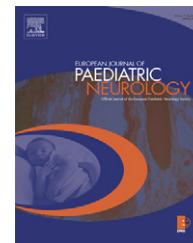




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## Original article

## Motor coordination in children with congenital strabismus: Effects of late surgery

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## ABSTRACT

**Background:** Strabismus is one of the most common visual disorders in infancy. While there is a great attention on the effects of the timing of surgery as to the development of binocular vision, little is known about the possible influence of congenital strabismus on perceptual-motor and more generally, on neuromotor development.

**Aims:** Aim of this study was to investigate perceptual-motor and motor coordination abilities of 19 children with essential congenital esotropia who underwent a late surgery (after 4 years), compared to 23 age-matched controls.

**Methods:** Children were tested using the Movement Assessment Battery for Children (Movement ABC) that were performed both 1-week before surgery (T1) and about 3 months ( $\pm 2$  weeks) after surgery (T2).

**Results and conclusions:** At T1, abnormal or borderline results were found in more than half of the children with strabismus, as opposed to only about 17% of the controls. At T2 none of the children showed abnormal Movement ABC total scores and there was no difference in global scores between the study and the control group. The two groups also did not show any significant difference in individual items of the movement ABC with the exception of those assessing ball skills.

Our results suggest that surgical correction of strabismus, even when performed after the 4th year of life, appears to be effective in improving perceptual-motor and motor function.

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## 1. Introduction

Strabismus is one of the most common visual disorders in infancy. A recent population-based study indicates that its prevalence is approximately 4% with an eso:exo rate of 5:1.<sup>1</sup> Strabismus is generally treated by surgery with the purpose to align the eyes both for aesthetic reasons and for allowing an appropriate development of binocular vision. The optimal age for surgery is, however, still controversial.<sup>2</sup> While some authors advocate early surgery to obtain a partial recovery of binocular cooperation,<sup>3,4</sup> others recommend observation and surgery at a later stage, in order to get a more precise correction and avoid multiple interventions.<sup>5</sup> In addition to the lack of consensus, it is conceivable that a certain amount of late surgeries are anyhow performed irrespective of surgeon opinions, simply because of late referrals.

Although emphasis in the debate on timing of intervention is mainly centred on prevention of further loss or restoration of binocular vision, other arguments have been also proposed. In particular, of great interest appears the possible influence of strabismus on neuromotor development, with particular regard to fine motor skills. In this respect, reliable data are lacking. An old report by Rogers and colleagues<sup>6</sup> explored the possible correlation between strabismus surgery and visual coordination abilities in infants of less than 1 year, showing a short-term significant positive effect of surgery in the areas of visual coordination assessed by the Bayley developmental scale.<sup>6</sup> More recently, Fronius and colleagues<sup>7</sup> reported an increase of pointing errors in four adults with alternating uncorrected strabismus, that were, however, unrelated to the angle of squint. In addition, an anecdotal description of motor improvements following surgery is often reported as referred by the parents or care-givers,<sup>6,8</sup> and this also reflects our personal experience.

In infants who undergo early surgical treatment the assessment of motor skills before and after therapy has mainly been performed using neurodevelopmental scales that are generally considered to have a poor sensitivity to identify specific neuromotor or perceptual motor abnormalities, both because they contain items assessing other aspects of development and also because of high variability of early developmental profiles.<sup>9–11</sup>

One of the advantages to assess children who had relatively late surgery, i.e. after the age of 4 years, is that at this age there are a few tools, that are well standardised with age-specific normative data that can provide more specific information on various aspects of perceptual motor and motor coordination abilities. In particular, the Movement Assessment Battery for Children (Movement ABC<sup>12</sup>) was found to be effective and sensitive in the assessment of fine and gross motor skills in children of three and a half years or older.<sup>13</sup>

In the present study, we have used the Movement ABC in a group of children with congenital strabismus performed after the age of 4 years with the purpose of (i) characterising their motor skills and (ii) exploring the possible effects of surgical treatment.

