Surgical Outcomes of Intracanal Cavernous Venous Malformation According to Their Location in Four Right-Angled Sectors

Min Ho Kim, MD, Ji Hyun Kim, MD, Sung Eun Kim, MD, and Suk-Woo Yang, MD, PhD

Abstract: The present study evaluated the surgical outcomes of intracanal cavernous venous malformation according to their location in 4 right-angled sectors. Data regarding the surgical method and approach, surgical outcome, and postoperative complications were retrospectively analyzed for 18 patients with intracanal cavernous venous malformations that were surgically treated at the authors’ center between March 2006 and May 2017. The lesion location was defined using 2 perpendicular lines connecting the optic disc and the 4 surrounding rectus muscles in the coronal plane, which resulted in the formation of 4 right-angled sectors (upper-outer quadrant, upper-inner quadrant, lower-inner quadrant, and lower-outer quadrant). Accordingly, there were 3, 3, 8, and 4 lesions in the upper-inner, upper-outer, lower-inner, and lower-outer quadrants, respectively. Ten patients received anterior orbitotomy and 8 received lateral orbitotomy. There were no recurrences during the follow-up period. All patients exhibited reduced proptosis after surgery. Vision improved in 4 patients and remained unchanged in 14. Five patients experienced ocular movement limitation (1 permanent and 4 temporary), 1 developed an inferomedial blowout fracture, 2 developed a temporary sensory deficit, and 1 developed temporary ptosis. The authors’ findings suggest that intracanal cavernous venous malformations are most frequently occur in the lower-outer quadrant. Although most lesions can be removed via anterior orbitotomy, large lesions located near the orbital apex or on the orbital wall require lateral orbitotomy. Ocular movement limitation is a common complication and can become permanent in rare cases, necessitating close monitoring. Lesion location and surgical approach do not seem to influence the postoperative complication rate.

Key Words: Complication, intracanal cavernous venous malformation, location, orbitotomy, surgical resection

Orbital cavernous venous malformations (CVMs)—previously known as cavernous hemangiomas—are the most common form of primary orbital lesions in adults, comprising 4% to 9% of all orbital tumors. Women in their 40s and 50s are predominantly affected, with the lesion often arising in an intracanal space and presenting with characteristic 1-sided, progressive, asymptomatic proptosis. Other accompanying signs and symptoms include decreased visual acuity, blurred vision, visual field defects, ocular movement limitation, choroidal folds, and optic disc changes (ie, papilledema).

Computed tomography (CT) and magnetic resonance imaging (MRI) are 2 frequently used imaging techniques for diagnosis of these lesions, which are observed as round, ovoid, or rarely lobulated lesions with clear boundaries and mild enhancement on contrast-enhanced CT or MRI.

For patients with an asymptomatic lesion or a small lesion that does not progressively increase in size, close observation is often sufficient. However, patients should receive surgical treatment if the lesion increases in size and causes symptoms such as progressive proptosis, pain, diplopia, or ocular movement limitation, or increases the risk of complications from optic nerve or eyeball compression, such as decreased vision or visual field defects.

Common surgical methods for the resection of intracanal CVMs include anterior orbitotomy, lateral orbitotomy, and an endoscopic transnasal approach. Recent reports suggest that transnasal endoscopic surgery using 3-dimensional intraoperative navigation provides favorable surgical outcomes. This surgical method enables reduction in the amount of bleeding during surgery or complication rate, as well as shorter hospitalization and minimal scarring after surgery, thereby providing a favorable aesthetic outcome. Multiple factors—including oral health, age, presence of pain and chronic disease, and postoperative disfiguring scars’—have been reported to affect health-related quality of life; therefore, shortening the duration of surgery through a minimally invasive approach will allow improved satisfaction for patients, because it allows better management of postoperative pain, more rapid recovery, and minimal scar formation.

