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CASE REPORT

Trigeminocardiac reflex preceding development of postoperative superior orbital fissure syndrome

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ABSTRACT

The superior orbital fissure syndrome (SOFS) is a rare condition presenting as painful unilateral opthalmoplegia, ptosis, pupillary dilatation and anesthesia of the upper eyelid and forehead. It has been described after facial trauma. Other etiologies include infections, tumors or spontaneous hemorrhages in the retro-orbital space. Occurrence of SOFS after surgery in the skull base has not been described before. We recently encountered a case where a young female developed SOFS following surgery for cerebrospinal fluid (CSF) rhinorrhea. The occurrence of SOFS was preceded by episodes of trigemino-cardiac reflex (TCR) during the surgery. This is the first case report associating TCR with SOFS. We managed the case report with megadose methylprednisolone and the patient showed improvement in the deficit over time.

Key words: Megadose methylprednisolone, superior orbital fissure syndrome, trigemino-cardiac reflex

Introduction

The superior orbital fissure syndrome (SOFS), a rare cause of unilateral opthalmoplegia, reflects injury to the neurovascular structures traversing the superior orbital fissure (SOF).[1] It is characterized by palsies of the 3rd, 4th, 6th, and the first division of the 5th cranial nerves, which leads to opthalmoplegia, ptosis and proptosis of the eye, reflex dilatation of the pupil, and loss of sensation of the upper eyelid and forehead.[2] Etiologies include trauma, infections, tumors or hemorrhage. Literature is scarce on the occurrence of postoperative SOFS and its management.

We encountered a case where a patient, who was operated for transcranial repair of the floor of the skull, developed postoperative SOFS, presumably due to injury to the neurovascular structures situated in the vicinity, and was managed with corticosteroids. The complication was heralded by the occurrence of intraoperative trigeminocardiac reflex (TCR), which alerted us to the possibility of an impending neurological injury.

Case Report

A 28-year-old, female patient, with fibrous dysplasia of the skull, was operated for fronto- temporo-orbito-zygotomy with inner and outer table and orbital plate shaving, along with deroofing of optic nerve. Pre-anesthetic history was insignificant and the surgery under general anesthesia was uneventful. On the 3rd postoperative day, she developed cerebrospinal fluid (CSF) rhinorrhea, presumably from frontal sinus fistula, and had to be re-operated for the transcranial repair of the same. During surgery, as the surgeons were operating in the anterior cranial fossa, slight ooze was observed near the superior orbital fissure and the surgeons proceeded to coagulate the bleeding points with electrocautery. As cauterization was in progress, the mean arterial pressure (MAP) dropped from 92 mm Hg to 54 mm Hg, and simultaneously the heart rate (HR) dropped from 84 beats per minute (bpm) to 45 bpm. The intervention was ceased immediately. About 40 seconds later, the hemodynamics reverted to normal spontaneously. As electrocauterization was re-started, an immediate and similar drop in vital parameters

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was observed, which normalized on removal of the probe. The anatomical position was then reassessed and it was found that the cauterization was being done over the dura of the Vth nerve. Further electrocauterization over the area was stopped and hemostasis achieved with the conservative use of surgicel and wet gauzes. From here onwards hemodynamic changes were not observed, and the rest of the surgery proceeded uneventfully and the patient was extubated at the end of the surgery.

Postoperatively, the patient developed left sided ptosis, opthalmoplegia, pupillary dilatation, and loss of sensation over the left eyelid [Figure 1]. A presumptive diagnosis of SOFS was made based on the clinical signs and in association with the occurrence of intraoperative TCR. The patient was started on megadose methylprednisolone (30 mg/kg intravenous loading followed by 15 mg/kg every 6 hours) for the next 3 days. A computed tomography (CT) scan was done to exclude additional mechanical cause of the SOFS. She was discharged after 10 days with no improvement in the deficit, and was kept on regular follow-up. After 3 months, the patient showed improvement in the deficit (partial recovery of 3rd nerve function), which ruled out vascular injury and pointed towards neuropraxic injury [Figures 2-4]. At 6 months, she was having complete recovery of ptosis and numbness but partial 5th nerve palsy was still present in the form of conjunctival redness. This is unfortunate because the chief indication of surgery for this patient was cosmetic, and she paradoxically landed with further disfigurement. She is still being followed up, with the hope of further recovery.

