CASE REPORT

Sudden-onset monocular blindness following orbito-zygomatic craniotomy for a ruptured intracranial aneurysm

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SUMMARY

We report the first case of sudden-onset ipsilateral blindness following orbito-zygomatic craniotomy and clipping of a ruptured anterior communicating artery aneurysm. CT showed no new intracranial or intraorbital pathology. Visual evoked potentials testing, electroretinography and diffuse flash stimulation all indicated loss of right optic nerve function. Although the patient made an excellent neurological recovery, complete right-sided monocular blindness persisted at 6-month follow-up. We postulate that external pressure on the eyeball, resulting in posterior ischaemic optic neuropathy, was the primary cause of our patient's blindness. This has been hypothesised in the 3 previously published cases of blindness following pterional or frontal craniotomy for aneurysm repair. Intraoperatively, the surgeon must avoid unnecessary pressure on the eyeballs and handle the optic nerves with the utmost care. Incomplete understanding of the mechanisms of sudden visual loss postcraniotomy may result in under-reporting of this adverse event. Nevertheless, its seriousness warrants discussion during consent.

BACKGROUND

Sudden monocular visual loss following uncomplicated cranial operations performed in the supine position is rare when the underlying pathology does not involve the visual pathway. However, there have been reported cases of sudden visual loss following pterional and frontal craniotomy for an aneurysm repair and tumour resection. We report the first case of sudden monocular blindness following orbito-zygomatic craniotomy for a ruptured anterior communicating artery aneurysm. This was likely due to the intraoperative external pressure on the eyeball, causing posterior ischaemic optic neuropathy. This case serves to highlight that during craniotomy, the surgeon must avoid unnecessary pressure on the eyeballs and handle the optic nerves with the utmost care. Owing to lack of clear understanding regarding the mechanism of sudden visual loss postcraniotomy, this rare adverse event may be under-reported. Nevertheless, its seriousness warrants discussion during consent.

CASE PRESENTATION

A woman aged 42 years with a background of hypertension presented with severe sudden-onset headache and mild confusion. On examination, she had a Glasgow Coma Score of 14 (Eyes 4; Verbal 4; Motor 6) and no peripheral neurological deficit. She had CT-proven WFNS (World Federation of Neurosurgeons) Grade II subarachnoid haemorrhage. CT angiography demonstrated a multilobulated anterior communicating artery (AComA) aneurysm. After multidisciplinary discussions, it was agreed that surgical ligation was the optimum treatment approach. Two days postictus, the patient underwent an uneventful right orbito-zygomatic craniotomy (figure 1) and successful clip ligation of the AComA aneurysm (figure 2). The craniotomy was performed in two stages as per standard for the senior author, using an oscillating saw for the orbital rim cuts and thin osteotomes under direct vision to complete the orbital roof removal. No drilling of the anterior clinoid was performed.

The patient woke up from surgery fully alert without any neurological deficits except her complete inability to see from her right eye. Visual acuity showed no perception of light in the right eye, while acuity was 6/9 in the left eye. Light stimulus to the right eye showed no reflex pupillary constriction in the right or left eyes (direct and consensual responses, respectively). However, light stimulus to the left eye did demonstrate direct and consensual responses. Therefore, anterior segment examination of the eyes indicated a right-sided afferent pupillary defect and right optic nerve injury. External examination of the eye was unremarkable.

INVESTIGATIONS

CT did not show any new intracranial or intraorbital pathology. Funduscopy was normal on the left and showed a pale disc and attenuated vessels on the right consistent with ischaemic optic atrophy. However, the portion of the ophthalmic arteries visualised on postoperative CT angiography demonstrated symmetrical patency. MRI was not possible as the patient experienced symptoms of nausea and dizziness related to supine posture. The cause of these symptoms is unexplained. Examination of the ears, nose and throat was unremarkable, and examination of the vestibular system in other positions showed no abnormality. Monocular blindness was the only neurological deficit. The patient developed hydrocephalus 1 week into her admission, which required a ventriculoperitoneal shunt. This had no bearing on her symptoms.

