



Wall-eyed bilateral internuclear ophthalmoplegia syndrome from a brainstem haemorrhage

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Wall-eyed bilateral internuclear ophthalmoplegia (WEBINO) syndrome is characterized by bilateral internuclear ophthalmoplegia (INO) with alternating exotropia. The syndrome is a rare variant of INO, and was first reported in 1974 in a patient with vascular lesion.^{1,2} WEBINO has many distinct clinical findings, with the most common being primary gaze exotropia, adduction deficit, nystagmus of abducting eye, and vertical gaze-evoked nystagmus.³ And whilst the pathogenesis of WEBINO remains uncertain, the involvement of bilateral medial longitudinal fasciculus (MLF) is

known to play an important role.⁴ WEBINO is most often caused by a demyelinating process or ischemic stroke. Less common causes include trauma, brainstem tumour, and hydrocephalus.⁵ Additionally, there are several case reports of WEBINO caused by vasculitis in patients with systemic lupus erythematosus (SLE).⁶ Here, we report a patient with brainstem haemorrhage (pontine bleeding with spread into the mesencephalon) that led to WEBINO syndrome. A 55-year-old woman with history of uncontrolled hypertension (HTN) was referred into our hospital with loss of consciousness.

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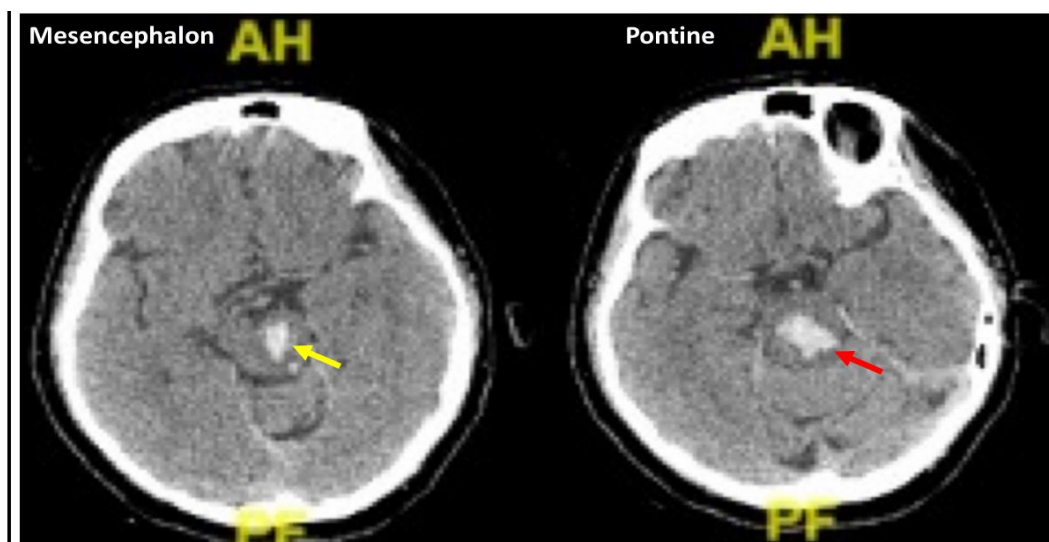


Figure 1. Non-contrast head computed tomography (CT) performed at stroke onset demonstrates a hyperdense lesion extending from the pontine region (right image, red arrow) to the mesencephalon (left image, yellow arrow), consistent with acute intracerebral haemorrhage

The patient initially experienced sudden headache during activity, followed by projectile vomiting, cramping, and right hemiparesis. On presentation, her blood pressure was 183/102 mmHg and her Glasgow Coma Scale (GCS) score was E3M4V2. The computed tomography (CT) scan of the brain on presentation showed hyperdense lesion in the brainstem (pontine-mesencephalon bleeding) with a volume of ± 9.3 cc (Figure 1). The patient received immediate neuro-intensive care to control hematoma expansion and prevent secondary brain injury. During treatment in our centre, she experienced multiple complications including pneumonia, sepsis, stress hyperglycaemia, acute

renal failure, and disseminated intravascular coagulopathy, which required intensive care at our centre. After 57 days of treatment, the patient was discharged with a modified Rankin scale (mRS) score of 5 (severe disability).

At the two-year follow-up, the patient had many sequels (WEBINO, dysarthria, dysphagia, right hemiparesis, left hemiataxia, right hemihypesthesia, and positive pathological signs such as Babinski and Clonus). Pupillary reflexes were intact bilaterally, and there were no visual field defects. On primary gaze position, the patient showed bilateral exotropia with right-sided dominance (Figure 2).

