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ORIGINAL ARTICLE

An Updated Characterization of Childhood Selective Mutism: Exploring Clinical Features, Treatment Utilization, and School Services

Kira Boneff-Peng[1,2] · Patricia C. Lasutschinkow[2] · Zachary A. Colton[2] · Carol R. Freedman-Doan[2]

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Abstract

Selective mutism (SM) is a severe but understudied childhood anxiety disorder. Most epidemiological research on SM was conducted decades ago and is limited by small sample sizes. This study analyzes parent-reported clinical data from 230 children with diagnosed and suspected SM to provide current information about the presentation of this disorder. Overall, anxiety and social anxiety symptoms were elevated. Gender ratio, comorbidities and family history of psychopathology were generally aligned with previous research. However, age of onset and diagnosis were both earlier than previously reported, with an average delay of 2 years between onset and diagnosis. The majority of children received therapy and school accommodations for their SM, yet there was large variability in types of interventions. This is the largest survey of children with SM conducted primarily within the US and it constitutes the first systematic inquiry into interventions and accommodations received within clinical and school settings.

Keywords Selective mutism · Child anxiety · Social anxiety

Introduction

Selective mutism (SM) is a severe childhood anxiety disorder characterized in the diagnostic and statistical manual of mental disorders, Fifth edition, text revision (DSM-5-TR) by a lack of speech in certain contexts (e.g., at school) despite speaking freely in other contexts (e.g., with parents; [1]). Onset typically occurs during early childhood, although most children are diagnosed after school entry [2–4]. Theories suggest that when children with SM are prompted to speak with unfamiliar people, they learn to reduce their anxiety and discomfort by remaining silent [5, 6]. The deficiency of speech associated with SM can lead to marked social and academic impairment (e.g., social isolation and teasing by peers), especially if children do not receive appropriate treatment, as symptoms typically do not remit spontaneously [1, 7, 8]. SM is often considered a rare disorder, although recent prevalence estimates vary widely from as low as 0.06% [9] to as high as 1.5% [10].

Research about SM has been lacking historically, despite these risks of lasting social and academic difficulties, the unlikelihood of remittance without treatment and reports that SM may be more common than generally believed. In particular, few recent studies have explored phenomenological patterns in populations of children with SM and SM has not been included as a diagnostic category in large-scale epidemiological studies of the prevalence of childhood psychiatric disorders [1]. Many of the epidemiological studies that have been conducted have had small sample sizes (e.g., [2, 11]). Furthermore, many of these studies were conducted abroad and/or were published decades ago, with the most recent prevalence and epidemiological study in the United States (US) occurring 20 years ago [7]. This is especially concerning given advancements in evidence-based treatments for SM during the last 20 years and efforts to increase awareness of SM and improve access to quality interventions (e.g., [5, 12, 13]). Thus, the purpose of this study is to inform the current understanding of this disorder by presenting data from a large sample of 230 children with SM. A thorough review of the literature on the topics explored in this paper, including data on prevalence and diagnostic patterns seen in SM, comorbidities, family history, treatment

✉ Kira Boneff-Peng
kiraboneffpeng@sunfieldcenter.com

[1] Sunfield Center, 3005 Boardwalk Dr, Suite 201, Ann Arbor, MI 48108, USA

[2] Department of Psychology, Eastern Michigan University, 341 Science Complex, Ypsilanti, MI 48197, USA

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and school services, is also included. To date, this is the largest known study examining clinical features of children with SM conducted primarily within the US and it constitutes a significant step forward in examining current clinical patterns in children with SM.

Prevalence and Diagnostic Patterns

As mentioned above, prevalence estimates of SM vary widely in the literature. Early studies that reported extremely low prevalence rates of 0.03% [14] and 0.06% (Fundudis et al. 1979, as cited in [7]) are still commonly cited, but key methodological limitations in these studies, as well as shifts in conceptualization of SM over time, compromise their utility. For example, these studies did not use specific diagnostic criteria for SM or standardized methodological processes to evaluate prevalence rates. Thus, it is important to look toward more recent studies of prevalence for a better understanding of the current rates of this disorder as defined today in the DSM-5-TR. More recent prevalence rates vary considerably, with estimates ranging from as low as 0.06% and 0.18% to as high as 1.5% and 1.9% [4, 8–10]. The only known study conducted within the US with an explicit aim of assessing the prevalence of SM reported a rate of 0.71% [7]. Importantly, even though SM is often considered rare, these rates are comparable to those of other pediatric psychiatric conditions such as autism spectrum disorder (1.5% [15]) and obsessive–compulsive disorder (0.25–4%, [16]).

SM typically presents in early childhood; mean age of onset has been reported to range from 2.7 years [2] to 4.6 years [4]. One study found that boys tended to have an earlier symptom onset than girls, but this has not been studied in depth [17]. Despite its early onset, children are often diagnosed at a later age, as they do not usually reach clinical attention until school entry [3]. Because school entry often acts as a precipitator to SM diagnosis, it is likely that diagnosis ages may differ between countries with varying ages of school entry. For example, a study based in Finland, in which children begin school at age seven, reported an average age of diagnosis of 7.9 to 8.1 years [9].

With regard to patterns between subgroups of the population, many studies have shown a higher rate of diagnosis in female children than in male children (e.g., [4, 8]), whereas others have reported approximately equal rates of diagnosis between female children and male children (e.g., [7, 18]). Some researchers have also suggested that immigrant status and/or bilingualism may be related to SM. One study found that prevalence rates of SM were significantly higher in a population of immigrant children than in the general population (2.2% and 0.76%, respectively, [18]. Similarly, a study conducted by Steinhausen and Juzi [17] also found high prevalence of SM in children from immigrant backgrounds. A more recent study by Starke [19] expanded on this finding, showing that bilingualism could not independently explain increased vulnerability to development of SM in immigrant children. Rather, anxiety, language skills and parental assimilation were all found to be major contributing factors in their sample.

