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Attention in selective mutism—An exploratory case-control study

Beate Oerbeck *, Hanne Kristensen

Centre for Child and Adolescent Mental Health, Eastern and Southern Norway, Norway

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Abstract

The aim of the study was to explore the association between selective mutism (SM) and attention. In SM social anxiety seems central but language impairment and motor problems are also reported. Attention problems have been described in parental behavioral ratings, while neuropsychological studies are lacking. A neuropsychological test (the Trail Making Test) and parental ratings of attention- and anxiety problems were administered to a clinical sample of 23 children with SM (aged 7–16 years, 12 boys and 11 girls) and 46 non-referred matched controls.

The SM group differed from controls on the Trail Making Test, but the group difference disappeared, when controlling for motor function and IQ. Parental ratings of attention problems were not significantly associated with the neuropsychological attention measure. Neuropsychological studies of attention controlled for IQ and motor function are needed as well as tests that measure different aspects of attention.

Keywords: Neuropsychology; Attention; Anxiety; Child; Selective mutism

Selective mutism (SM) is a childhood condition characterized by a persistent lack of speech in certain situations despite the ability to comprehend and use language. Children with SM usually speak to family members at home, but fail to speak in kindergarten or at school (American Psychiatric Association, 1994). The etiology of SM is still unclear but most probably SM represents a heterogeneous condition. SM is frequently associated with social anxiety both in children (Black & Uhde, 1995) and in adults (Steinhausen, Wachter, Laimbock, & Metzke, 2006) and it has been argued that SM represents a form of social anxiety disorder (SAD) (Anstendig, 1999; Sharp, Sherman, & Gross, 2007). However, the relationship between SM and SAD is still under debate (Yeganeh, Beidel, Turner, Pina, & Silverman, 2003).

SM is also associated with neurodevelopmental disorder/delay (Kristensen, 2000) in both language and motor function. Whether children with SM may suffer from subtle attention deficits is less studied. To explore the possibility of various neuropsychological impairments in SM may throw light on the etiology of the disorder and consequently have implications for treatment. Naturally, due to the mutism, language function has been most frequently studied in children with SM, and language impairment has been consistently reported in clinical studies (McInnes, Fung, Manassis, Fiksenbaum, & Tannock, 2004; Steinhausen & Juzi, 1996). However, language measures applied in SM studies may have required visuo-motor abilities as well (Manassis et al., 2003). Thus, in a former study we

* Corresponding author at: Centre for Child and Adolescent Mental Health, Eastern and Southern Norway, P.O. Box 4623, Nydalen, N-0405 Oslo, Norway. Tel.: +47 22586000; fax: +47 22586001.

E-mail address: beate.orbeck@r-bup.no (B. Oerbeck).

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explored aspects of nonverbal cognition and found that children with SM did not differ from controls on tests of visual memory span and visual memory, while verbal memory span was reduced (Kristensen & Oerbeck, 2006).

Motor deviance/delay is reported in studies including larger samples of children with SM (Kolvin & Fundudis, 1981; Steinhausen & Juzi, 1996) both with and without comorbid language disorder (Kristensen, 2002). Impaired motor function represents an important co-morbidity in language disorders without SM (Webster, Majnemer, Platt, & Shevell, 2005). In addition to motor impairment, language disorders are also associated with attention deficits and social difficulties (Snowling, Bishop, Stothard, Chipchase, & Kaplan, 2006). Whether children with SM may have subtle attention deficits is thus an important question to explore. Shyness, which is a hallmark of SM, has also been reported to co-vary with attention problems in normal girls (Caspi & Silva, 1995). Likewise, attention problems in early childhood years significantly predicted symptoms of anxiety and depression in a large cohort study of preadolescent boys and girls (Leech, Larkby, Day, & Day, 2006).

With regard to attention problems in SM, parental behavioral ratings suggesting attention deficits have been reported (Kristensen, 2001; Steinhausen & Juzi, 1996). To our knowledge, neuropsychological test results from case-control studies have not been published, and it is not known whether behavioral ratings of attention deficits and anxiety co-vary in this patient group. However, a study including two twin pairs with SM revealed an attentional problem in three of the four twins on a continuous performance test (Gray, Jordan, Ziegler, & Livingston, 2002).

In this article, we wanted to study the neuropsychological aspect of attention in a group of SM and matched controls and to examine whether motor function, performance IQ, and a diagnosis of language disorder and/or anxiety disorder were important predictors of performance on the attention test. We also wanted to explore whether the parental behavioral ratings co-varied with neuropsychological measures.

