

A rare pupillary phenomenon: Tadpole pupil

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The tadpole pupil is the most uncommon form of pupillary deformity. Erlenmeyer^[1] initially introduced it in 1912, while Thompson et al.^[2] delineated its clinical features in 1986. The term “tadpole” was assigned due to the resemblance of the pupil’s shape during the episode. The phenomenon arises from transient segmental spasms of the iris dilator muscle. It generally manifests in females and is unilateral.^[2] Identification is difficult owing to its benign characteristics and brief symptom duration.

A 33-year-old male patient was admitted to our neurology clinic with blurred vision and episodic pupillary deformity in the left eye. The patient’s anamnesis revealed a 1-cm deep injury to the anterior surface of the left axillary region caused by a sharp instrument after a fall 16 years ago, accompanied by reduced sweating in the left chest area after the incident. Aside from experiencing migraines, the patient’s overall health was satisfactory, and he reported no chronic illnesses or regular medication use. The patient reported experiencing blurred vision two years ago, occurring two to three times daily over four days, with each episode resolving within seconds.

Upon admission to our clinic, it was noted that the patient experienced episodes of blurred vision in the left eye lasting 15 to 30 sec, occurring two to three times daily over a period of 10 days. An image of the left eye, captured with a mobile phone during the attack, is presented in Figure 1. The patient had been diagnosed with

migraine; however, he reported no pain during these episodes. Four months later, the patient presented with similar complaints regarding the right pupil during follow-up. The patient expressed no concerns regarding the left side. The complaint persisted two to three times daily for a duration of two days before concluding.

The neurological examination was normal. The examination revealed no segmental pupillary defect. The patient’s gaze was normal in all directions; pupils were isochoric, and light responses were elicited (Figure 2). Ophthalmological evaluation revealed that near vision, distance vision, color vision, and visual field, as well as intraocular pressure measurements, were all within normal limits. A slit lamp examination and fundus evaluation were conducted, revealing no conditions such as iris atrophy, uveitis, or



Figure 1. Left eye of the patient during an episode of tadpole pupil.

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Figure 2. Examination images demonstrating normal gaze in all directions with isochoric pupils.

vitritis. No abnormalities were detected on brain magnetic resonance imaging (MRI), thoracic MRI, and brain-carotid computed tomography angiography. Small hernias were identified on cervical MRI. Brachial plexus needle electromyography and sympathetic skin response testing were normal limits. No abnormalities were detected in the blood tests. The pilocarpine and apraclonidine tests were unavailable in Türkiye, preventing their administration.

The attacks occur intermittently and unpredictably, resulting in a lack of data in the dark or light response during the attack. Based on the images from the attack, the patient's clinic, and our findings, we concluded that the patient exhibited a bilateral tadpole pupil. The patient was informed of the benign nature and course of this condition, and written informed consent for publication was obtained.

A review of all reported tadpole pupillary cases from 2019 indicated that the phenomenon occurred in individuals aged 2 to 48 years, with approximately 80% of cases involving females. Furthermore, reports indicate that over 90% of cases are unilateral. Instances exceeding 5 min in duration are uncommon. It demonstrates clustering characteristics at specific intervals, as illustrated in our case.^[3]

The etiology of segmental iris dilator muscle spasms remains unclear; however, multiple hypotheses have been suggested. In contrast to the denervation hypersensitivity hypothesis proposed by Hansen and Møller,^[4] Udry et al.^[3] presented their perspectives on pathophysiological mechanisms, focusing on hormonal changes and the contractile properties of the iris dilator muscle. Severe hyponatremia and autonomic

neuropathy were proposed as potential factors associated with a bilateral tadpole pupil in the literature.^[5] The impact of propofol used in this instance was not addressed. The initial case documented in Türkiye, characterized by the emergence of a tadpole pupil during anesthesia induction, highlighted the need for a more thorough investigation of anesthetic agents.^[6] Our patient exhibited no electrolyte imbalance or use of medications. A case of bilateral tadpole pupils that developed after exercise and resolved spontaneously in a 12-year-old patient was reported.^[4] The potential role of catecholamine hypersensitivity in the etiology was discussed.^[4] In our case, the attacks were not linked to exercise. Tadpole pupils, which exhibited side-switching in our study, were observed in six out of 43 patients in the literature.^[3]

Although the cause is unknown, tadpole pupil has been reported to occur in association with certain clinical conditions. Thompson et al.^[7] reported Horner syndrome in 46% of cases in their series, reported Adie's pupil in 15%, and noted an association with migraine in 31% of instances. Patients with symptoms such as anisocoria, ptosis, and miosis may be tested with the alpha-2 adrenergic receptor agonist apraclonidine. Patients with reversal of findings are diagnosed with Horner syndrome.^[8] A low-dose pilocarpine test is conducted in patients exhibiting no light response and dilated pupils. Pilocarpine functions as an agonist of the muscarinic M3 receptor in the iris sphincter muscle, leading to mydriasis. Adie's tonic pupil can be diagnosed using this method.^[9] These findings were absent in our case. It is advisable to evaluate the potential association

of these syndromes in the differential diagnosis or etiology of tadpole pupil and to conduct pharmacologic tests when feasible.

The axillary injury and unilateral hypohidrosis in our patient prompted us to contemplate sympathetic system involvement. Instances of posttraumatic Horner syndrome were documented in the literature.^[10,11] Furthermore, sympathectomy surgery may be conducted on individuals who have hyperhidrosis. This procedure may also be conducted via the axillary region.^[12] Given the significant time elapsed since the trauma in our case, we found no pathology that directly impacts or compresses the sympathetic chain. The patient's unilateral decreased sweating following axillary injury indicated an impact on the sympathetic system. Although our patient did not exhibit Horner syndrome, we propose that it may have influenced the sympathetic response through an unidentified mechanism.

A variety of components within the visual system receive innervation from the preganglionic cervical sympathetic division of the autonomic nervous system. The iris dilator muscles are included in this category. Sympathetic signals originate in the upper thoracic spinal segments, travel to the superior cervical ganglion, and subsequently reach the eye. Symptoms of Horner syndrome may arise if there is an impact on the sympathetic system.^[13] In some instances, findings of Horner syndrome emerged after the development of the tadpole pupil.^[2] No findings of Horner syndrome were observed in our case; however, they may develop in the future. Current findings suggest that our patient developed a tadpole pupil as a result of posttraumatic sympathetic pathway involvement, the mechanism of which remains unidentified.

The tadpole pupil is often overlooked due to its rarity, temporality, and benign nature. Requesting photographs from patients experiencing episodic blurred vision, as demonstrated in our case, will aid in the diagnostic process. It requires no treatment. This report presented the 50th documented case of tadpole pupil in the literature.

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