Received: 22 December 2020 | Revised: 14 March 2021 | Accepted: 15 March 2021

DOI: 10.1002/ccr3.4117

CASE REPORT

Clinical Case Reports Open Access WILEY

A caroticocavernous fistula without vascular injury following endoscopic transsphenoidal excision of a tuberculum sellae meningioma—A case report

Jesse Zen Ngui[1] | **George Higginbotham**[2] | **Davendran Kanesen**[1] | **Jia Him Lau**[3] | **Ing Ping Tang**[4] | **Donald N. S. Liew**[1]

[1]Neurosurgery Department, Sarawak General Hospital, Kuching, Malaysia

[2]Southmead Hospital, North Bristol NHS Trust, Bristol, United Kingdom

[3]Radiology Department, Hospital Kuala Lumpur, Kuala Lumpur, Malaysia

[4]ENT Department, Universiti Malaysia Sarawak Faculty of Medicine, Kuching, Malaysia

Correspondence
Jesse Zen Ngui, Neurosurgery Department, Sarawak General Hospital, Kuching, Malaysia.
Email: jzngui@yahoo.com

Abstract

Caroticocavernous fistulae can occur following transsphenoidal surgery even without evidence of carotid artery injury. A role of vascularized flap reconstruction may be contributory.

KEYWORDS

caroticocavernous fistula, endoscopy, meningioma, skull base

1 | INTRODUCTION

Caroticocavernous fistulae are rare complications following carotid artery injury during transsphenoidal surgery. The incidence of this complication without carotid injury has not been reported previously. The use of vascularized mucosal flaps may be contributory. This lesion can be treated with endovascular techniques.

The incidence of vascular complications following endoscopic surgery of the skull base is a rare but potentially devastating complication. This is usually associated with intraoperative injury to the adjacent internal carotid artery and/or cavernous sinuses, with one potential consequence being formation of a caroticocavernous fistula. However, the formation of such a fistula postoperatively in the absence of such an injury has not been reported in the literature, and may involve alternate mechanisms to direct trauma.

2 | CASE REPORT

A 49-year-old woman with underlying dyslipidemia who presented to us with a chronic progressive bilateral visual loss and headaches for 8 months, with the left eye being more severely affected. Examination revealed no light perception in the left eye, the right eye's visual acuity being 6/12 corrected, with a right temporal hemianopia. There were no other cranial nerve palsies. Fundoscopy of the left eye showed optic atrophy.

MRI Brain revealed a sellar enhancing mass with suprasellar extension measuring 3.4 × 2.9 × 2.8 cm (Figures 1 and 2) with a dural tail consistent with a tuberculum sellae meningioma. Pituitary hormonal profile was within normal ranges.

She underwent endoscopic endonasal transtubercular tumor excision with complete excision of the tumor with a multilayered repair of the dura utilizing a nasoseptal flap, tissue glue, and artificial dura (Duragen). The tumor was

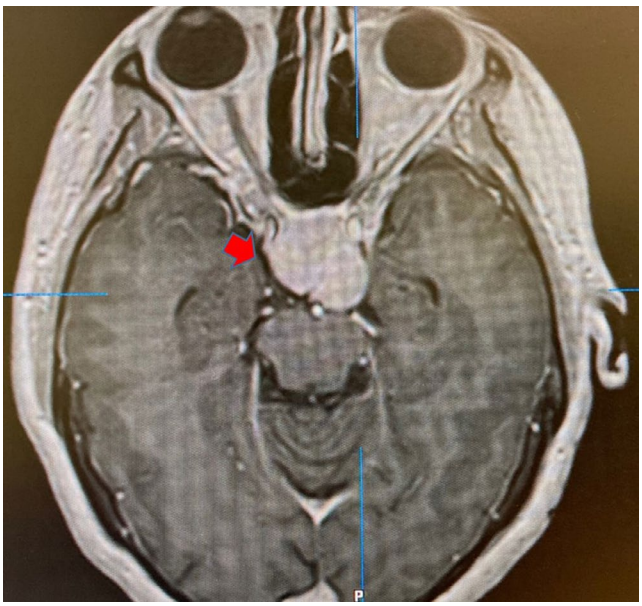

FIGURE 1 Preoperative contrast T1, axial MRI of the extra-axial meningioma at the sellar and suprasellar region (Arrow)

