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THESIS

Selective Mutism in Unilingual Children, Multilingual Children, and Children with a Co-morbid Autism Spectrum Disorder: Differences and Common Risk Factors.

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Preface

Bij de sollicitatie voor een klinische stageplaats binnen de zorglijn Autisme van het UMC Utrecht vroeg Maretha de Jonge aan mij of ik, naast mijn stage, mee wilde werken aan een follow up onderzoek naar selectief mutisme. Mede door mijn interesse in het wetenschappelijk onderzoek en het feit dat selectief mutisme een zeldzame en zeer interessante stoornis is, zei ik direct ja. Vanaf april 2010 heb ik als onderzoeksassistente met veel plezier meegewerkt aan dit onderzoek en ben vanaf februari 2011 begonnen met het schrijven van mijn thesis. In deze thesis heb ik geprobeerd een goed beeld te geven van deze bijzondere kinderen, waarvan een groot deel meertalig is en een ander deel een co-morbide autisme spectrum stoornis heeft. Ik hoop met dit onderzoek iets bij te kunnen dragen aan het klinisch beeld van deze stoornis. Dit had ik echter niet zonder hulp kunnen doen en ik wil dan ook de volgende mensen bedanken:

Allereerst wil ik Maretha de Jonge bedanken voor haar kennis en haar enthousiasmerende begeleiding. Ik heb het heel erg fijn gevonden om op het laatst, toen de tijd begon te dringen, bijna dagelijks met vragen bij haar binnen te kunnen lopen. Ik wil haar ook bedanken voor de vrijheid die ik gekregen heb om mijn eigen onderzoeksvragen te bepalen en ik heb hierdoor een thesis geschreven waar ik trots op ben. Ten tweede wil ik mijn tweede begeleider, mevrouw Liesbeth Aleva bedanken voor haar goede adviezen. Zij heeft me uitgedaagd diepgang in mijn thesis te brengen en ik hoop dat ik hierin geslaagd ben. Ik wil ook een aantal mensen in mijn persoonlijke omgeving bedanken. Mijn vriend Martin, die mij heeft gesteund in de afgelopen tijd en mij de energie heeft gegeven om door te gaan. Verder wil ik in het bijzonder mijn mede-stagiaire Laurien Passtoors bedanken, voor de lange gezamenlijke avonden in de Universiteitsbibliotheek, het doorlezen van mijn scriptie en het geven van zeer bruikbare tips. Mede dankzij deze mensen ben ik tot dit resultaat gekomen. Ten slotte rest mij niks anders dan de mensen die mijn thesis gaan lezen veel leesplezier toe te wensen!

Abstract

Objective: To evaluate characteristics of unilingual children with selective mutism, multilingual children with selective mutism and children with a co-morbid autism spectrum disorder (ASD). This research study will provide a much needed insight into the different risk theories of SM. **Method:** Information derived from the clinical files of 139 children, with SM referred for diagnosis and treatment to the academic hospital in Utrecht (UMCU) between 1973 and 2011, was analysed. 56% (78/139) of the SM children was unilingual, 28% (39/139) was multilingual, and 15% (21/139) had a co-morbid ASD. **Results:** The symptoms at school of unilingual children were significantly less severe when compared to the other groups. When unilingual and multilingual children were compared, the unilingual SM children appeared to experience more delays in the early development of language, whereas the multilingual SM children seemed to have a slightly (not significant) less extended vocabulary. The co-morbid ASD children showed significant language delays as well as a slightly (not significant) less extended vocabulary. The ASD group was found to be more anxious and showed more internalizing problems when compared to the other two groups. Compared to standardized scores, anxiety problems, multilingualism, gross motor and language developmental delays and a co-morbid ASD are found to be more present in our sample. Oppositional behavior is reported by parents in over half of the SM children. Teachers reported stubbornness in over a third of the SM children. Symptom severity, measured by the amount of situations the child refuses to speak in, is found to be the least at home. **Conclusion:** The ASD subgroup showed significantly more behavioral problems and a higher severity of SM symptoms compared to the other two groups. The multilingual subgroup only showed significantly more problems in language development and severity of the SM symptoms. Developmental delays (both in gross motor development and language development) could be a risk factor in developing SM. Anxiety problems, oppositional behavior, multilingualism, and ASD seem to be related to SM.

John is a 4 year-old boy who lives with his parents and two siblings. At home he is able to speak normally, but when he's at school he doesn't speak at all. John has selective mutism. Selective mutism is a paediatric psychiatric condition that is considered to be preceded by shy inhibited behaviour (Sharkey & McNicholas, 2008). It ranges in presentation from a reluctance to speak in certain situations to physical and social "frozen" unresponsiveness (Wintgens, Keen & Fonseca, 2008). According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-Text Revised, APA, 2001), selective mutism is a disorder that is primarily present in early childhood and can be diagnosed using the following criteria: there is a persistent failure to speak in specific situations in which speaking is expected, such as school and social gatherings, despite speaking appropriately in other situations. The disturbance interferes with educational or occupational achievement or with social communication. Selective mutism can not be diagnosed if the disturbance lasts for less than a month, is limited to the first month of school and if it is not due solely to a lack of knowledge of, and comfort with, the spoken language required in the social situation. It is also not diagnosed if the disturbance is better accounted for by embarrassment related to having a Communication Disorder (e.g., Stuttering) or if it occurs exclusively during a Pervasive Developmental Disorder, Schizophrenia, or other Psychotic Disorder. Diagnosis using the DSM has been possible since 1980. Before 1994, selective mutism was called elective mutism. This term remains the diagnostic label in ICD-10 (World Health Organisation, 1992), but the criteria for diagnosis are similar to selective mutism in the DSM-IV-TR. For the purpose of this study the term selective mutism (SM), will be used.

Prevalence

The prevalence of SM is estimated at around 0.76%, but ranges up to 2% in more recent school-based studies (Elizur & Perednik, 2003; Wintgens, Keen & Fonseca, 2008; Sharp, Sherman & Gross, 2007; Cohen et al., 2008). It has been hypothesized that variability among these estimates may be a function of differing diagnostic criteria used in each study and the age at which the children were sampled (Kumpulainen, 2002). The symptoms of selective mutism typically start at preschool age (Steinhausen & Juzi, 1996) and presentation peaks at 3 and 6 years of age (Wintgens, Keen & Fonseca, 2008). Elizur and Perednik (2003) found that native children with SM had a significantly earlier onset of symptoms, compared to immigrant children with SM.