## 2. Methods

All the subjects of this study were enrolled from the Department of Ophthalmology of the Meyer Children's Hospital in Florence. Inclusion criteria were: (i) age above or equal to 4 years; (ii) absence of specific neurological, cognitive or behavioural disorders; (iii) diagnosis of essential congenital esotropia, defined as convergent squint developed in the first 6 month of life with latent nystagmus, initial cross-fixation and asymmetric optokinetic nystagmus<sup>14,15</sup>; (iv) absence of amblyopia or significant accommodative errors at the time of surgery.

Nineteen children (14 males and 5 females) fulfilled the inclusion criteria and were enrolled for the study. The age ranged from 48 to 71 months, with a mean age of 59 months. Three of the 19 patients did not comply with the time window of the post-surgical assessment, and were therefore excluded from the comparative section of the study.

A control group of 23 children with no strabismus matched for sex (15 males and 8 females) and age (range: 48–70 months; mean: 57) was enrolled. A written consent was obtained in all cases. The research was approved by the Ethical Committee of the Meyer Children's Hospital.

### 2.1. Ophthalmological assessment

All patients underwent a full ophthalmological assessment. Best-corrected visual acuity was assessed by means of Albin's E chart and binocular vision by means of the Lang 1 Stereo-test.<sup>16</sup> Worth-four dot test for distance and near was used to measure if the patient had suppression scotoma. Cycloplegic refraction, ocular motility and fundus examination were also performed in all subjects.

Visual-field size was assessed using kinetic perimetry.<sup>17</sup> During central fixation of a centrally positioned white ball, an identical target is moved from the periphery towards the fixation point along one arc of the perimeter. Eye and head movements towards the peripheral ball are used to estimate the outline of the visual field. Normative data are available.<sup>18</sup>

### 2.2. Surgical procedure

All surgical operations were performed by two of the authors (CR and CL). Different surgical procedures were used to correct the angle of squint. In some cases a symmetrical posterior fixation suture on the medial recti (Faden operation) was the preferred technique.<sup>19</sup> Using a limbal approach, the medial rectus was isolated and a not absorbable suture was passed on both sides of the muscle, fixing it to the sclera at 13 mm from the original insertion. In few patients a recession of 2 mm of the same muscle was added during the same procedure. In the cases of patients with a unilateral preferred fixation, a procedure on the non-dominant eye was the procedure of choice: a recession of the medial rectus and resection of the lateral rectus were performed using a Vycryl 6-0 suture. Patients with elevation in adduction received an inferior oblique recession graded on the degree of vertical deviation. In all the surgeries, the conjunctiva was not recessed and sutured with Vycryl 8-0. Postoperative topical

medication with Desametazone and Tobramycine was prescribed for 2 weeks. Patients were checked at 1, 7 and 15 days after surgery. After surgery, all patients showed an alignment within  $\pm 10$  prism diopters.

### 2.3. Assessment of motor development

Each child was tested with the Movement ABC by two paediatric physiotherapist. As all the children were between 4 and 6 years, they were all assessed using the corresponding age band, i.e. age band 1 for children between 4 and 6 years. The test was administered both before and after surgery: the first assessment (T1) was performed within 1 week before surgery in children with strabismus and at a matched age in controls; the second assessment (T2) was performed at about 3 months ( $\pm 2$  weeks) after surgery in children with strabismus and at about 3 months plus 1 week ( $\pm 2$  weeks) in controls.

The Movement ABC is a battery consisting of eight items (posting coins, threading beads, bicycle trail, catching bean bag, rolling ball into goal, one-leg balance, jumping over cord, walking heels raised) grouped in four subscales, manual dexterity, ball skills, static balance and dynamic balance.<sup>12</sup> Raw scores on each item are converted to normative scores on a 0–5 scale. These scores are then summed to produce a total score ranging from 0 to 40, with high scores indicating poor performance. According to the distribution of scores obtained by the reference sample provided by the test, total scores of more than 5.5 (<15th centile) are considered as borderline and scores of more than 10.5 (<5th centile) as abnormal. The test also provides normative data for the four

subgroups: manual dexterity, ball skills, static and dynamic balance.

