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Case Report

Organized hematoma of the sphenoid sinus with epistaxis ☆,☆☆

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ABSTRACT

Few reports have been made on organized hematoma (OH) originating in the sphenoid sinus. We report the case of a 24-year-old man who presented to the hospital with consistent epistaxis for 1 month. Preoperative computed tomography and magnetic resonance imaging (MRI) detected an expansive heterogeneous mass with minimal bone destruction and hypointense peripheral rim in T2-weighted images of the left sphenoid sinus as a solitary lesion. An OH was suspected first based on the history of repeated epistaxis and characteristic imaging findings, although quite rare. Preoperative vascular embolism was not performed because its effects on the ocular artery should be considered. With endoscopic sinus surgery under hypotensive anesthesia, the mass was carefully and successfully removed without bleeding and diagnosed as OH based on postoperative pathological examination. When solitary lesions are found in the sphenoid sinus with epistaxis, this disease should be differentiated. If it increases, complications, such as visual impairment, frequently occur, and control of intraoperative bleeding is sometimes difficult. Not only missing the characteristic imaging findings, especially MRI with contrast administration but also evaluating the anatomical relationship between OH and other important organs is important for a successful intervention of sphenoid sinus OH.

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Introduction

Sinonasal organized hematoma (OH) is a rare, non-neoplastic, benign condition with expansive and destructive characteristics usually necessary to differentiate it from a malignancy. Organized hematoma can be curative when the lesion is completely removed. However, these tumors can be overtreated or misdiagnosed preoperatively because of the risk of malignancy [1]. Most OHs are derived from the maxillary sinus, but only four detailed reports have been published on OH of the sphenoid sinus [2–5]. A lot of important structures are adjacent to the sphenoid sinus, such as the dura, optic nerve, and internal carotid artery, which are vulnerable to injury with sphenoid disease [6], and visual impairment [3–5], severe adhesion [2], and intraoperative massive bleeding [5] are problematic. An accurate preoperative diagnosis and knowing the possibility of OH in what cases should be considered when solitary lesions are important. Herein, we report a case of OH in the sphenoid sinus with epistaxis, and the clinical assessment, imaging, and surgical approaches are also discussed for the early diagnosis and appropriate intervention.

Case report

A 24-year-old man had a history of bronchial asthma visited our hospital with a complaint of repeated left epistaxis for 1 month. He was referred to our department by a nearby otolaryngologist because of the unknown cause of epistaxis. Nasal endoscopy revealed bloody rhinorrhea persisting from the left sphenoid sinus orifice (Fig. 1A). No abnormal findings were observed in other otolaryngological examinations. He had no history of physical trauma or metabolic disease.

On computed tomography (CT), expansive mass with minimal bone destruction was observed only in the left sphenoid sinus (Figs. 2A–C), and the sphenoid sinus density was somewhat heterogeneous under soft-tissue conditions (Fig. 2A). The CT value in the lesion was derived from -43 to 106 Hounsfield unit (Fig. 2A). Magnetic resonance imaging (MRI) showed an internal heterogeneous high-intensity region in the left sphenoidal sinus on both T1- (Fig. 3A) and T2-weighted (Fig. 3B) images. A marked low-intensity region, hypointense peripheral rim, was also observed inside the sphenoid sinus mucosa on the T2-weighted image (Figs. 3B and C). Contrast enhancement examination could not be performed because adverse reactions to iodinated contrast media are more likely to develop in patients with asthma [7]. The patient was first suspected of diagnosing OH from the characteristic MR imaging findings described above. As a differential diagnosis, other lesions are rich in blood flow based on the history of repeated epistaxis. However, CT and MRI could not show a rich blood flow of this lesion without contrast enhancement examination.

Preoperative embolism was not performed by considering the effects on the ocular artery. After explaining to the patient that another surgery may be required if malignant findings were found by intraoperative rapid pathological diagnosis, endoscopic sinus surgery was performed 21 days after the

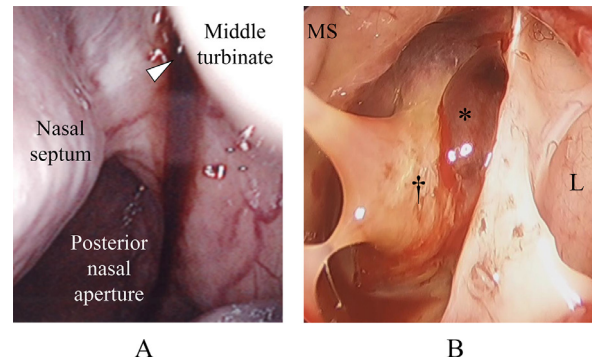


Fig. 1 – Endoscopic findings. (A) Nasal cavity (first visit), (B) Left sphenoid sinus (intraoperative). Persistent bloody rhinorrhea (arrowhead) was observed from the orifice of the left sphenoid sinus (A). In the left sphenoid sinus, a yellowish-white granulation-like part (†) and a dark red hematoma-like part (*) were mixed (B). The optic nerve is located laterally upward and is outside this field of view (B). MS, medial septum of sphenoid sinus; L, left side.

