Handwriting difficulties in juvenile idiopathic arthritis: a pilot study

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Abstract

Objective

The aim of the present study was to describe handwriting difficulties of primary school children with juvenile idiopathic arthritis (JIA), and to investigate possible correlations with hand function and writing performance.

Methods

In a cross-sectional approach, 15 children with JIA and reported handwriting difficulties were included together with 15 healthy matched controls. Impairments (signs of arthritis or tenosynovitis, reduced grip force and limited range of motion of the wrist (wrist-ROM)), activity limitations (reduced quality and speed of handwriting, pain during handwriting), and participation restrictions (perceived handwriting difficulties at school) were assessed and analysed.

Results

Although selected by the presence of handwriting difficulties, the majority of the JIA children (73%) had no active arthritis of the writing hand, and only minor hand impairments were found. Overall, the JIA children performed well during the short handwriting test, but the number of letters they wrote per minute decreased significantly during the 5-minute test, compared to the healthy controls. JIA patients had significantly higher pain scores on a 100 mm Visual Analogue Scale, compared to the healthy controls. The actual presence of arthritis, and limitation in grip force and wrist-ROM did not correlate with reported participation restrictions with regard to handwriting at school. The JIA children reported pain during handwriting, and inability to sustain handwriting for a longer period of time.

Conclusions

The results of this pilot study show that JIA children with handwriting difficulties, experience their restrictions mainly through pain and the inability to sustain handwriting for a longer period of time.
No correlations could be found with impairments.

Key words

Juvenile idiopathic arthritis, hand, handwriting, rehabilitation

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Introduction

Juvenile Idiopathic Arthritis (JIA) is an umbrella term, encompassing all forms of arthritis with unknown aetiology, with an onset of the disease before the age of 16 years, and symptoms persisting for at least 6 weeks (1).

Involvement of the upper extremity, and particularly the wrist, is among the early signs of a poor outcome for children with JIA, and special attention is required in the treatment of these patients (2). Due to arthritis, these children may suffer from pain and limited range of motion of the wrist or finger joints, in combination with reduced grip force (3, 4). Children with JIA often report difficulties in handwriting as a major restriction at school, and complain of 'slow, messy and painful' handwriting (5). For a satisfactory participation at school, adequate handwriting abilities are known to be an important factor (6). So far, however, there is limited knowledge about the actual handwriting performance and specific handwriting difficulties that children with JIA experience at school. While one might assume a straightforward relationship between handwriting difficulties and arthritis of the upper extremity, this has not yet been studied. In order to provide these children with optimal treatment, a clear understanding of this relationship is necessary.

The International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) (7) is a framework that describes the domains of body function and structure, activities, and participation. It is widely used in the rehabilitation of children for assessment, goal-setting, treatment planning and evaluation. We applied the ICF-CY to the handwriting difficulties of children with JIA and formulated the following research questions:

1) At the level of function and structure: Which impairments (*i.e.* arthritis, limited range of motion or reduced grip force) are present in JIA children with handwriting difficulties?
2) At the level of activities: Which specific limitations in activity (*i.e.* reduced quality and speed of handwriting, or pain during handwriting) can be observed in children with JIA?
3) At the level of participation (school): Which specific restrictions in handwriting do children with JIA report?
4) Which relationships can be found between impairments, activity limitations and participation restrictions?

Patients and methods

Setting and participants

Patients were recruited from Reade (formerly Jan van Breemen Instituut) in Amsterdam, which is a large clinic for rehabilitation and rheumatology in the Netherlands. The inclusion criteria were: 1) diagnosis of JIA, according to the International League of Associations for Rheumatology (ILAR) criteria (4), 2) self-reported handwriting difficulties, 3) at least one year learning to write at school, and 4) attending a regular Dutch primary school. The exclusion criteria were: other medical conditions which can interfere with handwriting. For this pilot study, 15 children referred to occupational therapy for treatment for their handwriting difficulties, who met the inclusion criteria, were selected. The control group consisted of 15 healthy children, matched with the patient group with regard to age, gender, writing left or right-handed, and school level (8). Eleven of the 15 children were in the same class at school as matched patients, and four were recruited from other local schools.