To help identify the level of the visual dysfunction, pattern visual evoked potential (PVEP) testing was conducted. This showed an absent evoked

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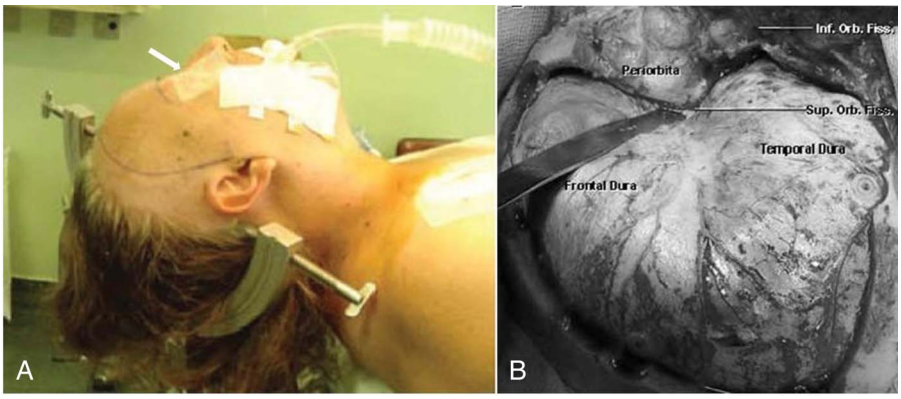

Figure 1 Patient positioning and right orbito-zygomatic craniotomy. (A) Patient was positioned supine, head fixed in a Mayfield clamp, and a thin protective (non-bulky) eye padding (white arrow) was used. (B) A right orbito-zygomatic craniotomy was performed with the scalp flap reflected without the use of spring hooks or bulky swabs.

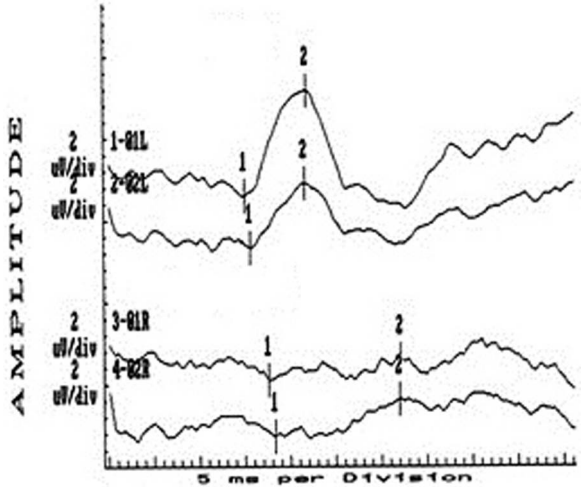

Figure 2 Postoperative pattern visual evoked potentials (PVEPs). Minimal amplitude PVEP in the right eye (lower graphs) versus normal amplitude PVEP in the left eye (upper graphs).

response on stimulation of the right eye, while a present and normal evoked response on stimulation of the left eye (figure 2). Diffuse flash stimulation evoked an anomalous and delayed response from the right eye compared with the left eye. Pattern electroretinogram (ERG) from the right eye showed mildly reduced P50 and markedly reduced N95, while the pattern ERG from the left eye was normal. These findings indicated marked loss of right optic nerve/retinal ganglion cell function.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis for the sudden visual loss following craniotomy includes central retinal artery thrombosis and ischaemic optic neuropathy due to spasm and/or surgical manipulation following prolonged hypotension or severe blood loss; intraoperative technical error resulting in mechanical or thermal damage to the intracranial portion of the optic nerve; postdecompressive optic neuropathy from sudden reduction in intracranial pressure and hypoperfusion to the optic disc (more commonly in tumour resection with preoperative papilloedema) and intraoperative external pressure on the eyeball causing posterior ischaemic optic neuropathy.

OUTCOME AND FOLLOW-UP

Although the patient made an excellent neurological recovery following operative treatment of her aneurysm, complete right-sided monocular blindness persisted, with no improvement in repeat visual evoked potentials testing at 6-month follow-up.

