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CASE REPORTS

Isolated and Transient Nuclear Midbrain Blepharoptosis in a Young and Healthy Adult

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ABSTRACT

A 32-year-old otherwise healthy man presented with acute-onset bilateral blepharoptosis of 6 days' duration. On examination, he had severe ptosis bilaterally and mildly restricted abduction in the left eye. Brain magnetic resonance imaging showed a 10-mm-diameter lesion in the dorsal midbrain. The ptosis resolved spontaneously within two weeks. Systemic investigation did not uncover any aetiological factor. During 70 months' follow-up, neither any systemic disease nor ptosis relapse developed. Isolated nuclear midbrain ptosis has been previously reported in a few patients and these had neoplastic or inflammatory causes. In this patient, spontaneous resolution of the nuclear ptosis within weeks suggested that the underlying cause might be isolated ischaemic damage to the central caudal nucleus.

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Nuclear blepharoptosis; mesencephalon; transient ptosis; midbrain lesion

Introduction

Isolated and transient nuclear midbrain blepharoptosis is very rare and is usually associated with nonvascular lesions.[1] We describe herein a young adult patient with isolated and transient nuclear ptosis that was presumably due to ischaemic damage, and the relevant literature is reviewed.

Case report

A 32-year-old man presented with bilateral ptosis of six days' duration. Ptosis occurred after somnolence and weakness lasting two days. The patient had no history of any systemic disorder, upper respiratory infection, trauma, use of medical or recreational drugs or familial ptosis. Inspection of the photograph on his identification card revealed no previous ptosis.

On examination, palpebral fissure heights were 2 mm OD and 1 mm OS. Levator excursions were 3 mm and 2 mm, respectively (Figure 1a). Except for mildly restricted abduction in the left eye, his extraocular eye movements were normal and the eyes were orthotropic. The pupils were isochoric and constricted to both light and near stimuli. The visual acuities were 20/20 OU. The other ocular findings were normal. Magnetic resonance imaging (MRI) demonstrated a lesion located predominantly on the left side of the dorsal caudal midbrain, which was 10 × 10 mm in diameter, isomildly hypointense on T1-weighted images and hyperintense on T2-weighted and fluid-attenuated inversion recovery images. The lesion showed contrast enhancement (Figure 2). Infectious diseases, immunology and haematology consultations, detailed blood tests and haemodynamic studies did not reveal any abnormalities.

Three days after presentation, the blepharoptosis started to improve spontaneously. On the 14th day, the ptosis resolved completely on the right side and near completely on the left side. During a follow-up period of 70 months, neither relapse of the ptosis nor any serious systemic disease occurred (Figure 1b).

Discussion

In the midbrain, nuclear or fascicular damage to the oculomotor nerve can cause bilateral or unilateral blepharoptosis, respectively. The subnucleus of levator muscle (central caudal nucleus) is in the caudal midbrain and is undivided. Other

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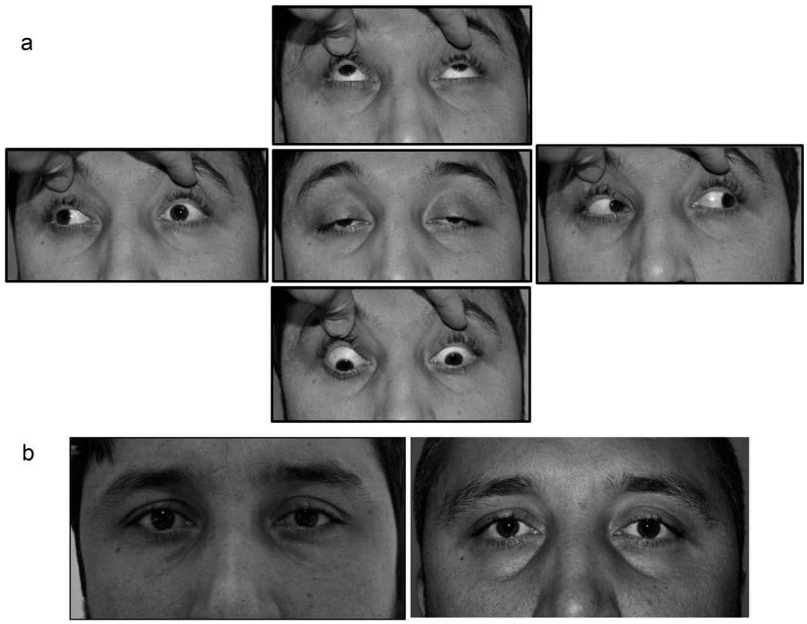

Figure 1. **a)** Patient with acute bilateral ptosis. Extraocular movements were normal, except for mildly restricted abduction in the left eye. **b)** Patient's photographs at 2 weeks (bottom, left) and 70 months after the first presentation (bottom, right). The ptosis resolved spontaneously.

