Exotropia-Hypotropia Complex in High Myopia

Sumit Monga, MS, DNB, FRCS; Ramesh Kekunnaya, MD, FRCS; Virender Sachdeva, MS

ABSTRACT

Purpose: To highlight the association of exotropia-hypotropia complex in cases of high myopia and its surgical outcome.

Methods: A retrospective study of 15 consecutive patients, diagnosed as having high myopia and exotropia-hypotropia, observed between January 2002 and May 2012 was performed. The main outcome measures were clinical presentation, orbital imaging features, and the surgical outcome of cases.

Results: Of the 15 patients, 9 were female. The median age was 23.5 years (range: 10 to 35 years). Thirteen cases had unilateral high myopia in the deviated eye. The mean axial length of the deviated eye was 29.43 ± 1.51 mm. Eight patients (53%) had a history of progressive eye deviation in adulthood. All patients had amblyopia of the deviating eye. The mean preoperative exotropia was 37.2 ± 8.72 prism diopters (PD) (range: 18 to 50 PD). The mean preoperative hypotropia was 12.73 ± 5.58 PD (range: 5 to 20 PD). An elevation deficit was noted in 6 patients (40%). On orbital imaging, high myopic eyes with deviation did not show significant displacement of rectus muscles in comparison to fellow eyes (P > .05, all rectus muscles). Eight cases (53%) were surgically managed with a median follow-up of 7 months. Intraoperatively, one case was found to have misplaced muscle insertion. Successful alignment was achieved by surgery on either horizontal rectus muscles alone (37%), or in combination with vertical muscles (62%).

Conclusions: Exotropia-hypotropia complex can occur in association with high myopia. No specific etiologic factors, clinical or radiological, were identified in this study. Successful alignment may be achieved in these cases using tailor-made strabismus procedures.

INTRODUCTION

There has been a continuing evolution in understanding the etiopathogenesis of strabismus in high myopia. The combination of exotropia-hypotropia complex, also called the heavy eye syndrome, has been shown to be caused by superotemporal globe prolapse due to axial distension of the globe. The latter has been shown to cause displacement of superior and lateral rectus muscles. In contrast, there has only been a brief mention of the association of exotropia-hypotropia complex in cases of high myopia. Herein, we report the occurrence of exotropia-hypotropia complex in cases of high myopia that presented at our tertiary care center.

PATIENTS AND METHODS

In this retrospective study, the medical records of 15 consecutive patients with high myopia...
(≥ 27 mm axial length) and combined exotropia and hypotropia were analyzed. All patients were seen at L. V. Prasad Eye Institute, Hyderabad, India, between January 2002 and May 2012. A total of 11,435 patients with high myopia was seen during the same period, which is equivalent to a hospital incidence of approximately 0.13%. We excluded cases of non-axial high myopia, history of previous intraocular or scleral buckling surgery, or any additional confounding ocular motility disturbance (eg, thyroid eye disease). Informed consent was obtained from all patients regarding use of their clinical data for research purposes. The Institutional Review Board of Hyderabad Eye Research Group approved the retrospective review of data and the study adhered to the tenets of the Declaration of Helsinki.

At presentation, comprehensive ophthalmic examination and A-scan echography (Ocuscan; Alcon Laboratories, Inc., Fort Worth, TX) were performed in all patients to determine axial length of the globe. The amount of ocular deviation and ocular motility at presentation were noted in all cases. All orbits were imaged using either a magnetic resonance imaging scanner (1.5 Tesla; GE Healthcare, Little Chalfont, UK) with a head coil or with computed tomography scan. The latter was obtained in some cases, owing to financial constraints. Fine coronal sections (2-mm cuts) of the orbit were taken. The plane containing the globe–optic nerve junction was designated as the reference plane. In addition, the positions of centroids of rectus extraocular muscles (EOMs) (horizontal and vertical distances in millimeters) were measured from the centroid of the optic nerve as the reference point. The orbital scans were evaluated for noting any obvious displacement of EOMs or any other gross ocular pathology. The corresponding scans of the fellow eyes of the patients were used as controls. Inter-eye comparison of rectus EOM position and statistical analysis were done using the paired Student’s t test (STATA version 11; StataCorp LP, College Station, TX).

