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Migraine presenting as Left Abducens nerve Palsy: A case report

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Abstract

Recurrent Painful Ophthalmoplegic Neuropathy, previously known as Ophthalmoplegic Migraine, is a poorly characterized disorder mainly because there are few cases described. We report a new case of Recurrent Painful Ophthalmoplegic Neuropathy with left abducens nerve involvement. A diagnosis of possible Recurrent Painful Ophthalmoplegic Neuropathy was made after excluding other possible mimicking disorders. Patient treated with migraine treatment and improved significantly in 6 weeks, which is documented by photographs.

Keywords: Ophthalmoplegic, Neuropathy, migraine.

Introduction

Ophthalmoplegic migraine (OM), more recently renamed recurrent ophthalmoplegic neuropathy (RPON) by the International Headache Society, is an uncommon and poorly understood condition with an incidence of 0.7 per million.[1] This rare condition manifests as episodes of ipsilateral headache followed by palsy of 1 or more ocular cranial nerves[2] that begins immediately or up to 14 days after onset of headache.[3] Ophthalmoplegia may persist for weeks to months and is typically reversible.[3] The most commonly affected cranial nerve is the oculomotor nerve, followed by the abducens nerve. Trochlear nerve involvement is rare (3).[4]

The demographics of OM/RPON differ from that of migraine. While migraine affects women more often than men, and prevalence peaks in middle-aged adults (2),[5] OM/RPON is primarily a pediatric condition seen in children less than 10 years old and affects males more often than females.[4]

Case presentation

40-year female with no underlying comorbidity and significant past history presented to casualty with chief complaint of acute onset double vision on looking towards left from last 3 weeks. There were no symptoms of giddiness, neck stiffness, photophobia, facial droop, slurring of speech, problems with swallowing, limb weakness, numbness or difficulty with gait.

On further questioning, the patient admitted to having pain over the left side of her head for the past 1 month.

The headache was described as a tight sensation of severe intensity (7-8/10 on the visual analogue scale) with associated nausea, vomiting and phonophobia. She has had similar headaches for 5 years, occurring once or twice a month. Prior to onset of pain, she usually experiences prodromal symptoms of fatigue and nausea. The pain is usually located over the forehead bilaterally and is described as tight discomfort of moderate severity (5/10 on the visual analogue scale). There is associated nausea, vomiting and phonophobia. The pain usually lasts 12 hours interfering with her daily activities. The headaches are partially relieved by paracetamol. She could not identify any precipitating or exacerbating factors. She had similar episode of double vision last year which resolved spontaneously.

Physical examination

On examination left 6th cranial nerve palsy was present. Examination of the other cranial nerves was normal. Tone, power, reflexes and sensation of all limbs were normal as was the cerebellar examination. Meningeal signs were absent. Funduscopic examination was normal. Her gait including tandem gait was normal.

Investigation

All blood investigations including a fasting glucose, FT with electrolyte, LFT, ABG, HIV, HBsag, HCV, erythrocyte sedimentation rate, C-reactive protein, Autoimmune screen and serum ACE level were normal. Imaging study like NCCT HEAD, CEMRI Brain, CT Angiography with CECT base of skull was also normal (Fig. 1-3). Detail CSF examination was Normal.

