

## Association of Handwriting Impairment in Elementary School-aged Children with Autism Spectrum Disorders

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**CLINICAL SCENARIO:** In the writer's school-based occupational therapy practice, students with autism spectrum disorders (ASD), who comprise a significant part of the caseload, are often referred for intervention because of handwriting challenges. Of the many articles on handwriting intervention for school aged students, including a recent systematic review (Hoy, Egan & Feder, 2011), the diagnostic category of ASD was not identified. This led the writer to wonder whether handwriting impairments are associated with ASD in school-aged children. This would substantiate what I have seen in practice. If this hypothesis was supported by the literature this could provide evidence to further inform clinical practice as well as having resource and service implications. Additionally this would help to justify that this is an appropriate population to target for future research including type, intensity and duration of intervention for best efficacy of handwriting treatment.

**FOCUSED CLINICAL QUESTION:** Are handwriting impairments associated with autism spectrum disorders in elementary school aged children?

### **SUMMARY of Search, 'Best' Evidence' Appraised, and Key Findings:**

- Four studies met the inclusion criteria. All were analytic (attempting to quantify the relationship between two factors), observational, case-control or cross-sectional studies of Level 4 evidence. No prospective cohort studies were found. Cartmill, Boccthy, Rodger and Medst (2009) found accuracy of letter formation was significantly worse for children with ASD than comparison children. Fuentes, Mostofsky and Bastian (2010) found adolescents with ASD have significantly poorer handwriting quality relative to controls. Myles et al. (2003) reported that a group of children with Asperger's Disorder produced significantly less legible letters and words than a control group. Lastly, Fuentes, Mostofsky and Bastian (2009) found that children with ASD show overall worse performance on a handwriting task than do age and intelligence-matched controls.

**CLINICAL BOTTOM LINE:**

Limited weak evidence shows an association between autism spectrum disorder and handwriting difficulties in elementary school-age children.

**Please note:** Handwriting in this CAT refers only to manuscript printing as evaluation of cursive handwriting was not used as an outcome measure in any of the included studies.

**Limitation of this CAT:** This critically appraised paper (or topic) was prepared for a graduate course assignment and has /has not been peer-reviewed by one other independent person/an instructor.

**SEARCH STRATEGY:****Terms used to guide Search Strategy:**

- **P**atient/Client Group: Elementary school-aged children
- **I**ntervention (or Assessment) ( or Issue that influences the prognosis): Autism spectrum disorders
- **C**omparison: Children of a similar age without autism
- **O**utcome(s): Handwriting difficulties

<b>Databases and Sites Searched</b>	<b>Search Terms</b>	<b>Limits Used</b>
Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) 1946 to Present	autism or autism spectrum disorder or Asperger*  AND  handwriting or handwriting difficult* or dysgraphia	English language  NO limits for date
CINAHL (1982- present)	AND	
EMBASE	child, preschool/ or child/ or child* or student	
Google Scholar		

\*denotes truncation for any variant spellings or word endings

Natural language, key words and index terms (including MeSH) were used as well as both Autism, Asperger's Syndrome and linked related terms such as autistic disorder.

### **INCLUSION and EXCLUSION CRITERIA**

- **Inclusion:** Studies involving male and female children/students with ASD with use of handwriting as an outcome measure.
- **Exclusion:** Studies involving adults with ASD, narrative or scoping reviews, studies that did not look specifically at the association of handwriting difficulties with ASD, studies that did not have handwriting as an outcome measure and studies with the main question being identification of underlying factors predicative of handwriting challenges in children with ASD.

### **RESULTS OF SEARCH**

4 relevant studies were located and categorised as shown in Table 1

**Table 1:** Summary of Study Designs of Articles Retrieved

Study Design/ Methodology of Articles Retrieved	Level*	Number Located	Author (Year)
Case-control	4	3	Fuentes, Mostofsky and Bastian (2009)
			Fuentes, Mostofsky and Bastian (2010)
			Myles, Huggins, Rome-Lake, Hagiwara, Barnhill and Griswold (2003)
Cross-sectional	4	1	Cartmill, Boccthy, Rodger and Medst (2009)

\*Based on Oxford Centre for Evidence-Based Medicine 2009 Levels of Evidence (Phillips et al. 2009)

### **BEST EVIDENCE**

The following study by Fuentes et al. (2009) was identified as the 'best' evidence and selected for critical appraisal. Reasons for selecting this study were:

- A case-control study design can be used for investigating the relationship or association between an outcome and a risk factor.
- This study is comparable to all the other included studies in terms of design type and level of evidence (4). The Cartmill et al. study (2009) was labelled "cross-sectional experimental" however it was very similar in design to the other 3 studies.
- The ages of the subjects were closest to the typical age of students on the writer's caseload (i.e. elementary age rather than adolescents).

