The Journal of Laryngology & Otology
April 2002, Vol. **116,** pp. 317–318

Pathology in Focus

Respiratory epithelial adenomatoid hamartoma of the maxillary sinus

Yuji Himi, M.D., Tomokazu Yoshizaki, M.D.[*], Katsuaki Sato, M.D.[†], Mitsuru Furukawa, MD.[*]

Abstract
A case of respiratory epithelial adenomatoid hamartoma of the maxillary sinus is reported. Glandular hamartomas involving the sinonasal tract have received only limited documentation in the literature. The differential diagnosis of adenomatoid hamartoma includes schneiderian papilloma of the inverted type and adenocarcinoma. Limited but complete surgical resection is the treatment of choice.

Key words: Adenomatoid Tumour; Hamartoma; Maxillary Sinus

Case report

A 56-year-old Japanese man had had exacerbated right nasal obstruction, rhinorrhoea and headaches for three months. He had a history of bronchial asthma. The middle nasal meatus was found closed on sinonasalscopy. Endoscopic observation and biopsy were performed under local anaesthesia. The maxillary sinus was occupied by a purulent fluid, and the mucosa was hyperaemic and hypertrophic. On the basis of histological findings, the biopsy specimen was diagnosed as respiratory epithelial adenomatoid hamaratoma.

Magnetic resonance imaging (MRI) revealed diffuse mucosal thickening of the right maxillary sinus. The tumour showed low intensity on a T1-weighted image, high intensity on a T2-weighted image and homogeneous gadolinium enhancement (Figure 1). Under a diagnosis of respiratory epithelial adenomatoid hamartoma of the maxillary sinus, the tumour was resected by the Caldwell-Luc procedure. The maxillary sinus was covered with solid and hypertrophic mucosa. The ethmoid sinus was occupied by oedematous polyps. The entire maxillary mass was diagnosed as adenomatoid hamartoma (Figure 2) and the ethmoid mass was diagnosed as an inflammatory polyp by histological examination. There has been no evidence of tumour recurrence for 12 months.

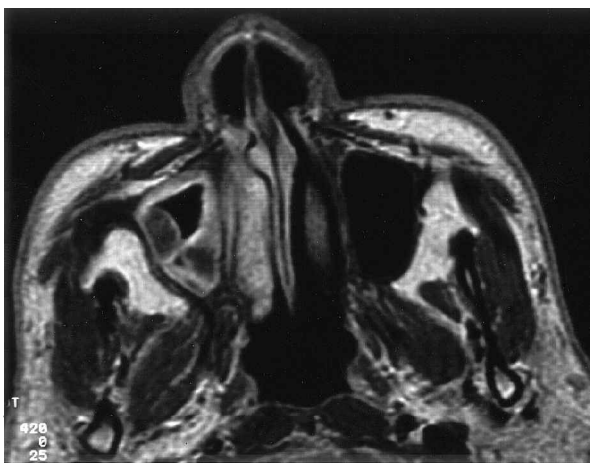


Fig. 1
MRI of the nasal cavity and paranasal sinus.

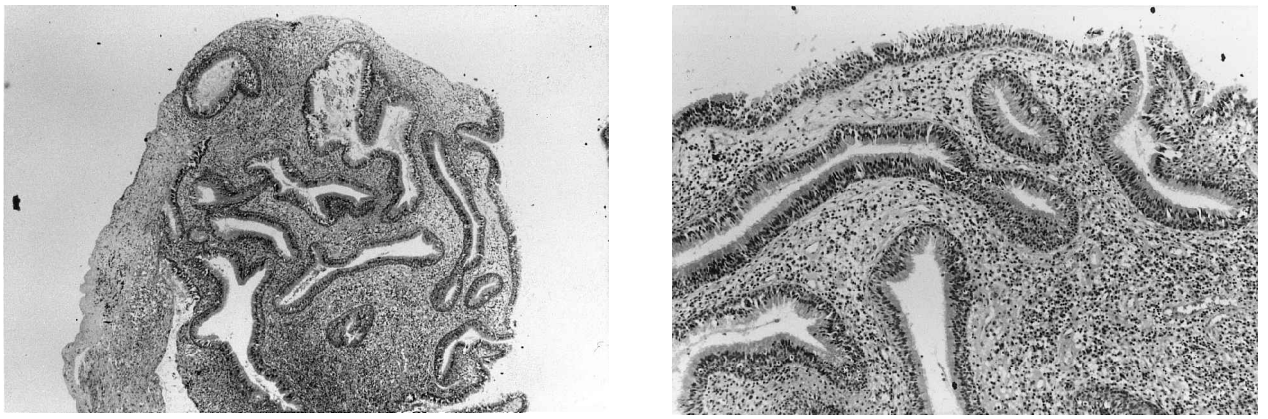
Pathological findings

The mucosa presented with a polypoid lesion originating from proliferating glands and inflammatory stroma (Figure 2(a)). The glands were moderately dilated and composed of ciliated respiratory epithelial cells directly continuous with the surface epithelium (Figure 2(b)). The proliferation of the glands was adenomatoid. No malignancy was found.

