OPEN

Sunken Eye Induced by Superior Orbital Wall Defect After Craniofacial Surgery

Sung-Pil Joo, MD,* Sung-Hyun Kim, MD,* Hong-Ju Park, DDS,† Seunggon Jung, DDS,† Jeong Joon Han, DDS,† and Tae-Sun Kim, MD, PhD*

Abstract: Enophthalmos after a ventriculo-peritoneal (V-P) shunt placement is very rare. Previous defects of the orbital wall with intracranial hypotension can cause enophthalmos after V-P shunting. The authors present 2 patients of enophthalmos with orbital wall defects resulting from anterior clinoidectomy that was performed during previous aneurysmal surgery. Both patients received a V-P shunt for hydrocephalus after subarachnoid hemorrhage. Although the hydrocephalus was improved by V-P shunts in both patients, sunken eyes were observed. The patients received reconstructive surgery of the superior orbital wall using titanium mesh and recovered after surgery without any neurological deficits. Here, the authors present 2 patients of enophthalmos with orbital wall defects treated by orbital wall reconstruction.

Key Words: Intracranial hypotension, orbital wall defect, reconstruction, sunken eyes

(*J Craniofac Surg* 2016;27: 1789–1791)

The causes of enophthalmos include trauma, infection with human immunodeficiency virus, and tumor.[1] Sunken eyes after the placement of a ventriculo-peritoneal (V-P) shunt can also occur, but this is very rare. However, in patients with orbital wall defects, enophthalmos can occur through the defect after V-P shunting induces intracranial hypotension. A defect in the superior orbital wall can also occur as a result of anterior clinoid process removal during the craniotomy procedure for aneurysmal surgery. Enophthalmos should be treated because of the potential for an eyeball movement disorder or a cosmetic problem. In this report, we present 2 patients of enophthalmos caused by orbital wall defects.

From the *Department of Neurosurgery, Chonnam National University Medical School; and †Department of Oral and Maxillofacial Surgery, Chonnam National University, School of Dentistry, Gwangju, Korea.
Received March 23, 2016; final revision received May 8, 2016.
Accepted for publication May 15, 2016.
Address correspondence and reprint requests to Tae-Sun Kim, MD, PhD, Professor, Department of Neurosurgery, Chonnam National University Hospital, 8 Hak-dong, Dong-gu, Gwangju 501-190, Republic of Korea; E-mail: taesun1963@yahoo.co.kr
This study was supported by a grant (CRI13902-21) from the Chonnam National University Hospital Biomedical Research Institute.
The authors report no conflicts of interest.

ISSN: 1049-2275
DOI: 10.1097/SCS.0000000000002956

CLINICAL REPORTS

Patient 1

A 54-year-old woman was transferred to our hospital with drowsiness. She had undergone craniotomy and clipping of an aneurysm in another hospital due to the rupture of a basilar tip aneurysm. A standard pterional craniotomy and anterior clinoidectomy were performed to repair the ruptured aneurysm. On admission, the patient was treated with antibiotics for meningitis related to the external ventricular drain. After the infection had improved, a follow-up computed tomography (CT) scan revealed hydrocephalus. We performed a V-P shunt to treat the hydrocephalus using a programmable valve. A follow-up CT scan showed that the hydrocephalus had not improved after placement of the V-P shunt. After reducing the pressure setting, the hydrocephalus improved (Fig. 1A). However, a superoposteriorly deviated eyeball had newly developed through the orbital wall defect (Fig. 1B–D). Contouring of Medpore and titanium (Ti)-mesh was achieved using a preoperative three-dimensional CT scan (Fig. 2A). The superior orbital rim was exposed by a bicoronal incision, followed by dissection of the internal orbital cavity while peeling the supraorbital nerve that passes through the supraorbital notch (Fig. 2B). The dura mater and orbital septum were separated from the defective orbital bone to the superior orbital fissure because of their adhesion. Herniation of the superior rectus muscle and orbital fat had occurred through the orbital wall defect. The Ti-mesh was inserted after checking the accuracy of the location in the orbital matrix using navigation. A forced-duction test confirmed the lack of limited eye movement. The Ti-mesh was fixed on the superior orbital margin using a screw, and Medpore was put on the depressed temporal muscle. The enophthalmos showed an improvement compared with the previous condition (Fig. 3A). We confirmed on postoperative CT that the orbital wall was properly reconstructed (Fig. 3B and C). The sunken eye improved after surgery, and there was no neurological deficit including blindness, or restriction of eyeball movement.