Disease history and clinical characteristics, including data on disease duration, age at onset of the disease, disease activity, current active joint count, medication, and ILAR classification, were collected from the medical charts. Disease activity was assessed on a 100 mm Visual Analogue Scale (VAS-doctor) (0 mm=inactive, 100 mm=extremely active), and a 5-point Likert scale (Likert-doctor) (0=none, 1=mild, 2=moderate, 3=severe, 4=extremely severe). We assessed 0–71 joints for clinical active arthritis (9), and previous involvement of joints of the dominant upper limb, including tenosynovitis, was recorded.

The data-collection was carried out between May 2009 and September 2009. The study protocol was approved by the Medical Ethics Committee of our institute, and all children and their parents gave written informed consent.

Impairments

– Assessment of hand function

In the JIA children, the hand and wrist of the writing hand were assessed for signs of active arthritis or tenosynovitis (*i.e.* swelling and pain).

The range of motion of the wrist (wrist-ROM) was measured on the dorsal side of the hand with a hand-held goniometer (E-LINK R500 Range of Motion Kit, Biometrics Ltd). Manual goniometry has shown high intra-rater reliability (10). All children were tested by one tester (HH).

Maximal isometric grip force was measured with a JAMAR-dynamometer (E-LINK H500 Hand Kit, Biometrics Ltd), and counted in Newtons (N). The children were asked to grip as strongly as possible three times. The average of the three grips and the maximum value per child were used for further analysis. The average value was compared with published norm data (11): a score was considered abnormal if is this was below the mean norm value, minus two standard deviation (SD).

Activity limitations

– Handwriting assessment

For this assessment, the children were seated at a standard table with a horizontal surface, and both the chair and the table were adjusted to the anthropometrics of each individual child. A Dutch method for the assessment of children's handwriting ("Beknopte beoordelingsmethode voor kinderhandschriften = BHK) was applied (12). The BHK is used to identify handwriting problems in primary school children. The writing task consisted of copying a standard text that was printed on a card, within five minutes, on a sheet of A-4 paper fixed to the surface of an x-y digitising tablet (WACOM Intuos GD-0912-R). The children wrote with a wireless electronic inking pen with a force-sensitive tip (Intuos inking pen), which is similar in size and weight to a normal pen. For the assessment, 13 handwriting characteristics, such as insufficient word-spacing, acute turns in connecting letters, irregularities in connections, or their absence, and collisions of letters, were scored with the BHK (12). The total BHK score was used to determine whether the child was 1) not dysgraphic (score 0-21), 2) at risk (score 22–28), or 3) dysgraphic (score 29 or higher). Writing in print was also scored, although this test is generally intended for cursive handwriting. In block-letter handwriting, the worst possible score for the connection between letters was 5 points. Writing speed was determined by counting the number of letters produced in exactly five minutes. The scores were then converted into decile scores related to norm values, corrected for the children's grades. Scores falling within the lower two deciles were considered to reflect slow writers, and within the upper two deciles, fast writers (12). Inter-rater reliability of the BHK has been reported to vary between r=0.71 and r=0.89; intra-rater reliability was r=0.79 to r=0.94 (12). In the present study, the handwriting products were all assessed by one tester experienced in handwriting assessment (HH). The BHK is commonly used for scientific and clinical purpose (13, 14).

The number of letters written per minute was also counted. These values provide information about the development of the number of letters written in time. The functionality of the pencil grip was also evaluated during handwriting. The dynamic tripod, lateral tripod, dynamic quadrupod, lateral quadrupod and four-finger pencil grips were all considered as functional pencil grips (15).

– Pain assessment

The children were asked to rate the upper extremity pain they experienced on a 100 mm VAS-pain (0 mm = no pain, 100 mm = extremely severe pain). End-points were a happy smiley face and a sad face. Pain was rated before and after the handwriting task, and also after the grip force measurements.

