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Case Report

A report of 2 patients with transient blindness following Le Fort I osteotomy and a review of past reported cases

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ABSTRACT

Blindness following a LeFort I osteotomy is a rare but extremely serious complication. Ten cases have been reported to date. None of these patients recovered vision. Optic neuropathy is believed to be the cause but the exact mechanism has not been settled. We report the first, and the only two, documented cases of complete loss of vision that recovered subsequently. The first patient was a 19-year-old male with repaired bilateral cleft lip and palate. He developed loss of vision in the right eye on the second postoperative day. The second patient was a 22-year-old male with repaired unilateral cleft lip and palate. He developed complete loss of vision in the left eye on the day of surgery. Both these patients underwent ongoing studies, which did not show any abnormalities. Both were treated with methylprednisolone. Both the patients gradually showed improvement in their vision. The first patient recovered normal vision several months postoperatively. The second patient's vision improved to 4/60 by 4 months postoperatively. We discuss the probable mechanisms of optic nerve injury and also the possible reasons why sight was restored in these patients. This is a rare but serious complication following a fairly common procedure. Through this article we wish to create an awareness of this complication and also a possible way of avoiding such a disaster.

KEY WORDS

Blindness, complications, hypotensive anaesthesia, LeFort I osteotomy,maxillary regression

INTRODUCTION

Blindness following a Le Fort I osteotomy, though rare, is an extremely serious complication. Ten cases have been reported to date. (Laniganet *al*., Bendor-Samuel *et al*.,Girotto *et al*., Wilson *et al*., Lo *et al*., Cruz *et al*., Cheng *et al*.).[1-7] It may occur in isolation or with other ophthalmic or intracranial complications. These include: Lachrymal gland injuries; cranial nerve III, IV and VI palsies; traumatic aneurysms; and arteriovenous fistulae (Lanigan *et al*., Girotto *et al*., Cruz *et al*., Steel and

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Cope).[1,3,6,8] It is generally agreed that optic neuropathy is the mechanism that leads to loss of vision in a Le Fort I osteotomy, but exactly how this occurs in an elective surgery is a question of debate. Direct injury to the nerve from atypical fractures of the base of skull or orbital walls has been found to account for only 3 of the 10 reported cases in the literature (Lanigan *et al*., Bendor-Samuel *et al*., Cruz *et al*.),[1,2,6] while indirect trauma to the optic nerve or its blood supply has been considered in the other 7 cases (Lanigan *et al*. Girotto *et al*.,Wilson *et al*., Lo *et al*. Cheng *et al*.).[1,3,4,5,7]

We report the first 2 documented cases of complete loss of vision that was restored, following Le Fort I osteotomies. The reported cases were in cleft patients and were presumably straightforward cases with no untoward difficulties intraoperatively. In one case, full vision was regained, while near-complete vision has been recovered in the second, more recent case.

CASE REPORT

Case 1

A 19-year-old male with bilateral cleft lip and palate presented to the centre in February 2006 for correction of his maxillary regression. The patient's medical history was unremarkable. A Le Fort I osteotomy with advancement was performed under controlled hypotensive anaesthesia with the mean arterial pressure maintained at about 70 mmHg. The duration of hypotensive anaesthesia was approximately 20 min. The osteotomy, pterygoid dysjunction and down-fracture of the maxilla were done without any difficulty. The estimated blood loss for the procedure was 500 mL and the duration of surgery was 90 min. His recovery from anaesthesia was uneventful.

On the second postoperative day, the patient complained of loss of vision in the right eye. On examination, there was no perception of light in the right eye, while the visual acuity for the left eye was 6/6.The right pupil was sluggishly reactive to light. The patient was immediately started on intravenous methylprednisolone 500 mg, twice daily. Meanwhile, an emergent computed tomography (CT) scan of the orbits, base of skull and brain, and Doppler study of the carotid and vertebral arteries were done.