Patient 2

A 43-year-old woman was transferred to our hospital with an altered mental state. Diffuse subarachnoid hemorrhage and an internal carotid artery aneurysm (ophthalmic segment) were found on a CT scan. A right-sided pterional craniotomy was performed. Because bleeding reoccurred after the initial surgery, a subsequent decompressive craniectomy and Guglielmi detachable-coil embolization were performed. A V-P shunt was placed because of hydrocephalus that was detected on follow-up CT. The hydrocephalus improved, but enophthalmos was observed in this patient. A superior orbital wall defect had also occurred in the course of the pterional approach. It was difficult to increase the shunt pressure setting due to the hydrocephalus, which prompted a reconstruction, as in patient 1. The enophthalmos improved without any other neurological abnormalities.

DISCUSSION

Sunken eyes are a relative posterior displacement of the eyeball that occurs within the orbital cavity. When enophthalmos occurs in only

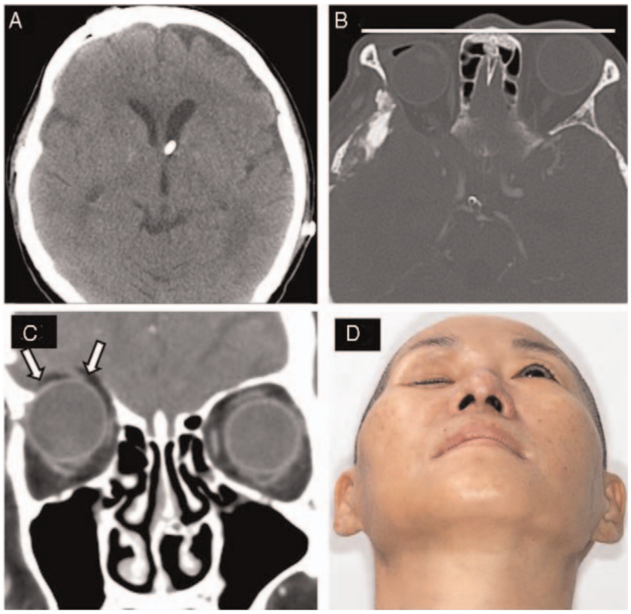

FIGURE 1. Hydrocephalus improved in the follow-up computed tomography after adjusting the pressure setting (A). The eyeball deviated superoposteriorly through the orbital wall defect (B, C). Enophthalmos observed as the patient looked up with head extension (D).

1 eye, it can be diagnosed based on a difference between the eyes that exceeds 2 mm.[1] Causes of enophthalmos include trauma, human immunodeficiency infection, chronic maxillary atelectasis, orbital varix, and breast cancer metastasis.[1] The development of sunken eyes after the placement of a V-P shunt is very rare. After V-P shunting, the decreased mechanical stress induced by chronic intracranial hypotension remodels the skull. The balance is upset between bone absorption and formation that is mediated by mechanical stress. Bone formation toward the intracranial side occurs, and this is an area of relatively low pressure. As a result, the increased orbital volume causes enophthalmos without intraorbital soft tissue atrophy.[2,3]

Enophthalmos has been reported to improve after the correction of intracranial hypotension in the short and long terms.[2] Therefore, it is important to maintain adequate intracranial pressure after V-P shunt placement. For this reason, frequent observation is necessary through examinations like CT. However, enophthalmos can occur despite the maintenance of adequate intracranial pressure, as shown in patient 1. We speculate that patients with orbital defects are highly likely to develop enophthalmos even though the intracranial pressure might be maintained at an appropriate level. Most orbital defects are caused by trauma. As in our patients, anterior clinoid process removal during aneurismal craniotomy can result in an excessive deficit of the superior orbital wall without trauma. It is

