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Website: www.e-tjo.org
DOI: 10.4103/tjo.tjo_110_17

Oculomotor palsy as a single presenting sign of midbrain hemorrhage

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Abstract:

We report a case presenting with bilateral oculomotor nerve palsy (ONP) resulting from a midbrain hemorrhage. The patient visited the ophthalmological clinic due to the sudden onset of horizontal diplopia for a week. Bilateral ONP spared the left eye levator and bilateral pupils were found while the remaining results of examinations were unremarkable. Cerebral magnetic resonance imaging revealed a midbrain hemorrhage. Mono-ocular occlusion was arranged, and the patient was referred to a neurologist for further management. No further ophthalmic signs developed, but the patient became bedridden due to brainstem hemorrhage and died of aspiration pneumonia 9 months after the onset of the oculomotor signs. It is advocated that midbrain hemorrhage should be included in the differential diagnosis once diplopia develops and that careful neurological investigation of the origin of diplopia is warranted.

Keywords:

Brainstem hemorrhage, diplopia, midbrain hemorrhage, oculomotor nerve palsy

Introduction

The third cranial nerve controls various movements of the eyeball as well as the eyelid and sends parasympathetic fibers to the ciliary body and pupil.^[1] Due to the proximity, midbrain lesions are often associated with other ocular symptoms, such as ptosis, diplopia, hemiplegia, and conscious disturbance.^[1,2] On the other hand, oculomotor nerve palsy (ONP) is a common feature of intracranial hemorrhage or ischemic changes in the midbrain.^[1,3]

The chance of isolated ONP induced by intracranial hemorrhage is rare which occurs more often in diabetic neuropathy, aneurysm, or basal meningitis.^[4] Approximately 80 cases with midbrain hemorrhage presented with isolated oculomotor paresis accompanied by levator muscle dysfunction and pupillary sphincter impairment in most cases.^[3-5] Now, we report a 39-year-old

male patient presenting with bilateral pupil-sparing ONP resulting from midbrain hemorrhage.

Case Report

A 39-year-old ethnic Han Taiwanese male was healthy until he visited the ophthalmological clinic at Chang Gung Memorial Hospital due to the sudden onset of horizontal diplopia for a week and stated that the ocular symptom was more prominent in the early morning. On ocular examination, mild ptosis of the right eye with exotropia of 25 prism diopters was detected using Krimsky method, and limitation of eye movements in all directions was identified by diplopia test. Importantly, adduction, supraduction, and infraduction of both eyes were found to be impaired [Figure 1]. Moreover, the left eye levator and bilateral pupils were unaffected. The remaining results of his ocular and physical examinations were unremarkable. The complete blood counts,

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How to cite this article: Lee CY, Chen HC, Lin HY, Kao LY. Oculomotor palsy as a single presenting sign of midbrain hemorrhage. Taiwan J Ophthalmol 2018;8:108-10.

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Submission: 29-10-2017
Accepted: 16-01-2018

prothrombin time, activated partial thromboplastin time, blood chemistry profile, including serum vitamin level, and thyroid function test were all within normal limits. Although an initial diagnosis of myasthenia gravis was suspected, cerebral magnetic resonance imaging was arranged 3 weeks after the onset of diplopia, and the result revealed a midbrain hemorrhage [circles, Figure 2]. Mono-ocular occlusion was arranged with improved symptoms, and the patient was referred to a neurologist for further management where edrophonium test showed negative result. Two months after the onset of diplopia, sudden onset dizziness accompanied by nuchal pain as well as bilateral leg muscle weakness were noted, and then, cerebral magnetic resonance angiography [circle, Figure 3] revealed a hemorrhage over the brainstem that was associated with mass effect and hydrocephalus, but no vascular anomaly was noted [Figure 3]. Then, the patient became bedridden, and the follow-up neurological examination identified headache insomnia, dysarthria, dysphagia, and decreased muscle power (Grade 4 at upper limbs and Grade 2 at lower limbs). Nine months after the onset of diplopia, the patient exhibited poor appetite, yellowish sputum, and general weakness after choking before being admitted to our internal medicine ward under the suspicion of aspiration pneumonia. The patient died as a result of aspiration pneumonia-induced respiratory failure within the same month.

