

# A Case Study and Literature Review of Coexisting Dissociated Vertical and Horizontal Deviations

Cassandra Norris, BSc BA MOrth<sup>1</sup>  
Linda Santamaria, MAppSc DipAppSc(Orth)<sup>2</sup>

<sup>1</sup>Department of Clinical Vision Sciences, La Trobe University, Melbourne, Australia

<sup>2</sup>Department of Surgery, Monash University, Melbourne, Australia

## ABSTRACT

Adequate treatment of dissociated deviations has eluded eye care professionals since its discovery in 1895. This case-based investigation presents a review of the various treatment methods that have been advocated for both dissociated vertical and dissociated horizontal deviations,

particularly in the presence of a consecutive exotropia and inferior oblique overaction. This difficult case provided an excellent opportunity to elucidate how surgery options change in the presence of multiple ocular anomalies.

**Keywords:** infantile esotropia, consecutive exotropia, dissociated vertical deviation, dissociated horizontal deviation

## INTRODUCTION

**E**ssential infantile esotropia, previously termed congenital esotropia, is defined as a manifest eso-deviation with an onset between birth and six months.<sup>1</sup> It is the most common form of strabismus, with a reported incidence of 0.1% to 1.0%.<sup>2</sup> Early treatment is aimed at surgically straightening the eyes in order to improve cosmesis and promote the development of binocular vision, though the outcome of orthotropia with full stereoacuity is considered rare.<sup>1-3</sup> Prior to surgical intervention, non-surgical treatment includes correction of any refractive error and occlusion for amblyopia until equal visual acuity or alternation of fixation is achieved.<sup>1</sup> After surgery, children require continued review for observation of amblyopia and visual development and also stability of the deviation, as further operations may be required for residual esotropia, consecutive exotropia or vertical muscle imbalance.

Infantile esotropia includes a constellation of associated features including primary inferior oblique overaction with or without an associated V-pattern, dissociated vertical deviation and latent nystagmus<sup>1,4-6</sup> which are rarely evident at the time of diagnosis but generally occur after two years of age.<sup>1,4</sup> Primary inferior oblique overaction is defined as increased elevation in the adducted eye on version testing without an associated superior oblique palsy.<sup>7,8</sup> These overactions have been reported in 63% to 78% of children with infantile esotropia,<sup>4,8</sup> the incidence depending upon the age of the children, most frequently presenting in the

second year of life and generally apparent by 7 years of age.<sup>4</sup>

Dissociated vertical deviation (DVD) is characterised by a slow elevation, abduction and extorsion of the non-fixing eye under cover, or when the patient is inattentive or fatigued.<sup>9</sup> DVD has been reported in 49% to 76% of children with infantile esotropia,<sup>3,4,8-11</sup> with the mean onset reported as 2.8 years with none developing after 6 years of age.<sup>9</sup> Infantile esotropia with both inferior oblique overaction and DVD has been reported in 45% to 59%.<sup>4,8</sup>

Another less frequently reported variant of a dissociated strabismus complex<sup>12,13</sup> is dissociated horizontal deviation (DHD), defined as a change in horizontal alignment associated solely with a change in the balance of input from the two eyes, usually manifesting as a spontaneous exo-deviation of greater magnitude in one eye during alternate prism cover test.<sup>13-15</sup> Generally the movement is slow, variable and asymmetrical in the two eyes.<sup>13-15</sup> Keskinbora and Puler,<sup>5</sup> in a long-term follow-up study of children with infantile esotropia noted DHD in 3% to 6%, however it was reported in 50% of 28 children with consecutive exotropia when performing the reversed fixation test specifically looking for DHD.<sup>15</sup>

## CASE REPORT

A five-year-old girl presented to the clinic, having been managed elsewhere as an infant. She was initially diagnosed at 8 months of age with a congenital esotropia (ET) and was prescribed a high hypermetropic correction at this time, although with poor results. Bimedial rectus recession surgery was undertaken at 12 months of age and no further problems were noted until 4 to 5 years of age. She then