Risk factors: SM and anxiety

SM has been highly correlated with anxiety disorders (Vecchio & Kearney, 2005; Dummit et al., 1997; Cunningham et al., 2004), especially social anxiety disorder (Dummit et al., 1997; Cohan et al., 2008; Cunningham, McHolm, & Boyle, 2006; Carbone et al., 2010; Ford et al.,

1998; Cunningham, McHolm & Boyles, 2006) and more internalizing problems have been reported (Elizur & Perednik, 2003) when comparing SM children to a matched control group. This could explain why selective mutism is usually preceded by extreme shyness and children are described by their parents as having a 'slow-to-warm' temperament in social situations (Sharp, Sherman & Gross, 2007). In one study (Cunningham et al., 2006) it was concluded that the SM children were more obsessive, prone to somatic complaints, and depressed than controls. Also, achievement and social stress may be particularly problematic for children with high levels of self-oriented perfectionism (Hewitt et al., 2002). Therefore it is likely that perfectionism could be a common factor in SM children. Limited research has been done to explore a possible link between SM and perfectionism but it is suggested that parenting styles could be an influence. Immigrant parents are often more performance-driven than Dutch parents (Pels, 2004), so their (multilingual) children could be more perfectionistic than unilingual children.

Risk factors: SM and oppositional behavior

Although most research suggests that SM is an anxiety related disorder, some studies found a rather strong relation between oppositional behaviour and SM. When a child withholds speech in certain situations while speaking freely in others, the behavior appears to be manipulative and controlling (Leonard & Topol, 1993). Many of the early case studies focused on the underlying oppositional and defiant etiology which often characterises children with SM (Sharp, Sherman & Gross, 2007). Descriptions used are 'manipulative', 'dominating', 'negative', 'disobedient' 'stubborn' and/or 'aggressive' (Kristensen, 1997; Sharp, Sherman & Gross, 2007; Anstendig, 1999). In some studies (Krohn, Weckstein & Wright, 1992) up to 90% of the selectively mute children were described as controlling, negative, or oppositional. Perednik and Elizur (2003) found a significant effect on being "stubborn, sullen, or irritable" when comparing SM children with a normal control group. Omdal (2007) has interviewed six recovered ex-patients and they all reported being very strong-willed and, they were consciously determinated not to speak.

Risk factors: SM and language problems

Another plausbile explanation for SM has been a hesitation to speak because of specific language problems. In 2003, Elizur and Perednik found that half of the children with SM had speech problems and Kirstensen (2000) found that SM children had significantly more mixed receptive-expressive language disorders and phonological disorders than their matched controls. Also, verbal social skills deficits were found in SM children (Cunningham et al., 2006). A relatively small amount of research has been conducted to explore the influence of multilingualism for children with SM because some studies have excluded multilingual

children, (Toppelberg et al., 2005). But a family background of migration was demonstrated to be a common risk factor (Dummit et al., 1997). In an Israeli study (Elizur and Perednik, 2003) selective mutism is up to four times more prevalent in immigrant versus native populations. In this study, immigrant children with SM had sufficient time in school, hence their mutism was not due to an inability to speak Hebrew.

Risk factors: SM and developmental problems

Developmental delays, like not reaching the milestones of motor development in time, are not the first thing that comes to mind when a child has selective mutism. But besides language developmental problems (Cohan et al., 2008; Kirstensen, 2002; Steinhausen & Juzi, 1996), an elevated rate of other developmental problems have been reported for children with SM (Kristensen 1997; Elizur & Perednik, 2003). Kristensen (2000) has found in a comparative study of unilingual children with SM and their matched controls, significant delays in language development as well as on gross motor skills. Immigrant children with SM were found to have lower scores on markers for (neuro)developmental delay/disorder (Elizur & Perednik, 2003).

Risk factors: SM and autism spectrum disorders (ASDs)

Very limited research is available when it comes to the co-morbidity of SM and ASDs. An extensive review (Fombonne, 2009a) has indicated that the prevalence of ASD the population worldwide is around 0.7%. An ASD is characterized by social and communicational problems, and by restricted, stereotypical behaviors (APA, 2001). Many children with an ASD have an IQ below average (de Bildt, et al., 2005) and a high percentage of them (except for children with Asperger's Syndrome) have significant language delays growing up (APA, 2001). Anxiety is also common among children with an ASD (Reaven, 2011). Although a pervasive developmental disorder is an exclusion criterion for SM in the DSM-IV-TR, some studies have reported associations between an ASD and SM (Kirstensen, 2000). It is a rare co-morbid diagnosis, but because of recent results in research it deserves our attention. There is a link between ASD and problems in social communication (Njiokiktjien, 2006). Very recently; an article has been published about a possible genetic relationship between SM and ASDs (Stein et al., 2011). A gene (CNTNAP2), that has been implicated in autism and, in the developmental language delayed component of autism, was found to be associated with risk for SM in a family-based sample and with social anxiety-related traits (behavioral inhibition and social anxiety) in a separate sample of young adults. They suggest that the SM syndrome could be more closely allied with the ASD spectrum than with social anxiety disorders. Although more research has yet to be done and the authors' conclusions are suitably cautious, this is an interesting hypothesis.

Present study

Because there is limited research about multilingualism and SM, the fact that multilingualism is up to four times more prevalent in children with SM (Elizur & Perednik, 2003; Toppelberg et al., 2005), and the growing percentage of bilingual children in the Netherlands[1], this study will use multilingualism as a factor of importance. In this study multilingualism in children will be operationalized as the capacity of a child to speak different languages, due to the fact that he or she is spoken to in a different language than Dutch at home, but can speak Dutch fluently. That last condition is very important, to make sure that children, who are not speaking because of a language acquisition period, are not misdiagnosed with SM (as mentioned by Toppelberg and colleagues (2005) that in a period of language proficiency a child can undergo a 'silent period'). Multilingualism and emotional and behavioural problems are related to each other (Pavlenko, 2005; Toppelberg et al., 2002). Also, Elizur and Perednik (2003) have revealed differences between immigrant and native children with SM on neurodevelopmental delays and (social) anxiety. Therefore it is interesting to compare a group of multilingual and a group of unilingual children with SM in the Netherlands to see if these results can be replicated in a larger sample. Because there seemed to be a genetic link between ASDs and SM, and very limited research is available about these children, a group of SM children with a co-morbid ASD were also compared to the other two groups.

The aim of the present study was to compare multilingual children with SM, unilingual children with SM, and children with SM and a co-morbid ASD. These groups were compared on several characteristics, namely anxiety, internalizing behavioral problems, stubbornness, perfectionism, disobedience at home, developmental problems, language difficulties, age at onset of symptoms, age at referral to the UMC Utrecht, total problems, and non-verbal IQ. If standardized scores are available, the SM group as a whole will be compared to those, to explore common risk factors within the entire SM group. Because of the importance of a transactional view on childhood disorders, if available, both parental and teacher ratings on behaviors were taken into account.

When significant differences between the three groups are found, recommendations for treatment can be made.

Research questions and hypotheses

Are there differences in onset of symptoms, age at referral, symptom severity, anxiety and other behavioral problems, developmental milestones, passive language IQ, or non-verbal IQ between unilingual, multilingual and autistic children with selective mutism?

