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The outcome of children with selective mutism following cognitive behavioral intervention: a follow-up study

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Abstract Selective mutism (SM) is a relatively rare child-hood disorder and is underdiagnosed and undertreated. The purpose of the retrospective naturalistic study was to examine the long-term outcome of children with SM who were treated with specifically designed modular cognitive behavioral therapy (MCBT). Parents of 36 children who met diagnostic criteria of SM that received MCBT treatment were invited for a follow-up evaluation. Parents were interviewed using structured scales and completed questionnaires regarding the child, including the Selective Mutism Questionnaire (SMQ). Twenty-four subjects were identified and evaluated. Their mean age±SD of onset of SM symptoms, beginning of treatment, and age at follow-up were 3.4±1.4, 6.4±3.1, and 9.3±3.4 years, respectively.

There was robust improvement from beginning of treatment to follow-up evaluation in SM, social anxiety disorder, and specific phobia symptoms. The recovery rate from SM was 84.2 %.

Conclusion: SM-focused MCBT is feasible in children and possibly effective in inducing long-term reduction of SM and comorbid anxiety symptoms.

What is Known:

- There are limited empirical data on selective mutism (SM) treatment outcome and specifically on cognitive-behavioral therapy, with the majority of studies being uncontrolled case reports of 1 to 2 cases each.
- There is also limited data on the long-term outcome of children with SM following treatment.

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What is New:

- Modular cognitive behavioral treatment is a feasible and possibly
 effective treatment for SM. Intervention at a younger age is more
 effective comparing to an older age.
- Treatment for SM also decreases the rate of psychiatric comorbidities, including separation anxiety disorder and specific phobia.

Keywords Anxiety · CBT · Comorbidity · Outcome · Prognosis · Selective mutism questionnaire

Abbreviations

ADHD Attention deficit hyperactivity disorder

ADIS-IV- Anxiety disorders interview schedule for DSML IV: Lifetime version

MCBT Modular cognitive behavior treatment

CGI Clinical global impression

CGI-I Clinical global of impression—improvement scale

CGI-S Clinical global of impressions—severity scale

DSM-IV Diagnostic and statistics manual IV
DSM-IV- Diagnostic and statistics manual IV—text

TR revision

ODD Oppositional defiant disorder SAD Social anxiety disorder

SCARED Screen for child anxiety related emotional

disorders

SD Standard deviation SM Selective mutism

SMQ Selective mutism questionnaire

Introduction

Selective mutism (SM) is a relatively rare disorder first evident in childhood. It is characterized by persistent failure to speak in situations where speaking is socially expected (e.g., kindergarten), while being able to speak freely in other situations (e.g., at home) [3]. According to population surveys, the prevalence rate of SM in children of the ages of 4–7 years ranges from 0.7 to 2 % [6, 17] and the prevalence is higher in girls than in boys [27]. The onset of SM usually occurs before the age of 5 years and most commonly between 2.5 and 4 years [8].

Children with SM have high rates of comorbidities, including other anxiety disorders such as social anxiety disorder (SAD) (65 to 100 %), separation anxiety disorder (17 to 32 %), specific phobias (30 to 50 %), history of communication disorders (50 %), and elimination disorders (16 to 29 %) [2, 8, 14, 15]. The comorbidity rate of SM with externalizing disorders, such as oppositional disorders, is much lower (6 to 10 %) than the comorbidity rate with anxiety disorders [18]. Thus, SM seems to be linked more often to anxiety spectrum

than to externalizing disorders, thereby questioning the previously held link between SM and oppositionality [1, 8]. There are limited data regarding the long-term course of SM. Results from studies of clinically referred children indicate that a substantial number will experience a chronic course of mutism while others will continue to experience marked discomfort in speaking situations [22]. Results from a study of nonreferred children with SM suggest that although some improvement may occur spontaneously, the majority remain symptomatic [6]. Thus, it seems that without intervention, most children are likely to chronically suffer and tend to show severe impairments in the areas of academic, social, familial, and personal functioning [8]. Despite the chronic nature of SM and empirical data that early intervention may be effective [24], treatment is usually sought years later after impairment and negative impact on social and academic development has occurred [8, 22].

