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RESEARCH ARTICLE

Understanding the Outcome of Children who Selectively Do not Speak: A Retrospective Approach

Zehra Kamani MA[1], Suneeta Monga MD[2,3]

Abstract

Background: Little is known about the longer-term outcomes of children diagnosed with selective mutism (SM) and/or social anxiety disorder (SAD); two anxiety disorders characterized by difficulties speaking in social situations despite being able to speak in other contexts. **Objective**: This retrospective study aimed to descriptively evaluate the long-term SM and SAD diagnostic and symptom severity outcomes in a clinical youth sample. **Methods**: Retrospective follow-up interviews were conducted with 31 parents of children/youth aged four to 14 years previously referred to a specialized anxiety clinic and diagnosed with SM and/or SAD (mean follow-up 4.2 years). Clinician and parent-report measures were used to determine follow-up diagnosis and symptom severity. **Results**: The majority (71%; n=22) of participants still met criteria for SM and/or SAD. Of these, 11 had SAD only; nine had a comorbid diagnosis of SM and SAD; and two had SM only. At follow-up 42% (n=13) were receiving school supports. Close to half (48%; n=15) of parents continued to express concerns about their child's anxiety. Almost all (90%, n=28) youth had attempted some form of treatment, with group cognitive behavioural therapy (CBT) reported as the most common form of treatment tried (48%, n=15). Almost thirty percent (29%, n=9) reported taking anxiety medications in the past with several (13%, n=4) still on medications at follow-up. **Conclusion:** Study results suggest that symptoms of SM and SAD persist in the longer-term. Further investigation into the differences between diagnostic groups and their long-term treatment outcomes is clearly warranted.

Key Words: *selective mutism, social anxiety disorder, cognitive behaviour therapy (CBT), anxiety, school supports*

Résumé

Contexte: Nous en savons peu sur le résultat à long terme des enfants ayant reçu un diagnostic de mutisme sélectif (MS) et/ou de trouble d'anxiété sociale (TAS); deux troubles anxieux caractérisés par des difficultés à parler dans des situations sociales, malgré la capacité de parler dans d'autres contextes. **Objectif:** Cette étude rétrospective visait à évaluer de façon descriptive le diagnostic de MS et de TAS à long terme ainsi que les résultats de la gravité des symptômes dans un échantillon clinique d'enfants. **Méthodes**: Des entrevues de suivi rétrospectives ont été menées auprès de 31 parents d'enfants/adolescents âgés de 4 à 14 ans, précédemment adressés à une clinique spécialisée en anxiété et ayant reçu un diagnostic de MS et/ou de TAS (moyenne du suivi à 4,2 ans). Les mesures des cliniciens et des déclarations des parents ont servi à déterminer le diagnostic et la gravité des symptômes au suivi. **Résultats**: La majorité (71%; n = 22) des participants satisfaisaient encore aux critères du MS et/ou du TAS. Sur ceux-ci, 11 avaient le TAS seulement; 9 avaient un diagnostic comorbide de MS et de TAS, et 2 avaient le MS seulement. Au suivi, 42% (n = 13) bénéficiaient de soutiens scolaires. Près de la moitié (48%; n = 15) des parents exprimaient encore des préoccupations au sujet de l'anxiété de leur enfant. Presque tous (90 %, n = 28) les jeunes avaient essayé une forme de traitement quelconque, et déclaraient que la thérapie cognitivo-comportementale (TCC) était la forme de traitement la plus communément essayée (48%, n = 15). Près

[1]Department of Neurosciences and Mental Health, Hospital for Sick Children, Toronto, Ontario
[2]Department of Psychiatry, Hospital for Sick Children, Toronto, Ontario
[3]Department of Psychiatry, University of Toronto, Toronto, Ontario

Corresponding E-Mail: zehra.kamani@gmail.com

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de 30 pour cent (29%, n = 9) ont dit avoir pris des médicaments contre l'anxiété par le passé, et plusieurs (13%, n = 4) en prenaient encore au moment du suivi. **Conclusion**: Les résultats de l'étude suggèrent que les symptômes du MS et du TAS persistent à long terme. Plus de recherche sur les différences entre les groupes diagnostiques et leurs résultats de traitement à long terme est clairement indiquée.

