An Exploratory Study of the Kinematics of Girls with Turner Syndrome in a Visuo-motor Task

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In this paper a study is reported in which a group of children with Turner Syndrome (TS) is compared on motor performance tests and real-time kinematic data with a control group. The objective of this study was to identify possible kinematic variables that characterize the movement patterns utilized by this group of children in achieving their optimal performance. The underlying idea is that by comparing test results and movement kinematics one might gain more insight into the movement production of children than by just looking at the test results or by just using clinical movement observation. The children performed a pen and paper task, the flower trail (one of the sub-tests of the Movement ABC) (Henderson and Sugden, 1992), on an XY-tablet. We explored kinematic variables, such as trajectory length, movement time, velocity and pen pressure. It was found that, although girls with TS have a severe motor deficit, they are able to draw a line accurately between two narrow boundaries just as well as the control children (no significant difference in mean number of spatial errors). However, quantitative analysis revealed that girls with TS made a large amount of deviations from the optimal path such that their trajectory length increased with more than 10% per flower trail compared to the control group children. They also made more pen lifts and more changes in velocity. The study showed that children from the two groups may reach the same basic accuracy performance level by using very different strategies. Kinematic registration of fine motor tasks holds promise in gaining more
Although Turner Syndrome (TS) has a high incidence (one in 2000–5000 live born females; Rosenfeld, 1990) and girls with this syndrome show an overall low motor performance (Salbenblatt et al., 1989; Nijhuis-van der Sanden et al., 2000) to date little theory-driven research is available to found specific movement therapy programs on for these children. Turner Syndrome is a chromosomal disorder in which the second sex chromosome is missing (XO). All children with Turner Syndrome have a female phenotype. Although the most common karyotype in girls with Turner Syndrome is 45,X there are also many girls who have mosaic chromosome constitutions (45,X/46,XX or 45,X/46,XY). Turner Syndrome in its full expression consists of short stature, streak gonads, short or webbed neck, low posterior hairline, shield chest, bicuspid aorta valve, co-arctation of the aorta and renal anomalies, hypoplastic nails and short metacarpals. Turner Syndrome, however, is not always accompanied by distinctive features and is most often not diagnosed in infancy but later in childhood (4–5 years of age), primarily because of short stature. Other prominent features in teenage years are delayed puberty and delayed menarche, and in adult women anovulation and infertility.

If children with TS are referred for movement therapy, norm-referenced tests are commonly used to quantify their motor performance compared to their peers. However, the same poor results on a test can be obtained in different ways, e.g. by slowing down, by breaking up a movement, by being less accurate or by using more force. The neuromotor system continuously adapts itself to cope with internal and external constraints especially during motor development (Edelman, 1987; Merzenich et al., 1993). The existence of multiple solutions for attaining a given output is a challenge to the clinician. On the other hand, it shows the limitation of using a norm-referenced test, since it gives no information about the kind of movement solution the children explored or the way the motor system handles the disease-related constraints. Although studies using norm-reference tests indicate that Turner Syndrome girls have difficulties in motor functioning (Salbenblatt et al., 1989; Nijhuis et al., 2000) to date no investigation on the kinematics of their movements has been reported.

In this paper an exploratory study is reported in which one specifically chosen item of a standardized test is performed on an XY digitizer to search for possible differences in trajectory formation in children with TS compared to their controls. Many authors report decreased visuo-spatial processing abilities in girls with TS (Downey et al., 1991; Money, 1993; Rovet, 1993; Temple and Carney, 1995; Ross et al., 1996). Since the Movement Assessment Battery for Children (M-ABC) (Henderson and Sugden, 1992) is the most frequently used test for children with mild movement disorders (Geuze et al., 2001), one of the visual motor tasks of this test, the flower trail, could be a good choice for a first kinematic analysis. The flower trail has high spatial constraints. The trajectory and distance travelled in the task are constrained by the figure boundaries (see Figure 1a). Since no time limit is set for the task, children can be expected to show their most accurate performance. This task was chosen because it tests various fine motor abilities like movement planning and precise visually guided trajectory
formation. Also by its form, the flower trail gives adequate testing opportunity as it consists of two large curved parts demanding gradual steering while moving between the lines and eight straight lines with seven acute angles, asking for correct decelerating and accelerating abilities. An advantage of the task is that it resembles graphic tasks that children use while playing or while in school and that it has normative data for large populations (Henderson and Sugden, 1992), including for the Dutch on which the present study was performed (Smits-Engelsman, 1998).