### 2.4. Statistical analysis

Student t-test was used (i) to compare the results of the ABC Movement test between the two groups at T1 and at T2; (ii) to compare the results of the ABC Movement test within each group, between T1 and T2; (iii) to compare the degree of changes observed at T2 between the two groups. A *p*-value below .05 was taken as significant.

## 3. Results

### 3.1. Ophthalmologic and surgical data

Detailed information on the ophthalmologic and surgical data are reported in Table 1. Ten children had no fixation preference, five had a right eye fixation preference and four had a left eye fixation preference. In all children binocular testing showed a complete absence of stereo-acuity as measured with the Lang Test, and a suppression scotoma at the Worth-four dot test; no improvements were observed after 3 months following surgery. No improvements in visual-field size were observed in any of the subjects.

Nine patients underwent a posterior fixation suture on the medial recti (Faden operation), eight received a unilateral surgery and in two it was necessary both posterior fixation suture and medial rectus recession. In six cases an inferior oblique recession was performed to reduce the elevation in adduction, in addition to the horizontal procedures.

**Table 1 – Description of type of strabismus and surgery**

| No. | Gender | Age (ms) |    | Strabismus | Angle (PD) | Stereopsis |     | Visual fields (R;L) |        | Surgery |
|-----|--------|----------|----|------------|------------|------------|-----|---------------------|--------|---------|
|     |        | T1       | T2 |            |            | T1         | T2  | T1                  | T2     |         |
| 1   | m      | 44       | 47 | ●          | 30         | Abs        | Abs | 70; 40              | 90; 90 | F       |
| 2   | m      | 50       | 53 | ◐■         | 30         | Abs        | Abs | 90; 60              | 90; 90 | F       |
| 3   | m      | 58       | 61 | ●          | 30         | Abs        | Abs | 90; 90              | 90; 90 | F       |
| 4   | m      | 55       | 59 | ◐          | 20         | Abs        | Abs | 80; 70              | 90; 90 | RR      |
| 5   | m      | 68       | 71 | ◐■         | 20         | Abs        | Abs | 70; 80              | 90; 90 | RM      |
| 6   | m      | 68       | 71 | ●■         | 35         | Abs        | Abs | —                   | —      | F       |
| 7   | m      | 68       | 71 | ◐          | 35         | Abs        | Abs | 60; 65              | 90; 60 | RR      |
| 8   | f      | 69       | 72 | ◐■         | 25         | Abs        | Abs | 80; 90              | 90; 90 | RM      |
| 9   | m      | 58       | 62 | ●          | 30         | Abs        | Abs | 90; 85              | 75; 75 | F+RM    |
| 10  | f      | 65       | 69 | ◐          | 30         | Abs        | Abs | 90; 90              | 90; 90 | RR      |
| 11  | m      | 62       | 65 | ●■         | 40         | Abs        | Abs | 90; 90              | 90; 90 | F       |
| 12  | m      | 52       | 55 | ◐          | 14         | Abs        | Abs | 85; 85              | 90; 90 | RM      |
| 13  | f      | 63       | 69 | ●          | 30         | Abs        | Abs | 80; 90              | 90; 90 | F+RM    |
| 14  | m      | 58       | 62 | ◐          | 35         | Abs        | Abs | 70; 75              | 90; 90 | RR      |
| 15  | m      | 62       | 65 | ●■         | 35         | Abs        | Abs | 90; 90              | 90; 90 | F       |
| 16  | f      | 61       | 64 | ●          | 40         | Abs        | Abs | 90; 90              | 90; 90 | F       |
| 17  | f      | 60       | —  | ●          | 30         | Abs        | —   | 60; 65              | —      | F       |
| 18  | m      | 69       | —  | ◐          | 35         | Abs        | —   | 90; 80              | —      | RM      |
| 19  | m      | 68       | —  | ●          | 40         | Abs        | —   | 70; 70              | —      | F       |

m = male; f = female; ms = months; ◐ = left eye; ◑ = right eye; ● = both eyes; ■ = vertical strabismus; PD = prism dioptres; Abs = Absent; R = Right; L = Left; F = Faden Post-Fix suture; RR = medial rectus and lateral rectus recession; RM = medial rectus recession; ms = months; — = missing data.