1. Methods

1.1. Subjects

The index group consisted of 23 children with SM, 12 boys and 11 girls, mean age 11.6 years (S.D. 2.7, range 7–16 years) while the control group comprised 24 boys and 22 girls, mean age 11.8 years (S.D. 2.6, range 7–16 years). The present study sample consists of children from age 7 years who were able to complete The Trail Making Test and had CBCL parental behavioral ratings (SM group: $n = 23$; control group: $n = 46$) from a larger sample of referred children with SM (age range 4–16 years, $N = 54$ and 108 matched controls). The children with SM were recruited nation-wide from out-patient clinics and school psychology services. The control group consisted of children who were asked to participate by the teachers of the children with SM. For each child with SM, two non-referred children (without SM) matched for gender, age, geographical site and SES were recruited. SES was assessed by the occupations of the parents and rated in accordance with the guidelines of the governmental statistics Norway and collapsed into four classes ranging from: 1 = upper status unto 4 = lower status, with no significant group differences [χ^2 (3,69) = 2.1, ns]. Details on the recruitment and SES procedures are described in a previous paper (Kristensen, 2000). As one of the matched controls failed to complete the most complex part of the Trail Making Test (part B), the number of controls was reduced from 46 to 45 on this subtest. The parents of one matched control did not fill out the Child Behavior Checklist, hence N was reduced to 45 on this measure.

1.2. Background assessment and present measures

1.2.1. Diagnostic procedure

A DSM-IV diagnosis of SM (APA, 1994) was assigned using the following procedure:

1. Discussion of the SM symptoms with the referring therapists. 2. A structured interview with parents and teachers on SM symptoms (Cline & Baldwin, 1994). 3. Direct observation.

1.2.2. Assessment of intelligence, motor function and behavior problems, and whether a diagnosis of a language disorder and/or an anxiety disorder was present

In addition to the neuropsychological test presented in this paper (the Trail Making Test), the examination included assessment of nonverbal IQ, as measured by performance IQ (PIQ) and motor function. PIQ was assessed by the Wechsler Scale of Intelligence-Revised (WISC-R; Wechsler, 1992). For further details concerning the WISC-R, see Kristensen (2000).

Fine and gross motor function was evaluated by sets of age-dependent motor items designed for the study (6–7 years: 8 items; 8 years: 12 items; 9–16 years: 13 items). The sets of items were chosen from three different motor assessment batteries (Gillberg, 1995; Oseretsky, 1936; Touwen, 1979). Each item was scored on a scale from 1 to 4. (1: failing the test; 2: poor performance or not at expected age level; 3: medium performance at expected age level; 4: good performance at expected age level). Each child was given a total motor score (sum score × 10 divided by number of items) with a maximum score of 40. To give an example: For age 6 through 7 years these items were included: Pencil grip, cutting a paper circle, diadochokinesis, sequential opposition of thumb to same-hand fingers, standing on one leg, jumping on one foot, walking on lateral aspects of feet, and balancing along a line. For more information about the motor tests applied, see Kristensen (2002). The mothers filled out the Achenbach Child Behavior Check List; CBCL (Achenbach, 1991). We present data from two CBCL syndrome scale scores related to anxiety (anxious/depressed and withdrawn) and the attention syndrome scale. Parents rate their child for how true each item is now or within the past 6 months using the following scale: 0 = not true (as far as you know); 1 = somewhat or sometimes true; 2 = very true or often true. Items are summed up to form the syndrome scales, and these raw scores are reported. For further details concerning the CBCL, see Kristensen (2001).

Table 1 present the performance IQ, the motor score, and the CBCL scores of the two groups.

The children were diagnosed as to whether or not they had a language disorder using different tests, due to a large age span of the total sample and varying response modes in the SM group. Tests used for the language assessment included the verbal IQ of the WISC-R (Wechsler, 1992), the Boston Naming Test (Kaplan, Goodglass, & Weintraub, 1983), the Reynell Scales (Reynell, 1977) the Peabody Picture Vocabulary Test-revised (Dunn & Dunn, 1981) and audiotapes of a conversation at home. Eleven out of 23 SM patients (48%) were diagnosed with a language disorder, in contrast to 7 of the 46 controls (15%), [χ^2 (1,69) = 8.46, $p = .004$]. The possible presence of an anxiety disorder was assessed by the parent version of the Child Assessment Schedule (CAS; Hodges, Kline, & Stern, 1982) adjusted according to DSM-IV criteria [regarding separation anxiety, social phobia, specific phobia, overanxious disorder (generalized disorder in DSM-IV), and obsessive-compulsive disorder]. Nineteen out of 23 SM patients (83%) fulfilled the criteria of an anxiety disorder, in contrast to four of the 46 controls (7%) [χ^2 (1,69) = 37.67, $p < .001$].

