IS SELECTIVE MUTISM ASSOCIATED WITH DEFICITS IN MEMORY SPAN AND VISUAL MEMORY?: AN EXPLORATORY CASE–CONTROL STUDY

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Our main aim in this study was to explore the association between selective mutism (SM) and aspects of nonverbal cognition such as visual memory span and visual memory. Auditory–verbal memory span was also examined. The etiology of SM is unclear, and it probably represents a heterogeneous condition. SM is associated with language impairment, but nonspecific neurodevelopmental factors, including motor problems, are also reported in SM without language impairment. Furthermore, SM is described in Asperger’s syndrome. Studies on nonverbal cognition in SM thus merit further investigation. Neuropsychological tests were administered to a clinical sample of 32 children and adolescents with SM (ages 6–17 years, 14 boys and 18 girls) and 62 nonreferred controls matched for age, gender, and socioeconomic status. We used independent t-tests to compare groups with regard to auditory–verbal memory span, visual memory span, and visual memory (Benton Visual Retention Test), and employed linear regression analysis to study the impact of SM on visual memory, controlling for IQ and measures of language and motor function. The SM group differed from controls on auditory–verbal memory span but not on visual memory span. Controlled for IQ, language, and motor function, the SM group did not differ from controls on visual memory. Motor function was the strongest predictor of visual memory performance. SM does not appear to be associated with deficits in visual memory span or visual memory. The reduced auditory–verbal memory span supports the association between SM and language impairment. More comprehensive neuropsychological studies are needed. Depression and Anxiety 23:71–76, 2006. © 2006 Wiley-Liss, Inc.

Key words: selective mutism; neuropsychology; memory span; visual memory

INTRODUCTION

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. The etiology of SM is still unclear, but most probably SM represents a heterogeneous condition. SM is frequently associated with not only social anxiety [Black and Uhde, 1995] but also neurodevelopmental disorder/delay [Kristensen, 2000].

To date, only two studies have examined whether SM simply reflects an extreme level of social anxiety by comparing children with SM and children with social phobia [SP; Manassis et al., 2003; Yeganeh et al., 2003]. The results showed surprising similarity between the
two groups on anxiety measures and thus failed to confirm unequivocally the hypothesis that SM merely reflects the extreme end of a social anxiety continuum. However, one of the studies reported that the SM group showed some language impairment relative to the SP group [Manassis et al., 2003].

Language impairment has consistently been reported in clinical studies of children with SM [Andersson and Thomsen, 1998; Steinhausen and Juzi, 1996], but most of these have been retrospective studies based on case reports. A case–control study including direct assessment of 54 children with SM found that as many as 50% of subjects were assigned a lifetime diagnosis of a language disorder, with phonological disorder being the most frequent category [n = 23; Kristensen, 2000]. Furthermore, studies as to whether the reluctance to speak may be a result of an inability to process speech or auditory stimulation have been recommended [Yeganeh et al., 2003]. Intriguingly, a recent case–control study detected reduced auditory afferent feedback activity in some children with SM [Bar-Haim et al., 2004]. The authors suggest that this deficiency may lead to restricted vocalization in contexts that require complex auditory processing. In the study by Manassis et al. [2003] comparing SM and SP, the children with SM performed less well than children with SP on a task measuring discrimination of speech sounds.

In accordance with the fact that muteness is the core symptom in SM, the language function of these children has been most frequently studied. Unfortunately, more comprehensive neuropsychological studies of SM are scarce. However, Gray et al. [2002] have described results from two sets of dizygotic twins with SM. Both sets of twins were prematurely born. One of the twin pairs was characterized by reduced verbal intelligence, along with considerably reduced expressive and expressive language. The other pair of twins had normal intelligence but difficulties with expressive language and motor sequencing, including oromotor coordination deficits.

Motor deviance/delay is also reported in studies including larger samples of children with SM [Kolvin and Fundudis, 1981; Steinhausen and Juzi, 1996]. This is not surprising, because impaired motor function represents an important comorbidity in language disorders [Webster et al., 2005]. However, nonspecific neurodevelopmental factors such as a reduced pre- and perinatally optimality score, increased frequency of minor physical anomalies, and deficient motor skills in SM do not seem to be restricted to the children with a comorbid language disorder [Kristensen, 2002].

