Improvement in motor development following surgery for infantile esotropia

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PURPOSE
Infantile esotropia is associated with abnormal visual development and thus may delay the achievement of developmental milestones. Although early surgery can improve visual function, less is known about its effect on motor development. Here we address whether early surgery can improve motor development.

METHODS
Recently, our lab devised the Infant Developmental Skills Survey, a 25-item questionnaire designed to assess sensorimotor and gross motor development. The questionnaire was completed by the parents of 3- to 10-month-old patients with infantile esotropia prior to surgery (n = 11005) and the parents of 6- to 11-month-old patients following surgery (n = 58). A subset of parents (n = 40) completed the questionnaire both before and after surgery. For comparison, the questionnaire was completed by the parents of infants with normal ocular alignment (n = 194).

RESULTS
Before surgery 4-, 5-, 6-, 7-, 9-, and 10-month-old patients showed delayed achievement of sensorimotor milestones (p < 0.01), and 5-, 9-, and 10-month-old patients demonstrated delayed attainment of gross motor milestones (p < 0.05) compared with normal children. However, following surgery, patients demonstrated a greater rate of sensorimotor development than age-matched controls (p < 0.0001) and caught up with normal children on both sensorimotor and gross motor skills.

CONCLUSIONS
Prior to surgery, patients with infantile esotropia were delayed in their achievement of developmental milestones. However, following surgery, a comparison group of patients showed rapid development and possessed motor skills comparable to those of normal children, suggesting that early surgery is beneficial to both visual and motor development. (J AAPOS 2008;12:136-140)

It is widely accepted that infantile esotropia is associated with severe deficits of stereopsis and fusion.1,2 Given the normal rapid development of stereopsis and fusion between 3 and 8 months of age,3-5 it is possible that infantile esotropia negatively affects the achievement of early developmental motor milestones (grasping objects, walking, and crawling), which rely, at least to an extent, on normal visual development.6 Poor stereovision has been shown to impede motor skills in older children. Recently, in a study of preschoolers investigating the effects of unilateral visual impairment, Hrisos et al7 found that reduced stereovision is associated with poor performance on tasks assessing hand-eye coordination and visuomotor skills.7

The visual outcomes of patients with infantile esotropia are substantially improved if the misalignment is surgically corrected early in life. Recent reports suggest that early muscle surgery is associated with greater prevalence of stereopsis and fusion along with reduced severity of amblyopia.1,8-14 Thus, it is possible that early surgery is also beneficial to subsequent development. To date, only two small sample studies have investigated this possibility, with mixed results.15,16

In the present study, we assess whether infantile esotropia delays the achievement of developmental milestones and investigate the potentially beneficial effects of early surgery on subsequent motor development in a large sample of patients. To address these issues, the parents of children with infantile esotropia completed the Infant Developmental Skills Survey (IDSS), a new questionnaire developed and validated (e-Supplement 1, available at jaapos.org) at the Retina Foundation of the Southwest (Morale & Birch, “Rapid achievement of developmental milestones following early surgery for infantile esotropia,” Annual Meeting, ARVO, Ft. Lauderdale, FL May 2002). The IDSS, available online at http://www.retinafoundation.org/Adobefiles/IDSS.pdf,
consists of 25 questions that evaluate an infant’s sensorimotor and gross motor skills. The questionnaire was completed before surgery, after surgery, or both.

Methods

Participants
Participants included 161 patients diagnosed with infantile esotropia with constant esodeviations of $\pm 30^\circ$. Patients ranged from 3 to 11 months of age and were referred by 14 pediatric ophthalmologists in the Dallas-Fort Worth area. All patients were diagnosed by 6 months of age and average age at surgery was 7.1 months (SD = 1.8 months). All patients were free from neurological and systemic disorders. For comparison, a control group of 194 three- to 11-month-old, full-term, healthy infants with normal ocular alignment also participated in the study. Informed, written consent was obtained from the parents of all participants after an explanation of the nature and any possible consequences of the study. The research protocol followed the tenets of the Declaration of Helsinki and was approved by the Institutional Review Board of the University of Texas Southwestern Medical Center. This research was compliant with the Health Insurance Portability and Accountability Act.

Patients with infantile esotropia were placed in one of two groups based on whether their parents completed the IDSS (described below) before surgery (ie, presurgery group) or after surgery (ie, postsurgery group). Parents of children in the presurgery group completed the IDSS a median of 1.6 months prior to surgery, whereas the parents of children in the postsurgery group completed the IDSS a median of 1.8 months following surgery. Only patients who were orthotropic or only intermittently heterotropic ($<10^\circ$ of heterotropia), with or without spectacle correction, at the 6- to 8-week postoperative visit were included in the postsurgery group (five patients were excluded from the postsurgery group due to constant heterotropia $\pm 10^\circ$). A subset of parents completed the questionnaire for their children ($n = 40$) both before and after surgery. These infants are included in both patient groups. The distribution of ages at which the parents completed the IDSS is provided for each patient group and the control group in Table 1.

