

Ophthalmoplegic migraine: report of two cases

Antônio Souza Andrade Filho,* Isolda Amado Nonato,** Paulo Maurício Menezes Nunes,*** Atalah José Haun Sobrinho,*** Camila Teles Machado Pereira,*** Nathanael de Freitas Pinheiro Júnior,*** Marise Silva Varjão****

* Department of Neuropsychiatry, Federal University of Bahia. President of the Foundation of Neurology and Neurosurgery. Brain Institute (FNN-BI).

** Department of Internal Medicine. Bahiana School of Medicine and Public Health.

*** Foundation of Neurology and Neurosurgery. Brain Institute (FNN-BI)/Students.

**** Foundation of Neurology and Neurosurgery. Brain Institute (FNN-BI)/Doctor.

ABSTRACT

Ophthalmoplegic migraine is a rare pathology, characterized by headache associated to paresis or palsy of the oculomotor nerves (oculomotor, trochlear and abducent). The diagnosis can be made after investigations of all demonstrated intracranial lesions. In this article, two cases of patients with headache and complete ophthalmoplegia are reported. The study of the cranium computed tomography, the digital cerebral angiography and the cerebrospinal liquid analysis were normal. Probable etiologies for this enfermity are discussed, since vascular compression of the *vasa nervorum* until dilation and hyperpulsation of the cranium's basis arteries. The differential diagnosis and the treatment proposed for this pathology are emphasized.

Key words: Migraine, ophthalmoplegic migraine, painful ophthalmoplegia.

INTRODUCTION

Ophthalmoplegic migraine (OM) is a rare pathology, characterized by headache associated to paresis or palsy of one or more of the cranial oculomotor nerves (oculomotor, trochlear and abducent).¹ According to the classification of the Brazilian Headache Society (BHS),² the diagnosis must be done only when all the demonstrated intracranial lesions are excluded. The differential diagnosis must be done with the carotid aneurysms, inflammatory, in-

Migraña oftalmopléjica: reporte de dos casos

RESUMEN

La migraña oftalmopléjica es una patología rara, caracterizada por dolor de cabeza y asociada a paresis o parálisis de los nervios oculomotores (oculomotor, troclear y abducente). El diagnóstico puede ser después de haber demostrado todas las investigaciones de lesiones intracraniales. En este artículo se reportan dos casos de pacientes con dolores de cabeza y oftalmoplejia completa. El estudio de tomografía computarizada de cráneo, la angiografía cerebral y el análisis del líquido cerebrospinal fueron normales. Se discute la probable etiología para estos enfermos desde la compresión vascular del vasa nervorum hasta la dilatación e hiperpulsación de las arterias de la base del cráneo. Se enfatiza el diagnóstico diferencial y el tratamiento propuesto para esta patología.

Palabras clave: Migraña, migraña oftalmopléjica, oftalmoplejia dolorosa.

feccious or expansive processes of the cranium's basis, neuritis, orbital periostitis, sphenoidal sinusitis, Tolosa-Hunt syndrome and Foix syndrome.^{3,4}

In this article, two cases of patients with headache and complete ophthalmoplegia are reported. The cranium computed tomography, the angiography and the cerebrospinal liquid analysis were normal in both cases. The importance of the differential diagnosis in these situations and the treatment proposed for these cases are emphasized.

CASES REPORT

Case 1

J.L.S., male, 51 years old, born in and proceeding from Maragogipe-Bahia, Brasil, watch-man,

Correspondencia:

Foundation of Neurology and Neurosurgery. Brain Institute of Bahia. Brazil. Rua Deocleciano Barreto, 10 Graça. Salvador, Bahia. Brazil.

E-mail: andradeneuro@uol.com.br

graduated on Elementary School, mulatto, married. He looked for the Neurology's Service of the Professor Edgard Santos University Hospital (Faculty of Medicine, Federal University of Bahia), complaining of intense headache and alteration in the left ocular movement.

