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Ophthalmoplegic migraine with onset in adulthood – a case report

Migrena okoporaźna o początku w wieku dorosłym-analiza przypadku

Ewa Papuć, Anna Brzozowska-Jaśkiewicz, Zbigniew Stelmasiak, Konrad Rejdak

Chair and Department of Neurology of Medical University of Lublin

Abstract

Ophthalmoplegic migraine (OM) is a rare form of primary headache characterized by migraine attack accompanied by oculomotor nerve palsy, with typical disease onset in childhood. Nevertheless there are some reports on adult-onset of OM. The cranial nerve most commonly affected is oculomotor nerve (55%), but the palsy may affect different nerves in the same patient. The prevalence of the disease is less that 1 case in a million and in adults the diagnosis is exceptional as requires exclusion of other alternative causes of painful oculomotor nerve palsy such as vascular aneurysm, myasthenia gravis, oculomotor nerve schwannoma, pituitary apoplexy, Tolosa-Hunt syndrome, mucomycosis, diabetes mellitus or idiopathic cranial palsy. The pathophysiology of the disease remains unknown. The aim of this study was to describe an rare adulthood-onset case of OM.

Streszczenie

Migrena okoporaźna to rzadki rodzaj migrenowego bólu głowy, któremu towarzyszą objawy uszkodzenia nerwu okoruchowego, z typowym początkiem zachorowania w dzieciństwie. Istnieją jednak doniesienia o możliwym początku zachorowania w wieku dorosłym. Częstość występowania szacuje się na mniej niż 1/1000 000 osób w populacji. Diagnostyka migreny okoporaźnej u dorosłych wymaga wykluczenia innych alternatywnych przyczyn bolesnego uszkodzenia nerwu okoruchowego, takich jak: tętniak, miastenia gravis, nerwiak nerwu okoruchowego, udar przysadki mózgowej, zespół Tolosy-Hunta, mukomykoza czy idiopatyczne porażenie nerwu czaszkowego. Patofizjologia tej jednostki chorobowej pozostaje nadal nieznana. Celem pracy było przedstawienie przypadku migreny okoporaźnej z atypowym początkiem w wieku dorosłym.

Słowa kluczowe: ophthalmoplegic migraine, oculomotor nerve
Key words: migrena okoporaźna, nerw okoruchowy

Introduction

Ophthalmoplegic migraine (OM) is a rare form of primary headache. Typical characteristic of headache is identical to usual migraine without oculomotor nerve palsy. The prevalence of the disease is less than 1 in million population [1]. The disease has usually onset in childhood, but there are also some reports on beginning the disorder in adulthood [2,3]. The cranial nerve most commonly affected is oculomotor nerve (55%), but the palsy may affect different nerves in the same patient. In great majority of cases it is characterized by a single attack of ophthalmoplegia. In this article we describe a case of ophthalmoplegic migraine with onset in adulthood and discuss differential diagnosis of headaches with concomitant oculomotor nerve palsy.

Case description

A 49–year-old man was admitted to the Department of Neurology of Medical University of Lublin because of the third cranial nerve palsy which was preceded by one day lasting headache. The pain was hemicranial on the right supraorbital side, throbbing with moderate to severe intensity. The patient had a history recurrent attacks of headache with migrainous characteristic without aura since adolescence. The previous migraine attacks in our patient had typical manifestation with unilateral or frontal pulsating pain on the same side on which the 3rd nerve palsy later occurred. The patient have already had one episode of migraine headache followed by the third cranial nerve palsy. Family history of migraine attacks was negative in our patient. The 3rd nerve paresis lasted for about 4 days and was gradually resolving so that on the 4th day recovery was complete.

On admission to our department neurological examination revealed symptoms of third cranial nerve palsy. The dropping of eyelid on the right side, moderate anisocoria: dilated and sluggishly reactive right pupil, and diplopia were found.

There was no abnormality on neurological examination. Gait and stance were normal.