Recently studied biomaterials include marine collagen, which exhibits osteoconductive abilities and recombinant human bone morphogenetic protein (rhBMP-2) (Infuse Bone Graft—Medtronic, Memphis, TN), both of which are characteristics that induce osseous regeneration; these materials can be utilized in a complementary manner in bone defects, including blowout fracture. In addition,
possible postoperative complications of enophthalmos may be corrected using these biomaterials. Therefore, appropriate use of biomaterials may provide good cosmetic outcomes.19,20

The anatomical location of the lesion and its relationship with nearby orbital structures are the 2 most important factors for deciding the surgical method.2 Previous studies have determined the frequency and surgical outcomes of these lesions after grouping them according to their location using various methods7,21-23 including the 8-sector (medial, lateral, superior, inferior, inferior lateral, superior lateral, inferior medial, and superior medial),22 4-sector (superior, medial, inferior, and lateral),23 and 3-sector (three 120° sectors)13 methods. However, no study has determined the location of the lesions on the basis of 4 right-angled sectors (upper-inner, upper-outer, lower-inner, and lower-outer quadrants). Considering that all the existing methods are sound methods for dividing the location of the lesions, it would be difficult to claim that any one of these analysis methods is better than the others. However, if the location of the lesions is divided with too much anatomical precision when analyzing the surgical approach and outcomes according to the location of the lesions, then it may be too ambiguous in defining the margins (boundaries) of the lesions. For these reasons, we took a simple grouping method with 4 right-angled sectors, using the optic disc in the center and 4 surrounding rectus muscles in the coronal plane. With this method, the lesions can be analyzed by simply categorizing their location into upper-inner, upper-outer, lower-inner, and lower-outer quadrants depending on whether the lesions are located in upper, lower, inner, or outer side. By grouping the lesions with such criteria, the margins can be defined more clearly, which is expected to make it easier to analyze the surgical outcomes according to the location of the lesions.

Therefore, the aim of the present study was to determine the surgical approach and outcome and postoperative complications for intraconal CVMs stratified according to their location in 4 right-angled sectors.

METHODS
In total, 18 patients diagnosed with and surgically treated for intraconal CVMs at Seoul St. Mary’s Hospital, The Catholic University of Korea between March 2006 and May 2017 were included in this study. Patients with extraconal CVMs and other intraorbital tumors were excluded. The Institutional Review Board/Ethics Committee of Seoul St. Mary’s Hospital approved this study and it was performed in accordance with the tenets of the Declaration of Helsinki.

All surgeries were performed under general anesthesia, with the surgical method and approach determined according to the location of the lesion. Lateral orbitotomy was performed for large tumors (≥3 cm) located near the optical apex because these lesions are associated with a higher risk of optic nerve damage during surgery. Tumors on the orbital wall that were difficult to access or visualize were also resected via lateral orbitotomy. All other lesions were removed via anterior orbitotomy (Figs. 1 and 2). Anterior orbitotomy was performed using a transcutaneous or transconjunctival approach based on the location of lesion and its accessibility. If accessibility was hindered by the eyelids, anterior orbitotomy with the vertical lid split procedure was performed (Fig. 2).

Data regarding the surgical method and approach, surgical outcomes, and postoperative complications were retrospectively analyzed for all 18 patients. We defined the location of the intraconal CVMs using 2 perpendicular lines connecting the optic disc and the 4 surrounding rectus muscles in the coronal plane. This resulted in the formation of 4 right-angled sectors (upper-upper quadrant, upper-inner quadrant, lower-upper quadrant, and lower-inner quadrant; Fig. 3). The best corrected visual acuity, intraocular
pressure (IOP), ocular motility, and degree of proptosis (using Hertel exophthalmometer) before and after surgery were also recorded. Other accompanying signs and symptoms that could affect the visual acuity were analyzed using anterior segment examination and fundus examination. Computed tomography or MRI studies were performed before and after surgery for all patients (Fig. 4).

RESULTS

There were 4 men and 14 women with a mean age of 44.8 ± 10.4 years (range: 31–62 years). The patients visited our center with the following chief complaints: proptosis (n = 13), decreased vision (n = 2), ocular pain (n = 1), and upward deviation of the eyeball (n = 1).

Clinical signs included progressive axial proptosis (n = 16), extraocular muscle movement limitation (n = 6), decreased visual acuity (n = 5), relative afferent pupillary defect (n = 3), papilledema of the optic nerve (n = 2), strabismus (n = 2), retinal folding (n = 1), and optic disc pallor (n = 1).