Based on the strabismus and radiological evaluation, an appropriate surgical plan was devised for all patients. Surgical outcomes were recorded in all cases. Patients who chose conservative management for their strabismus were advised regarding appropriate regular follow-up.

RESULTS

The clinical characteristics of the included cases are depicted in Table 1. The median age at presentation was 23.5 years (range: 10 to 35 years). All patients had a varying degree of amblyopia in the deviating eye. At presentation, only two cases (13%) had bilateral high myopia. The mean axial length of the deviated eyes (29.60 ± 1.51 mm) was significantly more than the axial length of the fellow eyes (24.69 ± 1.87 mm; P < .0001). The mean preoperative exodeviation and hypodeviation were 37 ± 9 prism diopters (PD) (range: 18 to 50 PD) and 13 ± 6 PD (range: 5 to 20 PD), respectively (Table 1).

On orbital imaging, the positions of the EOMs could be interpreted reliably in 14 cases (93%). No significant displacement of the rectus muscles of the deviating eyes was found compared to the fellow or fixing eyes, which served as controls (P > .05 for all EOMs) (Table 2).

Eight patients (53%) complied with the surgical advice. The median postoperative follow-up was 7 months. Successful surgical outcome (horizontal deviation < 8 PD, vertical deviation < 5 PD) could be obtained in seven (87%) cases (Figure 1). Only one case (13%) required a second surgery for residual exodeviation and hypotropia (case 2, Table 1). In one case, the inferior rectus insertion was found to be temporally displaced intraoperatively (case 4, Table 1) and a nasal transposition of the inferior rectus muscle was done.

DISCUSSION

To the best of our knowledge, this is the largest case study reporting the occurrence of exotropia and hypotropia in patients with high myopia (http://www.ncbi.nlm.nih.gov/pubmed, English literature, accessed April 8, 2013). In this study, 15 young patients with high myopia and exotropia-hypotropia complex were diagnosed over a decade. The majority of cases had unilateral high myopia with amblyopia. Almost half of the cases had onset of progressive strabismus in adulthood. In this study, no specific radiologic or intraoperative findings were identified that could explain the etiopathogenesis of the exotropia-hypotropia complex along with high myopia. Almost half of the cases underwent surgical correction of strabismus with good postoperative alignment.