Furthermore, SM is also described in children with Asperger’s syndrome [Bankier et al., 1999; Gillberg and Billstedt, 2000]. A high level of concordance in neuropsychological profile, including assets of auditory memory and vocabulary and deficits in visual memory and motor skills, have been found between Asperger’s syndrome and nonverbal learning disabilities [NLDs; Klin et al., 1995]. Finally, language measures applied in SM studies may have required visuomotor abilities as well [Manassis et al., 2003]. Thus, nonverbal cognition in SM merits further investigation.

In this study we wanted to explore aspects of nonverbal cognition in SM by examining whether children with SM differ from controls on measures of visual memory span and visual memory. Because of the reported association between SM and language impairment, we also wanted to examine auditory–verbal memory span in the two groups.

METHODS

SUBJECTS

The children in this study (SM group: n = 32; control group: n = 62) constituted a part of a larger sample consisting of 54 referred children with SM and 108 controls. The index group was recruited nationwide from outpatient clinics and school psychology services. The control children were asked to participate by the teachers of the children with SM. For each child with SM, two nonreferred children (without SM) were recruited, matched for gender, age (±10 months), geographical area (same or neighboring kindergarten or school), and socioeconomic status (SES; parents’ occupation belonging to the corresponding SES group). The recruitment procedure has been described in detail in a previous article [Kristensen, 2000].

The subsample described in this article consists of all the children with SM and their matched controls who completed an assessment of both memory span and visual memory. For various reasons, such as age, declining to participate, and so forth, only 32 of 54 children with SM completed this part of the assessment. Because two of the matched controls also failed to complete these tests, the number of controls is reduced from 64 to 62.

Sociodemographic characteristics are presented in Table 1. The index group consisted of 32 children with SM [14 boys and 18 girls, ages 6–17 years; mean age, 10.6 years (3.1)]. A total of 24 (75%) children with SM were recruited from various outpatient clinics, and 8 children with SM (25%) were recruited from different school psychology services.

BACKGROUND ASSESSMENT

Diagnostic procedure. We assigned a DSM-IV diagnosis of SM [American Psychiatric Association, 1994] using the following procedure: (1) discussion of the SM symptoms with the referring therapists; (2) interviews of parents and teachers about SM symptoms using a structured format [Cline and Baldwin, 1994]; and (3) direct observation.

Assessment of cognition, language, and motor function. In addition to the measures of memory span and visual memory presented in this article, the
examination included assessment of performance IQ (PIQ), and speech/language and motor function. PIQ was assessed by the Wechsler Preschool and Primary Scale of Intelligence [WPPSI; Wechsler, 1989] or the Wechsler Intelligence Scale for Children—Revised [WISC-R; Wechsler, 1992]. Language skills had to be evaluated with various language tests due to a large age span of the total sample and varying response modes in the SM group. However, the Peabody Picture Vocabulary Test [PPVT; Dunn and Dunn, 1981] was administered to all participants and was therefore chosen as a language measure in this study. Other tests used for the language assessment, but not included in this study, were the Verbal IQ section of the WPPSI or WISC [Wechsler, 1989, 1992], the Boston Naming Test [Kaplan et al., 1983], the Reynell Scales [Reynell, 1977], and audiotapes of a conversation at home.

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; Oseretisky, 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 motor score (sum score × 10 divided by number of items), and the maximum score was 40. For further details concerning the tests applied, see Kristensen [2000, 2002]. The PIQ, PPVT, and motor scores in the subsample are presented in Table 2.

### MEASURES

**Auditory–verbal memory span.** Auditory–verbal memory span was measured using the Digit Span forward subtest from the WISC-R [Wechsler, 1992]. The index children chose whether to answer verbally (n = 12), write down (n = 13), or point to corresponding figures in a digit row written on a sheet of paper (n = 7). All of the children in the control group answered verbally.

**Visual memory span.** Visual memory span was examined by having the children point to a series of 2.5 cm cubes placed randomly on a 30 cm tray made for the study. When administering this task, the examiner points to a series of cubes at a rate of one cube per second. Then the child reproduces the same sequence by pointing. Items gradually increase in length from sets of two to sets of nine cubes.

