American Journal of Ophthalmology Case Reports 32 (2023) 101947

Contents lists available at ScienceDirect

American Journal of Ophthalmology Case Reports

journal homepage: www.ajocasereports.com/

A case of bilateral horizontal gaze palsy and concurrent esotropia

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ARTICLE INFO

Keywords:
Bilateral horizontal gaze palsy
Gaze palsy
Esotropia
Cross fixation
Strabismus

ABSTRACT

Purpose: To report a unique case of bilateral horizontal pontine gaze palsy with concurrent esotropia, surgical management, and post-operative follow-up.
Observations: A 39-year-old male presented with diplopia and a history of neurocysticercosis. He was found to have bilateral horizontal gaze palsy and concurrent esotropia, R > L. Classic bimedial rectus recess-resect surgery was done to include resection of the right lateral rectus muscle. Follow-up three months post-op demonstrates markedly improved diplopia.
Conclusion and importance: We present a recommended therapeutic approach for the rare case of concurrent bilateral horizontal gaze palsy and esotropia, which should be further evaluated in longitudinal studies.

1. Introduction

Gaze palsies are defined by limitations in the conjugate movements of the eyes in a given direction. Horizontal movement may be limited by lesions in the supranuclear, nuclear, or internuclear regions. The dorsal pons houses nuclei for the abducens and facial cranial nerves, the paramedian pontine reticular formation (PPRF), and the medial longitudinal fasciculus (MLF). The PPRF projects to the ipsilateral abducens nucleus, which gives rise to the abducens nerve, providing the sole innervation to the lateral rectus muscle. Through rostral projections to the contralateral MLF, the PPRF also gives rise to interneurons that synapse at the contralateral oculomotor nucleus, which provides innervation of the contralateral medial rectus muscle.[1,2] Notably, a lesion in the abducens nucleus, not just of the abducens nerve, better describes the horizontal pontine gaze palsy.

A unilateral horizontal gaze palsy is defined by the inability to move both eyes to a given direction past the midline. This may result from lesions that can be localized to the contralateral FEF, ipsilateral PPRF or ipsilateral abducens nucleus.[3] To accommodate a horizontal gaze palsy, patients often adopt a compensatory head posture. In a unilateral palsy, this posturing demonstrates a gaze preference contralateral to the PPRF lesion.[4] Furthermore, patients with horizontal pontine gaze palsies can present with esotropia in the eye contralateral to the site of the lesion. This can be attributed to continued innervation of the contralateral medial rectus muscle from other regions of the brain.[2]

To the best of the authors' knowledge, there have been only reports of bilateral horizontal pontine gaze palsy with ipsilateral esotropia.[2] Here we report a unique case of bilateral horizontal pontine gaze palsy with concurrent esotropia with cross-fixation, surgical management, and post-operative follow-up. The difficulty of this case lies in the differentiation of a gaze palsy versus an abducens nerve palsy, or both. Treatment differs significantly depending on the etiology of the esotropia.

2. Case report

A 39-year-old male presented to the eye clinic with a one-year history of double vision. He describes a horizontal misalignment of images in his field of vision. His ocular history is otherwise unremarkable.

The patient's medical history is notable for neurocysticercosis, which was likely contracted during his time spent as an exotic meat taster in South America. Magnetic resonance imaging from November of 2020 revealed the development of cysts along the fourth ventricular outflow tract and a significant amount of edema involving the dorsal pons and periventricular cerebellar hemispheres (Fig. 1). Taken together, the calcified granulomas and obstructive hydrocephalus are consistent with neurocysticercosis. The man underwent resection of cysts in the cerebellar vermis in 2021.

On clinical examination in June of 2022, visual acuity was found to be 20/40 OD and 20/60 OS without correction and 20/40 OU. Pupils were equally reactive and showed no signs of afferent pupillary defects. Visual field testing was grossly full. Slit-lamp examination showed left-

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https://doi.org/10.1016/j.ajoc.2023.101947
Received 29 May 2023; Received in revised form 12 October 2023; Accepted 18 October 2023
Available online 26 October 2023

sided lagophthalmos, and bilateral trace nuclear sclerosis. Fundoscopic examination was unremarkable for hemorrhage or other structural abnormalities.

