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Respiratory epithelial adenomatoid hamartoma in the nasopharynx

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Abstract
We present a case report of a female patient with complaints of single-sided nasal obstruction. A polypoid structure was seen in the nasopharynx. Histologic examination showed a respiratory epithelial adenomatoid hamartoma – a rare, benign lesion. Therapy consisted of complete excision. In line with previous reports, the lesion did not recur during 13 months of follow up. The clinical and pathological features of this abnormality are discussed.

Key words: Hamartoma; Nasopharynx; Endoscopy; Surgical Procedures

Introduction

Single-sided nasal obstruction can be caused by a mass of tissue in the nose and nasopharynx. Nasal polyps and adenoidal hypertrophy are common causes, followed by antrochoanal polyps and papillomas. More infrequent, but important in differential diagnosis, is adenocarcinoma. In the patient we describe, a respiratory epithelial adenomatoid hamartoma was found. This is a benign tumour consisting of a proliferation of glandular spaces lined by ciliated epithelium and, less often, goblet cells.

Case report

A 68-year-old woman, previously seen in our out-patient clinic with sensorineural hearing loss, reported feeling something in her nose on the left side. Nasal endoscopy revealed a mass posterior to the left nasal cavity, originating from the tail of the inferior concha and covered in normal, intact epithelium. Biopsy revealed a respiratory epithelial adenomatoid hamartoma (REAH). After computerized tomography (Figures 1 and 2), complete endonasal endoscopic excision of the lesion, including the posterior part of the inferior turbinate, was performed

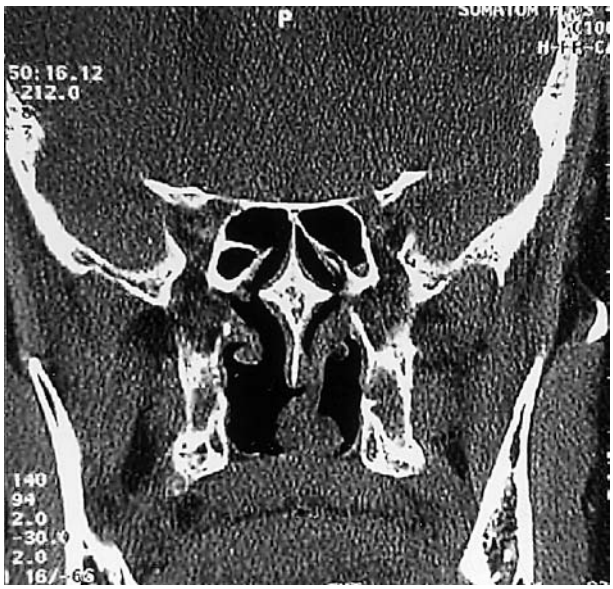

FIG. 1
Coronal CT view of the nasal cavity showing the tissue mass and its relation to the inferior concha.

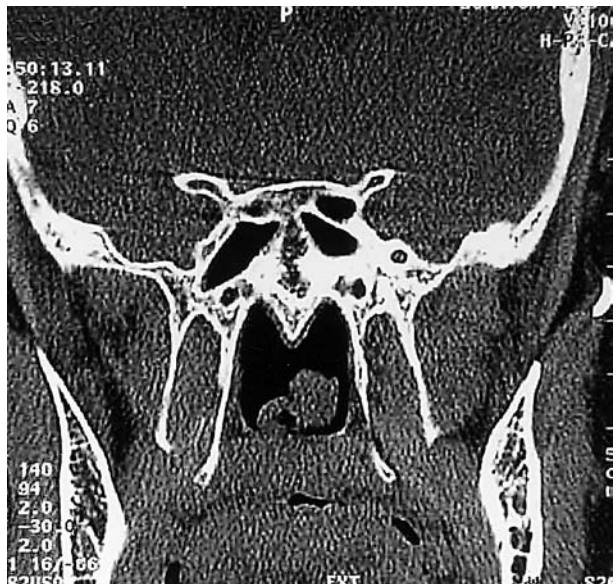

FIG. 2
A more posterior coronal CT slice shows the extension of the lesion in the nasopharynx.

