

SURGICAL NEUROLOGY INTERNATIONAL

SNI: Neurovascular, a supplement to Surgical Neurology International

OPEN ACCESS

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Case Report

Sphenopalatine arteriovenous fistula complicating transsphenoidal pituitary surgery: A rare cause of delayed epistaxis treatable by endovascular embolization

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Received: 11 January 16

Accepted: 17 June 16

Published: 21 December 16

Abstract

Background: Vascular injuries in transsphenoidal surgery for pituitary adenomas are uncommon but can result in serious disability or death.

Case Description: A 46-year-old man, who underwent resection of a pituitary adenoma with suprasellar extension via a transsphenoidal approach, presented with massive epistaxis five days postoperatively. Angiography revealed an arteriovenous fistula (AVF) between the right sphenopalatine artery and a deep vein draining to the right internal jugular vein, as well as contrast agent extravasation at the fistula point. The AVF was catheterized and successfully occluded with N-butyl-2-cyanoacrylate.

Conclusions: Transsphenoidal pituitary surgery can be complicated by massive epistaxis from a lesion of a small branch of the external carotid artery. Airway protection through intubation and investigation with conventional digital subtraction angiography is recommended. The treatment of choice is endovascular embolization because it can be done immediately at the angiography suite.

Key Words: Complication, epistaxis, pituitary adenoma, sphenopalatine artery, transsphenoidal surgery

Access this article online Website: www.surgicalneurologyint.com DOI: 10.4103/2152-7806.196369 Quick Response Code:

INTRODUCTION

Transsphenoidal pituitary surgery has a low complication rate. However when complications do occur they can be devastating and, in the case of vascular injury, potentially lethal. When postoperative epistaxis occurs, a much feared cause is a ruptured pseudoaneurysm of the internal carotid artery. Immediate angiographic investigation should be performed on all patients that develop postoperative epistaxis in the aftermath of transsphenoidal pituitary surgery. [10,11]

This article presents, to the best of the authors' knowledge, the first case of postoperative delayed massive epistaxis from an iatrogenic sphenopalatine arteriovenous fistula after transsphenoidal pituitary surgery.

CASE REPORT

A previously healthy 46-year-old man was referred to our clinic because of visual symptoms and a nonfunctional

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How to cite this article: Eneling J, Karlsson PM, Rossitti S. Sphenopalatine arteriovenous fistula complicating transsphenoidal pituitary surgery: A rare cause of delayed epistaxis treatable by endovascular embolization. Surg Neurol Int 2016;7:S1053-6.

http://surgical neurology int.com/Sphenopal atine-arteriove nous-fistula-complicating-transsphenoidal-pituitary-surgery:-A-rare-cause-of-delayed-epistaxis-treatable-by-endovascular-embolization/

pituitary adenoma with suprasellar growth that compressed the optic chiasm [Figure 1]. The tumor was resected via a transnasal, transsphenoidal microsurgical approach through the right nasal cavity. The tumor was tightly attached to the sellar diaphragm and there was a large opening to the suprasellar cistern after tumor removal. Tumor removal was macroscopically radical. Water-tight closure of the sella was achieved in layers using autologous fat tissue, collagen dural substitute, polyethylene sellar implant, fibrin glue, and repositioning of the saved remnants of the opened sphenoidal sinus mucosa. The nasal septum was realigned, as were the medial nasal turbinates on both sides. No bleeding from the sphenopalatine artery was encountered during the procedure. The patient had a momentary leakage of blood-tinted fluid from the nose when he was first mobilized from the supine to erect position. Four days post-peratively, as the patient was about to be discharged from our clinic to his local hospital, he had a brief transient episode of moderate epistaxis and was made to stay an extra night at the hospital. Early the next morning, 5 days postoperatively, the patient developed massive, life-threatening epistaxis from the right nasal cavity, as well as hematemesis. The patient was intubated, and a nasopharyngeal tamponade was applied. Computed tomography (CT) angiography did not reveal any injury to the internal carotid artery on either side. Digital subtraction angiography (DSA) of the right common carotid artery revealed irregularities in the wall of the right sphenopalatine artery and an arteriovenous shunt to a vein directed toward the internal jugular vein, with contrast agent extravasation at the fistula point [Figure 2]. A microcatheter was navigated into the sphenopalatine artery [Figure 3], and embolization of the arteriovenous fistula was done using 0.5 ml glue (N-butyl-2-cyanoacrylate and Lipiodol in proportion 1:2).

