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Recurrent painless ophthalmoplegic neuropathy with onset in advanced age – Case report

Deniz Selcuki, Ü. Serpil Sari, Ayşin Kisabay, Nihal Doğan, Bilge Oktan, Melike Batum *

Cello Bayar University, Department of Neurology, Manisa 45000, Turkey

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ABSTRACT

Recurrent painless ophthalmoplegic neuropathy is a clinical condition characterized by attacks of recurrent migraine headaches accompanied by ophthalmoplegia. It is a type of migraine in which single or multiple cranial nerves, most commonly the cranial nerve III, are involved within 4 days following headache. It is rare and more common in men. There are patients with onset in childhood but cases have rarely been reported beginning in the adulthood. The subject being reported here was considered to deserve to be presented because of attacks of recurrent painless ophthalmoplegic neuropathy were seen in advanced age.

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1. Background

Recurrent painless ophthalmoplegic neuropathy (RPON) is a clinical condition characterized usually by oculomotor nerve paralysis after recurrent migraine like headaches.[1] Most commonly, the third cranial nerve is involved but it is also likely that the fourth and sixth nerve may be involved.[1]

In 1860, Gubler reported that the attacks of migraine were associated with oculomotor paralysis.[2] The disease was first described as ``Ophthalmoplegic Migraine'' by Charcot in 1890.[3]

Definition of ophthalmoplegic migraine has been substituted by recurrent painless ophthalmoplegic neuropathy with ICDH-3 beta criteria

It is rare condition.[4,5] Its incidence has been reported as 0.7 per million.[2] It usually begins in the childhood.[6] The syndrome

* *Corresponding author*. Tel.: +90 02762240000.
E-mail addresses: dmselcuki@yahoo.com (D. Selcuki), dr.serpilsari@hotmail.com (an), blgdmrl@yahoo.com (B. Oktan), drmelikeyaman@hotmail.com (M. Batum).

Abbreviations: RPON, recurrent painless ophthalmoplegic neuropathy; VAS, visual analog scale; MRI, magnetic resonance imaging; NSAI, non-steroidal anti-inflammatory; PTZ–INR, partial thrombin time–International normalized ratio; aPTT, activated partial thrombin time; HbA1c, hemoglobin A1c; OGTt, otal glucose tolerance test; CRP, C-reactive protein; RF, rheumatoid factor; ANA, anti-nuclear antibody; anti-DNA, anti-deoxyribonucleic acid antibody; ASMA, anti-smooth muscle antibody; AMA, anti-mitochondrial antibody; ANCA, anti-nuclear cytoplasmic antibody; THS, Tolosa–Hunt syndrome; CSF, cerebro-spinal fluid; AVM, arterio-venous malformation; ACE, angiotensin converting enzyme; ENMG, electro-neuromyography.

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has been defined to be painless in children while it is painful in the adults.[6]

Subjects with onset in adulthood have rarely been reported in the literature.

2. Case report

A 66 years old man presented to Outpatient Clinic of Headaches-Algology of Neurology Department of Celal Bayar University with complaints of diplopia occurring after headache and ptosis of the right eyelid. The patient reported that he had hemicranial throbbing headache beginning over the last week around his right eye and being more prominent retro-orbitally and localized to right half of the head accompanied by nausea and photo- and phono-phobia with visual analog scale (VAS) score of 8–9 points on a 10-point VAS. Three days after the headache had decreased with analgesic (paracetamole) and anti-emetic drugs, ptosis in the right eyelid and diplopia complaints began. He reported that his complaints did not fluctuate in day.

Medical history of the patient revealed that he had diplopia and ptosis in his right eyelid following migraineous headache with retro-orbital localization in 1997 (at the age of 51). Cranial MRI (magnetic resonance imaging) taken in another center had been considered to be normal and the patient had been started corticosteroid treatment and full recovery had been achieved.

Medical history of the patient included attacks of migraine with or without aura occurring once or twice monthly for the last 30 years. He had no history of smoking, alcohol use, or trauma. He used to take paracetamole and/or non-steroidal anti-inflammatory (NSAI) drugs for migraine attacks.

Family history of the patient revealed migraine with aura in his mother.