first visit. After sufficiently enlarging the left sphenoid sinus orifice, a broad base mass was observed. The mass had both a yellowish-white granulation-like part and a dark reddish hematoma-like part (Fig. 1B). Fortunately, no lesions were detected on the optic nerve (Fig. 1B). Intraoperative rapid pathological examination showed no possibility of neoplasm; thus, the mass was successfully removed piece-by-piece without severe adhesion. To reduce bleeding, the intraoperative blood pressure was maintained at 80–90 mm Hg, and the total blood loss was 10 mL.

Postoperative histopathological examination revealed an OH diagnosis with deposition of hemosiderin, fibrous connective tissue, and squamous and ciliated cylindrical epithelium in part (Fig. 4A). No neoplastic lesions or malignancies were observed. Postoperatively, epistaxis promptly disappeared, and a postoperative CT scan showed that the aeration of the left sphenoid sinus was recovered (Fig. 4B). No recurrence occurred for >5 years.

Discussion

Among the many case reports of OH, those derived from the maxillary sinus account for most cases: 15/17(88.2%) [8], 82/84(97.6%) [9], and 15/23(65.2%) cases [10]. Excluding the nasal cavity origin, OH from the frontal [8] and ethmoid sinuses [10] is rare with only 1 case each reported. Those from the sphenoid sinus are also rare with only 4 previously reported cases [2–5], except for one of the seven reported cases (a 75-year-old woman) with unknown details [11], and one of 17 cases (a 35-year-old man, [8]), which is considered to be the same as in Yoon's report [4]. Pathologically, blood retention caused by trauma, surgery, hemorrhagic diathesis, and mechanical damage to arterial branches is not reabsorbed due to insufficient ventilation and excretion, and repeated intra-

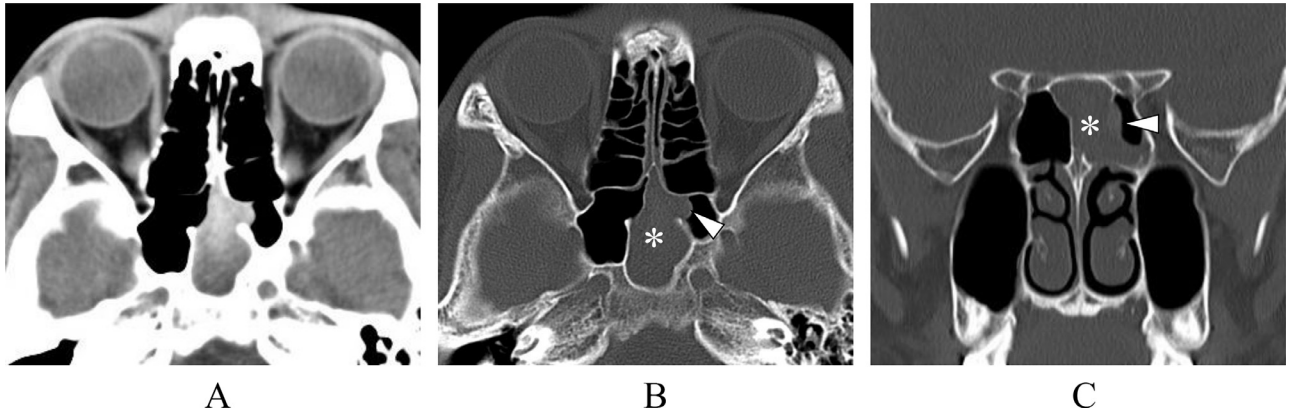


Fig. 2 – Preoperative plain CT findings. (A) Soft-tissue condition (axial image), (B) Bone condition (axial image), (C) Bone condition (coronal image). An internal heterogeneous solitary lesion was observed in the left sphenoid sinus (A), and an expansive mass (*) with bone thinning (arrowhead) was observed laterally anterior to the lesion in the left sphenoid sinus (B and C).