In addition to the 10 flower trails, the full age-appropriate M-ABC and a norm referenced performance test for cursive handwriting were administered to the children in this study. The underlying idea of this study was that by comparing test results (on overall motor performance and writing performance) and movement kinematics, one might gain more insight into the movement production of children with TS than by just looking at the motor performance test results or by just using clinical movement observation. Comparing test performance and the kinematic profiles of girls with TS to peers without motor problems, is a way to look at adaptive solutions of the non-optimal movement system. Furthermore, kinematic analysis, in addition to performance tests, may help to develop effective training programs for girls with TS like it did for other diagnostic groups (Smits-Engelsman et al., 1997, 2001).

Fig. 1. (a) Flower trail made by one of the girls with TS. (b) Enlarged part with acute angles. (c) Flower trail as recorded on the XY-digitizer with Oasis software (De Jong et al., 1996).
METHOD

Subjects
A total of 36 children aged 7–12 years took part in the study. These children belonged to two groups: children with TS (n = 12) and a control group of 12 boys and 12 girls (n = 24). Informed consent was obtained from all participants and their parents.

Children with Turner Syndrome (TS)
Twelve children with TS (mean age 9.8 years, range 7.0–12.4 years) participated in the study. Subjects were recruited from the TS-patients in the paediatric endocrine clinic of the University Hospital Nijmegen St Radboud. The TS-karyotype is diagnosed by cytogenetic studies on cultured peripheral lymphocytes according to routine procedures. From each patient at least 30 GTG-banded metaphases were studied to rule out any (low-grade) mosaicism. The karyotype was 7 times 45XO and 5 times mosaic. None of the girls was treated with oestrogen, only with non-biologic grow-hormones. Normal IQs were found in this group (Mean Full scale IQ 92.9, S.D. 8.9; Verbal IQ 94.0, S.D. 7.6; Performance IQ 93.2, S.D. 13.2).

Control children
Twelve boys and 12 girls matched for age (mean age 9.6, range 7.7–12.0 years) to the diagnostic groups were selected from regular elementary schools in the vicinity of the University. The children had no known problems in overall academic achievement or health.

Measuring Instruments
Flower trail
One of the manual dexterity items of the M-ABC for children between 7 and 12 years of age is the flower trail. For different age groups the same figure is used with different width of the trail (2, 1.5 and 1 mm for 7–8, 9–10 and 11–12+ years of age, respectively). To make a kinematic comparison possible the flower-trail width meant for 7- and 8-year-old children was chosen for the dynamic recording for children of all ages. The children were instructed to draw a line between the two solid lines of the flower trail as accurately as possible, choosing their convenient drawing speed without lifting the pen. It is hypothesized that the freely chosen speed might be the optimal speed. All children had to complete 10 flower trails after practice. The child was allowed to draw the line in either the clock or anti-clockwise direction. A spatial error is defined as in the manual, by crossing the outline of the figure or lifting the pen without making continuous the pen trajectory (Henderson and Sugden, 1992).

The Movement Assessment Battery for Children
The Movement Assessment Battery for Children (M-ABC) was developed by Henderson and Sugden (1992) and validated for the Dutch population by Smits-Engelsman (1998). This instrument was developed to provide an indication of a child’s everyday-life motor function. Research has shown that the M-ABC is a very sensitive instrument for identifying children with motor difficulties (Henderson and Hall, 1982; Sugden and Wann, 1987; Henderson et al., 1989;
Smits-Engelsman, 1998). The test consists of four age bands of eight items that measure different aspects of motor ability; three items measure manual dexterity, two items measure ball skills and three items measure static and dynamic equilibrium (see Table 1). Children can score between 0 and 5 on each item and total scores will vary from 0 (very good) to 40 (extremely impaired). The total scores can also be translated to percentile scores that show the child’s level of performance in comparison with the child’s peers. The test has an acceptable validity and reliability (Henderson and Hall, 1982; Lam and Henderson, 1987).