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[1] Centraal Bureau voor de Statistiek, Den Haag/Heerlen (2006)

It is expected that there will be significant differences between the three groups. The ASD group is expected to have more anxiety problems, and to show more other behavioral problems. For instance, restricted behaviors, present in children with ASDs (APA, 2001), could be perceived as stubborn or disobedient behavior and are therefore more likely to be seen in the co-morbid ASD group. They are also expected to have more gross motor and language delays, and to have a lower non-verbal and passive language IQ. The multilingual SM children are expected to have a later onset of SM symptoms, to be more perfectionistic and have a lower passive language IQ, compared to unilingual SM children. Also, because immigrant families usually have less access to highly needed medical services (Toppelberg et al., 2002), they are expected to have a higher age at referral. No previous research has explored differences between the three groups on symptom severity. Despite that, expectations are that symptoms are more severe for multilingual children at school and in other places outside the home, because outside their home they face the difficult task of having to choose between two languages, if they try to speak at all.

What are common risk factors in children with selective mutism in our sample?
Anxiety problems, oppositional behavior, language problems, gross motor and language developmental delays and a co-morbid ASD are expected to be more present in our sample, compared to normalized scores. Also multilingualism is expected to be a risk factor for developing SM. No prior research has indicated that, but maybe in multilingual children the anxiety to speak is partly due to a somewhat lower passive language IQ. Symptom severity, measured by the amount of situations the child refuses to speak in, is expected to be the least at home. This is hypothesized because the DSM-IV-TR (APA, 2001) criteria refer to school situations and social gatherings as being common places the child doesn't speak in.

Method

Participants and procedures

A Dutch sample of 139 children with selective mutism was examined. They were all diagnosed with SM between 1973 and 2011 in an academic hospital (UMC) in Utrecht. Clinical and diagnostic information of these children was obtained from their clinical files. The group of 139 children was a priori split into three groups; the first group consisted of 78 unilingual SM children (56%), the second group consisted of 39 multilingual SM children (28%) and the third group consisted of 21 SM children with a co-morbid diagnosis of an ASD (15%). The last group consisted of only 2 children that are multilingual. Because of this small amount, and the limitations for data analysis with such small groups, the multilingual and unilingual co-morbid SM and ASD children were considered as one group.

Diagnosis has been established on a behavioral level at the UMC Utrecht by certified psychiatrists or psychologists, following DSM-criteria. A co-morbid diagnosis of ASD has only been made after careful examination of the behavior at home, so that silence or anxiety in unknown situations did not lead to false ASD classifications. When in doubt a final diagnosis often was given after a period of behavioral therapy for selective mutism symptoms. Among personal and socio-demographic data, the following were used in the present study: age, gender, onset of symptoms, age at referral, and school type.

Measures

Non-verbal IQ

The Non-verbal IQ-scores of the SM children at referral were obtained in the last 33 year using a variety of tests. These tests were different editions of the Wechsler Intelligence Scales (WNV-NL and performance scales of WISC-RN, WISC-III-NL, WPPSI-RN and WPPSI-III-NL; Wechsler, 1974, 1989, 1991, 2002, 2008), the simultaneous processing scale of the Kaufmann Assessment Battery for Children (K-ABC-NL, Kaufman & Kaufman, 1983), the total IQscore of the Revised Amsterdam Child Intelligence Scale (RAKIT; Bleichrodt, Drenth, Zaal & Resing, 1984) and the total IQscore of the Snijders-Oomen Nonverbal Intelligence Scale for Young Children (the SON-R 2½-7 and SON-R 5½-17; Tellegen, Winkel, Wijnberg & Laros, 1998). All of these tests allowed to calculate standardized IQ-scores ($\mu = 100$, SD = 15).

Behavioural information and anxiety

The Child Behavior Checklist, filled in by parents, and the Teacher Report Form, filled in by teachers, (CBCL and TRF) for ages 2-3, ages 4-18, ages 1,5-5 and ages 6-18[2] were used for all referrals since 1986. The parent(s) and teacher responded along a 3-point scale, with 0 = "not true", 1 = "somewhat or sometimes true", and 2 = "very true or often true." Both questionnaires consisted of multiple questions about internalizing and externalizing behavior. The subscales Anxiety problems, Internalizing problems, and Total problems and the items stubborn, perfectionism and disobedience were included in data analysis. The normalized *t* scores on subscales and the raw scores the single items were used for this study, with a score of 0 = "behavior is not present" and a score of 1 or 2 = "behavior is present". The data on the Anxiety problems scale was obtained from equal versions of the CBCL and TRF (namely the CBCL1½-5 and 6-18 and TRF1½-5 and 6-18). Therefore the cut-off scores were used that indicate if the mean level of anxiety problems was elevated compared to normalized control groups. The cut-off score for average behavioral problems is 65, for borderline problems the *t* scores must be between 66 and 69 (between the 95th and 97.5th percentile), and for clinically significant problems the *t* scores must be above 70 (above the 97.5th percentile).

SM symptom severity

The selective mutism symptom severity at referral was measured using a short speech situation questionnaire, comprising a parent and a teacher version. This instrument was developed in the UMC Utrecht in Dutch language for clinical purposes, to investigate the situations in which a child speaks. The situations were divided into three categories: 11 items represented speaking behavior at home, 9 items represented speaking behavior at school and 4 items represented speaking behavior in other environments outside the home. For example, one of the questions in the home-situation was: *"Does the child speak to unfamiliar adults?"* Items were rated on a 3 point scale indicating how frequent the child speaks in the specified situation, ranging 0 (doesn't speak), 1 (rarely speaks) and 2 (does speak). The average scores on the three situation-scales of the unilingual, multilingual and ASD group were compared. Due to insufficient research, factor structure and internal consistency remain unknown. Total scores were calculated by adjusting the mean scores of the child on each category.

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[2] Copyright of both CBCL and TRF is T.M. Achenbach, Burlington, U.S.A. Dutch translation: F.C. Verhulst, Academisch Ziekenhuis Rotterdam/Sophia Kinderziekenhuis. Versions from 1983 through 2002 have been used.

Motor and language development

In order to compare the three groups on developmental delays, milestones for language development, motor development and smiling were used. We've compared the three groups on the percentage of children with a delayed development in the categories: smile, sit, crawl, walk and speak (first words and first two-word sentences). Delays were based on the milestones of SMOCK (Sociaal Medisch Onderzoek Consultatiebureau; Herngreen et al., 1992) and represent the age when around 90% of all children in the Netherlands reached that milestone. These milestones were based on 2151 children, all born between 1988 and 1989.

Passive language abilities

For measuring passive language abilities, the Peabody Picture Vocabulary Test (PPVT-III, Dunn & Dunn, 1997, Dutch translation by Schlichting, 2004) and the Reynell Developmental Language Scales (Reynell, 1977/1985, Dutch translation by Eldik, Schlichting, Spelberg, Meulen & Meulen, 1995) were used. When both scores were available, the test date closest to the date of referral were used. When that information was unknown, the PPVT-score were used. This choice is based on the psychometric qualities of both tests, assessed by COTAN (Commissie Testaangelegenheden Nederland, 2006). These tests also allowed to calculate standardized IQ-scores ($\mu = 100$, SD = 15).

