Auris Nasus Larynx 35 (2008) 569–571

AURIS NASUS LARYNX
INTERNATIONAL JOURNAL OF ORL & HNS

www.elsevier.com/locate/anl

Rhino sinusal bilateral hamartoma: A case report

Pasqualina Maria Picciotti [a,*], Lea Calò [a], Antonino Mulè [b], Claudia Maggiore [b], Emanuele Scarano [a]

[a] *Institute of Otolaryngology, Catholic University of the Sacred Heart, Rome, Italy*
[b] *Histopathology, Catholic University of the Sacred Heart, Rome, Italy*

Received 3 July 2007; accepted 16 October 2007
Available online 22 January 2008

Abstract

The hamartoma is a benign rare tumor constituted by a mixture of tissue. It is very unusual in the nasal cavity.

The objective of the study is to describe an unusual case of bilateral nasal hamartoma. We report a 52-year-old male patient with a bilateral paranasal hamartoma of the ethmoid and maxillary sinus. Functional endoscopic sinus surgery was performed to completely remove the masses.

The reported localization is unusual because the most common site in the nose is the posterior septum. Although hamartoma arising from the rhino sinusal region is very rare, head and neck surgeons must know this entity in order to differentiate it from inverted papilloma and adenocarcinoma. Misinterpretation of this lesion may result in aggressive surgery for a benign lesion.

Keywords: Hamartoma; Rhino sinusal region; Polyposis

1. Introduction

Hamartomas can be defined as non-neoplastic malformation or inborn errors of tissue development constituted by a mixture of tissue [1]. They have no capacity of continuous growth and their proliferation could be considered self-limiting. They can be sited in any part of the body, commonly originating from the lung, kidney, liver, spleen and intestine but rarely in the upper aero digestive tract.

Respiratory epithelial hamartomas, firstly described by Wenig and Heffner in 1995, are rare and take origin from the rhino sinusal epithelium. While the most common site is the nose and nasopharynx, paranasal involvement is quite rare. In particular, approximately 70% of nasal hamartomas occur in the posterior nasal septum [2].

Histologically, these lesions are characterized by a prominent glandular proliferation lined by ciliated respiratory epithelium originating from the surface epithelium. The differential diagnosis of hamartomas includes primary schneiderian papillomas of the inverted type, adenocarcinomas, secondary hemangiomas, gliomas, dermoids, squamous cell carcinomas, olfactory neuroblastomas, and lymphomas.

There are no instances of recurrence or persistence or progressive disease in literature.

This tumor has no tendency to regress spontaneously, so the complete local surgical excision by means of a functional endoscopic approach is the treatment of choice for this lesion [3].

In this paper we describe a case of a bilateral rhino sinusal hamartoma.

2. Case report

A 52-year-old smoker Italian man presented at our observation with a bilateral nasal obstruction, anterior nasal discharge, hyposmia, and headache and facial pain. Physical examination showed polypoid masses in both nasal cavities.

* Corresponding author. Tel.: +39 06 30154439; fax: +39 06 3051194.
E-mail address: pmpicciotti@rm.unicatt.it (P.M. Picciotti).