The M-ABC was administered by an experienced paediatric physician to collect normative data for the Turner group. Part of the Control Children were also tested (n = 13), although reference data are available for the Dutch population (n = 580) (Smits-Engelsman, 1998).

Table 1. Items of the Movement-ABC

<table>
<thead>
<tr>
<th>Manual dexterity</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Speed task for both hands separately</td>
</tr>
<tr>
<td>2 Bi-manual coordination</td>
</tr>
<tr>
<td>3 Eye-hand coordination with preferred hand (drawing with preferred hand)</td>
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</tbody>
</table>

<table>
<thead>
<tr>
<th>Ball skills</th>
</tr>
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<tbody>
<tr>
<td>4 Catching moving object</td>
</tr>
<tr>
<td>5 Aiming at goal</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Balance</th>
</tr>
</thead>
<tbody>
<tr>
<td>6 Static balance (stance)</td>
</tr>
<tr>
<td>7 Dynamic balance while moving fast (jumping and hopping)</td>
</tr>
<tr>
<td>8 Dynamic balance while moving slow (walking)</td>
</tr>
</tbody>
</table>

The Concise Assessment Method for Children’s Handwriting

The Concise Assessment Method for Children’s Handwriting (CAMCH) by Hamstra-Bletz et al. (1987) is a screening tool to recognize poor handwriting or dysgraphia on the basis of a completed piece of handwriting. This writing task consists of copying a standard text in 5 min or at least the first five lines if the child is a very slow writer. The standard text gradually increases in difficulty as the text proceeds, while at the same time the size of the print decreases for each consecutive paragraph. The first five sentences have a degree of difficulty equal to a grade 1 level reading exercise (Hamstra-Bletz, 1993). The text is to be copied on unruled paper and is not read by the child before it starts coping the text. Handwriting quality is evaluated using the dysgraphia scale of the CAMCH. This scale is based on the assessment of 13 dysgraphia features, i.e. deviations from the standard hand or writing course. The first two items are scored on the basis of the entire written work. Both items are measured on an ordinal scale with six categories resulting in a score from 0 to 5. For the remaining 11 items, the first five sentences are scored in terms of whether or not a particular feature is present in that sentence. A score of 0 is given if the dysgraphic feature is absent. The total possible score for a feature is 5. Each child’s total score for all 13 items is then used to determine if the child is dysgraphic. The manual provides the following norms: (a) not dysgraphic: a score from 0 to 21; (b) ambiguous dysgraphic or poor writer: a score from 22 to 28; (c) dysgraphic or very poor writer: a score of 29 or higher.
To determine copying speed the number of letters written by the child in the first 5 min, including corrections, are counted. This score is translated to a decile score scaled to the norm for the child’s grade. Hamstra-Bletz (1993) defines as slow writers, children with scores in the first or second decile while those with scores in the eight to tenth decile are said to be fast writer. Hamstra-Bletz (1993) reports satisfactory results regarding inter-rater reliability on the items ($r = 0.71–0.89$) and intra-rater reliability for grades 2 ($r = 0.87–0.94$) and 3 ($r = 0.79–0.88$). The CAMCH is used in this study to determine the level of writing performance.

APPARATUS

Data was gathered using a Wacom UD-1218-RE digitiser with a wireless inking pen and the OASIS software (De Jong et al., 1996) that was running on a PC. The equipment recorded the position of the pen tip, and the force exerted along the axis of the pen at a rate of 206 times a second and a spatial accuracy of 0.2 mm (see Figure 1c).

PROCEDURE

The children with TS were tested on the M-ABC (Henderson and Sugden, 1992; Smits-Engelsman, 1998). Part of the Control children were administered the M-ABC as well. All children were administered the CAMCH except for two children, who would not write cursive (one Control Boy and one Girl with TS).

For the experimental tasks, all the children ($n = 36$) were seated at a table with an XY digitiser. Prior to the experiment, each child was given the opportunity to practice in order to make sure (s)he understood the task. The subjects were instructed to draw the figures as accurately as possible on a sheet of paper attached to the XY digitizer. After the child had drawn three flowers a new paper was attached to the digitizer.

