Tonic pupil following surgical removal of an orbital cavernous haemangioma

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ABSTRACT

Orbital trauma and surgery are recognised aetiological factors of tonic pupil. Tonic or Adie's pupil is an efferent pupil defect in which light reactions to one or more segments of the iris sphincter are lost due to the postganglionic parasympathetic nerves damage from ciliary ganglion. There is loss of part or all of the light reflex and decrease in accommodative functions at near. We report a case of a 42-year-old lady who developed a tonic pupil after a successful surgical removal of an orbital cavernous haemangioma.

Keywords: Adie's pupil, cavernous haemangioma, Holmes-Adie's syndrome, tonic pupil

INTRODUCTION

A tonic pupil is usually caused by damage to the postganglionic parasympathetic pupillomotor fibers. An idiopathic tonic pupil, usually seen in young female patients is termed Adie's pupil. Orbital trauma and surgery are known aetiological factors of tonic pupil. Rarely there can be a systemic association. We report a case of tonic pupil developing after a successful removal of an orbital haemangioma.

CASE REPORT

A 42-year-old Burmese lady presented to our eye casualty clinic with a week history of pain around her right eye. She gave a history of temporary obscuration of vision lasting for few minutes (amaurosis fugax), occurring recurrently for the past few months. She also gave a history of using hyperopic glasses in the past two years.

On examination, her corrected visual acuity was 6/9 [OD] 6/6 [OS] and her colour vision was normal. She was orthophoric and extra-ocular movements were normal. There was an axial proptosis of the right eye of about 4 mm (Fig. 1). Anterior segment examination of the right was otherwise normal. Both pupils were of equal and normal size, briskly reacting to direct and consensual reflex and also had brisk response to near reflex. Posterior segment examination revealed an optic nerve head oedema with dilatation and tortousity of retinal vessels. Anterior

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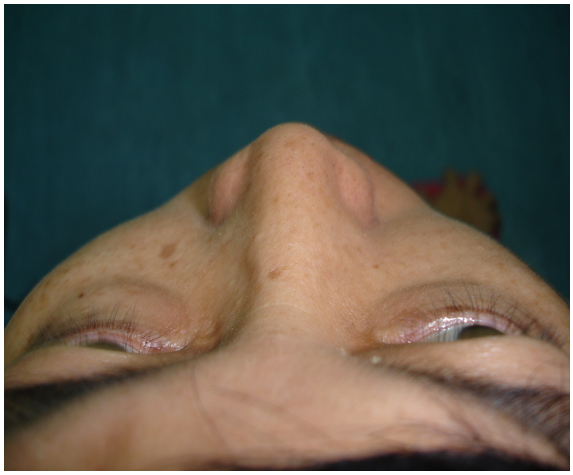

Fig 1: Examination showing right eye proptosis .

and posterior segment evaluations of the left eye were within normal limits. The intraocular pressures were 21 and 16 mmHg in the right and left eye respectively, with open angles. Visual field examination showed a reduction in sensitivity in the central field.

A computed tomography scan (CT) of the orbits and brain revealed a well-defined, oval, intraconal mass, in the superoposterior orbit of the right side, displacing the optic nerve nasally. The optic canals were normal and intracranial structures were normal (Fig. 2). These findings were later confirmed by magnetic resonance imaging (MRI).

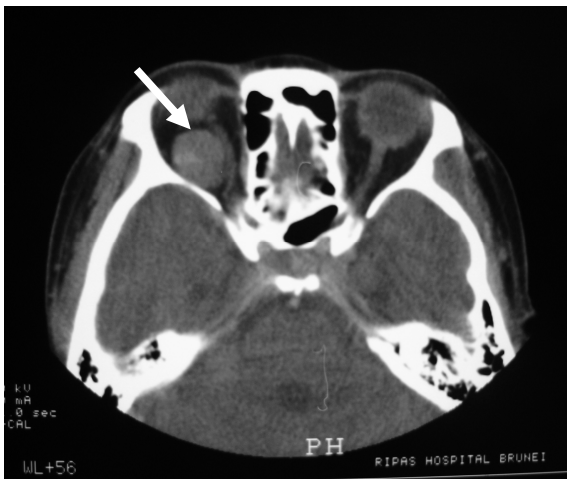

Fig. 2: Axial computed tomography of the orbit showing a right intraconal mass (arrow).

After proper evaluation of her physical condition, she underwent an excision biopsy of the orbital mass thorough a lateral orbitotomy. Histopathological examination of the mass revealed thin and thick walled vascular spaces lined by endothelial cells in a desmoplastic stroma with occasional smooth muscle cells. Some of the vessels contained thrombus (Fig. 3).

Postoperative evaluation one week later, showed her corrected visual acuity to be 6/9 in the right eye. The proptosis had resolved almost completely. Fundus examination showed a resolving optic nerve head oedema with normal retinal vessels. She had an anisocoria, with larger pupil on the right. The right eye pupil reacted poorly to light and accommodation, showing slow constriction and re-dilatation. On slit lamp examination, the right pupil also had an abnormal shape. It showed superior segmental paralysis with flattening of the normal iris surface crypts. There was bunching of the pupillary border inferiorly.