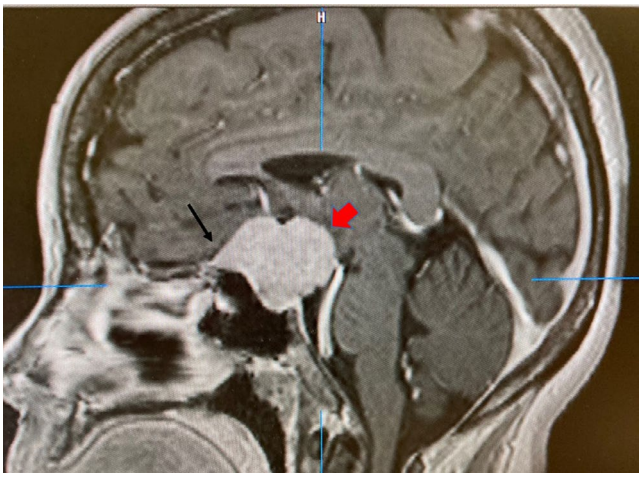

FIGURE 2 Preoperative contrast T1, sagittal MRI of the extra-axial suprasellar meningioma (Broad red arrow). The tumor occupies the sellar region with some suprasellar extension. Note anteriorly the dural tail finding characteristic of meningiomas (thin black arrow)

fibrous and both medial opticocarotid recesses were drilled bilaterally to access the tumor laterally. There was no excessive bleeding from the operative site seen during the procedure, and the patient did not require any blood transfusions during or after the procedure. Postoperatively, the patient had transient diabetes insipidus and required hydrocortisone replacement but was otherwise well and discharged after 8 days. Histology confirmed a grade I meningothelial meningioma. The patient then presented back 2 months later with CSF rhinorrhea. Endoscopic exploration and repair of the flap was performed. Intraoperatively, there was a small defect at the level of the tuberculum sellae on elevation of the mucosal flap, which was covered and reinforced with tissue glue.

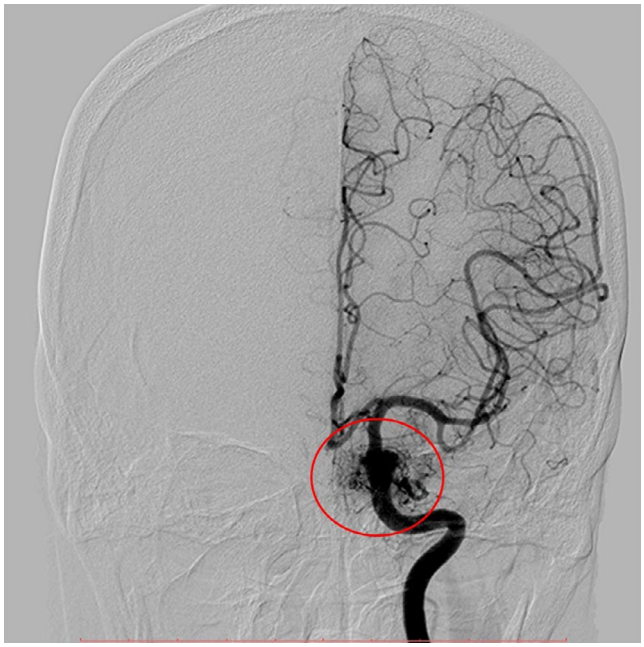

FIGURE 3 Internal Carotid angiogram, anteroposterior view, demonstrating left sided caroticocavernous fistula with supply from left internal carotid artery. Note the early filling of the left cavernous sinus (red circle)

No evidence of tumor recurrence was seen. Postoperatively, the patient was well.

The patient presented back 6 months after the initial surgery with new onset generalized tonic-clonic seizures. She was also noted to have left eye redness, pain, and swelling for 1 week prior to the seizure associated with intermittent headaches. Examination revealed left eye chemosis and proptosis, associated with multiple corkscrew vessels of the sclera, but no ophthalmoplegia. The left eye pupils was dilated. CT Brain revealed no tumor remnant. CT angiography revealed dilatation of the left superior ophthalmic veins with significant enlarged leptomeningeal vessels in the left cerebral hemisphere. Cerebral angiography confirmed a left caroticocavernous fistula supplied bilaterally by branches of both external carotid arteries and left internal carotid artery with no evidence of a pseudoaneurysm or venous thrombosis (Figures 3 and 4).