During the present hospitalization of the patient in the Neurological Clinic blood laboratory tests (hemogram, erythrocyte sedimentation rate, C-reactive protein, coagulogram, glucose level, renal and liver parameters, antophospholipid antibodies and antibodies anainst Borrelia burgdorferi-all these were within normal limits) as well as neuroimaging examinations have been carried out. The lumbar puncture was also performed, analysis of cerebrospinal fluid (CSF) revealed increased protein level. Neuroimaging of the head on brain contrast-enhanced computed tomography angiography and magnetic resonance imaging did not reveal any pathological vascular or focal findings. We performed brain MRI using 1.5 Tesla unit (Siemens vision). During the hospitalization the symptoms observed in the patient resolved gradually and on the 4th day the recovery was complete. The patient was not on any migraine prophylactic treatment before admission to hospital for low frequency of migraine attacks.

Discussion

Ophthalmoplegic migraine was firstly described by Gubler in 1860 according to Pierce [4]. As the disease is a rare disorder, the diagnosis of OM is exceptional because all alternative causes of painful oculomotor nerve palsy have to be excluded. It is worth noticing that most of oculomotor nerve palsy cases in the course of hemiplegic migraine are pediatric or in young adults [5]. The pathophysiology of the disease remains unknown. Some authors report the presence of contrast enhancement of the oculomotor nerve in the pericerebral region which is thought to be a special sign of this type of migraine [6]. Nevertheless, this symptom is not always present. In our patient headache was present before and during ophthalmoplegia, but in some patients it may be present only before the symptoms of the 3rd nerve palsy [7]. It is also worth mentioning that oculomotor nerve is the most commonly affected nerve during attacks of ophthalmoplegic migraine, however different nerves may be affected in the same patient subsequently during migraine attacks [Giraud]. Nevertheless, paresis of abducens nerve is rare [2]. The time between the presence of headache and the onset of nerve palsy may vary from hours to days [8,9]. The cranial nerve palsy and headache are usually reversible within weeks.

OM still remains a diagnosis of exclusion in clinical practice, even if it has some strict criteria proposed in the International Classification of Headache Disorder [7]. The criteria are based on clinical signs and require repetition of the attacks without abnormality in additional examinations. We performed biological blood tests which allowed us to rule out other disorders which could explain the described symptoms, such as diabetes mellitus, inflammatory process. Presently there is no consensus on the required tests for OM and the ICHD II does not require making these blood tests [7]. But it is necessary to rule out by appropriate examinations, parasellar tumor, orbital fissure or posterior fossa lesion (either by performing CT or brain MRI). Our patient fulfills the criteria for definite migraine according to the International Headache Society (IHS) from 2004 [7] as he had 2 episodes of migraine headache with oculomotor nerve palsy. Patients with a single attack of OM fulfilling the criteria of HIS for OM are considered as "probable OM".

It is important to highlight the difficulty of OM diagnosis even with the new IHS criteria because of rarity of having the full characteristics. Other disorders which can mimic OM are vascular aneurysm, myasthenia gravis, pituitary apoplexy, oculomotor nerve schwannoma. Differential diagnosis should also incorporate Tolosa-Hunt syndrome, in which both the headache and cranial nerve palsy occur simultaneously, but spontaneous good resolution without corticosteroid treatment rule out the diagnosis of this syndrome. Repetition of symptoms is also necessary for the diagnosis of OM, which was present in our patient. Other possible causes of 3rd nerve palsy is acute ocular mononeuropathy caused either by brain ischemia, viral infection, metabolic problems or idiopathic. Thus additional examinations are always required [5,6].

Conclusions

It is advisable to perform biological blood test and neuroimaging studies even though they are not required by the HIS, as they help the clinicians exclude alternative aetiology of headache with cranial nerve palsy and to make a proper diagnosis. Clinicians should also remember that late onset of OM (in adulthood) is also possible.

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Correspondence address

Ewa Papuć
Klinika Neurologii SPSK 4 w Lublinie
ul. Jaczewskiego 8
20-954 Lublin
Tel. (081)-7244-720, Fax: (081) 7244-540
e-mail: ewapap@yahoo.pl