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Case Report / Olgu Sunumu

Benign Episodic Unilateral Mydriasis (Case Report)

Benign Epizodik Unilateral Midriazis (Olgu Sunumu)

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Summary

Benign episodic unilateral mydriasis is a descriptive condition with recurrent unilateral mydriasis in adults, especially women with migraine. We report a 20 year-old male presenting with paroxysmal left pupil mydriasis and diagnosed as benign episodic unilateral mydriasis after exclusion of other reasons causing anisocoria. *(Turkish Journal of Neurology 2012; 18:111-3)*

Key Words: Anisocoria, benign, mydriasis

Özet

Benign epizodik midriazis çoğunluğu migrenöz kadınlarda olmak üzere genç erişkinlerde ortaya çıkabilen tekrarlayan izole unilateral midriazis atakları için kullanılan tanımlayıcı bir terimdir. Yazımızda aralıklı olarak sol gözbebeğinde büyüme şikayeti ile başvuran ve anizokori etiyolojisine yönelik tetkiklerin sonucunda benign epizodik unilateral midriazis tanısı alan 20 yaşında erkek hasta sunulacaktır. Benzer olgularda anizokori etiyolojisinin benign sebeplerinin akılda tutulması hastalara gerekli olmayan girişimsel tanısal yöntemlerin uygulanmaması konusunda uyarıcı olabilir. *(Türk Nöroloji Dergisi 2012; 18:111-3)*

Anahtar Kelimeler: Anizokori, benign, midriazis

Introduction

Anisocoria can present as a sign of potentially hazardous conditions including aneurisms, intracranial bleeding, cerebral neoplasms / compression, meningeal infiltration or tentorial herniation (1). Therefore, it is of utmost importance that the cause of anisocoria should be determined in patients. Benign episodic mydriasis, a relatively benign and quite rare cause, is a descriptive term used for recurrent isolated unilateral mydriasis attacks that may occur in young adults, mostly women with migraine (2,3). Although mydriasis typically presents in the same eye, the location may occasionally vary and it may last a few hours to days (4). As the mechanism is not clarified yet, diagnosis is by exclusion. While mydriasis may accompany a migraine or cluster headache attack or may occur following the attack, it may also temporarily occur in healthy individuals (5).

Case

A twenty-year old male patient presented at our neurology outpatient clinic with complaints of intermittent enlargement in his left iris for the last four years, explaining that this condition could last between 10 minutes and 1-2 days. It was found that the patient also

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had blurred vision with enlargement in the left iris. There was no headache, autonomous symptoms or epileptic attacks in his medical history. Physical examination revealed the diameters of his left and right pupils to be 4.5 mm and 2 mm, respectively, and anisocoria was observed to increase during examination under light (Figure 1). The patient's image after the disappearance of anisocoria can be seen in Figure 2. There was no migraine in family history. No pathology was identified in functions of presbyopia, vision acuity, colour vision or accomodation. Following a consultation by an ophthalmologist, iris deformity or glaucoma were not identified. Investigations and magnetic resonance imaging performed to examine intracranial, retroorbital and cervical pathologies that may cause anisocoria were normal. VEP and field of vision results performed to assess a potential optic nerve dysfunction were found to be normal. There was no clear change in anisocoria following pylocarpin 0.1% and 1% test. Bilateral median, ulnar and sural motor and sensory nerve conduction studies assessed for Adie's tonic pupil and autonomous testing results were normal.

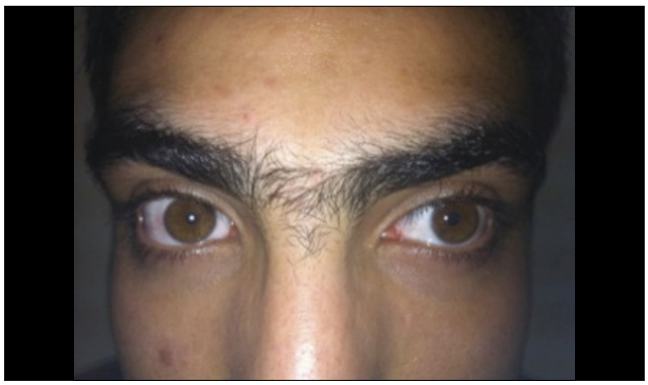

Figure 1. During a mydriasis episode: right pupil=2mm, left pupil=4.5mm

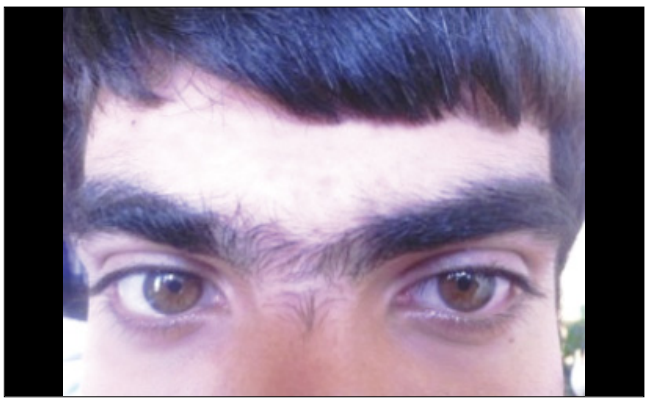

Figure 2. After mydriasis episode: right pupil=2.2 mm, left pupil=2.2 mm

Discussion

The causes of anisocoria may vary from physiologic anisocoria seen in 20% of normal individuals to life-threatening conditions requiring acute treatment including major stroke, aneurisms, infetions, III. cranial nerve paralysis, closed-angle glaucoma and trauma (6,7). Therefore, a detailed history, complete physical examination and use of imaging methods when indicated may sometimes be life-saving, because in cases with anisocoria treatment and prognosis depend on the underlying condition (8). Isolated benign episodic mydriasis, on the other hand, is a condition with a good neurological prognosis, that does not require further diagnostic imaging examinations.

To ensure that anisocoria is episodic, pupil function must be confirmed to be normal between attacks under light and in dark. In cases where pupillar function is normal, it must be remembered that physiologic anisocoria and benign episodic unilateral mydriasis may be the cause of transient anisocoria. There may be no clear etiologic pathology in 20% of anisocoria cases (2). In some episodes pupil dilatation may last several minutes to several days and the frequency of episodes may vary between several times a week and once in a few years (4).

Although the pathophysiology of benign episodic unilateral mydriasis is not exactly clarified yet, it is thought that the parasympathetic dysfunction or sympathetic hyperactivation of the iris may be the cause (9). It may commonly occur with migraine and some authors believe this may be a limited form of ophthalmoplegic migraine (4). This condition is thought to be associated with the demyelination due to the edema and inflammation caused by neuropeptides secreted in the Willis polygon by parasympathetic fibre dysfunction within the III. cranial nerve or by trigeminovascular system activation in the III. cranial nerve where it exits the brainstem (3,10,11). Benign episodic unilateral mydriasis seen rare in clinical practice and keeping this diagnosis in mind may avoid unnecessary interventional investigations (3). In our case, patient's interview did not reveal a history of headache or epileptic attacks, but the patient was planned to be closely monitored for headache and attacks for follow-up.

In conclusion, this case was found to be worth presenting to stress the importance of a rare clinical condition, benign episodic mydriasis as a diagnosis to keep in mind for the etiology of anisocoria.

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