

Case Report

Spontaneous Intraorbital Hemorrhage : A Case Report

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Intraorbital hemorrhage is a rare clinical condition caused by orbital trauma, surgery around the orbit, intraorbital vascular abnormalities, and neoplasm. It was reported to occur spontaneously without any known causes and in association with orbital pseudotumor in a very few cases. A 59-year-old, female patient admitted with sudden onset of severe exophthalmos and pain on the left eye. Orbital CT and MR imaging suggested hemorrhage in the upper part of retrobulbar area of the left orbit. Cerebral angiography was taken to rule out any possible vascular abnormalities. On the left carotid cerebral angiography, the run-off of the distal ophthalmic artery was not seen and the engorgement of the supraophthalmic artery was noted. Systemic administration of corticosteroid did not improve the clinical status and craniectomy was done and retrobulbar hematoma was removed, and the clinical symptoms and signs were improved. Authors report a case of spontaneous intraorbital hemorrhage with the clinical features similar to those of orbital pseudotumor, requiring surgical decompression.

KEY WORDS : Intraorbital hemorrhage · Orbital pseudotumor · Surgical decompression.

INTRODUCTION

Intraorbital hemorrhage may occur after orbital trauma, surgery around the orbit, in patients with sinusitis^{6,10,14}. It may occur in patients with various vascular abnormalities such as intraorbital arteriovenous malformation, cavernous angioma, or aneurysms of the intraorbital ophthalmic artery, aneurysms of the ophthalmic vein^{6, 8,10,12,15}. Also, it may occur in association with neoplasms or idiopathic inflammatory orbital pseudotumor^{5,9,11,13}.

Intraorbital hemorrhage with vascular anomalies present with sudden or gradual onset of severe ophthalmic pain, exophthalmos, decreased visual acuity, diplopia, etc^{6,8,10,12,15}. Orbital pseudotumor with hemorrhage and spontaneous intraorbital hemorrhage without any causes was reported in very rare occasions. Most of the intraorbital hemorrhage developed symptoms mimicking orbital pseudotumor.

Authors experienced a case of spontaneous intraorbital hemorrhage requiring surgical decompression. The etiology of the hemorrhage could not be explained by existing knowledge, so we suggested the possible pathogenesis on the basis of vascular factors demonstrated by cerebral angiography and report this rare case of intraorbital hemorrhage.

CASE REPORT

A 59-year-old, otherwise healthy, female patient admitted with sudden onset of severe exophthalmos and pain of her left eye. Visual acuity of left eye was markedly decreased to the level of detecting hand motion and ocular motility was almost limited at all directions. The eyelid was edematous and severe chemosis was also evident. Intraocular pressure was 34 mmHg and no significant change was noted on ophthalmoscopic examination.

History of systemic lymphadenopathy, thyropathy, thrombocytopeny, other autoimmune disease, and previous trauma were not seen. Laboratory examinations were within normal limits without evidence of bleeding tendency. Endocrine function evaluations including thyroid function test showed no abnormalities. Computed tomography of orbit revealed elliptical high density mass in the upper part of retrobulbar area of the left orbit (Fig. 1A). It was seen as inhomogenous signal intensity on the T1-weighted image with diffuse enhancement after injection of Gd-DTPA (Fig. 1B, C). On the left carotid cerebral angiography, the run-off of the distal ophthalmic artery was not seen and the engorgement of the superior ophthalmic artery was noted (Fig. 2).

Systemic corticosteroid was administered, but patient's clinical symptoms and signs did not improve. On the 10th hospital day, operation was done. Bicoronal scalp incision was made and craniectomy was extended down to the orbital roof

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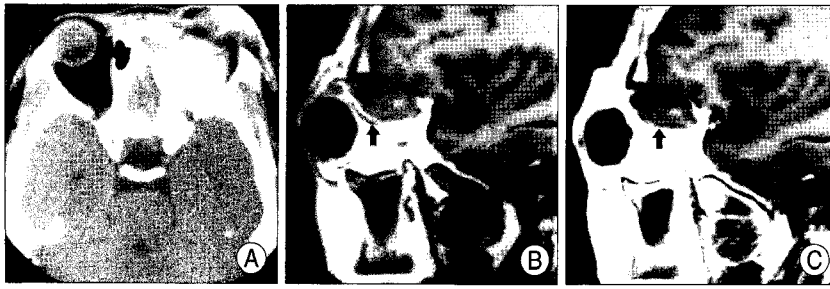


Fig. 1. Preoperative findings of the orbital computed tomography (CT) and magnetic resonance image. A : Orbital CT revealed high density mass in the upper part of the left orbital cavity. B : On T1-weighted sagittal image, superior orbital muscular structures are compressed by inhomogenous signal intensity mass with elliptical margin. C : On Gd-enhanced sagittal image, the crescent shaped margin of the mass is enhanced inhomogeneously (arrow).

