

Case Report

Seromucinous hamartoma of the bilateral nasal cavities: a case report

Bo-Nien Chen^{1,3}, Kuo-Ming Chang²

Departments of ¹Otolaryngology-Head and Neck Surgery, ²Pathology, Hsinchu MacKay Memorial Hospital, Hsinchu, Taiwan; ³Mackay Junior College of Medicine, Nursing, Management, New Taipei, Taiwan

Received January 17, 2017; Accepted June 6, 2017; Epub July 15, 2017; Published July 30, 2017

Abstract: Objectives: Seromucinous hamartoma, first described by Baillie and Batsakis in 1974, is a rare benign glandular proliferation of the sinonasal tract and nasopharynx. Herein, we present a case of seromucinous hamartoma arising in the bilateral nasal cavities. Case report: We present a 43-year-old woman with seromucinous hamartoma of the bilateral nasal cavities who presented with nasal obstruction and anosmia. The patient underwent endoscopic sinus surgery and was disease-free at the most recent follow-up, 3 years later. Results: In this paper, we review the literature and compare it to our case. Based on our research, this is the first case report of seromucinous hamartoma of the bilateral nasal cavities. Conclusions: This case report indicates that seromucinous hamartoma should be considered in the differential diagnosis of bilateral nasal tumors, especially those that arise from the posterior nasal cavities medial to the middle turbinates.

Keywords: Seromucinous hamartoma, respiratory epithelial adenomatoid hamartoma, low grade sinonasal adenocarcinoma

Introduction

Seromucinous hamartoma (SH), first described by Baillie and Batsakis in 1974, is a rare benign glandular proliferation of the sinonasal tract and nasopharynx [1]. To date, 24 cases of SH have been reported in the English language literature (**Table 1**) [1-4]. Herein, we present a case of SH arising in the bilateral nasal cavities. Based on our research, this is the first case report of SH of the bilateral nasal cavities.

Case report

A 43-year-old woman visited our hospital presenting with a long-standing bilateral progressive nasal obstruction and 5-month anosmia. Sinoscopy revealed papillomatous masses in the bilateral nasal cavities. Computed tomography of the sinonasal cavity revealed polypoid lesions in the bilateral nasal cavities medial to the middle turbinates. The bilateral olfactory clefts were obliterated by the lesions (**Figure 1**). The lesions were excised using endoscopic sinus surgery assisted by microdebrider. Intraoperatively, a pedunculated mass based in the sphenoid recess was discovered bilaterally.

The histopathological sections of the lesions of the bilateral nasal cavities showed a proliferation of individual medium- to large-sized glands lined by pseudostratified respiratory epithelia (of both mucinous and ciliated types), arising in direct continuity with the surface epithelium and invaginating downward into the stroma. Many of these glands were enveloped by prominent thickened hyalinized basement membranes (**Figure 2A**). Some individuals and clusters of various amounts of small serous glands were observed among or near the medium-sized mucous glands in a mildly haphazard scattered or, to a rarer extent, mildly crowded pattern without typical lobulated architecture (**Figure 2B**). Thus, SH of the bilateral nasal cavities was diagnosed.

The patient recovered well from the surgery and was disease-free at the most recent follow-up, 3 years later.

Discussion

Sinonasal hamartomas are considered uncommon and have been clearly defined only in recent years. Sinonasal hamartomas have been designated as epithelial, mesenchymal, and

Seromucinous hamartoma of the bilateral nasal cavities

Table 1. Summary of 24 cases of SH have been published in the English literature

Authors	Cases	Age	Gender	Location
Baillie et al. (1974)	1	26	M	Nasopharynx
Zarbo et al. (1983)	1	32	M	Nasopharynx
Graeme-Cook et al. (1992)	3	57, 63, 78	F = 3	Posterior nasal cavity = 2, nasopharynx = 1
Chuang et al. (2000)	1	54	M	Paranasal sinus
Weinreb et al. (2009)	7	14-86	M = 4, F = 3	Posterior septum = 6, lateral nasal wall = 1
Ambrosini-Spaltro et al. (2010)	5	49-66	M = 2, F = 3	Nasal cavity = 4, septum = 1
Figures et al. (2010)	1	72	M	Anterosuperior septum with skull base involvement
Khan et al. (2011)	2	59, 61	F = 2	Nasal cavity = 1, nasopharynx = 1
Fleming et al. (2012)	1	53	F	Posterior nasal cavity extending to nasopharynx
Huang et al. (2015)	2	34, 35	F = 2	Nasal cavity = 2



Figure 1. Computed tomography of the sinusal cavity of the patient. Coronal computed tomography revealed lesions in bilateral posterior nasal cavities medial to the middle turbinate (arrows). The bilateral olfactory clefts were obliterated.

mixed. The majority of sinonasal hamartomas is of the epithelial type and includes respiratory epithelial adenomatoid hamartoma (REAH) and SH [4]. However, mixed type of sinonasal hamartomas, such as chondro-osseous respiratory epithelial adenomatoid hamartoma (COREAH), is an extremely rare, developmental malformation [5]. REAH and SH lesions may belong to the same spectrum rather than being two separate entities. Foci of SH are frequently observed in REAH, and vice versa [2]. Their pathogenesis remains uncertain, with inflammatory and neoplastic origins proposed [6].

The male-to-female ratio of SH occurrence is approximately 1:1. Patients are most often middle-aged to elderly [2].