The physical examination and vital function were unremarkable, the patient had ptosis in his right eyelid, upward and downward eye movements and those to inner-right side were restricted. There were direct and indirect light reflexes. There was no pupillary involvement (Fig. 1). The pupils were normoisocoric. Margins of the optic disk were normal in the fundoscopic examination. Visual acuity was bilaterally full (20/20) and visual field was within normal limits. Fatigue and ice tests were negative. Muscle strength was 5/5 in all limbs, flexion/extension of the neck was 5/5, deep tendon reflexes were normoactive quadrilaterally, and examination of the cerebellar and sensorial system was normal. There was no pathological reflex.

Liver function tests, renal function tests, the electrolytes, lipid profile, thyroid function tests, level of vitamin B12 and folic acid, complete blood counting, partial thrombin

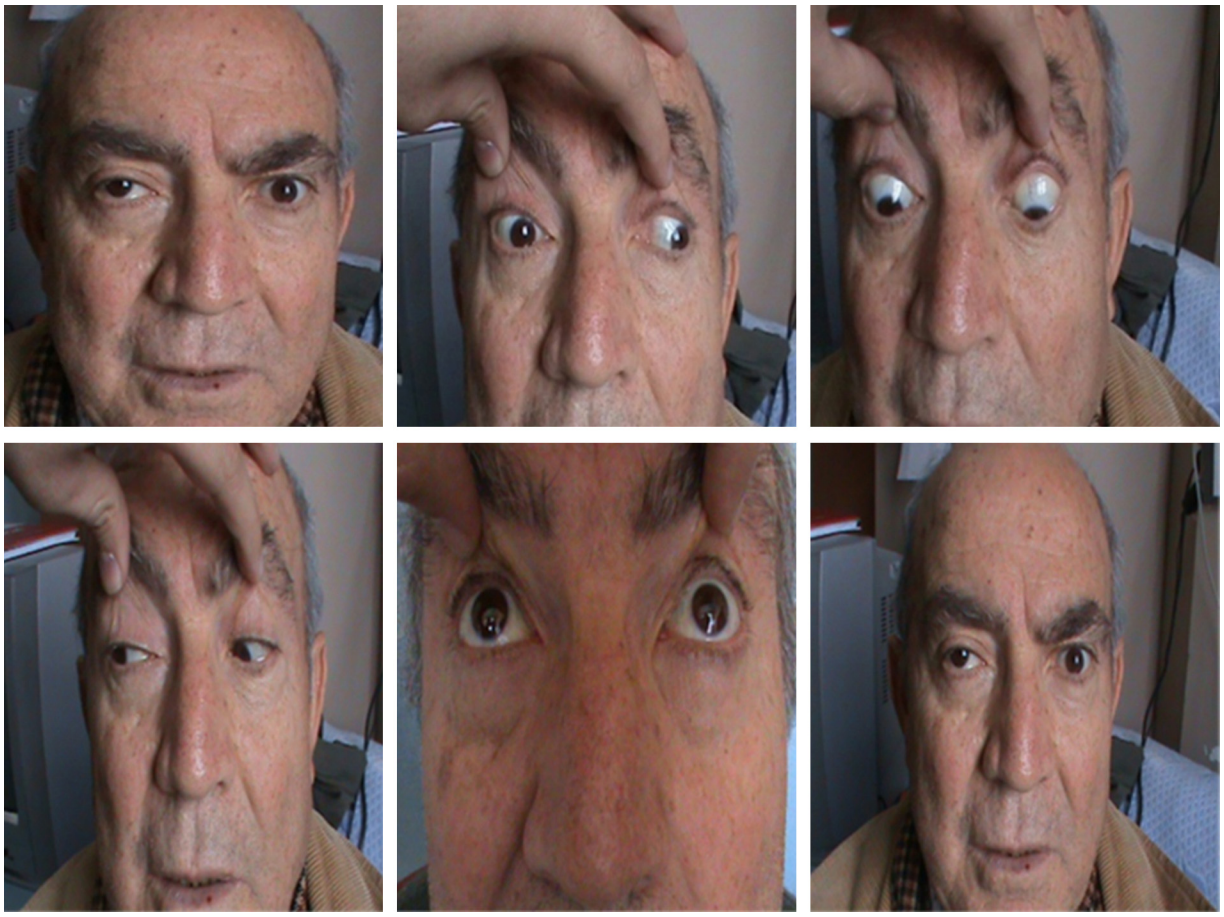

Fig. 1 – Neurological examination on admission. The patient had ptosis in his right eyelid, upward and downward eye movements and those to inner-right side were restricted.

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