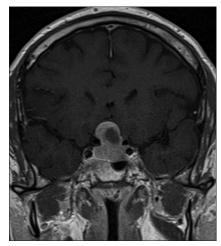


Figure 1: Contrast-enhanced T1-weighted magnetic resonance imaging in coronal view showing a nonfunctioning pituitary adenoma. The tumor size was 2.5 × 2 × 2 cm, with suprasellar extension and compression of the optic chiasm

The microcatheter was removed. Angiography of the right common carotid artery did not show any remaining arteriovenous shunt or contrast agent extravasation; the posterior septal branch of the sphenopalatine artery was occluded [Figure 4], indicating this branch as the lesion site. Angiography of the right internal carotid artery showed no sign of arterial injury or angiographic complications. Angiography of the left common, internal, and external carotid arteries revealed normal vessel anatomy. After embolization, all tamponades were removed and there was no active bleeding. The patient was stable overnight, he was successfully extubated the following day and he remained neurologically intact. On clinical follow-up, 2 months later, the patient was doing very well.

DISCUSSION

Transsphenoidal pituitary surgery is considered to be a safe procedure with reported mortality rates at approximately 1%. Complications include anterior pituitary insufficiency, diabetes insipidus, cerebrospinal fluid fistulas and, less commonly, carotid artery injuries, hypothalamic injuries, loss of vision, and meningitis.[4] Vascular injuries in transsphenoidal surgery for pituitary adenomas are particularly feared because they can result in serious disability, and even death. The most commonly reported vascular injuries are those concerning the internal carotid artery (reported to occur in 1.1% of all transsphenoidal operations), and include laceration, perforation, avulsion of dural branches, thrombosis, and other forms of injury that can cause massive hemorrhage, stroke, or lead to the formation of pseudoaneurysms and carotid-cavernous fistulas. [1,3,4] Risk factors for iatrogenic internal carotid artery injury include large invasive adenomas, previous transsphenoidal surgery, previous radiation therapy, medical therapy (prolonged dopamine agonist treatment), and aggressive surgery of lesions invading the cavernous sinus.[4]

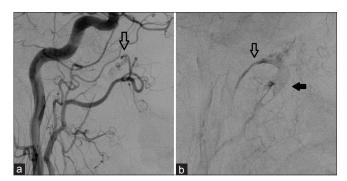


Figure 2: Digital subtraction angiography of the right common carotid artery in lateral view: (a) image in arterial phase showing a lesion in the sphenopalatine artery (arrow); (b) a later image showing arteriovenous shunt with venous filling (arrow) and extravasation of contrast agent (black arrow)

Arteriovenous fistulas are direct connections between high-pressure arteries and low-pressure veins that bypass the capillary bed. A post-traumatic (or, as in the current case, iatrogenic) arteriovenous fistula can be caused by both penetrating and nonpenetrating trauma to an artery and an adjacent vein, and can develop immediately following injury or in a delayed fashion from a post-traumatic pseudoaneurysm.^[7,15] We interpret the arteriovenous fistula in our case to have developed in the latter fashion.

