CASE REPORT

Severe Intraoperative Orbital Venous Congestion during Resection of a Frontal Meningioma Presenting with Post-operative Vision Loss and Ophthalmoplegia: A Case Report

Victoria Leung^a, Ari Aharon Shemesh^a, Laila Al Shafai^b, Timo Krings^b, Taufik Valiante^c, and Edward Margolin^a

^aDepartment of Ophthalmology and Vision Sciences, University of Toronto, Toronto, Ontario, Canada; ^bDepartment of Medical Imaging, University of Toronto, Toronto, Ontario, Canada; ^cDepartment of Neurosurgery, University of Toronto, Toronto, Ontario, Canada

ABSTRACT

We describe a unique case of a middle-aged man who noticed complete vision loss in the right eye after awakening from resection of a large right-sided frontal meningioma. Visual acuity was hand motions, and there were multiple signs of right orbital venous congestion. Magnetic resonance imaging and venography (MRI/V) of the brain and orbits demonstrated expected post-operative findings with no evidence of cavernous sinus thrombosis or fistula. Empiric treatment with intravenous antibiotics and intravenous methylprednisolone were ineffective.

Immediate post-operative computerised tomography (CT) images were re-reviewed and revealed right restricted diffusion of the entire intraorbital right optic nerve. Discussion with the neurosurgical team revealed that during craniotomy, a prominent diploic venous plexus in the frontal bone adjacent to the meningioma was identified and coagulated with bone wax. Review of pre-operative imaging revealed large diploid flow voids in the right frontal bone, corresponding to the intraoperative findings. This prominent venous plexus appeared to drain from the meningioma posteriorly into the vein of Labbe. A second pathway drained anteriorly through the right angular vein into the orbit.

We hypothesise that the posterior outflow pathway was coagulated intraoperatively, causing redirection of all venous outflow from the meningioma into the right orbit through the anterior pathway. This resulted in significant orbital hypertension with manifest signs and symptoms. Furthermore, sudden rise in intraorbital pressure led to infarction of the optic nerve, leaving the patient with hand motions vision. We suggest that pre-operative vascular imaging should be performed in patients with large meningiomas, as pre-operative embolisation of venous outflow channels may prevent severe post-operative complications.

ARTICLE HISTORY

Received 18 June 2018
Accepted 20 September 2018

KEYWORDS

Post-operative complication;
meningioma resection; optic
neuropathy

Case

A 47-year-old man presented with a history of new onset severe headaches for the past month. Neuroimaging revealed a large right-sided frontal lobe lesion causing significant mass effect with subfalcine herniation. Imaging features were most compatible with meningioma (Figure 1a). Right-sided frontotemporal craniotomy was performed to resect the tumour, which on pathological analysis was determined to be WHO grade 1.

Surgery was performed under general anaesthesia with the use of frameless stereotactic image guidance. No intraoperative complications were reported.

Pre-operatively, the patient had experienced no visual complaints. However, when the patient was extubated and awoke on post-operative day 3, he immediately complained of right-sided vision loss. On examination, visual acuity was hand motions on the right and normal on the left. The intraocular pressures were 13 on the right and 12 on the left. There was an obvious right relative afferent pupillary defect. There was significant right-sided chemosis, 3 mm proptosis, and painless ophthalmoplegia. Funduscopy on the right revealed optic nerve head oedema, diffuse retinal whitening, and a large macula-off exudative retinal detachment; on the left there was mild optic nerve head oedema present.

CONTACT Edward Margolin edmargin@gmail.com Department of Ophthalmology and Vision Sciences, University of Toronto, 801 Eglinton Ave W, Suite 301, Toronto, Ontario, Canada M5N 1E3.

Color versions of one or more of the figures in the article can be found online at www.tandfonline.com/ioph.

© 2018 Taylor & Francis

Figure 1. a (left). Preoperative MRI. Axial T1 post-gadolinium sequence of the head demonstrates a large right frontal meningioma with mass effect and subfalcine herniation (white arrow) and mass effect on the lateral ventricles. b (right). Axial T1 post-gadolinium sequence shows prominent enlarged diploic venous plexus (white arrow).

