Blepharospasm in a Patient With Pontine Capillary Telangiectasia

Aubrey L. Gilbert, M.D., Ph.D.*, William P. Dillon, M.D.†, and Jonathan C. Horton, M.D., Ph.D.*

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...
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. A recent estimate of blepharospasm prevalence for patients aged 30 to 49 years is 0.0024%. 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. 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%. 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.

REFERENCES


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

Manju Meena, M.D., Milind Naik, M.D., and Santosh Honavar, M.D., F.A.C.S.

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. 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. 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. 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 papillary 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.