be useful for inferior field visual function activities such as literacy and feeding.

Tracking of the fundus also identified areas of retinal function amenable to structural study by OCT. In our patient the PRL presented with defined neurosensory retinal anatomy. It is also noteworthy that the PRL does not always develop on the border of the lesion in other diseases,[8] probably because of subclinical disease. In our patient, the retinal thickness per se might not have been the decisive factor for the definition of the PRL; rather, the presence of an anatomically healthy choroid and partial preservation of anatomic features of the retina may have been the critical factors. This has been previously described in children after intensive treatment of a macular tumor.[10]

The cooperation needed for the examination was essentially the ability to sit comfortably at the machine and to press the buzzer to indicate appearance of the stimuli. Our patient was 9 years of age, within the age range recommended for conventional perimetry in children without cognitive impairment.[11] Applied to children with only one remaining eye, microperimetry combined with SS-OCT testing offers clinicians a new approach in understanding the adaptive mechanisms after macular damage and may have a role in future visual rehabilitative treatments in older children with early loss of central vision by retinoblastoma.

References

1. Kivelä T. The epidemiological challenge of the most frequent eye cancer: retinoblastoma, an issue of birth and death. Br J Ophthalmol 2009;93:1129-31.
2. Stacey AW, Clarke B, Moraitis C, et al. The incidence of binocular visual impairment and blindness in children with bilateral retinoblastoma. Ocul Oncol Pathol 2019;5:1-7.
3. Acton JH, Greenstein VC. Fundus-driven perimetry (microperimetry) compared to conventional static automated perimetry: similarities, differences, and clinical applications. Can J Ophthalmol 2013;48:358-63.
4. Reinhard J, Messias A, Dietz K, et al. Quantifying fixation in patients with Stargardt disease. Vision Res 2007;47:2076-85.
5. Vingolo EM, Salvatore S, Cavarretta S. Low-vision rehabilitation by means of MP-1 biofeedback examination in patients with different macular diseases: a pilot study. Appl Psychophysiol Biofeedback 2009;34:127-33.
6. Watts P, Westall C, Colpa L, et al. Visual results in children treated for macular retinoblastoma. Eye (Lond) 2002;16:75-80.
7. Narang S, Mashayekhi A, Rudich D, Shields CL. Predictors of long-term visual outcome after chemoreduction for management of intraocular retinoblastoma. Clin Exp Ophthalmol 2012;40:736-42.
8. Greenstein VC, Santos RA, Tsang SH, Smith RT, Barile GR, Seiple W. Preferred retinal locus in macular disease: characteristics and clinical implications. Retina 2008;28:1234-40.
9. Crossland MD, Culham LE, Kabanarou SA, Rubin GS. Preferred retinal locus development in patients with macular disease. Ophthalmology 2005;112:1579-85.
10. Samara WA, Pointdujour-Lim R, Say EA, Shields CL. Foveal microanatomy documented by SD-OCT following treatment of advanced retinoblastoma. J AAPOS 2015;19:368-72.
11. Patel DE, Cumberland PM, Walters BC, Russell-Eggitt I, Rahi JS, group Os. Study of Optimal Perimetric Testing in Children (OPTIC): feasibility, reliability and repeatability of perimetry in children. PLoS One 2015;10:e0130895.

Long-term follow-up of torsional augmentation surgery in a case of congenital ocular tilt reaction with head tilt

Henry Liu, HBSc,[a] and J. Raymond Buncic, MD, FRCSC[b,c]

Torsional augmentation surgery was used to correct the anomalous head position (AHP) in a child with congenital ocular tilt reaction (OTR). The underlying neuropathology was hypoplasia involving the right hemicerebellum and contralateral brainstem. Postoperatively there was an acceptable and variable resolution of head tilt sustained over a 25-year follow-up period. These findings suggest that early torsional augmentation surgery can effectively correct stable OTR head tilt in congenital cases over the long term.