Mots clés: *mutisme sélectif, trouble d'anxiété sociale, thérapie cognitivo-comportementale (TCC), anxiété, soutiens scolaires*

Introduction

Selective mutism (SM) and social anxiety disorder (SAD) are anxiety disorders characterized by a lack of speech in settings where speech is expected, such as at school or in social situations, despite having the ability to speak in other contexts, such as at home. These disorders are usually diagnosed in early childhood, as young as age of 2.7 years (Gensthaler et al., 2016). Symptoms are often only recognized when children enter the school setting and are required to interact with teachers and peers. When left untreated, evidence suggests that children with SM and SAD may go on to have deficits in social skills (Cunningham et al., 2006, 2004), difficulties in academic functioning (Bergman et al., 2002), psychiatric conditions in adulthood (Hirshfeld-Becker & Biederman, 2002) and higher rates of unemployment later in life (Kolvin, 1994; Patel et al., 2002; Remschmidt et al., 2001). The high comorbidity between SM and SAD, with estimates ranging from 68% to 100% (Dummit et al., 1997; Kristensen, 2000), the similarity in symptomatology, and response to similar pharmacological and psychotherapeutic treatments have led many to suggest that SM and SAD share a common etiology and mechanism (Black & Uhde, 1995; Muris & Ollendick, 2015; Yeganeh et al., 2003, 2006). However, to date, little is known about the long-term clinical outcome of these two disorders.

In one retrospective follow-up study (Remschmidt et al., 2001) of 41 children with SM (mean age = 8.7 years; 29 males) assessed on average 12 years post-referral (mean age at follow-up = 20.5 years), only 39% showed complete remission of SM symptoms. The majority (61%) still presented with significant fears and difficulties with communication, including fear of unknown situations and speaking with strangers (27%), and fear of speaking on the telephone or in shops or offices (15%), while 12% showed no change in symptoms over time.

In one of the few controlled long-term outcome studies of SM (Steinhausen et al., 2006), 33 young adults diagnosed with SM in childhood (mean age at follow-up 21.6 years; 13 males) were compared to two age- and gender-matched comparison groups: those diagnosed with a childhood anxiety disorder other than SM, and those with no childhood psychiatric disorder diagnosis. Fifty-eight percent of the young adults diagnosed with SM in childhood were in remission; however, 42% (n=14) had a phobic disorder at follow-up, most often SAD (71%). Of note, in the group diagnosed with a non-SM anxiety disorder in childhood, similar rates of phobic disorder were seen (42%), as compared to zero instances of phobic disorder in the control group with no psychiatric disorders (Steinhausen et al., 2006). Similarly, Oerbeck and colleagues (2018) evaluated the long-term effects of a school-based cognitive behaviour therapy (CBT) program in the first prospective 5-year SM follow-up study in a sample of 32 children (mean age at baseline 6 years; 10 males). Thirty percent of the 30 participants who completed the 5-year follow-up assessment still met some or all diagnostic criteria for SM. Minimal progress was seen in 10% (n=3) of participants, with one relapsing after being in partial remission at 1-year follow-up, and two (7%) in partial remission after speaking fluently at 1-year follow-up. Also, interestingly, 23% of the sample met criteria for SAD at 5-year follow-up.

Collectively, these studies suggest that symptoms of SM and SAD persist over time, despite treatment. Additionally, it appears that SM and SAD symptomatology are mutable over time. This can be seen in children previously diagnosed with SM who show improvement in SM symptoms, however go on to display more prominent features of social anxiety in the longer-term. There is, to date, insufficient evidence in the literature to make any strong conclusions about the relationship between SM and SAD given the interchangeable nature of their symptomatology.

The aim of the present study was to address this dearth in literature by evaluating the long-term outcome (mean follow-up was 4.2 years) of SM and SAD in a clinical sample of children diagnosed with these disorders. Clinical outcomes, specifically symptom/diagnostic severity and global functioning, inclusive of parental impressions of overall functioning as well as treatments and/or supports received during the follow-up period were our main areas of interest.

Methods

Participants

One hundred and twelve children diagnosed with SM and/or SAD in a tertiary anxiety clinic were eligible for the study. Of these, one declined participation, and 68 (61%)

were not contactable (i.e., did not respond). Ultimately, thirty-one (28%) parents participated in this clinical retrospective follow-up study. Children were initially referred to the clinic for assessment and diagnosis. Some children had received treatment within the clinic; others were referred to treatment programs closer to home. Mean age of children at initial baseline assessment was 6.8 years (*SD* = 2.0; range 4 to 14 years; 7 males). At baseline, 14 (45%) of the 31 were diagnosed with SM only, nine (29%) were diagnosed with SAD only, and 8 (26%) had a comorbid diagnosis of both SM and SAD after a complete psychiatric evaluation, which involved a clinical interview with the child and parent, using the parent-interview of the Anxiety Disorders Interview Schedule for DSM-IV (ADIS-P; Silverman & Albano, 1996).