Data analysis

A one-way between-subjects ANOVA design was employed on the normally distributed scaled variables (CBCL and TRF *t* scores, non-verbal IQ scores, passive language scores, and symptom severity (home and other)) using the Statistical Package for the Social Sciences; version 16.0 (SPSS Inc., 2008). The between-subjects factor had three levels: unilingual children with SM (unilingual group), multilingual children with SM (multilingual group) and, children with SM and a co-morbid ASD (ASD-group). Normality was measured using QQ-plots. In case of significant differences between the group means, Bonferroni Post Hoc tests were performed. All *p* values were calculated as two-tailed, and *p* values less than .05 were reported as significant. Due to skewness, four variables (age at onset, age at referral, CBCL *t* scores on the scale Anxiety problems, and symptom severity (school)) were analyzed using the Kruskal-Wallis Test, a non-parametric equivalent of the one-way between-subjects analysis of variance. In case of significant differences between the mean ranks, post hoc Mann-Whitney tests were performed, using a *p* value of .0167, to assess which group differed significantly from the other groups. Single-itemscores from the CBCL and TRF on the items stubborn, disobedience and perfectionism and the developmental milestones were compared using Chi-square analysis.

Results

Personal and socio-demographic data

The male-female ratio in our group was 1:1.5. 89.7% of our sample attended regular elementary school (N=122) and 10.3% attended special education (N=14). The mean age when symptoms first occurred was 2.8 years (SD 1.03; range 1-7.75; N=90). The mean age at referral was 6.0 years (SD 1.89; range 3-12). For age at onset, there was no significant effect of group (χ^2 (2, N =90) = .3.608, p = .17). For age at referral there was no significant effect of group (χ^2 (2, N = 134) = .507, p = .78). Mean results of ANOVA – and Kruskall-Wallis tests are presented in table 1.

Table 1. Mean scores for SM children's variables, split into three groups: unilingual, multilingual and with a co-morbid diagnosis of an autism spectrum disorder.

Scale	SM (unilingual)			SM (multilingual)			SM and ASD			*p* value
	Mean	SD	N	Mean	SD	N	Mean	SD	N	
Age at onset[1]	2.66	1.11	50	3.01	0.83	26	2.87	1.05	14	NS
Age at referral[1]	5.93	1.84	74	5.85	1.69	39	6.34	2.45	21	NS
Non-verbal IQ-scores[1]	103.38	15.84	42	102.13	16.07	24	92.53	15.76	15	NS
Passive language IQ[1]	100.32	11.03	22	93.64	18.31	14	95.50	17.33	6	NS
Anxiety problems CBCL[1]	**60.50**	**9.41**	**46**	**60.19**	**8.21**	**21**	**68.27**	**7.40**	**11**	**.046***
Total problems CBCL[1]	**55.57**	**9.03**	**67**	**54.14**	**10.29**	**29**	**63.44**	**7.43**	**16**	**.004****
Internalizing problems CBCL[1]	**61.18**	**9.03**	**67**	**60.55**	**8.96**	**29**	**68.25**	**6.14**	**16**	**.009****
Anxiety problems TRF[1]	62.62	9.83	42	63.95	9.05	20	69.55	5.26	11	NS
Total problems TRF[1]	**56.31**	**7.23**	**48**	**57.71**	**5.13**	**28**	**62.07**	**9.51**	**15**	**.027***
Internalizing problems TRF[1]	**62.67**	**9.14**	**48**	**63.25**	**7.32**	**28**	**69.33**	**6.95**	**15**	**.025***
SM symptom severity (Home)[2]	1.36	0.35	31	1.27	0.37	19	1.18	0.38	8	NS
SM symptom severity (School)[2]	**0.84**	**0.49**	**30**	**0.52**	**0.50**	**19**	**0.51**	**0.35**	**8**	**.035***
SM symptom severity (Other)[2]	1.12	0.43	30	1.00	0.60	19	0.75	0.48	8	NS

Note: Results for the Age at onset, Age at referral, Anxiety problems CBCL and SM symptom severity (school) are obtained using Kruskal-Wallis tests. Other results are obtained using ANOVA.
1: A higher score represents a higher age, higher IQ, and more problems
2: A lower score represents more severe symptoms
* p < .05; **p < .01; ***p < .001 (two-tailed). NS = not significant.

Non-verbal and passive language IQ

As shown in table 1, for non-verbal IQ, there was no significant effect of group ($F(2,78)$ = 2.660, p = .08). The ASD group had a slightly lower mean on non-verbal IQ when compared to standardized non-verbal IQ-scores (μ_{ASD} = 92.53, μ_{normal} = 100). The range of all three groups, shown in SD (standard deviation), was almost similar to the standardized scores ($SD_{unilingual}$ = 15.84, $SD_{multilingual}$ = 16.07, SD_{ASD} = 15.76, and SD_{normal} =15.0). For passive language IQ, there was no significant effect of group ($F(2,39)$ = .942, p = .40). The multilingual and ASD groups did show some deviations in mean and range when compared to standardized passive language IQ-scores ($\mu_{multilingual}$ = 93.64, μ_{ASD} = 95.50, μ_{normal} = 100.0;

$SD_{multilingual} = 18.31$, $SD_{ASD} = 17.33$, and $SD_{normal} = 15$). The different ranges mean that the scores were more widely spread in the distribution, meaning there was more variety among the scores. For instance, in current data 68% of unilingual SM children had a passive language IQ between 89 and 111, multilingual SM children had a passive language IQ score between 75 and 111, and SM children with an ASD had a passive language IQ score between 78 and 112.

Parent-reported problems

Anxiety problems

As shown in table 1, for the anxiety problems scale of the CBCL, there was a significant effect of group (χ^2 (2, $N = 78$) = 6.146, $p < .05$). Post Hoc Mann-Whitney U testing showed that there was no statistically significant difference between unilingual and multilingual children with SM in *t* scores for Anxiety problems on the CBCL ($U = 558.000$, $N_1 = 56$, $N_2 = 22$, $p = .52$, two-tailed). Considering the unilingual and multilingual children as one group, there was a significant difference between the ASD group and the total SM group with no co-morbid ASD on the Anxiety problems scale of the CBCL ($U = 196.000$, $N_1 = 66$, $N_2 = 11$, $p = .02$, two-tailed). Comparing the scores on parent reported anxiety problems in the entire group to the percentiles in normal development, much more borderline and clinical scores were seen in the SM group. These percentages are presented in table 2.