The sphenopalatine artery is the terminal branch of the internal maxillary artery and provides arterial supply to the nasal mucosa. It passes through the sphenopalatine foramen and divides into medial (septal) and lateral branches, the latter giving off branches to the superior, middle, and inferior nasal conchae.[18] The septal branches pass to the posterior nasal septum and often require coagulation and division when surgical access is obtained between the middle turbinate and nasal septum for an anterior sphenoidotomy. Injury to the sphenopalatine artery can occur during enlargement of the sphenoidal opening because the artery is located in the hidden inferior lateral corner of the sphenoidal sinus. If it retracts toward the maxilla upon avulsion it can be difficult to coagulate. [6,11,17] Despite its diminutive size in comparison to the internal carotid artery, sphenopalatine artery injuries can result in massive epistaxis. There are reports of sphenopalatine artery pseudoaneurysms having

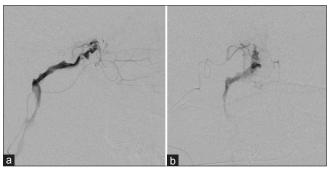


Figure 3: Digital subtraction angiography after microcatheterization of the right sphenopalatine artery showing the arteriovenous fistula before embolization: (a) Anterior posterior view; (b) lateral view

occurred postoperatively after endoscopic sinus surgery and orthognatic surgery (notably Le Fort I osteotomy) as well as post-traumatically. [6,2,8,12,16] Bleeding from the sphenopalatine artery, as from other branches of the external carotid artery, has been reported to have been successfully attenuated using detachable balloons, N-butyl-2-cyanoacrylate, particles, and platinum coils. [9,13,14,19]

Reports of sphenopalatine artery injuries in relation to transsphenoidal pituitary surgery are much less common than those of the internal carotid artery. We have found a total of 4 patients in two reports who developed angiographically diagnosed sphenopalatine artery pseudoaneurysms after transsphenoidal surgery for pituitary adenomas. [5,17] The case characteristics are presented in Table 1.

The decision to let the patient undergo a CT angiography instead of taking him straight to the endovascular suite for DSA can, in retrospect, be viewed as questionable. It did cause a delay in definitive treatment and added an unnecessary contrast bolus. In a patient with life-threatening epistaxis, CT angiography should not be

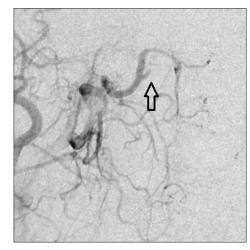


Figure 4: Digital subtraction angiography of the right common carotid artery in anterior posterior view:The arteriovenous fistula is occluded and the posterior septal branch of the sphenopalatine artery (arrow) is occluded

Table 1: Sphenopalatine artery injuries related to transsphenoidal pituitary surgery

| Reference | Sex/age | Clinical syndrome | Previous transsphenoidal surgery | Presenting bleeding episode | Sphenopalatine artery occlusion achieved by |
|--------------------|---------|------------------------|----------------------------------|-----------------------------|---|
| Raymond et al.[17] | F/34 | Galactorhea | No | Postoperative day 11 | Fiber microcoils |
| Raymond et al.[17] | M/40 | Acromegaly | No | Postoperative day 13 | Cyanoacrylate glue |
| Cockroft et al.[5] | F/52 | Prolactinoma | Yes | Postoperative day 15 | Polyvinyl alcohol particles |
| Cockroft et al.[5] | F/40 | ACTH-secreting adenoma | Yes | Postoperative day 17 | Spontaneous occlusion at angiography (vasospasm, dissection, or thrombosis) |

M: Male, F: Female

allowed to delay DSA and final treatment of the source of bleeding.

CONCLUSION

Massive postoperative epistaxis following transsphenoidal pituitary surgery requires swift treatment with immediate intubation to secure the airway, bleeding control through nasopharyngeal packing, and investigation with angiography. In this situation, a CT angiography is superfluous and a DSA is the investigation of choice. Although less common, massive epistaxis can result from lesions of a small branch of the external carotid artery such as the sphenopalatine artery. The treatment of choice, because it can be done almost without delay after diagnosis at the angiography suite, is endovascular embolization. There are many endovascular techniques available such as liquid embolics, particles, or coils that can be used. In the present case, embolization with N-butyl-2-cyanoacrylate was a successful method.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

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