DATA ANALYSIS AND DESIGN

Signal analysis

The drawn flower trails were automatically pre-segmented. The first and last segments were deleted because of the large variation in trajectory length due to differences in the start and end points (no exact beginning or end point is defined in the task). The remaining segments were subsequently checked for movement artefacts. These segments were excluded from the analysis (<1% of the total data). For each trail, values of a number of dependent variables were generated: trajectory length, movement time, velocity peaks, pen pressure, number and time of the pen lifts.

Data analysis

To search for gender and group differences the data were compared on the movement performance variables (spatial errors in the flower trail, M-ABC, writing speed, writing quality) and kinematic variables by using contrast analyses $t$-tests (corrected for equality of means) and alpha was set at 0.05. The contrasts were used to determine differences between Control Boys and Control
Results

Performance Measures

Differences between groups in performance on the experimental flower trails
Mean number of spatial errors (S.D. in brackets) in ten flowers trails were 20.3 (16.9) for Control Boys, 10.1 (9.9) for Control Girls and 17.3 (13.5) for children with TS, respectively (Table 2). These differences, although large, were not significant ($p = 0.078$) due to large standard deviations in all groups.

Differences between groups on motor performance
No differences on any of the Movement Performance variables were found between Control Boys and Control Girls. As a group, the Control Group scored close to the 50th percentile of the reference group on the M-ABC (means for the Control Group 48th, Control Boys 41th and Control Girls 55th percentile).

In contrast, none of the children with TS scored within the normal range on the M-ABC. The mean score on the M-ABC for the children with TS was two standard deviations below the norm score for their age group (2nd percentile) indicating a severe motor impairment. They performed significantly worse compared to the Control Girls and the Control Group, respectively, on all variables: Manual Dexterity ($t = -3.76, p < 0.001$; $t = -4.11, p < 0.001$), Ball skills ($t = -2.04, p < 0.05; t = -3.35, p < 0.05$) Balance ($t = -4.93, p < 0.001; t = -5.58, p < 0.00001$) and Total Score ($t = -4.80, p < 0.001, t = -5.93, p < 0.00001$). Table 3 presents the means and standard deviation for each group.

Table 2. Means and standard deviations of the Writing Performance, including spatial errors on the flower trail for the groups. Significant different values are printed bold

<table>
<thead>
<tr>
<th>Writing and drawing performance</th>
<th>Turner girls ($n = 12$)</th>
<th>Control boys ($n = 12$)</th>
<th>Control girls ($n = 12$)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Errors in 10 flower trails</td>
<td>17.3 (13.5)</td>
<td>20.3 (16.9)</td>
<td>10.1 (9.9)</td>
</tr>
<tr>
<td>Writing quality CAMCH</td>
<td>14.7 (3.8)</td>
<td>12.8 (3.6)</td>
<td>13.8 (3.6)</td>
</tr>
<tr>
<td>Writing speed CAMCH (decile)</td>
<td>7.0 (2.6)</td>
<td>5.3 (3.5)</td>
<td>7.7 (2.1)</td>
</tr>
</tbody>
</table>

Table 3. Means and standard deviations of the Motor Performance for the groups. Significant different values are printed bold

<table>
<thead>
<tr>
<th>Motor performance</th>
<th>Turner girls ($n = 12$)</th>
<th>Control boys ($n = 7$)</th>
<th>Control girls ($n = 6$)</th>
<th>Dutch reference group ($n = 580$)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Manual dexterity</td>
<td>8.2 (4.0)</td>
<td>3.6 (2.1)</td>
<td>2.1 (2.0)</td>
<td>2.1 (2.6)</td>
</tr>
<tr>
<td>Ball Skills</td>
<td>5.6 (3.5)</td>
<td>0.0 (0.0)</td>
<td>2.5 (3.7)</td>
<td>1.4 (1.8)</td>
</tr>
<tr>
<td>Balance</td>
<td>8.3 (3.2)</td>
<td>2.6 (2.8)</td>
<td>1.3 (2.0)</td>
<td>1.3 (2.0)</td>
</tr>
<tr>
<td>Total score MABC</td>
<td>22.17 (7.8)</td>
<td>6.2 (3.7)</td>
<td>5.9 (7.1)</td>
<td>4.5 (6.0)</td>
</tr>
<tr>
<td>Percentile MABC</td>
<td>2.4 (2.4)</td>
<td>41.3 (29.8)</td>
<td>54.8 (29.8)</td>
<td>50.0 (34.0)</td>
</tr>
</tbody>
</table>