Discussion

TCR manifests as sudden cardiac dysrhythmias, with a fall in the HR and MAP of more than 20% from the values before the stimulus, and coinciding with the manipulation of the trigeminal nerve. Cessation of the stimulus should result in the restoration of MAP and HR to normal levels and the phenomenon recurs when compression or traction is repeated.[3] It occurs in different craniofacial or skull base surgeries as well as neuroradiological interventions, due to the stimulation of the sensory portions of the trigeminal

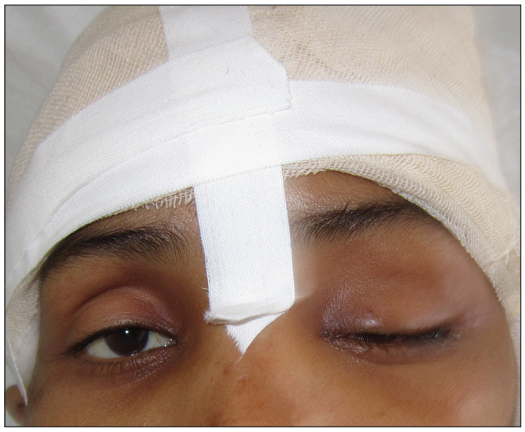

Figure 1: The patient developed ptosis, opthalmoplegia, chemosis, pupillary dilatation, and loss of sensation over the left eyelid in the immediate postoperative period

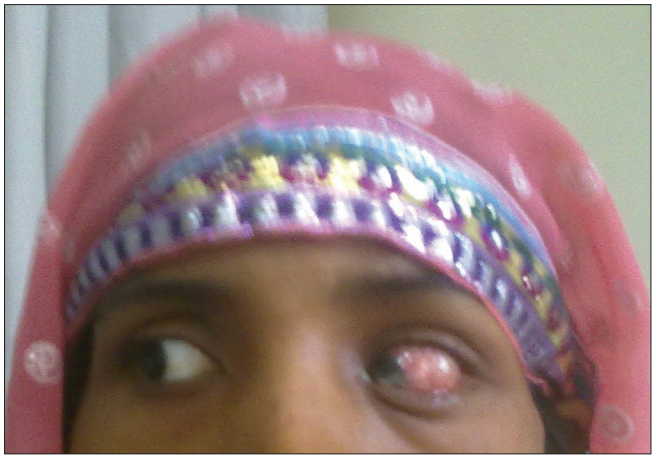

Figure 2: Three months after the surgery, there was partial recovery of ptosis and opthalmoplegia

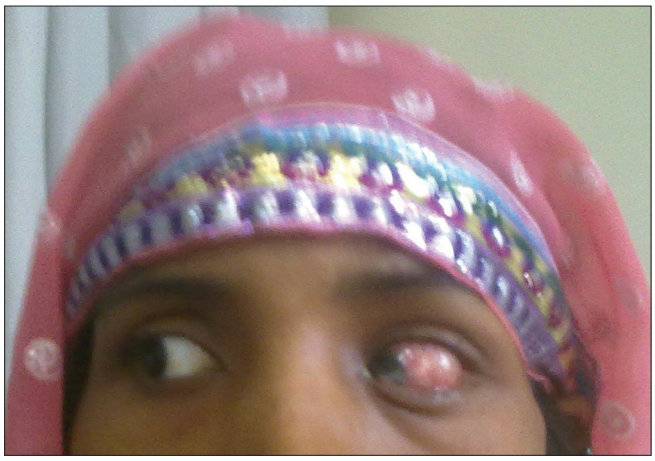

Figure 3: Abduction of the left eye has not recovered in three months

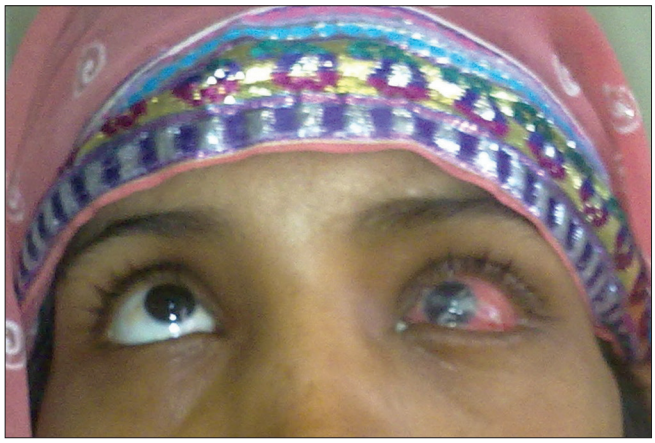

Figure 4: Three months after the surgery, chemosis is still present while opthalmoplegia has partially recovered