Comorbidities

Children with SM often have comorbid clinical conditions. A recent meta-analytic review found that, within the 22 studies meeting criteria and 837 children diagnosed with SM, 80% were diagnosed with a comorbid anxiety disorder [20]. Social anxiety disorder/social phobia (SAD/SP) has particularly high comorbidity with SM, with rates reportedly ranging from 67 to 100% ([2, 3, 17, 21], Oerbeck et al. 2013). However, some recent studies have found lower rates of comorbidity between SM and SAD/SP, which may reflect progress in conceptualizing SM as distinct from SAD/SP and improving differential diagnostic tools (18.2%, [22–25]). Driessen et al. [20] meta-analytic review summarized relevant literature covering other common comorbid anxiety disorders in children with SM. They found that comorbidity with specific phobias ranged from 0 to 45.8% and comorbidity with separation anxiety disorder ranged from 0 to 41.7%. Additionally, developmental disorders have been found to occur commonly in children with SM (13% [2], 68.5% [3]). Some studies have also identified high rates of comorbid elimination disorders in children with SM (17% [2], 24% [17], 31.5% [3]) Although findings are less consistent with regards to externalizing disorders, some earlier studies have found rates of comorbid oppositional defiant disorder and other externalizing disorders to be somewhat elevated in children with SM (i.e., 10% [2], 21% [17]). Additionally, approximately 30% to 50% of children with SM have communication, speech or language deficits [3, 4, 17, 26].

Family History

There is considerable support for high rates of social anxiety and SM symptoms in families of children with SM. Researchers have found rates of SAD/SP in first degree relatives of children with SM to be between 37 and 70% [2, 4, 27]. Additionally, Black and Uhde [2] found that 37% of children with SM had a first-degree relative with SM as well. When compared to parents of children with generalized anxiety disorder (GAD), parents of children with SM reported higher rates of stressful life events and higher psychopathology on a global severity index [28]. Although formal twin/adoption studies have not been conducted for SM, case studies have suggested both genetic and environmental impacts on the development of SM in both monozygotic and dizygotic twins [29, 30].

Clinical Treatments

Over the past 40 years, both the conceptualization and common treatments for SM have significantly changed. Traditionally, SM was commonly viewed through either a psychodynamic or family systems lens. However, recent studies have supported a conceptualization of SM as an anxiety disorder, resulting in an increased focus on cognitive-behavioral therapies (CBT). While 43.5% of treatment studies published from 1990 to 2005 saw inclusion of psychodynamic techniques, that decreased to 13.0% of studies from 2005 to 2015 [13, 31]. In a recent review of SM treatment studies published between 2005 and 2015, behavioral therapy (BT) and CBT were utilized and found effective across most treatment studies [13]. More recently, social skills training, parent child interaction therapy (PCIT) and intensive group treatments have also been the focus of scientific inquiry [32–34].

In addition to psychological treatments, pharmacological interventions for SM have been studied. Selective serotonin reuptake inhibitors (SSRIs) and monoamine oxidase inhibitors (MAOIs) were the most commonly prescribed medications for SM in treatment research and psychiatric practice [35]. Although a number of treatments, both psychological and pharmaceutical, have been found to be effective for SM, treatment outcome research is still in its preliminary stages. Furthermore, recent studies have not investigated which of these interventions are being utilized in the treatment of children with SM in clinical practice.

School Services

Children with SM are often highly symptomatic in school [2, 36], therefore, treatment for SM is often conducted partially or fully in schools. While various studies have demonstrated successful outcomes for several different models of school-based intervention (e.g., [12, 37–39]), little is known about the actual utilization of school-based interventions in the treatment of children with SM. Furthermore, very little is known about how school personnel conceptualize children with SM to qualify them for accommodations. Based on clinical experience, Kotrba [6] asserts that the most common designations given to children with SM to obtain individualized education plan (IEP) services are Speech/Language Impairment, Other Health Impairment, and Emotional Disturbance/Disability. However, research studies on SM have not investigated the prevalence of specific school plans, such as IEPs and section 504 plans, nor have they looked at the specific interventions being utilized in the treatment of SM within schools.

Study Aims

Compared to most other childhood psychiatric disorders, research on SM is quite limited. Much of what is known about SM currently is drawn from seminal cross-sectional design studies that may be outdated (e.g., [2, 11]) or more recent research that does not cover the wide breadth of these early studies. For example, a recent meta-analysis by Driessen et al. [20] focuses specifically on comorbid anxiety in children with SM, but it does not cover other aspects of the disorder in detail. Furthermore, many seminal studies on SM were conducted using international samples and cannot be generalized effectively to the US population (e.g., [4, 17, 18]). Thus, there is not enough recent, large-scale research in the US to provide an accurate consensus about the current phenomenology and course of SM.

To address this gap in the literature, the first aim of the current study was to analyze data from a large sample of children with SM to gain a more updated and comprehensive understanding of the current landscape of SM in the US and internationally. Specifically, this study analyzed data related to the course and clinical features of SM, including age of onset, age of diagnosis, rates of comorbidities, severity of anxiety symptoms, rates of family history of mental illness, and severity of SM symptoms. A second aim of this study was to identify the types of treatments and services received by children with SM in both clinical and school settings, the first known study to do so. The data presented in this article will hopefully be utilized by researchers and clinicians alike, leading to a better understanding of the risk factors, developmental course, and associated features of SM, as well as information about access to treatment and school services. This article is intended to act as a point of departure to spur more research on this disorder and broaden clinical training in evidence-based treatments for SM.