Table 2. Percentage of SM children's scores on the CBCL anxiety scale and TRF anxiety scale compared to a normal population

Anxiety problems	Average	Borderline	Clinical
Normal population	92.5	5	2.5
SM children (parent reported)	56.4	14.1	29.5
SM children (teacher reported)	53.4	17.8	28.8

Total problems

For the total problems scale of the CBCL, the means of the three groups differed significantly ($F(2,109) = 5.861$, $p < .01$). Post Hoc Bonferroni testing showed that for the ASD-group significantly more total problems were reported by parents than for the unilingual group ($p < .01$) and the multilingual group ($p < .01$). Between the multilingual and unilingual groups there were no significant differences ($p > .05$).

Internalizing problems

For the internalizing problems scale of the CBCL, there was a statistically significant effect of group ($F(2,109) = 4.866$, $p < .01$), with the ASD group significantly differing from the

unilingual (p = .01) and multilingual (p = .02) group. The multilingual and unilingual groups did not differ significantly (p > .05).

Oppositional behaviors (stubborn behavior and disobedience) and perfectionism

As shown in figure 1, for the CBCL-item stubborn, 72.5% of the unilingual children (N = 51), 56.5% of the multilingual children (N = 23), and 85.7% of the ASD children (N = 14) were reported as being stubborn by their parent(s). Chi-square analysis showed no significant relationship between stubbornness and group (χ^2(2, N = 88) = 3.818, p = .152). On perfectionism, 45.0% of the unilingual children (N = 20), 41.7% of the multilingual children (N = 12), and 55.6% of the ASD children (N = 9) were reported as being a perfectionist by their parent(s), but there was no significant relationship between perfectionism and group (χ^2(2, N = 41) = .427, p = .845). For disobedience, 51.0% of the unilingual children (N = 49), 37.5% of the multilingual children (N = 24), and 50.0% of the ASD children (N = 14) were reported as being disobedient by their parent(s), but there was no significant relationship between disobedience and group (χ^2(2, N = 87) = .1.237, p = .529). 70.5% of the entire group of SM children (N = 88) was reported stubborn by their parents, 46.3% (N = 41) was reported being a perfectionist, and 47.1% (N = 87) was reported being disobedient at home (figure 2).
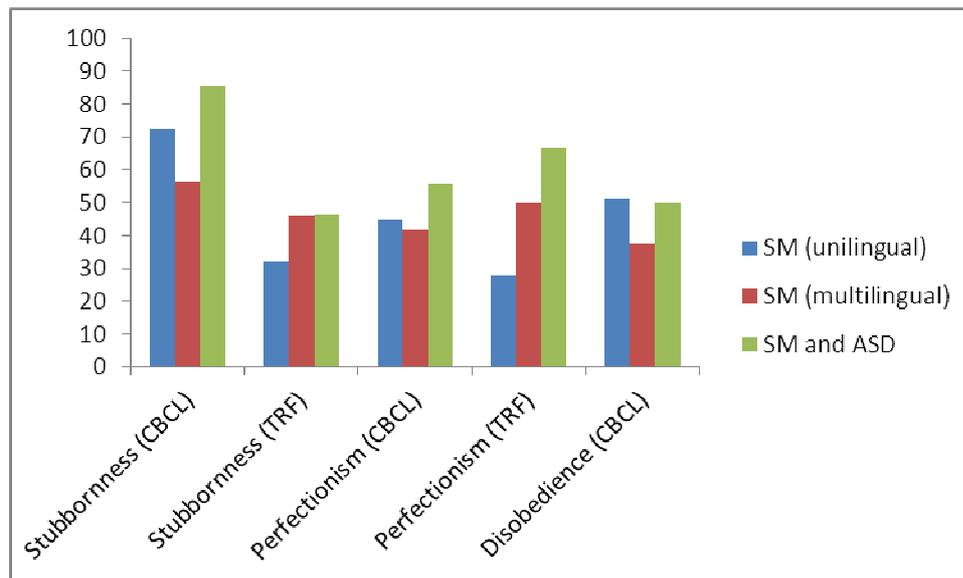

Figure 1. *Percentage of children with SM that were reported as being stubborn, being perfectionist, and being disobedient at home, split into three groups: unilingual, multilingual and with a co-morbid diagnosis of an autism spectrum disorder.*
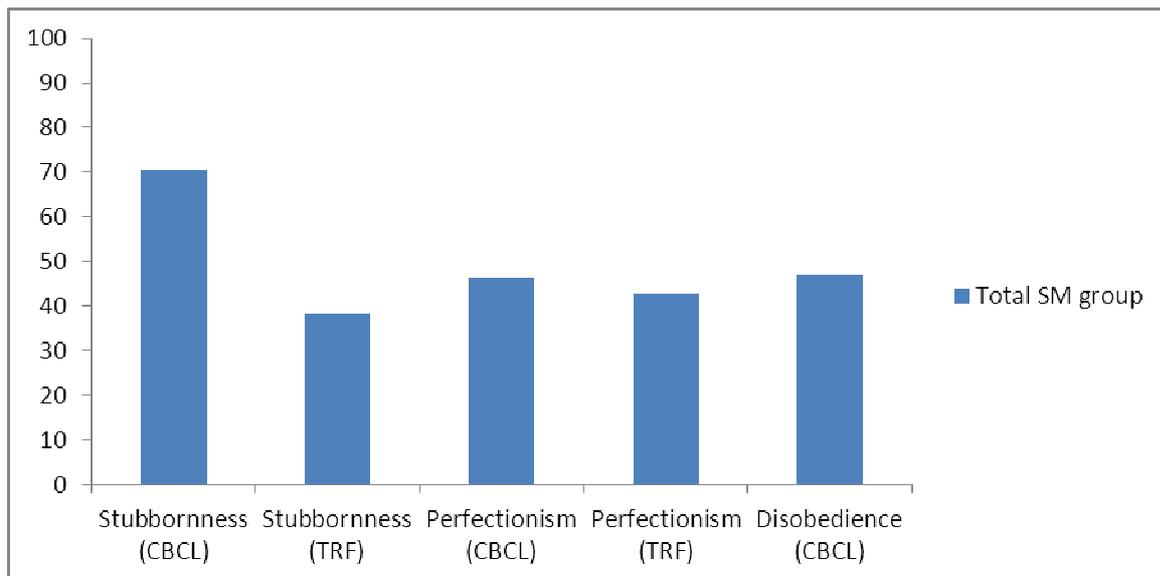
Figure 2. *Percentage of total group of children with SM that were reported as being stubborn, being perfectionist, and being disobedient at home.*

SM symptom severity

For SM symptom severity on the 'home' situation, there was no significant effect of group ($F(2,55) = .890$, $p = .42$). On all groups, the lowest mean scores were obtained in the school situation, meaning overall severity at school was higher, with the SM children speaking less in school. On the 'school' situation, Kruskal-Wallis testing showed there was a significant effect of group (χ^2 (2, $N = 57$) = 6.697, $p = .04$). Post Hoc Mann-Whitney U testing, using a p value of .0167, showed that there is no statistically significant difference between the multilingual and unilingual children on the 'school' situation of the SM severity scale ($U = 241.000$, $N_1 = 37$, $N_2 = 20$, p .03, two-tailed). Also, there was no significant difference between the ASD group and the multilingual group ($U = 60.000$, $N_1 = 7$, $N_2 = 19$, $p = .72$). The two multilingual children with an ASD were taken out of this equation. Between the ASD group and unilingual group, there were significant differences as well ($U = 72.000$, $N_1 = 8$, $N_2 = 30$, $p = .09$). For the 'other places outside the home' situation, there was no significant effect of group ($F(2,54) = 1.799$, $p = .18$).