Measures

The Anxiety Disorders Interview Schedule for DSM-IV (ADIS; Silverman & Albano, 1996), a semi-structured, clinician-administered, interview of parents (ADIS-P), was used to establish diagnosis at both baseline and follow-up. The Clinician Severity Rating (CSR) of the ADIS-P, a clinician measure of child symptom severity using a 9-point scale ranging from 0 to 8, with 0 being no impairment or interference and eight being significant impairment or interference, was determined for both SM and SAD at follow-up only. A CSR of four (moderate degree of impairment) or greater indicates a clinical diagnosis. The ADIS-P CSR was used to establish primary diagnosis at follow-up (SM and/or SAD).

The Children's Global Assessment Scale (CGAS; Shaffer et al., 1983) is a clinician rating of overall adaptive functioning rated on a 100-point scale, with 1 being the most impaired and 100 being least impaired.

The Selective Mutism Questionnaire (SMQ; Bergman et al., 2008) is a 17-item parent report questionnaire that assesses the degree and frequency of speech in school-aged children. Parents report on their child's speaking behavior in the past two weeks across three broad domains (or subscales): at school, at home/with family, and in social situations. Items are rated using a three-point scale ranging from 0 (never) to 3 (always). Higher scores on the SMQ indicate greater speaking behaviours.

The Screen for Child Anxiety Related Disorders (SCARED; Birmaher et al., 1999) is a well-validated parent-report questionnaire of anxiety symptoms in children aged eight years or older. The SCARED includes items that assess anxiety over several domains, including generalized anxiety, separation anxiety, social anxiety, panic, and school refusal, which constitute the 5 factors of the SCARED. Higher scores indicate greater symptoms of anxiety.

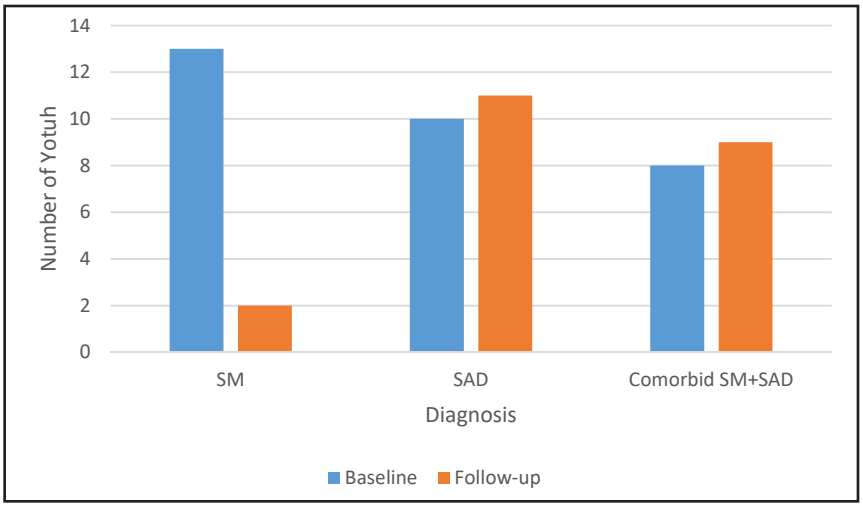
Procedure

Patient charts of children that were assessed and diagnosed with either SM and/or SAD in a tertiary care anxiety clinic between January 2010 and June 2015 were reviewed for eligibility. Ethical approval by the Research Ethics Board was obtained to conduct the chart review and contact eligible parents. Exclusionary criteria included the presence of autism spectrum disorder (ASD) or global developmental delay in the child, or difficulties with parent ability to speak and/or understand the English language in order to complete the necessary research study components. A research coordinator (RC) contacted parents of eligible children over the telephone and obtained informed consent verbally for participation, which involved a 45- to 60-minute follow-up telephone interview with the RC. Typically, these phone interviews were scheduled for a subsequent date. The RC, trained on the ADIS-P and experienced with the SM/SAD population, completed the ADIS-P sections for SM and SAD with the parent over the phone, and completed the SMQ and SCARED questionnaires with the parents. Additionally, using a scripted interview list of questions, parents were asked about their child's functioning at the time of follow-up, including participation in activities within and outside of school, as well as their verbal and non-verbal interactions with family members and friends. Parents were also asked about current or past treatment interventions their child received, such as medications or cognitive behavior therapy (CBT), school supports such as individual education plans (IEPs) or resource time, psychoeducational assessments and speech-language assessments. Finally, parents were also asked whether they had any concerns about their child's functioning at the time of follow-up. Parents were mailed two movie tickets as compensation for their time and effort. Based on parents' responses from the interview, the trained RC independently completed a CGAS score and ADIS-P CSR for both SM and SAD for each participant. Responses from each interview were then reviewed and discussed with an experienced clinician (study PI) and a consensus ADIS-P CSR and CGAS score for each participant was determined.

