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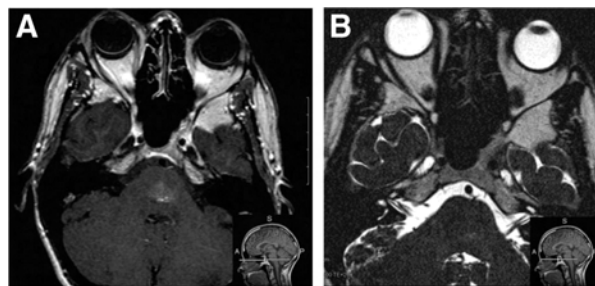
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## Blepharospasm in a Patient With Pontine Capillary Telangiectasia

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**Abstract:** Blepharospasm is rarely due to an identifiable etiology. In the majority of cases, imaging fails to reveal any structural lesion. Here we describe an otherwise healthy patient with blepharospasm who was found to have pontine capillary telangiectasia. We propose a potential association between blepharospasm and pontine capillary telangiectasia.

The cause of blepharospasm is unknown in almost all patients. If neuroimaging is obtained, it seldom reveals a structural lesion. We describe a patient with blepharospasm who



Magnetic resonance images revealing pontine capillary telangiectasia. An axial, contrast-enhanced T1-weighted image (3-mm slice, TR 500, TE 25) (A) shows feathery, stippled enhancement in the pons with dilated veins and no mass effect. Only slightly increased signal intensity is visible on an axial T2-weighted image (1.4 mm, TR 1500, TE 250) (B). TR, time to repeat; TE, time to echo.

was found to have pontine capillary telangiectasia, a heretofore unrecognized potential association.

### CASE REPORT

An otherwise healthy 30-year-old woman presented with a 1-year history of typical bilateral blepharospasm. She had an increased rate of blinking, and occasional spasms of involuntary eyelid closure. These varied in frequency, but occurred often enough to impair her daily activities. No facial nerve weakness, apraxia of eyelid opening, or sensory tics were found. She was treated with botulinum toxin injections by a dermatologist. She later consulted an ophthalmologist to exclude any underlying eye disease that might be contributing to her blepharospasm. No significant personal or family medical history, nor any history of dry eye, corneal exposure, or previous ocular trauma was known. She had no alcohol or drug addiction, perinatal asphyxia, or exposure to neuroleptic drugs. The patient did not complain of light sensitivity. Her visual acuity was 20/20 without correction in both eyes. The pupils, extraocular eye movements, and ocular alignment were normal. Facial sensation and movement were intact and symmetric. Apart from intermittent blepharospasm, no other abnormalities were noted on neurologic examination. The patient had a magnetic resonance imaging scan that demonstrated an abnormality in the pons. An axial, contrast-enhanced T1-weighted image (3-mm slice, TR 500, TE 25, see Fig. A) showed feathery, stippled enhancement with dilated veins. Only slightly increased signal intensity was visible on an axial T2-weighted image (1.4 mm, TR 1,500, TE 250, see Fig. B), and no mass effect or distortion of nearby structures was seen. These findings are consistent with pontine capillary telangiectasia.<sup>2</sup> Gradient-echo imaging was not performed in this case.

### DISCUSSION

Pontine capillary telangiectasia usually exhibits a benign clinical course, but it has been described in association with symptoms such as hearing loss, tinnitus, vertigo, unsteadiness, and headache.<sup>2</sup> Blepharospasm has been reported in cases involving other pontine lesions such as demyelinating plaques, lacunar strokes, and neoplasm.<sup>3,4</sup> The normal blink reflex involves a circuit consisting of an afferent limb, some central integration, and an efferent limb. It is generally accepted that blepharospasm results from some disruption in this circuit. In the case presented, the abnormality is located in the ventral pontine tegmentum. Although the facial nuclei are located in this region and may be affected, it is also possible that the telangiectasia is interfering with interneuronal circuits afferent to the facial nuclei. Descending corticobulbar inputs could also be affected.