Magnetic resonance imaging (MRI) and venography (MRV) of the brain and orbits with time-resolved imaging of contrast kinetics protocol were performed. Findings were in keeping with expected post-operative changes with no evidence of cavernous sinus thrombosis or fistula. The neurosurgical team confirmed there was no intraoperative surgical entry into the orbit. Given the timing of presentation, the patient was empirically treated with intravenous ceftazidime and vancomycin for possible post-operative infection, despite lack of surgical entry into the orbit. Posterior scleritis (as a variant of idiopathic orbital inflammatory syndrome) was also considered in the differential diagnosis given that the route of infection from the frontal lobe surgical site to the orbit was not clear. Thus, in addition to intravenous antibiotics, intravenous methylprednisolone (1 g per day for 3 days) was also administered. There was no improvement in central acuity or orbital congestion.

Immediate post-operative CT images were reviewed again and revealed right orbital proptosis and intramuscular hematoma in the right medial and inferior rectus muscles ([Figure 2a,b](#)). MRI and MRV performed 4 days after surgery demonstrated enlargement of the medial rectus with prominent hypointense signal on T2 and susceptibility weighted imaging compatible with recent hematoma ([Figure 2c,d](#)). There was restricted diffusion of the entire intraorbital right optic nerve extending

to the orbital apex ([Figure 2e](#)). This constellation of findings implicated orbital venous hypertension to be the culprit. However, its cause was unclear since there were no neuroradiologic signs of cavernous sinus thrombosis or carotid-cavernous fistula.

Further discussion with the neurosurgical team revealed that when performing the craniotomy a prominent diploic venous plexus was identified within the frontal bone adjacent to meningioma. It bled copiously and was initially retracted, but was eventually coagulated with bone wax. The remainder of the surgery was performed as planned.

The atypical nature of the patient's post-operative findings prompted detailed collaborative review of pre- and post-operative imaging. Careful re-examination of pre-operative MRI imaging demonstrated large diploid flow voids within the right frontal bone adjacent to dural base of the meningioma ([Figure 1b](#)). Furthermore, the meningioma's venous drainage appeared to diverge into anterior and posterior pathways. The anterior pathway drained venous blood from the diploic plexus into the right orbit via the angular vein, which was enlarged ([Figure 2c](#)), while the posterior pathway drained into the vein of Labbe.

We hypothesise that when the meningioma's venous drainage through posterior pathway was coagulated intraoperatively to enable surgical access, all venous drainage was at once diverted into the anterior pathway ([Figure 2f](#)). This caused

Figure 2. a. Immediate post-operative brain CT without contrast. Coronal reformatting demonstrated enlargement of the right medial and inferior rectal muscles (white arrows) and diffuse fat stranding in the orbits including intra and extraconal compartments. b. Immediate post-operative brain CT without contrast. Axial cut through the orbits demonstrating right orbital proptosis and preseptal thickening (white arrow). c. Post-operative MRI. Axial T2 sequence demonstrating right orbital proptosis, prominent right angular vein (white arrow), and T2 hypointensity (hematoma) in the right medial rectus muscle (black arrow). d. Post-operative MRI. Axial fat saturated T1 post-gadolinium sequence demonstrating heterogeneous enhancement of the right inferior rectus muscle (white arrow). e. Post-operative MRI. Axial diffusion weighted sequence demonstrating bright signal along the entire right intraorbital optic nerve (white arrow). f. "Banana Model" depicting how intraoperatively the entire venous outflow of the meningioma may have diverted to drain anteriorly into the right orbit after the posterior pathway was coagulated.

the entire venous drainage of a very large frontal lobe meningioma to flow into the right orbit for the duration of surgery, which required several hours.

This rapid and sustained intraoperative right orbital hypertension can explain the numerous ophthalmic manifestations of our patient, including right proptosis, chemosis, ophthalmoplegia, optic neuropathy, central retinal artery occlusion, and large exudative retinal detachment.

After the tumour was resected and inflow of venous blood from the meningioma stopped, right orbital pressure began to normalise. Two weeks after surgery, there was complete resolution of the patient's proptosis, chemosis, ophthalmoplegia, and exudative retinal detachment. Unfortunately, there was no improvement in the visual acuity. This was expected as there was restricted diffusion of a large portion of the right intraorbital optic nerve, which signifies irreversible infarction. Repeat MRI of the brain and orbits six months after surgery revealed atrophy of the right optic nerve.

Discussion

We present a unique case of monocular vision loss and ophthalmoplegia due to unexpected orbital venous hypertension during resection of a frontal meningioma.