Statistical analyses

Descriptive statistics of patient demographics, ADIS-P CSR, CGAS ratings, SMQ and SCARED scores at follow-up included means with standard deviations (SD) and ranges or frequencies with percentages as appropriate. A consensus ADIS-P CSR was determined for both SM and SAD at follow-up, and follow-up primary diagnosis was classified based on the diagnosis with greater severity (i.e., a higher ADIS-P CSR). Means with SD were also calculated to observe trends between the four measures (ADIS-P CSR, CGAS ratings, SMQ and SCARED scores) and various demographic characteristics, such as follow-up diagnostic group (SM SAD, or comorbid SM+SAD), baseline age (<7 years vs. ≥ 7 years of age), number of years between

Figure 1. Distribution of diagnoses at baseline and at follow-up



SM = selective mutism; SAD = social anxiety disorder.

Table 1. Mean ADIS-P CSR and CGAS scores by Follow-up Diagnostic Group

Diagnostic group at Follow-up	ADIS-P CSR, Mean (SD)	CGAS score, Mean (SD)
SM (n = 2)	5.0 (1.4)	60.0 (9.9)
SAD (n = 11)	5.5 (1.0)	54.8 (6.1)
Comorbid SM+SAD (n = 9)	6.1 (0.8)	48.1 (6.3)

ADIS-P CSR = Anxiety Disorders Interview Schedule for the DSM-IV parent interview Clinical Severity Rating; CGAS = Children's Global Assessment Scale; SM = selective mutism; SAD = social anxiety disorder.

baseline and follow-up assessment (2-4 years vs. 5-6 years), presence or absence of school supports at follow-up, and presence or absence of ongoing parental concern. All analyses were conducted using SPSS Statistics 24 (IBM Corp, Armonk, NY).

Results

At follow-up assessment, mean age of participants (n=31) was 11.0 years (*SD* = 2.4; range = 7 to 16 years). Follow-up interviews took place on average 4.2 years (*SD* = 1.4; range = 2 to 6 years) from initial assessment. Mean ADIS-P CSR of primary diagnosis was 4.5 (*SD* = 2.2; range = 0 to 7) and mean CGAS score was 62.1 (*SD* = 17.0, range = 42 to 97). Mean SMQ score across the sample was 1.7 (*SD* = 0.63) and mean SCARED total score was 27.8 (*SD* = 15.39).

Only nine of the 31 participants (29%) no longer met criteria for either SM or SAD, while 22 (71%) continued to meet DSM-IV criteria for either SM, SAD or both diagnoses: 2 had SM only; 11 had SAD only, and nine had both SM and SAD (see Figure 1). As compared to the group with SAD only at follow-up, those with a comorbid diagnosis of SM and SAD had a higher mean ADIS-P CSR (6.1, *SD* = 0.8 vs. 5.5, *SD* = 1.0). Of note, the CGAS scores at follow-up were significantly different between diagnostic groups, *F* (19) = 4.2, $p = .031$; children with comorbid SM and SAD had lower mean CGAS scores (48.1, *SD* = 6.3) than those with SAD only (54.8, *SD* = 6.1; *U* = 20.00, $p = .023$). Comparisons were not made with the SM-only group given the small sample of only two individuals in this group at follow-up. Mean ADIS-P CSR and CGAS scores by follow-up diagnostic group are reported in Table 1.

To evaluate the relationship between severity of diagnosis and time between baseline and follow-up, correlations between the primary diagnosis ADIS-P CSR and time (number of years) between baseline and follow-up interview were conducted, which were non-significant ($p > .30$) and remained non-significant even when a median split on number of years (2-4 vs. 5-6) between baseline and follow-up interview was used ($p > .30$).