Methods

Participants

Participants were parents of a child with diagnosed or suspected SM between the ages of 3 years, 9 months and 12 years. This age range was chosen to capture clinical data on preadolescent children of varying ages and stages of the disorder, as SM is most typically seen in children. Participants were primarily self-selected and recruited via flyers. Electronic flyers were circulated in groups online, including international SM advocacy groups, social media groups related to SM and clinical organizations serving children with SM. Paper flyers were posted in pediatric mental health clinics in Southeast Michigan.

Due to the selective nature of recruitment, it is assumed that this study sample largely represents families who are well informed about SM and have had the resources for diagnosis and treatment. Thus, in hopes of including families with less resources, as well as families who are less likely to have received treatment yet, this study also included children who were not yet formally diagnosed with SM but whose parents reported concerns about SM. Specifically, participants were screened for inclusion by asking if their child was formally diagnosed with SM and if parents reported no diagnosis, they were asked screening questions to confirm if they suspected their child had SM and if their child had a pattern of inhibiting speech in certain settings due to anxiety. Parents who reported no diagnosis of SM, no suspicions of SM and no pattern of inhibiting speech due to anxiety were screened out of the study. Participants with comorbid DSM-5-TR diagnoses were not excluded from this study due to the documented likelihood of children with SM meeting criteria for another disorder [3].

The number of parents who participated in this study via online recruitment was 226. Eight instances of duplicated data were deleted. Additionally, five participants were deleted due to ineligibility (e.g., parents of a child older than 12 years). Thus, the final sample consisted of 213 online-recruited families. Additionally, 17 participants were included in the sample who were recruited for a separate research study and consented to having their data included in the current study as well. These 17 participants were from a sample of treatment-seeking families who were participating in an intervention study. Thus, the total combined sample consisted of 230 participants. Of these 230 participants, 24 partially completed the study. Depending on the specific missing data, these 24 participants sometimes were excluded from certain analyses.

Participating parents were between the ages of 25 and 54 ($M = 39.83$, SD = 5.17). Most of the parents reported being White or European American (83.9%), female (95.2%) and married (90.4%). Almost all participating parents reported being the biological mother of the child they were reporting on (95.7%). Families were predominantly from a high socioeconomic status (SES), with 75.4% of families reporting yearly income over $75,000 and only 2.2% of families reporting yearly income less than $25,000. This was an international sample, with families from 15 different countries across five continents participating. However, most of the sample reported living in the US (86.5%). Of participants who did not live in the US, the most common countries of origin included Canada (3.9%), the United Kingdom (1.3%) and Argentina (1.3%). Most participating families lived in a country where English is the primary language spoken (92.6%). However, 20.8% of the sample reported speaking a language other than English in the home. The most common languages reported were Spanish (7.4%) and Chinese (3.5%), which included both Mandarin and Cantonese.

Parents could only report on one child in their home with suspected or diagnosed SM, regardless of whether they had more than one child with SM. The children were between the ages of 3 and 12 ($M = 7.36$, SD = 2.44). Most of the children were White or European American (81.9%), non-Hispanic (87.8%) and female (67.4%). Over half of the children were reported to have at least one additional comorbid psychiatric diagnosis besides SM. See Table 1 for a detailed review of demographic information about the participating children.

Procedures

Data were collected between November, 2019 and April, 2020. Interested participants first completed a screening questionnaire to verify eligibility for the study, as described above. Eligible participants completed the study via an online questionnaire after giving informed consent to participate. Regardless of participant's country of origin or primary language, all participants completed the survey in English. Parents were given the option to enter their personal information to be entered into a raffle to win one of four $25 gift cards as compensation for their participation.

Measures

Demographic and Family History Questionnaire

Parents were asked to complete a demographic and family history questionnaire to gain information about the participating parent, their child and their family. The questionnaire collected basic demographic information (e.g., ages of parent and child, family income, language(s) spoken at

Table 1 Descriptive statistics about participating children

	M (SD)	*n* (%)
Child age	7.36 (2.44)	
Child biological sex		
Female		155 (67.4%)
Male		74 (32.2%)
Not listed		1 (0.4%)
Child race		
White or European American		186 (81.9%)
Black or African American		1 (0.4%)
Asian		12 (5.2%)
Biracial or multiracial		21 (9.3%)
Not listed		7 (3.1%)
Child ethnicity		
Hispanic or Latino/a		28 (12.2%)
Not Hispanic or Latino/a		202 (87.8%)

home). Additionally, this measure asked questions related to the mental health history of the child, including information related to the diagnosis of SM, comorbid diagnoses and previous or ongoing treatments the child has received for SM. Finally, this questionnaire included questions about mental health history in the participating parent and the child's other family members. When reporting on comorbid diagnoses in the participating child and family history of mental health disorders, participants were asked to indicate from a list of common mental health disorders if each disorder was diagnosed (a) in the participating child and (b) in someone else within the family. If participants reported a comorbid diagnosis in the participating child, they were asked to list the age of diagnosis. If participants indicated a disorder was present in the family, they were asked to state who was affected by the disorder. Participants could identify more than one disorder in the child or family and they were instructed to report on everyone in the family. They were also able to report other comorbid diagnoses and disorders present in the family that were not on the list of common disorders. This questionnaire was developed by the first author of this study following a thorough review of the SM literature and identification of variables relevant to the clinical presentation of SM. Interested readers may contact the first author for a copy of this questionnaire.