Participation restrictions

– Questionnaire

The children were asked in a structured way about handwriting difficulties at school, and their use of handwriting aids (*e.g.* a laptop or an adapted pencil). The questions were designed following the formulation used in the Dutch version of the Childhood Health Assessment Questionnaire (CHAQ), which is available as a child-administered version from 8 years on (16). Children were asked to rate their answers on a 100 mm VAS, which is possible because from 8 years on, children are expected to understand the concept of rating a VAS (17). We asked questions about following handwriting features: 1) frequency of their handwriting difficulties, 2) severity of their handwriting difficulties 3) frequency of difficulties in finishing handwriting tasks on time, 4) severity of their difficulties in finishing handwriting tasks on time, 5) appearance of their handwriting, 6) frequency of pain during handwriting, 7) severity of pain during handwriting, and 8) frequency of their difficulties with sustaining handwriting throughout the day. A higher score indicated a worse situation. End-points were marked with a happy smiley face and a sad face. The values of the second question, about the severity of handwriting difficulties at school were used for the analyses of correlations between reported handwriting difficulties and hand function (arthritis, ROM and grip force).

Statistics

All statistical analyses were performed in SPSS 15.0. Descriptive statistics were expressed as means with SDs. Ordinal scales (*e.g.* handwriting questionnaires, functional pencil grip, BHK scores) were tested with the nonparametric Mann-Whitney U-test. Wrist-ROM and grip force measurements were analysed with t-tests. The three measurement values of the VAS-pain (before handwriting, after handwriting, after grip force measurement), were analysed with repeated measurements ANOVA, with age as covariate. The same procedure was performed to compare the groups with regard to number of the letters written per minute. The within-factor was set for each minute, and the group difference, time difference and the interaction effect of group with time were calculated. Correlations were defined via rank order correlations (Spearman's rho) for ordinal scales and the Pearson's correlation coefficient for interval and ratio scales. The increase

or decrease in number of letters written per minute was defined by the slope of linear regression through the datapoints. One missing wrist-ROM measurement of a JIA child and one missing VAS-pain measurement of a child from the control group were imputed, using expectation maximisation (EM). Significance set at two-sided *p*-values below 5% ($p<0.05$).

Results

Study population

Each group consisted of 13 girls and 2 boys, and 13 right-handed and 2 left-handed children. The JIA group had a mean age ± SD of 10.8±1.5 (range 8.0-13.1) years and did not differ from the control group, which had a mean age ± SD of 10.7±1.5 (range 7.9-12.9) years, (for details of the clinical features of the JIA children, see Table I). All JIA children were on tight disease control schedules and, overall, they had mild disease activity. They also had all had clinical arthritis of the upper extremity joints, including hand or wrist joints, during the course of the disease.

Table I. Demographics and clinical features of the 15 children with JIA.

	no. (%)	mean ± SD	range
Gender: female	13 (87)		
ILAR category			
RF-negative polyarthritis	8 (53)		
Extended oligoarthritis	7 (47)		
Age at disease onset in years		6.5 ± 3.3	1.4–10.2
Age on study inclusion in years		10.8 ± 1.4	8–13.1
Disease duration, years		4.3 ± 3.2	0.3–9.8
Disease activity (Likert scale)			
0=none	2 (13)		
1=mild	10 (67)		
2=moderate	2 (13)		
3=severe	1 (8)		
4=extremely severe	0 (0)		
Disease activity[$]		24 ± 16	0–64
Active joint count			
0–4	10 (67)		
5–10	4 (27)		
>10	1 (7)		
Current medication			
NSAID	15 (100)		
Methotrexate	15 (100)		
Etanercept	4 (27)		
Active clinical arthritis of upper extremity joints during the course of the disease	15 (100)		

[$]on 100 mm visual analogue scale; 0: no activity; 100: extremely active.
JIA: juvenile idiopathic arthritis; no.: number; %: percentage; SD: Standard Deviation; ILAR: International League of Associations for Rheumatology; RF: Rheumatoid factor; NSAID: non-steroidal anti-inflammatory drugs.

Impairments

– Hand function

On inclusion, four patients (27%) had current arthritis and/or tenosynovitis of the writing hand. In a comparison of the wrist-ROM between the JIA patients and the controls, it was found that wrist flexion was significantly decreased in the patient group, but that there was no difference in wrist extension between the two groups.

There was no significant difference in isometric grip force between the two groups, but there was a trend towards reduced grip strength in the JIA group. The average isometric grip force values of 4 JIA patients (27%), compared to one control (7%), were lower than age and gender-related norm values (11). For more details concerning hand function, see Table II.

Table II. Impairments.