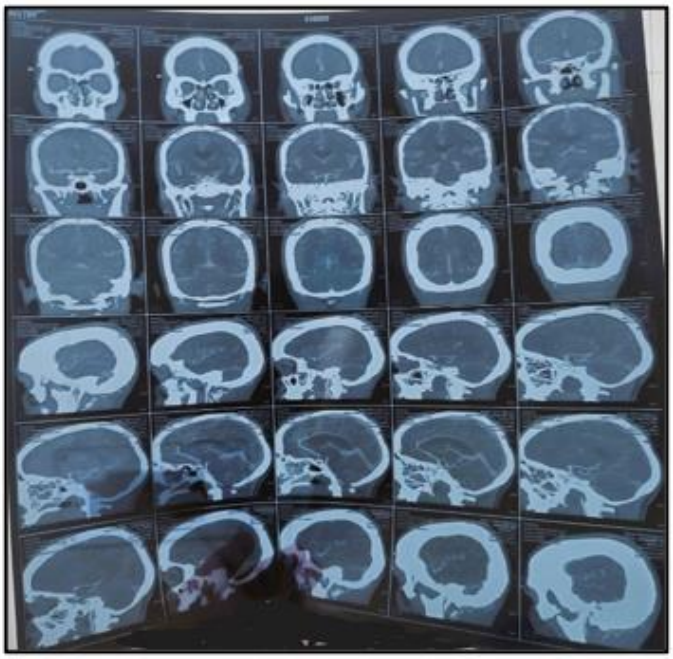

Fig.1: CECT base of skull

Fig.2: CT Angio brain

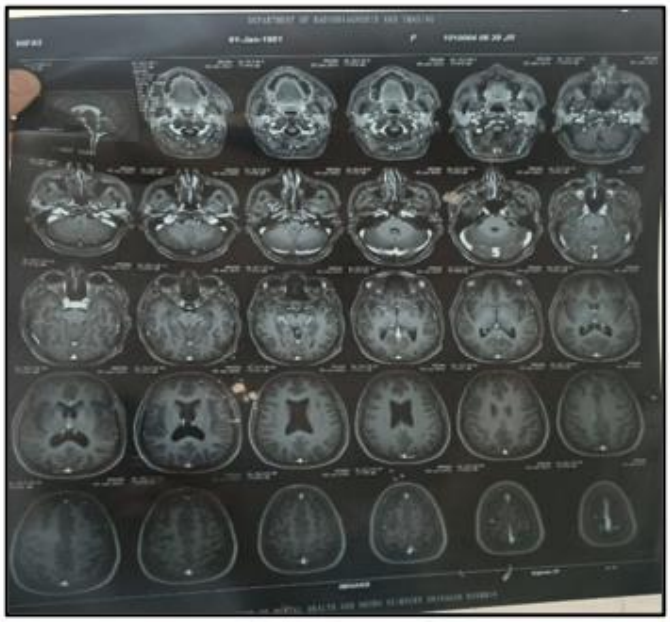

Fig.3: Cemri Brain

Differential diagnosis

In suspected RPON, the first diagnostic question to address, especially in adult-onset cases, is ruling out a mimicking lesion. From a review of the literature, it emerges that schwannomas are the most commonly implicated masses [6-10], followed by venous angiomas and hamartomas. Concerning the latter, Akimoto et al. proposed in them work that hamartomas may cause RPON through trigeminovascular-mediated vasodilation and subsequent nerve strangulation[11]. Being a rare entity, RPON can be easily mistaken for other, more common causes of painful ophthalmoplegia. Moving from neoplastic etiologies, disorders causing painful ophthalmoplegia constitute a vast group with different causes, comprising inflammatory, vascular, infectious, and traumatic ones, as highlighted in Gladstone's review[12]. Among the various disorders, Tolosa-Hunt syndrome (THS), in which granulomatous inflammation of the cavernous sinus leads to corticosteroid-responsive painful ophthalmoplegia, stands out as a mimicker of RPON[13-14] , especially in the rarest cases where MRI does not show typical features of THS. In summary, diagnosis of RPON requires exclusion of other more common disorders: this can be achieved by laboratory testing on blood and CSF, ruling out inflammatory and infectious diseases, and neuroimaging with MRI excluding lesional, vascular or traumatic causes.

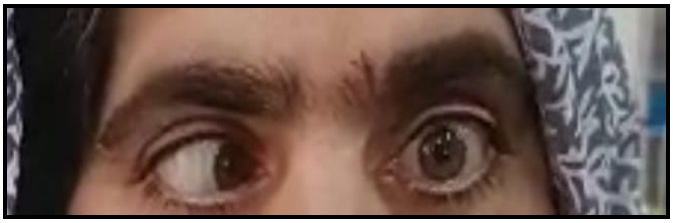

Fig. 4: AT Admission

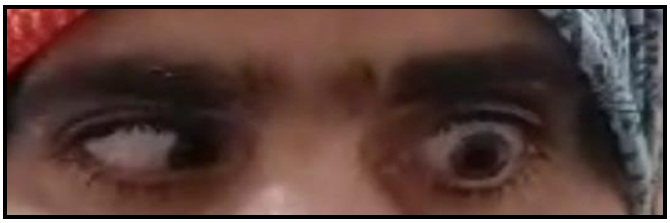

Fig. 5: 3 Weeks After Admission

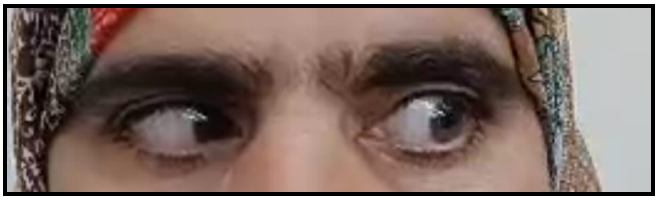

Fig. 6: 6 weeks after admission

Treatment And Outcome

Patient on discharge started on Amitriptyline 10mg, Divalproex 250mg and kept on OPD follow up. Within six weeks there is gradual and complete improvement in lateral rectus palsy which is documented by photographs (Fig. 4 -6).

Discussion

The RPON is a diagnosis of exclusion; no other etiology should better account for the findings [15]. Also, RPON is mainly a clinical diagnosis with imaging being an adjunct [15]. The RPON is a rare disorder occurring predominantly in children [16]. It is associated with recurrent headaches and ophthalmoplegia[16]. The pathophysiology of RPON is poorly understood [17]. The various hypotheses proposed include chronic inflammatory demyelination,

autoimmune process, reversible ischemic state of the nerve, simple nerve compression, and even oculomotor schwannoma with palsy due to the release of chemicals from the tumor causing inflammation, demyelination, and remyelination[17-18]. Pathological evaluation is usually limited due to the self-resolving, benign nature of the episodes. Hence, verification of these hypotheses is difficult[19].Imaging finding reported with RPON includes thickening and enhancement of one or more of the ocular cranial nerves, most commonly the oculomotor nerve.

As already outlined in several reviews[20-21], the current debate focuses on whether RPON should retain an association with migraine or be definitely considered as a neuropathy. While current evidence points more to a neuropathic mechanism, a relationship to migraine cannot be completely excluded in several patients [21], . In 2018, the third edition of ICHD changed the name of OM to RPON in order to reflect this view.

Diagnostic criteria (ICHD III)[15]

A. At least two attacks fulfilling criterion B

B. Both of the following:

1. Unilateral Headache

2. Ipsilateral paresis of one, two or all three-ocular motor nerves1

C. Orbital, parasellar or posterior fossa lesion has been excluded by appropriate investigation

D. Not better accounted for by another ICHD-3 diagnosis

Recurrent Painful Ophthalmoplegic Neuropathy is a poorly understood disorder, whose pathophysiology still remains elusive We provide a case report with atypical features (adult onset, female with abducen nerve palsy) in which diagnosis has been made by exclusion. More "atypical" cases might have been missed in the past.

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