Selective Mutism Questionnaire

The selective mutism questionnaire (SMQ; [5, 40]) was used as a measure of child SM symptoms for all participants, regardless of the child's current diagnosis. The SMQ is a 23-item parent-report measure for children aged 3 to 11 that assesses for the core symptoms of SM—that is, the absence of speech in a variety of settings and contexts. The SMQ consists of three symptom subscales that provide information about speech in a specific context: At School, Home/Family, and In Social Situations (Outside of School). For each of these three subscales, a variety of speaking tasks are listed and the parent rates how often their child speaks appropriately in each situation using a 4-point Likert scale ranging from 0 to 3 points. Lower scores on the SMQ indicate less frequent speech, and therefore more severe symptoms of SM. Each of these subscales are scored by calculating an average of the items within each subscale (range 0–3). The SMQ also provides a Total Score, which is calculated by adding together the sum of all items on these three symptom subscales (range 0–51).

The average Total Score for a child with SM is 12.99 (SD = 7.23), whereas the average total score for a child without SM is 46.00 (SD = 5.94; [40]). The measure does not include a cutoff score for diagnosing SM. The SMQ also has an Interference/Distress Subscale which is not included in the Total Score. The SMQ has been found to have good psychometric properties, including good convergent validity, discriminant validity, and incremental validity [40, 41]. Furthermore, the SMQ has been found to have excellent internal consistency (Total Scale $\alpha = 0.97$, School subscale $\alpha = 0.97$; Home/Family subscale $\alpha = 0.88$; Public/Social $\alpha = 0.96$; [40]).

Screen for Childhood Anxiety-Related Emotional Disorders

The parent-report version of the Screen for Childhood Anxiety-Related Emotional Disorders (SCARED; [42]) was administered as a measure of children's anxiety symptoms. The SCARED is a 41-item parent-rated measure of a child's anxiety symptoms across a variety of domains. Parents are asked to rate how often their child displays a range of anxiety symptoms using a 3-point Likert scale. The SCARED yields a Total Score as well as five subscale scores for symptoms related to generalized anxiety, social anxiety, school avoidance, panic/somatic symptoms and separation anxiety. Higher scores indicate more severe anxiety. The SCARED is scored using a clinical cutoff system designed for use with children ages 8 to 18. Due to the inclusion of children younger than 8 in this sample, this study does not rely heavily on use of the cutoff scores. In evaluating the psychometric properties of the SCARED, researchers have identified good internal consistency across scales and subscales, with α values ranging from 0.78 to 0.87 [42]. Additionally, the SCARED has been found to have good discriminant validity with anxiety disorders versus depressive disorders as well as with anxiety disorders versus disruptive behavior disorders [42].

Data Analyses

Frequency statistics were conducted to examine the characteristics of the sample with regard to categorical demographic variables (e.g., gender, comorbidities). Additionally, descriptive statistics were conducted to analyze characteristics of the sample regarding continuous variables (e.g., age of onset). Further, independent samples *t*-tests were run to compare continuous demographic variables across subgroups (e.g., age of diagnosis and gender). Finally, bivariate correlations were run to examine relationships between continuous variables (e.g., age and symptom severity).

Finally, content analysis was performed on a question included in the demographic questionnaire asking parents: "Has your child ever received special education services in school (e.g., special education, IEP plan, 504 plan)? If yes, describe services received." Initially, two members of the research team went through the 98 responses and created a frequency count based on mentions of services. Members of the research team then reviewed the initial responses and discussed creation of categories. Using the agreed-upon

categories, the initial two researchers coded responses accordingly. Outlying responses were reviewed for placement until consensus was reached.

Results

Aim 1: Course and Clinical Features of SM

Prevalence and Diagnostic Patterns

The majority of children in the sample had received a diagnosis of SM (91.7%). Based on parent's retrospective reports, the average age of diagnosis was 5.08 years (SD = 1.83) and the average age of onset of SM symptoms was 2.85 years (SD = 1.24). Parents most commonly reported that their child was first diagnosed with SM by a psychologist (35.2%) or a pediatrician (10.0%). Others reported their child was first diagnosed with SM by multiple providers (8.7%) or at a hospital/specialty clinic (8.7%). About two-thirds of participating children were female (67.4%). Male children ($M = 3.22$ years, SD = 1.34) had a significantly later onset of symptoms than female children [$M = 2.67$ years, SD = 1.15; $t(225) = 3.14$; $p < 0.001$]. Similarly, male children ($M = 5.53$ years, SD = 2.04) were diagnosed significantly later than female children [$M = 4.87$ years, SD = 1.69; $t(198) = 2.42$; $p < 0.05$].

Comorbidities

A little over half of this sample (50.9%) had at least one comorbid psychiatric disorder, as reported by the parent. Of the children with at least one comorbid diagnosis, 85.1% had at least one comorbid anxiety disorder. The most common comorbid disorders were SAD/SP, GAD and speech/language disorders (34.3%, 19.6% and 11.7% of the total sample, respectively). There were no significant differences in the presence of comorbid SAD/SP, separation anxiety, GAD or speech/language disorders between male and female children. See Table 2 for a review of comorbidities in this sample.

Scores on the SCARED were used to further evaluate comorbid anxiety in this sample. Total Scores on the SCARED varied from 1 to 72, with an average score of 30.49 (SD = 14.18). It should be noted that this average score is above the cut-off indicative of clinically elevated anxiety symptoms (≥ 25; [42]). Children showed the most notable elevations on the social anxiety subscale ($M = 11.06$, SD = 3.18), which was also above the cut-off suggestive of social anxiety disorder (≥ 8; [42]). Elevations on the other subscales were variable. Age was not significantly correlated with total anxiety scores. However, age was significantly correlated with symptom severity on both the separation anxiety subscale [$r(210) = -0.25$, $p < 0.001$] and social anxiety subscale [$r(209) = -0.18$, $p < 0.05$), with younger children being more severely symptomatic on both of these. No differences in the severity of anxiety symptoms based upon parent-reported gender were found. See Table 3 for details about the severity of anxiety symptoms in this sample.

