



Bilateral tonic pupil during a migraine attack: a case report

Ana Carolina Montouro Storarri , Annelise Akemi Higa Lee , Renan Domingues 

Neurology Department, Santa Casa de Misericórdia de São Paulo, São Paulo, Brazil



Ana Carolina Montouro Storarri
anastorarri@gmail.com

Edited by:

Raimundo Pereira Silva-Néto

Keywords:

Tonic pupil
Adie pupil
Mydriasis
Migraine

Abstract

Introduction

Tonic pupil or Adie's pupil occurs due to parasympathetic denervation, and it is characterized by mydriasis with little or no response to light, with pupillary contraction to accommodation. It is caused by eye pathologies, such as infections, trauma, neoplasms, inflammatory diseases, and systemic diseases with autonomic dysfunction. Few cases have been reported of bilateral tonic pupils associated with migraine attacks.

Case report

Our aimed to describe the case of a young female patient with a history of chronic migraine without aura, who presented acutely with bilateral pupillary mydriasis during a migraine attack, characterized as tonic pupil, and to discuss the possible causes of mydriasis during a migraine attack.

Submitted: March 6, 2024
Accepted: March 25, 2024
Published online: March 28, 2024



Introduction

Tonic pupil or Adie's pupil occurs due to parasympathetic denervation and is characterized by mydriasis with little or no pupillary response to light, with pupillary contraction to accommodation. It is often unilateral but bilateral involvement is observed in up to 20% of patients. It occurs predominantly in young women (1). It is caused by eye pathologies, such as infections, including syphilis (2), trauma, neoplasms, inflammatory diseases (3) and may be associated with systemic diseases with autonomic dysfunction.

Cases of tonic pupil during migraine attacks have been previously described (4-10).

In 2003, Mylius et al. (11) evaluated the pupillary function of 42 patients with migraine versus 42 controls and showed a reduction in the speed and amplitude of pupillary constriction up to two days after the migraine attack, thus suggesting post-ganglionic parasympathetic dysfunction during the migraine attack.

Barriga et al. (12) proposed the term ciliary ganglioplegic migraine in 2011 to describe persistent mydriasis that can occur ipsilateral to pain during a migraine attack.

Case report

We report a case of a 30-year-old woman, without comorbidities, with chronic migraine without aura for 1 year, according to the criteria of the International Classification of Headache Disorders (ICHD-III) (13). The patient sought care at the emergency room of the "Santa Casa de Misericórdia de São Paulo" during a migraine attack. During this crisis she had sudden, bilateral visual blurring. She did not have eye pain, diplopia, ophthalmoparesis, or eyelid ptosis.

The neurological examination revealed regular pupils, with bilateral mydriasis, absent direct and consensual photomotor reflex and preserved miosis at convergence. No other abnormality was found the neurological examination. A 0.125% pilocarpine test was performed, which was positive, showing bilateral postganglionic parasympathetic denervation.

The patient underwent investigation with neuroimaging, cerebrospinal fluid, and laboratory tests, ruling out structural, infectious, including syphilis, and metabolic causes of pupillary abnormalities. The pupillary abnormality was attributed to parasympathetic dysfunction related to migraine.

The patient was reassessed two months after hospital discharge, when she reported no visual blurring since a few days after hospital discharge. The neuro-

ophthalmological examination showed reactive to light and accommodation pupils.

Discussion

Cranial autonomic symptoms (CAS) are common in migraine, including pupillary abnormalities, ptosis, eye redness, tearing, lid edema, nasal congestion and rhinorrhea - ocular CAS are the most common. Migraineurs with CAS present with higher headache intensity and frequency and chronic migraine is more frequent in these subjects (14-16).

Anisocoria may occur in migraine because of either abnormal miosis or mydriasis, indicating an autonomic imbalance between parasympathetic constriction and sympathetic dilation of the pupil. Miosis is the most reported pupillary abnormality, frequently associated to other CAS such as ptosis, due to oculosympathetic hypofunction (17, 18).

Mydriasis during a migraine attack can represent different situations: a comorbid concurrence of Adie's pupil and migraine, an ophthalmoplegic migraine (with a predominance of parasympathetic paresis), headache as a painful manifestation of ganglionitis and ganglioplegic migraine. In the present case the first possibility was more likely, considering the lack of other neuro-ophthalmologic abnormalities and the fact that it seemed to be an isolated episode.

The frequency of Adie's pupil during migraine attacks is still unknown. Its relationship with other pathophysiological elements, including other autonomic nervous system abnormalities (19) in migraine, is not yet clear. Therefore, more studies are needed to evaluate the relationship between parasympathetic dysfunction of the ciliary ganglion and migraine.

Author's contributions: All those designated as authors gave substantial contributions to this manuscript, drafted and revised it, approved its final version and agreed to be accountable for all aspects of the work.

Conflict of interests: The authors declare that there is no conflict of interest.

Funding: This research did not receive specific funding from any funding agency in the public, commercial, or not-for-profit sectors.