0385-8146/$ – see front matter
doi:10.1016/j.anl.2007.10.006

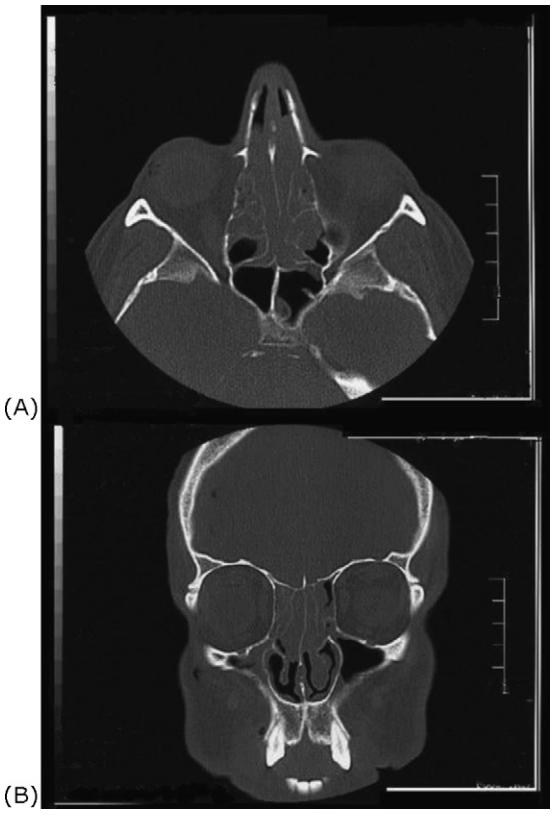

Fig. 1. Axial (A) and coronal (B) CT scan of the skull showing a bilateral nasal homogeneous mass occupying the ethmoid and the ostiomeatal complex bilaterally.

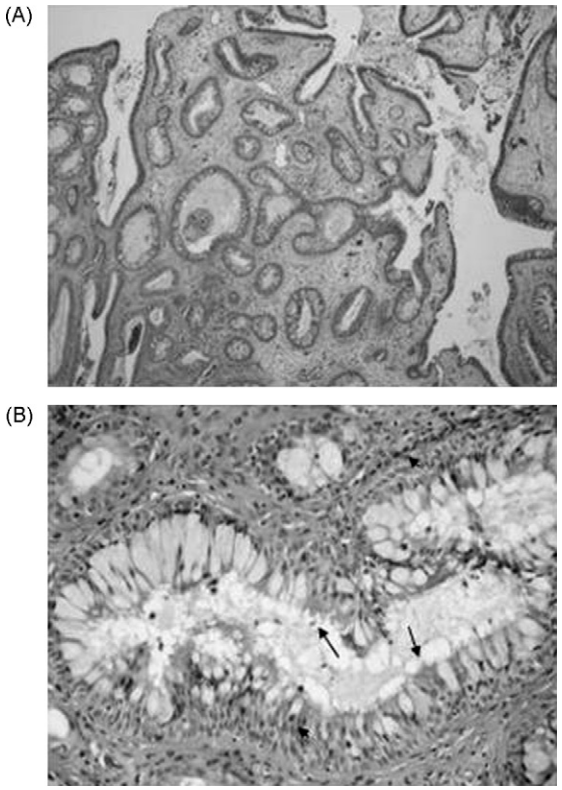

Fig. 2. (A) Branching glands with extensive mucous metaplasia embedded in aedematous stroma. Note the continuity between glandular and surface epithelium (H&E, original magnification 40×). (B) Respiratory cells with luminal cilia intermingled with mucous goblet cells. Scattered eosinophils are evident in the stroma and within the glandular epithelium (H&E, original magnification 100×).

Axial and coronal CT scan showed the presence in the right and left nasal cavity of a mass causing ostiomeatal complex obstruction (Fig. 1A and B). Ethmoid sinus opacification was associated. At the endoscopic observation the masses were neither attached to the septum, nasal vestibule nor floor and nasopharynx. The nasal masses were removed endoscopically under general anesthesia performing a bilateral ethmoidectomy and antrostomy. The excised masses were elastic and hard with a smooth surface, and an aedematous appearance. Histological examination revealed numerous branching glands filled with abundant mucus and embedded in aedematous or collagenized stroma. Ciliated respiratory cells with extensive mucous metaplasia composed the glandular epithelium. A moderate to scanty inflammatory infiltrate with a prevalent eosinophilic component was detected throughout the stroma and within epithelial cells. A diagnosis of bilateral respiratory epithelial adenomatoid hamartoma was rendered (Fig. 2).

The patient's postoperative course was uneventful. He is well and without any sign of local recurrence at a 12 months follow-up evaluation.