Table 2 Rates of comorbid disorders in participating children

Disorder	*n* (%)
Any comorbid mental health disorder	117 (50.9%)
Any anxiety disorder	100 (43.5%)
Social anxiety disorder	79 (34.3%)
Generalized anxiety disorder	45 (19.6%)
Separation anxiety disorder	19 (8.3%)
Panic disorder	3 (1.3%)
Obsessive–compulsive disorder	9 (3.9%)
Attention-deficit/hyperactivity disorder	13 (5.7%)
Autism spectrum disorder	4 (1.7%)
Speech/language disorder	27 (11.7%)
Specific learning disorder	9 (3.9%)
Depressive disorder	1 (0.4%)
Oppositional defiant disorder	1 (0.4%)
Other comorbid disorder	17 (7.4%)

Parents could report multiple comorbid diagnoses

Table 3 Severity of SM and anxiety symptoms in participating children

Measure	*M* (SD)	Range
SMQ total score	22.78 (9.84)	4–51
SMQ at school subscale	1.13 (0.83)	0–3
SMQ home/family subscale	2.03 (0.61)	0.5–3
SMQ social situations subscale	0.80 (0.69)	0–3
SMQ interreference/distress subscale (not in total)	1.63 (0.65)	0–3
SCARED total score	30.49 (14.18)	1–72
SCARED panic disorder/somatic subscale	4.07 (4.47)	0–26
SCARED GAD subscale	7.46 (4.69)	0–18
SCARED separation anxiety subscale	5.84 (3.95)	0–15
SCARED social anxiety subscale	11.06 (3.18)	0–14
SCARED school avoidance subscale	2.05 (2.11)	0–8

SCARED total score cut-off score is 25. SCARED Subscale cut-off scores are 7, 9, 5, 8 and 3, respectively

SMQ selective mutism questionnaire, *SCARED* screen for childhood anxiety-related emotional disorders

Family History

Approximately two-thirds of the participants (64.3%) reported a family history of at least one psychiatric disorder. Family history of GAD (33.0%), depression (30.0%),

attention-deficit/hyperactivity disorder (16.0%) and SAD/SP (15.7%) were the most frequently reported. A family history of SM was reported by 10.0% of participants. See Table 4 for details about reported psychiatric disorders in first-degree family members, as well as extended family.

Selective Mutism Symptomatology

Severity of SM symptoms were somewhat variable in this sample. Total Scores on the SMQ ranged from 4 to 51, which is the maximum possible score. Higher scores indicate less severe SM symptoms, suggesting some participants were in remission from SM at the time of the study. The average score on the SMQ for this sample was 22.78 (SD = 9.84), lower than that of children without SM in the standardization sample ($M = 46.00$, SD = 5.94; [40]) but higher than the average of the SM standardization sample (12.99, SD = 7.23; [40]). Severity of SMQ symptoms was significantly correlated with child age, such that younger children had more severe SM symptoms than older children, $r(214) = 0.22$, $p < 0.01$. On subscales of the SMQ, children had the most severe SM symptoms in social settings outside of school ($M = 0.80$, SD = 0.69) and the least severe SM symptoms at home and with family ($M = 2.03$; SD = 0.61). There were no differences in the severity of SM symptoms based upon parent-reported gender. See Table 3 for details about the severity of SM symptoms in this sample.

Aim 2: Types of Treatments and Services Received

Clinical Treatments

The vast majority of children (83.9%) had reportedly received therapy for their SM, either in the past or ongoing at the time of the study. The most commonly reported type of therapy was individual CBT therapy (61.7%). Additionally, almost half of the parents (47.5%) reported that their child had participated in play therapy at some point in the course of their SM treatment. Finally, participation in intensive CBT treatments through either individual or group modalities was reported by 29.6% of parents. Children who had never received therapy for their SM ($M = 18.06$, SD = 7.08) had significantly more severe overall SM symptoms on the SMQ than those who had received therapy [$M = 23.69$, SD = 10.07; $t(222) = 3.21$; $p < 0.01$]. Similarly, children who had never received therapy ($M = 12.15$, SD = 2.23) had significantly more severe social anxiety symptoms on the SCARED than therapy recipients [$M = 10.87$, SD = 3.30; $t(217) = 2.17$; $p < 0.05$].

In addition to psychotherapy, about one-third of parents in the study reported that their child has taken psychiatric medication for their SM (30.8%). The most commonly prescribed medication was Fluoxetine (17.0% of the total sample), followed by Sertraline (14.8% of the total sample). Only a small portion of parents reported that their child took a medication for their SM that was not an SSRI (2.6% of total sample). About one-quarter of parents reported that their child was currently taking at least one medication for