The patient referred that he had a pulsating headache of moderate intensity in the left hemicranium, 23 days before the admission. He got better after taking common analgesic. Twenty-four hours later the pain became worse, and he presented bleary-eyed and progressive ophthalmoplegia that got completed in four days. Since then, the headache became intense and it didn't respond to common analgesics. There is no history of trauma, infection or lost of weight.

Positive risk factor for Chagas' disease and schistosomosis. There is no history of diabetes or systemic arterial hypertension. The patient's sister has got migraine with aura.

The patient denies being tabagist, etilist or making use of illicit drugs.

At the physical exam, he was in good general and nutritional conditions, with no alterations at the segmentar exam.



Figure 1.

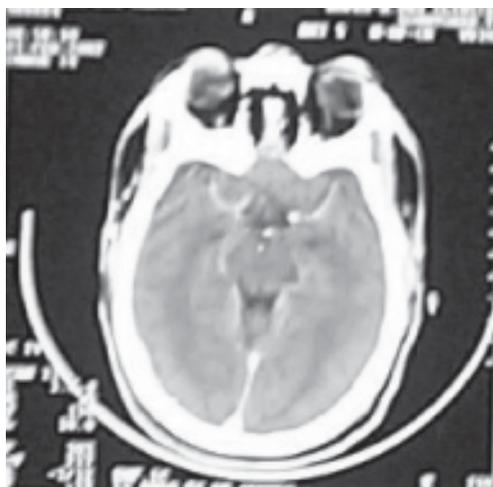


Figure 2.

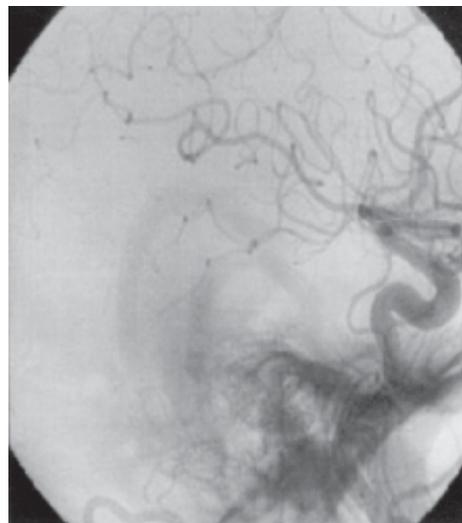


Figure 3.



Figure 4.

Arterial pressure: 120×80 mmHg, radial pulse: 80 bpm, respiratory frequency: 18 ipm.

Neurologic exam: left palpebral ptosis with complete ophthalmoplegia (Figure 1).

Laboratorial exams: hematocrit 44%; hemoglobin 12.3 g/dL; glycemia 86; Na 138; K 3.5; prothrombin time 100%; urea 30; creatinine 0.8.

Image exams (Cranium computed tomography with contrast, Figure 2; Digital cerebral angiography by subtraction, Figures 3 and 4) and the cerebrospinal liquid analysis were normal.

Case 2

A.S., female, 22 years old, born in and proceeding from Salvador-Bahia, Brasil, maid, graduated on Elementary School, mulatta, married. She looked for the

emergency service of the State's General Hospital (HGE) complaining of a right hemicranial headache, strong and pulsating, that started five weeks ago. The patient didn't get better after making use of common analgesics. The pain got worse and the patient evaluated with palpebral ptosis associated to right ocular movement alteration two weeks later. She denies other associated symptoms like nauseas, vomits or conscious level alteration. She reports previous history of headache since childhood. She denies history of diabetes or systemic arterial hypertension, and denies being tabagist, etilist or making use illicit drugs.

At the physical exam, the patient was in good general and nutritional conditions, with no alterations at the segmentar exam.

Arterial pressure: 120×80 mmHg; radial pulse: 85 bpm; respiratory frequency: 16 ipm.

Neurologic exam: right palpebral ptosis with complete ophthalmoplegia (Figure 5).

The laboratorial and image exams (cranium computed tomography and digital cerebral angiography by subtraction, Figures 6 and 7) were normal. It wasn't found any alteration in the cerebrospinal liquid analysis.