Until recent years, there were limited empirical data on SM treatment outcome and specifically on cognitive behavioral therapy [28], with the majority of studies being uncontrolled case reports of one to two cases each [11] and two old retrospective studies that assessed the outcome of psychosocial interventions in SM without using standardized outcome measures [16, 26]. The treatment in these studies combined behavioral therapy (CBT) with other techniques (e.g., psychodynamic) and did not employ structured measures. Recent publications on treatment outcomes of SM implemented more rigorous CBT treatments, and their assessments included structured questionnaires [4, 19, 21, 23]. In one case study, an 8-year-old child with SM was treated with modular CBT for childhood anxiety disorders. Based on comprehensive psychiatric assessments, the child recovered from SM following 21 treatment sessions and remission was maintained according to a follow-up 6 months later [23]. In another study, four children with SM ages 5-10 years underwent a manualized treatment based on guiding the parents and teachers conjointly to implement behavioral techniques. Following treatment, the children showed modest improvement in mutism behaviors but results did not generalize to the children's behavior at school [19]. In the most rigorously designed psychological intervention for SM to date, Bergman et al. [4] conducted a randomized controlled study comparing the efficacy of behavioral treatment in children with SM to waiting list controls. In this study, 21 children with SM ages 4-8 years, who received 24 sessions of integrated behavioral therapy, experienced significant improvement compared to waiting list controls, and improvement was maintained for at least 3 months [4]. Lastly, Oerbeck et al. [21] developed a CBT home and school-based intervention and treated 24 children with SM ages 3-9 years for 6 months (24 sessions). At a follow-up 1 year after the end of treatment, 50 % of children no longer fulfilled criteria for SM and there was no significant improvement in the children's comorbid



psychiatric disorders. Of the younger children, 78 % recovered while recovery rate was much lower for older children (33 %).

The aim of the present retrospective naturalistic study was to examine the long-term outcome of children with SM who were treated with SM-focused cognitive behavioral therapy. We hypothesized that there would be significant long-term improvement in SM symptoms and a significant decrease in the rate of psychiatric comorbidities including SAD in these children.

Methods

The sample included 36 children who were 5 to 15 years old at the time of follow-up. All children met DSM-IV criteria for SM and who were treated by SM-focused CBT treatment in a clinic that specializes in treating anxiety disorders and SM. All the children were evaluated at least 1 year after the end of the treatment. Written informed consent was obtained from parents of all participants. The study was approved by the Institutional Review Board and by the Ministry of Health.

All children were treated by one therapist (C.L.), a clinical psychologist. We have two time points in our study: (1) baseline—a retrospective assessment of the children's clinical status at entry to the modular cognitive behavior treatment (MCBT); (2) follow-up—the clinical status of the children as assessed at follow-up on average 2.90 ± 3.23 years following baseline, which is 1.94 ± 2.10 years following end of treatment.

Intervention

The treatment approach used in this study is based on MCBT for childhood anxiety disorders [9]. This therapy approach utilizes cognitive behavioral techniques divided into separate modules, which are flexibly used by clinicians to create individualized treatment plans. MCBT was found effective in 7-13-year-old children with anxiety disorders [10]. MCBT has also been shown to be efficacious in the treatment of an 8year-old boy with SM [23]. The flexibility of MCBT, often lacking in manualized treatments, is much needed in order to address the variable multidimensional developmental components of SM [23]. In line with the abovementioned studies [9, 10, 23], the modular approach implemented in our study included the following components: (1) psycho-education for children and parents—defining SM as an expression of anxiety and specifically of social anxiety; (2) physiological training-breathing and muscle relaxation; (3) cognitive training—externalizing the symptoms and cognitive restructuring; (4) behavioral training—contingency management, development of an exposure hierarchy, modeling, shaping, and gradual desensitization; (5) parent training—enhancing parents' skills in assisting their child and gradually discontinuing the mutism behaviors, in enhancing their child's motivation,