Correspondence: **Cassandra Norris**  
Department of Clinical Vision Sciences,  
La Trobe University, VIC 3086, Australia  
Email: cassandra.norris12@gmail.com

presented to this clinic when her mother noticed her left eye turning out intermittently, approximately 50% of the time. She had continued wearing the glasses prescribed by her previous ophthalmologist (RE +7.25DS and LE +8.25DS) and her current visual acuity was RE 3/4 and LE 3/6 with single optotypes. On examination, with glasses she had a consecutive left exotropia (XT) of 4-6 prism dioptres (PD) at near increasing to 45-50 PD at distance. Without correction a moderate alternating ET of 20 PD was evident. Asymmetric DVD was present bilaterally, greater in the right eye. On ocular movements, bilateral inferior oblique overaction and a significant Y-pattern were noted. Without glasses a chin-down head posture was apparent, which in the presence of the Y-pattern resulted in a reduction of the esotropia. She demonstrated no stereopsis or evidence of binocular single vision.

Cycloplegic refraction demonstrated that her current glasses did not include her astigmatic correction and overcorrected her left hypermetropia. It was decided to prescribe a one-dioptre undercorrection of RE +7.00/-1.50 x 180 and LE +7.00/-2.50 x 180 in order to encourage accommodative convergence to reduce the size of the exotropia. Two months later with the new glasses, visual acuity was 6/9 each eye with Snellen chart. With glasses a 25 PD left to alternating exotropia was present at distance, with 12 PD LXT measured at near. Also noted was the presence of a dissociated strabismus complex, with both horizontal and vertical components. For distance fixation, she tended to alternate fixation and when fixing with the right eye the left exotropia was greater, however when fixing with the left eye the exotropia was less but the right hypertropia was more apparent. At this visit a 10 PD V-pattern was noted on ocular movements as opposed to the Y-pattern described at the previous visit.

She was scheduled for bilateral inferior oblique anterior transposition surgery, aiming to reduce both the DVD and inferior oblique overaction. Although it was not clinically significant, this operation may have been expected to reduce the V-pattern and could also decrease the exotropia. Postoperatively, a 2 PD LXT was present at near with glasses, increasing to 8 PD at distance, with both the V-pattern and inferior oblique overaction reduced.

## DISCUSSION

Initially the patient was thought to have an infantile esotropia, however this diagnosis in the presence of high hypermetropia lead to questioning the possibility of an alternate diagnosis such as an early-onset accommodative esotropia.

As she presented at 8 months of age, it would generally be considered that her esotropia was of the infantile esotropia variety. The more common presentation for accommodative

esotropia is generally between the ages of one and three years,<sup>16</sup> though there are instances where it has been reported earlier than 6 months of age. One study of a group of children with esotropia with an onset before 6 months found 14% had hypermetropia of greater than 2.5 dioptres, and that almost half of these had an accommodative esotropia in which control was achieved with refractive correction alone, while the remainder required surgical correction and as such, would be diagnosed with an infantile esotropia.<sup>6</sup> Another study reported 3.6% of their group of children with unilateral infantile esotropia were controlled with hypermetropic correction.<sup>17</sup>

The majority of children with an infantile esotropia have an insignificant refractive error, however a small number of children have high hypermetropia. Several studies have reported the incidence of hypermetropia in children with infantile esotropia; 11% to 17% greater than 2.5 or 3.0 dioptres,<sup>4,6,17,18</sup> 18% greater than 4.0 dioptres,<sup>5</sup> and 9% greater than 6.0 dioptres.<sup>19</sup> In the presence of her high hypermetropia and the early onset, she would be considered to have a partially accommodative esotropia; an infantile esotropia with an accommodative element superimposed, where the deviation is decreased by 10 prism dioptres or more when the accommodative component is corrected.<sup>1,16</sup> As her esotropia did not improve significantly with hypermetropic correction, bimedial rectus recession surgery was performed at 12 months of age.