The preoperative best corrected visual acuity [logarithm of the minimum angle of resolution (LogMAR)] was 0.2 ± 0.5, IOP was 15.3 ± 3.8 mm Hg, and degree of proptosis was 3.3 ± 1.7 mm (range: 1–6 mm; Table 1).

All patients had unilateral lesions. The lesions were located on the right side in 8 patients and left side in 10 patients. The lowerouter quadrant was the most common lesion location (n = 8; 44.4%), followed by the lower-inner (n = 4; 22.2%), upperouter (n = 3; 16.7%), and upper-inner (n = 3; 16.7%) quadrants (Table 2). Among the 8 patients with lowerouter quadrant lesions, 2 received anterior orbitotomy and 6 received lateral orbitotomy. All 4 patients with lowerinner quadrant lesions and all 3 patients with upperinner quadrant lesions received anterior orbitotomy. One of the 3 patients with upperouter quadrant lesions received anterior orbitotomy; the remaining 2 received lateral orbitotomy. Details of the surgical approaches are summarized in Table 2.

After the surgery, all patients exhibited significantly reduced proptosis. Visual acuity improved in 4 patients (22.2%) and remained unchanged in 14 (77.8%); none of the patients experienced deteriorated vision. However, there was no significant difference between the preoperative and postoperative visual acuities. Similarly, although IOP exhibited an overall decrease after surgery, the difference was not statistically significant (Table 1).

Postoperative complications included ocular movement limitation (n = 5), temporary sensory deficit (n = 2), inferomedial blowout fracture (n = 1), and temporary ptosis (n = 1). In total, 50% of the patients exhibited postoperative complications. The ocular movement limitation was temporary in 4 patients and spontaneously resolved during the follow-up period. The remaining patients exhibited permanent limitation and required strabismus surgery. The patient with the inferomedial blowout fracture had a lower-inner quadrant lesion. This patient was originally taken up for surgery via a transethmoidal endoscopic approach by neurosurgeons and ear-nose-throat specialists. However, the patient was transferred to the ophthalmology department with incomplete resection and eventually underwent complete resection using a transconjunctival approach, along with surgical repair of the inferomedial blowout fracture (Table 3).

The mean postoperative follow-up period was 11.2 ± 10.9 months and no recurrence was observed during this period.

DISCUSSION

In this study, we used a simple anatomical localizing system to group intracanal CVMs and characterize the treatment approach and follow-up for these lesions, according to location. We have shown that intracanal CVMs most frequently occur in the lowerouter quadrant. We have also highlighted that although most lesions can be removed via anterior orbitotomy, large lesions located near the orbital apex or on the orbital wall require lateral orbitotomy. We found ocular movement limitation to be one of the commonest

| Location of Lesion | Surgical Approach |
|-------------------|-------------------|
| Lower-outer quadrant (n = 8) | Anterior orbitotomy (n = 2) |
|                    | Lateral orbitotomy (n = 6) |
| Lower-inner quadrant (n = 4) | Anterior orbitotomy (n = 4) |
| Upper-outer quadrant (n = 3) | Anterior orbitotomy (n = 1) |
| Upper-inner quadrant (n = 3) | Anterior orbitotomy (n = 3) |

Two perpendicular lines connected the optic disc and the 4 surrounding rectus muscles in the coronal plane, resulting in 4 right-angled sectors (upper-inner, upper-outer, lowerinner, lower-outer).

After surgery, all patients exhibited significantly reduced proptosis. Visual acuity improved in 4 patients (22.2%) and remained unchanged in 14 (77.8%); none of the patients experienced deteriorated vision. However, there was no significant difference between the preoperative and postoperative visual acuities. Similarly, although IOP exhibited an overall decrease after surgery, the difference was not statistically significant (Table 1).

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The mean postoperative follow-up period was 11.2 ± 10.9 months and no recurrence was observed during this period.