facilitating behavioral interventions, and promoting healthier coping skills; (6) educational and/or recreational staff training—concentrating on their role in facilitating behavioral interventions and promoting social speech within the school and recreational setting. The modular nature of the treatment allows for flexibility in the therapy setting. Very young children (ages 3–5 years) received parent training and school guidance. Older children (ages 6–9 years) were seen together with their parents, whereas children age 10 years and up were seen individually while their parents received separate parent training sessions. All of the children, regardless of setting, received parental and school guidance. These allowed for ecologically sound interventions, allowing the treatment to effectively move beyond the clinical setting and into the natural settings in which the SM symptoms occur.

Measures

Parents of participants were recruited by phone and were then invited to the clinic to undergo structured interviews by trained clinicians using the standardized psychiatric tools at the Anxiety Clinic in a large tertiary medical center. The evaluation measures included the following:

The anxiety disorders interview schedule for DSM-IV: lifetime version (ADIS-IV-L) [20] The ADIS-IV-L is a semistructured interview that yields DSM-IV diagnoses for children including anxiety disorders, mood disorders, and externalizing disorders. The ADIS-P was administered to the parents in a face-to-face interview by interviewers who were trained until achieving good reliability with the senior authors (DG).

The selective mutism questionnaire (SMQ) [4, 5] The SMQ is a parent report instrument designed to measure a child's frequency of nonspeaking behavior across situations in which children are expected to speak. It includes 17 items that yield a quantifiable measure of the severity and interference of the mutism across different settings, yielding three situational domains: "at school" (6 items), "at home/with family" (6 items), and "in public/social settings" (5 items). Parents rated the frequency of each item on a 4-point Likert scale—0=never, 1=seldom, 2=often, 3=always, which are averaged to obtain a mean item and scale scores. The internal consistency of SMQ was within acceptable range (Cronbach's alpha=0.77).

The clinical global impression (CGI) [12] The CGI is a widely used scale completed by a trained clinician to obtain a global rating of illness severity (CGI-S) and a global rating of improvement (CGI-I). Each scale is rated on a seven-point Likert scale from "not at all ill" to "extremely ill" (CGI-S) or from "very much improved" to "much worse" (CGI-I). The



inter-rater reliability for the ADIS-IV-L and CGI assessments was good (kappa≥0.80).

Statistical analysis

Improvement in CGI-S and SMQ scores was measured using paired *t* tests. Changes in the rates of recovery from each of the psychiatric diagnoses were analyzed using the McNemar test. Clinical improvement between treatment completers and noncompleters was compared using independent sample *t* tests.

Results

Sample characteristics

Of the 36 children included in the sample, 7 parents could not be reached due to parental inaccessibility and 5 did not consent to participate. The 12 children who did not participate in the follow-up study were similar to the 24 who did participate in severity of SM symptoms and rates of comorbid psychiatric diagnoses. Table 1 summarizes the characteristics of the sample (N=24) that was recruited for follow-up evaluation. The sample consisted of 12 girls and 12 boys. The mean±SD age of onset of SM symptoms was 3.4 ± 1.4 years. The mean±SD age at the beginning of treatment was 6.40 ± 3.06 years, and the mean±SD age at follow-up was 9.30 ± 3.40 years.