Both horizontal and vertical strabismus have been reported to occur in high myopes. In a large study of 636 patients with pathologic myopia, Tanaka et al. reported the prevalence of exotropia and vertical heterotropia in 8.8% and 16.2% of patients,
<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age/Gender</th>
<th>Onset of Progressive Deviation</th>
<th>BCVA (Decimal VA)</th>
<th>SE (D)</th>
<th>AL (mm)</th>
<th>Preoperative Deviation at Presentation (PD)</th>
<th>Ocular Motility Deficit in Deviating Eye</th>
<th>Surgery</th>
<th>Postoperative Primary Gaze Alignment at Last Follow-Up</th>
<th>Follow-up (mo)/Remarks</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>10/F</td>
<td>Childhood</td>
<td>1</td>
<td>0.041</td>
<td>-1.5</td>
<td>-13.5</td>
<td>Yes, OS-1 elevation deficit</td>
<td>OS LR Rc 8 mm, OS MR Res 4.5 mm</td>
<td>Central HBR</td>
<td>26 (M)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>OD OS</td>
<td>OD OS</td>
<td>OD OS</td>
<td></td>
<td></td>
<td>OD IO Myectomy</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>11/F</td>
<td>Childhood</td>
<td>0.015</td>
<td>1</td>
<td>-11.5</td>
<td>plano</td>
<td>Yes, OD elevation deficit (-1) and OS limitation of depression in adduction (-2)</td>
<td>1st Surgery: OD LR Rc 8.5 mm, MR Res 5.5, OU IO recession</td>
<td>16 PD XT, RHoT 22 PD after 1st Surgery/8 PD XT, Right HoT 4 PD after 2nd surgery</td>
<td>58 (M) initially misdiagnosed as OS SOP/Re-operated for residual strabismus</td>
</tr>
<tr>
<td>3</td>
<td>23/F</td>
<td>Adulthood</td>
<td>1</td>
<td>0.1</td>
<td>-0.25</td>
<td>-24</td>
<td>No</td>
<td>OS LR Rc 8, OS MR Res 5 (HTU); OS SO tenotomy (posterior 7th/8th)</td>
<td>ET 6 PD</td>
<td>19 (F) Preop OS SO tight/Postop 'A' pattern collapsed</td>
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<tr>
<td>4</td>
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<td>Adulthood</td>
<td>0.8</td>
<td>0.125</td>
<td>-1.25</td>
<td>-19.5</td>
<td>No</td>
<td>OS LR Rc 8 mm, OS MR Res 4.5 mm (FTU), OS IR Rc 5 mm (FTN)</td>
<td>5 PD XT, LHT 8 PD</td>
<td>4 (M) Preop IR insertion temporally placed by nearly 10 mm</td>
</tr>
<tr>
<td>5</td>
<td>32/M</td>
<td>Childhood</td>
<td>0.008</td>
<td>0.1</td>
<td>-22.5</td>
<td>-1.25</td>
<td>Yes, -4 limitation of elevation in abduction, -2 elevation in up gaze and adduction in OD</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>6</td>
<td>25/M</td>
<td>Childhood</td>
<td>0.06</td>
<td>1</td>
<td>-25.75</td>
<td>-12</td>
<td>No</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Case</td>
<td>Age/Gender</td>
<td>Onset of Progressive Deviation</td>
<td>BCVA (Decimal VA)</td>
<td>SE (D)</td>
<td>AL (mm)</td>
<td>Preoperative Deviation at Presentation (PD)</td>
<td>Ocular Motility Deficit in Deviating Eye</td>
<td>Postoperative Surgery</td>
<td>Postoperative Primary Gaze Alignment at Last Follow-Up</td>
<td>Follow-up (mo)/Remarks</td>
</tr>
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<td>------------------------</td>
</tr>
<tr>
<td>7</td>
<td>26/F</td>
<td>Childhood</td>
<td>0.625</td>
<td>-0.75</td>
<td>-25</td>
<td>40 PD XT</td>
<td>No</td>
<td>OS LR Rc 8 mm, OS MR Res 5 mm (HTU)</td>
<td>Central HBR</td>
<td>8</td>
</tr>
<tr>
<td>8</td>
<td>35/F</td>
<td>Adulthood</td>
<td>0.1</td>
<td>-8</td>
<td>-0.5</td>
<td>50 PD XT</td>
<td>No</td>
<td>OD LR Rc 7 mm, MR Res 5 mm (FTU)</td>
<td>Right HoT 4 PD</td>
<td>6</td>
</tr>
<tr>
<td>9</td>
<td>17/F</td>
<td>Childhood</td>
<td>0.1</td>
<td>-25.25</td>
<td>-3</td>
<td>18 PD XT</td>
<td>Yes, -2 limitation of elevation</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>10</td>
<td>24/M</td>
<td>Childhood</td>
<td>0.008</td>
<td>0</td>
<td>-11</td>
<td>30 PD XT</td>
<td>No</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>11</td>
<td>22/F</td>
<td>Adulthood</td>
<td>0.25</td>
<td>-11</td>
<td>-0.5</td>
<td>40 PD XT</td>
<td>Yes, -2 limitation of elevation in up gaze and abduction</td>
<td>OD LR Rc 6 mm, MR Res 5 mm (FTU)</td>
<td>Central HBR</td>
<td>5</td>
</tr>
<tr>
<td>12</td>
<td>21/F</td>
<td>Adulthood</td>
<td>0.008</td>
<td>-14</td>
<td>-1</td>
<td>40 PD XT</td>
<td>No</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>13</td>
<td>19/M</td>
<td>Adulthood</td>
<td>0.5</td>
<td>0.25</td>
<td>MH</td>
<td>30 PD XT</td>
<td>Yes, OS-1 limitation of elevation deficit</td>
<td>–</td>
<td>–</td>
<td>OU surgery done for complicated cataracts</td>
</tr>
<tr>
<td>14</td>
<td>17/F</td>
<td>Adulthood</td>
<td>0.12</td>
<td>0.66</td>
<td>-18</td>
<td>40 PD XT</td>
<td>No</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>15</td>
<td>27/M</td>
<td>Adulthood</td>
<td>0.008</td>
<td>1</td>
<td>-19.5</td>
<td>25 PD XT</td>
<td>No</td>
<td>OD LR Rc 5 mm, MR Res 4 mm (FTU, IR Rc 5 mm)</td>
<td>ET 4 PD</td>
<td>3</td>
</tr>
</tbody>
</table>