Except for opacities seen in all the paranasal sinuses suggestive of haemorrhage and the left pterygoid separation that had occurred anterior to the pterygomaxillary fissure, the CT scans were normal. The Doppler study of the carotid and vertebral arteries was also normal [Figure 1a and b].

The patient's vision in the right eye began to improve gradually from hand movements on the 11th postoperative day, to a visual acuity of 6/9 by 1 month postoperatively. Fundus examination later showed disc pallor in the right eye, with normal findings in the left eye. A diagnosis of ischaemic optic neuropathy was made. Several months postoperatively, 6/6 vision was reported for the right eye and has remained so, to date.

Case 2

A 22-year-old male patient with unilateral cleft lip and palate presented with maxillary regression. His medical history was not significant. A Le Fort I osteotomy with advancement was performed in August 2012.

The procedure was uneventful and the pterygoid dysjunction and down-fracture were done without any difficulty. Controlled hypotensive anaesthesia was given for less than 20 min, with the mean arterial pressure maintained at 70 mmHg.The duration of surgery was 45 min and the estimated blood loss was approximately 700 mL.

Postoperatively in the recovery room, he complained of complete loss of vision in the left eye. He was immediately started on intravenous methylprednisolone 1 g, twice daily. CT scans and an urgent ophthalmology consultation were requested immediately. Eye examination revealed

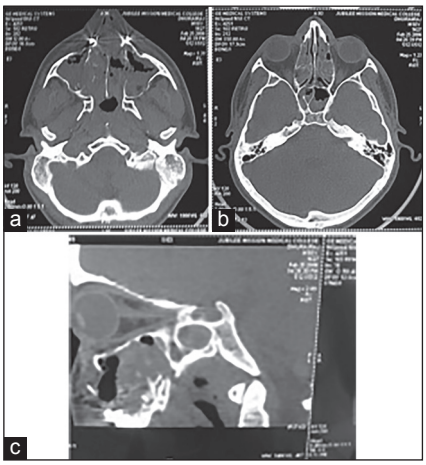

Figure 1:(a and b) Coronal section of the first patient showing blood in the spheno-ethmoidal and maxillary sinuses (c) Sagittal section of the first patient showing intact optic canal

defective vision in the left eye, while the right eye was normal. The left pupil was sluggishly reactive to light.

CT scans showed no atypical fractures to the base of skull or orbits, but showed opacities in the spheno-ethmoidal air cells. The pterygoid dysjunction had occurred anterior to the pterygomaxillary fissure on the right side. The carotid Doppler report was normal [Figure 2a and b].

The patient's left eye vision improved gradually, with a visual acuity of 1/60 on the 6th postoperative day to 4/60 by 4 months postoperatively. Disc pallor of the left eye was noted on fundoscopy 1 month postoperatively, and a diagnosis of ischaemic optic neuropathy was made.

DISCUSSION

The complication of blindness following an elective surgery is indeed devastating to both the patient and the surgeon, especially when one cannot explain to the patient exactly how it has occurred. Fortunately, blindness is a rare complication. We report 2 cases of blindness out of 690 Le Fort I surgeries done from 1996 to 2012 by the same surgeon, for an overall incidence of 0.29%. Lanigan *et al*.[1] reported an incidence of 0.25% in a survey of North American oral and maxillofacial surgeons, while Lo *et al*.[5] reported an incidence of 2.1% for a centre in Taiwan.