- The wider diagnostic grouping of ASD (rather than only Asperger's Syndrome) best corresponds to the writer's typical caseload.
- The sample size was similar to the other included studies, with only one study (Myles et al. study of only students with Asperger's Syndrome) being slightly larger.
- The outcome measure is a standardized handwriting assessment (manuscript printing) and has well established validity and reliability (Reisman, 1993).
- This study represents low level evidence, but is the best study available for this critical appraisal.

## SUMMARY OF BEST EVIDENCE

**Table 2:** Description and appraisal of the case control study by Fuentes et al. (2009)

**Aim/Objective of the Study:** The objective was to identify if children with ASD show overall handwriting impairments when compared to age and intelligence matched controls and if so, whether these impairments are in specific qualitative categories. As well the study aimed to identify predictors of handwriting performance.

**Study Design:** A case-control design with outcomes measured at one point in time was used.

**Setting:** The study was set in an outpatient clinic in a large urban centre.

**Participants:** 28 subjects between 8 and 13 years of age participated. 14 subjects with ASD (3 females; mean age 10.2) and 14 typically developing controls (5 females; mean age 11.1) were enrolled. Based on performance on the Wechsler Intelligence Scale for Children (WISC-IV) all children had full-scale IQs (FSIQ) greater than 80, with the exception of 2 children with ASD who had marked discrepancies between factor scores. The two groups appeared to be comparable on the key demographic variables listed.

A purposive sample was used. Children with ASD were recruited from outpatient clinics at a large urban institute, local autism society chapters, schools, social groups, and paediatricians' offices. Children met DSM-IV criteria for ASD according to the Autism Diagnostic Observation Schedule–Generic (ADOS-G) and the Autism Diagnostic Interview–Revised (ADI-R).

Subjects in the control group were free of neurologic, developmental, or psychiatric disorders and had no immediate family member with a pervasive developmental disorder.

Informed consent was obtained from children, and written consent was obtained from a parent or guardian. Protocols were approved by an institutional review board.

**Phenomenon Investigated:** Handwriting abilities of children with ASD relative to controls were investigated as well as predictors of handwriting.

**Outcome Measures:****Primary Outcome:**

Handwriting impairment was investigated using the Minnesota Handwriting Assessment (MHA). For this test a sample of words is presented and subjects are instructed to copy the words on a standardized interlined test sheet. The MHA was scored according to the MHA scoring protocol including scoring letters on an individual basis in 5 categories (Legibility, Form, Alignment, Size and Spacing ) as well as a rate score (speed). The 5 qualitative category scores were then combined with the rate score for a total score (maximum total score of 204).

**Secondary Outcome:**

Subjects' motor skills were assessed using the Revised Physical and Neurological Examination for Subtle (Motor) Signs (PANESS). The PANESS consists of several categories, including stressed gaits, balance, and timed movements.

**Data Analysis:** Mean total handwriting scores, as well as mean scores in the 5 qualitative categories of the MHA and rate scores were compared between ASD and control groups with Student's t tests. Student's t tests were also used to compare age, intelligence scores and PANESS scores between the 2 groups. Within- and across-group stepwise multiple regressions were performed with handwriting scores as the dependent variable and independent variables such as: age, gender, IQ, and PANESS scores as predictor variables.

**Main Findings:****Primary Findings:**

Children within the ASD group demonstrated a range of handwriting abilities; both low-and high-scoring. The authors found that children with ASD showed overall worse performance on a handwriting task than did age-and intelligence-matched controls. Overall, total handwriting scores were lower in the ASD group than the control group (p 0.025). More specifically, children with ASD demonstrated worse quality of forming letters (p 0.006), but did not show significant differences in their ability to correctly size, align, and space their letters.

**Secondary Findings:**

Within the ASD group, motor skills were significant contributors to handwriting performance, whereas age, gender, IQ, and visuospatial abilities were not.

**Original Authors' Conclusions:**

The authors concluded that the ASD group scored significantly worse on motor measures including overall handwriting legibility than the control group demonstrating general motor impairments. The authors stated that the "results suggest that training targeting letter formation, in combination with general training of fine motor control, may be the best direction for improving handwriting performance in children with autism." (p. 1523).

**Critical Appraisal:****1) Validity:**

The case-control study is an appropriate method to answer the question of association of variables, as described in this study. However the authors did not include calculation of odds ratios in their analysis which is typically done in a case-control study. In terms of clarity of study purpose, the authors listed a second aspect to the study that was not clearly stated in the outset (i.e. identification of predictor variables of handwriting performance).