After many years of retrospective studies, case series and prospective studies considering the surgical treatment for infantile esotropia and the choice of bimedial rectus recession or unilateral recession/resection, a randomised, controlled, multicentre trial has found no significant difference in the postoperative angle using either surgical approach.<sup>20</sup> In this study all of the children were operated on between the ages of 3 and 8 years, which would be considered late surgery.

Similar to the discussion regarding type of surgery, there has been long-standing debate regarding the optimum timing of surgical intervention to allow for the potential development of binocularity. The Early vs Late Infantile Strabismus Surgery Study (ELISS), a large controlled, prospective multicentre study comparing early (6 to 24 months) and late surgery (32 to 60 months), reported that gross stereopsis with the Titmus Fly was achieved by 13.5% of the early group and 3.9% of the late group, however finer levels of stereopsis were achieved by only 3.0% and 3.9% respectively.<sup>11</sup> Earlier studies have suggested 12 months as the optimal time, reporting gross random dot stereopsis in 35% to 41% of those operated before 12 months, however the number achieving finer levels of stereopsis was small.<sup>10,17</sup> One report of 'very early' surgery examined a small group of children operated prior to 6 months and found gross stereopsis in 75%, but none with finer levels.<sup>3</sup> In summary, the authors generally supported surgery prior to 24 months<sup>2,11</sup> or 12 months of age<sup>3,10,17</sup> as with our case,

however despite this optimal timing she did not achieve binocularity. There appears no strong justification for surgery before 6 months, noting the difficulty of diagnosis, assessment and intervention in this short timeframe and the finding that those who have early surgery are more likely to require subsequent operations.<sup>3,10,11,17</sup>

Consecutive exotropia such as that exhibited by the subject, is a manifest exo-deviation that develops in a previously esotropic patient<sup>19</sup> and is reported in 12% to 26% of children who have undergone surgery for esotropia.<sup>4,5,10,19,20</sup> This may not develop until some years after surgery, with many occurring more than 5 years later, often after years of 'successful' alignment,<sup>4,18</sup> as was the case with our child. The presence of limited adduction and multiple operations is associated with an increased risk of consecutive exotropia,<sup>18,19</sup> however neither of these factors were present in this case. Reduction of the hypermetropic correction to induce accommodative convergence is recommended as nonsurgical management.<sup>1,16</sup> In our patient's case this had some effect, but further treatment was still required.

In 1895, Stevens first described dissociated vertical deviation, explaining the presence of an 'alternating vertical strabismus'.<sup>12,21</sup> The pathophysiology of dissociated strabismus complexes are still under investigation, however they are known to have strong associations with nystagmus and the presence of strabismus early in life.<sup>12,22</sup> DVD is generally asymptomatic, usually bilateral and frequently asymmetrical,<sup>23</sup> and as shown by our subject, frequently becomes apparent after strabismus surgery. Interestingly, the timing of initial strabismus surgery appears to have no impact on the likelihood of developing DVD.<sup>9</sup> Simonsz et al suggested that decompensated DVD occurred more frequently with consecutive exotropia (72%) than with residual esotropia (50%),<sup>11</sup> such as was demonstrated by this patient, where the dissociated deviations coexisted with a consecutive exotropia.

DVD does not respond to orthoptic management,<sup>23</sup> so surgery is the only viable option available to alleviate this problem,<sup>22</sup> but is considered only when the deviation becomes cosmetically unacceptable.<sup>1,23-25</sup> Various procedures aim to weaken the elevator muscles or strengthen the depressor muscles as a means of correcting DVD, albeit with varying degrees of success.<sup>22,25</sup> One earlier study reported the success of large inferior rectus resections for children with DVD, stating that they were as valuable as superior rectus recessions and easier to perform.<sup>21</sup> However, the complications of restriction of elevation and palpebral fissure changes were generally considered to be unacceptable,<sup>23,25,26</sup> though correction may be considered as a secondary procedure.<sup>23,26</sup>

