

A randomized controlled trial of a home and school-based intervention for selective mutism – defocused communication and behavioural techniques

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Background: Randomized controlled psychosocial treatment studies on selective mutism (SM) are lacking. **Method:** Overall, 24 children with SM, aged 3–9 years, were randomized to 3 months treatment ($n = 12$) or wait list ($n = 12$). Primary outcome measure was the School Speech Questionnaire. **Results:** A significant time by group interaction was found ($p = .029$) with significantly increased speech in the treatment group ($p = .004$) and no change in wait list controls ($p = .936$). A time by age interaction favoured younger children ($p = .029$). Clinical trial registration: Norwegian Research Council INCT01002196. **Conclusions:** The treatment significantly improved speech. Greater improvement in the younger age group highlights the importance of an early intervention.

Key Practitioner Message

- Early detection and treatment of children with selective mutism is crucial due to the elevated risk for chronicity of symptoms.
- The preschool arena is an important setting for the recognition of anxious and withdrawn behaviour that often precedes selective mutism.
- Evidence supports the use of a home- and school-based psychosocial intervention to increase speech.
- Greater improvement in younger children (preschool) highlights the importance of early intervention.

Keywords: Selective mutism; randomized controlled trial; behavioural intervention; social phobia; childhood anxiety

Introduction

Children with SM are characterized by a consistent lack of speech in specific social situations in which there is an expectation for speaking (e.g. school) despite speaking in other situations (e.g. home) (American Psychiatric Association, 2000). SM is found to be associated with comorbid anxiety diagnoses, in particular with social anxiety disorder, as well as neurodevelopmental disorders (Kristensen, 2000). Age of onset is typically before age 5 years (Black & Uhde, 1995). SM is relatively rare, with a prevalence of about 0.7–0.8% in childhood, somewhat more frequent in girls (Bergman, Piacentini, & McCracken, 2002). A 0.1% prevalence of SM was found in a representative sample of Norwegian 4 year olds using a preschool diagnostic interview (Wichstrom et al., 2012).

SM is considered to be hard to treat and the literature on treatment for SM is dominated by case studies or case series. One review describes that the few existing studies (since 1980) with samples above five children have

mainly used behavioural, cognitive-behavioural or multimodal interventions (Manassis, 2009). Another review states that behavioural interventions (including stimulus fading, desensitization, shaping, modelling and contingency management) are most frequently used (Cohan, Chavira, & Stein, 2006). Only two studies include a comparison group. The first study examined retrospectively the records for 25 children with SM treated either with behavioural therapy or a school-based remediation programme. The children treated with behavioural therapy showed a greater improvement compared to the group which received the school-based remediation intervention (Sluckin, Foreman, & Herbert, 1991). In the second and more recent study (Vecchio & Kearney, 2009) an alternating treatment design was applied in nine children with SM. Greater effectiveness was described for exposure-based practice versus contingency management. Children, parents and teachers rated outcome in terms of words spoken, and the reported effect sizes suggested improvement by children

and parents with somewhat less favourable teacher ratings. More treatment studies with sound methodology, larger sample sizes, and systematic descriptions of the treatment content and duration, diagnostic procedures and evaluation instruments, are needed. To date no prospective psychosocial randomized controlled treatment (RCT) studies of children with SM are published.

In Norway, the first and last author established a multidisciplinary project group in 2007 and subsequently developed a home and preschool/school-based intervention for children with SM 3–9 years of age.

The intervention includes defocused communication, psychoeducation and behavioural techniques.

Defocused communication (developed based on clinical experience with a considerable number of children with SM) was used as a general treatment principle to decrease social anxiety (see Content of sessions, page 8).

In line with the previously mentioned review of the treatment literature (Cohan et al., 2006) psychoeducation was chosen along with behavioural interventions [stimulus fading in the form of gradually increased exposure as well as contingency management (use of positive reinforcement for speaking behaviour)] that were applied in a joyful play activity inspired by the Selective Mutism Resource Manual (Johnson & Wintgens, 2007).

We found favourable treatment outcome in a pilot study of seven preschool children (five girls and four bilinguals) diagnosed with long-standing SM (mean 20 months) (Oerbeck, Johansen, Lundahl, & Kristen-

sen, 2012). Six children spoke freely in all preschool settings after a mean of 14 weeks treatment, and at follow-up 1 year after end of treatment. These children were recruited from the local Oslo area, and the members of the project group treated six of the seven children.