Table 4 Rates of family history of mental health disorders

	n (%) in any relative	*n* (%) in first-degree relatives (parents and siblings)
Any family history of mental health disorders	148 (64.3%)	
Any anxiety disorder	116 (50.4%)	
Selective mutism	23 (10.0%)	19 (8.3%)
Social anxiety disorder	36 (15.7%)	28 (12.2%)
Generalized anxiety disorder	76(33.0%)	46 (20.0%)
Separation anxiety disorder	5(2.2%)	4 (1.7%)
Panic disorder	27(11.7%)	19 (8.3%)
Obsessive–compulsive disorder	12 (5.2%)	8 (3.5%)
Attention-deficit/hyperactivity disorder	39 (17.0%)	30 (13.0%)
Autism spectrum disorder	17 (7.4%)	8 (3.5%)
Speech/language disorder	19(8.3%)	14 (6.1%)
Specific learning disorder	13 (5.7%)	6 (2.6%)
Depressive disorder	69 (30.0%)	33 (14.3%)
Oppositional defiant disorder	1(0.4%)	1 (0.4%)
Posttraumatic stress disorder	14 (6.1%)	9 (3.9%)
Bipolar disorder	13 (5.7%)	3 (1.3%)
Schizophrenia	10 (4.3%)	1 (0.4%)
Other disorder	5 (2.2%)	3 (1.3%)

their SM at the time of participating in the study ($n = 58$, 25.2%). Children who were taking medication for their SM at the time of the study ($M = 26.28$, SD = 10.26) had significantly less severe overall SM symptoms on the SMQ than those who were not taking medication [$M = 21.57$, SD = 9.42; $t(223) = 3.20$; $p < 0.01$]. See Table 5 for detailed information about interventions reported in this sample.

School Services

The majority of children received accommodations and/or interventions within the school setting ($n = 142$, 61.7%). The kinds of services received included IEP plans (44.0%), section 504 Plans (34.8%) and informal, personalized or other types of services (21.3%). This category of informal, personalized or other services included services received by children located outside of the US. Of the children who had IEP plans, the most common IEP classifications were Other Health Impairment (29.8%), Emotional Impairment (20.8%) and Speech/Language Impairment (13.4%).

Parents were also asked to describe the specific interventions, services and accommodations that their child received in school. The two coders had 80.1% agreement with an interrater reliability found to be kappa = 0.93. There was considerable variability in the services that children were reported to receive for their SM. The most commonly listed services were speech therapy ($n = 39$, 39.8%) and in-school therapy/counseling ($n = 22$, 22.4%). Several other modalities of therapy were reported at lower rates, including physical therapy ($n = 6$, 6.1%), occupational therapy ($n = 6$, 6.1%), and play-based therapy ($n = 2$, 2.0%). In terms of specific interventions for SM, stimulus fading/"fading in" ($n = 8$, 8.2%), and exposures ($n = 16$, 16.3%) were most frequently reported. In terms of accommodations, both academic-specific and SM-specific accommodations were reported. For academic support, the most commonly reported accommodations were special education classes for specific topics such as math and writing ($n = 5$, 5.1%). SM-specific accommodations included oral accommodations for presentations and spoken exams (5, $n = 5.1$%) and a buddy system where a friend would attend all of the same classes as the child with SM (6, $n = 6.1$%). Lesser-mentioned accommodations of note included allowing the substitution of writing for speaking (2, $n = 2.0$%), not requiring/pushing the child to speak (3, $n = 3.1$%), and allowing the child to work in or present to smaller groups (2, $n = 2.0$%).

Table 5 Interventions and treatments received by participating children

Treatment	*n* (%) overall
Any therapy for SM	193 (83.9%)
Individual CBT	142 (61.7%)
Individual play therapy	109 (47.4%)
Other individual therapy	25 (10.9%)
Family therapy	22 (9.6%)
Group CBT	32 (13.9%)
Other group therapy	7 (3.0%)
Any intensive treatment	68 (29.6%)
Intensive CBT	41 (17.8%)
Group intensive/SM "Camp"	44 (19.1%)
Other therapy for SM	28 (12.2%)
Any medication taken for SM	80 (34.8%)
Fluoxetine	39 (17.0%)
Sertraline	34 (14.8%)
Citalopram or escitalopram	10 (4.3%)
Other SSRI medication	4 (1.7%)
Other non-SSRI medication	6 (2.6%)

Parents could report multiple types of therapy

SM selective mutism, *CBT* cognitive behavioral therapy, *SSRI* selective serotonin reuptake inhibitor

Discussion

Prevalence and Diagnostic Patterns

Regarding basic demographic patterns, the results of this study are generally consistent with previous literature. First, the results show an average age of SM symptom onset before age 3, which is consistent with previous data based in the US (2.7 years, [2]). However, this finding is somewhat earlier than several studies conducted in Europe (3.7 years [3], 4.6 years [4], 4.15 years [17]). It is possible that this finding reflects cultural differences between the US and European countries, such as the high prevalence of early preschool attendance in the US, particularly among higher SES families. Further, this sample was about two-thirds female, which is consistent with previous literature finding approximately a 2:1 female to male ratio (e.g., [2, 11, 17]). Male participants in our sample were reported to show onset of SM symptoms and be diagnosed with SM significantly later than female participants, which is contrary to previous studies that found male children tended to show symptoms and be diagnosed earlier [9, 17]. This cohort was diagnosed around age 5 on average, earlier than cohorts in previous studies conducted in Europe (e.g., 5.5 years [3], 7.9–8.1 years [9], 8.8 years [43]). This earlier age of diagnosis may be partially due to improvements in awareness and identification of SM during the last 20 years, as well as cultural differences in age of school entry in the US and Europe.

This finding that SM diagnosis appears to be occurring earlier than previously reported is encouraging, as research has indicated that early intervention is most effective [12, 44, 45]. However, despite apparent improvements, this study

still found a discrepancy between symptom onset and diagnosis of about two to three years. Clinicians, educators and researchers working with selectively mute children should continue focusing on finding ways to ensure earlier referral to treatment. Since children are usually highly symptomatic in school settings, earlier identification, diagnosis and treatment will rely, in part, on increasing awareness and education about SM in schools and preschools.