3. Discussion

Hamartoma of the nasal cavity is rare and its common site is the nasal septum, in the posterior area or anterior nasopharynx [4]. Although paranasal involvement is quite rare, recently different cases have been reported in literature. Wenig and Heffner described 31 cases of nasal hamartoma: 22 in the nasal cavity, 3 in the nasopharynx, 1 in the ethmoid and finally only 1 in both ethmoid and frontal sinuses. Kessler and Unterman [5] and Himi et al. [6] reported two cases of unilateral hamartoma in the maxillary sinus associated with an inflammatory polyp of the ethmoid in the second case. On the other hand, Malinvaud et al. described a unilateral ethmoidal hamartoma in a man [7]. Finally, Terris et al. in a child reported a similar nasosinusal unilateral localization [8].

Our patient differs from the previous described cases of nasosinusal hamartomas, because of the bilateral localization of the tumor in the ethmoid and maxillary sinuses.

The mechanisms responsible of the occurrence of hamartomas are still unknown and it has been speculated that they could be induced by inflammatory process [2]. The frequent association with the polyposis [9,10] supports this hypothesis. The association of hamartoma and chronic rhino sinusitis without polyps characterizes our case. This is the reason why in our opinion, it seems more likely for hamartoma to induce a chronic inflammatory condition which favorites the development of inflammatory polyps. Moreover, congenital nature of this tumor cannot be justified

with an inflammatory hypothesis. In fact, hamartomas are considered as a malformation or congenital errors of tissue development, characterized by an abnormal mixture of tissue indigenous to that area of the body, but with an excess of one or more tissue types [8]. An alternative possible explanation for this association is that some ''hamartomatous'' lesions represent inflammatory polyps with exuberant hyperplasia of the epithelium.

This report stresses the importance to be aware of a possible unusual localization of hamartomas in the paranasal cavity, in order to distinguish them from papillomas and adenocarcinomas to avoid useless and destructive surgery.

References

[1] Kacker SK, Dasgupta G. Hamartomas of ear and nose. J Laryngol Otol 1973;87:801–5.

[2] Wenig BM, Heffner DK. Respiratory epithelial adenomatoid hamartomas of the sinonasal tract and nasopharynx: a clinicopathologic study of 31 cases. Ann Otol Rhinol Laryngol 1995;104:639–45.

[3] Braun H, Beham A, Stammberger H. Respiratory epitheloid adenomatoid hamartoma of the nasal cavity—case report and review of the literature. Laryngorhinootologie 2003;82:416–20.

[4] Graeme-Cook F, Pilch BZ. Hamartomas of the nose and nasopharynx. Head Neck 1992;14:321–7.

[5] Kessler HP, Unterman B. Respiratory epithelial adenomatoid hamartoma of the maxillary sinus presenting as a periapical radiolucency: a case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;97:607–12.

[6] Himi Y, Yoshizaki T, Sato K, Furukawa M. Respiratory epithelial adenomatoid hamartoma of the maxillary sinus. J Laryngol Otol 2002;116:317–8.

[7] Malinvaud D, Halimi P, Cote JF, Vilde F, Bonfils P. Adenomatoid hamartoma of the ethmoid sinus: one case report. Rev Laryngol Otol Rhinol (Bord) 2004;125:45–8.

[8] Terris MH, Billman GF, Pransky SM. Nasal hamartoma: case report and review of the literature. Int J Pediatr Otorhinolaryngol 1993;28:83–8.

[9] Endo R, Matsuda H, Takahashi M, Hara M, Inaba H, Tsukuda M. Respiratory epithelial adenomatoid hamartoma in the nasal cavity. Acta Otolaryngol 2002;122:398–400.

[10] Delbrouck C, Fernandez Aguilar S, Choufani G, Hassid S. Respiratory epithelial adenomatoid hamartoma associated with nasal polyposis. Am J Otolaryngol 2004;25:282–4.