	Patients no. (%) or mean ± SD	range	Controls no. (%) or mean ± SD	range	Difference *p*-value
Arthritis and/or tenosynovitis of writing hand	4 (27%)		None (0%)		
Wrist ROM					
Wrist flexion (°)	50 ± 6	40–60	60 ± 9	43–60	0.03
Wrist extension (°)	58 ± 12	40–77	56 ± 12	33–78	0.742
Isometric grip force measurements					
Maximum value (N)	177 ± 49	118–275	196 ± 49	137–314	0.22
Average value of three trials (N)	167 ± 49	98–255	186 ± 49	128–304	0.15

SD: Standard Deviation; ROM: Range of Motion; N: Newton; °: degree.

Activity limitations

– Handwriting

There was no difference between the JIA children and the control children with regard to the BHK scores (*i.e.* quality of handwriting), handwriting speed, or functionality of pencil grip (for details see Table III).

With regard to the number of letters written (mean ± SD) in 5 minutes (JIA: 273±87; control: 258±92), no difference was found between the groups ($p=0.658$), but when analysing the influence of time, a significant interaction effect of time with group ($p=0.023$) was found. On average the control children started with fewer letters (mean ± SD in the minute: 50±19) and the number of letters increased, whereas the JIA children started with more letters (mean ± SD in the first minute: 57±19), but the number of letters decreased slightly during the 5-minute test. There was a clear decrease in the number of letters written per minute in 7 JIA children, compared to 1 control child (slope <-1) (Fig. 1).

– Pain

The JIA group rated their perceived pain significantly higher than the control

Table III. Activity limitations: handwriting and pain assessment.

	Patients no. or mean ± SD	Controls no. or mean ± SD	Difference *p*-value
BHK (not dysg. / at risk / dysg.)	(15 / 0 / 0)	(12 / 2 /1)	0.073
BHK writing speed (slow / normal / fast)	(1 / 6 / 8)	(0 / 8 / 7)	0.869
Pencil grip (functional/non functional)	(11 / 4)	(13 / 2)	0.369
VAS-pain before handwriting (mm)	17.7 ± 17.8	6.8 ± 13.2	
VAS-pain after handwriting (mm)	51.9 ± 24.6	33.2 ± 27.4	0.008*
VAS-pain after force measures(mm)	63.2 ± 24.2	38.4 ± 28.4	

*derived from repeated measurements ANOVA with age as covariate; BHK: concise assessment method for children's handwriting; dysg: dysgraphic; VAS: 100 mm visual analogue scale.

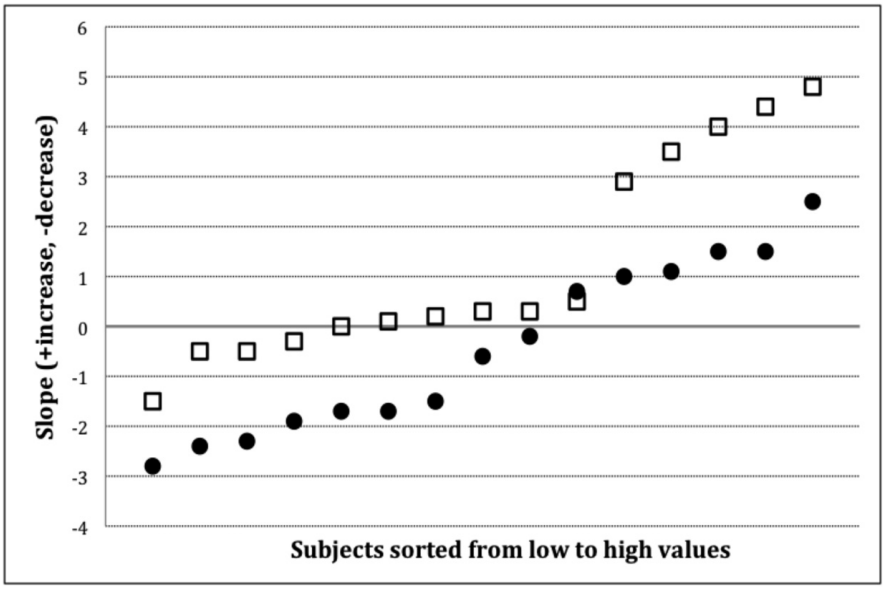

Fig. 1. Slope indicating letter written per minute during five minutes of handwriting.
In children with JIA there was a decrease of written letters per minute in a 5-minute handwriting test, and this was more pronounced in the patient group than in control group (reflected in the negative slopes). Although there were some children in the JIA group who increased their speed over time, these increases were smaller than in the control group.
●: patients □: controls.