Girls, between Children with TS and Controls, and between Girls with TS and Control Girls. For the comparison on the M-ABC, also the Dutch reference data were available.
Since group data obscure individual differences between subjects, we also report how many children in the two groups really deviated from the norm values (defined as below the 15th percentile). From the 13 Control Children that were administered the M-ABC, two scored below the 15th percentile on manual dexterity, one on ball skills, one on balance and one on the total score. None of the TS girls scored above the 15th percentile on the total score. Nine scored below the 15th percentile of manual dexterity, 10 on ball skills, and 10 on balance.

**Differences between groups on writing performance**

Control Girls produced more letters in 5 min (7.7 decile) than the Control Boys (5.3 decile) \((t = -2.11, p < 0.05)\). However, they did not differ on the writing quality of the CAMCH, indicating that although the girls were faster than the boys, they produced equal quality script (see Table 2).

The children with TS did not score differently as compared to the Control Group on the writing quality and writing speed of the CAMCH. This shows that although the girls with TS show a severe overall motor impairment, they are able to produce a good quality script at normal speed. Looking at individual differences revealed that one of the Control Boys and one of the girls with TS scored in the ambiguous dysgraphic range.

**Preliminary conclusion based on the results of the performance measures**

No differences between the groups were found on the number of times that the children in the three groups crossed the boundaries of the flower trail. However, on the bases of results on the MABC it can be stated that children with TS have a severe motor deficit. On the sub score of manual dexterity nine of the 12 children score below the 15th percentile. Nevertheless, they are able to meet the task requirements in writing (CAMCH) and in drawing within boundaries.

**KINEMATIC MOVEMENT ANALYSIS OF THE FLOWER TRAIL**

No significant differences emerged on the kinematic variables between Control Boys and Control Girls. Like in the CAMCH, the girls tended to move faster but this difference did not reach the level of significance \((p = 0.09)\).

Children with TS, however, moved differently from the Controls (Table 4). Without crossing the movement boundaries more often, they made 10% longer movement trajectories than the Controls \(\text{‘trajectory length’, } \(t = -2.26, p < 0.05)\). They also lifted the pen more frequently \(\text{‘number of lifts’, } \(t = -2.47, p < 0.05)\) and longer off the paper than the Control Children \(\text{‘lift time, } \(t = -2.81, p < 0.01)\).

**Table 4.** Means and standard deviations of the Kinematic Variables of the flower trail for the groups. Significant different values are printed bold

<table>
<thead>
<tr>
<th>Kinematic variables</th>
<th>Turner girls ((n=12))</th>
<th>Control boys ((n = 12))</th>
<th>Control girls ((n = 12))</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trajectory length (cm)</td>
<td>39.3 (6.8)</td>
<td>36.1 (4.9)</td>
<td>34.5 (1.8)</td>
</tr>
<tr>
<td>Movement time (s)</td>
<td>38.6 (12.1)</td>
<td>39.8 (11.2)</td>
<td>32.3 (7.3)</td>
</tr>
<tr>
<td>Number of lifts</td>
<td>4.0 (3.6)</td>
<td>1.9 (2.5)</td>
<td>1.4 (0.7)</td>
</tr>
<tr>
<td>Lift time (s)</td>
<td>3.8 (3.7)</td>
<td>1.5 (2.1)</td>
<td>1.1 (0.9)</td>
</tr>
<tr>
<td>Pen pressure (N)</td>
<td>1.72 (0.7)</td>
<td>2.29 (0.6)</td>
<td>2.17 (0.9)</td>
</tr>
<tr>
<td>Velocity peaks (per s)</td>
<td>6.9 (0.6)</td>
<td>5.9 (0.5)</td>
<td>6.0 (0.6)</td>
</tr>
</tbody>
</table>

They used more changes in velocity per second to accomplish the task (‘velocity peaks’, $t = -4.01$, $p < 0.001$). Girls with TS tended to used less pen pressure ($p = 0.06$) while moving through the trail (‘pen pressure’).