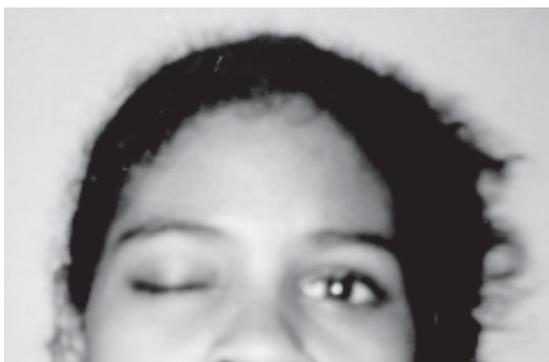


Figure 5.

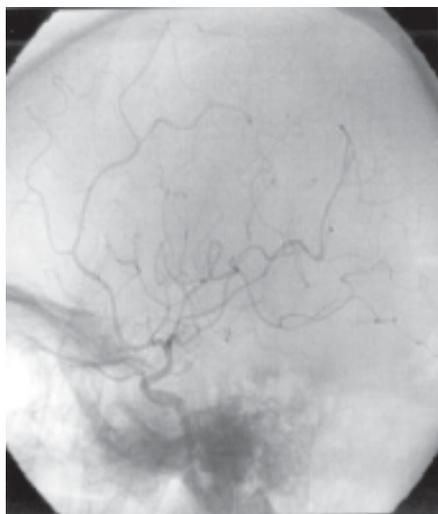


Figure 6.



Figure 7.

In the cases above, indomethacin and dexamethasone were used for eleven and ten days, respectively. Both patients developed with headache improvement. The patient 1 presented only discreet improvement of the ocular movement, while the patient two recovered completely. Once the patients were released from hospital, prednisone was prescribed with orientation to gradual reduction of the doses. Indomethacin was prescribed in case of headache. Ambulatory accompany is being provided to the patients. The first one is still getting ocular physiotherapeutic treatment.

DISCUSSION

In 1860, Gluber described a clinic syndrome characterized by hemicranial headache and palsy of the oculomotor nerves, which was evidenced to be a cranium basis' meningitis at necropsy. Moebius, in 1884, affirmed that this clinic feature was in fact a proper disease of either the oculomotor nucleus or their correspondent nerves, but not a migraine. He called it "periodic oculomotor palsy's syndrome". In 1890, Charcot was the first one to name this syndrome as ophthalmoplegic migraine.

The recorency of double vision with migraine, associated to sinais of paresis or palsy of the extraocular muscles has been named ophthalmoplegic migraine. The III cranial pair is the most commonly affected. The paresis often persists even after the migraine for days or weeks, and it can rarely become permanent after repeated episodes.⁵ The pain is always unilateral and often at the same side. The palsy of the eye's extrinsic muscles installs during the crisis.

In 1960, Walsh and O'Doherty⁶ reported distal straitment of the internal carotid artery compatible with edema of the vessel's wall, through arteriographic studies. From these findings, the authors suggested that either the direct vascular compression of the III, IV and VI cranial nerves inside the cavernous sinus or the ischemia due to the interference with the blood flux of the *vasa nervorum* were the main mechanisms involved in the OM's etiology.

Vijayan,⁷ in 1980, defends the possibility of ischemia as etiology, after finding partial or totally preserved pupillary reaction to light in two-thirds of the patients with migraine and palsy of the oculomotor nerve. In his paper, he affirms that in the ischemic neuropathies, like the diabetic, the pupillary reactions are often preserved. It's more common that the pupillary fibers that are in an external situation are firstly involved in the cases of direct compression. Besides that, the angiographies are commonly normal. Confirming that, Lance,⁵ in 1999, affirmed that the internal ophthalmoplegia (pupillary dilation without extra-ocular palsy) can be a characteristic of the basilar migraine.

