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Clinical characteristics of internal carotid artery pseudoaneurysms in the sphenoid sinus

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ABSTRACT

Background: Internal carotid artery (ICA) pseudoaneurysms are associated with high mortality if lack of proper management. Patients with ICA pseudoaneurysms in the sphenoid sinus often visit a hospital's ear, nose and throat (ENT) department due to nasal bleeding. In such cases, simple examination and therapy will lead to misdiagnosis.

Objective: This study sought to investigate the clinical characteristics, diagnostic methods and treatment of ICA pseudoaneurysms in the sphenoid sinus.

Methods: Various data, including clinical features, imaging examination results, and treatment and prognosis information, were collected and analyzed for 8 patients who visited the Department of Otolaryngology, Head & Neck Surgery of West China Hospital from March 2008 to January 2017.

Results: The patients included 6 males and 2 females (ages 16 to 56 years). Repeated epistaxis was a common symptom in six of the eight patients (6/8), whereas monocular blindness and binocular blindness were observed in the other two patients. Head trauma was found to play a role in the induction of ICA pseudoaneurysms, given that five patients (5/8) exhibited a specific history of head injury. CT examination tended to result in misdiagnosis, whereas MRI and digital subtraction angiography (DSA) were helpful for obtaining a definite diagnosis with all diagnoses were confirmed via DSA. Coated stent intervention was performed in five patients, while carotid artery ligation was performed in two patients in emergency situations: one of whom exhibited paraplegia, but recovery was ultimately observed after rehabilitation. Moreover, failure of coated stent intervention in one patient was resolved via additional unilateral common carotid artery ligation. Furthermore, one patient with vision loss experienced vision restoration. One patient discontinued treatment for personal reasons and was lost to follow-up. No recurrence was observed in the other 7 patients.

Conclusion: ICA pseudoaneurysms in the sphenoid sinus are uncommon. To accurately identify ICA pseudoaneurysms, collaboration between otolaryngologists and imaging specialists is essential. On the other hand, both surgical and interventional treatments can achieve good results; therefore, otolaryngologists should enhance their cooperation with neurosurgery and intervention departments. Accurate diagnosis and rapid treatment are keys to managing ICA pseudoaneurysms.

1. Introduction

Aneurysms include true aneurysms and pseudoaneurysms. Although aneurysms have various causes, the most common causes are intracranial atherosclerosis and hypertension [1]. In addition, infected lesions in the body, such as bacterial endocarditis and pulmonary infection, cause the infectious emboli to fall off and erode the wall of the cerebral artery to form infectious aneurysms. Head trauma can also lead to aneurysms [2]. Traumatic pseudoaneurysms are fragile and prone to rupture. Internal carotid artery (ICA) pseudoaneurysms are associated with 30% mortality from massive epistaxis if left untreated [3].

Pseudoaneurysms arising from the cavernous segment of the ICA have long represented a management challenge for neurosurgeons due to their unique anatomical location, surrounding critical neurovascular structures and often fusiform and/or giant morphologies [4]. However, when an ICA pseudoaneurysm occurs in the sphenoid sinus, patients often visit the otolaryngologist due to nasal bleeding, which is a challenge for the otolaryngologist. To avoid serious medical accidents,

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otolaryngologists should keep in mind that in such patients, nasal bleeding might be due to an ICA pseudoaneurysm.

2. Methods

In this retrospective study, all patients were diagnosed with ICA pseudoaneurysm via digital subtraction angiography (DSA) from March 2008 to January 2017 at West China Hospital, Sichuan University. All patients underwent head computed tomography (CT) and magnetic resonance imaging (MRI), and we recorded the patients' medical history, initial diagnosis and treatment. The mean follow-up period was 20.1 months (range, 12 to 36 months). This retrospective review of medical records was approved by the Institutional Review Board of West China Hospital.