Ocular tilt reaction (OTR) is a pathologic syndrome characterized by an abnormal synkinesis of skew deviation (supranuclear hypertropia), associated head tilt, and primary position paradoxical ocular torsion.[1,2] OTR is typically a result of asymmetric disruption to the utriculo-ocular reflex in response to injury of the vestibular system in the periphery, brainstem tegmentum, or cerebellum.[3,4] This imbalance is associated with tilt in the ipsilateral direction of the perceived subjective visual vertical (SVV) with respect to the earth's graviceptive vertical plane.[5] Brodsky[6] reported horizontal transposition of the vertical rectus muscles to augment the ocular torsion in the direction of the pathological head tilt (torsional augmentation) resolving the anomalous head position (AHP) and associated lateropulsion in a patient with acquired ocular head tilt in a case of partial OTR (near absent 1^{Δ} skew). We describe the long-term result in a case of congenital OTR–associated severe head tilt (associated with upbeating nystagmus and ataxia) that was successfully treated with torsional augmentation surgery.

Author affiliations: [a]Faculty of Medicine, University of Ottawa, Ottawa, Ontario, Canada; [b]Department of Ophthalmology and Vision Sciences, University of Toronto, Toronto, Ontario, Canada; [c]Ophthalmology and Vision Sciences, Hospital for Sick Children, Toronto, Ontario, Canada

The study did not have external funding except incidental personal funding by the senior author Buncic.

Submitted July 14, 2018.

Revision accepted November 11, 2018.

Published online November 26, 2018.

Correspondence: J. Raymond Buncic, MD, FRCSC, Professor, University of Toronto, Staff Ophthalmologist, Hospital for Sick Children, 555 University Avenue, Toronto, ON, M5G 1X8 (email: ray.buncic@sickkids.ca).

J AAPOS 2019;23:117-119.

1091-8531/$36.00

https://doi.org/10.1016/j.jaapos.2018.11.002

Case Report

A 4-year-old girl with a history of OTR with upbeating nystagmus and ataxia from cerebellar hypoplasia was followed-up at the Neuro-ophthalmology service at the Hospital for Sick Children, Toronto. She had a normal birth history. Developmental milestones and muscle tone were reduced. Severe esotropia was noted at infancy, along with severe right head tilt (approximately 45°). The esotropia responded partially to two horizontal strabismus surgeries (medial rectus recession and lateral rectus resection in each eye, performed sequentially).

The child was unable to sit until 1 year of age and started walking independently at age 5, after extensive physiotherapy. Coordination and balance were poor, and her overall limb movements were ataxic. The rest of her neurological and cranial nerve examinations were unremarkable. X-rays showed no cervical spine abnormalities. A T2-weighted magnetic resonance imaging (MRI) of the brain demonstrated a small vestigial right cerebellar hemisphere with associated hypoplasia of the right posterior fossa. The contralateral brainstem appeared reduced in size, particularly the upper pons and mesencephalon.

Augmentation torsion surgery was performed (JRB) in 1993, when the patient was 6 years of age to correct the AHP by rotating both eyes ipsilateral to the direction of head tilt. The right superior rectus muscle was transposed one full tendon width nasally, and the right inferior rectus muscle was transposed one full tendon width laterally. The left superior rectus was transposed one full tendon width temporally, and the right inferior rectus was transposed by one tendon width medially.

Postoperatively the AHP was reduced from 45° to 25°. She continued to exhibit a right hypotropia on follow-up at age 13, measuring between 2^{Δ} and 8^{Δ}, as well as a moderate esotropia. Fundus photography documented an estimated 25° right excyclotorsion and 10° left incyclotorsion. There was also a significant reduction of her primary position upbeat nystagmus in her new head position. The patient's skew deviation decreased from 2^{Δ} to 0^{Δ} and cyclotorsion resolved on double Maddox rod following supine positioning as previously reported in OTR.[6] The upbeat nystagmus was notably dampened as well. Unfortunately, her earlier operative clinical records from infancy were no longer accessible.