**Visual memory.** We examined visual memory with the Benton Visual Retention Test [BVRT; Sivan, 1992] using form C and administration A. This test consists of 10 cards with geometric figures. The subjects are allowed a 10-s exposure to each card before an immediate recall by drawing is required. The number

| TABLE 1. Sociodemographic characteristics in children with SM and control children |
|----------------------------------|----------------------------------|-----------|
|                                  | SM                                | Control   |
| Age (years) Mean (SD)            | 10.0 (2.6)                       | 10.7 (3.0) |
| Gender                           | 18 (56.2)                        | 36 (58.1) |
| SES                              |                                  | 62 (100)  |
| 1 (upper)                        | 3 (9.4)                          | 11 (17.7) |
| 2                                | 7 (21.9)                         | 20 (32.3) |
| 3                                | 14 (43.8)                        | 24 (38.7) |
| 4 (lower)                        | 8 (25.0)                         | 7 (11.3)  |
| Gender                           | 18 (56.2)                        | 36 (58.1) |
| SES                              |                                  | 62 (100)  |
| 1 (upper)                        | 3 (9.4)                          | 11 (17.7) |
| 2                                | 7 (21.9)                         | 20 (32.3) |
| 3                                | 14 (43.8)                        | 24 (38.7) |
| 4 (lower)                        | 8 (25.0)                         | 7 (11.3)  |

*aMean (SD) if indicated in the first column.

| TABLE 2. PIQ, PPVT, and motor scores in children with SM and control children |
|----------------------------------|----------------------------------|-----------|
|                                  | SM                                | Control   |
| PIQ* Mean (SD)                   | 93.9 (19.1)                      | 111.6 (13.9) |
| PPVT score* Mean (SD)            | 94.4 (20.6)                      | 115.9 (21.4) |
| Motor score* Mean (SD)           | 31.1 (5.0)                       | 36.5 (3.2)  |

*aSM total < Control total, P < .001.
of correct scores (range, 0–10) and the number of errors (one per figure) may be reported. In this study, we report the number of correct scores. Performance on the BVRT is dependent upon several different functions, such as visuospatial perception, conceptualization, memory, and motor response [Lezak et al., 2004], and must be evaluated in relation to IQ [Lezak et al., 2004; Sivan, 1992].

PROCEDURE

One examiner (H. Kristensen) assessed all the children in both groups. The participating families chose the location for assessment where they thought the child would feel most comfortable. The children also chose whether one of the parents should be present during the assessment. In the SM group, one child was assessed at home, 13 at school, and 18 at their outpatient clinic. The majority of the control children were seen at school, and a minority at home. The order of presentation of the different tests was identical for all participants. The Benton results were scored by an experienced neuropsychologist (B. Oerbeck) blind to group status.

STATISTICS

Data were analyzed with the Statistical Package for the Social Sciences (SPSS) version 11 [Norusis, 2002]. Means and standard deviations are reported, and the significance level was set to .05. We used independent means and standard deviations are reported, and the Social Sciences (SPSS) version 11 [Norusis, 2002].

DATA ANALYSIS

The results presented in Table 4 show that motor function was the most significant predictor of the visual memory results. The language measure was also a significant predictor, whereas IQ no longer was. As in the first analysis, no group differences were detected.

DISCUSSION

This is the first case-control study exploring memory span and visual memory in a relatively large sample of children with SM. Because studies on SM

| TABLE 3. Memory span and visual memory (BVRT) in children with SM (n = 32) and controls (n = 62), and results from group comparison using independent t-tests |
|---------------------------------|-----------------|-----------------|-----------------|-----------------|
|                                | SM (n = 32)     | Controls (n = 62) | Mean difference |
|                                | Mean (SD)       | Mean (SD)        | (95% CI)         | t (df)          | P-value         |
| Auditory–verbal memory span    | 5.2 (1.1)       | 5.9 (1.3)        | 0.7 (−1.2 to −0.1) | 2.5 (92)       | .01             |
| Visual memory span             | 5.2 (1.3)       | 5.2 (1.0)        | 0.01 (−0.5 to 0.5)| 0.03 (92)      | .98             |
| BVRT correct score             | 4.8 (3.0)       | 6.3 (2.6)        | 1.5 (−2.7 to 0.3)| 2.52 (92)      | .01             |
including comprehensive neuropsychological assessment with sufficient samples sizes are lacking, reports on specific aspects of nonverbal cognitive functioning in SM may be important. Such knowledge may contribute to the understanding of possible pathways of symptom development and to the identification of subgroups of children with SM.