3. Results

3.1. Patients

The patients were aged 16 to 56 years with 6 males and 2 females. Repeated epistaxis was a common symptom in six of the eight patients (6/8), while monocular blindness was observed in one patient and binocular blindness in another patient. Five patients had a history of a blunt injury to the head, and a linear fracture of the head or skull base was visible on CT for three of these patients. One patient had a history of prior nasal surgery. Based on CT examinations, the following misdiagnoses were identified: sphenoid sinus tumor was diagnosed in 3 of the 8 patients, a sphenoid sinus cyst was diagnosed in one patient, sphenoid sinus hematocele after optic canal tube fracture was diagnosed in one patient, fungal sphenoid sinusitis was diagnosed in one patient, nasopharyngeal fibroadenoma was diagnosed in one patient, and postoperative wound hemorrhage was diagnosed in one patient. All patients were definitively diagnosed by DSA. The details are shown in Table 1.

3.2. Imaging examination

All eight patients underwent CT and MRI examinations. The CT examinations of five patients (5/8) showed soft tissue or cystic lesions with sphenoid cystic degeneration and bone destruction, with a linear fracture of the head or skull base visible for three of these patients; three patients (3/8) only had soft tissue lesions in the sphenoid sinus. The common MRI findings (8/8) were cystic lesions with a flow void in the center. Four patients underwent magnetic resonance angiography (MRA), which showed a sphenoid sinus lesion connected to the ICA (Fig. 1). All patients (8/8) were diagnosed with ICA pseudoaneurysms upon DSA, in which the sphenoid sinus lesion exhibited a circular shape and was connected to the ICA (Table 2).

3.3. Treatment and follow-up

Five patients underwent coated stent treatment (Fig. 2); four of these patients were cured, while one received ligation treatment because of repeated bleeding after coated stent treatment. Two patients underwent emergency unilateral common carotid artery ligation after balloon test occlusions were performed, and another patient discontinued treatment for personal reasons During the mean follow-up period of 20.1 months, one patient experienced paraplegia after emergency unilateral common carotid artery ligation but ultimately recovered after rehabilitation, and one patient was lost to follow-up; the other patients did not relapse. One patient with vision loss experienced vision restoration.

4. Discussion

Intracranial aneurysms in the cavernous sinus account for approximately 5–8% of intracranial aneurysms [5]. This segment is adjacent to the sphenoid sinus, and aneurysms commonly burst into the sphenoid sinus. The bony wall of the sphenoid sinus is < 0.5 mm thick in 50% of people and may be dehiscent in 4%, leaving only the sphenoid sinus mucosa covering the carotid artery. This relationship allows ICA pseudoaneurysms to expand anteromedially into the sphenoid sinus or posterior ethmoid air cells and rupture into the nose via the sphenoethmoid recess [6].

Head trauma, monocular vision loss and delayed massive nasal hemorrhage are referred to as ‘Maurer's triad’ of traumatic carotid pseudoaneurysms [7]. Patients with internal carotid pseudoaneurysms in the sphenoid sinus often visit the hospital for nasal bleeding. However, epistaxis is a common disorder, accounting for 33% of all otolaryngology-related emergency admissions [8]. Otolaryngologists often consider arterial damage in cases of massive hemorrhaging but ignore the possibility of ICA pseudoaneurysms when a low level of nasal bleeding is present. We typically used the following criteria to identify patients who could potentially have had ICA pseudoaneurysms: 1) repeated nasal hemorrhage with no abnormalities revealed upon rigid endoscopy and coagulation assessment; 2) nasal hemorrhage after head trauma; and 3) visit to the hospital due to decreased vision, with tissue in the sphenoid sinus revealed via CT. In our study, five of the six patients with nasal bleeding had a history of chronic slight rhinorrhagia, which was one of the reasons that we misdiagnosed the condition as inflammation or tumor. During endoscopic optic nerve decompression surgery, ignoring the possibility of a pseudoaneurysm after trauma and injury to the pseudoaneurysm will lead to a serious crisis, such as the situation experienced by patient 5. Therefore, otolaryngologists should be aware of the possible occurrence of ICA pseudoaneurysm in patients with recurrent bleeding and impaired vision after trauma.