### 3.2. Movement ABC

#### 3.2.1. First assessment (T1)

In the group of the 19 children with strabismus, 4 (21%) had total scores below the 5th centile, 6 (32%) had borderline (between the 5th and the 15th centile) and the remaining 9 (47%) had normal scores.

In the control group, one of the 23 children (4%) had a total score below the 5th centile, 3 (13%) had borderline scores and 19 (83%) had normal scores (Fig. 1).

A significant difference in the total score ( $p < .005$ ) was found between the study and the control group. A significant difference was also found in the subscale assessing manual dexterity ( $p < .005$ ). The analysis of single items showed statistically significant differences in posting coins, threading beads, catching bean bag and walking heels raised (see  $p$ -values in Table 2).

#### 3.2.2. Second assessment (T2)

None of the 16 children with strabismus had total scores below the 5th centile, 4 (25%) had borderline scores (between the 5th and the 15th centile) and 12 had normal scores.

None of the 23 children in the control group had a total score below the 5th centile, 1 (4%) had borderline scores and 22 (96%) had normal scores (Fig. 1).

There was no significant difference in total scores between the study and the control group. When analysing subscores and individual items, a significant difference between the two groups was only observed in the subscale assessing ball skills ( $p < .005$ ) and in the single item for catching bean bag (see  $p$ -values in Table 2).

#### 3.2.3. Comparison between T1 and T2

In order to evaluate the changes on the ABC Movement after surgery, a score was obtained in each subject based on the difference between the result of the first assessment and the result on the second assessment. Accordingly, a positive score corresponded to an improvement of performance. In the group of children with strabismus, a significant difference between the pre-surgical and post-surgical assessment was observed in the total score and in the subscales assessing manual dexterity, static and dynamic balance. When evaluating the single items, a significant difference between T1 and T2 was observed in the following: threading beads, one leg

balance and walking heels raised. In the group of controls, no difference between T1 and T2 was found with the exception of the item assessing catching.

When comparing the degree of change between the two groups, significant differences were found in the total score and in 3 of the 4 subscales, namely the manual dexterity, static balance and dynamic balance (Fig. 2). For the details on single items see Table 3.

## 4. Discussion

In this study we were able to show that children with untreated congenital strabismus, when assessed after the age of four, frequently show on the Movement ABC difficulties in perceptual motor skills and motor coordination. When examined before surgery, abnormal results (below the 5% centile) were found in 20 percent and borderline scores (between the 5th and the 15th centile) were observed in an other 32 percent. As the normative data of the Movement ABC were obtained in other countries, we also assessed a control group matched for age and sex. The comparison between strabismic children and controls confirmed the abnormal motor performance in children with strabismus, showing significant differences in the global scores of the Movement ABC. When we analysed the individual items or the subscales, we found that the subscale in which children with strabismus appeared to show the lowest abilities was the one assessing manual dexterity. In particular, there was a significant difference in two of the three items of the subscale, namely posting coins and threading beads, while in the remaining one, the bicycle trail, the score of both groups fell into normal range. It is of interest that the two items in which the strabismic children failed are those with the highest load of three dimensional information, as they both require an interaction with 3D objects and an action into all dimensions of space, while the third item is mainly carried out on a single plane, and therefore requires a lower amount of 3D information processing. Another factor, which may have also played a role, is the speed required for the task, as the score in the first two items is also based on the time spent to complete the task. The possibility that motor performances of children with strabismus might also be affected by the speed of the action is also suggested by the good performance found