The patient was most recently seen at age 30 years to assess the degree of resolution for her OTR symptomatology. Her best-corrected visual acuity was 20/32 in the right eye and 20/40 in the left eye. There were no complaints of diplopia, oscillopsia, or tilt of the perceived vertical. A right esotropia of 20^{Δ} and hypotropia of 10^{Δ} (left eye fixation) alternated with a left esotropia of 14^{Δ} and hypertropia of 3^{Δ} (right eye fixation). Extraocular eye movements showed a slight abduction deficit of the right eye and moderately limited abduction and adduction of the left eye. The vertical nystagmus was controlled by a residual mild right head tilt and became somewhat evident in the erect position of the head. Eye closure did not affect the head tilt angle. The patient demonstrated sustained improvement of her right AHP at approximately ≤25° since surgery. She is able to voluntarily straighten her head to graviceptive or normal vertical during eye closure and approximated her SVV to be 1.2° (within normal limits) to the left on the bucket test.[7-9]

Discussion

The intent of rotating the eyes in the direction of the head tilt and the tilted SVV (torsional augmentation) is to drive the return of the initial vision by compensatory tilt adjustment toward her perceived vertical. In contrast to traditional surgical correction of the head tilt with cyclotropia described by von Noorden[10] in patients with idiopathic ocular head tilt, torsion augmentation surgery was recently utilised successfully by Brodsky[6] reported in the short term in an adult with OTR with lateropulsion. Our patient experienced an acceptable and lasting improvement in her rightward head tilt and associated upbeat nystagmus, which was sustained at 25 years' follow-up. It should be noted that this surgery followed the original bilateral horizontal strabismus surgery, which might influence the interpretation of these findings.

We postulate that the correction of the AHP following surgical augmentation of binocular torsion involves further rotation of the retinal image in the direction of head tilt, and that this induces a compensatory torsional vestibular ocular reflex counteroll that serves as the driving force to realign the head back toward her subjective vertical. The postoperative ocular adjustment drive is different from the direct reestablishment of the visual vertical in idiopathic cycloptropia.

Unlike Brodsky's case, our patient had a congenital cerebellar and brainstem malformation in contrast to a diffusely, acquired process involving small vessels of subcortical white matter. Further, our patient exemplified a complete OTR with accompanying skew deviation rather than partial OTR (nearly absent skew). Postoperatively, our patient was able to maintain a transient voluntary upright head position for the sake of appearance but had no resolution of her ataxic symptoms apart from amelioration of the blur associated with her nystagmus.

Clinically, in suspected cases of OTR, the upright-supine test can be applied to evaluate whether the utriculo-ocular pathway is implicated in causing the AHP.[6] Structural neuroimaging, including MRI, would further elucidate the localization of the lesion along the central and peripheral vestibular pathway and distinguish focal versus diffuse neurological involvement in surgical planning. Moreover, fundus photography can document the severity and symmetry of the paradoxical OTR torsion pre- and postoperatively and allow comparison with the relative degree of head tilt.

References

1. Brandt T. Vertigo: Its Multisensory Syndromes. Munich: Springer Science & Business Media; 2013.
2. Westheimer G, Blair SM. The ocular tilt reaction—a brainstem oculomotor routine. Invest Ophthalmol 1975;14:833-9.
3. Brandt T, Dieterich M. Skew deviation with ocular torsion: a vestibular brainstem sign of topographic diagnostic value. Ann Neurol 1993;33:528-34.
4. Parulekar MV, Dai S, Buncic JR, Wong AM. Head position–dependent changes in ocular torsion and vertical misalignment in skew deviation. Arch Ophthalmol 2008;126:899-905.
5. Dieterich M, Brandt T. Ocular torsion and tilt of subjective visual vertical are sensitive brainstem signs. Ann Neurol 1993;33:292-9.
6. Brodsky MC, Holmes JM. Torsional augmentation for the treatment of lateropulsion and torticollis in partial ocular tilt reaction. J AAPOS 2012;16:141-4.
7. Wong AM, Colpa L, Chandrakumar M. Ability of an upright-supine test to differentiate skew deviation from other vertical strabismus causes. Arch Ophthalmol 2011;129:1570-75.
8. Frisén L. Practical estimation of ocular torsion and the subjective vertical. Neuro-ophthalmol 2000;23:195.
9. Zwergal A, Rettinger N, Frenzel C, Dieterich M, Brandt T, Strupp M. A bucket of static vestibular function. Neurology 2009;72:1689-92.
10. von Noorden GK, Jenkins RH, Rosenbaum AL. Horizontal transposition of the vertical rectus muscles for treatment of ocular torticollis. J Pediatr Ophthalmol Strabismus 1993;30:8-9.