Discussion

ONP generally leads to adduction impairment, vertical gaze palsy, ptosis, and mydriasis in the affected eye.^[5] Cases of nuclear third palsy, which is usually accompanied by unilateral or bilateral third nerve palsy, bilateral ptosis, and bilateral mydriasis,^[6] and ONP caused by midbrain hemorrhage have been reported elsewhere, but nearly all of those cases presented with pupillary sphincter impairment.^[3-5,7,8] Our patient presented with bilateral ONP but without bilateral eyelid and pupil involvement, which is a very rare presentation in the context of midbrain hemorrhage that only one patient was reported previously.^[8] To our knowledge, this is a rare case of ONP with diplopia sparing the pupil but without significant risk factors of intracranial hemorrhage. The pupillary sphincter is innervated by the Edinger–Westphal nucleus, which is the central nucleus controlling the levator palpebral superioris, and the other extraocular muscles are innervated by the central, medial, and lateral subnuclei. The presenting signs of our patient suggest that the lesion site was probably located in the lower part of midbrain and did not involve the Edinger–Westphal nuclei.

Various disorders may cause ONP, and in our patient, the midbrain hemorrhage was confirmed by magnetic



Figure 1: Ocular examination indicating bilateral oculomotor nerve palsy

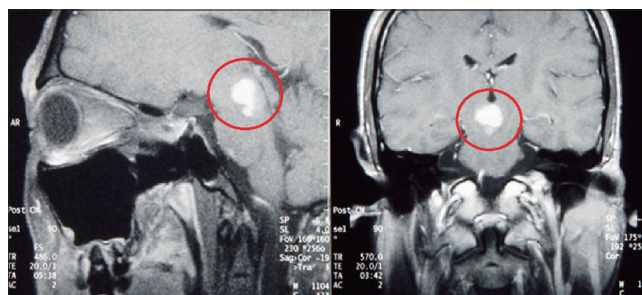


Figure 2: Magnetic resonance imaging indicating the midbrain hemorrhage

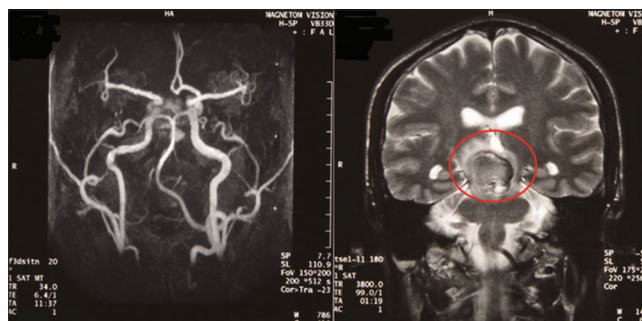


Figure 3: Magnetic resonance angiography indicating the brainstem hemorrhage with mass effect and hydrocephalus

resonance imaging, and the laboratory examinations also excluded the possibility of Wernicke's encephalopathy. In general, the most common causes of midbrain hemorrhage are vascular malformation, hypertension, and bleeding diathesis.^[5,7] Considering the absence of vascular anomaly according to magnetic resonance angiography and the normal blood pressure value as well as the laboratory data, a spontaneous midbrain hemorrhage has the highest probability (30%) in our patient.^[5,7]

Spontaneous brainstem hemorrhage accounts for 5%–10% of intracerebral hemorrhage, and secondary brainstem hemorrhage develops from cerebral hemorrhage with a frequency of 45%.^[9,10] Thus far, no study has evaluated the incidence of brainstem hemorrhage occurring after midbrain hemorrhage.

Nonetheless, all intracerebral hemorrhages share similar risk factors, such as hypertension, aneurysm, and vascular malformation, and the prognosis of brainstem hemorrhage is generally poor.^[10] Although our patient had a relatively stable period, he eventually died from aspiration pneumonia. The patient's bedridden status resulting from the brainstem hemorrhage may be the cause of death.

Treatment options for diplopia include strabismus surgery, ocular occlusion, monovision optical correction, prism glasses, and chemodenervation by botulinum toxin.^[2] Surgical interventions are usually deferred for a minimum of 6 months after the onset of symptoms, and prisms have a limited role in the treatment of deviations caused by ONP.^[2] As a consequence, we chose mono-ocular occlusion as the initial treatment. Since the mono-ocular occlusion improved the diplopia in the following months, we did not perform strabismus surgery.

Conclusion

We report a rare case of midbrain hemorrhage presenting with pupil-sparing ONP without predisposing factors of intracranial hemorrhage. Accordingly, we suggest that midbrain hemorrhage should be included in the differential diagnosis once diplopia develops and that careful neurological investigation of the origin of diplopia is warranted.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgment

This study was supported by the Chang Gung Memorial Hospital (CMRPD1C0251-3 and CMRPG3G0031-3)

and the Ministry of Science and Technology (MOST 106-2314-B-182A-042-).

Ethics approval and consent to participate

All the procedures and managements performed in the current report that involved a human patient adhered to the Declaration of Helsinki (1964). An informed consent form was signed by the patient's next of kin who agreed to enroll in this report after an explanation of the report was provided. A copy of the written consent form is available for review by the editor of this journal.

Financial support and sponsorship

Nil.

Conflicts of interest

The authors declare that there are no conflicts of interests of this paper.

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