Our current study failed to find any deficits in visual memory span or visual memory (when controlling for PIQ, language, and motor function) in children with SM compared to controls. The visual memory span result is in line with a previous study reporting that children with SM (n = 14) and children with SP (n = 9) did not differ in visual memory span or visual working memory [Manassis et al., 2003]. To our knowledge, no other study has examined visual memory as measured with BVRT in SM, and these results need to be replicated. Visual memory deficits are frequent in Asperger's syndrome and NLDs [Klin et al., 1995]. Thus, the present visual memory results do not indicate a strong relationship between these conditions and SM as a group. The fact that SM has been reported in several cases of Asperger's syndrome [Bankier et al., 1999; Gillberg and Billstedt, 2000] may reflect the notion of SM as a heterogeneous condition. However, in this study we only explored just a few aspects associated with a NLD neuropsychological profile. Moreover, the SM group performance was poorer than that of controls on the PIQ measure, indicating nonverbal deficits. The subtests comprising the PIQ are also dependent on several different neuropsychological skills, and the results emphasize the need for studies including a more comprehensive neuropsychological assessment in larger SM samples.

The visual memory results also indicate that visual skills may not explain previously reported group differences between SM and controls on language measures requiring visuomotor abilities [Manassis et al., 2003]. Nevertheless, our finding that motor function explained a significant portion of the BVRT underlines the need of controlling for motor function, if the tests applied also require motor abilities. Furthermore, it emphasizes the fact that motor deficits may be important in SM.

Even though very few studies have included direct assessment of motor skills, motor disorder/delay has consistently been reported in SM [Kolvin and Fundudis, 1981; Steinhausen and Juzi, 1996]. In general, studies on the relationship between subtle motor impairment and social anxiety are sparse. However, children with motor coordination problems have been reported to have a threefold prevalence of mental disorders compared to a normal population [Meltzer et al., 2000]. Neurological soft signs, including motor items at age 7, have also been found strongly to predict anxiety/withdrawal at age 17 [Shaffer et al., 1985]. Moreover, reduced athletic achievement has been found to be an antecedent of avoidant personality disorder, which is held to reflect the more severe end of the social anxiety spectrum [Retrrew et al., 2003]. Subtle motor impairment and not anxiety alone may be one reason for withdrawal from some social situations in some children with SM. Furthermore, oromotor coordination deficits have been demonstrated in two pairs of twins with SM [Gray et al., 2002], and the most prevalent language disorder in SM is found to be articulation disorder [Kristensen, 2000], which also reflects motor problems.

The difference in auditory–verbal memory span between children with SM and controls supports the notion that deficits in speech and language processing are associated with SM. The methodological weakness of different response mode in the index group and between the index and control group did not seem to influence the results substantially. The children with SM who answered verbally (n = 12) still performed poorer than their controls.

However, anxiety has been shown to influence memory functions [Eysenencck and Calvo, 1992] and may thus account for the deficit findings. If so, one still has to explain why the anxiety has a greater influence on auditory as compared to visual processing. 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 in which they thought the child would feel most comfortable. The children also chose whether one of the parents should be present during the assessment. It is crucial to establish rapport with children with SM to carry out a comprehensive assessment. Even though this lack of uniformity may seem nonoptimal methodically, we found this procedure quite essential in recruiting an adequate sample size. The assessment was also conducted by a single clinician, aware of the children's group status. Thus, investigator bias is another possible limitation, but conducting this study blindly cannot be done for obvious reasons.

The representativity of the sample is another important issue. It is a clinical sample mainly recruited from outpatient clinics and may thus represent children...
with increased comorbidity. However, the inclusion of children with SM referred from primary care services (the school counseling service) somewhat reduces the bias of a clinical sample. Still, the results cannot be generalized to nonreferred children with SM. Interestingly, an association between SM and neurodevelopmental disorder/delay has recently been confirmed in a community study [Elizur and Perednik, 2003]. The mean age [10.6 (3.1)] in our sample may also seem relatively high compared to other clinical samples, but it is due to the age limits for the visual memory test applied in the study (>8 years). (Children with SM who were younger than 8 years were also included if they, and one or both their age-matched controls, managed to perform the Benton test). The total sample of this study corresponds fairly well with other clinic-based samples with regard to gender ratio, age, SES, age at symptom onset, and symptom duration at assessment.

CONCLUSION

This study explored the issue of specificity of language deficits in SM by examining whether children with SM differ from controls on measures of memory span and visual memory. The results show that children with SM differ from controls with regard to auditory–verbal memory span, but not visual memory span. The impaired performance on the visual memory test in the SM group compared to controls disappeared when statistically controlling for language and motor function. The results support the association between SM and problems in speech and language processing. Studies including a more comprehensive neuropsychological assessment are needed.

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