**TABLE 1. Ophthalmological Parameters Before and After Surgery for Intraconal Cavernous Hemangiomas in Eighteen Patients**

|                      | Before Surgery | After Surgery | P Value |
|----------------------|---------------|---------------|---------|
| BCVA (LogMAR)        | 0.2 ± 0.5     | 0.1 ± 0.5     | 0.059   |
| IOP (mm Hg)          | 15.3 ± 3.8    | 13.9 ± 3.1    | 0.122   |
| Proptosis (mm)       | 3.3 ± 1.7     | 0.9 ± 1.0     | 0.000   |

BCVA, best corrected visual acuity; IOP, intraocular pressure; logMAR, logarithm of the minimum angle of resolution.

*Statistically significant difference (paired t test; SPSS 22.0).

**TABLE 2. Surgical Approaches for Intraconal Cavernous Hemangiomas (n = 18) According to Their Location in Four Right-Angled Sectors**

| Location of Lesion | Surgical Approach |
|-------------------|-------------------|
| Lower-outer quadrant (n = 8) | Anterior orbitotomy (n = 2) |
|                    | Lateral orbitotomy (n = 6) |
| Lower-inner quadrant (n = 4) | Anterior orbitotomy (n = 4) |
| Upper-outer quadrant (n = 3) | Anterior orbitotomy (n = 1) |
| Upper-inner quadrant (n = 3) | Anterior orbitotomy (n = 3) |

Lower transconjunctival approach (n = 2), Eyelid crease approach (n = 5), Transcanthal approach (n = 1), Lower transconjunctival approach (n = 2), Medial and inferior caruncular approach (n = 2), Vertical lid split procedure (n = 1), Eyelid crease approach (n = 2), Vertical lid split procedure (n = 1), Transconjunctival approach (n = 2).

**TABLE 3. Postoperative Complications in Patients With Intraconal Cavernous Hemangiomas Treated by Anterior or Lateral Orbitotomy Using Various Approaches**

| Complication                      | Number of Patients | Percentage (%) |
|-----------------------------------|--------------------|----------------|
| Ocular movement limitation        | Temporary          | 4              | 22.2           |
|                                   | Permanent          | 1              | 5.6            |
| Temporary sensory deficit         | 2                  | 11.1           |
| Inferomedial blowout fracture     | 1                  | 5.6            |
| Temporary ptosis                  | 1                  | 5.6            |
| Total                             | 9                  | 50.1           |

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comparisons, with the lesion’s location and the surgical approach used not influencing the postoperative complication rate. Our findings provide important data to inform the treatment and follow-up of patients with intraconal CVMs approached surgically.

Orbital CVMs are the most frequent orbital lesions in adults and are classified as low-flow venous malformations under The International Society for the Study of Vascular Anomalies classification system.2,24

The expression of progesterone receptors in the epithelial cells of orbital CVMs is a known cause for the frequent occurrence in women in their 40s and 50s.2,6,8,25 In the present study, 77.8% of patients were women with a mean age of 44.8 years, consistent with the demographics in previous studies.

The most frequent clinical sign of orbital CVMs is progressive and asymptomatic proptosis, with possible accompanying conditions including decreased visual acuity, pain or pressure in the eyes, blurred vision, visual field defects, diplopia, headache, and ptosis.2,3–6 Orbital CVMs often occur in the intracranial space,2,6,8 with progressive axial proptosis as the most frequently observed clinical sign. Furthermore, patients are often not aware of the lesions because of their slowly progressive nature.2,3,11,26 We only included patients with intraconal CVMs in the present study and observed that proptosis was the most prevalent clinical sign, followed by blurred vision, decreased visual acuity, pain in the eyes, and upward deviation of the eyeball.

Computed tomography and MRI are 2 frequently used imaging studies for the diagnosis of the lesions, and ultrasound and nuclear imaging are also occasionally used. In the present study, CT or MRI was performed before and after surgery for diagnosis and evaluation of the surgical outcomes, respectively.