The cases were recruited from a large variety of settings which might assist with generalization however it was not stated exactly how many cases were enrolled from each setting. Additionally selection bias is a concern as it was not stated how the cases were recruited (i.e. whether there was any mention of the purpose of the study during recruitment that might lead to children with handwriting and/or motor problems being overly represented). The cases were well defined with standardized measures of intellectual and perceptual function provided in addition to detailed ASD diagnostic information. They were well matched to controls on parameters such as age and intellectual functioning. The non-response rates in both groups were not discussed. Concerns also exist regarding selection bias of the controls, as the variety of settings of recruitment was not indicated nor was there any mention of a reliable established system for selecting the controls. It is challenging to know if sufficient cases were selected in both groups as this was not discussed and a power calculation was not mentioned.

The MHA's validity and strength as a strong measurement tool was viewed with both high interrater and intrarater reliability as well as strong construct validity reported. To minimize bias, 2 raters, both blind to group classifications, independently scored each handwriting assessment and the averages of these scores were used in the analyses. High intrarater reliability was reported. The examiner's proficiency was not described for any of the measures. Additionally the validity of the PANESS and the WISC-IV was not reviewed nor was it indicated if there was blinding used for these secondary outcome measures. The likelihood of Type I errors (false positives) in testing were not mentioned.

Confounding factors were not reviewed explicitly in design, techniques or analysis. One additional confounding factor that the authors did not address is the lack of socio-economic data. This might lead to certain socioeconomic groups being over represented in either or both populations. There was also no mention of treatment intervention (i.e. did any of the cases receive any type of fine motor or handwriting treatment in the past that might have impacted their handwriting abilities and underestimated the handwriting difficulties).

**2) Interpretation of Results**

The analysis was not fully appropriate to the design of the study. As mentioned above, typically odds ratios are calculated in case-control studies to determine risk. This was not done. Thus, it is difficult to ascertain the strength of the association between the 2 variables in the primary outcome. The findings can

only be interpreted as an association and the many confounding biases listed previously weaken the strength of the association. The p values did support the significant differences that were seen between the ASD group and the controls for motor scores as well as overall handwriting legibly and form scores. Confidence intervals were not reported and could not be calculated with the data provided. Stepwise multiple regressions did isolate predictors of handwriting scores (secondary findings).

### **3) Summary/Conclusion:**

Despite the many limitations of this study, the results that indicate handwriting impairments are associated with ASD do appear clinically valid as they are consistent with what is typically seen in practice. With caution the results could be transferable and generalizable to the writer's clients. This would include measurement to individually evaluate client outcomes to substantiate findings as the strength of these results along with study limitations indicates limited and weak evidence. Additionally the secondary findings that motor skills are significant contributors to handwriting difficulties in children with ASD are also consistent with practice.

## **IMPLICATIONS FOR PRACTICE, EDUCATION and FUTURE RESEARCH**

There is little research on the association of handwriting difficulties in children with ASD despite the clinical observations that these children are known to exhibit handwriting difficulties. This study, despite its many limitations, does provide limited low level evidence to support this common finding in occupational therapy practice. The results of this study are consistent with what is typically seen in practice and do concur with the findings of a recent scoping review of handwriting difficulties in children with ASD (Kushki, Chau and Anagnostou, 2011). This review concluded that there is evidence that handwriting difficulties do exist in children with ASD however the studies are scant. Kushki et al. (2011) stated that overall legibility (specifically letter formation) is poorer in children with ASD than their typically developing peers and the evidence suggests that deficits in fine motor skills are significant contributors to handwriting difficulties in children with ASD. Cautious use of current findings in the writer's practice, with use of outcome measures to substantiate findings would be appropriate. Disseminating this knowledge to school staff for early screening and ongoing identification would also be prudent as studies have found that handwriting challenges persist beyond the elementary years (Beverdors et al. 2009; Breivik and Hemmingsson 2013; Fuentes, Mostofsky, and Bastian 2010; Myles et al. 2003). Further, this study also does lend some support to targeting this population for future research of handwriting intervention. The findings of this study highlight the need for well-designed prospective longitudinal studies over extended periods of time, preferably from large population-based samples. Such studies would further clarify the population-based prevalence, extent, and developmental trajectory of handwriting difficulties in those with ASD.

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### Critical Appraisal Checklists used:

Critical Appraisal Skills Programme (CASP) Case Control Study Checklist  
Retrieved Nov 07, 2013, from Critical Appraisal Skills Programme  
<http://www.casp-uk.net/wp-content/uploads/2011/11/CASP-Case-Control-Study-Checklist-31.05.13.pdf>