1.2.3. Neuropsychological measure of attention

Attention was measured using the child version of the Trail Making Test, with number of seconds to complete the test as our test variable (Reitan, 1971). The Trail Making Test require in the first and simpler part A that a line is drawn from digit 1 through 15 on a piece of paper where the numbers are scattered within circles. In the more complex part B, the subject is instructed to draw the lines in sequential shifts between digits and letters (; 1 – A, A – 2, 2 – B and so forth up to 8 – H). The Trail Making Test provides information regarding attention and visual scanning along with speed of both eye–hand coordination and information processing, and it is sensitive to cerebral dysfunction (Reitan & Wolfson, 1985). Studies have reported associations with motor function and IQ scores, and part B can also be seen as a measure of executive functioning as it requires alternate shifts between sets of stimuli; (Lezak, Howieson, & Loring, 2004; Mitrushina, Boone, & Delia, 1999; Spreen & Strauss, 1998; Warner et al., 2006).

1.3. Procedure

One examiner (the second author) assessed all the children. The participating families were asked to choose the location where they thought the child would feel most comfortable. The children were left to choose whether or not one of the parents should be present during the assessment. In the SM group, one child was assessed at home, nine at school and 13 at their outpatient clinic, while the control children were mainly seen at school. All participants received the different tests in the same order.

Table 1
Performance IQ, motor scores and CBCL subscale raw scores in children with SM and controls

	SM ($n = 23$) mean (S.D.)	Controls ($n = 46$) mean (S.D.)	t-test
Wechsler performance IQ	96.39 (18.34)	108.35 (14.74)	$t = -2.93$, $p = .01$
Motor function score	31.43 (4.68)	37.15 (3.18)	$t = -5.28$, $p < .001$
CBCL withdrawn[a]	6.83 (3.68)	1.0 (1.38)	$t = 7.34$, $p < .001$
CBCL anxious-depressed[a]	7.13 (5.93)	1.60 (2.65)	$t = 4.26$, $p = .01$
CBCL attention[a]	3.78 (4.09)	1.20 (1.31)	$t = 2.95$, $p = .008$

[a] Higher score indicates more problems.

Table 2
Mean results (time to finish in seconds; raw scores) on the Trail Making Test (part A and part B) in children with SM ($n = 23$) and controls ($n = 46$) and results from group comparison using independent t-tests on logtransformed data

	SM ($n = 23$) mean (S.D.)	Controls ($n = 46$) mean (S.D.)	Mean difference (95% CI) on logtransformed data	t (df[a])	p-value
TMT, part A	26.0 (12.5)	19.9 (7.4)	0.22 (0.002 to 0.45)	2.01 (67)	.05
TMT, part B	62.4 (31.8)	43.1 (16.9)	0.31 (0.09 to 0.54)	2.48 (66)	.008

[a] df: degrees of freedom.

1.4. Statistics

The data were analyzed using the Statistical Package for the Social Sciences; SPSS version 11 (SPSS, 2002). Means and standard deviations are reported and the significance level was set to 0.05. Due to skewness, the Trail Making Test data were log transformed. Independent t-tests were used to assess possible group differences. Linear regression analysis was employed to study the impact of SM on attention controlled for motor function, performance IQ, and the presence of language- or anxiety disorders. We used bivariate correlation analysis (Pearson) to study whether the neuropsychological test data and the behavioral rating data co-varied. The χ^2 test was used to assess whether the groups differed significantly in SES and the presence of deviant scores on the individual CBCL attention subscale items.

2. Results

2.1. Performance on the Trail Making Test

The SM group differed significantly from the control group on the Trail Making Test. Mean results (raw scores reported as seconds to complete the test) and analysis of group differences with independent t-tests (using log transformed TMT data) are presented in Table 2.