The majority (90%; n=28) of parents interviewed reported attempting or receiving some form of treatment or support for their child, such as group or individual CBT, speech-language therapy, medications, school support services, or other individual counselling services with a psychologist or social worker (see Table 2). Fourteen (45%) youths had been prescribed medications for anxiety, typically a selective serotonin reuptake inhibitor (SSRI). Of those started on medication, four (29%) were still on medication at

Table 2. Types of treatments or supports received

Treatment/Support Type	Number (%) of youths
CBT – individual	8 (26)
CBT – group setting	15 (48)
Speech-language therapy	9 (29)
Anxiety medication (i.e. SSRI) – current; past [a]	4 (13); 9 (29)
School supports (e.g. resource time, IEP) – current; past [a]	13 (42); 5 (16)
Private therapy	8 (26)
Individual counselling with therapist, social worker, or psychologist	6 (20)
Play therapy	2 (6)
Other – school drop-in program for parents and children	1 (3)
Other – community improv/drama	1 (3)
Other – Big Brother/Big Sister	1 (3)

CBT = cognitive behaviour therapy; SSRI = Selective serotonin reuptake inhibitor; IEP = individual education plan.
[a] Past refers to treatment/support received anytime between baseline assessment and follow-up, but not received currently.

follow-up interview. CBT was reported as the most common form of intervention, with twenty parents (65%) reporting that their child had received CBT intervention in either a group (n=15) or individual (n=8) setting. Although only two parents reported their child as having speech-language difficulties, nine parents (29%) reported their child as having received speech-language therapy during the ensuring follow-up period. Parents also reported that to assist their child with symptoms of anxiety, they had attempted a variety of community programs, such as Big Brother/Big Sister, improvisation/drama class, and programs within the school that included parent and child involvement (Table 2). Seven (23%) parents reported that their children were not participating in any extracurricular activity within or outside of school at follow-up, reflecting their ongoing anxiety.

At follow-up, mean CGAS score for children who were older (≥ 7 years of age) at baseline was 55.4 (*SD* = 12.0), whereas mean CGAS score for children who were younger (< 7 years of age) at baseline was notably higher at 69.3 (*SD* = 18.8). Mean follow-up ADIS-P CSR for older children was 5.4 (*SD* = 1.3), while mean follow-up ADIS-P CSR for children who were younger at baseline was notably lower at 3.5 (*SD* = 2.6).

At follow-up, 13 (42%) youths were still receiving school supports, such as IEPs or resource time (see Table 2). These youths were rated as having a mean ADIS-P CSR of 5.5 (*SD* =1.8) at follow-up and mean CGAS score of 52.7 *(SD* = 12.9), while those youths without school supports had a lower mean ADIS-P CSR of 3.7 (*SD* = 2.2) and a higher mean CGAS score of 68.9 (*SD* = 16.5). Amongst the parents of children receiving school supports, mean SCARED total score was 35.9, (*SD* = 16.8) whereas parents of those not receiving school supports reported a lower mean SCARED total score of 22.0 (*SD* = 11.51; see Table 3).

Ongoing concerns about their child's anxiety at follow-up was expressed by 15 (48%) of the interviewed parents. The children of these parents were rated as having a mean primary ADIS-P CSR of 5.1 (*SD* = 2.3) and mean CGAS score of 56.3 (*SD* = 16.7), whereas children whose parents no longer expressed concern were rated as having a lower mean ADIS-P of 3.5 (*SD* =2.0) and higher mean CGAS score of 70.7 (*SD* =15.9). Similarly, mean SMQ score for concerned parents was 1.5 (*SD* = 0.7) and mean total SCARED score of 36.1 (*SD* = 16.4), whereas for those parents who no longer expressed concerns, mean SMQ score was higher at 2.1 (*SD* =0.41) and mean total SCARED score was lower at 20.4 (*SD* =10.3 see Table 4).

Of particular interest, five (16%) parents were of the opinion that changing schools helped their child in allowing them to have a fresh start and begin speaking to new peers and teachers. Other parents remarked that there is "not enough help out there" and that "teachers do not understand SM".

Discussion

The overall aim of this study was to evaluate the long-term outcomes (between 2 and 6 years; mean = 4.2 years) of a clinical sample of children diagnosed with SM and/or SAD. Our findings suggest that symptoms of SM and SAD persist over time. Of importance, although the majority of participants remained symptomatic at follow-up, their diagnoses changed over time. At baseline, the majority of children had a diagnosis of SM only, whereas at follow-up only two

Table 3. Follow-up outcome measure scores between youths currently receiving school supports and those with no school supports