Teacher-reported behavioral problems

Anxiety problems

As shown in table 1, for the anxiety problems scale of the TRF, there was no significant effect of group ($F(2,70) = .2.527$, $p = .09$). A comparison of the total SM group no a normal population is presented in table 2.

Total problems

For the total problems scale of the TRF, the means of the groups differed significantly (F(2,88) =, p = .03). Post Hoc testing showed that for the ASD-group significantly more total problems were reported by the teachers in comparison to the unilingual group (p = .02) but not compared to the multilingual group (p = .18). Between the multilingual and unilingual groups there were no significant differences (p > .05).

Internalizing problems

For the internalizing problems scale of the TRF, there was a statistically significant effect of group (F(2,88) = 3.839, p = .03), with the ASD group significantly differing from the unilingual group (p = .02), but not from the multilingual group (p = .08). The multilingual and unilingual groups did not differ significantly (p > .05).

Oppositional behaviors (stubborn behavior) and perfectionism

As shown in figure 1, for the TRF-item stubborn, 31.9% of the unilingual children (N = 47), 45.8% of the multilingual children (N = 24), and 46.2% of the ASD children (N = 13) were reported as being stubborn by their teacher(s), but these differences were not significant (χ^2 (2, N = 84) =1.729, p = .431). For the item perfectionism, 27.8% of the unilingual children (N = 18), 50.0% of the multilingual children (N = 8), and 66.7% of the ASD children (N = 9) were reported as being a perfectionist by their teacher(s), but these differences were not significant as well (χ^2 (2, N = 35) = 3.921, p = .170). For the entire SM group, 38.1% of the children were reported as being stubborn by their teachers (N = 84) and 42.9% was considered a perfectionist (N = 35) (as shown in figure 2).

Motor and language development

The percentages and total amount of children that have delayed gross motor and/or language development are presented in table 3. A more graphic overview of the results is given in figure 3. Considering that 90% of normal developing children reached these milestones for that given age, all groups showed developmental delays for the items smile, walk, and speak (shown in figure 4). The differences between the groups were not significant for the items smile, (χ^2 (2, N = 74) = 5.396, p = .063), sit (χ^2 (2, N = 81) =2.208, p = .366), crawl (χ^2 (2, N = 63) = 3.052, p = .282) and walk (χ^2 (2, N = 96) = 2.666, p = .277). The unilingual and ASD group showed more delays in language development for the item speak (words). The ASD group showed rather large delays in both items of language skills (speak words and speak 2-word sentences). Chi-square testing showed that there is a relationship between speaking first words and group (χ^2 (2, N = 85) = 6.500, p = .041) and between speaking of the first two-word sentences and group (χ^2 (2, N = 76) = 7.881, p = .019).

Analysis of the results implicates that for speaking of the first word, less multilingual and more ASD children have delayed speech than was expected. Also, for speaking of the first two-word sentence, less unilingual and multilingual children had delayed speech compared to chi-square expectations.
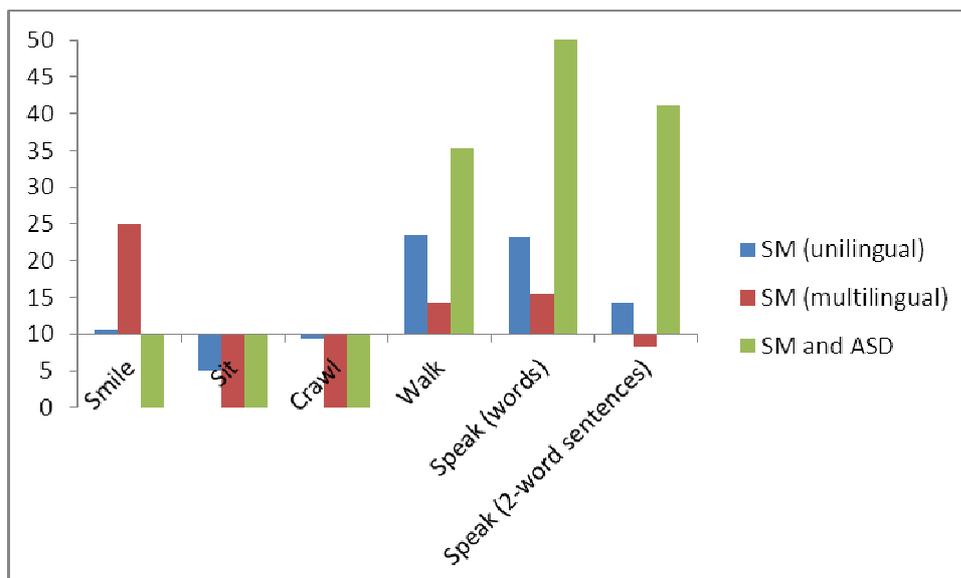
Table 3. Percentage of SM children with a delayed motor and/or language development, split into three groups: unilingual, multilingual and with a co-morbid diagnosis of an autism spectrum disorder.

Developmental Milestone	SM (unilingual) Percentage delayed	N	SM (multilingual) Percentage delayed	N	SM and ASD Percentage delayed	N	*p* value
Smile	10,5	38	25	20	0	16	NS
Sit	5,1	39	0	26	0	16	NS
Crawl	9,4	32	0	20	0	11	NS
Walk	23,5	51	14,3	28	35,3	17	NS
Speak (words)	**23,3**	**43**	**15,4**	**26**	**50**	**16**	**.041***
Speak (2-word sentences)	**14,3**	**35**	**8,3**	**24**	**41,2**	**17**	**,019***