This article will present results from the RCT component of a study using this intervention involving 24 children with SM 3–9 years of age recruited from Southern Norway and treated by local therapists at community health clinics. The children were randomized to 3 months of treatment or to 3 months as wait list controls.

Due to the situational nature of SM, we hypothesized that the children in the active treatment group, compared to controls would show increased speaking behaviour (rated by teachers and parents) in preschool/school, where the targeted problem behaviour and the treatment took place.

Given an increased neurodevelopmental plasticity and less entrenched mutism in young children, we further hypothesized that younger children would benefit more than older ones.

Method

Design

This is a randomized controlled trial (RCT) of a psychosocial intervention for SM where participants were allocated to the intervention group or to the wait list controls. Figure 1 shows the participant flow through the trial.

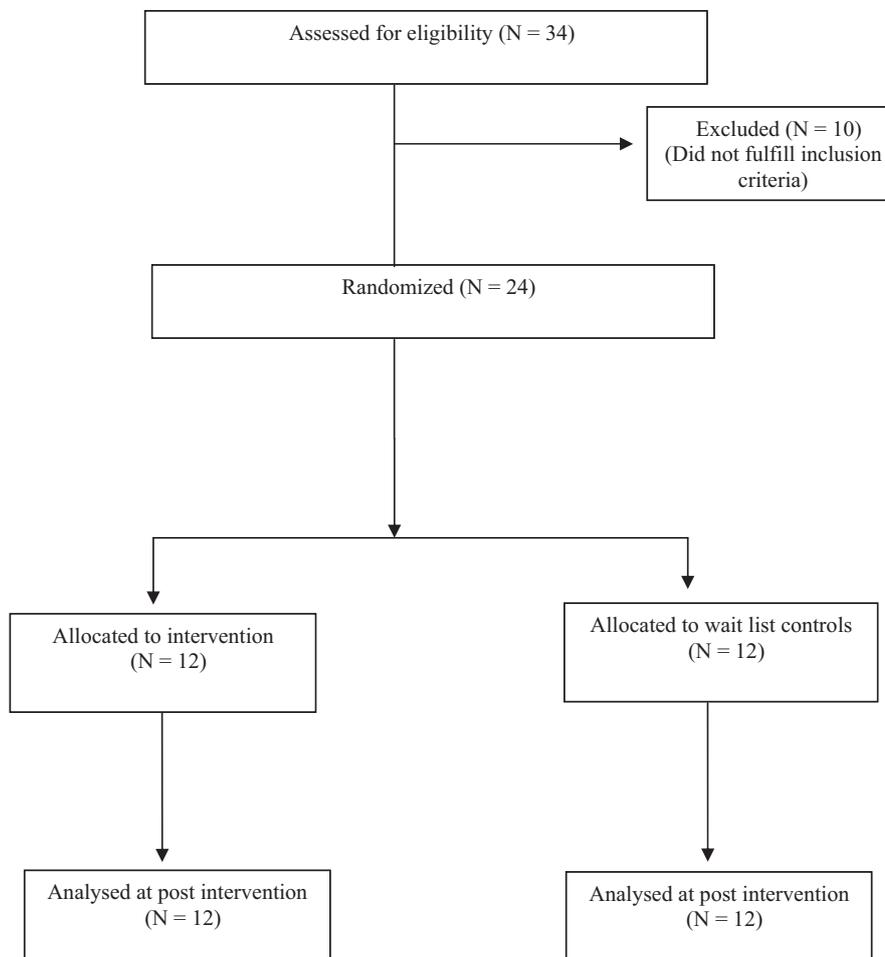


Figure 1. The flow diagram of participants in each stage of the RCT

Participants

The sample consists of 24 children with SM, 3–9 years of age [16 girls, mean age 6.5 years ($SD = 2.0$), 9 children in preschool; age 3–5 years, 15 school children; age 6–9 years]. The exact onset of SM was hard to pinpoint for the parents, especially for the older children, but the long-standing nature of SM was highlighted, and most mentioned the start in preschool as a crucial point in symptom development. Six children were bilingual. Twenty-two children lived with both biological parents (one adopted child, one child with divorced parents). The educational level of the parents was ≤ 12 years ($N = 23$) and >12 years ($N = 25$) (See Table 1 for background variables).

