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Antrochoanal polyp concomitant with turbinoethmoidal osteoma: A case report

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ABSTRACT

INTRODUCTION: Antrochoanal polyps (ACPs) have several unique features that distinguish them from other nasal polyps. Despite many investigations, the pathogenesis of ACP has not yet been elucidated. Sinonasal osteoma is the most common type of tumor of the sinonasal tract and can be caused by trauma, infection and developmental anomaly.
PRESENTATION OF CASE: We report the case of a 35-year-old man with left nasal obstruction for more than 20 years. Examination revealed ACP concomitant with an osteoma that arose from the middle turbinate and ethmoid sinus. The osteoma had an air cell in its anterosuperior area and was in a position of being pulled downward. The intranasal part of the ACP covered the posterior area of the osteoma.
DISCUSSION: Considering the radiological and surgical findings, the intranasal part of the ACP seems to have affected the turbinoethmoidal osteoma during its growth.
CONCLUSION: The authors describe a very rare condition in which an ACP was connected with a turbinoethmoidal osteoma.

1. Introduction

Antrochoanal polyps (ACPs) are benign polypoid lesions that originate from the inside of the maxillary sinus and extend to the posterior choana through the natural or accessory ostium. ACP is thought to be caused by conditions that cause cystic changes to the glands of the antrum, such as chronic inflammation or allergies [1]. However, the increased density of lymphatic vessels found at the origin sites of ACPs suggests primary or secondary lymphatic obstruction and lymphatic malformations as the cause of ACP [2]. Previous studies have reported on ACPs showing atypical stromal cells and vascular reactive processes such as neovascularization, thrombosis, hemorrhage and vascular hyperplasia [3,4].

Sinonasal osteoma is the most common type of benign tumor of the sinonasal tract and is found in approximately 3% of the population [5]. Several cases of osteoma in the middle turbinate have been reported thus far [6]. In line with the SCARE criteria, we describe a rare case of ACP combined with osteoma arising from the middle turbinate and ethmoid sinus, in which the intranasal part of the ACP covered the posterior area of the osteoma [7]. To the best of our knowledge, this condition has never been reported.

2. Presentation of case

A 35-year-old male patient presented with left nasal obstruction for more than 20 years. He had no other symptoms. He did not complain of post-nasal drip, headache, or epiphora and had no symptoms of allergies, such as sneezing or itching. He denied chronic systemic illness and history of head trauma. Approximately 5 months prior, he had visited a private ENT clinic to undergo a polypectomy under local anesthesia. However, the procedure was unsuccessful because of the hard consistency.

Endoscopy revealed a mass lesion that had a bony consistency and adhered to the anterior end of the inferior turbinate.

Computed tomography of the paranasal sinuses showed a soft tissue density lesion occupying the maxillary sinus and nasal cavity on the left side. This lesion extended to the nasopharynx through the posterior choana (Fig. 1). A $3.5 \times 3 \times 2$ cm irregularly shaped calcified mass was observed inside the soft tissue density lesion. The anterosuperior area of the calcified mass contained a 9 mm oval cell. The air cells of the ethmoid sinus were not seen. The middle turbinate was not observed either, except for a remnant of the lamellar portion anteriorly and the insertion site to the skull base posteriorly (Fig. 2A and B). The frontal and sphenoid sinuses were normally pneumatized.

Endoscopic sinus surgery was performed under general anesthesia. The mass lesion was connected to the anterior remnant of the middle turbinate by a fibrotic mucosal fold. Polypoid mucosa covered the posterior area of the bony lesion and was a part of the

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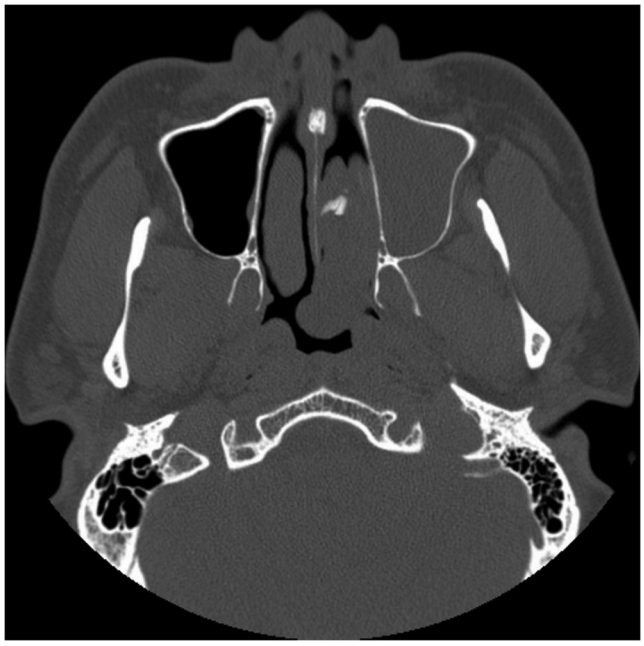