Cavernous venous malformations are reported to occur more frequently on the left side and within the intracranial space of the eye socket. Different studies have used different methods for defining the locations of these lesions as there is currently no standard method. However, the most frequent site of origin for CVMs is the lateral part of the optic nerve.2,22,23 and the inferior quadrant.21 Although it remains unclear why the lesion frequently occurs on the left side, the reason for the frequent onset at the lateral part of the optic nerve within the intracranial space is that this region is thought to be the arterial side of the circulation, with extensive distribution to be the arterial side of the circulation, with extensive distribution of small arteries.2,22,32,32 Similarly, there were more lesions on the left side (n = 10) than on the right side (n = 8) in the present study. In addition, the lower-inner quadrant of the intracranial space was the most common location (44.4%), followed by the lower-inner (22.2%), upper-inner (16.7%), and upper-inner (3 patients, 16.7%) quadrants; these findings were in agreement with those of previous studies reporting a greater frequency in the lower-inner quadrant of the optic nerve.

Cavernous venous malformations, in general, are slow-growing tumors. Therefore, if the lesion is asymptomatic, close observation of the patient is often sufficient without additional treatment. However, if the patient exhibits signs and symptoms such as progressive proptosis, decreased visual acuity, visual field defects, pain, diplopia, or ocular movement limitations, surgical treatment is required.2,4,8,10,11 All the patients in the present study exhibited clinical signs or symptoms that were indications for surgical treatment. In general, anterior orbitotomy and lateral orbitotomy are frequently used surgical procedures, with the endoscopic transnasal approach being the more frequently used approach of late.12–14 Anterior orbitotomy is an appropriate surgical procedure for most intraconal lesions located in front of the orbital space or at the base of the optic nerve without intrusion into the orbital apex. The transconjunctival or transcutaneous approach can be utilized for this procedure.2,18,28–31 For better exposure of the lesion during surgery, lateral canthotomy can be performed in parallel.

Furthermore, the lesion can be conveniently removed with a cryoprobe.29,30,32,33 In the present study, 10 patients received anterior orbitotomy and 8 received lateral orbitotomy (Table 2). A cryoprobe was used in 8 of the 10 (80%) patients who received anterior orbitotomy. Lateral orbitotomy can be used if the lesion is located near the orbital apex, in the outer or upper/lower region of the optic nerve, and in the posterior third of the eye socket. This method was introduced by Kronlein for the first time in 1889 and several modifications have been made since then, allowing for frequent use of the procedure in current clinical practice.5,8,11,31,34–37 This approach can be considered if the lesion is a large tumor (>3 cm in diameter) without clear boundaries or with suspicious malignant changes, or if the lesion is attached to the orbital wall or surrounding tissues, consequently leading to poor accessibility and a poor visual field. The use of this approach can minimize postoperative complications in such cases.29,34–40

Among the 10 patients who received anterior orbitotomy, 8 received surgeries using a transconjunctival approach and medial/inferior caruncular approach without any difficulty in lesion exposure. However, it was difficult to access the tumor in the remaining 2 patients because of the upper eyelids; therefore, we performed anterior orbitotomy using the vertical lid split procedure. All 8 patients who received lateral orbitotomy had large tumors (>3 cm) located in the upper-inner or lower-inner quadrants and attached to the orbital walls and tissues. We used this approach to secure a clear view and minimize potential postoperative complications in these patients (Table 2).

If the lesion is located in the outer regions of the optic nerve, is small in size, and can be easily exposed, anterior orbitotomy would be the best surgical method regardless of its vertical position (upper or lower). However, if the lesion is large in size, located near the optical apex, or attached to the orbital walls and tissues, lateral orbitotomy would be a better option to ensure patient safety, regardless of the lesion’s vertical position (upper or lower). Lesions located in the upper-inner region of the optic nerve are often not easily accessible because of the upper eyelids. For these lesions, anterior orbitotomy with the vertical lid split procedure would be the best surgical approach. Meanwhile, lesions located in the lower-inner region of the optic nerve can normally be removed using anterior orbitotomy with extensive dissection of the episcleral venous plexus and the use of beveling. This approach also involves less dissection of extraocular muscles, with the endoscopic transnasal approach can also be used for lesions closer to the orbital apex.