However, the group difference disappeared in multiple regression analysis including IQ and motor function. Motor function and IQ were both significant predictors of the TMT A performance, while IQ significantly predicted the TMT B performance. The results from the regression analysis are presented in Table 3.

One could question the use of performance IQ as a control for IQ in our study, since this score to some extent is time dependent (better scores with quick performance), as is the Trail Making Test itself. We therefore also performed the regression analyses with another measure related to IQ (the Peabody Picture Vocabulary Test-revised), that is time independent, rendering similar results (results not shown).

Analysis of whether or not the presence of a language disorder or an anxiety disorder influenced the attention test results by including them in the regression analysis (using the enter method) proved that none were significant predictors (all standardized betas <.23, ns).

Analysis of whether or not the presence of the parental CBCL ratings influenced the attention test results by including them in the regression analysis (using the enter method) proved that none were significant predictors (all standardized betas <.30, ns).

2.2. The relation between neuropsychological measures and questionnaire data

The neuropsychological measure of attention problems (the Trail Making Test) did not co-vary with the parental ratings of problems with attention or anxiety as measured by the CBCL syndrome scales. However, the

Table 3
Multiple regression analysis (enter method): effects of patient status and performance IQ and motor score on log transformed Trail Making Test (part A and part B) data

	Dependent variable: Trail Making Test, part A		Dependent variable: Trail Making Test, part B	
	Standardized coefficient (β)	p-value	Standardized coefficient (β)	p-value
Patient status	.154	.226	−.114	.423
Performance IQ	−.356	.002	−.268	.029
Motor score	−.463	<.001	−.193	.173
R^2 adjusted	.320		.170	
Significance of model	$F(3,65)$ 11.65, $p < .001$		$F(3,64)$ 5.43, $p = .002$	

Table 4
Correlations between parental ratings (three CBCL syndrome scales) and neuropsychological measures (log-transformed Trail Making Test score, motor score and Performance IQ)

Parental ratings	Neuropsychological measures			
CBCL syndrome scales	TMT A	TMT B	Motor score	Performance IQ
Attention problems	.168, ns	.079, ns	$-.514^{**}$, $p < .001$	$-.253^{*}$, $p = .037$
Anxiety/depression	.054, ns	.057, ns	$-.412^{**}$, $p < .001$	−.139, ns
Withdrawn	.041, ns	.068, ns	$-.485^{**}$, $p < .001$	−.232, ns

* $p < .05$.
** $p < .001$.

Table 5
The number and percentage of children with SM ($n = 23$) and controls ($n = 45$) with positive item scores (1 = somewhat or sometimes true, 2 = very true) on the CBCL attention syndrome subscale items

	SM ($n = 23$) N (%)	Controls ($n = 45$) N (%)	χ^2	p-value
Item 1, Acts too young for his age	10 (43)	4 (8)	11.14	.004
Item 45, Nervous	10 (43)	5 (11)	10.42	.005
Item 13, Confused, seems to be in a fog	6 (26)	1 (2)	9.49	.009
Item 62, Poorly coordinated, clumsy	4 (17)	0	8.32	.004
Item 8, Can't concentrate	10 (43)	8 (18)	7.01	.031
Item 17, Daydreams, gets lost in his thoughts	11 (48)	10 (22)	6.91	.032
Item 80, Stares blankly	2 (9)	0	4.03	.045
Item 61, Poor schoolwork	7 (30)	5 (11)	4.76	.093
Item 46, Nervous movements or twitching	3 (13)	1 (2)	3.59	.17
Item 41, Impulsive, acts without thinking	6 (26)	12 (27)	2.11	.35
Item 10, Can't sit still, restless, hyperactive	1 (4)	4 (9)	0.68	.71

Group differences explored with Chi-square analysis (descending χ^2).

CBCL parent reported attention problems were associated with the two other neuropsychological measures; motor problems and reduced nonverbal IQ (Table 4).

The parent reported syndrome scale scores on attention and anxiety (anxiety/depression and withdrawn) also correlated significantly ($R = .67$ and .58, respectively, $p < .001$).

The specific items from the CBCL syndrome scale of attention problems that best differentiate SM from controls are presented in Table 5.

3. Discussion

3.1. Performance on the Trail Making Test

This is the first case-control study exploring a neuropsychological measure of attention in children with SM. Because studies on SM that include comprehensive neuropsychological assessment with sufficient samples sizes are lacking, our report on specific aspects of attentional functioning in SM may be important. This added knowledge may contribute to the understanding of possible pathways of symptom development and to the identification of possible subgroups of children with SM.