Of the 48 parents, 37 were born in Israel and 11 were immigrants (4 from Russia, 2 from the USA, and 1 each from Denmark, Romania, and Turkey). The mean years of education were 16.15 ± 2.13 for mothers and 14.95 ± 1.88 for fathers. Out of the 24 children with SM, 5 had a history of language and speech problems (3 with language delay, 1 with stuttering, and 1 with articulation problems). None of the children had these language and speech problems at follow-up. Three children were bilingual; 2 children speak Hebrew and Russian, and 1 child speaks Hebrew and English. History of SM was reported in only one first-degree relative, a dizygotic twin of one of the children with SM. Other anxiety and depressive symptoms reported by the parents included shyness (14 mothers, 2 fathers, and 5 siblings), anxiety or excessive worrying (6 mothers, 5 fathers, and 5 siblings), and depression (1 mother and 1 sibling).

Out of the 24 children included in the sample, 19 children (79 %) completed the treatment. Two children were receiving psychiatric medications during the treatment period and follow-up (both were on fluvoxamine). The mean±SD length of the CBT treatment was 12.58±9.96 months for subjects who completed the treatment and 7.80±4.09 weeks for noncompleters. Noncompleters were characterized by low motivation on the part of the children, and the parents were

Table 1 Improvement in selective mutism and social anxiety disorders from baseline to follow-up in 24 children with selective mutism

	Baseline	Follow-up	Statistics	
Age	6.40±3.06	9.30±3.40		
Selective mutism				
CGI-S CGI-I	5.70 ± 0.95	2.30±1.51 83 %	t(23)=10.12, P<0.0001	
Social anxiety disorder				
CGI-S CGI-I	5.62 ± 0.77	2.54±1.61 83 %	t(23)=9.70, P<0.0001	
SMQ				
School	0.52 ± 0.99	2.49 ± 0.92	t=19.18, P<0.001	
Home	1.63 ± 1.15	2.83 ± 0.49	t=10.73, P<0.001	
Public	0.42 ± 0.83	2.24 ± 1.01	t=12.40, P<0.001	
Total	0.88 ± 1.15	2.53 ± 0.87	<i>t</i> =23.43, <i>P</i> <0.001	

SMQ selective mutism questionnaire

unsuccessful in applying the behavioral techniques with their children. In general, duration of treatment was relatively long as the therapy aimed to cure the SM rather than simply improve/alleviate symptoms. In some cases, therapy aimed to continue therapeutic support through grade changes (e.g., from preschool to first grade), thereby elongating the therapy by several months.

We did not find a correlation between length of treatment and the degree of clinical improvement as measured by the CGI-S (r=-0.20, P=ns) and Δ SMQ total scores (r=-0.07, P=ns).

Improvement in symptoms of selective mutism

Table 1 summarizes symptom improvement based on the CGI of SM and SAD. Overall, there was a robust improvement in SM and SAD symptoms. CGI-S scores declined from 5.70 ± 0.95 and 5.62 ± 0.77 at baseline to 2.30 ± 1.51 and 2.54 ± 1.61 at follow-up for SM [t(23)=10.12, P<0.0001] and SAD [t(23)=9.7, P<0.0001], respectively. Marked improvement in SM and SAD was also seen by CGI-I scores, 83 % of the sample evaluated as much (CGI-I=2) or very much improved (CGI-I=1).

Symptom improvement was also analyzed separately for completers versus noncompleters (Table 2). At baseline, the five noncompleters did not significantly differ in their CGI-S scores compared to the 19 completers: 6.20 ± 0.45 and 5.58 ± 1.02 , respectively (P=0.26). At follow-up treatment, completers compared to noncompleters showed more significant improvement in SM and SAD symptoms based on CGI-S and CGI-I scores, recovery rates measured by the ADIS-IV-L, and SMQ scores (Table 2).