Table IV. Participation restrictions at school: Results on 100 mm visual analogue scale scores of children on handwriting questions.

	Patients mean ± SD	Controls mean ± SD	Difference *p*-value
1) Frequency of handwriting difficulties	53 ± 20	15 ± 17	0.000
2) Severity of handwriting difficulties	63 ± 22	15 ± 18	0.000
3) Frequency of difficulties in finishing handwriting tasks on time	30 ± 25	20 ± 20	0.383
4) Severity of difficulties in finishing handwriting tasks on time	35 ± 31	18 ± 22	0.109
5) Appearance of handwriting	47 ± 33	33 ± 19	0.135
6) Frequency of pain during handwriting	59 ± 19	18 ± 18	0.000
7) Severity of pain during handwriting	58 ± 15	16 ± 15	0.000
8) Frequency of difficulties with sustaining handwriting throughout the day	56 ± 19	16 ± 18	0.000

A high score means a negative scoring of children; SD: Standard Deviation.

group (p=0.008) (see Table III). The increase in pain rating after handwriting and after grip force measurements was similar in both groups. Thus, no interaction effect of group with time was found for VAS-pain after handwriting (p=0.353), or for VAS-pain after grip force measurement (p=0.199).

Participation restrictions

The JIA children rated almost all restrictions in participation as high (mean ± SD: 57.5±19 mm), and significantly higher than their controls, except for 'finishing handwriting tasks on time' and 'appearance of their handwriting' (the means ± SDs and p-values are shown in Table IV). Fourteen JIA children reported that they used aids for handwriting (mainly adapted pencils and laptops).

Relationship between impairments, activity limitations and participation restrictions

In the JIA children, the severity of handwriting difficulties was not correlated to current arthritis or to tenosynovitis of the writing hand (Spearman's rho=0.087; p=0.757), nor to wrist flexion (Spearman's rho= -0.290; p=0.294) or maximum isometric grip force (Spearman's rho=0.197; p=0.482).

In both groups, pain after handwriting correlated with the number of letters written in 5 minutes (Pearson's correlation coefficient: 0.459; p=0.011), *i.e.* the more they wrote, the more pain they experienced. The correlation was found to be significant in the JIA group (Pearson's correlation coefficient: 0.779; p=0.001), but not in the control group (Pearson's correlation coefficient: 0.202; p=0.471) (Fig. 2).

Discussion

The present study focused on a group of JIA children who experienced handwriting difficulties at school.

Contrary to our expectations, more than two thirds of the JIA children who reported current handwriting difficulties showed no clinical signs of tenosynovitis or arthritis in any joint of the wrist or hand on clinical assessment. This result must be interpreted with care. By means of ultrasound it has been shown that arthritis in the wrist and small hand joints can be missed in clinical observations (18, 19). Overall disease activity in the JIA group was mild.

Results at the level of function and structure: The JIA children differed

slightly from the healthy controls in wrist flexion. This was unexpected, because a decrease in wrist extension, and not wrist flexion, is usually reported in JIA (20-22). The JIA children did not have significantly worse grip force, compared to the control group, but they had a tendency towards lower values, suggesting that a significant difference may not have been observed due to small sample size. We found that 27% of JIA children had reduced grip force, compared to age-related norm values (11).

Results at activity level: No differences were found in the quality of handwriting between the children with JIA and handwriting difficulties and the healthy controls. Moreover, the JIA children did not write less letters in 5 minutes time, and they even started with more letters per minute than their controls. When analysing the development in time, the number of letters written per minute decreased during the 5 minute test in 7 children in the JIA group, compared to 1 child in the control group. We can hypothesize that the observed decrease would continue after the test time of 5 minutes. Furthermore, the JIA children reported higher pain scores on a 100 mm VAS after the 5-minute handwriting test compared to the healthy controls. Therefore, it is questionable whether the strategy to start fast and to write as quickly as possible for a short period of time with a high pain level is appropriate in daily school situations.