DISCUSSION

Aneurysms in the region of AComA and internal carotid arteries have been known to present with pre-existing visual deficits of compressive aetiology, although these preoperative deficits usually improve following successful surgical decompression. Even if the aneurysm ruptures into the optic apparatus, substantial delayed recovery can be expected. Iatrogenic postoperative blindness following neurosurgical procedures is associated with a poor visual prognosis.[1] Consistent with this, we have observed no visual recovery, to date, in our case of sudden postoperative monocular blindness.

Thrombosis of the central retinal artery (an end-artery) due to spasm and/or surgical manipulation was considered a possible cause of the ischaemic optic neuropathy. However, the ophthalmic arteries visualised on postoperative CT angiography demonstrated good patency bilaterally.

Prolonged hypotension and severe intraoperative haemorrhage are possible causes of ischaemic optic nerve damage, but these were both highly unlikely since on thorough review of the anaesthetic charts, our patient was normotensive throughout the procedure and had minimal blood loss <100 mL. Furthermore, other than hypertension, this patient did not have predisposing comorbidities, such as diabetes mellitus, atherosclerotic vascular disease, polycythaemia, renal failure, narrow-angle glaucoma or collagen vascular disorders.[2]

We (including two consultant neurosurgeons not directly involved in this case) have reviewed the recorded footage of the operation, and confirm that there was no advertent direct mechanical trauma or thermal injury to the intracranial portion of the optic nerve. Therefore, intraoperative technical error was unlikely.

Postdecompressive optic neuropathy from sudden reduction in intracranial pressure and hypoperfusion to the optic disc has been associated with tumour resection with preoperative papilloedema.[3–7] In our case, there was no radiological evidence of mass effect or aneurysmal compression of the optic apparatus, and there was no preoperative papilloedema. Thus, this mechanism for visual loss was unlikely in our case.

By process of elimination, we consider that the most likely primary cause of our patient's blindness was possible external pressure on the eyeball resulting in posterior ischaemic optic neuropathy. This has been postulated in the three previously published cases of postaneurysm surgery (table 1),[1 8 9] although the exact point where this has occurred during our operation is

Table 1 Cases in the literature of sudden-onset visual loss following craniotomy[1 3–12]

Reported case	Primary pathology	Operative approach	Clinical features	Proposed mechanism
Our case	Ruptured anterior communicating artery aneurysm	Orbito-zygomatic craniotomy	Sudden ipsilateral visual loss, limited recovery	Posterior ischaemic optic neuropathy from external pressure on ipsilateral eye
Vahedi *et al*[7]	Olfactory groove meningioma	Bifrontal craniotomy	Sudden bilateral blindness, no recovery	Posterior ischaemic optic neuropathy from reduction of ICP and hypoperfusion
Takahashi *et al*[9]	Ruptured anterior communicating artery aneurysm	Bifrontal craniotomy	Sudden irreversible bilateral blindness; severe globe tenting and ocular ischaemia	Bilateral orbital compartment syndrome secondary to external ocular pressure
Yamashita *et al*[12]	Frontal tumour	Frontal craniotomy	Sudden contralateral visual loss	Orbital compression from Bispectral index sensor leading to infarction
Choudhari and Pherwani[8]	Ruptured anterior communicating artery aneurysm	Pterional craniotomy	Sudden ipsilateral visual loss, limited recovery	Posterior ischaemic optic neuropathy from external pressure on ipsilateral eye
Maier *et al*[11]	Frontal tumour	Bifrontal craniotomy	Bilateral blindness, limited recovery	Bilateral orbital infarction due to external compression
Boström *et al*[6]	Intraventricular tumour with papilloedema	Bifrontal craniotomy	Bilateral blindness, limited recovery	Postdecompressive optic neuropathy, hypoperfusion to optic disc
Kang *et al*[1]	Ruptured middle cerebral artery aneurysm	Pterional craniotomy	Sudden ipsilateral visual loss, limited recovery	Posterior ischaemic optic neuropathy from external pressure on ipsilateral eye
Beck and Greenberg[5]	Intracranial tumour with papilloedema	Frontal craniotomy	Bilateral visual loss, limited recovery	Postdecompressive optic neuropathy, hypoperfusion to optic disc
Obenchain *et al*[4]	Frontal tumour	Parietal craniotomy	Bilateral visual loss, limited recovery	Postdecompressive optic neuropathy
Rinaldi *et al*[10]	Parietal tumour	Parietal craniotomy	Sudden ipsilateral visual loss	Occipital infarction
Sachs[3]	Cerebellar tumour and papilledema	Posterior fossa craniotomy	Sudden ipsilateral blindness	Ischaemic optic neuropathy from reduction in ICP and hypoperfusion