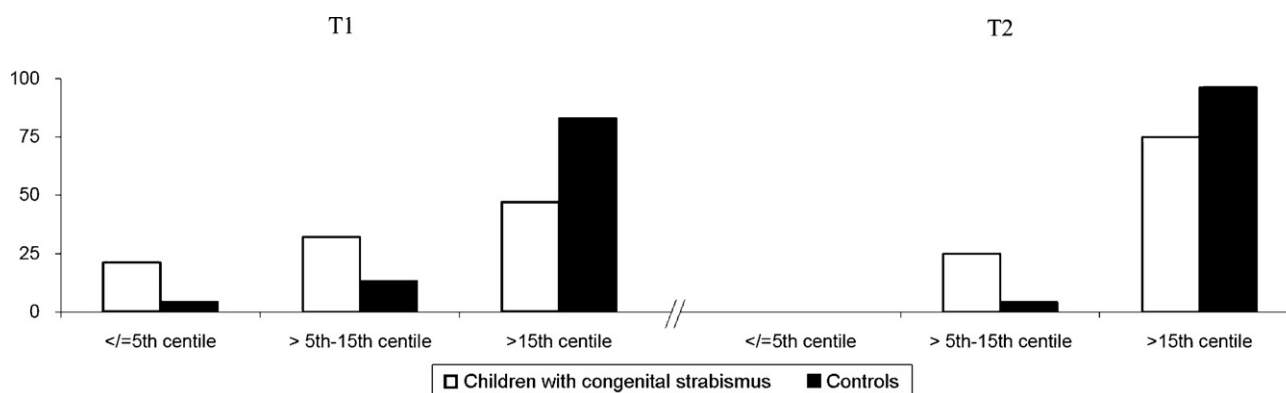
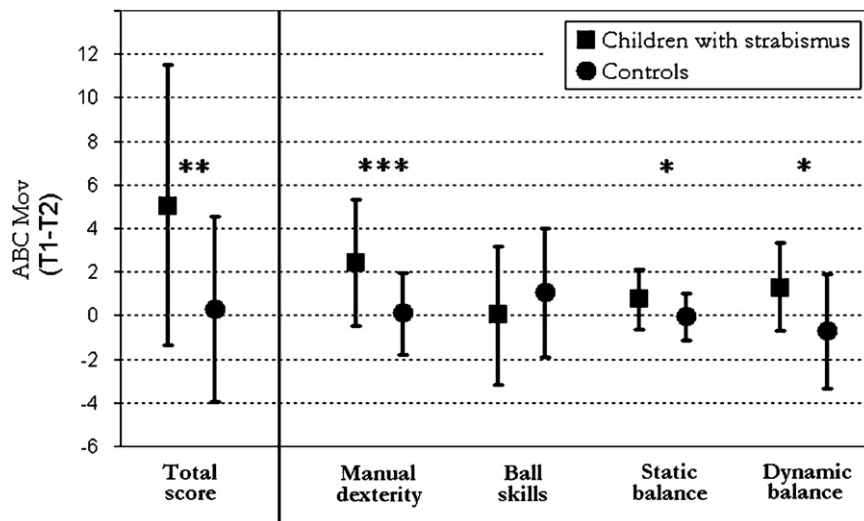


Fig. 1 – Distribution of the subjects with strabismus and controls according to centile values at T1 and T2.

**Table 2 – Mean values and statistical differences between children with strabismus and controls at T1 and T2 assessments**