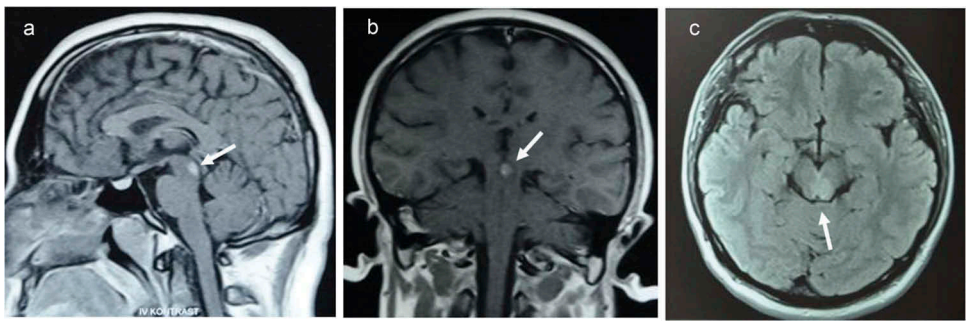

Figure 2. Magnetic resonance imaging shows a 10 mm diameter lesion located predominantly on the left side of the dorsal midbrain (white arrows), that prominently enhanced on contrast-enhanced T1-weighted images (sagittal, a and coronal, b) and was hyperintense on the axial fluid-attenuated inversion recovery image (c).

oculomotor subnuclei (including the pupillary sphincter subnucleus) are paired and located laterally and innervate muscles ipsilaterally (except the contralaterally innervated superior rectus and superior oblique muscles). The subnuclei are so small and at such close proximity that their isolated involvement is extremely rare. The somatic lateral column of the oculomotor nerve nucleus is, on average, 2.4 × 1.2 mm in diameter on transverse section. The average central caudal nucleus is only about 0.8 × 0.6 mm in diameter.[2]

Vascular infarcts and nonvascular abnormalities in the midbrain can cause blepharoptosis. Possibly the most common cause is large cerebral infarcts, also involving the midbrain. Isolated pure midbrain infarcts are rare and frequently cause a fascicular (unilateral) ptosis, accompanied by neurologic deficits. We found five patient series in the English literature, including a total of 101 patients with pure midbrain infarcts. Among these, 17 (17%) had unilateral ptosis and eight (8%) had bilateral ptosis.[3–7] Six of the eight patients with bilateral ptosis, whose ages were reported, were between 50 and 88 years of age (mean age, 69.5 years). In the largest series in which the neuro-ophthalmological deficits were described in detail in 40 patients with pure midbrain infarct, 21 (52.5%) had oculomotor involvement and 12 had ptosis (10 unilateral, 2 bilateral).[3] Two patients with

bilateral ptosis were 65 and 50 years of age, and the infarct mechanism was small-vessel disease in one patient and unknown in the other. In these two patients, neurological abnormalities accompanied the ptosis, including superior rectus paralysis, mydriasis, dysarthria, sensorial symptoms, gait ataxia, facial paralysis and clumsiness in movements. In a recent study by Ogawa et al., 10 (91%) out of 11 patients with midbrain infarct-induced oculomotor nerve palsy had unilateral ptosis and one patient (9%) had bilateral ptosis.[8] Except for the patient's age (88 years), no other information about the patient with bilateral blepharoptosis was reported in that study. Typically, ptosis and other oculomotor palsies related to midbrain ischaemic infarct resolve spontaneously within days or weeks.

Isolated bilateral midbrain blepharoptosis has been reported in a few cases. The nuclear damage had structural or inflammatory pathologic mechanisms in these cases and included metastatic breast cancer, lymphoma, glioma, haematoma, tuberculosis and neurocysticercosis[1,9–13] To our knowledge, isolated and transient nuclear midbrain ptosis in a healthy young adult has not been previously reported. Martin et al. described bilateral isolated midbrain ptosis in a 41-year-old woman, which quickly resolved in two days, as in the case presented herein.[10] However, the MRI studies did not show any midbrain lesion in this patient, despite having definite multiple sclerosis.

In about 38% to 89% of patients with midbrain infarct, the possible aetiopathogenesis is classified as small-vessel disease or idiopathic.[3–7] In these patients, basilar or vertebral artery hypoplasia could cause a predisposition to midbrain infarction.[7] In the current case, spontaneous resolution of ptosis and MRI findings suggests that the origin could be a transient vascular occlusion associated with small-vessel disease. The small size of the lesion and absence of an embolic source or another predisposing factor supports this assumption.[7] This study may also support the finding that the central caudal nucleus has a special arterial supply in some individuals.[2] Symptoms like somnolence, as seen in the current patient, before the outset of ptosis, may be an initial symptom of midbrain infarction and likely caused by impaired function in the reticular activating system.[14] In the differential diagnosis, vertebrobasilar artery stroke syndromes may have been considered; however, this case was easily distinguishable since the sensorimotor findings were extremely limited, consistent with the specific location of the lesion.

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