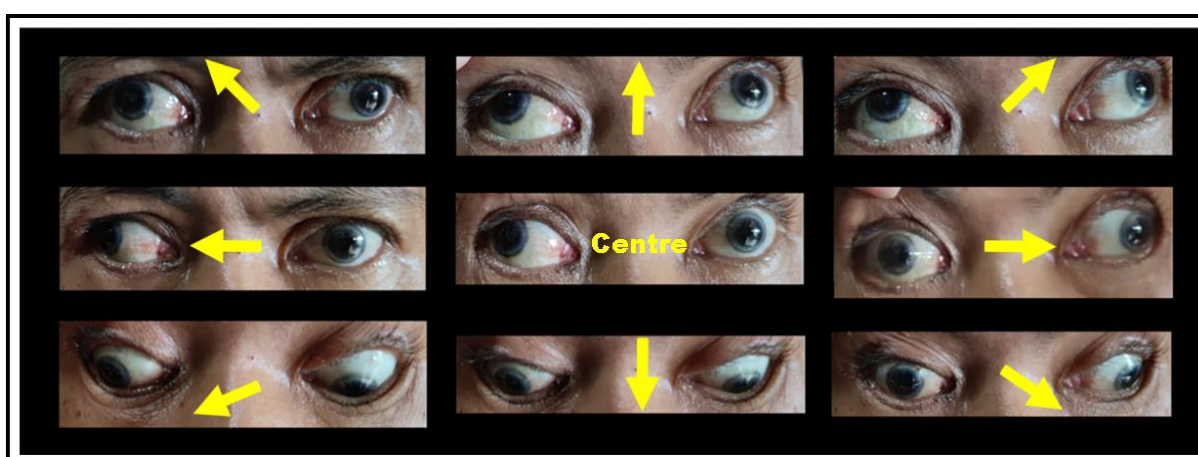


Figure 2. Neuro-ophthalmologic examination revealed primary gaze exotropia accompanied by bilateral adduction deficits with right-sided dominance, consistent with the diagnostic features of wall-eyed bilateral internuclear ophthalmoplegia (WEBINO) syndrome. These ocular motor abnormalities persisted chronically and remained evident on follow-up examination two years after the initial brainstem hypertensive haemorrhage, indicating a long-term deficit likely due to irreversible damage to the medial longitudinal fasciculi (MLF)

There was ocular bobbing. We also observed convergence, saccadic, and smooth pursuit disturbances. Additionally, during abduction in lateral gaze, the contralateral eye failed to adduct. The most recent mRS score at follow-up was 4 (category: moderate-severe disability). Additional imaging examinations were also performed at the 2-year follow-up; the CT-scan showed complete absorption of hematoma in the brain stem, and cerebral angiography revealed no vascular malformations or other structural abnormalities.

In the WEBINO syndrome, exotropia is the main presenting sign of the ophthalmoplegia, and can be caused by many conditions that lead to brainstem lesions, particularly of the pontine tegmentum, pontine-mesencephalon, or mesencephalon areas.^{1,7} Many lesions involved in bilateral MLF dysfunction are ischemic, autoimmune [multiple sclerosis (MS)], inflammatory, infectious, toxic, metabolic, traumatic, and nutritional disorders in nature.⁸ In our patient, the WEBINO syndrome was due to a very uncommon cause, which was haemorrhagic stroke in the brainstem area.

Primary brainstem haemorrhage itself is a challenging clinical entity with a high mortality rate.⁹ In this instance, the management of our patient required multiple intensive care unit (ICU) admissions due to complications such as sepsis, renal failure, and coagulopathy. Due to the complexity of management and high mortality, it is challenging to assess what potential ophthalmic complications of brainstem haemorrhage may be. This case report adds to the currently limited literature on WEBINO as a sequela of brainstem haemorrhage. INO is caused by a lesion in the MLF. It affects conjugate horizontal gaze and causes impaired adduction ipsilateral to the lesion

and abduction nystagmus contralateral to the lesion.¹⁰ When the lesion involves the MLF bilaterally, it is then termed as WEBINO. There are several distinct clinical signs of WEBINO, such as exotropia in primary gaze, impaired adduction, dissociated nystagmus in the abducting eye, vertical gaze-evoked nystagmus, and impaired vertical vestibulo-ocular reflex (VOR).⁸ Several hypotheses have elucidated that the primary gaze exotropia is due to lesions in the MLF bilaterally as well as in the subnuclei of the medial rectus.¹¹ WEBINO should be distinguished from one-and-a-half syndrome, which shows ipsilateral conjugated horizontal gaze palsy (one) and ipsilateral INO (a-half), and is a result of lesions at the dorsal tegmental pontine that involve ipsilateral paramedian pontine reticular formation (PPRF) (or the abducens nucleus) and the ipsilateral MLF.^{12,13} On clinical exam, one eye is fixated in the midline while the other shows horizontal abduction.¹⁴ However, in this case, since both eyes moved laterally, we exclude one-and-a-half syndrome as a differential.

In conclusion, we believe that our rare case patient will provide us with greater insight and knowledge of brainstem haemorrhage as a rare cause of WEBINO syndrome, which is a rare variant of INO with the most common aetiology being cerebral ischemia or MS.

Conflict of Interests

The authors declare no conflict of interest in this study.

Acknowledgments

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