Fig. 1. An axial computed tomography image showing a soft tissue density occupying the left maxillary sinus and passing through the posterior choana to the nasopharynx.

intranasal polyp that extended continuously from the medial side of the maxillary sinus to the nasopharynx. After dissecting the mucosa using a microdebrider and an elevator, the bony mass was removed (Fig. 3). The maxillary sinus was occupied with cystic lesions from the same origin without any other pathologic findings.

Histopathologically, the bony mass was consistent with ivory type osteoma and the soft tissue was consistent with inflammatory polyp.

After surgery, the patient' symptoms disappeared, and he recovered without any complications. There was no recurrence after 2 years of follow-up.

3. Discussion

Sinonasal osteoma can be caused by trauma or infection, but it has recently been recognized as a developmental anomaly. It grows very slowly and occurs mainly in the frontoethmoidal region [5,8].

The osteoma in the present case appears to have arisen from the middle turbinate and ethmoid sinus, and it differs from those reported in the past in three main aspects. First, the intranasal part of the ACP covered the posterior area of the osteoma. Second, the osteoma was pulled downward and was not in an expected position. Finally, a single cell was identified inside the osteoma.

The adhesions to the inferior turbinate observed at the anterior end of the osteoma in the present case may have occurred during the attempted polyp removal 5 months prior. However, the mucosal continuity of the posterior area of the osteoma to the intranasal part of the ACP did not have an iatrogenic origin because the polypoid mucosa which was a part of the ACP covered most of the posterior area of the osteoma. Therefore, this condition seems to be associated with the two other findings.

The entire osteoma was not in a position where the ethmoid cells would be expected to be present, but it was in a shape that pulled downward from the insertion site of the middle turbinate. Additionally, the fact that the osteoma and the vertical lamellar part of the middle turbinate were thinly connected by only a fibrotic mucosal fold implies that a pulling force was acting from the posteroinferior side, which is consistent with the surgical finding that