Tonic pupil was confirmed by the presence of a hypersensitive sphincter to diluted pilocarpine solution, which does not affect normal sphincter. Pilocarpine (0.1%) was instilled into both eyes. Examination after 30 minutes showed the right pupil to be smaller in size than the left (Fig. 4). Postoperative central visual field showed normal sensitivity. Postoperative CT scan showed no residual lesion. Postoperatively, a gradual resolution of the proptosis (Fig. 5) and disc oedema were noted and she continued to be followed up regularly.

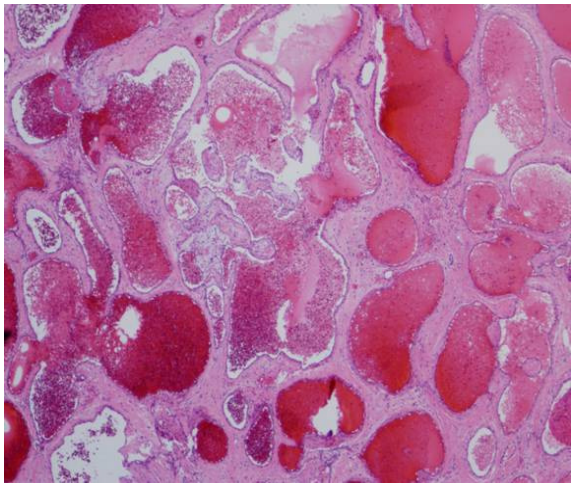

Fig 3: Histology showing thin and thick walled vascular spaces with thrombus. (H&E stain, x10).

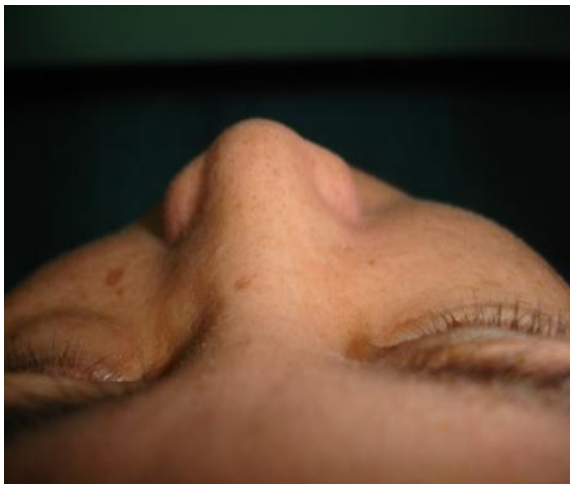

Fig 5: Postoperative picture showing resolution of proptosis of the right eye.

DISCUSSION

Orbital trauma is one of a number of causes for a tonic pupil.[1, 7-9] An idiopathic tonic pupil occurring in young adults either unilateral or bilateral is termed Adie's pupil. Holmes-Adie syndrome includes other features such as diminished deep tendon reflexes and orthostatic hypotension.[7] Other causes include herpes zoster, giant cell arteritis, diabetes mellitus, syphilis, alcoholism, laser or cryotherapy.[8] There are also reports of tonic pupil occurring bilaterally in acute pancreatitis, Sjogren's syndrome and Vogt-Koyanagi-Harada syndrome.[3-6]

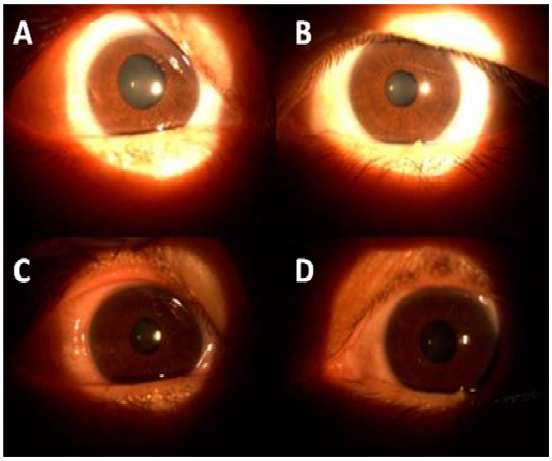

Fig. 4: Reaction of right and left pupils to light (A and B respectively) showing dilated right pupil; and pupils of the right and left eyes of the same patient following instillation of diluted pilocarpine (C and D respectively).

Tonic pupil is caused by postganglionic parasympathetic pupillomotor damage, either to ciliary ganglion or distal to it. In most cases the clinical appearance is very typical. In the initial stages, the tonic pupil is dilated, reacts poorly to light and shows slow constriction and redilatation to near reflex. Due to this slow redilatation, the tonic pupil might appear relatively smaller after near effort.

Slit lamp examination often reveals segmental palsy of the iris sphincter and vermiform movement of the pupillary border in the non-paralytic segment. The postganglionic denervation super-sensitivity can be demonstrated by instilling pilocarpine (0.1%), where the tonic pupil will be smaller than the normal fellow pupil. In most cases the clinical appearance is very typical and pharmacological confirmation is not needed.[8] Patients with tonic pupil may have asthenopia (symptoms arising from accommodative effort) or photophobia but most are asymptomatic. [7-9] Surgical trau-

ma to the short ciliary nerves or the ciliary ganglion itself may have perpetuated the development of a tonic pupil in our patient.

In conclusion, Adie's pupil or tonic pupil is a benign postganglionic parasympathetic denervation of diverse causes that include trauma, orbital surgery and infections. Despite meticulous dissection during orbital surgery, nerve injury can still occur (as in our patient). Hence, patients undergoing orbital surgery should be made aware of this rare complication preoperatively.

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