Imaging examinations play an important role in the diagnosis of sphenoid sinus diseases. CT is useful for the examination of bone fracture around the sphenoid sinus and the degree of destruction of

Table 1
Summary of reported case.

List	Sex	Age	Clinical manifestation	History of trauma	Initial diagnosis
1	M	28	Blood in nasal mucus for 2 years	Fell and hurt his head 8 years ago	Fungal sphenoid sinusitis
2	F	53	Left eyesight decreased for approximately 30 years	No	1. Sphenoid sinus tumor; 2. Optic atrophy
3	F	16	Blood in nasal mucus for 2 years	No	Sphenoid sinus cyst
4	M	24	Right eyesight decreased progressively for 2 days	Had right eye injury 2 days ago	1. Sphenoid sinus hematocele; 2. Right optic canal tube fracture; 3. Multiple skull base fractures
5	M	56	Lost sight in both eyes, epistaxis and nasal obstruction for 1 year	Head trauma 1 year ago	Sphenoid sinus tumor
6	M	21	Repeated epistaxis for 2 months	Automobile accident 2 months ago	Nasopharyngeal angiofibroma
7	M	47	Repeated epistaxis for 3 months	Automobile accident 3 months ago	Sphenoid sinus tumor
8	M	52	Epistaxis for 2 days	Underwent surgery to remove a tumor in the sphenoid sinus 10 days ago	Postoperative mucosal bleeding

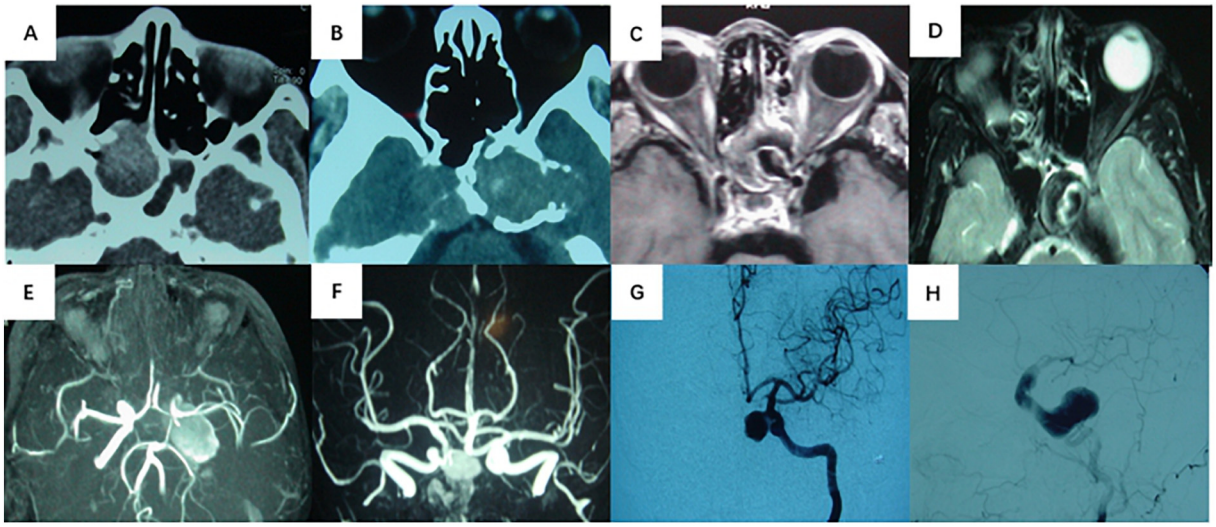

Fig. 1. Imaging examination in patients with internal carotid artery aneurysm. (A) CT showed soft tissue with sphenoid cystic degeneration, which was misdiagnosed as fungal sphenoid sinusitis. (B) CT showed cystic lesions in sphenoid sinus and bone destruction, which was misdiagnosed made as a tumor. (C) and (D) MRI showed cystic lesions with a flow void in the center. (E) and (F) MRA showed a sphenoid sinus lesion with high density connected to the internal carotid artery. (G) and (H) DSA showed that the sphenoid sinus lesion presented a circular shape and was connected to the internal carotid artery.