She underwent successful endovenous coil occlusion of the left caroticocavernous fistula. After procedure, her new ocular symptoms resolved completely, but the left eye remained blind. A repeat cerebral angiogram 2 months later showed no radiologic evidence of a residual caroticocavernous fistula (Figure 5). The patient is currently well and is still under our follow-up.

3 | DISCUSSION

This case report highlights a hitherto unreported complication of endoscopic endonasal transsphenoidal surgery. A Pubmed literature search did not find any previously such reported

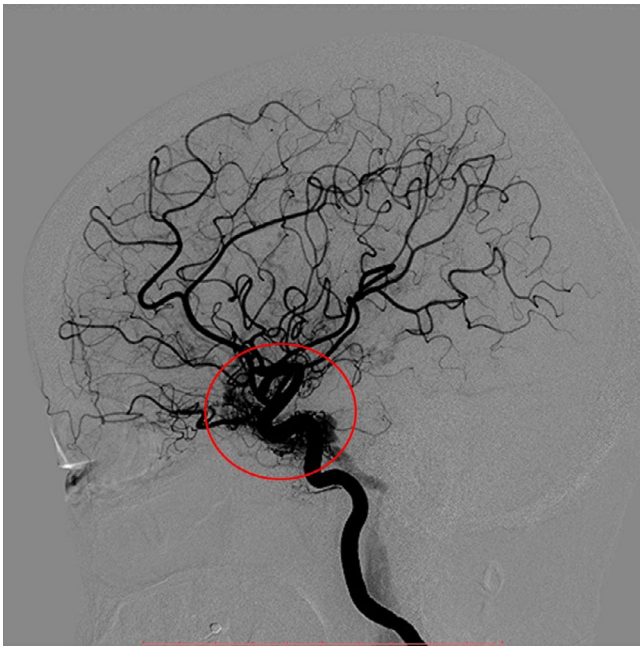

FIGURE 4 Internal carotid angiogram, lateral view, demonstrating left sided caroticocavernous fistula with early filling of the left cavernous sinus (red circle)

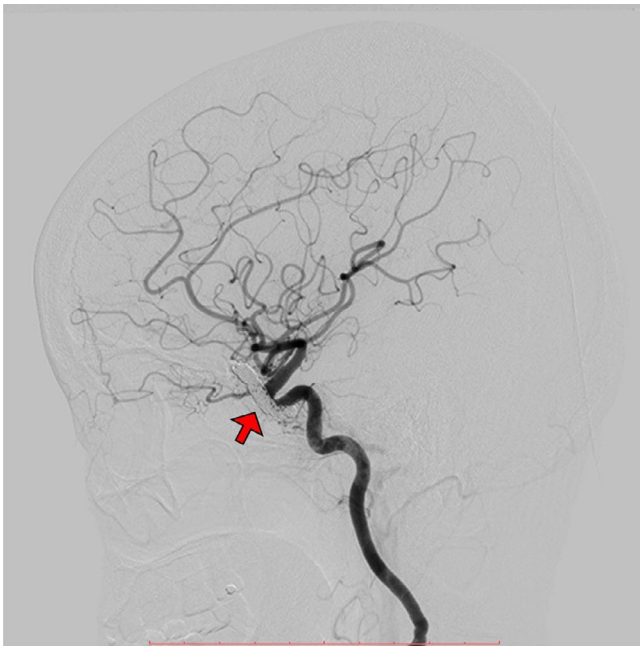

FIGURE 5 Repeat internal carotid angiogram, lateral view 3 months postcoiling, demonstrating resolution of fistula. The packed coils used for obliteration are visible (red arrow)