Infant Developmental Skills Survey
The IDSS is designed to evaluate specific aspects of an infant’s early motor development. It consists of 25 yes/no items that were selected carefully from a number of currently existing developmental questionnaires and checklists (O’Brien, “Alive... aware... a person: A developmental model for early childhood services with special definition for visually impaired children and their parents.” Rockville, MD: Montgomery Public Schools, 1976). The first 10 items assess sensorimotor skills, ie, fine motor skills such as the ability to manipulate small objects and hand-eye coordination tasks (eg, grasping a toy or holding a bottle). The final 15 items assess gross motor skills, which require balance along with the movement and coordination of large muscles groups (eg, sitting, standing, and walking). The sensorimotor and gross motor sections of the questionnaire are scored separately, and in each case, the score equals the number of “yes” responses.

Table 1. Age distribution of participants by study group

<table>
<thead>
<tr>
<th>Age (mo)</th>
<th>Presurgery group</th>
<th>Postsurgery group</th>
<th>Control group</th>
</tr>
</thead>
<tbody>
<tr>
<td>3</td>
<td>13</td>
<td>0</td>
<td>31</td>
</tr>
<tr>
<td>4</td>
<td>16</td>
<td>0</td>
<td>28</td>
</tr>
<tr>
<td>5</td>
<td>29</td>
<td>0</td>
<td>33</td>
</tr>
<tr>
<td>6</td>
<td>23</td>
<td>3</td>
<td>26</td>
</tr>
<tr>
<td>7</td>
<td>27</td>
<td>9</td>
<td>15</td>
</tr>
<tr>
<td>8</td>
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<tr>
<td>10</td>
<td>7</td>
<td>9</td>
<td>19</td>
</tr>
<tr>
<td>11</td>
<td>0</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td>N</td>
<td>143</td>
<td>58</td>
<td>194</td>
</tr>
</tbody>
</table>

Note that, for all participants, parents completed the questionnaires without supervision. In each case, they were instructed to respond to the questions as honestly as possible, and they were informed that their child was not expected to have mastered all milestones on the questionnaire.

Statistical Analyses
One of the goals of the present study was to determine whether infantile esotropia delays the achievement of developmental milestones prior to surgery, and whether any evidence of delay persists following surgery. Thus, to test for presurgery delay, two-way ANOVAs (group $\times$ age) were conducted to compare the sensorimotor and gross motor scores of the presurgery group to the control children across the same age range (ie, 3 to 10 months of age). Similarly, two-way ANOVAs (group $\times$ age) were conducted to test for differences between the postsurgery patients and control children across the same age range (ie, 6 to 11 months of age). If the above omnibus ANOVAs demonstrated significant differences, planned comparisons were carried out in 1-month age bins to determine at what ages attainment of milestones was delayed.

A second goal of the present study was to determine whether children with infantile esotropia exhibited greater than normal rates of achievement of developmental milestones following surgery. Thus, for each patient who was assessed both presurgery and postsurgery ($n = 40$), rate of development was determined by dividing his/her difference in scores on the IDSS from pre- to postsurgery by his/her difference in age. Each patient’s rate of development was then matched to the average rate of development of control children across the same age range. Finally, the patient group’s overall mean rate of development was then compared with the mean age-matched rate of development of control children using independent samples t-tests. Comparisons were conducted separately for sensorimotor and gross motor rates of development.

Results

Presurgery Group versus Control Group
Sensorimotor and gross motor scores of the presurgery group are compared with those of the control group as a function of age in Figure 1A and B. Relative to the control group, the presurgery group showed delayed achievement on both sensorimotor milestones ($F_{1,111} = 70.09, p <$
and gross motor milestones ($F_{1,311} = 13.75, p < 0.0005$). Planned comparisons indicated that the delay of sensorimotor milestones was significant at 4, 5, 6, 7, 9, and 10 months of age ($p < 0.01$). Delay of gross motor milestones was significant at 5, 9, and 10 months of age ($p < 0.05$).

**Post surgery Group versus Control Group**

Sensorimotor and gross motor scores of the postsurgery group are compared with those of the control group as a function of age in Figure 2A and B. Unlike the presurgery group, the postsurgery group showed no evidence of delayed achievement on sensorimotor milestones ($F_{1,148} = 1.59, p = 0.21$) or gross motor milestones ($F_{1,148} = 2.82, p = 0.10$).

**Rate of Development: Surgery Group versus Control Group**

Mean rates of development for those children assessed both presurgery and postsurgery and those of the control group are provided in Table 2. The table indicates that patients showed a significantly higher rate of sensorimotor development than age-matched control children ($p < 0.0001$). However, there was no significant difference between the groups on rate of gross motor development ($p = 0.11$). Note that, although the normal rate of development is higher for gross motor milestones than for sensorimotor milestones, this trend is reversed for patients as they displayed a higher rate of sensorimotor development.