Angiostrongylus—a technique for removing a rare parasite from the cornea of a child

Pradhnya Sen, MBBS, MS, Ashok Date, MBBS, MS, Elesh Jain, DOMS, DNB, and Amit Mohan, MS, FAICO

We report a case of an Angiostrongylus parasite in the corneal stroma in an 8-year-old boy and our technique for its removal. The parasite was identified on slit-lamp examination. Its location was confirmed on anterior segment optical coherence topography (AS-OCT). The parasite was found on the superficial corneal stroma in the periphery and was removed after lamellar dissection of the cornea following marking with trephine.

Author affiliations: Pediatric Ophthalmology and Strabismus, Sadguru Netra Chikitsalaya and Post Graduate Institute of Ophthalmology, Chitrakoot, Madhya Pradesh, India

Submitted August 24, 2018.
Revision accepted November 13, 2018.
Published online February 12, 2019.

Correspondence: Dr. Pradhnya Sen, MBBS, MS, Consultant and Head of Department, Pediatric Ophthalmology and Strabismus, Sadguru Netra Chikitsalaya and Post Graduate Institute of Ophthalmology, Jankikund, Chitrakoot, MP, India (email: drpradhnyasen@rediffmail.com).

J AAPOS 2019;23:119-120.

1091-8531/$36.00
https://doi.org/10.1016/j.jaapos.2018.11.005

Case Report

An 8-year-old boy presented at Children Eye Care Centre, Sadguru Netra Chikitsalya, Chitrakoot, with a complaint of foreign body sensation, redness, and diminished vision in his right eye for the previous 4 days, following traumatic injury with a wooden object. On examination, his best-corrected visual acuity was 20/60 in the right eye and 20/20 in the left eye. Intraocular pressure was 14 mm Hg in each eye by noncontact tonometer. Slit-lamp examination revealed circumcorneal congestion in the right eye, and two white, coiled structures with a tracklike appearance from 10 to 12 o'clock in the peripheral superficial corneal stroma, with surrounding infiltrates (Figure 1). The anterior chamber showed a fibrinous reaction, the pupil was round and reactive to light, and the lens was clear. The left eye was normal. Dilated fundus examination of both eyes was within normal limit.

Anterior segment optical coherence topography (AS-OCT) revealed two linear opacities in the superficial stroma (Figure 2). One was within 181 μm and the other within 131 μm of the anterior surface. Systemic examination did not reveal any abnormality. Routine blood investigations, x-ray chest, abdominal ultrasound, and stool examination were within normal limits. A provisional diagnosis of corneal parasite was made, and surgical excision under general anesthesia was planned with cover of systemic and topical medications (albendazole, 15 mg/kg tablet; prednisolone 1 mg/kg tablet; moxifloxacin and dexamethasone eye drops 4 times daily; atropine ointment at bedtime).

The area containing the parasite was removed with a 4 mm corneal trephine. The segment was elevated using a crescent blade (Figure 3), and the worm was removed whole; a sample was sent for histopathological examination. The parasite was identified as *Angiostrongylus cantonensis*. The patient's status improved significantly following treatment, with resolution of anterior uveitis and healing of the corneal defect within 7 days (Figure 4). At 6 weeks' follow-up the eye had quieted, and visual acuity was 20/20 in each eye.

Discussion

Angiostrongylus is a rodent lungworm commonly encountered in Southeast Asian countries. It is a well-recognized cause of eosinophilic meningitis in many Pacific islands and in Southeast Asia.[1] The parasite normally cycles between rodents and several land mollusks. Rodents are the definitive hosts; mollusks or snails are intermediate hosts. It is thought that human infection occurs following ingestion of undercooked infected snails or raw vegetables contaminated by larvae from mollusks.[2] Ocular