Based on parent reports, there was a significant improvement in SMQ symptom severity total scores (t=23.43, P<0.001) in all three situational domains: at school (t=



Table 2 Improvement in selective mutism and social anxiety disorders in treatment completers versus noncompleters

	Treatment completers $(n=19)$	Treatment noncompleters $(n=5)$	Statistics
CGI scale scores at follo	ow-up		
Selective mutism			
CGI-S	1.79 ± 0.98	4.20 ± 1.79	t(22)=2.90, P<0.04
CGI-I	1.37 ± 0.58	2.60 ± 1.14	t(22)=3.37, P<0.01
Very much improved	13 (68.4 %)	1 (20 %)	
Much improved	5 (26.3 %)	1 (20 %)	
Social anxiety disorde	er		
CGI-S	2.05 ± 1.13	4.40 ± 1.95	t(22)=3.55, P<0.01
CGI-I	1.42 ± 0.61	2.80 ± 1.30	t(22)=2.30, P<0.04
Very much improved	12 (63.2 %)		1 (20 %)
Much improved	6 (31.6 %)	1 (20 %)	
Recovery rate ^a			
Selective mutism	16 (84.2 %)	3 (60 %)	
Social anxiety disorder	13 (68.4 %)	2 (40 %)	
SMQ scale total scores	2.75±0.65	1.71±1.15	t(22) = -5.30, P < 0.0001

CGI clinical global impression, SMQ selective mutism questionnaire

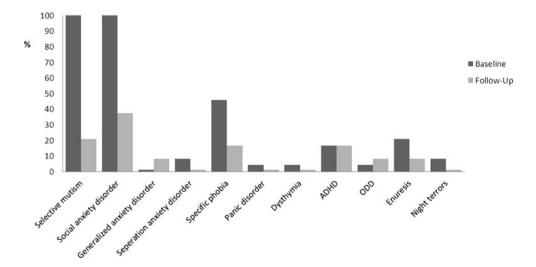
19.18, P<0.001), at home/with family (t=10.73, P<0.001), and in public/social settings (t=12.40, P<0.001).

respectively), social anxiety disorder (100 vs 37.5 %), and specific phobia (45.8 vs 16.7 %).

Change in psychiatric comorbidities from baseline to follow-up

The psychiatric comorbidities of the study sample at baseline and follow-up are presented in Fig. 1. At baseline, all children met DSM-IV-TR criteria for SM and SAD. Using McNemar's test, we found a significant decline (all Ps < 0.05) from baseline to endpoint in the rate of SM (100 vs 20.8 %,

Fig. 1 Change in psychiatric comorbidities from baseline to follow-up



Discussion

The overall aim of the present study was to examine the longterm course of children with SM who received SM-focused CBT. Our findings indicate that there is significant improvement in SM symptoms at follow-up, after SM-focused CBT. Beyond symptom improvement, at follow-up, after treatment



^a Based on the Anxiety Disorders Interview Schedule for DSM-IV (ADIS-IV-L)

was completed, the vast majority of the children (84 %) no longer met DSM-IV criteria for SM. In addition, a significant decrease was found in the rate of psychiatric comorbidities, including SAD, specific phobia, and enuresis.

Despite the debilitating short- and long-term consequences of SM, to date, it has been difficult to determine the effectiveness of SM treatments in children. Overall, compared to other anxiety disorders, there are relatively few publications on SM and specifically limited data on treatment outcome in children with SM. Another limiting factor is that most treatment studies consisted of retrospective record reviews reporting mostly on single cases. To our knowledge, to date, there are only three publications on treatment outcome in SM that consisted of samples similar in size to our sample [4, 16, 26]. There is also no information on the effectiveness of cognitive behavioral therapy in SM as these three studies mentioned above included only a behavioral and not a cognitive component.

Krohn (1992) and Sluckin (1991) retrospectively reviewed the records of 20 and 25 children with SM, respectively, years after the completion of treatment. The treatment examined was based on a nonstructural behavioral protocol that included other behavioral intervention modalities, such as psychodynamic components. Overall, the outcome of children in both studies was very good and most children that received the behavioral intervention described were reported to outgrow the SM [16, 26]. Yet, a limitation of both studies was that they did not use structured assessment tools to measure SM and other psychiatric symptoms.

The only randomized controlled treatment study to date in SM [4] compared behavioral therapy to a waiting list group and found that compared to waiting list controls (N=9), children with SM who received behavioral therapy (N=12) significantly improved. The improvement of SM symptoms was maintained 3 months following the end of treatment.

