CASE REPORT

Respiratory epithelial adenomatoid hamartoma of the maxillary sinus presenting as an antrochoanal polyp

Selcuk Mulazimoglu a,*, Yuce Islamoglu a,1, Suha Beton a,1, Hale Kivrak b,2, Serpil Dizbay Sak b,2, Irfan Yorulmaz a,1

a Department of Otorhinolaryngology, Ankara University Medical School, Ankara, Turkey
b Department of Pathology, Ankara University Medical School, Ankara, Turkey

Received 4 February 2015; accepted 25 February 2015
Available online 17 March 2015

KEYWORDS
Respiratory epithelial adenomatoid hamartoma; Maxillary sinus; Antrochoanal polyp; Sinonasal neoplasm; Hamartoma

Abstract Respiratory epithelial adenomatoid hamartoma (REAH) is a rare benign upper airway lesion that is characterized by abnormal glandular proliferation. Maxillary sinus localization is extremely rare. Maxillary sinus involvement along with the extension beyond the sinus could be a challenge in differential diagnosis. In this article, a case of an isolated maxillary sinus REAH extending to the choana is presented.

© 2015 Egyptian Society of Ear, Nose, Throat and Allied Sciences. Production and hosting by Elsevier B.V. All rights reserved.

1. Introduction

Hamartomas are benign, non-neoplastic lesions that occur secondary to tissue-development anomalies and they are composed of overgrowth of mature cells and tissues that normally occur in the affected location.1,2 Respiratory epithelial adenomatoid hamartomas (REAH) are rare benign upper airway lesions that are characterized by abnormal glandular proliferation.3,4 They are commonly reported in the nasal cavity and nasopharynx;4,5 maxillary sinus localization is extremely rare.4,5 Histopathological differential diagnosis of this benign lesion from inverted papilloma and adenocarcinoma is important.5 In this article, a case of an isolated maxillary sinus REAH simulating an antrochoanal polyp (ACP) is presented.

2. Case report

A 44 year old woman presented with nasal obstruction 3 years of duration. Endoscopic nasal examination showed left sided polypoid mass obstructing the left nasal cavity extending to the nasopharynx (Fig. 1). Computed tomography (CT) showed complete opacification of the left maxillary sinus and nasal cavity, with erosion of the medial wall of the maxillary sinus (Fig. 2A). Magnetic resonance imaging (MRI) showed a left...
sided maxillary sinus mass hypointense in T1, and hyperin-
tense in T2 weighted images, with peripheral contrast enhance-
ment (Fig. 2B, C). Incisional biopsy from the intranasal part of
the mass was consistent with histopathological findings of a
nasal polyp. The mass was removed by the endonasal
endoscopic approach, with the help of angled endoscopes
and instruments. The site of origin was the posterolateral wall
of the maxillary sinus and complete removal was confirmed.
Histopathological examination revealed glandular prolifera-
tions lined with the respiratory epithelium in continuity with
the surface epithelium (Fig. 3A). Around these glandular
formations, a thick, dense, eosinophilic basement membrane
like material was identified, separated by an edematous stroma
(Fig. 3B). The final diagnosis was respiratory epithelial
adenomatoid hamartoma.

3. Discussion

Hamartomas are malformations composed of an excessive
proliferation of one or more cellular components of a specific
tissue. They are self-limiting, non neoplastic lesions which
do not regress spontaneously. Although the mechanisms for
occurrence of hamartomas are still unclear, it is suggested that
they originate from inflammatory polyps. REAHs are rare
lesions and since they are unmentioned in most textbooks of
general pathology, otolaryngologists and pathologists should
be aware of their existence when diagnosing a sinonasal
mass. Recognition and differential diagnosis of REAHs
from more aggressive sinonasal pathologies such as inverted
papillomas and low-grade adenocarcinomas are crucial to save
the patient from unnecessary extensive surgical procedures.

Figure 1  Endoscopic examination of the left nasal cavity
showing polypoid mass obstructing the nasal cavity. (S: septum,
IT: inferior turbinate, asterisk: mass).

Figure 2  Coronal computed tomographic images (A), coronal (B) and axial (C) TIRM sequence magnetic resonance images of the
patient.
Antrochoanal polyps are benign lesions originating from the maxillary sinus mucosa, which grow into the nasal cavity to reach the choana. ACPs either enlarge the accessory or natural maxillary sinus ostia while reaching the nasal cavity. ACPs has an intramaxillary cystic component and a solid intranasal component. While resecting ACPs one should identify the typical cystic intramaxillary component and if it is missing the histopathologic diagnosis should be waited for differential diagnosis. If possible, the site of origin should be identified for further procedures.

Respiratory epithelial adenomatoid hamartomas are typically polypoid or exophytic lesions. Histologically, they are composed of small to medium sized, round to oval glands lined by the ciliated respiratory epithelium. The glands are characteristically surrounded by thick, dense, eosinophilic basement membrane like material and often in direct continuity with the surface epithelium. The glandular lumina contain mucinous or amorphous material. The stroma is well vascularized, edematous, or fibrous and may contain inflammatory cells. REAHs are most often confused with ordinary sinonasal polyps, inverted papillomas and low grade adenocarcinomas. Inverted papillomas arise from the surface Schneiderian epithelium and invaginate into the underlying stroma similar to REAHs. However, the cellular proliferation is a transitional squamoid epithelium in contrast to the adenomatoid respiratory epithelial-lined structures typically observed in REAHs. Sinonasal adenocarcinomas may originate from either seromucinous glands or the surface respiratory epithelium. The glands have a characteristic back-to-back cribriform growth pattern with no intervening stroma. In addition, pleomorphism, mitotic figures, and nuclear atypia are often present.

Maxillary sinus REAHs are rare sinonasal lesions, clinically simulating antrochoanal polyps. They may be clinically differentiated with the lack of a cystic component in REAHs. The exact differential diagnosis is made by histopathologic examination. They require complete resection, which is commonly achieved endoscopically.

Conflict of interest

None declared.

Acknowledgement

This study was presented as a poster presentation at the 11. International Congress of Otorhinolaryngology and Head & Neck Surgery, 17–19 April 2014, Ankara, Turkey.

References