|                        | T1                                 |                    |   | T2                                 |                    |   |
|------------------------|------------------------------------|--------------------|---|------------------------------------|--------------------|---|
|                        | Children with strabismus Mean (SD) | Controls Mean (SD) | Significant differences between groups p-Values | Children with strabismus Mean (SD) | Controls Mean (SD) | Significant differences between groups p-Values |
| Movement ABC           |                                    |                    |   |                                    |                    |   |
| Total score            | 10.50 (6.62)                       | 4.93 (4.75)        | 0.003   | 6.73 (4.73)                        | 4.63 (3.97)        | ns  |
| Manual dexterity       | 3.79 (2.88)                        | 1.37 (2.14)        | 0.003   | 1.97 (2.07)                        | 1.30 (1.93)        | ns  |
| Posting coins          | 0.68 (1.07)                        | 0.07 (0.23)        | 0.010   | 0.22 (0.5)                         | 1.8 (4.4)          | ns  |
| Threading beads        | 2.63 (2.06)                        | 0.87 (1.42)        | 0.002   | 1.47 (2.14)                        | 1.09 (1.59)        | ns  |
| Bicycle trail          | 0.47 (0.84)                        | 0.43 (1.44)        | ns  | 0.25 (0.68)                        | 0.43 (0.21)        | ns  |
| Ball skills            | 3 (2.03)                           | 1.65 (2.55)        | ns  | 3.06 (2.44)                        | 0.6 (1.44)         | 0.000   |
| Catching bean bag      | 2.68 (1.63)                        | 0.96 (1.69)        | 0.002   | 2.56 (2.06)                        | 0.44 (0.21)        | 0.000   |
| Rolling ball into goal | 0.32 (0.82)                        | 0.70 (1.46)        | ns  | 0.5 (0.89)                         | 0.56 (1.44)        | ns  |
| Static balance         | 1.47 (1.74)                        | 0.87 (1.38)        | ns  | 0.97 (1.1)                         | 0.94 (1.37)        | ns  |
| One-leg balance        | 1.47 (1.74)                        | 0.87 (1.38)        | ns  | 0.97 (1.1)                         | 0.94 (1.37)        | ns  |
| Dynamic balance        | 2.24 (2.49)                        | 1.04 (2.14)        | ns  | 1 (1.67)                           | 1.78 (2.21)        | ns  |
| Jumping over cord      | 1.11 (2.00)                        | 0.87 (1.77)        | ns  | 0.5 (1.32)                         | 1.22 (2.02)        | ns  |
| Walking heels raised   | 1.13 (1.65)                        | 0.17 (0.65)        | 0.014   | 0.5 (1.27)                         | 0.56 (1.5)         | ns  |



**Fig. 2 – Mean values and standard deviations of the improvement between T1 and T2. The total scores and the main subscores are shown (\* = p-value <.05; \*\* = p-value <.01; \*\*\* = p-value <.005).**

in the item of rolling ball in a goal, an other task requiring a good amount of motor accuracy but not affected by time constraints.

Another item in which strabismic children scored significantly worse than controls, was the one assessing the ability of catching. In the light of the considerations concerning poor

**Table 3 – Mean values and standard deviations of the changes between T1 and T2 in children with strabismus and normal controls**

|                          | Children with strabismus<br>Mean (SD) | Controls<br>Mean (SD)    | Significant differences between groups<br>p-Values |
|--------------------------|---------------------------------------|--------------------------|--|
| Movement ABC total score | 5.06 (6.44) <sup>a</sup>              | 0.3                      | 0.008  |
| Manual dexterity         | 2.44 (2.89) <sup>b</sup>              | 0.06 (1.87)              | 0.004  |
| Posting coins            | 0.56 (1.31)                           | –0.1 (0.45)              | 0.029  |
| Threading beads          | 1.56 (2.58) <sup>a</sup>              | –0.2 (1.56)              | 0.011  |
| Bicycle trail            | 0.31 (1.01)                           | 0.39 (1.3)               | ns   |
| Ball skills              | 0 (3.16)                              | 1.04 (2.96)              | ns   |
| Catching bean bag        | 0.12 (2.7)                            | 0.91 (1.62) <sup>a</sup> | ns   |
| Rolling ball into goal   | –0.12 (1.08)                          | 0.13 (2.2)               | ns   |
| Static balance           | 0.75 (1.39) <sup>a</sup>              | –0.06 (1.09)             | 0.047  |
| One-leg balance          | 0.75 (1.39) <sup>a</sup>              | –0.06 (1.09)             | 0.047  |
| Dynamic balance          | 1.32 (2.02) <sup>a</sup>              | –0.74 (2.63)             | 0.013  |
| Jumping over cord        | 0.5 (1.42)                            | –0.35 (2.19)             | ns   |
| Walking heels raised     | 0.75 (1.48) <sup>a</sup>              | –0.39 (1.7)              | 0.036  |

Intra-group and inter-group significant differences are indicated.

