▪ Headache Medicine ▪ ISSN 2178-7468 ▪ e-ISSN 2763-6178 ▪ v.11 ▪ Supplement ▪ p. 6 ▪ October 2020.
DOI: 10.48208/HeadacheMed.2020.Supplement.6

Headache Medicine

Case report: atypical recurrent painful ophthalmoplegic neuropathy

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Introduccion

Recurrent painful ophthalmoplegic neuropathy (RPON) is a rare disorder with repeated episodes of ocular cranial nerve neuropathy associated with ipsilateral headache in which secondary causes have first been excluded.

Case Report

Woman, 52 years old, no comorbidity. In 2001 she presented sudden onset intense (8-10) throbbing left-sided unilateral headache that irradiated to the ipsilateral eye. The crisis lasted for 04 days, without nausea, vomit, photophobia or phonophobia. The intensity of the pain was alleviated with 1g of dipyrone and sodium naproxen in one daily oral dose of 550 mg, without analgesics excess. About two days after the end of the crisis the patient noticed left-sided palpebral ptosis and vertical diplopy preceded by retro-orbital ipsilateral twinge pain. The neurological exam showed fixed mydriasis, left-sided hypotropia and exotropia, compatible with the compromising of the 3rd ipsilateral nerve CN III. At the occasion, the patient was submitted to laboratorial exams of the cerebrospinal fluid, CT scan of the orbit and the skull, digital cerebral angiography of the four vessels, with no abnormalities. Cranial MRI showed enhanced cisternal segment of the left side third nerve. In 2004, 2006, 2008 and 2010 the patient presented the same clinical manifestations, having been treated with 1mg/kg methylprednisolone with full relief in 15 days. However, on the last episode in june of 2019, the patient presented only left-sided oculomotor manifestation, showing residual vertical diplopy after 06 months of pulse therapy. At the moment, she is taking 5 mg of Prednisone, via oral, in protocol of weaning off and ambulatorial follow up at the specialized center of cephalalgy.

Comments

A case of RPON was described, diagnosed according to the ICHD-3. However, the patient presented unusual clinical aspects and age of symptoms onset. Unlike the descripitions found in literature, the first crisis occurred at the age 33. Besides, in the last event, only ocular manifestation ocurred, without cephalalgy, with persistent vertical diplopya, after pulse therapy and treatment with via oral steroids and 06 month- follow up. A case of RPON with atypical clinical manifestations and incomplete response to the treatment with steroids must be pointed out.

Keywords: Atypical Headache, Recurrent Ophthalmoplegic Neuropathy, Steroids