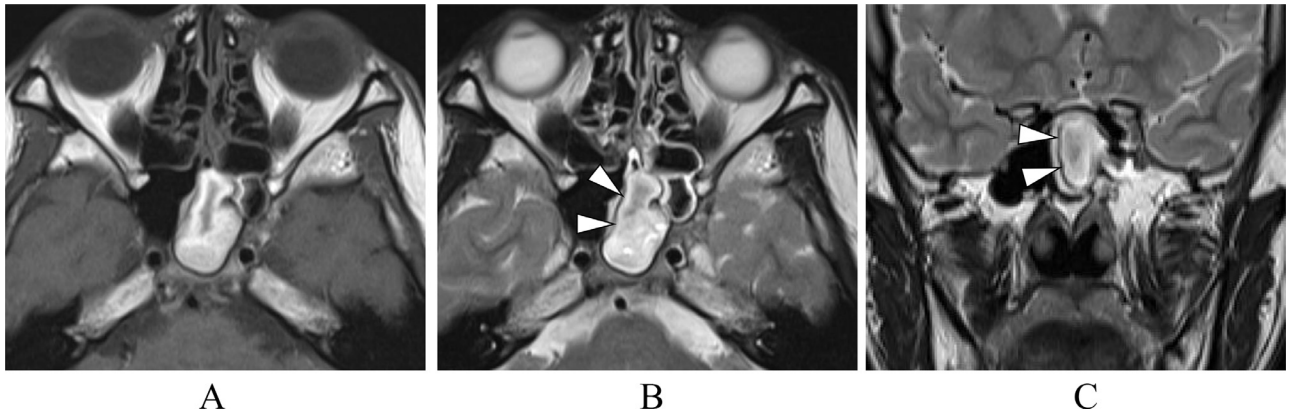


Fig. 3 – Preoperative nonenhanced MRI findings. (A) T1-weighted axial image, (B) T2-weighted axial image, (C) T2-weighted coronal image. Lesions of the left sphenoid sinus were heterogeneous high-intensity internally in both T1- (A) and T2-weighted images (B and C), and a marked low-intensity rim (arrowhead) was observed inside of high-intensity area which shows sphenoid sinus mucosa (B and C).

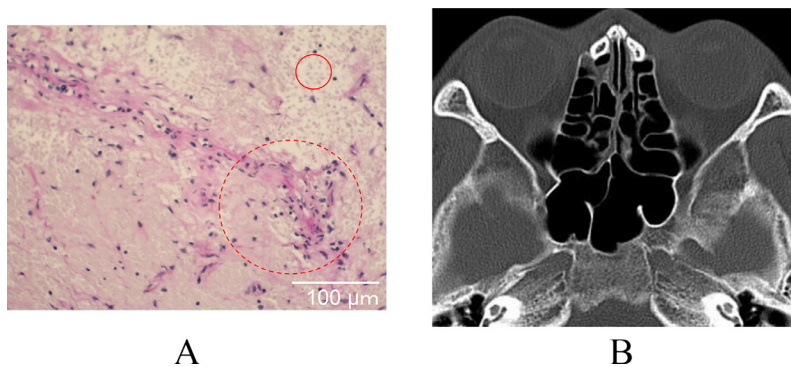


Fig. 4 – Postoperative findings. (A) Histopathological findings (PAS staining), (B) postoperative plain CT findings (bone condition, axial image). Both a collection of red blood cells reminiscent of bleeding (small red circle) and a mixture of bleeding, vessels, hemorrhage, hemosiderin, and fibrosis connective tissues (large dotted red circle) are observed (A). The aeration of the left sphenoid sinus was recovered, and no recurrence occurred (B).

Table 1 – Summary of previous reports of organized hematoma in the sphenoid sinus.

No.	Authors	Age (years)/sex	Side	Presentation	Past medical history	Computed tomography	Magnetic resonance imaging	Surgery
1	Nakagawa et al. [2]	85/F	L	Floating sensation	Hypertension, diabetes, hyperlipidemia	Expansile mass with bony erosion and patchy enhancement	T1: heterogenous T2: contained high-intensity and hypointense peripheral rim	Endoscopic sinus surgery
2	Lin YH et al. [3]	81/F	R	Headache, decreased VA, blepharoptosis	Chronic renal disease, sinus surgery	Bone destruction	T1: heterogenous T2: contained high-intensity and hypointense peripheral rim	Endoscopic sinus surgery
3	Yoon et al. [4]	35/M	L	Headache, orbital pain, decreased VA	Aplastic anemia, valvular heart disease	Expansile mass with bony erosion and patchy enhancement	N/A	Endoscopic sinus surgery
4	Lin et al. [5]	59/M	L	Headache, decreased VA, epistaxis	Sinus surgery	Middle cranial fossa defect, carotid artery erosion	T1: heterogenous T2: contained high-intensity and hypointense peripheral rim	Zygomatic extradural subtemporal approach
5	Kitaoka et al. (present case)	24/M	L	Epistaxis	None	Expansile mass with bony erosion	T1: heterogenous T2: contained high-intensity and hypointense peripheral rim	Endoscopic sinus surgery