Outcome Measure	Youths with school supports, Mean (SD)	Youths with no school supports, Mean (SD)
Primary ADIS-P CSR	5.54 (1.81)	3.72 (2.22)
CGAS	52.69 (12.89)	68.94 (16.51)
SCARED total	35.84 (16.84)	22.00 (11.51)
Generalized Anxiety Factor	9.92 (5.48)	5.28 (3.66)
Separation Anxiety Factor	6.38 (3.43)	3.83 (3.78)
Social Anxiety Factor	11.23 (3.30)	8.06 (2.84)
School Refusal Factor	1.77 (2.09)	0.94 (1.26)
Panic Factor	6.54 (5.03)	3.89 (4.50)
SMQ average	1.58 (0.55)	1.86 (0.66)
School subscale	1.69 (0.74)	1.98 (0.94)
Home subscale	1.91 (0.58)	2.08 (0.51)
Social Situations subscale	1.06 (0.81)	1.46 (0.88)

Table 4. Follow-up outcome measure scores between parents with and without current concerns about their child's anxiety

Outcome Measure	Parents with current concerns, Mean (SD)	Parents with no current concern, Mean (SD)
Primary ADIS-P CSR	5.13 (2.32)	3.46 (1.98)
CGAS	56.27 (16.68)	70.69 (15.87)
SCARED total	36.07 (16.40)	20.38 (10.33)
Generalized Anxiety Factor	9.60 (5.15)	5.38 (3.94)
Separation Anxiety Factor	7.13 (4.00)	3.00 (2.16)
Social Anxiety Factor	10.33 (3.68)	8.08 (2.99)
School Refusal Factor	1.87 (2.17)	0.62 (0.77)
Panic Factor	7.13 (5.85)	3.31 (2.53)
SMQ average	1.49 (0.71)	2.08 (0.41)
School subscale	1.57 (0.96)	2.32 (0.48)
Home subscale	1.77 (0.58)	2.19 (0.38)
Social Situations subscale	1.08 (0.96)	1.66 (0.68)

participants had a diagnosis of SM only, with the majority having a primary diagnosis of SAD at follow-up. These findings are similar to those of Oerbeck and colleagues (2018), who found that 23% of individuals with SM at baseline had a diagnosis of SAD instead at 5-year follow-up, and those of Steinhausen and colleagues (2006), who found that 42% of adults in their follow-up study of children diagnosed with SM had instead developed a phobic disorder, most often SAD. These findings of a change in diagnosis from SM to SAD at follow-up, in conjunction with evidence suggesting the same treatments (e.g., CBT, SSRIs) effectively treat both disorders, and the high comorbidity between SM and SAD, lends credence to the notion that these two disorders are mutable and may be conceptualized as falling on a single spectrum rather than distinguished as two distinct disorders (e.g., Black & Uhde, 1995; Yeganeh et al., 2003, 2006). SM has also been conceptualized as an early marker for developing a phobic disorder or other anxiety disorder later in life (e.g., Chavira et al., 2007), which could also be supported by the present study's findings. Furthermore, it is unclear as to whether SM represents a more severe, and thereby more impairing, form of SAD, or whether a comorbid diagnosis of SM and SAD is more functionally impairing. The present study suggests that a comorbid diagnosis of SM and SAD is more impairing than diagnosis of only SAD, given the significantly lower adaptive functioning of children with both diagnoses. Due to the

small sample size of only two participants with an exclusive diagnosis of SM at follow-up, direct comparisons could not be made with this group. As such, more comparative and long-term clinical and community follow-up studies with larger sample sizes are needed.

While formal statistical analyses were not conducted within the scope of this descriptive review, children who were older (≥ 7 years) at baseline appear to be more impaired at follow-up, with higher anxiety disorder severity ratings and lower overall adaptive functioning as compared to children who were younger (< 7 years) at baseline. Of note, there was no effect of time between baseline and follow-up assessment, which precludes the possibility that older children were assessed within a shorter span of time, allowing for less opportunity for treatment and symptom remission, than younger youths. These findings support the importance of earlier diagnosis of SM being associated with a more favourable prognosis. Early diagnosis and intervention for anxiety disorders is widely known to be linked to better outcomes (Hirshfeld-Becker & Biederman, 2002). Oerbeck and colleagues have shown that treatment of SM at a younger age is associated with greater post-treatment improvements (Oerbeck et al., 2014) and greater remission rates at 12 months post-treatment follow-up (Oerbeck et al., 2015); specifically, 78% remission rates in 3 to 5 year old children versus 33% in 6 to 9 year old children. Oerbeck and colleagues (2018) have also demonstrated that greater baseline symptom severity is seen in older children with SM. Unfortunately in our study, baseline symptom severity was not available. Interestingly, however, we found no effect of type of treatment; specifically, no differences were found between those youths who reported receiving CBT and those who did not.