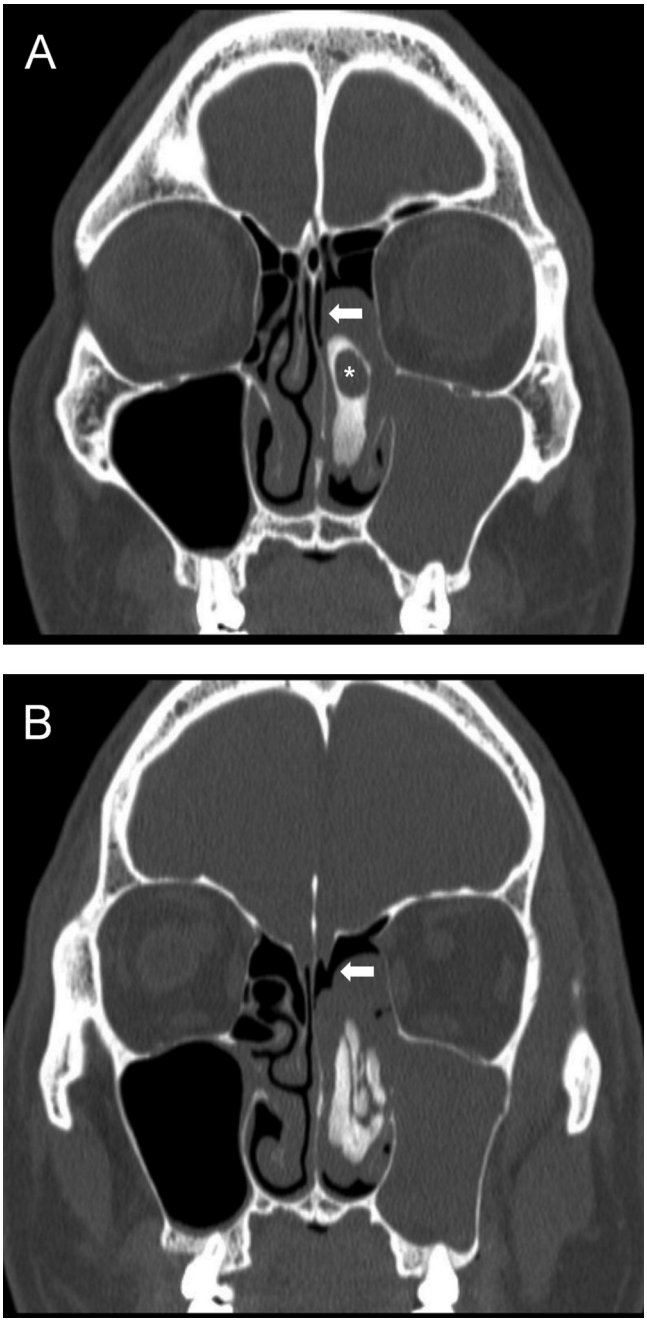

Fig. 2. Coronal computed tomography scans show a calcified mass in the left nasal cavity. (A) This mass showed a single cell in the anterosuperior area (asterisk) and a connection to the remnant of the middle turbinate (arrow). (B) The middle turbinate and ethmoid cells were not seen except for the insertion site to the skull base (arrow). Note that the calcified mass is positioned downward.

the intranasal part of the ACP covered the posterior area of the osteoma.

The cell present in the osteoma was presumed to originate from one of the frontoethmoidal air cells in the frontal recess area because of its location and size. This could indicate that the anterosuperior part of the osteoma might have been the last area to be involved. This condition might also be explained by the intranasal part of the ACP that covered the posterior area, exerting a pulling force on the osteoma during its slow and continuous development.

The frontal sinus is thought to originate from frontal furrows or pits from the anterior ethmoid cells and remains a small blind pocket until 1 year after birth. Secondary pneumatization begins

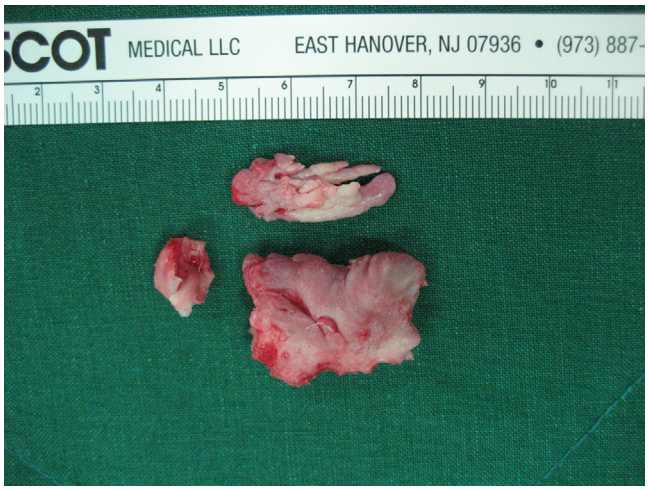

Fig. 3. The irregular shaped osteoma that was extirpated completely by endoscopic sinus surgery.

after the age of 2 years and can be radiologically identified after the age of 4 years. The frontal sinus continues to grow through puberty until about 20 years of age [9,10]. The well-pneumatized ipsilateral frontal sinus of the patient showed that the osteoma had not disturbed the development of the frontal recess and sinus.

Therefore, the intranasal part of the ACP appears to have had a sustained pulling effect during the growth of the osteoma. However, it is not clear when the ACP began to affect the osteoma. Further, the adhesion of the ACP to the developing ethmoid cells may have led to the development of a traumatic force, causing the osteoma to occur. If the ACP and the osteoma developed independently, it is not clear how the ACP, which originated from the inside of the maxillary sinus, become connected to the posterior area of the osteoma. It seems difficult to confirm the above-mentioned possibilities. However, considering that mature polyps do not spontaneously fuse with surrounding structures, it is possible that the ACP occurred in the vicinity of the osteoma at a very early age, possibly at the embryonic stage.

4. Conclusion

The authors successfully treated a patient with ACP concomitant turbinoethmoidal osteoma by endoscopic sinus surgery. The intranasal part of the ACP covered the posterior area of the osteoma and seems to have affected the osteoma during its growth.

Conflicts of interest

None.

Funding

None.

Ethical approval

This study was exempted by the Institutional Review Board of Veterans Health Service Daejeon Hospital.

Consent

Informed consent was obtained from the patient included in the study.

Author contribution

Yong Won Lee operated the patients.
Yong Won Lee prepared the first draft of themanuscript.
Yong Min Kim revised the manuscript for important intellectual content.
All authors have read and approved the final manuscript.

Guarantor

Yong Won Lee

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