case. The use of endoscopic endonasal transsphenoidal surgery for skull base tumors has become increasingly popular in the past two decades. This technique, however, has previously been reported to have its own unique set of complications. The most common of these is a CSF leak, reported to occur in up to 15% of patients.[1,2] Other reported complications include transient or permanent neurological deficit, intracranial infection including meningitis. Carotid artery injury is a feared complication of such procedures, with a review reporting a 0.2%-1% incidence.[3] This is usually marked by the presence of torrential bleed from the exposed artery, but no such signs were seen in the above case. The treatment of such injuries range from surgical packing, clip ligation to endovascular exclusion or repair of the defect. Postoperative complications from such injuries reported include pseudoaneurysms, caroticocavernous fistulae, and cavernous sinus thrombosis. Previous case reports of caroticocavernous fistulae[4] or pseudoaneurysms[5] following transsphenoidal surgery were, however, preceded by clear evidence of intraoperative vessel injury and bleeding which was not seen in our case.

A common method of dural defect reconstruction in skull base surgery is the utilization of a pedicled nasoseptal flap, which has been shown to reduce the incidence of CSF leak in this population. Additional benefits are also seen in comparison with other methods of repair, with a reduced incidence of meningitis.

Nasoseptal flap reconstruction is generally considered a well-tolerated procedure, with minimal morbidity.[6] Expected complications of nasoseptal flaps predominantly involve the nasal septal donor site, and include septal perforation, mucocele formation, flap necrosis, and collapse of the nasal dorsum.[7] Vascular complications of dural defect reconstruction are negligible in the literature.

Caroticocavernous fistula is vascular lesions caused by an abnormal communication between the carotid artery or its branches and the cavernous sinus. It is widely classified using the Barrow classification into direct and indirect variants.[8] The type A or direct variants are typically associated with traumatic injury to the cavernous ICA and present acutely. Reported mechanisms include skull base and head trauma, angiography, and surgical procedures in the nasal cavity or transsphenoidal techniques. An association with collagen vascular diseases such as Ehler-Danlos syndrome and fibromuscular dysplasia has been reported, but this patient showed no evidence of such pathology.

Cavernous sinus thrombosis has also been proposed to be etiologic in the formation of indirect caroticocavernous fistulae. This was highlighted in a recent case report,[9] with possible mechanisms include direct injury to the cavernous sinus, and a postsurgical hypercoagulable state. Two case reports[10,11] of postoperative cavernous sinus thrombosis following transsphenoidal surgery serve to highlight this possibility, but the presentations were acute and not associated with an eventual caroticocavernous fistula. This patient did not have a history of thrombophilia, and angiography did not reveal signs of cavernous sinus thrombosis.

A potential pathophysiologic mechanism in the development of arteriovenous fistulae (AVF) is the overexpression of angiogenic growth factors such as VEGF. Overexpression of these factors have been described in the connective tissue of patients with dural AVFs.[12] A rat

model supports the VEGF hypothesis, with reduced rates of dural AVF formation seen with the addition of a VEGFR antagonist.[13] We propose that this patient's caroticocavernous fistulae were formed after the provocation of local overexpression of VEGF in the region following the surgery, possibly due to neovascularization of the nasoseptal mucosal flap. It is also interesting to note case series of VEGF expression in meningiomas,[14] and we propose that this may be significantly associated in this case via a paracrine effect. Further analysis of the patient's serum and or local tissue for angiogenic factors would be beneficial in clarifying this hypothesis, but were not locally available.

4 | CONCLUSION

The formation of caroticocavernous fistulae following transsphenoidal surgery may occur even without overt vascular injury. The role of vascularized flap reconstruction may be contributory, though the mechanism remains unclear.

ACKNOWLEDGMENTS

We acknowledge the support of the Head of Department of Neurosurgery of Sarawak General Hospital, Mr Albert Wong Sii Hieng in preparing this paper. Published with written consent of the patient.

CONFLICT OF INTEREST

The authors of this paper reports no conflicts of interest.

AUTHOR CONTRIBUTIONS

JN and GH: contributed to manuscript drafting, data collection and interpretation, and literature review. LJH and TYP: contributed to the diagnosis and management of the case. DL and DK: contributed to diagnosis and management of the case, conception of the article, data interpretation, editing, and supervision.

ETHICS STATEMENT

Written informed consent for publication of their clinical details and clinical images was obtained from the patient.

DATA AVAILABILITY STATEMENT

Data used in this study are available from the corresponding author upon reasonable request.

