Case study

Adie's pupil (About a case)

ABSTRACT

The pupil of adie is unilateral mydriasis which does not respond to light stimulation. Generally, this syndrome is associated with many pathologies such as syphilis, autoimmune hepatitis, migraine or even sarcoidosis. The syndrome is caused by damage to the post ganglion fibers of the parasympathetic innervation of the eye. We report a case of a young man presenting for 1 year a pupil of adie. The pupil of adie is a rare manifestation, a complete clinical and paraclinical examination is necessarily associated with a test with the pilocarpine to establish a positive diagnosis.

INTRODUCTION

Adie's syndrome is a relatively common neurological disorder of unknown etiology comprising dilated unilateral or bilateral pupils with near-lumen dissociation. [1] The syndrome is caused by damage to the post ganglion fibers of the parasympathetic innervation of the eye. It is frequently observed in women with, in particular, tendon areflexia. [2] The diagnosis is obtained by means of the pilocarpine test diluted to 0.125%. We report the case of a young man with an isolated Adie pupil for a year.

Case Présentation

This is a young man aged 27, with no particular history, who consults for anisocoria discovered 10 months ago by the patient himself; without other accompanying signs. On examination we find a visual acuity of 10/10 in both eyes. The photomotor reflex examination found a right pupil in reactive semi-mydriasis, the consensual reflex is present, there is no PDAR (Figure 1). There was also no limitation of the oculomotor muscles. Eye tone and anterior

segment examination were normal in both eyes. The fundus examination was unremarkable. The examination of the osteotendinous reflexes was normal and the cerebral orbital MRI angiography is without abnormality. The pilocarpine test diluted to 0.125 is positive (Figure 2). The remainder of the neurological, cardiovascular and osteotendinous examination was normal. So we concluded that it is a pupil of Adie.

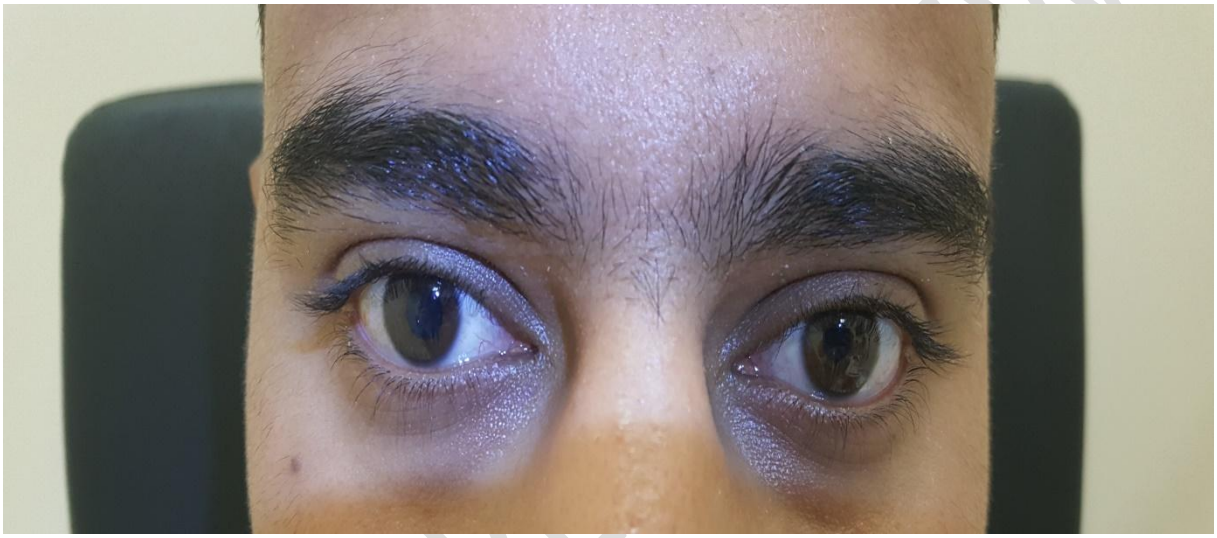

Figure 1 : Right eye showing a dilated pupil despite bright lighting (Adie's pupil)

Figure 2: Miosis pupil after the pilocarpine test confirming the diagnosis of the Adie's pupil

DISCUSSION

Adie syndrome is primarily idiopathic with no identifiable cause, but can rarely be caused by local disorders involving the orbit that affect the ciliary ganglion, including infections such as syphilis, chickenpox, human parvovirus B19, the virus human immunodeficiency and Lyme disease, [3] [4] ischemia due to lymphomatoid granulomatosis, migraine and giant cell arteritis, [5] [6] autoimmune diseases such as Sjögren's syndrome, polyarteritis nodosa, sarcoidosis, amyloidosis, Guillain-Barré and Vogt-Koyanagi syndrome -Harada disease, [7], local or general anesthesia, [8] orbital or choroidal tumors, [9] orbital surgery for orbital floor fractures, [10] neuromuscular conditions such as Lambert-Eaton myasthenic syndrome and paraneoplastic in association with anti-Hu antibodies. [11] It can also be caused by retinal photocoagulation which results in damage to the ciliary nerves in the suprachoroidal space. [12]

The incidence of Adie's syndrome is approximately 4.7 / 100,000 inhabitants / year with a prevalence of two cases / 1,000 inhabitants (approximately). [13] Young adults generally aged 25 to 45 are the most frequently affected, as is the case with our patient. On the other hand, there is a female predominance unlike our case, it was a male patient.