Conventional superior rectus recessions (3-5 mm) were found to be ineffective in treating DVD.<sup>26</sup> 1976 saw the introduction of an operation described by Cüppers which involved the weakening of an extraocular muscle by

suturing the muscle to the sclera posterior to its insertion, originally termed a fadenoperation, now commonly known as a Faden operation.<sup>23</sup> Sprague et al reported the outcomes of a 14 mm superior rectus Faden procedure combined with a graded 3-5 mm recession on 34 patients, 23 of whom gained good postoperative results. One later paper compared the outcomes of a 12-14 mm Faden procedure with or without a 3 mm superior rectus recession and reported that the combined procedure gave the best long-term effect, avoiding undercorrection particularly in cases of DVD greater than 14 prism dioptres.<sup>25</sup> In comparing a Faden/recession combination with a large superior rectus recession (7-9 mm), Esswein et al reported that though the results were similar in the short-term with 86% corrected or improved, the large recession produced the best long-term result, with 72% maintaining the result.<sup>26</sup> In contrast, Lorenz et al found the Faden/recession combination gave better long-term results compared to a 10 mm recession.<sup>25</sup> In cases where the DVD is asymmetric, bilateral surgery with a graded recession was recommended.<sup>23,25,26</sup> It must be noted that all of these reports were retrospective case series, reporting on the surgery of choice at any period in time.

A further method of reducing the overall elevation effect is an inferior oblique anterior transposition, which was performed on our patient to treat both her DVD and inferior oblique overaction. This procedure is based on the hypothesis that transposing the inferior oblique anteriorly to the temporal corner of the inferior rectus muscle insertion site increases the recession effect, converting the muscle into a depressor.<sup>27,28</sup> Kratz et al determined anteriorisation of the inferior oblique to be effective in the treatment of DVD, and that grading the anteriorisation improved the surgical outcomes,<sup>27</sup> a finding later contradicted by Engman et al who determined that grading is no more effective than the standard surgical procedure.<sup>29</sup> This procedure has been favoured for the treatment of DVD associated with inferior oblique overaction, such as in this case,<sup>28</sup> though it has also been reported that DVD improvement showed no significant difference between those with or without inferior oblique overactions.<sup>22</sup> One case series reported the use of bilateral inferior oblique anterior transposition as a treatment for bilateral inferior oblique overaction, and found a subsequent prevention or reduction in DVD, with only one case in 61 children requiring subsequent DVD surgery compared to nine in a control group of 60 children.<sup>30</sup> It was found to be more effective for smaller degrees of DVD, but less predictable and less stable long-term if the vertical deviation exceeds 15 prism dioptres.<sup>22,29,31</sup> A combination of inferior oblique anterior transposition and resection has been suggested for moderate to large DVD.<sup>24</sup>

It has been found that inferior oblique anteriorisation may result in adverse side effects, such as narrowing of the palpebral fissure and a bulging of the lower lid on elevation.<sup>28</sup> A mild limitation of elevation was noted in 20% to 27% of

cases, but was not of clinical or cosmetic concern.<sup>24,31</sup> The debate continues as to whether bilateral surgery should be performed in the presence of unilaterally manifesting DVD,<sup>32</sup> but it has been suggested that the limitation of elevation is less apparent with bilateral surgery.<sup>13-15,22</sup>

In 1976 Raab observed what is now known as dissociated horizontal deviation, terming it a horizontal variant of dissociated vertical divergence.<sup>14,15</sup> DHD is described as the occurrence of a slow and variable horizontal deviation, which changes in magnitude depending on which eye is fixing.<sup>14</sup> This ocular alignment anomaly is unrelated to accommodation, and needs to be distinguished from an unequal amount of accommodation exerted by the eyes by ensuring assessment with the full refractive correction.<sup>14,33</sup> Post-surgical changes to the extraocular muscles such as over-recession of the medial rectus, over-resection of the lateral rectus, fibrosis of the lateral rectus or a slipped medial rectus would result in differing primary and secondary deviations,<sup>14,33,34</sup> however the deviation would not be a slow movement as is seen in dissociated complexes. Ocular movements would also demonstrate gaze incomitance with these muscle weaknesses or restrictions.<sup>14,33,34</sup>

