) of paranasal sinuses

Sinonasal polyposis involving bilateral maxillary, ethmoidal, frontal sinuses and nasal cavities with extensions into sphenoid sinus with blockage of bilateral osteomeatal complex, spheno-ethmoidal and frontal recesses. Widened olfactory cleft.

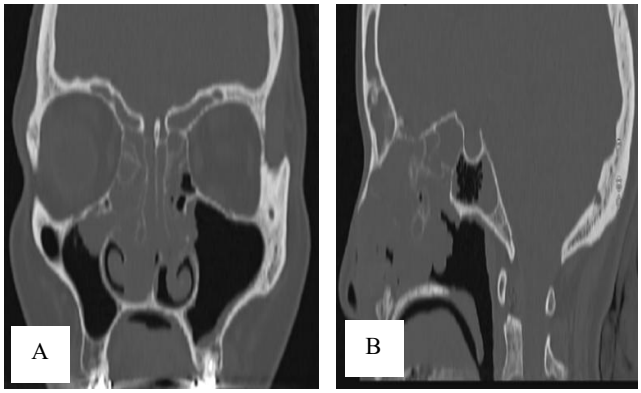

Figure 2: (A) plain computed tomography of paranasal sinuses coronal section: showing homogenous soft tissue opacification involving both nasal cavities with widened olfactory cleft and (B): plain computed tomography of paranasal sinuses sagittal section: showing homogenous soft tissue opacification involving nasal cavities with widened olfactory cleft.

Treatment

Under general anesthesia, through endoscopic approach single, pedunculated, globular mass originating from right side roof of the nose seen, the pedicle is cauterized with bipolar and removed in total and sent for biopsy.

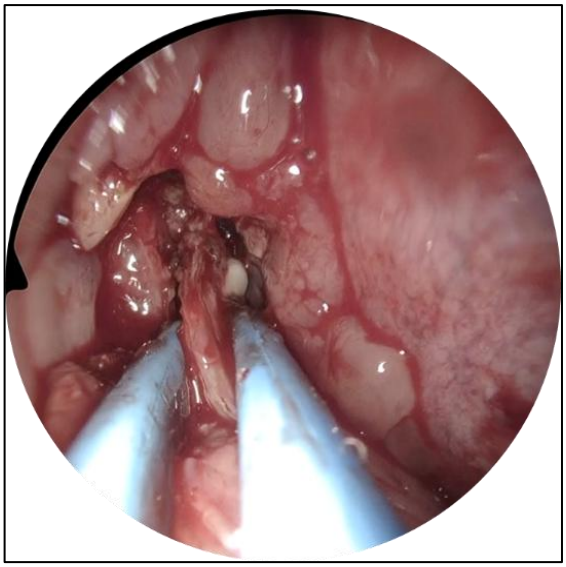

Figure 3: Intraoperative images showing endoscopic sinus surgery-posterior septectomy being performed after excision of polyps and nasal mass.

Bilateral full house functional endoscopic sinus surgery is done as polypoidal changes seen in middle meatus, nasal cavity, spheno-ethmoidal recess. Bilateral middle turbinectomy done. Posterior septectomy done as mass is

arising from posterior part of septum, cribriform area and olfactory cleft area.

Histopathological examination

Shows fragments of polypoidal tissue lined predominantly by pseudostratified ciliated columnar epithelium which is showing intervening goblet like cells at some areas. The underlying stroma is loose, edematous. Irregularly dilated small collections of seromucinous glands seen.

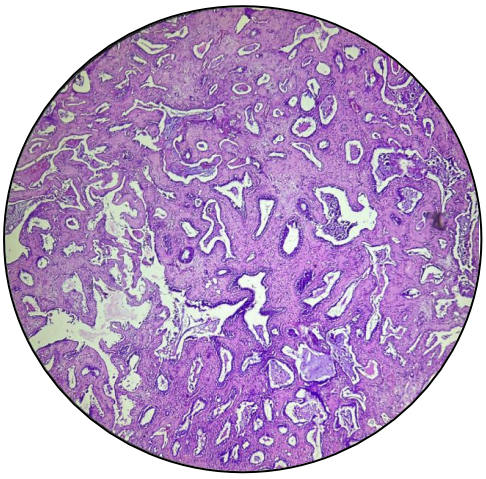

Figure 4: Histopathological examination of tissue sent for biopsy at 40X magnification showing histological features consistent with respiratory epithelial adenomatoid hamartoma with polyposis.

