Clinical Case Report

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Seromucinous hamartoma of inferior turbinate

A case report

Dong Hoon Lee, MD, Tae Mi Yoon, MD, Joon Kyoo Lee, MD, Sang Chul Lim, MD*

Abstract

Rationale: Seromucinous hamartoma is a rare benign glandular proliferation of the sinonasal tract and nasopharynx, and the majority of lesions occur on the posterior nasal septum.

Patient concerns: The patient had complaints of rhinorrhea, sneezing, and itching for a number of years, and recurrent right nasal bleeding for which the patient underwent endoscopic removal of a right inferior turbinate tumor. The biopsy result was low-grade, non-intestinal type adenocarcinoma, and the patient was referred to our hospital.

Diagnosis and Interventions: An endoscopic medial maxillectomy of the right nasal cavity was performed. The histopathological analysis of the nasal mass revealed a seromucinous (glandular) hamartoma.

Outcomes: The postoperative course was unremarkable. The patient has been followed up regularly for 1 year with no additional treatment and no recurrence.

Lessons: Seromucinous hamartoma of the nasal cavity is extremely rare, especially in anterior portion of nasal cavity. It is important to distinguish seromucinous hamartoma from adenocarcinoma and to maintain regular long-term follow-up.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging, PET-CT = positron emission tomography-CT.

Keywords: adenocarcinoma, glandular, hamartoma, surgical procedure, turbinate

1. Introduction

Glandular neoplasia of the sinonasal tract is divided into benign tumors and malignant tumors.[1–4] Distinguishing between benign hamartoma and adenocarcinoma is important because benign tumors have rarely been reported to recur.[1–5] Seromucinous hamartoma is a rare benign glandular proliferation of the sinonasal tract and nasopharynx, and the majority of lesions occur on the posterior nasal septum.[1–4] Herein, we report an unusual case of a patient with seromucinous hamartoma at the anterior end of the right inferior turbinate that was mistaken for low-grade, non-intestinal-type adenocarcinoma.

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2. Case report

A 40-year-old woman was referred to our hospital after endoscopic removal of a right inferior turbinate tumor at a local hospital. The patient had had rhinorrhea, sneezing, and itching for a number of years, and recurrent right nasal bleeding had occurred 2 months prior. On endoscopic image taken at the local hospital, there was a round, smooth-margined, polypoid 1-cm mass at the anterior end of the right inferior turbinate (Fig. 1). However, the biopsy result was low-grade, non-intestinal-type adenocarcinoma, and the patient was referred to our hospital. Physical examination of the nasal cavity, head, and neck was unremarkable except for an ulcerative lesion at the anterior end of the right inferior turbinate due to previous endoscopic removal at the local hospital (Fig. 2). Computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated no definite focal enhancing lesion in the nasal cavity. However, positron emission tomography-CT (PET-CT) showed a focal hypermetabolic lesion at the right inferior turbinate (Fig. 3).

Based on these observations, we performed endoscopic medial maxillectomy of the right nasal cavity, and the histopathological analysis of the nasal mass revealed a seromucinous (glandular) hamartoma (Fig. 4). The postoperative course was unremarkable. The patient has been followed up regularly for 1 year with no additional treatment and no recurrence. This study was approved by the institutional review board of the Chonnam National University Hwasun Hospital. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

3. Discussion

Seromucinous hamartoma in the nasal cavity is a rare lesion.[1–4] The etiology of hamartoma is unknown, but chronic inflammation of the nasal mucosa associated with chronic rhinosinusitis,

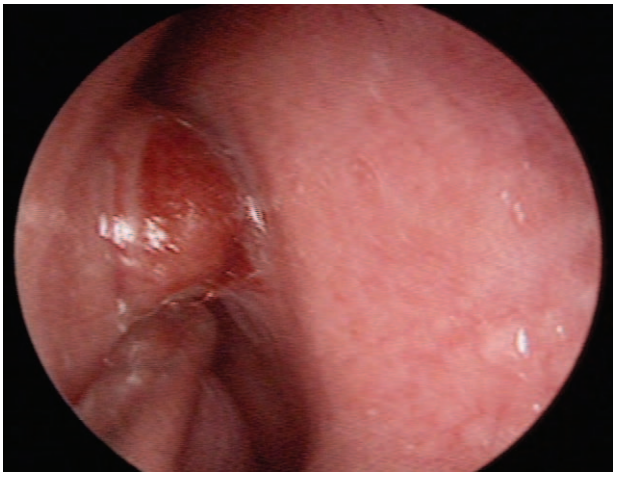

Figure 1. Nasal endoscopy reveals a round, polypoid mass in the right inferior turbinate.

allergic rhinitis, or bronchial asthma is the most accepted hypothesis.[5] The lesion occurs predominantly in women, and the most common symptom is unilateral nasal obstruction, followed by nasal bleeding.[1–4] The majority of these lesions occurred in either the posterior nasal cavity or the nasopharynx, and most have occurred from the posterior nasal cavity medial to the middle turbinate.[1–4] To our knowledge, this is the first case of seromucinous hamartoma to arise from the anterior nasal cavity.