To examine whether the event of surgery itself affected rate of development, the rates of sensorimotor and gross motor development of the children who had unsuccessful surgery ($n = 5$) were compared with those of control children across the same age range. Patients and control...
Table 2. Rate of sensorimotor and gross motor development by group

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>Surgery group</th>
<th>Control group</th>
<th>Difference</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rate of sensorimotor development</td>
<td>40</td>
<td>1.41 (0.99)</td>
<td>0.63 (0.17)</td>
<td>0.78*</td>
</tr>
<tr>
<td>Rate of gross motor development</td>
<td>40</td>
<td>1.25 (0.62)</td>
<td>1.08 (0.44)</td>
<td>0.17</td>
</tr>
</tbody>
</table>

Numbers in parentheses represent standard deviations. *p < 0.0001.

children did not differ significantly on either rate of development ($p_{\text{sensorimotor}} = 0.58$; $p_{\text{gross motor}} = 0.34$).

Discussion

Our findings indicate that children with infantile esotropia exhibited delayed achievement of developmental milestones prior to surgery. The impairment of sensorimotor development was particularly profound and was evident at virtually all ages assessed. This result is predictable as the majority of sensorimotor milestones included in the questionnaire are visually demanding (eg, grasping a toy, grasping tiny objects, following a moving toy). Therefore, if a child has abnormal binocular function (eg, no stereopsis and/or fusion), achievement of such tasks might be difficult. This appears to be the case with older children, as preschoolers with no stereopsis or gross stereopsis have been shown to perform poorly on visuomotor tasks and those relying on hand-eye coordination.

The delay of gross motor milestones in presurgery patients was also significant but not as severe. A possible explanation for this finding is that the majority of attainable gross motor milestones on the IDSS are more reflective of balance and coordination of large muscle groups rather than visual functioning. Note, however, several of the gross motor items likely require adequate binocular function but are well beyond the developmental capabilities of most 3- to 10-month-old infants regardless of binocular status (eg, walking without support, running, climbing stairs). Therefore, it would be interesting to assess whether infantile esotropia delays the achievement of these relatively advanced gross motor milestones in slightly older infants.

Following surgery, patients no longer showed impaired achievement of sensorimotor or gross motor milestones. In fact, they exhibited greater rates of development after surgery as their rate of improvement on sensorimotor items was twice that of the control children. However, there was no significant difference between patients and control children on gross motor items (means, 1.25 and 1.08, respectively). Once again, this may reflect the notion that, compared with the sensorimotor items on the IDSS, attainable gross motor milestones rely less on visual functioning.

Two prior studies have investigated motor development milestone achievement in children with infantile esotropia. Both studies had methodological limitations. Rogers et al reported that, compared with children with normal alignment, patients with infantile esotropia showed greater rates of improvement on the Mental Development Index of the Bayley Scale of Infant Development. Yet this study is limited by its small sample size (18 patients, 8 control children) and its failure to match the ages of the control group to patients with infantile esotropia. In addition, children were not evaluated on the Psychomotor Development Index, which assesses posture control, coordination of large muscle groups, and manipulation of the fingers and hands. Conversely, Wheeler et al found no improvement on Mental Development Index or Psychomotor Development Index scores in a very small sample (n = 11) of patients with infantile esotropia assessed before and after surgery.

Although the results presented here suggest that early muscle surgery is beneficial to subsequent sensorimotor development, this study possesses several limitations. First, it is possible that it is the event of surgery itself, rather than successful realignment, that led to improved sensorimotor development. To examine this possibility, we analyzed whether those patients who had unsuccessful realignment surgery also demonstrated improved achievement of developmental milestones following surgery. Although the results indicated that these patients did not show significant sensorimotor or gross motor improvement compared with control children, the analysis was limited by poor statistical power due to the small sample size (n = 5).

Second, the results presented here may simply reflect the parents’ expectation of post-surgery improvement. However, if this was the case, we might expect to see an increased rate of development for both scales. Instead, the rapid rate of development was seen only for the sensorimotor scale. Furthermore, the IDSS has been validated previously (Morale & Birch, “Rapid achievement of developmental milestones following early surgery for infantile esotropia,” Annual Meeting, ARVO, Ft. Lauderdale, FL, May 2002; e-Supplement 1); there was excellent agreement between scores on the IDSS and an independent masked examiner’s assessment of whether a child could demonstrate achievement of the milestones on the questionnaire.

Finally, it must be noted that the results presented here do not indicate clearly that sensorimotor improvement was the result of improved binocular function due to surgical realignment since the evaluation of stereopsis and fusion were not part of this study protocol. In the future, it would be wise to assess these aspects of binocular function in patients with infantile esotropia both before and after surgical alignment.

Despite these limitations, it is our opinion that the rapid rate of development shown in the present study is the result of improved binocular function following surgical alignment. Early surgery is associated with improved stereoaucuity outcomes, lower rates of subsequent misalignment, and reduced risk of moderate to severe amblyopia.

Because many of the items on the IDSS are visually demanding,
improved binocular function might be expected to facilitate the achievement of these items.

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References