Anterior orbitotomy with the vertical lid split procedure has been introduced as a method that allows effective exposure of superomedial orbit lesions—the most difficult lesions to access—and removal of huge intraconal CVMs in a relatively safe manner, without complications.29,41–43 Furthermore, postoperative eyelid management is performed using direct closure; therefore, this is considered a surgical option that can maintain normal eyelid function without complications, and with good cosmetic outcome.25

Furthermore, recent studies have suggested that sclerotherapy is both safe and highly effective as first-line treatment of orbital low flow vascular lesions.44–46 Agents currently used for such treatment include OK-432 (Picibanil; Chugai Pharmaceutical Co, Ltd., Tokyo, Japan), sodium tetradecyl sulfate, 5% sodium morrhuate, and bleomycin A5. In general, sclerotherapy is utilized as an alternative to surgery or to treat and shrink lesions prior to surgery.45 Intralesional injections are performed to increase treatment efficacy and the use of ultrasound guided or fluoroscopically guided puncture may allow for a safer surgical procedure.44–46 In addition, recent studies have demonstrated methods for easier and safer access to the orbital lesion by using navigation and endoscopy; the use of sclerotherapy might provide more effective treatment for patients.13,47–49 More specifically, transnasal endoscopic surgery using three-dimensional intraoperative navigation based on
Preoperative CT scans is a surgical method that can be utilized for orbital surgeries (eg, complex orbital fracture), as well as for other types of surgery (eg, skull base, orthognathic, sinus, trauma, oncology, and neurosurgery). With greater visualization of the lesion, this surgical method will likely become more accurate and safer than traditional methods.11-13

Postoperative complications include early (ie, lid swelling, periorbital edema, chemosis, transient ocular movement limitation, temporary ptosis, and diplopia) and late complications (ie, enophthalmos, permanently decreased visual acuity or visual field defects due to circulatory or optic nerve damage, permanent ocular movement limitation, permanent ptosis, pupil dilation, and diplopia).4,8,10,11,13,50,51 In the present study, 50% of patients who received surgical treatment exhibited postoperative complications, including ocular movement limitation, a temporary sensory deficit, an inferomedial blowout fracture, and temporary ptosis. Most patients showed spontaneous resolution of their complications. One patient with persistent ocular movement limitation required strabismus surgery, while the patient with the inferomedial blow out fracture required surgical repair. The blow out fracture occurred during surgery by neurosurgeons and ear-nose-throat specialists, who attempted to remove the lesion in the lower-inner quadrant using an endoscopic approach. The surgery was inadequate and the patient was transferred to the ophthalmology department for additional surgery. Using a transconjunctival approach, we completely removed the lesion and simultaneously performed the inferomedial blow out fracture repair surgery (Table 3).

With regard to the association between postoperative complications and the surgical method, we observed that 5 patients who received anterior orbitotomy and 4 who received lateral orbitotomy developed complications. Accordingly, the surgical approach did not appear to be a factor for increased postoperative complications. In contrast, complications were more frequently observed in patients with lesions in the lower quadrants (3 lower-quadrant lesions and 3 lower-inner quadrant lesions) than in those with lesions in the upper quadrants (2 upper-quadrant lesions and 1 upper-inner quadrant lesion; Figs. 5 and 6). This finding can be attributed to the greater number of lesions in the lower quadrants than in the upper quadrants. The greater number of surgeries performed in the lower quadrants probably contributed to the increased frequency of complications. Thus, the location of the lesion, based on 4 right-angled sectors, did not seem to have an impact on the increased frequency of complications. Nonetheless, damage to surrounding tissues (ie, the optic nerve) during surgery is more likely to increase the chance of complications, and securing a clear view during surgery is an important factor to reduce the frequency of postoperative complications.

This study has some limitations. It is difficult to generalize our findings because of the small sample size and relatively short follow-up period. In addition, all surgeries were performed by a single surgeon, and it is difficult to apply the surgical outcomes of patients treated by a single surgeon to all CVMs. Future long-term studies with larger sample sizes are necessary to clarify our findings.

In conclusion, the findings of our study suggest that intraconal CVMs most frequently occur in the lower-quadrant. Although most lesions can be removed via anterior orbitotomy, large lesions that are attached to the orbital walls or tissues require lateral orbitotomy to secure a clear view of the lesion and ensure the patient’s safety. Ocular movement limitation is a common complication. Although it is mostly temporary, it can become permanent in some patients; therefore, close monitoring is mandatory. Finally, the location of orbital CVMs and the surgical approach do not influence the postoperative complication rate.

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