ORCID

Jesse Zen Ngui https://orcid.org/0000-0003-1083-4706

REFERENCES

1. Kassam AB, Prevedello DM, Carrau RL, et al. Endoscopic endonasal skull base surgery: analysis of complications in the authors' initial 800 patients. *J Neurosurg*. 2011;114(6):1544-1568.
2. Naunheim MR, Sedaghat AR, Lin DT, et al. Immediate and delayed complications following endoscopic skull base surgery. *J Neurol Surg B Skull Base*. 2015;76(5):390-396. https://doi.org/10.1055/s-0035-1549308
3. Chin OY, Ghosh R, Fang CH, Baredes S, Liu JK, Eloy JA. Internal carotid artery injury in endoscopic endonasal surgery: a systematic review. *Laryngoscope*. 2016;126(03):582-590.
4. Ahuja A, Guterman LR, Hopkins LN. Carotid cavernous fistula and false aneurysm of the cavernous carotid artery: Complications of transsphenoidal surgery. *Neurosurgery*. 1992;31:774-779.
5. Tuchman A, Khalessi AA, Attenello FJ, Amar AP, Zada G. Delayed cavernous carotid artery pseudoaneurysm caused by absorbable plate following transsphenoidal surgery: case report and review of the literature. *J Neurol Surg Rep*. 2013;74(1):10-16. https://doi.org/10.1055/s-0033-1338164
6. Kessler RA, Garzon-Muvdi T, Kim E, Ramanathan M, Lim M. Utilization of the nasoseptal flap for repair of cerebrospinal fluid leak after endoscopic endonasal approach for resection of pituitary tumors. *Brain Tumor Res Treat*. 2019;7(1):10-15. https://doi.org/10.14791/btrt.2019.7.e19
7. Thorp BD, Sreenath SB, Ebert CS, Zanation AM. Endoscopic skull base reconstruction: a review and clinical case series of 152 vascularized flaps used for surgical skull base defects in the setting of intraoperative cerebrospinal fluid leak. *Neurosurg Focus*. 2014;37(4):E4. https://doi.org/10.3171/2014.7.FOCUS14350
8. Barrow DL, Spector RH, Braun IF, et al. Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J. Neurosurg*. 1985;62(2):248-256.
9. Al-Mufti F, Amuluru K, El-Ghanem M, et al. Spontaneous bilateral carotid-cavernous fistulas secondary to cavernous sinus thrombosis. *Neurosurgery*. 2017;80(4):646-654. https://doi.org/10.1093/neuros/nyw128
10. Miao Z, Zhang Z, Chen J, Wang J, Zhang H, Lei T. Cerebral venous sinus thrombosis following second transsphenoidal surgery—report of a rare complication and review of literature. *World Neurosurg*. 2018;110:101-105.
11. Sawarkar D, Satyarthee GD, Singh P, Gurjar H, Singh MM, Sharma BS. Developing cerebral venous infarct presenting with seizure occurring after lumbar drain placement following trans-sphenoidal surgery of Cushing's disease: review of literature. *J Neurosci Rural Pract*. 2017;8:148-150.
12. Uranishi R, Nakase H, Sakaki T. Expression of angiogenic growth factors in dural arteriovenous fistula. *J Neurosurg*. 1999;91(5):781-786.
13. Li Q, Zhang Q, Huang QH, et al. A pivotal role of the vascular endothelial growth factor signaling pathway in the formation of venous hypertension-induced dural arteriovenous fistulas. *Mol Med Rep*. 2014;9(5):1551-1558. https://doi.org/10.3892/mmr.2014.2037
14. Hou J, Kshettry VR, Selman WR, Bambakidis NC. Peritumoral brain edema in intracranial meningiomas: the emergence of vascular endothelial growth factor-directed therapy. *Neurosurg Focus*. 2013;35(6):E2. https://doi.org/10.3171/2013.8.FOCUS13301

How to cite this article: Ngui JZ, Higginbotham G, Kanesen D, Lau JH, Tang IP, Liew DNS. A caroticocavernous fistula without vascular injury following endoscopic transsphenoidal excision of a tuberculum sellae meningioma—A case report. *Clin Case Rep*. 2021;9:e04117. https://doi.org/10.1002/ccr3.4117