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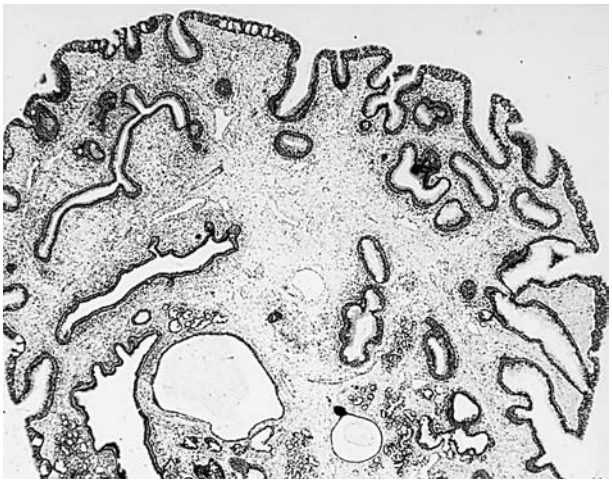

FIG. 3

Histological section of the excisional biopsy, showing a polypoid lesion consisting of an organoid proliferation of glands. Respiratory epithelial adenomatoid hamartoma has the low-power appearance of an inflammatory polyp but contains large numbers of glands lined by ciliated respiratory epithelium (H&E, ×25).

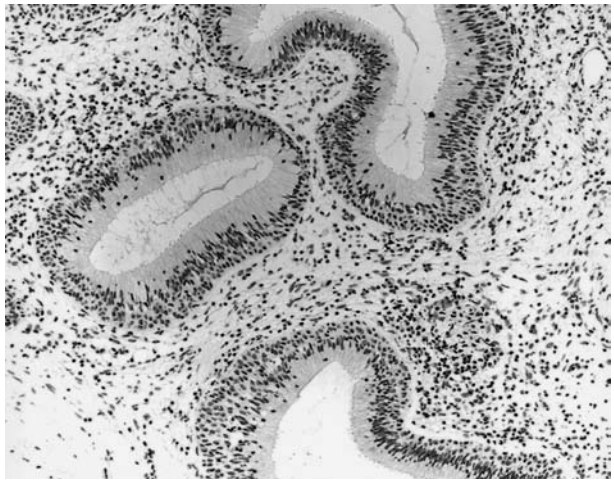

FIG. 4

High-power magnification showing organoid glands lined by ciliated cells (the cilia are not visible on the photograph but are easily detected under the microscope) (H&E, ×200).

under general anaesthesia. The excised specimen had a maximum diameter of 13 mm. Definitive histology confirmed the previous biopsy diagnosis. Afterwards, the patient was relieved of her complaints. She was well at follow up 13 months after surgery. More recent nasal endoscopy revealed no further abnormalities.

Histology

The biopsy and excision biopsy showed the same abnormality: a polypoid lesion consisting of an organoid proliferation of glandular spaces lined by ciliated epithelium (Figures 3 and 4). Neither atypia nor characteristics of an inverted papilloma were present.

- **In this patient unilateral nasal obstruction was caused by a respiratory epithelioid adenomatoid hamartoma, a rare, benign lesion**
- **Treatment was by endoscopic excision, with no recurrence at 13 months**
- **The differential diagnosis and treatment are discussed**

Discussion

Gradually increasing, unilateral nasal obstruction in adult patients should always be regarded as a symptom requiring further examination. Nasal endoscopy often reveals a mass of tissue in the nasal cavity or nasopharynx on which incisional biopsy can be performed. Computed tomography scanning can be helpful for diagnostic and surgical reasons. Treatment mainly depends on histological examination and ranges from conservative to radical surgical removal. Hamartomas in the nasal cavity or nasopharynx are quite rare. Thus far, only 37 cases have been presented in the literature. Diameters are reported up to a maximum of 4.9 cm.[1]

Hamartomas are defined as non-malignant malformations composed of excessive proliferation of one or more cellular components specific to a given tissue.[2] They can occur in all areas of the body, especially the liver, spleen, kidney and lung. In the upper aerodigestive tract, however, they are rare; most reports are limited to single case studies.[3–5]

According to Wenig and Heffner,[1] hamartomas in this location are histologically characterized by glandular components consisting of respiratory epithelium originating from the surface epithelium. The main morphological feature is a proliferation and accumulation of glands lined by pseudostratified ciliated epithelial cells. These are different from seromucinous glands, which are characterized by absence of ciliated epithelial cells. There are no signs of atypia or metaplastic changes. Hamartomas have no capacity for continuous unimpeded growth, so their proliferation is self-limiting. Hamartomas have no tendency to regress spontaneously.

Apart from inflammatory polyps, the differential diagnosis includes inverted papilloma and adenocarcinoma. Inflammatory polyps have mucous glands embedded in their oedematous stroma. Inverted papillomas are characterized by invagination of the surface epithelium in the underlying stroma. In adenocarcinoma, a proliferation of infiltrative, growing neoplastic tubular glands are recognized, lined by atypical epithelium. On tiny biopsies, the differential diagnosis between normal tissue, inflammatory polyp and REAH can be problematic for the surgical pathologist.

Complete surgical removal of the hamartoma is the treatment of choice. In the upper respiratory tract, endonasal endoscopic surgery is preferred because of its relatively limited morbidity. No recurrences are reported in the literature for a follow-up period of some months to several years.[1,6]

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