Our patient represents the fourth documented case of sudden blindness postaneurysm surgery and the first following orbito-zygomatic craniotomy.

not clear. Since we do not have any direct evidence for our assertion, it is also conceivable that a combination of factors (as discussed above) may have contributed to our patient's optic neuropathy with cumulative effect. In any case, all previously documented cases of sudden visual loss in the literature, for aneurysms or tumours, followed either a pterional or frontal craniotomy. To the best of our knowledge, our patient represents the first documented case of sudden-onset blindness following orbito-zygomatic craniotomy.

Choudhari and Pherwani[8] proposed that raised orbital venous pressure with prolonged ischaemia of the intraorbital structures results in loss of optic nerve and retinal ganglion cell function. This may be due to the overuse of protective eye padding, bulky swabs placed over the eyeball and underneath the scalp to keep the flap moist, or spring hooks used to retract the scalp inferiorly to facilitate a low craniotomy. Kang *et al*[1] recommend using protective eye shields rather than bulky and spongy eye pads to prevent posterior ischaemic optic neuropathy. In our experience, we have used eye pads without difficulties. In this case, we did not use spring hooks for flap retraction nor bulky swabs that may exert undue pressure on the eyeball (figure 1).

Embryologically, the orbit has dual supply from the supraorbital artery (which later becomes the middle meningeal artery) and ophthalmic artery, but variant anatomy exists. In up to 50% of patients, a communicating branch between the ophthalmic and middle meningeal artery is present and most frequently passes through the superior orbital fissure. In 15% of cases, multiple connecting branches are present—for example, recurrent meningeal artery or orbital branch of the middle meningeal artery—and pass through an additional foramen lateral to the superior orbital fissure (foramen of Hyrtl or the meningo-orbital foramen). In some cases, there is regression of the proximal ophthalmic artery, leaving the entire orbital supply to the middle meningeal artery. Other anatomical variants include origin of the middle meningeal branch from the ophthalmic artery, origin of the ophthalmic artery from the middle meningeal artery and less commonly cavernous sinus origin of the ophthalmic artery.[13] All these variants are important to recognise when considering positioning and craniotomy in order to minimise the risk of ischaemic injury to the orbit.

It must be emphasised that the risk of ischaemic optic neuropathy should be considered not only during vascular surgery via a skull base approach but also during other types of procedures. These include frontal and pterional craniotomies in the supine position for excision of parenchymal tumours and meningiomas

Learning points

- Owing to lack of clear understanding regarding the mechanism of sudden visual loss postcraniotomy, this rare adverse event may be under-reported.
- Postulated mechanisms for sudden visual loss following craniotomy include central retinal artery thrombosis and ischaemic optic neuropathy; intraoperative technical error resulting in mechanical or thermal damage to optic nerve; postdecompressive hypoperfusion to the optic disc; and intraoperative external pressure on the eyeball causing posterior ischaemic optic neuropathy.
- During craniotomy, the surgeon must avoid unnecessary pressure on the eyeballs and handle the optic nerves with the utmost care.
- Owing to its rarity, patients are not routinely warned of visual loss as a complication following orbito-zygomatic craniotomy. Nevertheless, its seriousness warrants discussion during consent.

where postdecompressive hypoperfusion to the optic disc may be involved; and spinal surgery in which the patient is positioned prone and at risk of reduced intraocular perfusion from direct pressure on the globe and raised central venous pressure secondary to downward tilt of the head and/or malposition of the abdomen.[2]

Intraoperative measures including maintaining a normotensive state, minimising haemorrhage, avoiding pressure on the eyeballs due to bulky swabs or excessive protective eye padding, meticulous and delicate manipulation of the optic nerves and related blood vessels within the surgical field, together with recognising variation in orbital blood supply, will all help minimise the risk of injury to the visual pathway and prevent this devastating complication.

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