<sup>a</sup> =  $p < .05$ ; <sup>b</sup> =  $p < .01$ .

results on manual dexterity, this result is not unexpected. In fact, the task of catching a beanbag, as assessed in the ABC movement test, requires a good ability to process 3D information at a high speed.<sup>20</sup>

In this study, we were also interested to establish the possible effects of surgery performed after the age of 4 years on motor function. Although the follow-up is relatively short, the results obtained reassessing children 3 months after surgical intervention surprisingly showed significant changes between pre- and post-surgery assessment. An important improvement of the total Movement ABC score was observed in patients after surgery, as, at variance with the pre-surgery assessment, there were no children with abnormal results and only four (21%) with borderline scores. This finding was further confirmed by the comparison of the second assessment on the Movement ABC between the study and the control group, which showed no significant differences with the exception of the items of the ball skill subscale. In order to exclude a possible learning effect due to the repeated assessment, we compared the difference in the Movement ABC test between the first and the second assessment in both study and control group. In all subscales, except the ball skills, the strabismic children showed larger improvements than controls, resulting in the loss of the previously observed discrepancy in the motor abilities of the two cohorts.

Our results therefore suggest that surgical correction of strabismus, even when performed after the 1st years of life, may help to improve some aspects of motor function and in particular fine motor actions and eye–hand coordination. This finding is in accordance with previous studies exploring the improvement of eye–hand coordination in adult subjects with strabismus after surgery.<sup>21</sup> In strabismic subjects, visuomotor function appears to be highly capable of an adaptation following surgery even in adulthood whereas sensory representation, namely stereopsis, seems to be less plastic.<sup>7,22</sup>

It is of interest that not all aspects of motor function have been positively affected by treatment. In particular, the scores on the ball skill subscale did not show any significant improvement following surgery, as if the underlying mechanisms of these tasks are poorly sensitive to changes in eye alignment. In fact, several studies have explored catching abilities in subjects with strabismus suggesting a high dependence of these skills on the presence of a good binocular vision.<sup>20,23</sup> This is essential particularly in the temporal processing of the action, as shown by the dependence of catching skills on the velocity of the object, with decreased performance at increased speed.<sup>24</sup> It may be thus hypothesised that the limited improvement of binocularity observed in the first 3 months after surgery in our cohort, can be at least to some extent responsible of the poor catching skills, as opposed to other motor coordination abilities. Longer follow-ups are, however, needed to explore the possible improvements of these abilities in those cases showing a good development of stereo-acuity.<sup>25</sup>

The understanding of the mechanisms underlying the pattern of improvements observed in our patients is not straightforward. The possibility of a positive influence on motor performance by a visual field enlargement following eye realignment is not supported by the absence in our cohort of changes at the field assessment. Although, we have not been able to perform an objective measure of visual fields, such as the computerised perimetry, due to the young age of the subjects, the methodology used has proven reliable in detection of visual field development in normal children.<sup>18</sup> Moreover, most of the tasks required by the Movement ABC are based on a good central visual acuity, which was indeed present in all our patients, thus making highly unlikely the contribution of field modifications in performance improvement. It is of interest that the degree of improvement in the different motor tasks seems to be related to the magnitude of stereo-acuity required and to the speed of 3D information processing. As a consequence, it might be suggested that the



pattern of improvements observed results from a partial recovery of a binocular cooperation, secondary to eye realignment, which determines different effects based on the load of 3D information required. Unfortunately, the binocularity tests used in our study were unable to detect any changes in stereo-acuity, thus indicating that modifications of binocularity, if present, were too small to be detected with normal paediatric clinical testing such as the Lang test for binocular vision. It has also to be underlined that other forms of binocular vision other than stereopsis, such as simultaneous perception, could have developed, possibly accounting for the observed improvements.

In conclusion, our findings support the hypothesis of a high incidence of abnormalities of motor function in children with congenital strabismus, which seem to be significantly reduced by surgical correction, even when performed after 4 years of age. The pattern of improvement is not uniform for the different aspects of motor functions, but is apparently related to the neurophysiological mechanisms underlying the accomplishment of the various motor tasks, with less effective results for the abilities requiring a higher degree of binocularity. These results are in favour of a rapid beneficial effect of late surgery, even in the absence of a documented benefit in binocular vision. This might be of relevance not only to help clinical decisional processes, but also to better shape the boundaries of health programs and insurance policies in this controversial matter. Studies with a longer follow-up are needed to explore the possible presence of further significant improvements, which might be expected after the rapid changes observed in the short term.

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