Figure 3. Percentage of SM children with a delayed motor and/or language development, split into three groups: unilingual, multilingual and with a co-morbid diagnosis of an autism spectrum disorder.
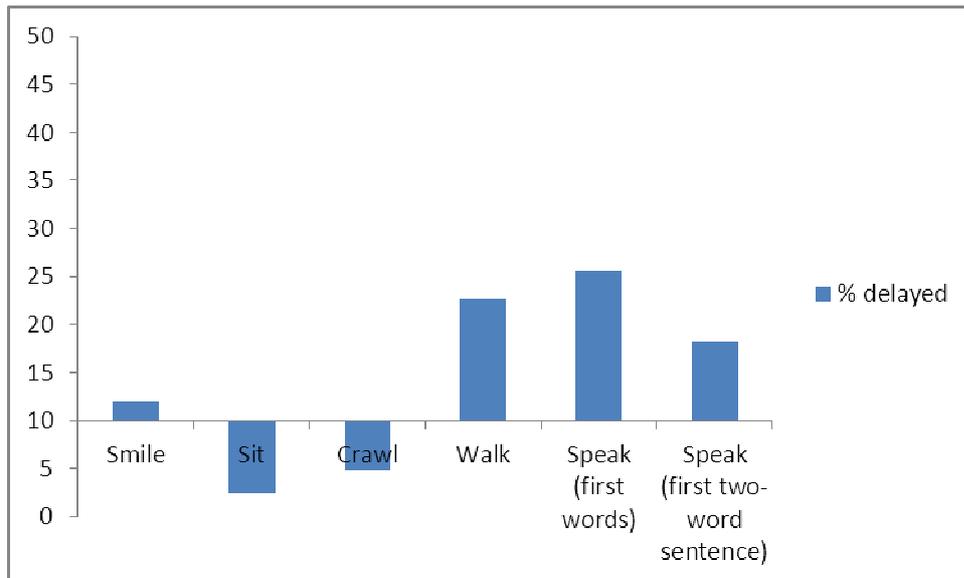
Figure 4. *Percentage of SM children with a delayed motor and/or language development.*

Discussion

This study has compared three groups of Dutch children with selective mutism: unilingual children, multilingual children, and children with a co-morbid autism spectrum disorder. Also, common risk factors derived from literature are explored for the total group of 139 SM children, referred for diagnosis and treatment to the academic hospital in Utrecht (UMCU) between 1973 and 2011.

Are there differences between unilingual, multilingual and autistic children with selective mutism?

Our results indicated that the three groups of SM children showed many similarities, but also showed some important differences. Similarities were found between the three SM groups on age at onset of symptoms, age at referral, on non-verbal IQ scores, and on gross motor developmental delays. Also, on parent reported disobedience, parent – and teacher reported stubbornness, and perfectionism, the groups did not differ significantly. Almost all significant differences were found when the ASD group was compared to the other two groups. On anxiety problems, internalizing behavioural problems, and total behavioural problems, the ASD group showed significantly more parent reported anxiety problems, and more parent – and teacher reported internalizing behavioural problems and total behavioural problems. On language development, the ASD children with SM showed significantly more language developmental delays compared to the other two groups. The only differences between the unilingual and multilingual SM children were found on language problems and severity of symptoms. Compared to multilingual SM children, more unilingual SM children were delayed in speaking their first words. On symptom severity, the unilingual children spoke significantly more in school compared to the children in the other two groups. On passive language IQ, both multilingual and ASD SM children showed more scores below average, and especially a wider range in scores, compared to the unilingual children with SM. However, these differences did not reach significance, which might be due to the relatively small sample size of these two groups.

What are common risk factors in children with selective mutism?

Almost 30% of all SM children showed clinically significant anxiety problems, as reported by both parents and teachers, which is over 10 times more common compared to a normal population. Around 15% of them showed borderline severe anxiety problems in parent – and teacher reports, which is also three times more common compared to a normal population. Therefore anxiety is highly common among children with SM and may be a risk factor for developing SM. Perfectionism, related to anxiety, seemed to be highly present among the total group, with almost half of the children reported as perfectionist by their parents and

teachers. In all SM children, the severity of the symptoms was most apparent in the school situation. Oppositional behavioural problems were very common, as almost three quarter of all SM children was reported as stubborn and half of all SM children as disobedient by their parents. Teachers reported over a third of the children as stubborn. Stubborn and disobedient behaviour could be factors related to SM. When compared to developmental milestones, based on the Dutch population, a higher percentage of SM children showed developmental delays. These delays were smiling, walking, speaking of the first word and first two-word sentence. They can be risk factors for developing SM, because these delays happened before the onset of symptoms. Multilingualism was present in almost a third of the children with SM, which seems to be a rather large amount and gives rise to the assumption that multilingualism is a risk factor for developing SM as well. Finally, ASDs are over 20 times more present in our SM group, compared to the population world-wide (Fombonne, 2009a). Because the onset of ASD is usually before the onset of SM, an ASD could be a risk factor for developing SM.

Conclusions with respect to previous research

The mean onset of SM symptoms in this study is 2.8 years, which is similar to previous research (Steinhausen & Juzi, 1996). Not all of the results were in line with earlier expectations. Because of no significant differences in age at referral, the statement that immigrant parents have less access to psychiatric facilities, is probably not true for the Netherlands (in contrast to Israeli and American studies, Elizur & Perednik, 2003; Toppelberg et al., 2002).

SM symptom severity

The SM symptoms in unilingual children in the school situation are severe, but less severe compared to the other groups. This could be because the multilingual SM children face an extra difficulty in having to decide in which language to speak, if they speak at all. For all ASD children, school can be an overwhelming place, and that could contribute to the severity of the SM symptoms in the school situation for the SM children with a co-morbid ASD.

SM and anxiety

As expected, anxiety problems were present among the entire group of SM children (Vecchio & Kearney, 2005; Dummit et al., 1997; Cunningham et al., 2004; Cohan et al., 2008; Cunningham, McHolm, & Boyle, 2006; Carbone et al., 2010; Ford et al., 1998), and more present for the ASD SM children (Reaven, 2011). Perfectionism was expected to be highly present in all children, because it correlates with social stress (Hewitt et al., 2002), and that seems to be the case in this study. To prove this assumption the group has to be compared

to a normalized, age – and gender matched control group. It was also expected that perfectionism was higher among multilingual children, because sometimes immigrant parents have more achievement-driven parenting styles (Pels, 2004). That was not the case in this study, so probably there are no differences between the groups on perfectionism. Another possibility is that it is not found in this study because of a rather small group size and because the results were based on just one question in the questionnaire. A third reason could be that perfectionistic behaviour is not recognised as being perfectionistic by parents with a high-achievement parenting style.

SM and oppositional behavior

In previous research (Kristensen, 1997; Sharp, Sherman & Gross, 2007; Anstendig, 1999; Perednik & Elizur, 2003; Omdal, 2007) SM children were described as stubborn and disobedient. High rates of stubbornness and disobedience at home were also found in our study and a high percentage of the SM children were, as was hypothesized, found to be more stubborn at home compared to school. This difference could be present because the SM children feel more freedom at home to show their apparently strong will. With the lack of a normalized, age – and gender matched control group, and the fact that these results are only based on a single item in a questionnaire, we cannot prove that these characteristics are more present in all SM children. In contrary to our expectations, which stated that children with a co-morbid ASD could be perceived as stubborn due to restricted behaviours, highly present in ASD children, they did not show more stubborn behaviours compared to the other groups. Although this is also based on a single-item in a questionnaire and the sample size in the ASD group is not very large, the assumption can be made that all SM children show the same (high) rate of stubbornness.