Inclusion criteria: Children aged 3–9 years, consecutively referred for SM during a 9-month period from outpatient Child and Adolescent Mental Health Clinics (CAMHS) or school psychology services in Southern Norway, whose parents consented to the randomization procedure and fulfilled DSM-IV diagnostic criteria for SM. In addition, we specified that the children should not speak to adults in preschool/school, and that mutism was present also in the native language for bilingual children.

Exclusion criteria: 1. Parents who did not speak Norwegian or 2. Children with IQ <50 , psychosis or a Pervasive Developmental Disorder. 3. Children who were on psychotropic medication or receiving another active treatment for SM.

Power

On the basis of the recruitment to our pilot study, we found it likely that a minimum of 24 children would be referred within the planned time frame and the extended geographical area of this study. The pilot study showed that six children started to talk in preschool within 3 months. A power analysis showed that with 24 children, 80% power was achieved if 75% of the children (9 of 12) in the treatment group started to talk within 3 months compared to the possibility of a spontaneous

onset of talking in 25% of the wait list controls (3 of 12) during the 3-month waiting list period.

Recruitment

The CAMHS and school psychology services in Southern Norway received written information about the treatment study resulting in 34 age-appropriate referrals. All parents were contacted by phone by the principal investigators (first and last author) and interviewed about the SM symptomatology and the general development of the child, and the mother completed the Selective Mutism Questionnaire (SMQ) (Bergman, Keller, Piacentini, & Bergman, 2008). This screening led to the exclusion of 10 children [Pervasive Developmental Disorder ($N = 2$), no mutism in native language ($N = 1$), use of speech to some adults in preschool/school ($N = 7$)]. A phone call to the local CAMHS was made to ensure that all remaining 24 children were registered, and that a local therapist would be made available. The final inclusion of the 24 children was based upon a confirmation of the SM diagnosis after a home visit where a diagnostic interview was conducted with the parents (by the last author), and the children were assessed (by the first author) to rule out severe intellectual and/or language problems. In addition, parents and the staff/teachers in preschool/school completed questionnaires. All the baseline assessments were made blind to the randomized group allocation.

Randomization procedure

The randomized group allocation was made by the fourth author based on information from the screening interview. To reduce the risk of imbalance regarding four predefined variables (symptom severity as assessed by SMQ, age group (preschool/school), bilingualism and gender) the children were allocated to the treatment or control group according to the algorithm of Hofmeijer, Anema, and van der Tweel (2008). This method is a modified minimization procedure that involves a random component. The parameter (between 0 and 1) that controls the degree of randomness was set to 0.4. When using this procedure the four variables had equal weight and the method was restricted to ensure 12 children in each group. The algorithm was implemented by using the Matlab programming language (Natick, MA). The results of the randomized group allocation was kept in a sealed envelope by the fourth author and revealed to the principal investigators after baseline assessments had taken place.

Initial psychoeducation of all participants

For each of the 24 participants, one initial psychoeducational session (including information about SM, as well as on how to use defocused communication) was held by phone with the staff/teachers from preschool/school and parents together. The practical arrangement for participants in the treatment group was also planned in this phone call, while the wait list group only received information on the assessment and treatment procedures that would take place after 3 months, with no further contact with the investigators or therapists during this time period.

Therapist training

The 12 children in the treatment group were treated by a total of 10 therapists registered to a local CAMHS who

Table 1. Background variables for children in the intervention group ($N = 12$) and waitlist group ($N = 12$)

	Intervention group $N = 12$	Wait list group $N = 12$
Gender	3 boys	5 boys
Age group	$N = 5$ in preschool	$N = 4$ in preschool
Severe SM (SMQ School subscale ≤ 0.5)	$N = 8$	$N = 8$
Bilinguals	$N = 3$	$N = 3$
Educational level mother ≤ 12 years/ >12 years	$N = 5/N = 7$	$N = 7/N = 5$
Educational level father ≤ 12 years/ >12 years	$N = 6/N = 6$	$N = 5/N = 7$
Age mean (SD)	6.59 (2.35)	6.47 (1.53)
Vocabulary (PPVT) mean (SD)	92.50 (8.98)	97.08 (9.23)
Non-verbal IQ mean (SD)	97.08 (11.77)	98.33 (8.35)
Children with comorbid diagnoses other than SM/Social Phobia	$N = 8$	$N = 8$
SM in other family members	$N = 5$	$N = 5$

received a detailed manual describing the use of defocused communication as a general treatment principle and how to arrange the behavioural interventions of the stimulus fading/sliding-in technique. The manual required the checking off of planned tasks and interventions in each session, and all therapists used the treatment manual under close guidance and supervision related to each session by phone from the first or last author. No further treatment adherence measures were included in this study. All but one therapist had a minimum of 10 years of clinical experience including some ($N = 4$) or extensive ($N = 5$) work with selectively mute children.