In the 7 cases in literature, and in our 2, where unfavourable fractures are absent, adverse transmission of forces to the base of skull during pterygoid separation and downfracture has been proposed as the mechanism of injury (Cruz *et al*., Polley).[6,9] Transmitted forces have been postulated to cause contusion or contrecoup type injuries to the nerve or compression of the nerve from haemorrhage and/or oedema around the optic canal, causing ischaemia of the nerve. It has also been considered that hypoperfusion of the optic nerve due to the controlled systemic hypotension might have a significant role in causing blindness (Lanigan *et al*. Cheng *et al*.).[1,7]

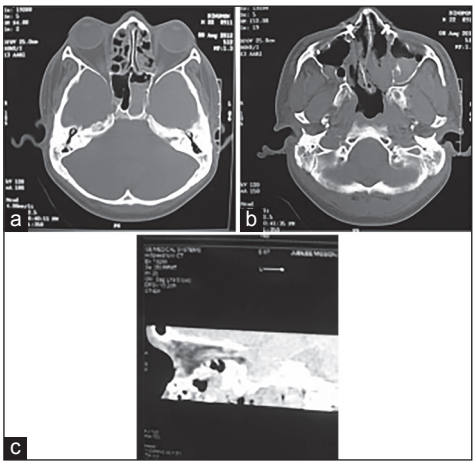

Figure 2: (a and b) Coronal section showing blood in the spheno-ethmoidal and maxillary sinuses of the second patient (c) Sagittal section of second patient showing intact optic canal

Injury to the ophthalmic artery from transmitted forces could be responsible for the haemorrhage within the spheno-ethmoidal sinuses in the CT scans of both our cases. The resultant hypoperfusion to the nerve, made worse by the hypotensive anaesthesia, may have caused the ischaemic optic neuropathy that led to unilateral loss of vision. We believe that loss of vision occurred unilaterally because the forces transmitted to the nerves and therefore the degree of injury inflicted on the nerves was unequal, as evidenced by the fracture pattern seen following both our osteotomies. The fractures had occurred anterior to the pterygomaxillary fissure, Type F separation (Ueki *et al*.),[10] on the opposite side of the affected nerves. Lanigan *et al*.,[1] stated that evidence of spheno-ethmoidal sinus haemorrhage should make one suspicious of the possibility of optic nerve injury, though they did not elucidate on the mechanism of injury.

We believe that the optic nerve and the ophthalmic artery suffer some degree of injury following this elective surgery due to their proximity to the surgical site. However, from our 2 cases, it appears that the nerve can recover from this insult, as long as it is not exposed to additional and prolonged ischaemic injury from controlled systemic hypotensive anaesthesia. Fortunately for our 2 cases, the duration of hypotensive anaesthesia was short and this was perhaps the reason the blindness was reversible. Lanigan *et al*.,[1] reported a case of blindness where the patient had been on hypotensive anaesthesia for 3 h; Lo *et al*.[5] reported that both their cases had been under hypotensive anaesthesia for 75 min and 95 min; and Wilson *et al*.,[4] though they did not mention the duration of hypotensive anaesthesia, had a 14-h surgery with a lowest recorded blood pressure (BP) of 80/40. Cheng *et al*.,[7] also reported a case of blindness where hypotensive anaesthesia had been used for 8 h. All these patients suffered irreversible blindness.

Controlled hypotensive anaesthesia in isolation does not cause blindness; it seems to aggravate the insult to an already injured nerve. This is evidenced by Kumar *et al*. (2004)[13], who reported a case of blindness following

spinal surgery due to compression of the globe by the head rest in the prone position. Cheng *et al*.[7] also reported a case of blindness where the effects of a hypoplastic carotid artery may have been made worse by hypotensive anaesthesia following Le Fort osteotomy. It has also been shown from animal studies that the optic nerve will not recover from ischaemia of over 60 min (Yoon and Marmor, Tsukahara *et al*.).[11,12] The duration of tolerable ischaemia to the optic nerve is quite possibly much lower in humans.

We therefore strongly recommend that hypotensive anaesthesia be avoided for this procedure or, if used at all, be used for a very short period of time. Proper surgical technique should also be employed to avoid unfavourable fractures and excessive haemorrhage, hence circumventing the need for hypotensive anaesthesia. It is also suggested that all patients undergoing Le Fort osteotomies undergo a preoperative ophthalmic assessment of vision.

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Conflicts of interest
There are no conflicts of interest.

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