Comorbidities

In looking at parent-reported patterns of comorbidities, these findings revealed similar diagnostic overlap between SM and other anxiety disorders as reported in previous studies [20]. However, specific rates of comorbidity in this sample are somewhat lower than previous research has revealed. For example, our finding of 34.3% comorbidity with SAD/SP is notably lower than many older studies have reported [2, 3, 17, 21]. It may be that conceptualizations of SM have shifted in the last 20 years such that SAD/SP is no longer commonly diagnosed alongside SM in clinical practice. Various research groups have addressed the diagnostic overlap between SAD/SP and SM and identified clinical distinctions in these two presentations. For example, compared to SAD/SP, children with SM have been reported to have higher anxiety about speech-demanding situations, significantly less verbal participation in social situations, higher symptoms reported in school settings, higher externalizing behaviors, and higher comorbidity of oppositional defiant disorder [17, 23–25]. Additionally, a cluster analysis identified three possible subgroups of children with SM that are also distinct from SAD/SP: those with high anxiety about speaking, those with increased presentation of mild oppositional behavior symptoms, and those with additional speech and syntax concerns [26].

Further research into the clinical and phenomenological differences between SM and SAD/SP may help broaden our understanding of the overlap between these two conditions and true rates of comorbidity. Additionally, speech/language disorders and separation anxiety disorder were commonly comorbid with SM in the present sample, but rates (11.7% and 8.9%, respectively) were also lower than in previous studies (30–50% and 13–32%, respectively; [3, 17, 26, 28]).

There were also some differences in comorbidity patterns in this sample compared to what has previously been reported. First, one of the most commonly reported comorbid diagnoses in this sample was GAD (19.6%), which had not been reported at such a high rate in previous studies (6%, [20]). Additionally, only 1.7% of the sample reported a comorbid diagnosis of an autism spectrum disorder (ASD), whereas previous studies have identified higher comorbid rates of ASD ranging from 7 to 63% [3, 43]. Little is currently known about the relationship between SM and ASD, although some researchers are beginning to address this unique clinical presentation [46]. Similarly, previous research identifying high rates of comorbid elimination disorders, oppositional behavior disorders and developmental delays with SM were not replicated in this sample [2, 3, 17]. These differences in comorbidities may reflect clearer diagnostic criteria for SM and other childhood psychiatric disorders since the introduction of the DSM-5 in 2013, as well as the development and use of more accurate assessment tools over time.

Family History

A growing body of literature has examined patterns of family history in children with SM, as well as risk factors for developing SM based on parental and family mental health characteristics. The present study replicated findings that a broad range of psychiatric disorders are present in family members of children with SM. Although these rates were lower than some previous studies have reported (e.g., [2, 27], the present study also found elevated rates of SM and SAD/SP in relatives, with the majority of these cases in first-degree relatives (i.e., parents and siblings). Looking beyond these closely related conditions, high rates of depressive disorders and other anxiety disorders, most notably GAD, in family members of children with SM were found. Recent findings have indicated that having a parent with any psychiatric diagnosis increases the odds of a child having SM, especially if both parents have a diagnosis [9] and that parents of children with SM had a higher severity of clinical symptoms than parents of children with GAD [28]. Thus, findings from the present study are consistent with literature supporting a high incidence of psychiatric disorders, especially SAD/SP, in families of children with SM.

Clinical Treatments

In addition to confirming and expanding upon previous research findings related to clinical patterns, this study also significantly expands the field of literature on SM by addressing less commonly researched phenomena related to treatment. Previous studies have identified shifts in the theoretical orientation of research on SM intervention in the last 40 years away from psychodynamic and family systems orientations and toward cognitive-behavioral orientations [13, 31]. However, little research to date has examined what treatments families with SM are receiving in clinical settings around the US and internationally. Recent treatment reviews have shown that behavioral and cognitive-behavioral treatments are the most effective treatments for SM [13, 31], which were the most commonly reported treatments in this sample. However, a wide variety of other treatment models and modalities were also reported in this sample, including

play therapy, family therapy and other/unspecified therapies, for which there is less consistent research support. These results demonstrate that, while evidence-based practices are being widely used clinically, there remains large variability in the types of treatment received and many families may still be receiving treatments that do not reflect the current state of the research. However, children in this sample who had never received treatment for SM had more severe SM and social anxiety symptoms than those who had received therapy, which suggests that receiving any therapy for SM may be better than receiving none, regardless of the treatment model. It is also important to note that, while these results seem to suggest that a large proportion of children who have access to treatment are receiving evidence-based care and their symptoms are improving through treatment, the present sample was relatively homogenous and may not be representative of all children with SM across demographic groups. Increased access to education and awareness of SM in lower SES school districts and communities is necessary to ensure earlier identification of SM and access to treatment for all children.

It is also interesting to note that a sizable proportion of this sample reported receiving intensive therapy for SM through individual and group modalities (29.6% of the total sample). Research interest in intensive treatments for SM has been increasing in recent years, with intensive group behavioral treatment recently showing promise in a preliminary randomized controlled trial [34]. Increases in the availability of intensive treatment programs for SM demonstrate a clinical and social interest in this modality, which could account for the high utilization of intensive treatments in this sample. These findings suggest that although intensive treatments may not be the standard of care for children with SM, they are being used relatively widely in clinical practice. It should be noted that 17 families (7.4% of the sample) were enrolled in an intensive one-week camp program that had both child and parent treatment components in a group format.

Regarding medication, reviews of the efficacy of psychotropic medications for SM have identified selective serotonin reuptake inhibitors (SSRIs) as being effective treatments [35]. Of the 80 children in the present sample who had taken psychotropic medication for their SM, only six (7.5%) reportedly ever were prescribed non-SSRI medication. Thus, again, it seems that the vast majority of children with SM who are taking medications are receiving treatments supported by research evidence.