BCVA = best-corrected visual acuity; VA = visual acuity; SE = spherical equivalent; D = diopters; AL = axial length; PD = prism diopters; OD = right eye; OS = left eye; XT = exotropia; HoT = hypotropia; LR = lateral rectus; Rc = recession, MR = medial rectus; Res = resection; IO = inferior oblique; HBR = Hirschberg reflex; SOP = superior oblique palsy; OU = both eyes; SR = superior rectus; HTU = half tendon upshift; SO = superior oblique; ET = esotropia; preop = preoperative; FTU = full tendon upshift; IR = inferior rectus; FTN = full tendon nasalization; LHT = left hypertropia; MH = media haze due to lenticular opacity

*Childhood < 12 years old, adulthood > 12 years old.
respectively. However, the nature of the type of vertical deviation and those having combined exotropia and vertical deviation have not been specified. Moreover, the study did not provide orbital imaging details of the included cases.

In the current study, because most of the patients had full or nearly full ocular versions, the strabismus did not qualify as myopic divergent fixus variety, as has been described previously.\textsuperscript{5-7} In our study, the majority of the cases had unilateral high myopia, whereas most reported cases of myopic convergent strabismus fixus were reported to be bilateral.\textsuperscript{8} Regardless of age of onset, all of our patients gave recent history of progression of their deviation. The progressive nature of strabismus was similar to the results in a large study of myopic convergent strabismus fixus, although the age of onset (mean age: 53.9 ± 16.4 years) of strabismus was much higher in the latter study.\textsuperscript{7} All patients in our study had amblyopia of the deviating eye, which could be explained by either anisometropia alone or combined mechanism of both anisometropia and strabismus. The presence of dense amblyopia may explain the absence of diplopia in our study, in contrast to its presence in patients with high myopia with esotropia-hypotropia complex.\textsuperscript{8}

Vertical deviations have been known to coexist with exotropic horizontal strabismus and have been described in up to 50% of the patients who present with exotropia.\textsuperscript{10} The small-angle vertical tropia can be present in patients with intermittent exotropia, consecutive exotropia, and constant exotropia.\textsuperscript{10} It has been shown that there is no advantage in eliminating a vertical component of 5 PD or less.\textsuperscript{11} In

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**TABLE 2**

<table>
<thead>
<tr>
<th>Eye</th>
<th>MR X</th>
<th>Y</th>
<th>LR X</th>
<th>Y</th>
<th>SR X</th>
<th>Y</th>
<th>IR X</th>
<th>Y</th>
</tr>
</thead>
<tbody>
<tr>
<td>Deviating eye</td>
<td>10 ± 3</td>
<td>1.57 ± 2.69</td>
<td>18.71 ± 7.18</td>
<td>2.42 ± 2.99</td>
<td>0.141 ± 3.93</td>
<td>10 ± 3.52</td>
<td>0.71 ± 2.28</td>
<td>17.28 ± 4.02</td>
</tr>
<tr>
<td>Fellow eye</td>
<td>8.57 ± 2.76</td>
<td>0.57 ± 2.57</td>
<td>18.85 ± 2.03</td>
<td>1.83 ± 2.78</td>
<td>0.57 ± 3.57</td>
<td>12.57 ± 2.37</td>
<td>1.28 ± 1.79</td>
<td>16.71 ± 4.30</td>
</tr>
<tr>
<td>$p^b$</td>
<td>.17</td>
<td>.31</td>
<td>.94</td>
<td>.58</td>
<td>.76</td>
<td>.08</td>
<td>.45</td>
<td>.71</td>
</tr>
</tbody>
</table>

MR = medial rectus; LR = lateral rectus; SR = superior rectus; IR = inferior rectus; X = horizontal coordinate distance in mm; Y = vertical coordinate distance in mm.

