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Case Report **Surgery**

Respiratory Epithelial Adenomatoid Hamartoma

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Abstract

Respiratory epithelial adenomatoid hamartoma (REAH) is an uncommon benign lesion of the nasal cavity and paranasal sinuses with an unclear etiology. Although rare, REAH should be considered in the differential diagnosis of nasal lesions. Limited but complete surgical resection is the treatment of choice. We present a 54-year-old male patient with REAH in the left nasal cavity, with differential diagnoses including antrochoanal polyp and inverted papilloma.

Keywords: Hamartoma; Maxillary Sinus; Antrochoanal Polyp; Inverted Papilloma.

INTRODUCTION

Respiratory epithelial adenomatoid hamartoma of the nasal cavity and sinuses is a histological entity [1] that is relatively rare. Indeed, only about fifty cases have been described in the literature [1-2]. REAH was first described in a series of 31 cases by Wenig and Heffner in 1995 [1]. Hamartomas do not clearly represent either a neoplastic or inflammatory disorder [4]. These lesions are benign and, unlike neoplasms, hamartomas lack the ability to grow continuously, resulting in self-limited proliferation [1–3]. They are commonly seen in the lung, kidney, liver, spleen, and intestines but are very rare in the upper aerodigestive tract. Hamartomas of the head and neck region, particularly in the nasal cavity and paranasal sinuses, are extremely rare [5]. In the nasal cavity, REAH predominantly involves the posterior nasal septum. Involvement of the maxillary sinus, ethmoid sinus, frontal sinus, and nasopharynx is extremely rare [6].

CASE REPORT

A 54-year-old male patient with no significant medical or surgical history presented to our department with a six-month history of unilateral left nasal obstruction, associated with epistaxis. He denied rhinorrhea, hyposmia, headache, or any ophthalmologic signs. Clinical examination did not reveal any facial deformities, and the lymph nodes were non-palpable. Nasal endoscopy revealed a cerebriform polyp reaching the lower edge of the inferior turbinate and extending to the contralateral choana. CT scan showed the polyp filling the entire left maxillary sinus, protruding from the accessory maxillary sinus ostium (Figure 1, Coronal). The same mass extended posteriorly to the nasopharynx (Figure 2, Axial), a presentation suggestive of an antrochoanal polyp. A biopsy of the tumor was performed, and the anatomopathological examination was consistent with inverted papilloma.

The treatment was based on endoscopic sinus surgery, combined with a Caldwell-Luc surgical approach. The nasal mass was completely excised, and the postoperative recovery was uneventful. The final anatomopathological examination confirmed a diagnosis of respiratory epithelial adenomatoid hamartoma.

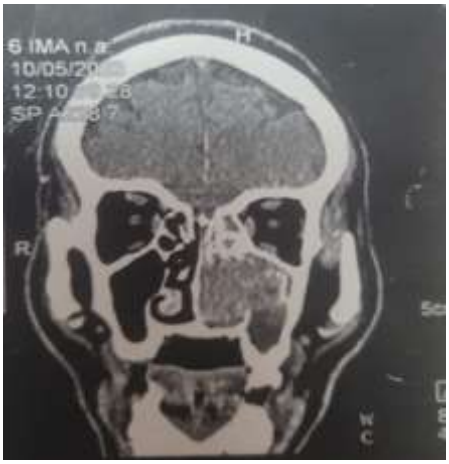

Figure 1: Coronal CT scan image showing the polyp filling the entire left maxillary sinus, protruding from the accessory maxillary sinus ostium

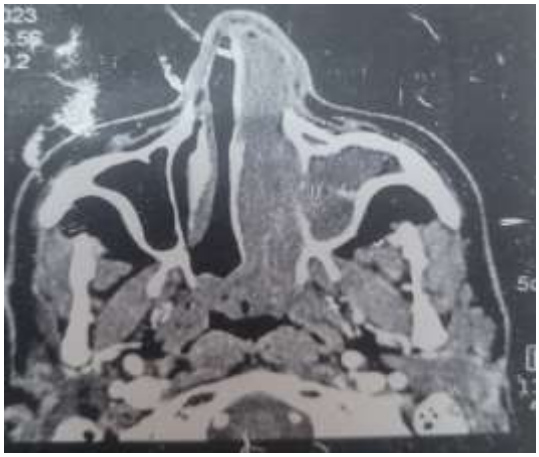

Figure 2: Axial CT scan image showing the mass extending posteriorly to reach the nasopharynx

DISCUSSION

Hamartomas are non-neoplastic malformations or congenital abnormalities of tissue development. They are defined as aberrant differentiation that may produce a mass of disorganized but mature, specialized tissue indigenous to the particular site [7]. Wenig and Heffner identified a subgroup of hamartomas and named it respiratory epithelial adenomatoid hamartoma (REAH). They defined REAH as a glandular component consisting of respiratory epithelium originating from the surface epithelium and a polypoid growth, resulting in a respiratory epithelial-lined adenomatoid proliferation [1]. They suggested that adenomatoid hamartomas arise in the setting of inflammatory polyps, and their development is most likely induced secondarily to the inflammatory process.

REAH predominantly affects male adults, with a male-to-female ratio of 5-7:1, and the age range typically spans the sixth to ninth decades of life [1, 10, 11]. It is relatively more common in the nasal cavity or nasopharynx [8, 9]. However, paranasal sinus involvement, particularly of the maxillary sinus alone, is quite rare. The clinical symptoms of adenomatoid hamartoma resemble those of chronic rhinosinusitis, such as nasal obstruction and epistaxis.

The histological differential diagnosis of REAH includes Schneiderian papillomas of the inverted type and adenocarcinoma. Inverted papillomas are characterized by a markedly thickened epidermoid or squamous epithelial proliferation mixed with mucocytes, intra-epithelial mucous cysts, and inflammatory cell infiltrates permeating the epithelial layer. Adenocarcinomas generally consist of a complex glandular growth, with glands growing in a back-to-back pattern without intervening stromal tissue [12].

Additionally, there is a scannographic differential diagnosis found in our case, with the presentation resembling an antrochoanal polyp. The description includes a hypodense mass arising from an enlarged, opacified maxillary sinus without bony destruction, enlarging the ostium, filling the nasal cavity, and extending to the choana.

Regarding the evolution of hamartomas, they lack the capacity for continuous, unimpeded growth, meaning their proliferation is self-limiting. Hamartomas have no malignant potential and do not tend to spontaneously regress [1, 14]. Therefore, it is crucial to distinguish REAH from more aggressive neoplastic diseases. Misdiagnosing this benign lesion may lead to unnecessarily aggressive surgical interventions [13]. The treatment of REAH is complete surgical resection [3]. No recurrence of nasal hamartoma has been reported in the literature [14].

CONCLUSION

Although adenomatoid hamartomas arising from the sinonasal tract are extremely rare, head and neck surgeons should be aware of this pathological entity as a differential diagnosis for inverted papilloma and adenocarcinoma. Furthermore, respiratory epithelial adenomatoid hamartoma of the maxillary sinus can be a differential diagnosis for antrochoanal polyp, especially on scannographic imaging.

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