nerve.[4] The incidence of TCR ranges from 8-18%.[4] Several risk factors for the intraoperative occurrence of TCR have been identified, such as light general anesthesia, young age, and the nature of the provoking stimulus (abrupt and sustained traction is more reflexogenic than smooth and gentle traction).[4-6] In addition, there are several known provoking agents such as potent narcotic agents (sufentanil and alfentanil), b-blockers, calcium channel blockers, hydrogen peroxide, and infections.[4-6] Since, in our case, a clear cause and effect relationship was present with the use of electrocautery, we postulated that the electrocautery-induced stimulation of the V nerve would have precipitated the occurrence of this phenomenon.

Most cases of intraoperative TCR are managed with immediate cessation of the stimulus. In case of persistent bradycardia or hypotension, atropine (0.6 mg) is administered intravenously and repeated as required. Unresolved cases may necessitate the use of intravenous epinephrine (6 mcg).[7] Clear correlation does not exist between intraoperative TCR and postoperative outcome. Few case series have revealed a poor outcome of the surgery in which intraoperative TCR was observed.[8,9] In our case, TCR was probably stimulated by attempts to cauterize over the dura of the V1 division of the trigeminal nerve. The injury involved other neurovascular structures in the vicinity, leading to the development of SOFS. The episode of TCR during the surgery alerted us to the possibility of injury to the structures of the SOF, and immediately after the appearance of ptosis, opthalmoplegia and anesthesia of the upper eyelid in the postoperative period, therapy was started.

A CT scan was done to exclude any small bone fragments or retro bulbar blood or clot causing the symptoms. Spiral CT (2 mm slices) is the preferred tool for the diagnosis of traumatic SOFS, as it can provide detailed information about the SOF in the axial plane, coronal projection and by the 3D reformatted images.[1,10] Although SOFS has been described since 1860,[2] no case of SOFS after skull base surgery has been reported. Chen *et al*.,[10] have discussed the management of post-traumatic SOFS. Treatment options for traumatic SOFS vary from conservative approach (wait and watch) to surgical exploration. The use of steroids in SOFS has been well investigated and studies report different doses of methylprednisolone and dexamethasone for this purpose.[10-12] In the absence of any case report especially mentioning postoperative SOFS, we followed the management strategy described by Chen *et al*., for traumatic SOFS. They described the use of high dose methylprednisolone and we administered the same dosage to our patient (30mg/kg loading dose followed by 15mg/kg every 6 hours for 3 days). Acarturk *et al*.,[12] also used megadose corticosteroids for the management of posttraumatic SOFS. Steroids are believed to reduce the tissue edema, thereby, treating the condition. We believe that the timely start of methylprednisolone, helped reduce the inflammation around the SOF, and lead to the gradual improvement in the deficit.

This case associates the occurrence of intraoperative TCR with postoperative SOFS. Intraoperative hemodynamic events, especially while operating near the SOF, may mark the development of SOFS following the surgery. Although postoperative SOFS is rare, it is cosmetically disfiguring, debilitating and may lead to blindness. We suggest that SOFS, occurring after skull base surgery, should be treated with high dose steroids. Based on the lessons learnt from this case, we would also like to add a word of caution for the neurosurgeons operating near the SOF, whereby signs of 5th nerve irritation, occurring during the surgery, should be taken seriously. During bone work around vital structures, the bone work/drilling should be restricted to what is necessary. The drilling should be done under high magnification with judicious saline irrigation at the time of drilling and coagulation. While securing hemostasis near vital structures, instead of using aggressive measures like electrocautery; use of non-mechanical methods like saline irrigation, surgicel, fibrin glue, and collagen should be preferred.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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