We followed 24 children with SM who were all treated with a manualized SM-focused cognitive behavioral therapy. The follow-up on the maintenance of treatment gains was relatively long—on average 2 years after treatment was completed. In addition, change in the rate of psychiatric comorbidities was also evaluated.

Previous long-term studies of children with SM found that by adulthood, many years following the onset of SM, 32–42 % continued to suffer from SM [22, 27]. Our findings of 84 % recovery rate from SM following MCBT suggest that the long-term course of treated SM is favorable following a CBT treatment.

We found that in addition to the 84 % recovery rate from SM at follow-up, high recovery rates of psychiatric comorbidities, including SAD, specific phobia, and enuresis, were noted. Improvement of some of the psychiatric disorders may be related to age. However, taken together, these findings may be promising suggesting that when children with SM are treated, in addition to their recovery from SM, they do not tend to

develop other anxiety disorders or other psychiatric comorbidities.

It is well known that there is a high comorbidity and overlap between SM and SAD [5, 7]. It is not clear whether SM represents a severe form of SAD or whether the two conditions are related but distinct disorders [8]. In our sample, there was significant improvement at follow-up following SM-focused CBT not only in SM symptoms but also in SAD symptoms. However, some children that recovered from SM still fulfilled the DSM-IV criteria of SAD. Taken together, our findings suggest that in some cases, the failure to speak in certain situations is a form of behavioral avoidance that successfully helps to decrease SAD and in other cases, SAD has broader aspects not related to speaking (e.g., performing) that are not targeted in SM-focused CBT.

In our study, there was a relatively low dropout rate (20.8 %). Children who dropped out from treatment had similar SM severity symptoms at baseline but significantly more severe SM symptoms at follow-up providing further evidence to the assumption that SM-focused CBT changes the long-term clinical course of SM.

More than half of the children in our cohort started the SM-focused CBT as preschoolers. In our sample, the onset of SM was around the age of 3 years, but children approached the CBT treatment only at the age of 6 years. This finding seems alarming given the clinical and research indications that the younger the child is when he receives treatment for SM, the better the prognosis [13, 25]. The effectiveness of the SM-focused CBT found in our sample of preschoolers refutes the assumption that it is impossible to employ cognitive techniques in preschoolers because of their immature cognitive functions.

Our study has several limitations that should be noted. The main limitations are the retrospective naturalistic design of the study. In addition, the lack of treatment control group limits our ability to conclude regarding the efficacy of the treatment which is beyond the natural course of SM. Reliance on one rater only in the SMQ assessment is another limitation. Since the symptoms are usually most severe at school, adding a teacher rating is important. The sample size was too small to identify other subgroups of responders. We did not have data of adherence measures and results at the end of treatment. Lastly, the fact that evaluations were not conducted blind to the diagnosis of the children potentially biases our results.

Yet, since SM is a relatively rare and neglected disorder in the literature, we believe that our study adds to the limited literature on treatment and long-term outcome of children with SM. We can cautiously conclude that SM-focused CBT is feasible in children and possibly effective in inducing a long-term reduction of SM and comorbid anxiety symptoms.

In the future, there is a need for larger randomized controlled trials to determine the relative efficacy of various types and length of duration of SM treatment.



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Authors' contributions Claudia Lang administered the treatment protocol,

Ziv Nir and Shoshi Domachevsky carried out the evaluations and administered the questionnaires.

Claudia Lang, Ayelet Gothelf, Lee Ginton, Jonathan Kushnir, and Doron Gothelf managed the literature search and statistical analyses

Ziv Nir and Claudia Lang wrote the first draft of the manuscript, and Ayelet Gothelf, Lee Ginton, Jonathan Kushnir, and Doron Gothelf assisted in further preparation of the manuscript.

All authors contributed to and have approved the final version of the manuscript.

Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

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