It is suggested that diagnosis of DHD include prism cover test in all positions of gaze, testing with both eyes fixing alternately to show that the size of exodeviation differs only depending on which eye is fixing,<sup>14,33</sup> though some note the difficulty in quantifying the angle in the same way as DVD<sup>33,34</sup> and suggest that an estimation of the angle may be required.<sup>34</sup> In the Bielschowsky Darkening Wedge Test patients will show a result similar to that observed in DVD, but in the horizontal plane.<sup>13,14</sup> There may also be an associated torsional component, DVD and/or latent or manifest latent nystagmus.<sup>14,15,34</sup> A positive reversed fixation test result is also indicative of the presence of DHD.<sup>14,15,33</sup> The slow movement will be observable on cover testing, so occluding the eye for a longer period allows time for the dissociated deviation to appear.<sup>33,34</sup> Also evident is an increase in the size of the deviation during periods of visual inattention,<sup>14,15,33,34</sup> and this is further shown when the deviation is a greater magnitude under anaesthesia than during prism cover testing.<sup>14,15,33</sup>

A study by Brodsky and Fray found 50% of 28 patients with consecutive exotropia following infantile esotropia surgery demonstrated DHD, and reported a correlation with the findings of DVD, but not with latent nystagmus, however eye movement recordings were not undertaken to determine the presence of subclinical latent nystagmus.<sup>15</sup> Brodsky then concluded that monocular fixation with either eye generates a dissociated esotonus in patients with DHD, superimposed upon a baseline exo-deviation in those with a consecutive exotropia.<sup>15</sup> He hypothesised that DVD, DHD and latent nystagmus present a trilogy of dissociated ocular motor responses to unequal visual input, and refuted the earlier theory presented by Zubcov et al that DHD could

be the manifestation of an asymmetric nystagmus blockage syndrome.<sup>35</sup>

Similar to DVD, treatment of DHD is only considered when the deviation is manifesting and cosmetically unacceptable, and the only non-surgical management available is to attempt to promote fixation with the eye that deviates most often so as to improve the cosmetic effect.<sup>34</sup> Wilson et al, after a long-term follow-up study of 32 patients who had surgery for DHD, recommended unilateral lateral rectus recession of 5-8 mm for those with DHD alone, and bilateral recessions of 5-8 mm in the presence of an exotropia, with 78% requiring only one operation and a residual latent DHD remaining in 31% long-term.<sup>34</sup> In contrast, Brodsky<sup>14</sup> reported that a medial rectus advancement procedure is a more effective treatment for consecutive exotropia than a lateral rectus recession, arguing that the horizontal alignment may apparently be improved, however the exotropia still exists under conditions of visual inattention. In the presence of DHD and consecutive exotropia he recommended bilateral medial rectus advancements when there is lateral incomitance or a greater deviation at near, or large bilateral rectus recessions if the distance deviation is greater, though no results are described.

As the majority of those with DHD also have DVD, simultaneous horizontal and vertical surgery may be required. In an analysis of a retrospective series of 20 patients with both DVD and DHD, 15 had surgery for DHD, with 10 of these having unilateral horizontal surgery combined with bilateral vertical surgery.<sup>33</sup> Wilson and McClatchey noted that DHD can be corrected with more conservative amounts than the large recessions recommended for DVD, again recommending simultaneous lateral and superior rectus recessions.<sup>13</sup>

## CONCLUSION

This case illustrates the complexity of managing coexisting dissociated horizontal and vertical deviations. Although it is recommended that a prism cover test is done in all positions of gaze with each eye fixing, the child was diagnosed with dissociated horizontal deviation following observation of a difference in the size of deviation depending on which eye was fixing. The difficulty lies in the decision as to whether to operate on the horizontal or vertical deviation, or to do a combined procedure in the presence of combined dissociated deviations. In this child's case, surgery on the inferior obliques to improve the inferior oblique overaction as well as the dissociated vertical deviation was undertaken; however, this surgery also decreased the appearance of the horizontal deviation and the V-pattern. This case study showed a successfully treated individual with a complex case of coexisting dissociated horizontal and vertical deviations, inferior oblique overaction and V-pattern.

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