Table 2
Image examination.

Examination result	Number of cases
CT	
Fracture of sphenoid sinus wall	5/8
Cystic lesion in the sellar region only	3/8
MRI	
Cystic lesion in the sellar region	8/8
Flow void in the lesion center	8/8
MRA	
Sphenoid sinus lesion connected to the internal carotid artery	4/4

bone as well as the initial diagnosis of sphenoid sinus disease. When patients exhibit typical clinical manifestations, CT reveals bone fractures in the sphenoid sinus or orbital apex. Traumatic pseudoaneurysms of the ICA should be considered, but the possibility of traumatic carotid pseudoaneurysms cannot be excluded if CT does not indicate fractures. However, CT is not as useful as MRI, MRA, DSA or CTA (computed tomography angiography) for the clinical diagnosis of pseudoaneurysms. MRI can reveal the features of the sphenoid sinus, including whether it is substantial or cavitated, the blood supply, the boundaries of the organization, and the relationship with intracranial tissue. Upon MRI, a pseudoaneurysm always appears as a round placeholder in the sphenoid sinus with a flow void in the center. MRA examination is helpful to further understand the relationship between the lesion and the surrounding vessels. If a lesion is strongly suspected of being a pseudoaneurysm, DSA examination can not only provide a diagnosis but can also simultaneously provide interventional therapy. However, it is still unclear whether emergency DSA can detect potential pseudoaneurysms after head trauma because the formation of traumatic pseudoaneurysms requires time. Pelliccia et al. [9] reported one patient with a traumatic pseudoaneurysm with normal intracranial findings upon early CT angiography; thus, these authors suggested that if the first angiographic evaluation reveals normal findings, repeated epistaxis should prompt a second angiographic evaluation because a pseudoaneurysm can gradually develop. However, Zheng et al. [10] suggested that compared to DSA, noninvasive MRA or CTA is safer and allows more effective examination. Because of its outstanding three-dimensional display capabilities, CTA can clearly show the positions and surroundings of pseudoaneurysms and intraluminal thrombi. Moreover, CTA is a non-invasive technology that requires less time than MRA and is safer than DSA. CTA has become more acceptable for diagnosing pseudoaneurysms [11,12]. However, for small aneurysms (< 3 mm), the sensitivity of CTA is 40–90%; therefore, choosing CTA as the first examination may lead to a missed diagnosis [13]. In our study, CT primarily showed soft tissue or cystic lesions with sphenoid cystic degeneration accompanied by bone destruction in 5 cases and soft tissue in the sphenoid sinus in 3 cases. All MRI examinations revealed a flow void effect, similar to the occurrence of hemangioma. The diagnosis of ICA pseudoaneurysms via CT is difficult for otolaryngologists;

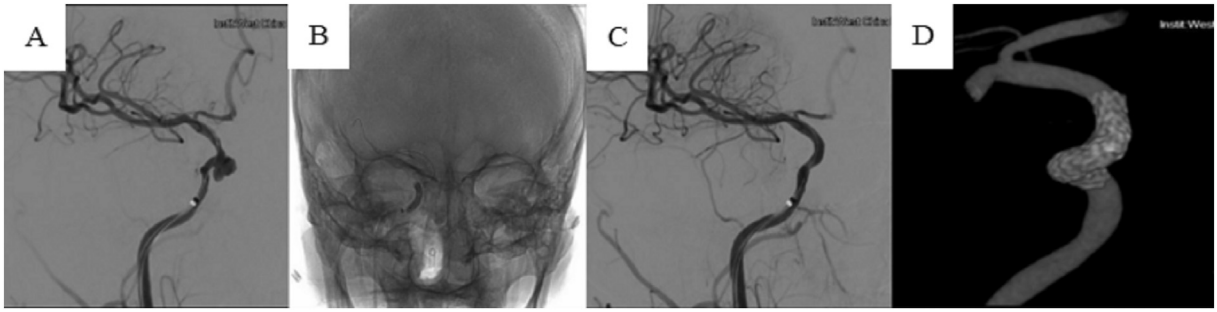

Fig. 2. Coated stent intervention treatment. (A) Before coated stent intervention, showing an arterial pseudoaneurysm in the right cavernous sinus segment. (B) The coated stent had been implanted. (C) After the coated stent intervention, the arterial pseudoaneurysm disappeared. (D) Revascularization after coated stent placement is shown.

thus, MRI, MRA, CTA or DSA is necessary as a supplemental examination.