\textsuperscript{a}Data are mean distances of the centroid of extraocular muscles from the centroid of optic nerve (millimeters ± standard deviation).

\textsuperscript{b}Statistical significance was determined between the parameters of deviating eyes as study group and the fellow eyes as control group (paired t test).

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![Figure 1](image-url)

**Figure 1.** Clinical presentation and management of exotropia-hypotropia complex in high myopia. (A) Preoperative diagnostic gaze positions of case 3, who presented with high myopia with pattern exotropia and large hypotropia in the left eye, along with its over-depression in adduction. (B) Postoperative nine-gaze photos of the same patient showing good surgical outcome, at the 2-month follow-up visit, after recession-resection procedure with half tendon up-shift of horizontal muscles of left eye, combined with superior oblique posterior tenotomy of the left eye.
contrast, the vertical deviation along with exotropia in our study of high myopes (> 5 PD in 80% of cases) was too significant to ignore. Several studies have examined the treatment of vertical deviations in association with horizontal strabismus.\textsuperscript{12,13} However, none of these studies have specified the type of refractive error or axial lengths of their patients. Hence, it is difficult to draw direct comparisons between these groups of patients and those in our study.

Over the past decade, extensive research has substantiated the fact that the development of exotropia-hypotropia complex in myopic strabismus fixus occurred due to the displacement of extracocular muscle paths.\textsuperscript{2,3} Similarly, Krizik et al. also reported the association of exotropia-hypotropia in two cases of high myopia, which had mislocated medial rectus muscles.\textsuperscript{2} However, in our study, we did not find any significant displacement of EOMs and the presence of associated significant hypotropia in some cases of sensory heterotopia, all of which had associated overactions of the oblique muscles.

Table 1

Twelve of the initial cases in this study were misdiagnosed as cases of congenital superior palsy (cases 1 and 2). It was presumed that the paretic eye was the fixating eye and the documented over-elevation in adduction of the opposite eye was probably pseudo-inferior oblique overaction.\textsuperscript{19} In case 2, the residual exotropia and hypotropia was managed by loop myopexy of the superior rectus and medial rectus muscles. In the latter case, the EOMs had normal insertions and muscle paths. However, we would not advocate the loop myopexy procedure in the absence of displaced EOMs. In retrospect, the management of these two initial cases at our tertiary care institute highlights the diagnostic and treatment dilemma that surgeons can face while dealing with exotropia-hypotropia complex in high myopia.

We understand that our study might have some limitations. Owing to the retrospective collection of data, we included single documented deviation and could not confirm the progression of strabismus with time, except by means of history. Additionally, we did not have uniform imaging protocols for this study. We did not use fixation controlled orbital scans or specialized computer simulation software, as described previously, to quantify the EOM positions.\textsuperscript{2,3,20} Moreover, owing to financial reasons, some of our patients had obtained computed tomography scans, instead of magnetic resonance imaging, for orbital imaging. Due to the retrospective nature
of the study, we could not take patients with high myopia and aligned eyes as controls. The patients in this study were treated by different surgeons, whose individual preferences might have influenced the management protocols. Moreover, our small sample size did not have the power to enable us to make any definite recommendations regarding surgical management of exotropia-hypotropia complex in high myopia. Nevertheless, we believe that the data from this study would be useful in highlighting the above-mentioned strabismus in high myopes.

The presence of exotropia-hypotropia complex is a possibility in high myopia. A majority of the patients with exotropia-hypotropia complex had unilateral high myopia with dense amblyopia. Most patients presented with progressive deviation at a young age. Care should be taken to distinguish the incomitant strabismus of high myopia from sensory deviations and paralytic strabismus, especially contralateral superior oblique palsy. None of the patients in this study showed any gross displacement of EOMs on orbital imaging. However, orbital imaging of all patients with high myopic strabismus is advisable. Based on the preoperative measurements and intraoperative inspection of rectus muscle insertions, the combined exo-hypodeviation can be managed by appropriate horizontal and vertical muscle surgery. The exact etiologic factors, either clinical or radiological, could not be identified in this study. However, future research aided with high resolution advanced imaging protocols would probably contribute to a better understanding of exotropia-hypotropia complex in high myopia.

REFERENCES