F, female; L, left; M, male; R, right; VA, visual acuity; N/A, not available.

capsular hemorrhage with angiogenesis and fibrosis changes leads to OH formation [12–15]. It commonly occurs in the maxillary sinus because the maxillary sinus is most likely to cause negative pressure and decreased ventilation [13]. In this case, histopathological examination showed no hemangioma and no obvious hemorrhagic predisposition such as trauma or surgery.

Table 1 summarizes previous detailed reports of sphenoid sinus OHs. Remarkably 3 of 4 cases [3–5] had decreased visual acuity during the course. Report of 132 isolated sphenoid disease cases (80 with inflammation including mucocele, 38 tumors, 10 trauma and developmental disorders, and 4 fibrotic diseases), the most common symptom is headache, followed by visual changes and cranial nerve palsy [6]. Although epistaxis rarely occurs, that is in only 5/132 cases (2 traumatic aneurysms and 3 malignant tumors) [6], 2/5 cases of sphenoid sinus OH showed epistaxis (Table 1), which might be an important clinical finding. Our diagnosis was triggered by the bloody secretion flowing from the orifice of the sphenoid sinus that was not overlooked. If isolated sphenoid sinus disease is found and accompanied by epistaxis, the possibility of OH should be considered.

In addition to malignant tumors and mucocele, the differential diagnosis includes inflammatory diseases, such as polyps, fungal infection, and papilloma. Imaging findings of OH are internally heterogeneous, reflecting pathological bleeding and fibrous changes [12–15]. In typical cases, T1-weighted MRI shows heterogenous intensity and T2-weighted

MRI contains high-intensity and hypointense peripheral rim [1,2,4,6,14,16–18] and, as in our case, helped for the diagnosis of OH [17]. Previous studies reported that the frond-like pattern of contrast enhancement is one of the characteristic imaging findings of OH that helps to differentiate from other simulating lesions [13,17]. In our case and the previous reports [2–5], the sphenoid impact was only one side, but possible factors that influenced this were unknown.

Diagnosing using CT images alone is difficult, and compressive bone destruction with bone thinning, unlike malignant tumors, was characteristic [8,9]; however, no bone destruction occurred in 61.3% of OH [8]. The “polyp-like” mass at the top or side of the sphenoid sinus may represent a rare entity, such as a brain or internal carotid aneurysm [6], and should be well evaluated in preoperative images. Contrast enhancement is also important, and reports of 1000 mL intraoperative bleeding via the 4-hand approach have been published [5]. Moreover, the internal carotid artery was found to be close to OH ruptured during removal [19].

As for maxillary sinus OH, surgery is the first choice of treatment. Preoperative vascular embolism is commonly used in maxillary sinus OH; however, its effects on the ocular artery should be considered in the case of sphenoid sinus OH [5]. Inadvertent electrocoagulation intraoperatively can also affect other important organs, such as the optic nerve or internal carotid artery, and the positional relationship between OH and these organs must be sufficiently confirmed preoperatively. In this case, bleeding could be stopped by compression with

epinephrin-containing gauze, and the bleeding volume was small. This is thought to be associated with hypotensive anesthesia, and the operation should be carefully performed in order not to touch the mass until the surgical field is sufficiently secured, and early diagnosis before the mass enlargement is essential.

Conclusion

When solitary lesions are found in the sphenoid sinus with epistaxis, OH should be differentiated, although quite rare. If it increases, severe complications, such as visual impairment and massive bleeding, can occur, and control of intraoperative bleeding is sometimes difficult because preoperative vascular embolism and intraoperative electrocoagulation are difficult due to the anatomical characteristics of the sphenoid sinus. Early and appropriate preoperative imaging diagnosis, especially MRI with contrast administration, is needed to miss the characteristic imaging findings and evaluate the positional relationship between OH with the optic nerve and arteries, which is important for the successful intervention of OH in the sphenoid sinus.

Patient consent

Written informed consent was obtained from the patient for publication of this case report.

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