Discussion

Hamartomas are non-neoplastic malformations or congenital errors of tissue development. They are characterized by an abnormal mixture of tissues indigenous to that area of the body, but with an excess of one or more of the tissue types.[1] They occur in all areas of the body, especially the liver, spleen, kidney and lung, but are rare in the upper aerodigestive tract. They are reported relatively more often in the nasal cavity or nasopharynx.[1–5] However, paranasal sinus involvement, especially of the maxillary sinus alone, is quite rare.

From the Departments of Plastic and Reconstructive Surgery, Kanazawa Medical College and the Department of Otolaryngology[*], and the First Department of Pathology[†], Faculty of Medicine, Kanazawa University, Kanazawa, Japan.
Accepted for publication: 8 October 2001.



(a) (b)

FIG. 2

Histological findings.
(a) Low-power microscopic findings of the paranasal mucosa. The mucosa shows polypoid growth of glands and inflammatory stroma (H&E; ×40).
(b) High-power microscopic findings of the paranasal mucosa. The proliferating glands are organized with ciliated respiratory epithelial cells originating from the surface epithelium (H&E; ×200).

Wenig and Heffner found a subgroup of hamartoma and named it respiratory epithelial adenomatoid hamartoma. They defined respiratory epithelial adenomatoid hamartoma as follows; the glandular component of the hamartoma consisted of respiratory epithelium originating from the surface epithelium, and polypoid growth was the result of respiratory epithelial-lined adenomatoid proliferation.[6] They suggested that adenomatoid hamartomas arose in a setting of inflammatory polyps and in all likelihood their development was induced secondary to the inflammatory process. Thus, clinical symptoms of adenomatoid hamartoma resemble chronic rhinosinusitis such as nasal obstruction, nasal stuffiness, and epistaxis.

In the case presented here, physical examination identified the presence of a polypoid mass lesion in the middle nasal meatus, which was histologically diagnosed as an inflammatory polyp, suggesting the co-existence of inflammatory disease.

The histology of the maxillary mucosa was typical of the adenomatoid hamartoma which was defined by Wenig and Heffner.[6] Thus, our case fitted in the category of respiratory epithelial adenomatoid hamartoma. The differential diagnosis of adenomatoid hamartoma includes schneiderian papillomas of the inverted type and adenocarcinoma. Inverted papillomas are composed of a markedly thickened epidermoid or squamous epithelial proliferation admixed with mucocytes, intra-epithelial mucos cysts, and the presence of an inflammatory cell infiltrate permeating through the epithelial layer. Adenocarcinomas are generally composed of a complex glandular growth, with glands growing in a back-to-back pattern without intervening stromal tissue.[7]

In contrast to neoplasms that also represent an overproliferation of cells endogenous to a given tissue, hamartomatous proliferations do not have the capability for continuous unimpeded growth and are self-limiting. Therefore, there are no instances of recurrent, persistent, or progressive disease in the literature.[2,6] Conservative surgical intervention is curative.

Although adenomatoid hamartoma arising from the sinonasal tract is very rare, head and neck surgeons should be aware of this pathological entity as a differential diagnosis for inverted papilloma and adenocarcinoma.

References

1 Mark HT, Glenn FB, Seth MP. Nasal hamartoma: case report and review of the literature. *Int J Pediatr Otorhinolaryngol* 1993;**28**:83–8
2 Graeme-Cook F, Pilch BZ. Hamartomas of the nose and nasopharynx. *Head Neck* 1992;**14**:321–7
3 Majumder NK, Venkataramaniah NK, Gupta KR, Gopalakrishnan S. Hamartoma of the nasopharynx. *J Laryngol Otol* 1977;**91**:723–7
4 Lapado AA. A case of benign congenital hamartoma of the nasopharynx. *J Laryngol Otol* 1978;*92*:1141–5
5 Mahindra S, Daljit R, Sohail MA, Bazaz Malik G. Hamartomas of the nose. *J Laryngol Otol* 1978;**92**:57–90
6 Wenig BM, Heffner DK. Respiratory epithelial adenomatoid hamartomas of the sinonasal tract and nasopharynx: A clinicopathological study of 31 cases. *Ann Otol Rhinol Laryngol* 1995;**104**:639–45
7 Heffner DK. Classification of human upper respiratory tract tumours. *Environ Health Perspect* 1990;**85**:219–29

Address for correspondence:
Tomokazu Yoshizaki, M.D.,
Department of Otolaryngology,
School of Medicine, Kanazawa University,
13-1 Takara-machi,
Kanazawa 920-8640, Japan.

Fax: +81-76-234-4265
E-mail: tomoy@orl.m.kanazawa-u.ac.jp

Dr T. Yoshizaki takes responsibility for the integrity of the content of the paper.
Competing interests: None declared