On sensorimotor examination without correction, a large angle esotropia was observed bilaterally, with a more pronounced angle in the right eye. A bilateral complete horizontal gaze palsy was observed concomitantly. This was reflected in the absence of the vestibulo-ocular and oculocephalic (Doll's eye) reflexes. The patient maintained cross fixation posturing for the duration of the visit, with extreme right face turning. Diplopia was relieved with a 45–50 prism diopter (PD) offset, but was no better with 60 PD. A fine upbeat jerk nystagmus was also observed, but optokinetic drum testing was non-informative.

A review of a December 2020 MRI revealed no significant findings pertaining to the bilateral lateral rectus muscles.

In June 22, 2022 the patient underwent a 5.0 mm recession of bilateral medial rectus muscles and a 5.0 mm resection of the right lateral rectus, both using adjustable sutures. At this point, forced duction testing revealed 3+ tightness of the right medial rectus muscle, and 2+ tightness of the left medial rectus muscle.

One month following the procedure, the patient was found to have orthophoria, with an equal and centered corneal reflex on Krimsky testing. However, he remained diplopic and required a 20 PD offset Fresnel prism. During post-operative month 2, examination revealed only mild, intermittent horizontal diplopia when viewing letters 20/50 and smaller. The offset was reduced to 10 PD, and use was limited to when viewing distant objects. By month 3, the offset was reduced to 5 PD and only used intermittently, by month 5 it was reduced to 3 PD, and by month 6 he no longer required the use of a prism. Throughout all follow-ups, motility was unchanged.

3. Discussion

In patients presenting with horizontal gaze palsy and concurrent esotropia the goal of surgical intervention is to regain binocular single vision and relieve abnormal head posturing. To this end, a survey of the literature highlights two studies of surgical intervention in cases of pontine gaze palsy and concurrent esotropia. Surgical intervention in patients with complete unilateral horizontal gaze palsies secondary to dorsal pontine lesions with concurrent unilateral esotropia resulting in abnormal head posturing is described by Somer et al.[5] Briefly, the authors demonstrated that application of an asymmetric surgical dose of standard recession-resection protocols to both eyes, favoring the esotropic eye, improved head posture and bilateral eye motility. Notably, this protocol produced more favorable outcomes than vertical rectus transposition, which is the favored surgical treatment in cases of complete loss of lateral rectus muscle function. Improved post-operative motility thus suggests "paresis, not paralysis" of the musculature. In a study of patients with horizontal gaze palsy with ipsilateral esotropia, Coats et al.[2] highlight the importance of an asymmetric recession-resection procedure. The asymmetric recess-resect procedure will help augment surgery to sufficiently cover for larger angles in asymmetric cases.

Clinically, the horizontal gaze palsies may masquerade as damage to the sixth cranial nerve.[2]

Unilateral palsies of the abducens nerve are among the most common cranial nerve palsies, and often arise from neoplasia, trauma, or microvascular damage,[6] but have also been reported in cases of neurocysticercal cyst development in the posterior fossa.[7] Acutely, patients present with horizontal binocular diplopia and ipsilateral eye abduction deficits, preventing the eye from moving past the midline.[8,9] In patients with only mild paresis of the abducens nerve, refractive errors are observed more frequently than diplopia, and can be acutely alleviated by occluding one eye, through the use of prism lenses,[10] or by injecting botulinum toxin into the medial rectus muscle.[11] If the nerve paresis persists for 6–12 months a surgical recession of the ipsilateral medial rectus muscle, and resection of the affected lateral rectus muscle (recess-resect) is indicated.[10,11]