According to Heyck⁸ (1965), the OM's etiology is probably involved with the pressure exercised by the dilation and hyperpulsation of the cranium basis' arteria (particularly the communicant and the posterior cerebral) on their adjacent nerves. There is still the hypothesis that an important edema in the ipsilateral cerebral hemisphere would be responsible for the herniation of the temporal lobe, compressing the oculomotor nerve.

The classification of the Brazilian Headache Society (BHS)² suggests the following diagnostic criteria:

- a) At least two crisis filling B.
- b) Headache accompanied with paresis of one or more of the cranial nerves III, IV and VI.
- c) Parasellar lesion is excluded by appropriated investigation.

In the differential diagnosis, Tolosa-Hunt syndrome and Foix syndrome must be considered. Tolosa (1954)⁹ reported a case of a patient with bilateral painful ophthalmoplegia and granulomatous periarteritis of the cavernous section of the internal carotid artery. Hunt et al.,¹⁰ in 1961, reported six cases of painful ophthalmoplegia with retrorbita-

rial pain. Foix syndrome or syndrome of the lateral wall of the cavernous sinus⁴ is, in fact, the thrombosis of this sinus. It is characterized by painful ophthalmoplegia, orbital and supraorbital pain, in association with palsy of the III, IV and VI cranial pairs. The ophthalmoplegia is often unilateral, and it's used to be assymmetric in the bilateral forms. The algic presentation is seen as the compromising of the ophthalmic section of the trigeminal nerve.

The BHS affirms that is unknown if the ophthalmoplegic migraine really is a migraine, since the headache lasts for one week or more. In most cases the complementary exams and the necropsy exclude the clinic diagnostics of OM. However, it can not be affirmed that there is no authentic cases. These two cases, for example, suggest the diagnosis of OM, since exams with good sensibility were realized and normal results were found. Therefore, other causes that could be leading to ophthalmoplegia were excluded, like carotid aneurysms, inflammatory, infeccios or expansive processes of the cranium's basis, neuritis, orbital periostitis, sphenoidal sinusitis, Tolosa-Hunt syndrome and Foix syndrome.

REFERENCES

1. Spierings LH. Management of migraine. Newton: Butterworth-Heinemann; 1996, p. 3-5
2. Comitê de Classificação das Cefaléias. International Headache Society. Classificação e Diagnósticos das Cefaléias, Neuralgias Cranianas e Dor Facial/Tradução de Carlos Alberto Bordini, et al.; coordenação Yara Dadalti Fragoso. São Paulo: PMK Consultoria e Marketing; 1999.
3. Farias-da-Silva W, Cruz IS, Costa J, Albuquerque H, Medeiros FL. Migrânea oftalmoplégica: revisão e apresentação de um caso. *Neurobiologia* 2001; 64: 39-40.
4. Foix CH. Syndrome de la paroi externe du sinus caverneux. A méloration considerable par lê traitement radiothérapique. *Ver Neurol (Paris)* 1922; 38: 827.
5. Lance JW, Goadsby PJ. Mechanism and Management of Headache. 6ª Ed. Oxford: Butterworth-Heinemann; 1999, p. 47
6. Walsh JP, O'Doherty DS. A possible explanation of the mechanism of ophthalmoplegic migraine. *Neurology* 1960; 10: 1079-84.
7. Vijayan N. Ophthalmoplegic migraine: ischaemic or compression neuropathy? *Headache* 1980; 20: 300-4.
8. Heyck H. Cefalalgias. Diagnóstico diferencial y tratamiento para el médico práctico. 1ª Ed. Barcelona: Salvat; 1965, p. 246
9. Tolosa E. Periarteritic lesions of the carotid siphon with clinical features of a carotid infraclinoidal aneurysm. *J Neurol Neurosurg Psychiatr* 1954; 17: 300.
10. Hunt WE, Meagher JA, Lefever HE et al. Painful ophthalmoplegia. Its relation to indolent inflammation of the cavernous sinus. *Neurology* 1961; 11: 56.

Recibido: Enero 18, 2006.
Aceptado: Marzo 20, 2006.