Intervention

Frequency, location and participants. The intervention group received a total of 21 sessions by the therapist over a 3-month period. The first three 1-hr sessions were carried out weekly in the child's home with the parents present, primarily to get to know the child, establish an alliance and train on procedures that later would be carried out in preschool/school. The next sessions took place at preschool/school twice a week, each lasting half an hour. The intervention at preschool/school was divided into six modules/speaking levels according to the progress of the child (see Table 2). The parents participated in the first module; the teachers from modules III to VI and peers/classmates from modules IV to VI.

Content of sessions. Defocused communication and behavioral interventions (including rewards) were our two main components.

1. Defocused communication was a general treatment principle in all sessions. Central components of defocused communication are: to sit beside rather than opposite the child; to create joint attention using an activity the child enjoys rather than focusing on the

child; to 'think aloud' rather than asking the child direct questions; to give the child enough time to respond rather than talking for the child, to continue the dialogue even though the child does not respond verbally; and try to receive a verbal answer in a neutral way rather than praising the child.

2. The behavioural intervention is presented in some detail below (see Second session). The principle for when to use rewards was to reward the child immediately if she or he talked to adults (with a normal or near-normal voice) in accordance with the levels described in Table 2.

The three home-based sessions

First session: The therapist (T) explained to the child (adjusted to the age of the child) the purpose of the visit; the fact that many other children also struggle with muteness outside home; that most do not know why and that they really want to start to speak, but do not know how to do it; that it is possible to work with the problem and practice in small steps and that they will be prepared for what to do and receive small gifts for speaking behaviour. T introduced a workbook (including talking map, stickers, drawing sheets) and audio tape (tell a joke, sing, count, etc.); both only for optional use as a tool to enhance report in the sessions. The workbook was similar for all ages. T, child and parents chose a favourite game with speech demand (e.g. counting or naming) and prepared the stimulus fading situation for the next session.

Second session: T and child reviewed session one and workbook/audiotape if these had been used between sessions. The stimulus fading situation was carried out. I. The stimulus fading situation was divided into six stages: I. Parent and child playing a game; T outside the room with the door closed; II. T outside the room with the door open; III. T visiting the room during the game; IV. T in the room but not playing; V. T sitting beside but not playing; VI. T participating in the game.

Third session: Similar to session two. The stimulus fading situation continued from where the child left off in the previous session. In addition, T prepared for the next session at preschool/school.

Sessions 4–21: Similar content with session three, but in another location (preschool/school). The intervention followed the six modules in Table 2 according to the child's progress. The sessions were held in a separate room until the child reached module V. In module V and VI the intervention was moved to other rooms/settings, including the classroom to facilitate generalization of speech.

Instruments

In addition to the outcome measures, the following instruments were included to give an adequate description of the sample.

Diagnosis of SM and comorbid problems. SM was diagnosed using the SM module from the Anxiety Disorders Interview Schedule (ADIS-IV) (Albano & Silverman, 1996). ADIS is a semi-structured interview with good construct validity (Langley, Bergman, McCracken, & Piacentini, 2004). The SM module relates to the speaking behaviour of the child in different social situations. In addition, we gathered detailed information on whether the child talked to adults in the preschool/school.

Table 2. Predetermined treatment goals reflecting increasingly difficult speaking levels (I–VI) to be obtained in the preschool/school setting from the basic level of zero; does not speak to adults (as defined by the diagnosis of SM in this study) and the number of children in the active treatment group ($N = 12$) who achieved the different treatment goals

Speaking Level	Description of the goal to be obtained in each speaking level	$N = 12$
I	Speaks to the therapist (T) in a separate room with parent (P) present	0
II	Speaks to T in a separate room without P present	3
III	Speaks to one teacher in a separate room with T present	2
IV	Speaks to other teachers/carers and children in a separate room with T present	4
V	Speaks to teachers/carers and children in some settings without T present (speaks to some adults but not to all or most adults; speaks in some groups but not in most or all groups)	3
VI	Speaks to teachers/carers and children in all settings without T present (normal speech, indistinguishable from other children)	0

To assess comorbidity, we used the revised version of the Schedule for Affective Disorders and Schizophrenia for School-Aged Children: Present and Lifetime Version (K-SADS-PL) (Kaufman et al., 1997). This study included nine children below age 6 years, but adequate diagnoses can be made as long as the behavioural concepts and the understanding of life interference is adapted to be relevant to a preschool child (Birmaher et al., 2009).