Results at participation level: The frequency and severity of the pain during handwriting at school was rated high by the JIA children, and they also reported difficulties with sustaining handwriting throughout the day. At the same time, however, JIA children appreciated the appearance of their handwriting at the same level as the healthy controls. This is in line with the observed quality of the handwriting throughout the handwriting assessment.

We found no relationship between activity limitations and participation restrictions. Current arthritis in the writing hand (body function and structure) did not correlate with the severity of difficulties of handwriting at school (participation), but this can be a result of under-estimation of arthritis, as we discussed before. Furthermore, children with more severe impairments (*i.e.* less grip force or a greater decrease in wrist-ROM) did not report more participation restrictions (*i.e.* more handwriting difficulties at school). Therefore, grip strength and wrist-ROM seem to be less important for handwriting in JIA children.

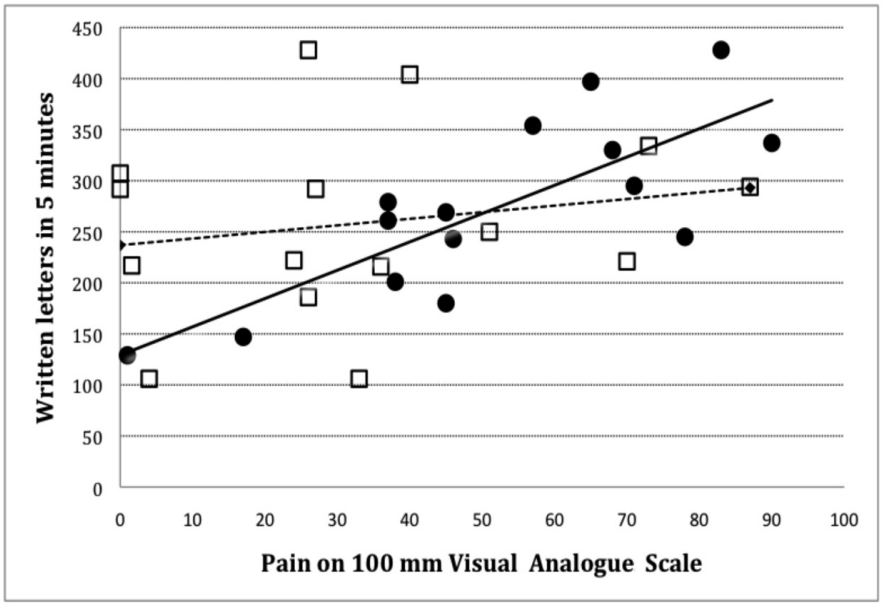

Fig. 2. Correlation between number of letters written and pain after handwriting.
The number of letters written by the children with JIA in 5 minutes was correlated with the pain they experienced after handwriting. This correlation did not exist in the control group.
●: patients □: controls; lines: linear regression.

In general, interventions should focus on participation restrictions that children experience at school. Advice on how to sustain handwriting throughout the day, and how to reduce pain during handwriting would seem to be beneficial for these children. Based on our results, suggestions could be: splitting handwriting tasks into small elements, taking breaks, writing slowly, allowing extra time for finishing writing tasks, providing a variety of writing utensils and, if necessary, providing adapted pencils, orthoses or a laptop. Of course, it is a prerequisite that the school and the teachers collaborate in this process.

Further research is required to fully understand the restrictions that children with JIA experience during handwriting at school and to assess the possibilities for intervention. Considering that the pain that JIA children experience seems to increase with the amount they write, their handwriting should be tested over a longer period of time. We limited our pilot study to primary school children, with only one measurement. It would therefore also be important to focus on how JIA children with handwriting difficulties fulfil their tasks in secondary school with an increasing amount of handwriting.

Conclusions

The results of this pilot study show that JIA children with handwriting difficulties experience their restrictions at school mainly because of pain and the inability to sustain handwriting for a longer period of time. The JIA children had high pain scores on a 100 mm VAS after 5 minutes of handwriting. Most of them started at a relatively high writing speed, but the number of letters written per minute decreased during the 5-minute test in half of the children. The handwriting difficulties were present both in children with and without clinical signs of active arthritis or tenosynovitis in the hand or wrist. No relationship was found between impairments, activity limitations and participation restrictions.

Interventions that focus on restrictions at school are needed for JIA children with handwriting difficulties.

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