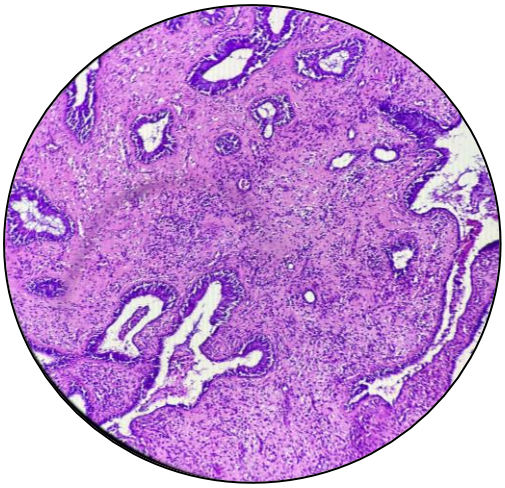

Figure 5: Histopathological examination of tissue sent for biopsy at 100X magnification showing histological features consistent with respiratory epithelial adenomatoid hamartoma with polyposis.

Irregularly branching and dilated glands lined by respiratory epithelium with thickened basement membrane and abundant intraluminal mucinous secretory material along with foamy macrophages. Intervening stroma appears hyalinized with mixed inflammatory infiltrate. No evidence of malignancy seen. Hence the diagnosis was confirmed as REAH with inflammatory nasal polyposis.

DISCUSSION

REAH is a rare and benign lesion that does not undergo spontaneous regression. First reported by Wenig and Heffner in 1995, it has since been increasingly recognized in sinonasal pathology.[1] Two primary etiological hypotheses exist: one proposing a congenital origin, and the other suggesting chronic inflammation, such as in chronic sinusitis, as a contributing factor.[1,2]

The majority of REAH cases present as polypoidal masses causing nasal obstruction, stuffiness, and hyposmia, clinically resembling chronic rhinosinusitis.[3,4] These lesions may be confined to the nasal cavity or diffusely involve the paranasal sinuses, often in association with other inflammatory changes.[2,4] Radiologically, REAH typically presents as a soft-tissue density within the sinuses or nasal septum, often mimicking nasal polyps. In our patient, similar imaging features were noted.

Lima et al. highlighted that enlargement of the olfactory cleft on CT of the paranasal sinuses is the most distinguishing radiographic feature of REAH, helping differentiate it from nasal polyposis.[4] The posterior nasal septum is the most commonly affected site.[1,4]

Histopathologically, REAH is characterized by round to oval-shaped glands separated by stroma, often distended with mucus. The basement membrane is thickened, and the stroma is typically edematous. Importantly, no atypical or metaplastic changes are observed.[1,3] distinguishing it from malignant lesions. The treatment of REAH involves complete local excision. When adequately removed, no recurrence has been reported, and there is no evidence of malignant transformation.[1,4,5]

CONCLUSION

Respiratory epithelial adenomatoid hamartomas (REAHs) are rare but distinct clinicopathological entities that should be considered in the differential diagnosis of sinonasal masses. They can clinically, radiologically, and histopathologically mimic other lesions such as inverted papilloma, sinonasal adenocarcinoma, olfactory neuroblastoma, and nasal polyposis. These lesions may present as isolated nasal cavity masses or be more diffusely distributed throughout the paranasal sinuses, often in association with chronic inflammatory conditions.

Regardless of the extent of clinical presentation, endoscopic surgical excision is curative, with no reported cases of recurrence following complete removal. Radiological findings, particularly enlargement of the olfactory cleft, should raise suspicion for REAH;

however, histopathological examination remains the gold standard for definitive diagnosis.

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