Questions were also asked pertaining to the motor- and language development of the child (whether it was considered to be delayed, normal or advanced, compared to peers), the family history of SM, and whether the parents described themselves as socially anxious in childhood years.

Interviews were conducted by the last author, with extensive experience with ADIS/K-SADS interviews from research and clinical work, who at the time of assessment was blind to treatment group membership.

Testing of non-verbal IQ and receptive vocabulary. The Stanford-Binet Nonverbal Fluid Reasoning subtest (Roid, 2003) was used as a screening tool for non-verbal IQ. The Stanford-Binet Scales show good psychometric properties and the abbreviated forms are highly related to full-scale IQ. The Peabody Picture Vocabulary Test (PPVT-IV) (Dunn & Dunn, 2007) assesses receptive vocabulary in the form of words read aloud to the child, who in turn points to the appropriate picture. The PPVT has acceptable correlations with standardized verbal IQ measures, such as the Vocabulary subtest of the Stanford-Binet Scale (Roid, 2003). A computed standard score (mean = 100; $SD = 15$) is reported for both the Stanford-Binet Scale and the PPVT.

The children were tested at home with a parent present by the first author, an experienced neuropsychologist, who at the time of testing was blind to treatment group membership.

Outcome measures. The *School Speech Questionnaire (SSQ)* (Bergman et al., 2002). Our primary outcome measure was the SSQ (based on speech frequency in the school context) rated by the child's teacher at baseline and after 3 months, as it was expected that teachers would have the most accurate information on speaking behaviour in this setting. The SSQ, a quantitative measure with no cut-off score, includes 10 questions and is modified from the SMQ (see below) with acceptable internal consistency. Six of the SSQ questions (identical to the SMQ) are used to compute a mean score (range = 0–3), computed as the mean of the valid items, if at least half the items were valid. We used the Norwegian translation with permission from Lindsey Bergman, the developer of the measure. Internal consistency was somewhat low ($\alpha = .64$).

The Selective Mutism Questionnaire (SMQ) (Bergman et al., 2008). We also included the SMQ rated by mothers at baseline and after 3 months for two reasons; to have multiple raters of speaking behaviour in the preschool/school setting, and to look at possible changes in two additional settings (at home and in public). The SMQ includes 32 questions scored from 0 to 3, where 0 indicates that speaking behaviour never occurs, and 1, 2 and 3 refer to seldom, often and always speaking respec-

tively. Seventeen of the SMQ questions are used to compute three subscale mean scores [preschool/school (six items), at home (six items) and in public (five items)] with the same 0–3 scoring range, computed as the mean of the valid items, if at least half the items were valid. The SMQ total factor score was computed from the sum of three subscales divided by three. In this study, one child had one missing item on one subscale. We used the Norwegian translation with permission from Lindsey Bergman, the developer of the measure. Acceptable internal consistency was found for the three subscales and the total score respectively ($\alpha = .68, .73, .76, .77$).

Finally, an additional outcome measure was the achieved treatment goal, ranging from I to VI rated by the therapist (Table 2). For the 12 children who received active treatment, the achieved treatment goal (I–VI) was compared to the baseline value of zero indicating that the child did not speak to adults in the preschool/school setting.

Ethical approval

Written informed consent was provided by the parents. The study was granted approval by the Norwegian Social Science Data Services and the Regional Committees for Medical and Health Research Ethics.

Data analysis

To account for clustering of the data, we used mixed effects models (Pinheiro & Bates, 2000) to investigate group differences at baseline and 3 months. As group differences were expected at 3 months, but not at baseline, a time by group interaction was included. The model was repeated with adjustment for age at baseline including a time by age interaction. In the age adjusted model, age was centred at 6.5 years, close to the mean age in our sample. The level of significance was defined as $p < .05$.

