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Respiratory Epithelial Adenomatoid Hamartoma and Inflammatory Nasal Polyps Arising from the Different Sides of the Nasal Cavity: A Case Report

CASE REPORT

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ABSTRACT

Respiratory epithelial adenomatoid hamartoma (REAH) is rare, non-neoplastic lesion characterized by an abnormal proliferation of the glandular part of the respiratory nasal mucosa. Occasionally, this polypoid mass can be bilaterally associated with inflammatory nasal polyps. We present a case of a 63-year-old woman with REAH that originated from the right-sided superior turbinate and that was associated with nasal polyposis of the left nasal cavity. Endoscopic examination results revealed a lobular, yellow, soft-tissue mass in the right nasal cavity and nasal polyps in the left nasal cavity. Therapy included complete excision. Histopathological examination results showed the characteristics of REAH in the right nasal cavity and inflammatory nasal polyps in the left nasal cavity. To our knowledge, only one case of simultaneous presentation of REAH and nasal polyps in opposite sides of the nasal cavity has been previously described. Appropriate cooperation between a rhinologist and pathologist enables a correct diagnosis of these rare and unusual lesions.

Keywords: Hamartoma, chronic inflammation, nasal cavity, nasal polyps, nasal surgical procedures

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INTRODUCTION

The respiratory epithelial adenomatoid hamartoma (REAH) of the nasal cavity and paranasal sinuses is a benign lesion that arises from the respiratory mucosa; it was first described by Wenig and Heffner (1). This lesion appears to be rare, and to date, only 60 cases have been reported in the literature (2). Hamartomas are defined as primary, non-neoplastic lesions that are caused by disturbance in tissue development. These anomalies develop because of disturbances in tissue development during the fetal period and errors in development of immature tissues during the postnatal period. REAHs are histopathologically characterized by glandular invaginations that originate from the surface respiratory epithelium (3). They may occur in isolation or in association with nasal polyposis (4). In this report, we present a case of a woman with REAH that originated from the right-sided superior turbinate and was associated with nasal polyposis of the left nasal cavity. To our knowledge, only one similar case of such an unusual clinical presentation of REAH has been described in the literature (5).

CASE REPORT

A 63-year-old woman was admitted to the Department of Otorhinolaryngology with clinical signs of chronic sinusitis. She suffered with bilateral nasal obstruction, mucopurulent nasal secretions, postnasal discharge, hyposmia, and right-sided epistaxis. Her symptoms worsened over 10 months, despite treatment with intranasal corticosteroids and oral antibiotics. Endoscopic examination results revealed a big, yellow, lobular polypoid lesion in the right nasal cavity and multiple polypoid masses in the left nasal cavity. Computed tomography revealed total opacification of the right nasal cavity and right maxillary and ethmoid sinus. In the left nasal cavity, we observed total opacity of the ethmoid sinus and partial opacity of the nasal cavity and maxillary sinus (Figure 1).

The patient underwent endoscopic excision of the lesions under general anesthesia. The right-sided mass originated from the medial portion of the superior turbinate and the left-sided polyps originated from the middle meatus. We performed bilateral ethmoidectomy and right-sided middle meatal antrostomy and found that the right maxillary sinus was filled with mucopurulent secretions. The excised right-sided lesion was stringy with a smooth yellow surface and lobular appearance (Figure 2a), and the excised left-sided masses had typical appearances of nasal polyposis (Figure 2b). Histopathological examination results showed a polypoid lesion that was covered with ciliated pseudostratified respiratory epithelium and that comprised many glandular spaces with ducts of various sizes, which were embedded in a fibrous stroma. The glands comprised columnar epithelial cells without metaplasia. These findings suggested a diagnosis of REAH (Figure 3a). Histological examination results of excised left-

sided polyps showed signs that were typical for inflammatory nasal polyps. The proliferation of ciliated respiratory epithelium, stromal edema, and strong inflammatory infiltrate, predominantly comprising eosinophils, was observed (Figure 3b). The patient did not have any complaints at the 1-year follow-u after treatment, and control examination showed no evidence of recurrence.

DISCUSSION

Respiratory epithelial adenomatoid hamartoma is a rare, benign lesion with an unclear etiology. The mechanisms that induce the development of hamartomas remain unknown. Two hypotheses have been postulated. One hypothesis suggests congenital origin, whereas the other one suggests an inflammatory origin (6). REAH may arise and coexist in the setting of inflammatory polyps, raising a possible developmental induction secondary to the inflammatory process. Cases of bilateral association of REAH and nasal polyposis, particularly those originating from the olfactory clefts, have been described in the literature (7). However, in our case, the development of REAH was independent of the development of inflammatory nasal polyposis. In our case, REAH originated from the right nasal cavity and inflammatory nasal polyps originated from the left nasal cavity. Careful histopathological analysis of our patient showed no evidence of the coexistence of REAH and nasal polyposis in both sides of the nasal cavity. In contrast, long-time chronic inflammation of the sinonasal mucosa could be an inducer for the development of the two different lesions in the two opposite sides of the nasal cavity. Histologically, REAH has no evidence of metaplasia and cell atypia, and also has no tendency to spontaneously regress (8). It most commonly originates from the nasal septum and olfactory cleft. The rare origin sites of REAH are paranasal sinuses, nasopharynx, and middle and inferior turbinate (8, 9). In our patient, the lesion originated from the medial part of the superior turbinate, which is the medial portion of the olfactory cleft. Complete surgical excision of the lesion is generally sufficient for its cure, and the recurrence rate of REAH is relatively small compared with that of inflammatory nasal polyps (10).