While less common, bilateral abducens palsies have been described, often in the context of increased intercranial pressure, brainstem infarction,[12] and in a case of transventricular migration of a neurocysticercal cyst.[13] Patients with bilateral abducens palsies present with bilateral abduction deficits,[12] in which testing of forced duction may be required to differentiate between sixth nerve palsy and paresis, with the former demonstrating poor force generation.[11] Complete abducens nerve palsies require a transposition procedure, such as the Jensen procedure, where the superior and inferior rectus muscles are partially joined to the lateral rectus muscle.[11,14]

In the presented case, the patient's dramatically improved diplopia, lasting at least six months after a recess-resect procedure implies that this was a true horizontal gaze palsy. Furthermore, the incremental

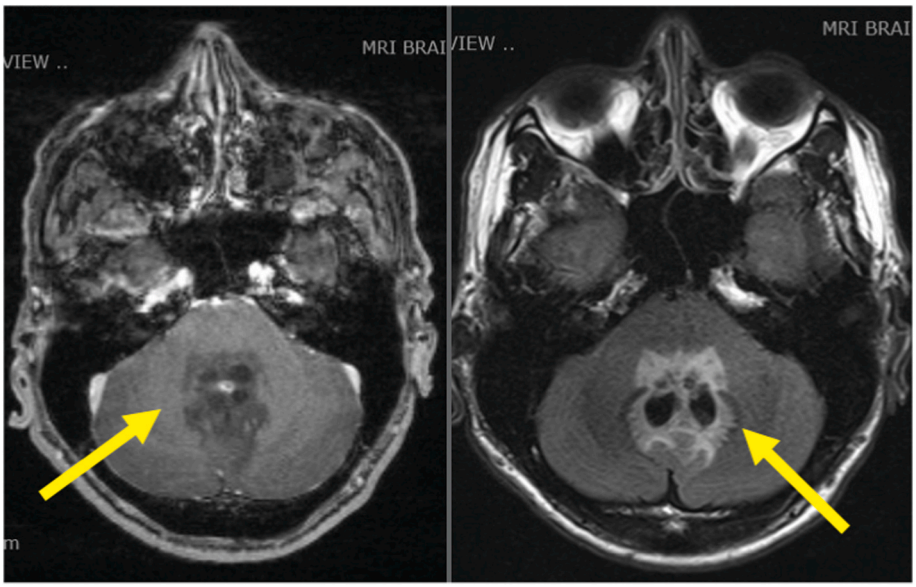

Fig. 1. Axial MRI of the brain showing subcentimeter peripherally enhancing cystic focus along the fourth ventricular outflow tract with substantial amount of surrounding edema involving the dorsal pons and periventricular cerebellar hemispheres. Reducing/involuting enhancement and disproportionate edema as well as concomitant obstruction of the fourth ventricular outflow system.

improvement post-operatively indicates that mild tone persisted in the lateral rectus muscles. The distinction between an abducens nerve palsy and a horizontal gaze palsy may not be clear prior to attempting treatment. In such cases, the authors suggest treating the patient under the assumption of a horizontal gaze palsy, as a specific etiology is not necessary to direct treatment. If a true horizontal gaze palsy is misdiagnosed and treated with a transposition procedure, as indicated for an abducens nerve palsy, there is a low likelihood that the desired result would be achieved due to the lack of ipsilateral lateral rectus tone. We would recommend a recess-resect procedure over a transposition in a horizontal gaze palsy, especially since a transposition procedure could serve to increase the risk of anterior segment ischemia in future operations,[15] and increase the risk of inducing a vertical strabismus.[6] Conversely, if a true abducens nerve palsy is treated with a standard recession-resection, as indicated for a horizontal gaze palsy, it is unlikely to cause undue harm. However, a transposition procedure would likely be indicated at a future timepoint. Importantly, surgical intervention for strabismus carries the risk of anterior segment ischemia and should be considered depending on patient risk factors.[16]

4. Conclusion

In situations of an inconclusive diagnosis of either horizontal gaze palsy or an abducens nerve palsy pre-operatively, risk to the patient can be reduced by first completing a recession-resection procedure. In cases of concomitant esotropia, an asymmetric dosing of this operation is recommended.

Funding

No funding or grant support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

None.

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