Results

Diagnosis of SM and comorbid problems

Mothers reported that 15 of the 24 children were somewhat delayed in their motor or language development, but all children now spoke freely in full sentences at home. The selective mutism diagnosis was confirmed using the appropriate module on the ADIS diagnostic interview. Using the K-SADS interview, all 24 children with SM suffered from social phobia. Furthermore, 16 children presented with one or more additional lifetime diagnoses of Separation Anxiety ($n = 7$), Specific Phobia ($n = 6$), Generalized Anxiety Disorder ($n = 2$), OCD ($n = 2$), Tics ($n = 2$), Enuresis ($n = 6$), Encopresis ($n = 1$). This did not only apply to older children, as seven of the nine children aged 3–5 years had one or more additional diagnoses. In 10 of the 24 families, there was a positive history of SM in family members (in parents: $n = 5$, other relatives: $n = 5$). In all but one family, one or both parents described the presence of social anxiety symptoms in their own childhood (see Table 1).

Testing of non-verbal IQ and receptive vocabulary. Results were within the average range (see Table 1).

Outcome

The School Speech Questionnaire (SSQ) rated by the teacher. On our primary outcome measure, the SSQ,

there was a significant difference favouring the intervention group [a significant time by group interaction ($F_{1,22} = 5.44, p = .029$)]. In the wait list control group, there was no significant change from baseline to 3 months [prescore = 0.44; postscore = 0.40 ($T_{22} = -0.08, p = .936$)], whereas in the treatment group there was a significant increase in speech [prescore = 0.68, postscore = 1.22 (0.54, 95% CI 0.19–0.89, $T_{22} = 3.22, p = .004$)].

In the model that also included a time by age interaction, using 6.5 years as a reference age, there was a significant time by age ($F_{1,21} = 5.47, p = .029$) interaction, indicating a more pronounced increase in speech in the treatment group for younger children. Again there was a significant time by group interaction ($F_{1,21} = 6.93, p = .016$) and in the treatment group there was a significant increase from baseline to 3 months (0.55, 95% CI 0.23–0.87, $T_{21} = 3.61, p = .002$), with no significant change for wait list controls ($T_{21} = -0.12, p = .908$).

Selective Mutism Questionnaire (SMQ) rated by the mother. For the SMQ School- and Total scores (Table 3) there was a significant difference in time changes between groups with an improvement in the intervention group, and this was essentially unchanged with age adjustment. For the other two SMQ subscales (Table 3) there was a similar but somewhat weaker pattern, without significant group differences in time changes.

The achieved treatment goal (Table 2) ranging from I to VI rated by the therapist. Compared to the baseline value of zero, all children started to speak to the therapist within the preschool/school setting (see Table 2 for more details on this second outcome measure). While three children spoke to the therapist only, not to other adults, another three children achieved level 5 (spoke freely in some but not all groups and/or to some but not all adults. The last three were aged 3–5 years.

Discussion

To our knowledge, this is the first published RCT of a psychosocial treatment for children with SM. We found that the home- and school-based intervention was, as hypothesized, significantly more effective than a wait list control. In addition, we expected that younger children

would benefit more than older ones. Support was found on our primary outcome measure, the SSQ rated by the teachers, and also by the fact that the three children who spoke freely in some settings were 3–5 years of age (therapist rated). The SMQ, our second outcome measure confirmed the findings on the SSQ. On the other hand, the SMQ school subscale showed that the mothers in the intervention group had the impression of increased speech in the school settings irrespective of age. With regard to this finding, one must remember that the parents did not have first-hand information on speech in this setting. So, in summary, although the treatment was also efficacious for children above age 5 years, the finding of greater improvement in the younger age group should be seen as highlighting the importance of early interventions in children with SM.

The children in this study seem to resemble children with SM from other studies in the sense that the group included a high percentage of bilinguals (Elizur & Perednik, 2003) children with comorbid anxiety disorders, and/or some developmental delay (Kristensen, 2000). In line with earlier studies, we found a parent-reported familial accumulation of SM (Black & Uhde, 1995) and of social phobia (Chavira, Shipon-Blum, Hitchcock, Cohan, & Stein, 2007). Our findings also support the new DSM-V classification of SM as an anxiety disorder and that it is upheld as a separate diagnosis from social phobia due to frequent comorbid language delays/disorders (American Psychiatric Association, 2013). Concerning severity of SM, there are no Norwegian norms on the SM questionnaires, but the present baseline data resemble the children in our pilot preschool study (Oerbeck et al., 2012) as well as in a sample of children aged 3–11 years (Bergman et al., 2008) with scores <1 indicating no or rare speaking behaviour.