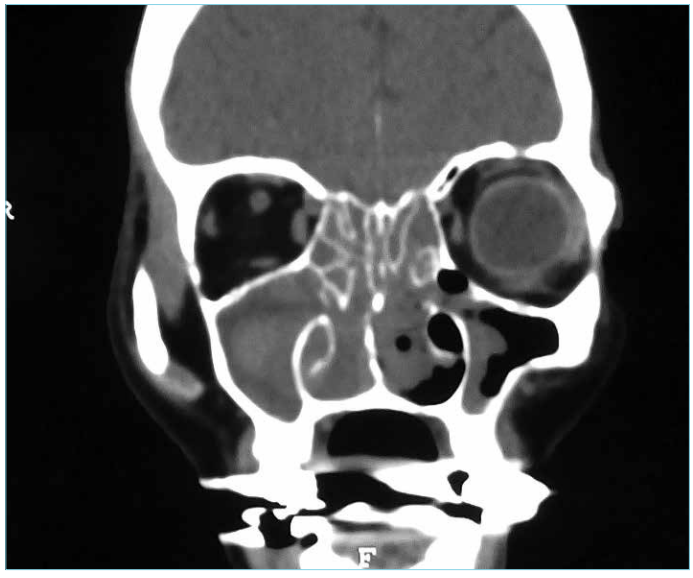

Figure 1. Coronal CT of the paranasal sinuses shows total opacification of both ethmoid sinuses and right maxillary sinus and partial opacification of the left nasal cavity and maxillary sinus

The main differential diagnostic problems with respect to REAH are nasal polyposis, inverted papilloma, and adenocarcinoma (10). REAH can be misdiagnosed as an inflammatory polyp because of its morphological and clinical similarities. REAH is characterized by submucosal proliferation of small-to-medium-sized, round-to-oval glands, lined with pseudostratified ciliated epithelium, and admixed with mucin-secreting cells. In contrast, inflammatory nasal polyps show the proliferation of pseudostratified respiratory epithelium with basement membrane thickening and stromal fibroblast proliferation and edema, with strong eosinophil, lymphocyte, and plasma cell tissue infiltration. Glandular proliferation can be found in nasal polyposis, but abnormal submucosal proliferation of the surface respiratory epithelium is more typically observed in REAH (9, 10). Although inverted papillomas often have a grossly polypoid appearance, they differ from "true" nasal polyps and REAHs. The lining epithelium of inverted papillomas most often is a non-ke-