SM and language problems

Although the elevated percentage of delayed language development in the total SM group is somewhat biased by the high amount of language delay in the ASD group, language problems seem to be present in all three groups. The fact that more children with SM had delayed language development, compared to developmental milestones, is in line with previous studies (Cohan et al., 2008; Kirstensen, 2002; Steinhausen & Juzi, 1996; Kristensen, 2000). Therefore language developmental problems, and multilingualism, seem to be a risk factor, which supports previous research (Elizur & Perednik, 2003; Kristensen, 2000; Dummit et al., 1997).

SM and developmental problems

As previous research indicated (Kristensen 1997; Elizur & Perednik, 2003) more children in our total SM group had gross motor delays, compared to a normal population. Because Elizur and Perednik (2003) found differences in characteristics between native and immigrant children with SM on motor development, similar results were expected in this comparison of unilingual and multilingual children. Only small differences were seen in our data, but probably due to a rather small sample size they were not significant.

SM and autism spectrum disorders (ASDs)

The fact that a higher percentage of language delays are also present in the other two groups supports previous research (Stein et al., 2011). They indicated that a gene (CNTNAP2), implicated in the language delay component of autism is associated with a risk for SM. As expected from DSM-IV-TR criteria for autism, the ASD children had significantly more language delays compared to the unilingual and multilingual SM children. Contrary to expectations, obtained from previous research (de Bildt, et al., 2005), the ASD group doesn't show a significantly lower non-verbal IQ score, compared to the other groups. This leads to the assumption that SM may be more present in children with ASD with an average IQ, but may as well be caused by the selection of patient care in the academic hospital (where the main focus is on high functioning autism).

Strengths and limitations

Data on SM children has been collected over a long period of time, and has therefore been able to explore data of 139 children with selective mutism. This sample size is considered to be one of the largest sample sizes known in international SM research in history. Also, because of the high amount of different characteristics that have been taken into account, this study gives a very broad insight in common risk factors and differences in unilingual, multilingual and ASD children with SM.

Besides these strengths, this study also has some limitations. A total group size of 139 is very large, but data was not available for every child in our group. This is because in 38 years some information was lost, or simply not obtained. When the group was split up in multilingual, unilingual and children with a co-morbid ASD, on the last group in some cases the group size was only 10 or less. Also, as mentioned earlier, because of the lack of a matched control group on stubbornness, disobedience and perfectionism, no conclusions can be drawn from these results.

Another point of consideration is the fact that passive IQ scores below 85 (clearly below average) have been reported for a small amount of the clinical group. Following the DSM-IV-TR criterion carefully, stating that children that have a lack of knowledge of, and

comfort with, the spoken language required, cannot be diagnosed with SM. Maybe these children should not have had a diagnosis of SM at all.

Because the data have been collected over a 38 year time period, it has not been possible to use the same psychometric instruments. For instance, the PPVT-III (Dunn & Dunn, 1997, Dutch translation by Schlichting, 2004) and Reynell Developmental Language Scales (Reynell, 1977/1985, Dutch translation by Eldik, Schlichting, Spelberg, Meulen & Meulen, 1995) have both been used to measure passive language IQ. Although the mean scores of both tests were comparable, the distribution was not. Also, to measure non-verbal intelligence, nine different tests have been used, with different psychometric qualities and limitations. Both the items 'passive language IQ' and 'non-verbal IQ' could have been biased, due to the use of different psychometric instruments.

Clinical implications

The severity of symptoms at school is higher in multilingual and ASD SM children, making them a more vulnerable subgroup. Especially anxiety and internalizing behavioral problems are higher in the ASD subgroup. With 28% multilingual children and 15% children with a co-morbid ASD in our database, it's hard to ignore the fact that SM is relatively common among multilingual and autistic children. As implicated, it is possible that the unilingual children speak significantly more in the school situation compared to the other groups, because of somewhat lower passive language scores in the ASD and multilingual SM group. Therefore it is advised to always obtain information about the passive language IQ on a child with SM, especially on a multilingual child or a child with a co-morbid ASD. Therefore, the effectiveness of the treatment could be increased by a more adjusted approach to the child with SM. The children could be split into these three groups beforehand and the characteristics that are more common among that specific group could be a factor that deserves more attention in treatment. For instance, when scores on passive language IQ are significantly lower for multilingual children, expanding a child's vocabulary could mean a significant diminishing of the symptoms. Because a high percentage of the children are perceived by their parents and/or teachers as being stubborn, perfectionistic, and disobedient, more education to the parents about this disorder, highly related to anxiety and not to externalizing behavioral problems, should be considered.

Also, because the severity of the SM symptoms is seen most in the school situation, it is recommended that careful observations are made at home as well as at school. For children with a co-morbid ASD, it is advised to be aware of the fact that these children show more internalizing behavioural problems and more anxiety problems, and a higher percentage of these children have delayed speech, compared to developmental milestones.

Implications for future research

The first recommendation for future research is to conduct a follow up study. If SM is characterized by developmental delays, future outcomes for these children should be collected and analyzed, especially with respect to motor development, language skills, anxiety, co-morbid disorders and behavioral problems. Omdal (2007) interviewed six recovered ex-patients and has given some interesting insights in the development of the disorder. All the informants described incidents they had found traumatic in their childhood, and in three cases it was associated with the onset of their selective mutism. In a follow up study on SM, traumatic experiences should be explored in recovered ex-patients.

In the present study, unilingual and multilingual SM children did not show significant differences on anxiety, oppositional behaviors, language problems and developmental problems. On these factors, the groups can probably be considered similar. Maybe with more extended measures for oppositional behavior, a larger sample size and more insight in parenting styles in these two groups of SM children, significant differences could be found. With a multiple regression analysis, the predictability of these factors on the development of SM can be explored. It is implicated for future research to conduct such a research study.

A higher percentage of SM children have shown language delays compared to children in the Dutch population, and there is no difference between the groups on the age at onset of the symptoms. Maybe SM and ASD are related to each other. The relation between SM, ASD and SM with co-morbid ASD is hard to explore because of the restrictions in the DSM-IV-TR (APA, 2001) criteria, but more research is advised on the possible relation between these two childhood disorders.

With respect to exploratory theories that state that SM is related to problems in parent dyads (Nowakowski et al., 2010), the attachment theory by John Bowlby (1907 – 1990) could be an underlying theory in the development of SM. In one study (Nowakowski et al., 2010) the SM children withdrew from their parents during stressful situations, in contrast to normal developing children and children with an anxiety disorder, which engaged in joint attention dyads with their parent(s) during stressful situations. Social smiling, delayed in a higher percentage of SM children, compared to developmental milestones, could mean an underlying attachment problem. For future research, more attention could be paid on parent dyads in SM, which could give more insight into the development of this rare and fascinating disorder.

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