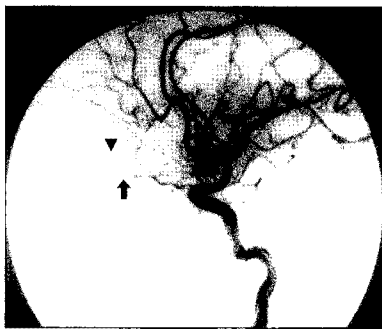


Fig. 2. Engorged supraorbital artery (arrowhead) is shown, and distal run-off of the ophthalmic artery (arrow) is not shown on left internal carotid angiogram.



Fig. 3. At 6 months after the operation, abnormal findings were not shown on T1-weighted axial magnetic resonance image.

surgery, the visual acuity was improved but oculomotor nerve palsy was still remained. There was no mass lesion noted on MRI (Fig. 3) and no episode of recurrence during the follow-up period of 10 years.

DISCUSSION

Intraorbital hemorrhage was reported to occur after surgery or trauma around the orbit, in patients with sinusitis^{6,10,14}. It may occur in patients with intraorbital arteriovenous malformation, cavernous angioma, or aneurysms of the intra-

and orbital unroofing was done. Clot-formed hematoma was found in the retrobulbar area and was removed. Retrobulbar fat tissue was excised enough for decompression. There was no gross identifiable tumorous condition. Histopathological examination revealed that most of the tissue consisted of fat with little inflammatory cells. Neither tumor cell nor abnormal vascular structure were identified.

Postoperatively, exophthalmos and ophthalmic pain were subsided. Six months after the

with recovery of clinical signs and symptoms especially when associated with orbital pseudotumor⁹. But, most of intraorbital hemorrhage cases required surgical or endoscopic decompression because of mass effect persisted in spite of medical treatment with unrelieving clinical symptoms¹⁰⁻¹⁵.

Orbital pseudotumor is a nongranulomatous inflammatory process, causing painful exophthalmos, conjunctival edema, and diplopia, etc^{1,3,7}. Most of them are confined to the orbit and involve extraocular muscles, periorbita, optic nerve, lacrimal gland, or the inflammatory process may be diffuse^{2,4}. Differential diagnosis include acute inflammation and tumorous condition. Orbital pseudotumor occur with sudden onset and associated hemorrhage was reported on rare occasion^{9,13}.

Intraorbital hematoma without any definite causes also has been reported^{10,13,14}. A case of persistent hematoma with organization was suspected to be tumor by imaging study. Surgical removal of the mass was performed and histopathological examination revealed organized hematoma without evidence of tumor or vascular anomalies¹⁴.

The case similar to our patient was reported by Husag et al.⁶ Acute exophthalmos was developed on the right eye in 13-year-old female patient, and treated by surgical removal of the hematoma with improvement of the clinical symptoms. The etiology was not identified. They suggested hemorrhagic diatheses, arterial, vasomotoric and toxic diseases, the congestive and traumatic causes as a possible causes. Another case was reported by Tsutsumi et al¹³. Sudden onset of retroorbital pain, visual loss, periorbital ecchymosis and double vision without preceding trauma, paranasal sinus surgery, or infections signs on the left eye in 75-year-old patient was seen, and treated with high dose steroid initially under a diagnosis of pseudotumor. But, surgery was performed because unresponsive to steroid therapy. Postoperatively, his visual acuity and other symptom were improved.

Authors report a case of spontaneous intraorbital hemorrhage in whom the hematoma was located in upper area of

orbital ophthalmic artery, aneurysms of the ophthalmic vein, and orbital pseudotumor, or other inflammatory disease^{5,8,9,10,11,12,15}.

The symptoms occur abruptly or gradually and diagnosis is made by cerebral angiography in addition to orbital computed tomography and MRI. Initial management of retrobulbar hemorrhage is usually made by administering high-dose corticosteroid^{9,12,13}. It is an effective treatment

the orbit. Computed tomography and MRI of the brain support the lesion to be the hematoma. Angiography revealed the run-off of the distal ophthalmic artery was not seen and the engorgement of the superior ophthalmic artery. Because of any definite causes were not found in our patient, our first diagnosis was orbital pseudotumor with hemorrhage, but not confirmed by pathologic studies. Any inflammatory condition of the orbit might have increased intraorbital pressure obstructing venous drainage. Consequently, increase in intraluminal pressure with resultant hemorrhage could have been taken place.

CONCLUSION

In patient with intraorbital hemorrhage, attention should be given to the various possible etiologies, and treatment should be based on each cause. Spontaneous intraorbital hemorrhage is a rare condition and surgical decompression is required in patient with steroid unresponsiveness. One of the suggested pathogenetic mechanism of hemorrhage is increased intraluminal pressure by inflammation of the retro-orbital tissue.

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