Compared to our pilot study that only included children aged 3–5 years who received treatment within the preschool, fewer children spoke freely after 3 months in this study. This could be due to a possibly less entrenched mutism in the younger children and/or the difference of the preschool and the school arena. At least in Norway the structure of the day (both in the form of activities, personnel, available rooms) is easier to change in preschool. A third plausible explanation was that the children were treated by local therapists rather than therapists expert in this treatment modality.

Table 3. SMQ results, analysed with mixed effects models

	SMQ school subscale Coefficient [95% CI], <i>p</i>	SMQ home subscale Coefficient [95% CI], <i>p</i>	SMQ public subscale Coefficient [95% CI], <i>p</i>	SMQ total score Coefficient [95% CI], <i>p</i>
Model 1^a				
Time × Group	$F_{1,22} = 10.9, .003$	$F_{1,22} = 2.8, .106$	$F_{1,22} = 3.4, .080$	$F_{1,22} = 9.1, .006$
Time, intervention	0.74 [0.40–1.08], <.001	0.47 [0.18–0.76], .003	0.34 [0.11–0.57], .006	0.52 [0.29–0.74], <.001
Time, wait list Ctr	−0.03 [−0.37 to 0.31], .867	0.14 [−0.15 to 0.43], .331	0.05 [−0.18 to 0.28], .661	0.05 [−0.17 to 0.28], .626
Model 2^b				
Time × Group	$F_{1,21} = 10.8, .004$	$F_{1,21} = 2.7, .112$	$F_{1,21} = 3.2, .088$	$F_{1,21} = 8.8, .007$
Time × Age	$F_{1,21} = 0.5, .506$	$F_{1,21} = 0.1, .823$	$F_{1,21} = 0.03, .863$	$F_{1,21} = 0.1, .713$
Time, intervention, 6.5 years	0.74 [0.40–1.08], .004	0.47 [0.18–0.77], .003	0.34 [0.10–0.58], .007	0.52 [0.29–0.75], <.001
Time, wait list ctr, 6.5 years	−0.03 [−0.37 to 0.32], .862	0.08 [−0.44 to 0.59], .763	0.05 [−0.19 to 0.29], .668	0.05 [−0.18 to 0.28], .637

^aModel by time, group with interaction.

^bModel by time, group, age and interaction/time by group and age.

One could question why the RCT period was only 3 months. In Norway, referred children have the right to receive treatment free of charge within CAMHS after a maximum waiting of 3 months. In our pilot study, the parents refused to participate in the planned design comparing our intervention with treatment as usual. To ensure an RCT design in this study, and to follow national regulations, we found wait list controls for 3 months to be the best alternative. Inclusion in the study was based upon acceptance of the possibility to enter the wait list control group.

To our knowledge, this is the first controlled study to demonstrate that a psychosocial treatment can be effective for SM in children aged 3–9 years. The therapists used treatment including defocused communication as a general treatment principle, and a home and formal day care/school-based intervention with gradual exposure to the feared situations in which speech is expected. Future research, including a larger and more methodologically rigorous efficacy trial is needed to ascertain the active treatment components. In this regard, the fact that we could observe a significant and substantial effect of the intervention – even in the hands of therapists who were not experts in SM – speaks to the robust nature of the intervention and the likelihood of being able to disseminate it in future.

Limitations

The relatively small sample size is an important limitation. This is nonetheless the only psychosocial RCT of SM we are aware of and, small sample notwithstanding, the results were positive. Second, the three outcome raters were not blind to whether treatment had taken place; only the baseline assessments were masked. Third, the internal consistency of the primary outcome measure (SSQ) was somewhat low. Fourth, the lack of a blind assessment of treatment adherence is also a limitation.

Conclusions

This is the first RCT of a psychosocial treatment for children with SM. A statistically significant and clinically meaningful group effect was found after 3 months of a treatment including defocused communication as a general treatment principle, and behavioural interventions in the form of stimulus fading/sliding-in technique. However, a cautious interpretation is needed given the lack of blinded outcome assessments, independent therapist integrity ratings and a relatively small sample size.

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The authors have declared that they have no competing or potential conflicts of interest.

References

Albano, A.M., & Silverman, W.K. (1996). Anxiety disorders interview schedule for DSM-IV. San Antonio, TX: Graywind Publications Inc.

American Psychiatric Association (2000). Diagnostic and statistical manual of mental disorders: DSM-IV-TR. Washington, DC: American Psychiatric Association.

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders: Fifth edition. DSM-V. Washington, DC: American Psychiatric Association.