Detailed review of pre-operative imaging revealed two plausible pathways of venous outflow from the meningioma: anterior and posterior. We believe that when the craniotomy was performed, transdural coagulation of the adjacent diploic venous plexus obliterated the posterior drainage pathway from the meningioma. This resulted in redirection of venous flow through the less dominant anterior pathway into the orbit via the angular vein. The anterior pathway became the sole channel for venous drainage from the highly vascular meningioma until it was entirely resected. As it took a few hours for the meningioma to be resected, excess inflow of venous blood resulted in a significant and sustained rise in right intraorbital venous pressure. This manifested

in several ways, including: extraocular muscle congestion, ischemic optic neuropathy, central retinal artery occlusion, and large exudative retinal detachment.

Exudative retinal detachment is a rare complication of orbital congestion, the mechanism of which is not clear. We hypothesise that in our patient, soaring intraorbital venous pressure caused compression of the choroidal vortex veins. This led to disruption of choroidal venous drainage, which resulted in diffuse leakage of fluid through retinal pigment epithelium into subretinal space.^{1,2} In addition, hypoxia of the choriocapillaris secondary to venous stasis likely resulted in widespread impairment of RPE function.³ Inability to pump excess fluid from the subretinal space, coupled with an abnormal increase in fluid accumulation, may have caused the retinal detachment.

This case represents a unique mechanism through which orbital compartment syndrome developed following frontotemporal craniotomy. Previous cases of orbital compartment syndrome following surgical procedures involving frontotemporal craniotomies have been reported, the mechanisms of which included: direct compression of the globe and orbit through a myocutaneous flap or instruments holding the flap,⁴ post-operative straining,⁵ and prolonged Trendelenburg positioning.⁶ In our case, the flap was retracted anteriorly without compression on orbital structures. There was no note of intraoperative or post-operative agitation. Furthermore, the patient was supine throughout the surgery and Trendelenburg positioning was not used. The mechanism of orbital venous congestion and subsequent orbital compartment syndrome described in our case is unique. Coagulation of the main pathway for venous outflow from the meningioma resulted in redirection of venous flow through an alternate drainage pathway, which fed into the adjacent orbit for several hours until the meningioma was entirely resected.

Currently, pre-operative embolisation of meningioma-associated feeder vessels is regarded as mostly unnecessary because the tumour's vascular supply is usually easily controlled through transdural coagulation that is performed after the craniotomy. Our case challenges this assumption and

demonstrates that preoperative venous vasculature assessment of convexity meningiomas may be indicated. Prior knowledge of the venous drainage of our patient's meningioma may have informed surgical planning and reduced the risk of visually significant complications.

We propose that all convexity meningiomas associated with dilated diploic veins on routine pre-operative imaging should be further evaluated with pre-operative computed tomography angiography and venography in order to evaluate the tumour's vascular supply and outflow. In cases where vascular communication between the tumour and surrounding structures is demonstrated, pre-operative embolisation should be considered in order to reduce the pressure flow within the tumour. This may assist in preventing tissue damage caused by unanticipated redirection of high flow venous drainage from the tumour into adjacent structures.

Conflicts of interest

All authors report no conflicts of interest.

References

1. Marmor MF. Control of subretinal fluid: experimental and clinical studies. *Eye*. 1990;4:340–344. doi:10.1038/eye.1990.46.
2. Escoffery RF, Bobrow JC, Smith ME. Exudative retinal detachment secondary to orbital and intraocular benign lymphoid hyperplasia. *Retina*. 1985;5:91–93.
3. Garg SJ, Regillo CD, Aggarwal S, Bilyk JR, Savino PJ. Macular exudative retinal detachment in a patient with a dural cavernous sinus fistula. *Arch Ophthalmol*. 2006;124(8):1201–1202. doi:10.1001/archophth.124.8.1201.
4. Pahl FH, de Oliveira MF, Dal Col Lucio JE, Souza E, Castro EF. Orbital compartment syndrome after frontotemporal craniotomy: case report and review of literature. *World Neurosurg*. 2018;109:218–221. doi:10.1016/j.wneu.2017.09.167.
5. Gauden AJ, Hardy T, Mack HG, Danesh-Mayer HV, Kaye AH. Orbital compartment syndrome following aneurysm surgery. *J Clinical Neuroscience*. 2012;19:1032–1036. doi:10.1016/j.jocn.2012.01.009.
6. Takahashi Y, Kakizaki H, Selva D, Leibovitch I. Bilateral orbital compartment syndrome and blindness after cerebral aneurysm repair surgery. *Ophthalmol Plast Reconstr Surg*. 2010;26(4):299–301. doi:10.1097/IOP.0b013e3181c062ca.