Glandular neoplasia of the sinonasal tract is divided into benign tumors, such as respiratory epithelial adenomatoid hamartomas, seromucinous hamartomas, and malignant tumors, including intestinal- and non-intestinal-type adenocarcinomas.[3,4] These lesions have similar clinical presentations and histopathologic and immunohistochemical features.[1–5] In addition, there are no criteria for definitive distinction between these lesions.[4]

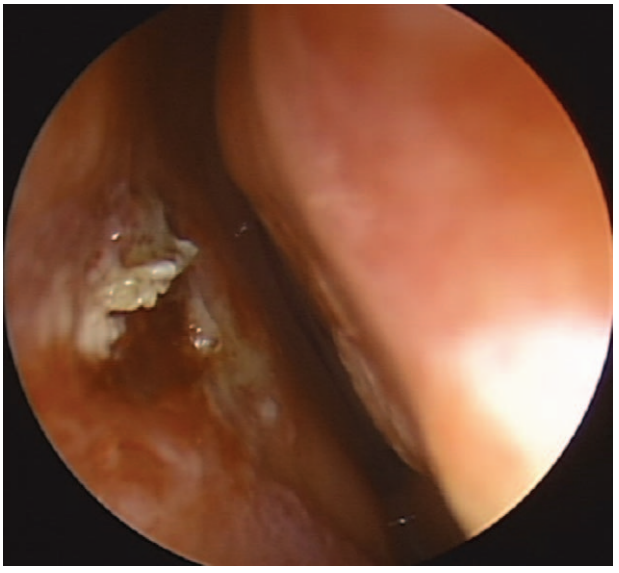

Figure 2. Nasal endoscopy shows an ulcerative lesion in the right inferior turbinate due to previous endoscopic removal at a local hospital.

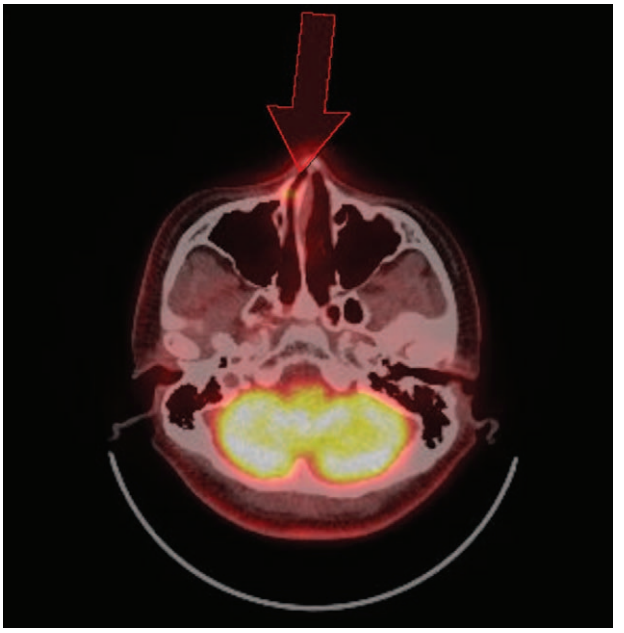

Figure 3. PET-CT shows a focal hypermetabolic lesion (arrow) on the right inferior turbinate. PET-CT = positron emission tomography-computed tomography.

However, distinguishing between benign hamartoma and adenocarcinoma is important because benign tumors have rarely been reported to recur, unlike adenocarcinomas.[1–5] Unlike respiratory epithelial adenomatoid hamartomas, seromucinous hamartomas consist of epithelial proliferations of mixed small and large glands.[2,4] Adenocarcinomas show crowded glandular proliferations, pleomorphism, mitoses, and varying degrees of cellular atypia.[5] However, histological distinction is sometimes difficult if the biopsy sample is too small, as was the case with our patient.

The treatment of choice for seromucinous hamartoma is complete surgical excision by transnasal endoscopic approach,[1,5] and following adequate endoscopic tumor excision,

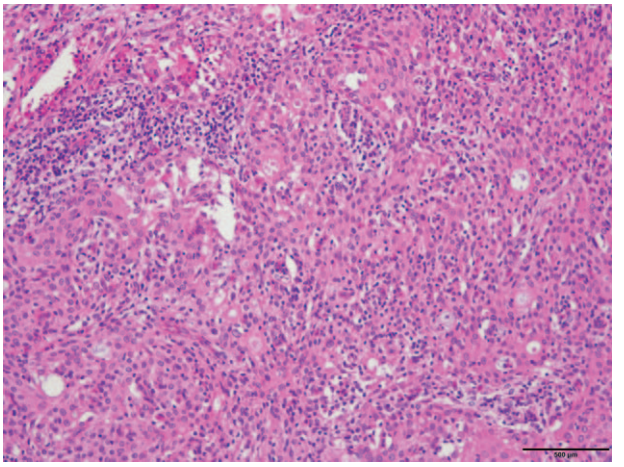

Figure 4. Histopathologic findings of a nasal tumor at the anterior end of the right inferior turbinate show that small tubules, ducts, and glands are present in lobular and more haphazard arrangements, lined by bland cuboidal cells (hematoxylin and eosin, original magnification ×200).

recurrence rates are very low.[1,4] In this case, we performed endoscopic medial maxillectomy because the initial diagnosis was low-grade, non-intestinal-type adenocarcinoma. As with our initial misdiagnosis, because there is no definitive distinction method, the biological potentials of glandular neoplasias of the sinonasal tract have not been investigated.[3] Therefore, long-term follow-up is necessary with endoscopic and radiologic examinations.

In conclusion, seromucinous hamartoma of the nasal cavity is an extremely rare benign disease, especially at the anterior end of the inferior turbinate. It is important to distinguish seromucinous hamartoma from adenocarcinoma, and to maintain regular long-term follow-up.

Author contributions

Data curation: Dong Hoon Lee, Tae Mi Yoon.
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Formal analysis: Joon Kyoo Lee.
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Investigation: Sang Chul Lim.

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