Ana Carolina Montouro Storarri:
<https://orcid.org/0009-0007-9460-206X>
 Annelise Akemi Higa Lee:
<https://orcid.org/0000-0002-0796-7156>
 Renan Domingues:
<https://orcid.org/0000-0002-6058-7937>



References

- Xu S-y, Song M-m, Li L, Li C-x. Adie's Pupil: A Diagnostic Challenge for the Physician. *Medical Science Monitor*. 2021;28. Doi 10.12659/msm.934657
- Sakai T. Bilateral Tonic Pupils Associated with Neurosyphilis. *Japanese Journal of Ophthalmology*. 2003;47(4):368-71. Doi 10.1016/s0021-5155(03)00058-3
- Bachmeyer C, Zuber M, Dupont S, Blanche P, Dhôte R, Mas JL. Adie Syndrome as the Initial Sign of Primary Sjögren Syndrome. *American Journal of Ophthalmology*. 1997;123(5):691-2. Doi 10.1016/s0002-9394(14)71084-0
- Woods D, O'Connor PS, Fleming R. Episodic Unilateral Mydriasis and Migraine. *American Journal of Ophthalmology*. 1984;98(2):229-34. Doi 10.1016/0002-9394(87)90359-x
- Purvin VA. Adie's tonic pupil secondary to migraine. *J Neuroophthalmol*. 1995;15(1):43-4.
- Massey EW. Pupillary Dysautonomia and Migraine: Is Adie's Pupil Caused by Migraine? *Headache: The Journal of Head and Face Pain*. 2005;21(4):143-6. Doi 10.1111/j.1526-4610.1981.hed2104143.x
- Iannetti P, Spalice A, Iannetti L, Verrotti A, Parisi P. Residual and Persistent Adie's Pupil After Pediatric Ophthalmoplegic Migraine. *Pediatric Neurology*. 2009;41(3):204-6. Doi 10.1016/j.pediatrneurol.2009.03.019
- Jacome DE. Status Migrainosus and Adie's Syndrome. *Headache: The Journal of Head and Face Pain*. 2002;42(8):793-5. Doi 10.1046/j.1526-4610.2002.02182.x
- Skeik N, Jabr F. Migraine with benign episodic unilateral mydriasis. *International Journal of General Medicine*. 2011. Doi 10.2147/ijgm.S18613
- Murphy SJX, Francis I, Nadarajan V. Prolonged Benign Episodic Unilateral Mydriasis in Hemiplegic Migraine. *Journal of Neuro-Ophthalmology*. 2024;44(1):e140-e. Doi 10.1097/wno.0000000000001661
- Mylius V, Braune HJ, Schepelmann K. Dysfunction of the pupillary light reflex following migraine headache. *Clinical Autonomic Research*. 2003;13(1):16-21. Doi 10.1007/s10286-003-0065-y
- Barriga FJ, de Silanes CL, Gili P, Pareja JA. Ciliary ganglioplegic migraine: Migraine-related prolonged mydriasis. *Cephalalgia*. 2010;31(3):291-5. Doi 10.1177/0333102410381144
- Headache Classification Committee of the International Headache Society (IHS) The International Classification of Headache Disorders, 3rd edition. *Cephalalgia*. 2018;38(1):1-211. Doi 10.1177/0333102417738202
- Cortez MM, Millsap L, Brennan KC, Campbell CL. Craniofacial Autonomic Dysfunction in Migraine: Implications for Treatment and Prognosis. *Journal of Neuro-Ophthalmology*. 2020;40(1):67-73. Doi 10.1097/wno.0000000000000876
- Singh A, Tiwari A, Maurya P, Qavi A, Kulshreshtha D, Thacker A. Cranial autonomic symptoms in migraine: An observational study. *Annals of Indian Academy of Neurology*. 2022;25(4). Doi 10.4103/aian.aian_948_21
- Riesco N, Pérez-Alvarez AI, Verano L, García-Cabo C, Martínez-Ramos J, Sánchez-Lozano P, et al. Prevalence of cranial autonomic parasympathetic symptoms in chronic migraine: Usefulness of a new scale. *Cephalalgia*. 2015;36(4):346-50. Doi 10.1177/0333102415593087
- Smith SV. Neuro-Ophthalmic Symptoms of Primary Headache Disorders: Why the Patient With Headache May Present to Neuro-Ophthalmology. *Journal of Neuro-Ophthalmology*. 2019;39(2):200-7. Doi 10.1097/wno.0000000000000790
- Yildiz MB, Yildiz E, Balci S, Hasirci Bayir BR, Çetinkaya Y. Effect of migraine attack on pupil size, accommodation and ocular aberrations. *European Journal of Ophthalmology*. 2020;31(6):3450-5. Doi 10.1177/1120672120975334
- Domingues RB, Fonseca KB, Ziviane LF, Domingues SA, Vassalo D. Altered Cardiovascular Reactivity to Mental Stress But Not to Cold Pressure Test in Migraine. *Headache: The Journal of Head and Face Pain*. 2010;50(1):133-7. Doi 10.1111/j.1526-4610.2009.01567.x