Almost half (n=13) of the participants in this study were receiving school supports at follow-up. These youths had clinically significant diagnoses for either SM or SAD at follow-up (average primary diagnosis ADIS-P CSR ≥ 4) in contrast to youths who were not utilizing school supports, who, on average, no longer met diagnostic criteria for SM or SAD (ADIS-P CSR < 4). These findings suggest that children diagnosed with SM and SAD continue to require additional resources to support their academic performance. Beyond the scope of this study is whether these youths were falling behind academically as a result of persistent and impairing symptoms, or whether school supports enabled them to perform at par with their peers. The availability of support within schools, however, was an ongoing theme in parent interviews, as several parents, commented that *teachers do not understand SM and should be provided with more training or resources to assist their students with these disorders*. The long-term efficacy of school-based CBT programs in which teachers play a central role in delivering treatment, are currently being explored and have shown promise (Oerbeck et al., 2014, 2018).

Interviews with parents also shed light on parents' perceptions about their child's wellbeing. At follow-up, parents with concerns about their child's anxiety generally reported more symptoms of anxiety on the SCARED and fewer speaking behaviours on the SMQ. Furthermore, clinicians' ratings of diagnostic symptom severity for this group of children appeared to be higher and overall global functioning appeared to be poorer compared to the children of parents who did not express such concerns about their child. These observations suggest that parents' impressions of their child's symptoms are in line with clinicians' judgments. The addition of child report measures would provide further insight into the extent to which youths are metacognitively aware of their symptoms and degree of impairment, as well as the extent to which their own self-ratings coincide with their parents' ratings.

Finally, five parents reported that a change in schools appeared to help their children as they were able to speak to new peers and teachers who were not familiar with their previous diagnosis and history of mutism. Although further evaluation is needed, it could be hypothesized that the change in social environment enabled these children to have a "fresh start" in their new schools in which they were not known for their mutism. Given that children who do not speak often have comorbid social anxiety and fear attention from others, it is possible that transitioning from "not speaking" to "speaking" at school is, in itself, anxiety-provoking, given the attention that such a change might bring upon the child. With a change in schools, however, this attention is avoided, perhaps facilitating speaking in the socially anxious child.

There are several limitations to this study. Our small sample size and the use of a clinical sample preclude generalizations on the outcomes of these disorders to non-clinical samples where there may be milder symptoms. A clinical sample also neglects to include those who experience barriers to accessing mental health resources, such as cultural, economic, or logistical barriers, which are important factors to consider in future studies. Unfortunately, studies to date on the longer-term outcomes of children with SM are comparable in size and typically have looked at clinical populations.

Given the methodology of this study, formal statistical analyses were not feasible to determine associations between our outcome measures as a function of the treatments and/or supports received during the follow-up time period. This presents an important avenue for future research. Another limitation posed by the methodology is the bias of self-selection, whereby those parents whose children did not show improvements over time may have self-selected to participate in this study. The retrospective nature of the study also lends itself to parental recall bias as it is possible that parents did not accurately recall details of their child's diagnostic or treatment history, such as the type, length and age

at which treatments or assessments were completed. For example, many parents described certain components of treatments obtained, such as learning relaxation techniques or playing one-on-one with a health-care professional, but were not consistently able to label the types and methods of treatment received, such as behaviour therapy, play therapy, or CBT.

Conclusion

This retrospective follow-up study of a clinical population of SM and SAD children adds to the literature on the long-term outcomes of these children. The majority of participants continued to meet diagnostic criteria for SAD or SM and remained symptomatic on average four years post initial assessment, suggesting that these disorders are persistent over time. This highlights the importance and need to develop a better understanding of best treatment options and necessary supports for these children to improve their functioning over time. Larger population-based and prospective follow-up studies are also needed to better recognize and diagnose SM and SAD, and to understand the long-term sequelae and the changing nature of these disorders over time.

Acknowledgements / Conflicts of Interest

The authors have no conflicts of interest to declare.

References

Bergman, R. L., Keller, M. L., Piacentini, J., & Bergman, A. J. (2008). The development and psychometric properties of the selective mutism questionnaire. *Journal of Clinical Child & Adolescent Psychology, 37*(2), 456-464.

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

Birmaher, B., Brent, D. A., Chiappetta, L., Bridge, J., Monga, S., & Baugher, M. (1999). Psychometric properties of the Screen for Child Anxiety Related Emotional Disorders (SCARED): A replication study. *Journal of the American Academy of Child & Adolescent Psychiatry, 38*(10), 1230-1236.

Black, B., & Uhde, T. W. (1995). Psychiatric characteristics of children with selective mutism: A pilot study. *Journal of the American Academy of Child & 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 & Adolescent Psychiatry, 46*(11), 1464-1472.