Bergman, R., Keller, M.L., Piacentini, J., & Bergman, A.J. (2008). The development and psychometric properties of the Selective Mutism Questionnaire. *Journal of Clinical Child and Adolescent Psychology*, 37, 456–464.

Bergman, R., Piacentini, J., & McCracken, J.T. (2002). Prevalence and description of selective mutism in a school-based sample. *Journal of the American Academy of Child and Adolescent Psychiatry*, 41, 938–946.

Birmaher, B., Ehmann, M., Axelson, D.A., Goldstein, B.I., Monk, K., Kalas, C., ... & Brent, D.A. (2009). Schedule for affective disorders and schizophrenia for school-age children (K-SADS-PL) for the assessment of preschool children—a preliminary psychometric study. *Journal of Psychiatric Research*, 43, 680–686.

Black, B., & Uhde, T.W. (1995). Psychiatric characteristics of children with selective mutism: A pilot study. *Journal of the American Academy of Child and Adolescent Psychiatry*, 34, 847–856.

Chavira, D.A., Shipon-Blum, E., Hitchcock, C., Cohan, S., & Stein, M.B. (2007). Selective mutism and social anxiety disorder: All in the family? *Journal of the American Academy of Child and Adolescent Psychiatry*, 46, 1464–1472.

Cohan, S.L., Chavira, D.A., & Stein, M.B. (2006). Practitioner Review: Psychosocial interventions for children with selective mutism: A critical evaluation of the literature from 1990–2005. *Journal of Child Psychology and Psychiatry*, 47, 1085–1097.

Dunn, L.M., & Dunn, D.M. (2007). Peabody picture vocabulary test-IV. San Antonio, TX: Pearson Assessments.

Elizur, Y., & Perednik, R. (2003). Prevalence and description of selective mutism in immigrant and native families: A controlled study. *Journal of the American Academy of Child and Adolescent Psychiatry*, 42, 1451–1459.

Hofmeijer, J., Anema, P.C., & van der Tweel, I. (2008). New algorithm for treatment allocation reduced selection bias and loss of power in small trials. *Journal of Clinical Epidemiology*, 61, 119–124.

Johnson, M., & Wintgens, A. (2007). The selective mutism resource manual. Brackley, UK: Speechmark Publishing Ltd.

Kaufman, J., Birmaher, B., Brent, D., Rao, U., Flynn, C., Moreci, P., ... & Ryan, N. (1997). Schedule for affective disorders and schizophrenia for school-age children-present and lifetime version (K-SADS-PL): Initial reliability and validity data. *Journal of the American Academy of Child and Adolescent Psychiatry*, 36, 980–988.

Kristensen, H. (2000). Selective mutism and comorbidity with developmental disorder/delay, anxiety disorder, and elimination disorder. *Journal of the American Academy of Child and Adolescent Psychiatry*, 39, 249–256.

Langley, A.K., Bergman, R., McCracken, J., & Piacentini, J.C. (2004). Impairment in childhood anxiety disorders: Preliminary examination of the child anxiety impact scale-parent version. *Journal of Child and Adolescent Psychopharmacology*, 14, 105–114.

Manassis, K. (2009). Silent suffering: Understanding and treating children with selective mutism. *Expert Review of Neurotherapeutics*, 9, 235–243.

Oerbeck, B., Johansen, J., Lundahl, K., & Kristensen, H. (2012). Selective mutism: A home-and kindergarten-based intervention for children 3–5 years: A pilot study. *Clinical Child Psychology and Psychiatry*, 17, 370–383.

Pinheiro, J.C., & Bates, D.M. (2000). Mixed-effects models in S and S-PLUS. New York: Springer.

Roid, G.H. (2003). Stanford-Binet IQ scales (5th edn). Itasca, IL: Riverside Publications.

Sluckin, A., Foreman, N., & Herbert, M. (1991). Behavioural treatment programs and selectivity of speaking at follow-up in a sample of 25 selective mutes. *Australian Psychologist*, 26, 132–137.

Vecchio, J., & Kearney, C.A. (2009). Treating youths with selective mutism with an alternating design of exposure-based

practice and contingency management. *Behavior Therapy*, 40, 380–392.

Wichstrom, L., Berg-Nielsen, T.S., Angold, A., Egger, H.L., Solheim, E., & Sveen, T.H. (2012). Prevalence of psychiatric

disorders in preschoolers. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 53, 695–705.

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