CASE REPORT

Abducens Nerve Palsy Following Expansion Cranioplasty with Distraction Osteogenesis

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ABSTRACT

Childhood abducens nerve palsy can occur as a result of trauma, tumour, vasculopathic disease, elevated intracranial pressure, infection, inflammation, and congenital or idiopathic causes. The authors present two cases of unilateral abduction deficit secondary to a recent trans-sutural distraction osteogenesis (TSuDO) operation for craniosynostosis. After distractor removal, the problem resolved spontaneously over 2–4 months in both cases. This is a first reported case of sixth nerve palsy as a complication of TSuDO operation.

Keywords: Abducens nerve palsy, distraction osteogenesis, expansion cranioplasty

INTRODUCTION

The various causes of acquired abducens (sixth) nerve palsy in childhood include trauma, tumour, elevated intracranial pressure (ICP), and viral infection.^{1–3} Rarely, it can occur as a result of surgery not related to tumour.¹ We report two cases of abducens nerve palsy associated with expansion cranioplasty and discuss possible mechanisms for this complication.

CASE REPORTS

Case 1

A 5-year-old boy was referred to the ophthalmic service with an inability to abduct his left eye associated with left postauricular pain. The problem was first noted 2 days before the consultation and has been preceded by expansion cranioplasty 3 weeks earlier with subsequent distraction osteogenesis. He had no prior history of ocular complaints or problems.

He underwent expansion cranioplasty for the surgical correction of his sagittal and lambdoid craniosynostosis. A burr hole was drilled at anterior

and posterior fontanelles, middle of the sagittal suture, and middle of the right lambdoid suture. This was followed by trans-sutural craniectomy of the sagittal and bi-lambdoid sutures. Three distractors for sagittal distraction and one distractor for lambdoid distraction were placed (Figure 1A). Subsequently, the distractors were activated 0.3–2.5 mm/day for 16 days.

On initial ocular examination, his visual acuity was 20/20 in each eye. Intraocular pressure was 11 and 10 mm Hg in the right and left eyes, respectively. There was a left esotropia of 15 prism dioptres (PD) at distance and near with decreased ability of abduction, just slightly passing the midline (3 minus). He also had a face turn to the left. Pupil showed normal size with prompt response to light stimuli in both eyes. Ophthalmoscopy revealed normal disc and fundus, and retinoscopy revealed a refractive error of emmetropia and –0.25 D sphere in the right and left eyes, respectively. There was no significant finding in serologic test, and a computed tomographic (CT) scan of the brain did not show any evidence of intracranial haemorrhage, ischaemic damage, and fluid collection. There were no neurological signs indicating involvement of other cranial nerves.

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FIGURE 1 (A) 3D reconstructed CT images before (upper row) and during distraction osteogenesis (lower row). CT images in lower row show three distractors for sagittal distraction and one distractor for lambdoid distraction. (B) A 5-year-old boy at the onset of left abducens nerve palsy with an abduction deficit of the left eye (4 minus). (C) Diagnostic gaze positions 2 months after the onset. Full range of versions in the both eyes is noted.

The findings were consistent with a diagnosis of isolated sixth nerve palsy. Two days after the initial ophthalmic examination, the patient developed perioral erythematous grouped vesicles, herpes simplex labialis was diagnosed and topical acyclovir prescribed. There was no clinical evidence of herpes simplex encephalitis, and no abnormality was found on otolaryngological examination. Subsequent evaluation after 5 days demonstrated a progression in the patient's limitation in ability of abduction. There was left esotropia of 35 PD in the primary position with inability to abduct past midline (4 minus; Figure 1B). It was presumed that a nerve stretch injury had occurred and accordingly activation of distractors was stopped. Plates, screws, and distractors used in the distraction and fixation were all removed. During the neurosurgical procedure, he underwent lumbar puncture with an opening pressure of 15.0 cm H₂O, with normal protein and glucose levels, and no cells. Four days later, ocular motility demonstrated much improvement. There was left esotropia of 12 PD at distance and orthophoria at near with ability to abduct (2 minus), but an abnormal face turn to the left persisted. Two months after the initial presentation, his ocular motility was completely recovered with orthophoria in all directions and his abnormal head posture also resolved (Figure 1C). At 1-year follow-up, he had developed neither new symptoms nor recurrence.