Connolly, S. D. & Bernstein, G. A. (2007). Practice parameter for the assessment and treatment of children with anxiety disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 46*(2), 267-283.

Cunningham, C. E., McHolm, A. E., & Boyle, M. H. (2006). Social phobia, anxiety, oppositional behavior, social skills, and self-concept in children with specific selective mutism, generalized selective mutism, and community controls. *European Child & Adolescent Psychiatry, 15,* 245-255.

Cunningham, C. E., McHolm, A., Boyle, M. H., & Patel, S. (2004). Behavioral and emotional adjustment, family functioning, academic performance, and social relationships in children with selective mutism. *Journal of Child Psychology & Psychiatry, 45*(8), 1363-1372.

Dummit, E. S., Klein, R. G., Tancer, N. K., Asche, B., Martin, J., & Fairbanks, J. A. (1997). Systematic assessment of 50 children with selective mutism. *Journal of the American Academy of Child & Adolescent Psychiatry, 36*(5), 653-660.

Gensthaler, A., Khalaf, S., Ligges, M., Kaess, M., Freitag, C. M., & Schwenck, C. (2016). Selective mutism and temperament: The silence and behavioral inhibition to the unfamiliar. *European Child & Adolescent Psychiatry, 25,* 1113-1120.

Hirshfeld-Becker, D. R., & Biederman, J. (2002). Rationale and principles for early intervention with young children at risk for anxiety disorders. *Clinical Child and Family Psychology Review, 5*(3), 161-172.

Kolvin, I. (1994). The origin of elective mutism. 13th International Congress of the International Association for Child and Adolescent Psychiatry and Allied Professions, San Francisco, USA, July 24-28, 1994, Abstracts, 181.

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

Muris, P & Ollendick, T. H. (2015). Children who are anxious in silence: A review on selective mutism, the new anxiety disorder in DSM-5. *Clinical Child and Family Psychology Review, 18,* 151-169.

Oerbeck, B., Overgaard, K. R., Stein, M. B., Pripp, A. H., & Kristensen, H. (2018). Treatment of selective mutism: A 5-year follow-up study. *European Child & Adolescent Psychiatry, 27*(8), 997-1009.

Oerbeck, B. Stein, M. B., Pripp, A. H., & Kristensen, H. (2015). Selective mutism: Follow-up study 1 year after end of treatment. *European Child & Adolescent Psychiatry, 24,* 1-10.

Oerbeck, B., Stein, M. B., Wentzel-Larsen, T., Langsrud, O. & Kristensen, H. (2014). A randomized controlled trial of a home and school-based intervention for selective mutism – defocused communication and behavioural techniques. *Child and Adolescent Mental Health, 19*(3), 192-198.

Patel, A., Knapp, M., Henderson, J., & Baldwin, D. (2002). The economic consequences of social phobia. *Journal of Affective Disorders, 68*(2-3), 221-233.

Remschmidt, H., Poller, M., Herpertz-Dahlmann, B., Henninghausen, K., & Gutenbrunner, C. (2001). A follow-up study of 45 patients with elective mutism. *European Archives of Psychiatry and Clinical Neuroscience, 251,* 284-296.

Shaffer, D., Gould, M. S., Brasic, J., Ambrosini, P., Fisher, P., Bird, H., & Aluwahlia, S. (1983). A children's global assessment scale (CGAS). [Research Support, U.S. Gov't, P.H.S.]. *Archives of General Psychiatry, 40*(11), 1228-1231.

Silverman, W. K., & Albano, A. M. (1996). *The Anxiety Disorders Interview Schedule for Children for DSM-IV: Clinician Manual (Child and Parent versions).* San Antonio, TX: The Psychological Corporation.

Steinhausen, H.-C., & Juzi, C. (1996). Elective mutism: An analysis of 100 cases. *Journal of the American Academy of Child & Adolescent Psychiatry, 35*(5), 606-614.

Steinhausen, H-C., Wachter, M., Laimbock, K., & Metzke, C. W. (2006). A long-term outcome study of selective mutism in childhood. *Journal of Child Psychology and Psychiatry, 47*(7), 751-756.

Yeganeh, R., Beidel, D. C., & Turner, S. M. (2006). Selective mutism: More than social anxiety? *Depression and Anxiety, 23,* 117-123.

Yeganeh, R., Beidel, D. C., Turner, S. M., Pina, A. A., & Silverman, W. K., (2003). Clinical distinctions between selective mutism and social phobia: An investigation of childhood psychopathology. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*(9), 1069-1075.