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A case of Adie’s tonic pupil

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The authors stated that the written consent was obtained from the patient presented with images in the study.

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Abstract

Adie's tonic pupil, the round unilateral tonic pupil, is a neuro-ophthalmologic disorder with dilation and no light response. Although usually idiopathic, it may be associated with infection, inflammation, and postganglionic paraneoplastic nerve damage. It is called Holmes-Adie syndrome when deep tendon reflex loss accompanies Adie’s tonic pupil. The diagnosis is made by observing a miotic response after dilated pilocarpine instillation. Spontaneous recovery depends on etiological factors.

Keywords: Adie syndrome, Tonic pupil, Pilocarpine, Anisocoria

Introduction

Adie's tonic pupil (Holmes-Adie syndrome) is usually characterized by unilateral pupillary dilatation and decreased light reflex. It may be accompanied by a decrease in deep tendon reflexes [1]. Its annual incidence is 4-7/100.000, and it is more common among females in their thirties [2, 3]. While many diseases including ocular infections and inflammations, peripheral and autonomic neuropathies, paraneoplastic syndromes, toxicity, tumors, and trauma can cause tonic pupil, idiopathic cases are most encountered. The tonic response seen during convergence is more distinct than that seen during light reflex and the diagnosis is based on tonic constriction after pilocarpine instillation [4]. It is believed that Adie's tonic pupil occurs due to damage in the postganglionic parasympathetic nerves of the eye [3, 4]. We aimed to review the basic and clinical features of the subject through a case diagnosed with Adie’s tonic pupil.

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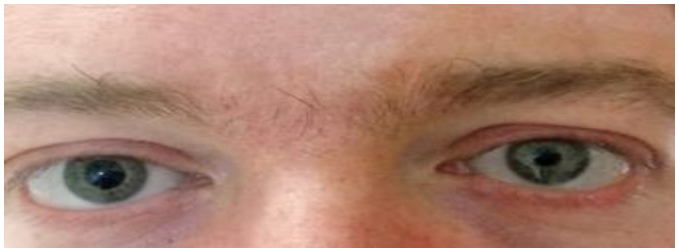
Case presentation

A 34-year-old male patient visited our neurology outpatient clinic in the past month with the complaint of blurred vision, especially when close to an object. The patient was evaluated in the ophthalmology clinic with complaints of itching and watering in his right eye two months ago, diagnosed with allergies, and prescribed betamethasone + sulfacetamide and fusidic acid drips. Neurological examination revealed a dilated right pupil (Figure 1) which was unresponsive to light and lost deep tendon reflexes.

Figure 1: Anisocoria – mydriasis on the right



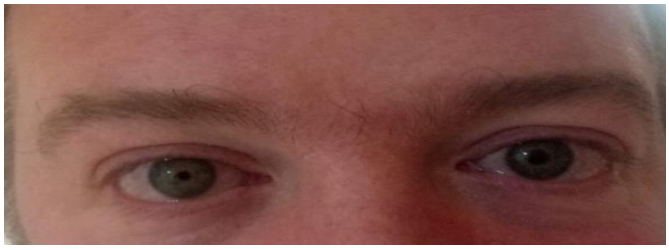
Ocular trauma, inflammation, migraine, benign episodic pupillary mydriasis, Adie's tonic pupil, oculomotor nerve paralysis, pharmacological agent use, and paraneoplastic conditions were considered as the preliminary diagnoses in the patient with anisocoria. The patient's cranial magnetic resonance (MR), cranial MR angiography and orbital MR imaging, hemogram, sedimentation, biochemistry, paraneoplastic, infectious parameters were within normal limits.

A 0.1% drop prepared by diluting 1% pilocarpine (Pilosed®) drop with ringer lactate was instilled in both eyes equally, after which the left pupil remained unchanged, but significant shrinkage was observed on the right (Figure 2). Hence, the patient was diagnosed with Adie's tonic pupil.

Clinical signs and symptoms of the patient were completely improved within two months.

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Figure 2: After 0.1% pilocarpine drop, there is myosis in the right pupil, and no change on the left.



Discussion

A difference of more than 0.1 mm between the pupil diameters is called anisocoria. In all patients with anisocoria, the shape of the pupil, its dimensions at dark and light, response to light and near stimuli, eyelid conditions, and eye movements should be evaluated. If anisocoria is more pronounced in the dark, the smaller pupil should be considered the problem and pathologies related to the sympathetic system should be investigated [5]. If the anisocoria is more prominent in a bright environment, parasympathetic system pathologies should be assessed. Unilateral, isolated anisocoria causes include ocular trauma, inflammation, Adie's tonic pupil, oculomotor nerve paralysis, pharmacological agent exposure, keratoplasty operation, paraneoplastic syndromes, syphilis, benign episodic pupillary mydriasis, migraine, and seizures [6]. In our case, Adie's tonic pupil was diagnosed due to the absence of similar complaints before this visit, no pathology in imaging studies, near-light dissociation, and response to 0.1% pilocarpine in the affected eye. Denervation hypersensitivity refers to the increased miotic response observed in the tonic eye compared to the unaffected eye when 0.1% pilocarpine solution is dropped on both. Thompson et al. suggested that cholinergic hypersensitivity is not typical but pathognomonic for a tonic pupil [7].

William et al. [8] stated that following ciliary ganglion injury, light-near dissociation develops in the iris sphincter of the tonic eye due to postganglionic parasympathetic denervation. Other causes of light-close dissociation include Argyll Robertson pupil following Herpes Zoster Ophthalmicus, aberrant degeneration of the third nerve, juvenile diabetes, myotonic dystrophy, dorsal midbrain syndrome of Parinaud, pituitary tumors, encephalitis, and chronic alcoholism [8].

Although the prognosis of Adie's tonic pupil disease is benign, cases with angle-closure glaucoma have been reported in the literature [9-10].

Conclusion

Patients with anisocoria should undergo a thorough neurological examination and other underlying diseases should be investigated with cranial MR, cranial MR angiography, and orbital MR before making the diagnosis of Adie's tonic pupil. Adie's tonic pupil is a rare disease that should be considered in patients with anisocoria.

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