Case 2

A 4-year-old boy was referred with an abrupt inability to abduct his right eye during the distraction. He underwent expansion cranioplasty for genetically confirmed Crouzon syndrome 3 weeks prior to his eye problem (mutation (c.314A→G) in the *FGFR2* gene). Five distraction devices were applied: three across the osteotomised sagittal suture, one across the coronal suture, and one across the lambdoid suture. Subsequently, distraction was initiated 2 weeks prior to the presentation (Figure 2A). Apart from mild inferior oblique overaction in the right eye, the remainder of his preoperative ophthalmic examination was normal.

On ophthalmic examination, his visual acuity and intraocular pressure was 20/40 and 14 mm Hg in each eye, respectively. There was a right esotropia of 8 PD in the primary position, increasing to 30 PD on right gaze with decreased ability of abduction (2 minus; Figure 2B). He also had a face turn to the right. Pupil showed normal size with prompt response to light stimuli, and ophthalmoscopy revealed normal disc and fundus in both eyes. Laboratory data including viral titres showed normal result. A CT scan of the brain did not show any evidence of other possible etiology of sixth nerve palsy, and there were no other

neurological signs. With suspicion of a nerve stretch injury, distractors were deactivated immediately and all devices were removed 2 weeks after the initial presentation. Prior to the removal procedure, he underwent lumbar puncture with an opening pressure of 11.5 cm H₂O. On the day after surgery, he showed almost normal abduction in the right eye and a right esotropia of 12 PD. Four months after the initial presentation, ocular motility was completely resolved with orthophoria. At 10-month follow-up, he had no eye problem despite a high opening pressure of 32 cm H₂O in lumbar puncture.

DISCUSSION

Childhood abducens nerve palsy can occur due to neurosurgery not related to tumour.¹ However, this complication has been infrequently reported and there are no documented reports of such a complication following distraction osteogenesis for craniosynostosis.

Distraction osteogenesis is the surgical induction of bone growth by applying distractive force across an osteotomy site.⁴ This technique for surgical correction of craniosynostosis was introduced by Barone and colleagues in the mid-1990s⁵ and is among several recently introduced techniques for managing craniosynostosis. The trans-sutural distraction osteogenesis (TSuDO) method is a new distraction technique with simple suturectomy and vector-controllable distraction, which is applicable for all types of craniosynostosis. The basic principle of the TSuDO method is dissection and distraction of the prematurely fused sutures.⁶ Complications that have been reported are focal wound infection, discharge, device dislocation or distortion, device exposure, bone fracture, shaft slippage, and distractor breakage or malfunction.⁶⁻⁸ Although abducens nerve palsy after maxillary osteotomy and distraction osteogenesis have previously been described,⁹ to our knowledge, this is the first report of abducens nerve palsy associated with the TSuDO method.

The pathophysiological mechanism resulting in cranial nerve injury during distraction osteogenesis is not well understood. Possible explanations include increased ICP, localised ischaemic damage secondary to the procedure, compressive effect caused by a space-occupying lesion such as intracranial haemorrhage, neurapraxic injury, and nerve stretch caused by anatomic rearrangement of Dorello's canal associated with widening of the subdural space during cranial expansion. In our cases, ICP was within the normal range and the concurrent herpes viral infection in the first case appeared to be unrelated to the acquired abducens palsy. Because there were no localised ischaemic or space-occupying lesions on CT scan, and no neurological signs of herpes viral central

FIGURE 2 (A) 3D reconstructed CT images before (upper row) and during distraction osteogenesis (lower row). CT images in lower row show three distractors for sagittal distraction, one distractor for coronal distraction, and one distractor for lambdoid distraction. (B) A 4-year-old boy at the onset of right abducens nerve palsy with slight abduction deficit of the right eye (2 minus).

nervous system (CNS) infection, we suspect a neurapraxic or stretch injury may have been the cause. Neurapraxia is characterised by a focal conduction block at the site of the lesion, with normal neuronal conduction distally.¹⁰ It may occur during distraction osteogenesis due to stretching or kinking of the abducens nerve, or an anatomical change in its

position, and resolve rapidly once the distraction force is removed.^{10,11} In previous studies, Dorello's canal has been suggested as a potential site of abducens nerve injury, because the abducens nerve is most vulnerable to kinking as it passes over the bony ridge of the petrosphenoidal junction.¹¹ Injury could occur at dural entry or exit points or at the petrous ridge

within Dorello's canal.¹¹ A compressive effect, possibly caused by a sudden surge of cerebral spinal fluid into rearranged Dorello's canal, may also impair the function of the abducens nerve.¹²

Our cases may represent a combination of events following distraction osteogenesis producing shearing and/or stretching force of the abducens nerve, which was reversible after removal of the distraction hardware.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Note: Figures 1 and 2 of this article appear in colour online at <http://informahealthcare.com/oph>.

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