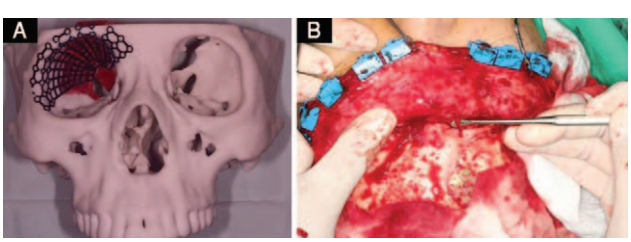

FIGURE 2. Contouring of the titanium-mesh was done using preoperative three-dimensional computed tomography (A). Peeling of the supraorbital nerve passing through the supraorbital notch (B).

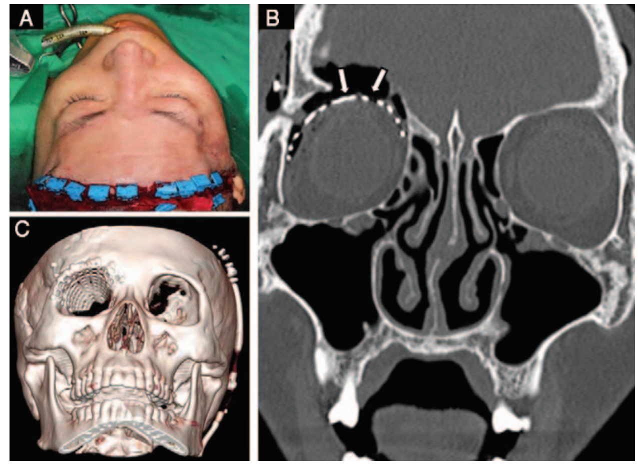

FIGURE 3. Enophthalmos showing a marked improvement compared with the previous condition (A). The orbital wall was well reconstructed and the enophthalmos had improved on postoperative computed tomography (B, C).

therefore important to avoid creating an orbital wall defect during the removal process to prevent enophthalmos. Of course, enophthalmos dose not occur after VP shunt in all patients with iatrogenic superior orbital wall defects such as syndromic craniosynostosis. Hydrocephalus and intracranial hypertension are common complications in syndromic craniosynostosis.[4] As a result of posterior fossa constriction, resistance to cerebrospinal fluid outflow is increased and venous outflow obstruction occurs.[4] In this patient, enophthalmos did not appear after VP shunt even though a superior orbital wall defect was present due to craniofacial surgery. We assume that the volume of brain gradually increased during growth after fronto-supraorbital surgery and intracranial hypertension has been already existing for a long time after birth in syndromic craniosynostosis patient. Therefore, structural intracranial hypotension is unlikely to have occurred after surgery. Because the incidence and mechanism of enophthalmos are unknown in the literature, further studies are needed to confirm. If the patient does not display any symptoms or if the bony defect is small, conservative treatment is sufficient.[5] In our patients, the bony defects measured 3.6 and 2.1 cm^2. Even if the bony defect is small, enophthalmos can still occur after V-P shunt placement. Therefore, it is difficult to determine the criteria for quantifying a defect that requires orbital wall reconstruction. Surgical treatment is considered if the patient has problems such as enophthalmos, double vision, loss of vision, or cosmetic concerns. In many patients in whom surgery is delayed for a variety of reasons, incomplete correction of the orbit can be caused by soft tissue atrophy, scar contracture, or remnant scar tissue attached to the fracture site.[6] For these reasons, the prognosis for early surgery is good. If an operation is indicated for the patient, surgery should be performed as soon as possible. The goal of surgery is the restoration of the herniated orbital contents and reconstruction of the orbital rim. The most common complication that occurs in the reconstruction process is damage to the optic nerve. The optic nerve is vulnerable to ischemic injury because of the narrow bony structure surrounding it. Careful subperiosteal dissection during surgery is crucial. Customized mesh and accurate implantation are essential to prevent various complications. Mesh implantation using a navigation system has been introduced to protect the orbital structure and its safety has been proven.[7–9] The navigation system provides valuable information about orbital plate fitting including the size and shape, visualization of dissection, and implantation in the area adjacent to the optic canal.[10] Several materials are used in reconstructive surgery including autologous bone, absorbable implants, and Ti-mesh. Each has its own advantages,[11,12] but Ti-mesh is superior to the other materials in terms of cost and effectiveness.[13] Another important aspect is the forced-duction test. In the test, the eye is moved by grasping the conjunctiva with forceps, providing information about mechanical restrictions and the release of herniated muscle intraoperatively.[14]