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A wide range of values has been published for the prevalence of focal dystonia, and the actual number of cases is difficult to estimate, as the studies differ greatly in design.<sup>5</sup> A recent estimate of blepharospasm prevalence for patients aged 30 to 49 years is 0.0024%.<sup>6</sup> This value falls in the lower end of the published data, which range from 0.0016% to 0.0133%, but many findings indicate that the prevalence of blepharospasm increases with age.<sup>7</sup> Those studies that do stratify patients by age show similar rates for the young age group in which the described patient belongs. The prevalence of pontine capillary telangiectasias is also uncertain, as these vascular malformations are usually asymptomatic and are often found only incidentally, but it has been estimated at between 0.1% and 0.7%.<sup>8,9</sup> Based on these prevalence data, less than 1 in 1 million patients will have both blepharospasm and pontine capillary telangiectasia as a matter of chance. Thus we cannot exclude a coincidence, but the occurrence of both phenomena in our patient raises the possibility of a causative association.

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## Acute Recurrence of Orbital Cavernous Hemangioma in a Young Man: A Case Report

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**Abstract:** A 29-year-old man presented with a history of prominent left eye of 6 months' duration. He also reported fluctuating blurred vision since 15 days ago. On examination, proptosis of 3 mm was noted in the left eye. Computed tomography (CT scan) of the orbits showed a well-circumscribed, hyperdense, intraconal

mass lesion in left orbit, located in the inferotemporal quadrant. Orbitotomy was performed, and the tumor was delivered with an intact capsule. The clinical diagnosis of cavernous hemangioma was confirmed on histopathologic examination. The presence of intact capsule was confirmed grossly and with histopathology. The patient returned 6 weeks after surgery with recurrent proptosis. Repeated CT scan showed a recurrent intraconal mass of similar characteristics as in the primary presentation. A repeated orbitotomy was done, and the tumor was removed intact. Histopathology of the recurrent tumor confirmed the diagnosis of cavernous hemangioma. We report a rare case of orbital cavernous hemangioma with short-term recurrence (6 weeks).

Cavernous hemangioma is the most common benign tumor of the orbit in adults.<sup>1</sup> It is typically single and unilateral and does not generally recur after complete surgical excision. Recurrent multiple cavernous hemangiomas coexisting with concurrent systemic tumors have been reported.<sup>2</sup> Cavernous hemangioma is believed to recur from intrinsic vasculature in response to a proliferated stimulus for years after removal. Recurrence has been reported even after a decade.<sup>3</sup> However, short-term recurrence (6 weeks) of cavernous hemangioma has not been reported to the best of our knowledge. We present one such case of cavernous hemangioma that recurred within 6 weeks after complete removal.

## CASE REPORT

A 29-year-old man presented with a history of prominent left eye of 6 months' duration. He also reported fluctuating blurred vision for the past 15 days. His visual acuity in both the eyes was 20/20 N6 with normal color vision. The right eye was essentially normal. Proptosis of 3 mm with hyperglobus of 2 mm was noted in the left eye (Fig. 1). The ocular movements were full and free. A grade 1 relative afferent pupillary defect was seen; the rest of the anterior segment was normal. Dilated fundus examination showed blurring of optic disc margins in the left eye. The CT scan of the orbits showed a well-circumscribed, hyperdense, intraconal soft-tissue mass in the left orbit in the inferotemporal quadrant. The lesion was seen to be displacing the optic nerve nasally and abutting the eyeball (Fig. 2A,B). A clinical diagnosis of orbital cavernous hemangioma was made. Swinging transconjunctival inferior orbitotomy was planned to remove the tumor. The tumor, measuring 20 × 15 ×



**FIG. 1.** Clinical photograph of the patient showing proptosis of left eye and hyperglobus eye at the time of initial presentation.

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