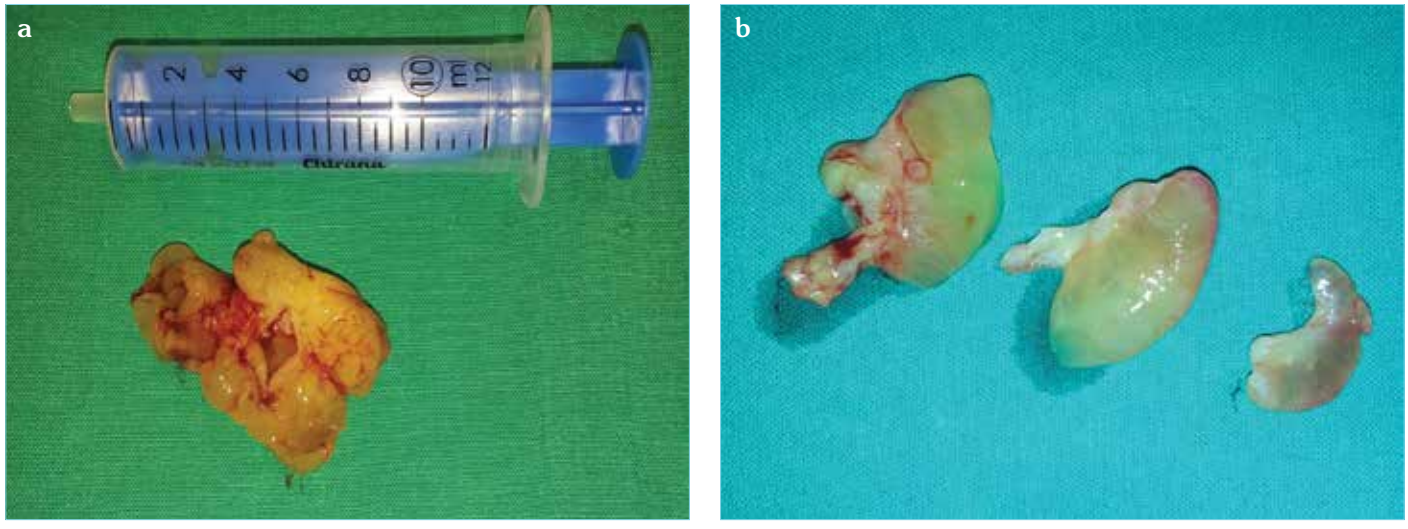

Figure 2. a, b. Macroscopic view of the excised right-sided lobular, yellow-colored, smooth surface mass (a) and of the excised left-sided polypoid lesions (b)

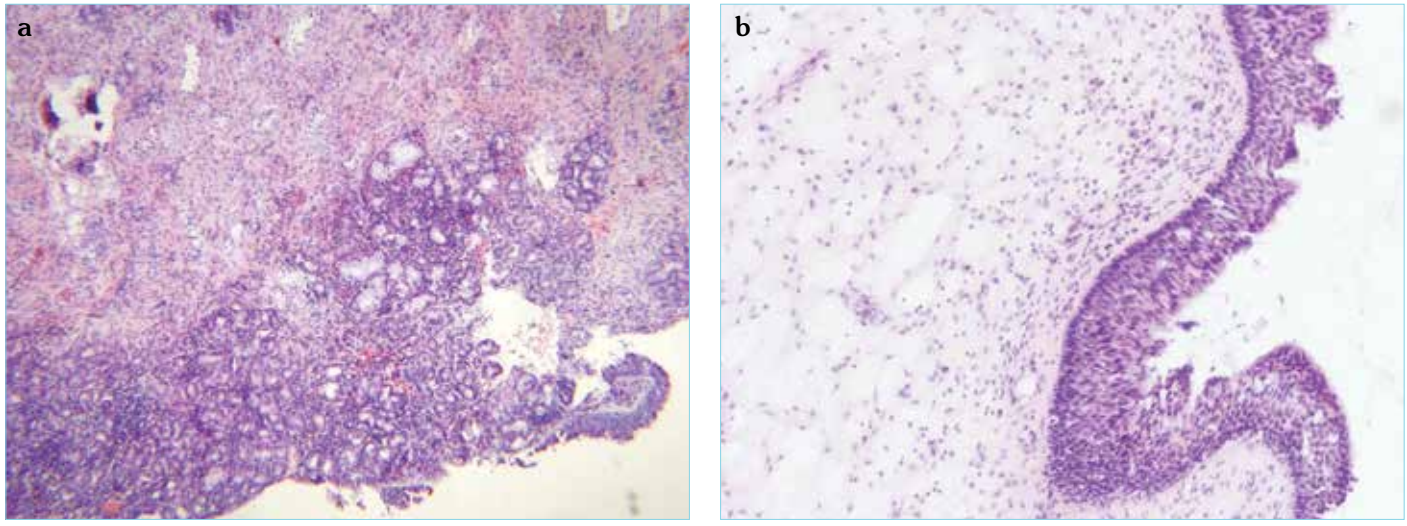

Figure 3. a, b. Microphotograph of the excised right-sided lesion (a). Histopathological examination results showed a lesion that was covered with ciliated pseudostratified respiratory epithelium and that comprised proliferated glandular spaces with ducts of various sizes (hematoxylin and eosin staining, magnification 40×). Histopathological findings of the excised left-sided nasal polyps (b). The microphotograph shows a proliferation of ciliated respiratory epithelium with stromal edema and strong inflammatory infiltrate predominantly comprised eosinophils (hematoxylin and eosin staining, magnification 100×).

ratinizing, stratified squamous type of epithelium that invaginates in the underlying stroma. The main histological characteristic of adenocarcinoma is the proliferation of infiltrating and fast-growing tubular glandular formations, which are covered by epithelium with different levels of cellular atypia (10). REAH must be histologically differentiated from other lesions. Mistaking REAH for an inverted papilloma and adenocarcinoma may result in an overly aggressive treatment of this benign lesion. Conversely, a misdiagnosis of REAH as chronic rhinosinusitis with nasal polyps may result in an inadequate medical treatment usually comprising intranasal/systemic glucocorticoids and/or macrolide antibiotics.

CONCLUSION

Respiratory epithelial adenomatoid hamartoma is a well-defined pathological entity but is unknown to many clinicians and pathologists. This lesion may accompany inflammatory nasal polyposis. However, the simultaneous presentation of REAH and inflammatory nasal polyps in the opposite sides of the nasal cavity is an extremely rare clinical finding. Appropriate cooperation between a rhinologist and pathologist enables a correct diagnosis.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Military Medical Academy Faculty of Medicine (MFVMA 06/16-18/).

Informed Consent: Written informed consent was obtained from patient who participated in this study.

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