Adie's syndrome is a clinical diagnosis. A low concentration pilocarpine test (one-eighth to one-tenth percent) may be useful in demonstrating supersensitivity to cholinergic denervation (80% prevalent) in the tonic pupil. [14] After administration of diluted pilocarpine, a more miotic response will be observed in the affected eye compared to the other normal eye since these low concentrations are generally ineffective in normal pupils. [15]

There is no treatment and the pilocarpine, which contracts the pupil, is generally poorly tolerated; however, wearing tinted glasses does reduce photophobia. [16]

We often see a stability of mydriasis, sometimes it decreases slightly over time. Sometimes we can see bilateralization (rare). [17]

CONCLUSION

Adie's pupil is a rare manifestation, the eye examination is important and the pilocarpine test remains the fundamental diagnostic element.

REFERENCES

[1] Adie WJ. PSEUDO-ARGYLL ROBERTSON PUPILS WITH ABSENT TENDON REFLEXES: A BENIGN DISORDER SIMULATING TABES DORSALIS. Br Med J. 1931 May 30;1(3673):928-30.

[2]Thompson HS. Adie's syndrome: some new observations. Trans Am Ophthalmol Soc. 1977;75:587-626.

[3] ROSS AT. Progressive selective sudomotor denervation; a case with coexisting Adie's syndrome. Neurology. 1958 Nov;8(11):809-17.

[4] Weller M, Wilhelm H, Sommer N, Dichgans J, Wiethölter H. Tonic pupil, areflexia, and segmental anhidrosis: two additional cases of Ross syndrome and review of the literature. J Neurol. 1992 Apr;239(4):231-4.

[5] Sakai T, Shikishima K, Mizobuchi T, Yoshida M, Kitahara K. Bilateral tonic pupils associated with neurosyphilis. Jpn J Ophthalmol. 2003 Jul-Aug;47(4):368-71.

[6] Stricker RB, Winger EE. Holmes-Adie syndrome and Lyme disease. Lancet. 2001 Mar 10;357(9258):805. [PubMed]

[7] Cerny R, Rozsypal H, Kozner P, Machala L. Bilateral Holmes-Adie syndrome as an early manifestation of the HIV neuropathy. Neurol Sci. 2010 Oct;31(5):661-3.

[8] Foroozan R, Buono LM, Savino PJ, Sergott RC. Tonic pupils from giant cell arteritis. Br J Ophthalmol. 2003 Apr;87(4):510-2.

[9] Purvin VA. Adie's tonic pupil secondary to migraine. J Neuroophthalmol. 1995 Mar;15(1):43-4.

[10] Bennett JL, Pelak VA, Mourelatos Z, Bird S, Galetta SL. Acute sensorimotor polyneuropathy with tonic pupils and an abduction deficit: an unusual presentation of polyarteritis nodosa. Surv Ophthalmol. 1999 Jan-Feb;43(4):341-4.

[11] Garza Leon M, Herrera-Jimenez IP, González-Madrigal PM. Complete Vogt-Koyanagi-Harada disease and Holmes-Adie syndrome: case report. Ocul Immunol Inflamm. 2014 Aug;22(4):336-40.

[12] Guaraldi P, Mathias CJ. Progression of cardiovascular autonomic dysfunction in Holmes-Adie syndrome. J Neurol Neurosurg Psychiatry. 2011 Sep;82(9):1046-9.

[13] Kobayashi M, Takenami T, Kimotsuki H, Mukuno K, Hoka S. Adie syndrome associated with general anesthesia. Can J Anaesth. 2008 Feb;55(2):130-1.

[14] Goldstein SM, Liu GT, Edmond JC, Katowitz JA, Rorke LB. Orbital neural-glial hamartoma associated with a congenital tonic pupil. J AAPOS. 2002 Feb;6(1):54-5.

[15] Stromberg BV, Knibbe M. Anisocoria following reduction of bilateral orbital floor fractures. Ann Plast Surg. 1988 Nov;21(5):486-8.

[16] Bruno MK, Winterkorn JM, Edgar MA, Kamal A, Stübgen JP. Unilateral Adie pupil as sole ophthalmic sign of anti-Hu paraneoplastic syndrome. J Neuroophthalmol. 2000 Dec;20(4):248-9.

[17] Wirtz PW, de Keizer RJ, de Visser M, Wintzen AR, Verschuuren JJ. Tonic pupils in Lambert-Eaton myasthenic syndrome. Muscle Nerve. 2001 Mar;24(3):444-5.