The most common presenting symptoms are nasal obstruction and epistaxis, although most patients are asymptomatic and the lesions are discovered incidentally [2, 4]. However, the only presenting symptom of the patient of this report was nasal obstruction. The patient suffered from anosmia subsequently and we speculated that the anosmia was caused by the obliteration of bilateral olfactory recesses. Finally, the olfaction of the patient was recovery after surgical excision of SH.

With rare exceptions, most cases of SH have arisen in either the posterior nasal cavity medial to the middle turbinate or the nasopharynx [2, 3]. This feature was also observed in our case. Moreover, Lima et al. suggested hamartoma must be suspected on CT scan enlargement of the olfactory clefts [7].

Histopathologically, the most crucial differential diagnosis for SH is low-grade sinonasal adenocarcinoma, of the nonintestinal type (LGSNAC), because the latter may require more extensive surgery and possibly radiotherapy [2, 4]. Immunohistochemistry does not contribute toward differentiating SH and LGSNAC [2]. However, in contrast to SH, LGSNAC typically exhibits complex growth patterns including micropapillary architecture, and glands that are back-to-back or fused [2, 8].

SH is typically cured by simple surgical excision. After treatment with surgical excision, there has been only one report of a recurrence [2-4]. Our case was disease-free at the most recent follow-up, 3 years later.

Conclusion

Our case demonstrates two pertinent points. First, this is the first case report of SH of the

Seromucinous hamartoma of the bilateral nasal cavities

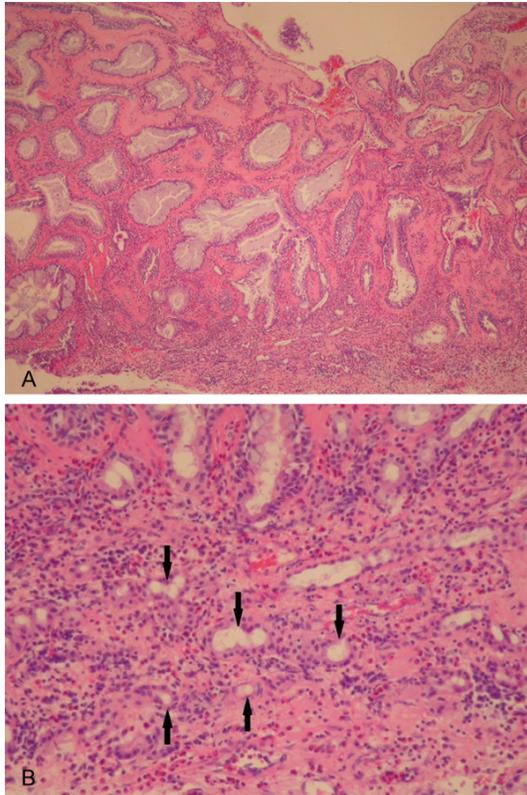


Figure 2. Histopathological features of the lesion. A. Proliferation of respiratory glands arising in direct continuity with the surface epithelium and invaginating downward into the stroma (H&E 200 \times). B. High power field of the lower part of the Figure; some individuals and clusters of serous glands without normal lobulated architecture (arrows) (H&E 300 \times).

bilateral nasal cavities. Second, SH is extremely rare but should be considered in the differential diagnosis of bilateral nasal tumors, especially those that arise from the posterior nasal cavities medial to the middle turbinates.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Bo-Nien Chen, Department of Otolaryngology-Head and Neck Surgery, Hsinchu MacKay Memorial Hospital, No. 690, Sec. 2, Guangfu Road, East Dist, Hsinchu 30071, Taiwan. Tel: +886 36119595; Fax: +886 36110900; E-mail: chenbonien@yahoo.com.tw

References

- [1] Baillie EE, Batsakis JG. Glandular (seromucinous) hamartoma of the nasopharynx. *Oral Surg Oral Med Oral Pathol* 1974; 38: 760-762.
- [2] Fleming KE, Perez-Ordoñez B, Nasser JG, Psooy B, Bullock MJ. Sinonasal seromucinous hamartoma: a review of the literature and a case report with focal myoepithelial cells. *Head Neck Pathol* 2012; 6: 395-399.
- [3] Huang CC, Lee TJ, Huang CC, Wu PW. Seromucinous hamartoma in the nasal cavity medial to the middle turbinate: report of 2 cases and review of the literature. *Head Neck* 2015; 37: E15-18.
- [4] Khan RA, Chernock RD, Lewis JS Jr. Seromucinous hamartoma of the nasal cavity: a report of two cases and review of the literature. *Head Neck Pathol* 2011; 5: 241-247.
- [5] Perić A, Vukomanović-Durđević B, Stanojević I. Middle turbinate chondro-osseous respiratory epithelial adenomatoid hamartoma. *B-ENT* 2015; 11: 239-243.
- [6] Bullock MJ. Low-grade epithelial proliferations of the sinonasal tract. *Head Neck Pathol* 2016; 10: 47-59.
- [7] Lima MB, Jankowski R, Georgel T, Grignon B, Guillemin F, Vignaud JM. Respiratory adenomatoid hamartoma must be suspected on CT scan enlargement of the olfactory clefts. *Rhinology* 2006; 44: 264-269.
- [8] Jo VY, Mills SE, Cathro HP, Carlson DL, Stelow EB. Low-grade sinonasal adenocarcinomas: the association with and distinction from respiratory epithelial adenomatoid hamartomas and other glandular lesions. *Am J Surg Pathol* 2009; 33: 401-408.