Overall in can be stated that the movement trajectories executed by the Girls with TS were more irregular (see Figure 1b) and segmented. On account of the increased number of pen lifts, it is important to notice that these lifts were not detectable on the test sheets. According to the manual of the M-ABC, pen lifts should only be counted as mistakes if the writing trajectory is not continued at the same place. By just looking at the test sheets these pen lifts would have been missed.

DISCUSSION AND CONCLUSIONS

In this paper a study is reported in which a group of children with Turner Syndrome (TS) was compared with an aged-matched control group of boys and girls. A comparison was made on fine motor performance data like drawing and writing, on an overall motor performance test and on kinematic data collected with an XY-tablet during the performance of a pen and paper task, the flower trail. The results show that, although girls with TS have a severe motor deficit, they are able to draw a line accurately between two narrow boundaries just as well as the control children. Hence, in this study it is shown that children can perform rather well under one specific constraint (accuracy in the flower trail) and very poorly in another situation (general motor performance). These results indicate that the motor system, albeit hampered by non-optimal function, still has opportunities to achieve good performance on a visuo-motor task like the flower trail. The most important finding is that the group of children with pathology used a different movement pattern or strategy to accomplish the same task performance as the control children.

One objective of this paper was to identify possible differences in movement patterns on the basis of quantitative measures. We explored whether the dependent kinematic variables, such as trajectory length, movement time, velocity, velocity peaks and pen pressure, were significantly different between the groups of children and if these data provide additional information about the motor solution the children used as compared to test performance results or observation. The study shows that control boys and girls did not differ on their movement profile. However, children with internal constraints like TS have their own, significantly different, way to perform the task, despite similarities in total outcome of the test item.

Girls with TS were able to meet the same scores on spatial mistakes in the flower trail as the controls by frequently correcting their movement and through lifting the pen more often, so they would not cross the boundary of the trail. From a motor control point of view, the task should be seen as a two-dimensional accuracy task under visual control allowing iterative correction during the movement. One of the strategies normally used by children to reduce errors and to keep spatial variability of movements within the limits of task requirements is to move slower. Girls with TS, however, tend to use segmentation of movements since they built the line from many different small curved pieces. A possible explanation for the segmentation strategy is that it results from freezing of one or more joints. This leads to piecemeal trajectory formation, lifting and to more curved trajectories but it makes movement processing easier (Bernstein, 1967). Similar segmentation has been found in other studies on motor impaired children. In children with cerebral palsy, Eliasson et al. (1991) found segmentation...
in a study on arm co-ordination. They attributed this segmentation to a deficit in motor programming. The children in their study showed an impaired capability to develop motor programs and to program the features of the movement in advance, thereby causing typical pauses between the subsequent parts of a movement pattern.

Overall, the present data clearly illustrate that motor competence is not a unitary construct (e.g. Fleishman, 1966; Smits-Engelsman et al., 1998). In the present study it was observed that general manual dexterity or gross motor functions are not prerequisites for good performance on grapho-motor tasks. All of the girls with TS had impaired overall motor function and nine showed decreased performance on the manual dexterity items yet they wrote legibly within a normal time limit.

In summary, the fact that equal performance in terms of spatial errors in a norm-referenced item of the M-ABC can be obtained despite large differences in kinematic data reveals the risk of looking at performance measures alone. In addition, the kinematic analysis provides insight into differences in movement solutions in movement tasks. These insights may prove useful in the future beyond the borders of the laboratory in developing training programs (Smits-Engelsman, 1995).

In conclusion, kinematic registration of fine motor tasks holds promise in gaining more insight into how clinical groups move, especially under circumstances where performance levels fail to discriminate. Experimental manipulations of likely constraints (speed, accuracy, parallel processing) can be a fruitful way to make progress towards theory-driven training programs for different diagnostic groups.

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