CONCLUSION

Enophthalmos after V-P shunt placement is rare but it can develop with intracranial hypotension when an orbital wall defect exists. A bony defect of the orbital roof may occur as a result of clinoidectomy regardless of the level of surgical skill. Experienced neurosurgeons who can drill the skull base competently should be more careful not to drill the orbital roof excessively to preserve the eggshell thick compact bone. They should also be very careful not to injure the orbital fascia allowing orbital fat protrusion. Not only those who are young and inexperienced but also skilled neurosurgeons should be careful not to destroy the orbital roof to avoid delayed enophthalmos. Because enophthalmos is likely to occur at lower intracranial pressures, it is necessary to maintain adequate pressure. Early reconstruction of the orbital wall is recommended when enophthalmos occurs in a situation where it is difficult to control the pressure setting due to hydrocephalus.

REFERENCES

1. Hamedani M, Pournaras JA, Goldblum D. Diagnosis and management of enophthalmos. *Surv Ophthalmol* 2007;52:457–473
2. Hwang TN, Rofagha S, McDermott MW, et al. Sunken eyes, sagging brain syndrome: bilateral enophthalmos from chronic intracranial hypotension. *Ophthalmology* 2011;118:2286–2295
3. McCulley TJ. Sphenoid sinus expansion: a radiographic sign of intracranial hypotension and the sunken eyes, sagging brain syndrome (an American Ophthalmological Society thesis). *Trans Am Ophthalmol Soc* 2013;111:145–154
4. Collmann H, Sorensen N, Krauss J. Hydrocephalus in craniosynostosis: a review. *Childs Nerv Syst* 2005;21:902–912
5. Sung YS, Chung CM, Hong IP. The correlation between the degree of enophthalmos and the extent of fracture in medial orbital wall fracture left untreated for over six months: a retrospective analysis of 81 cases at a single institution. *Arch Plast Surg* 2013;40:335–340
6. Kim YH, Ha JH, Kim TG, et al. Posttraumatic enophthalmos: injuries and outcomes. *J Craniofac Surg* 2012;23:1005–1009
7. Gabrielli MF, Monnazzi MS, Passeri LA, et al. Orbital wall reconstruction with titanium mesh: retrospective study of 24 patients. *Craniomaxillofac Trauma Reconstr* 2011;4:151–156
8. He W, Zhang Y, He Y, et al. Correction of enophthalmos following orbital fracture with computer-assisted navigation system. *Zhonghua Kou Qiang Yi Xue Za Zhi* 2014;49:641–644
9. Metzler P, Ezaldein HH, Pfaff MJ, et al. Correction of severe enophthalmos by simultaneous fat grafting and anatomic orbital reconstruction. *J Craniofac Surg* 2014;25:1829–1832
10. Kim YH, Jung DW, Kim TG, et al. Correction of orbital wall fracture close to the optic canal using computer-assisted navigation surgery. *J Craniofac Surg* 2013;24:1118–1122
11. Clauser L, Galie M, Pagliaro F, et al. Posttraumatic enophthalmos: etiology, principles of reconstruction, and correction. *J Craniofac Surg* 2008;19:351–359
12. Fernando Gonzalez Magana D, Rodrigo Menendez Arzac MD, Laura De HilarioAviles MD. Combined use of titanium mesh and resorbable PLLA-PGA implant in the treatment of large orbital floor fractures. *J Craniofac Surg* 2011;22:1991–1995
13. Nkenke E, Vairaktaris E, Spitzer M, et al. Secondary reconstruction of posttraumatic enophthalmos: prefabricated implants vs titanium mesh. *Arch Facial Plast Surg* 2011;13:271–277
14. Metz HS. Forced duction, active force generation, and saccadic velocity tests. *Int Ophthalmol Clin* 1976;16:47–73