School Services

This study constitutes the first systematic inquiry into interventions and accommodations being received within the school setting by children with SM. Research has consistently highlighted the benefits of targeting mutism in the school setting as part of treatment for SM, since children with SM tend to be highly symptomatic at school [36]. This study confirmed that children with SM commonly qualify for IEPs with a designation of Other Health Impairment, Emotional Impairment and/or Speech/Language Impairment [6]. Almost two-thirds of the children in this study were reported to be receiving some form of accommodations or interventions in the school setting through IEPs, section 504 Plans, or other plans. However, there was a great degree of variability in the types of services provided, and thus consistent patterns regarding the nature of school-based services for SM were difficult to discern. These findings have implications for schools and clinicians serving children with SM, as they suggest a relatively widespread precedent around the US for children with SM to qualify for an IEP due to their diagnosis. They also open the door for researchers and clinicians to work toward establishing standardized evidence-based recommendations for in-school services for SM.

Limitations and Future Directions

The present study has two primary limitations worth addressing. First, the current sample was predominantly White/European American with high income and SES. This sample was recruited from existing social media pages and advocacy groups devoted to SM, as well as specialty clinics for treatment of SM and other anxiety disorders. Due to these avenues of recruitment, this sample largely represents families who are well-informed about SM and have had the resources to secure a diagnosis and treatment for their child. Further, while English as a first language was not required to participate, surveys were administered in English and therefore written competency in English was necessary for completion of the surveys. This may have impacted the diversity of individuals who were able to participate in our study, especially internationally. To attempt to combat these biases and diversify the sample, this study included children for whom parents reported problematic levels of SM symptoms, but were not yet formally diagnosed (suspected SM). These children tended to be younger and have more severe symptoms. The suspected SM sample was recruited into this study in the hopes of including families that may have fewer resources or less awareness of the disorder. It is likely that families from lower income brackets are seeing continued underdiagnosis, lack of awareness of the disorder, and limited access to evidence-based treatment. In fact, past research from Europe has shown high rates of SM in low-SES families [9], but these patterns have yet to be explored within the US. As such, it is imperative that researchers not only investigate the prevalence of SM in low-SES samples, but also the discrepancy in availability and utilization of services between higher and lower income families. While the demographics of this sample limit the generalizability of the results, the findings of this study offer new and useful

information about SM that will hopefully increase awareness of this disorder and assist in increasing diagnosis and treatment for underserved populations.

Another limitation of this study is the sole use of parental reports to gather data about demographic and treatment patterns. Relying exclusively on parent-report data could have impacted the validity of these results due to measurement bias, and thus findings should be interpreted through this lens. Additionally, information about the age of SM onset, comorbid diagnoses, treatments received, and family history of psychiatric disorders were all obtained via parent's retrospective reporting about their child and family. Parents who were not heavily involved in their child's treatment or are naïve to treatment and diagnosis may not have understood the theoretical orientation of these services or comorbid diagnoses given, for example. Thus, future studies would benefit from more varied methods of data collection to verify the results of this study. Another limitation related to the methodology of this study was the use of the SCARED to evaluate anxiety symptoms in children younger than age 8, as the SCARED is designed for use with children aged 8 to 18. Due to our use of the SCARED for younger children, we did not rely heavily on the cut-off scores, which were set based on symptoms of children aged 8 to 18.

As a result of the contributions of the current study to the SM literature, a couple avenues for future research directions are recommended. First, as discussed earlier, the literature is still unclear as to how multilingualism and immigrant status relate to SM symptomatology and prevalence [18, 19]. A relatively large subset of children in this study reportedly spoke two or more languages, although the present study was unable to fully explore links between SM symptoms and multilingualism. Thus, the field of SM literature would benefit from more rigorous, high-powered studies investigating the link between multilingualism, immigrant status and SM. Second, data collection for this study primarily occurred before the COVID-19 pandemic began. As such, pandemic-related factors that might impact SM diagnosis and symptomatology—such as school attendance, participation in online learning environments, age of school entry, school re-entry and changes to family systems—were not investigated. As engagement with peers and adults outside of the home environment is key to understanding and treating SM, researchers should investigate immediate, delayed, and possible future impacts of the pandemic on both rates of diagnosis and protracted treatment outcomes.

Summary

Examinations of data from a sample of 230 children with SM and suspected SM between the ages of 3 and 12 revealed updated information about clinical patterns for this disorder and common interventions and services received by children with this condition. Gender ratio, comorbidities, and patterns of family history generally were aligned with previous research. We found a female to male ratio of 2 to 1, similar comorbid anxiety disorders, and similar family history of anxiety, depression and other mental health conditions as previously reported. Reported age of SM onset and age of diagnosis were both earlier than previously reported, with an average delay of about 2 years between onset and diagnosis. Overall, anxiety symptoms and social anxiety symptoms were elevated in this sample of children with SM, with elevations higher in children who had not received treatment. The majority of children in this sample received therapy and school accommodations for their SM, and history of treatment with psychiatric medications was reported in about one-third of the sample. There was large variability in the types of treatments and services received by these children, but the majority of participants reported a history of treatment with evidence-based interventions, suggesting success in disseminating evidence-based treatments for SM in clinical and school settings. This study offers information about the presentation of SM to help treating professionals assess for the disorder, secure and advocate for evidence-based interventions in school and clinical settings, and further increase awareness about SM.

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Declarations

Conflict of interest The authors declare that they have no relevant financial or non-financial interests to disclose.

Informed Consent Informed consent was obtained from all individual participants included in the study.

Research Involving in Human and Animal Participants All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

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