Common approaches for nasal bleeding when excluding medical diseases include surgical or interventional therapy. Emergency surgical ligation in the neck has traditionally been an acceptable approach but can lead to death or stroke. Nasal packing may be useful for addressing hemorrhage of ICA pseudoaneurysms but can cause bleeding when the packing is removed; in certain studies, a muscle patch has been reported to be extremely effective as a permanent packing material in the management of hemostasis of the ICA [14–16]. Other valuable packing tools, such as a thrombin-gelatin matrix, have even been recommended [17]. However, "overpacking" should be avoided; otherwise, important vessels are compressed, leading to decreased perfusion [18]. Success with bipolar coagulation via endoscopy has also been described, especially for microaneurysms (aneurysms with a maximum diameter ≤ 3 mm) [19], but this approach has a risk of secondary bleeding and appears to be less reliable than the use of packing or clipping [16]. Certain neurosurgeons have also attempted to clip aneurysms with a neuroendoscope via a nasal pathway and have achieved hemostasis while maintaining vascular patency [20,21]. On the other hand, due to the deep location of such pseudoaneurysms in the sphenoid sinus and the narrow ostium, interventional therapy to manage these pseudoaneurysms could be an effective approach. As an interventional therapy, coated stent treatment can maintain arterial ventilation and isolate the lesion. A coated stent is highly suitable for segments of the ICA without important branches, such as the petrous, cavernous, and clinoidal segments. In 1997, Singer first reported the application of coated stents for the treatment of giant pseudoaneurysm of the ICA [22]. Then, Kocer used this method to treat carotid-cavernous fistulas caused by ICA injury [23]. In this study, 5 patients were treated with stent implantation, and good results were achieved. One patient continued to experience bleeding after two stent treatments, possibly due to the following factors: 1) the stent was improperly placed or moved after placement; 2) the stent was inadequately expanded; and 3) support was retracted. If treatment is unsatisfactory after placement of a second coated stent, the affected ICA can be balloon embolized. Because the ICA is tortuous, which presents a challenge for the surgeon, rational use of a coated stent is a suitable method for treating pseudoaneurysms.

One consideration is that we simply searched for all patients diagnosed with ICA pseudoaneurysms in our hospital's ENT department; however, some patients with pseudoaneurysms may have first visited the neurosurgery, ophthalmology or other departments and subsequently received treatment. In addition, a subset of patients who achieved hemostasis after blind nasal packing (with no definite bleeding point found) may have refused imaging examinations. Both of the aforementioned factors could have led to certain biases in our study. Therefore, prospective studies with larger samples of patients diagnosed and treated for ICA pseudoaneurysms in the sphenoid sinus remain necessary.

5. Conclusion

ICA pseudoaneurysms in the sphenoid sinus are uncommon. Improper diagnosis can have disastrous consequences, such as fatal epistaxis. To accurately identify ICA pseudoaneurysms, collaboration between otolaryngologists and imaging specialists is essential. When CT shows an expansile, hyperdense and destructive mass in the sphenoid sinus or MRI displays a flow void effect in the sphenoid sinus, particularly for patients who exhibit repeated epistaxis or experienced prior head trauma, ICA pseudoaneurysms should be considered. A subsequent examination using CTA, MRA or DSA will be necessary, and biopsy should be avoided. On the other hand, both surgical and interventional treatments can achieve good results; therefore, otolaryngologists should enhance their cooperation with neurosurgery and intervention departments. Accurate diagnosis and rapid treatment are key to managing ICA pseudoaneurysms.

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