The present study found an attention deficit in SM, as measured by the Trail Making Test. However, motor problems and reduced nonverbal IQ were central factors in explaining this deficit, emphasizing the need to control for these factors in future measures of attention. Our results are in line with a study of 38 anxious/depressed boys (age 9–11 years) who performed the Trail Making Test part B significantly slower than the control group (Emerson, Mollet, & Harrison, 2005). No attempts were made to assess the impact of IQ and motor function in this study. Hence whether anxiety actually explained the mentioned deficit, is not known.

3.2. The relation between neuropsychological measures and questionnaire data

The neuropsychological measure of attention did not co-vary with the parent reported attention or anxiety problems. This finding, along with the fact that neither a language disorder, nor the presence of an anxiety disorder significantly predicted the Trail Making Test performance, could suggest that the neuropsychologically measured attention deficit represent an important dimension in SM worth further studying. Whether possible attention deficits will present as a separate problem or embedded within motor and intellectual impairment, as in our study, remains to be seen.

Whether acute anxiety influenced the results in our study is not known, since we regretfully did not include a state measure of anxiety. Interestingly, in anxious adults presented with somewhat threatening auditory stimuli, only state anxiety but not trait anxiety influenced attention, as measured by event-related potentials (Mercado, Carretié, Tapia, & Gómez-Jarabo, 2006). Gray et al. (2002) described an attentional test pattern indicative of what he calls an anxious response style in three of the four twins with SM he studied. This response style was characterized by slow reaction times, numerous omission errors (errors due to non responding) along with hardly any commission errors (false positive errors). This could suggest over-cautiousness and anxiety linked to being afraid of making mistakes in the test situation. However, an anxiety state measure was not included in this study either.

The parent reported attention problems were not associated with the neuropsychological measure of attention problems used in our study. However, the parent reported attention problems was associated with the parents' own ratings of anxiety problems and with a reduced nonverbal IQ and motor problems (Table 4). That reduced IQ and motor problems were associated with parental ratings of attention problems is not surprising given the individual items comprising the CBCL attention problem syndrome scale that significantly differed between SM and controls in our study (immaturity, clumsiness, being nervous and being confused; Table 5). That parental CBCL rating of anxiety and attention problems were associated with each other, is in line with a recent study on CBCL as a screen for psychiatric co-morbidity in pediatric patients with ADHD (Biederman, Monuteaux, Kendrick, Klein, & Faraone, 2005), where anxiety disorders were predicted both by the anxious/depressed and the attention CBCL syndrome scales.

3.3. Limitations

The present study has several limitations. It is a clinical sample mainly recruited from out-patient clinics and may thus represent children with increased co-morbidity. However, the inclusion of children with SM referred from primary care services (the school counselling service) somewhat reduces the bias inherent to clinical samples. Still, the results cannot be generalised to non-referred children with SM. Interestingly, an association between SM and neurodevelopmental disorder/delay has recently been confirmed in a community study (Elizur & Perednik, 2003).

The mean age, 11.6 years, in the present sample may also seem relatively high compared to other clinical samples, but is due to the age limit set for the test applied in the study. Our total study sample (Kristensen, 2000) corresponds fairly well with other clinic-based samples with regard to gender ratio, age, SES, age at symptom onset and symptom duration at assessment.

Furthermore, the assessment was conducted by a single clinician aware of the children's group status. Thus, investigator bias is another possible limitation. However, this study cannot be conducted blindly, for obvious reasons. One could also argue that the differences within and between groups with regard to location of assessment may have influenced the results. However, all the participating families chose the location which they thought the child would feel most comfortable with, and the children themselves chose whether one of the parents should be present during the assessment. It is crucial to establish rapport with children with SM in order to be able to carry out a comprehensive assessment. Even though this lack of uniformity may seem non-optimal methodically, we found the procedure essential in recruiting an adequate sample size.

4. Conclusion

The present study explored attentional functioning in SM, as measured by the Trail Making Test. We found that children with SM differed from controls on the Trail Making Test, but this deficit disappeared when controlling for motor function and IQ in multivariate analyses.

Parental CBCL ratings of attention problems were associated with reduced IQ and motor problems, not with the neuropsychological attention measure.

Further neuropsychological studies controlling for motor function and IQ, and including measures of different aspects of attention, are needed.

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