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Selective mutism: A review and integration of the last 15 years ☆

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ABSTRACT

Selective mutism (SM) is a rare childhood disorder characterized by a lack of speech in one or more settings in which speaking is socially expected. A comprehensive and uniform theory about the etiology, assessment, and treatment of SM does not exist. Historically, varying definitions and criteria have been applied to children with SM, therefore making comparisons between studies somewhat difficult. Accumulating findings on the phenomenology of SM point to a complex and multidetermined etiology. Developmental psychopathology represents a useful heuristic for conceptualization of SM and serves as an integrative framework for organizing the sometimes disparate findings that permeate the SM literature. The purpose of this review is to summarize the literature on SM, including phenomenology, assessment, and treatment, with the main goals of clarifying its clinical presentation, offering a theoretical understanding of SM from a developmental psychopathology perspective, and highlighting both research and practice gaps that may exist. Recommendations for future research are made with the goal of expanding the current knowledge base on the etiology of SM.

Contents

Selective mutism (SM) is a rare childhood disorder characterized by a lack of speech in settings in which speaking is socially expected (e.g., school). Such lack of speech occurs despite the presence of normal or nearly normal speech in other situations. A child with SM may not talk to teachers or peers at school, but speak fluently to

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parents and siblings at home. In order to meet *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text revision; American Psychiatric Association [APA], 2000) criteria for SM, the child's failure to speak in a particular setting/situation must have been present for at least one month and must not be better accounted for by the presence of a pervasive developmental disorder, communication disorder, or a psychotic disorder (e.g., schizophrenia). Because many young children are apprehensive in new settings, a diagnosis of SM is not assigned if the child is in the first month of school, as a certain level of anxiety – including a lack of speech – could be developmentally expected. Finally, the disorder must cause social and academic impairment.

In part because of the rarity of SM, population-level prevalence rates have been difficult to establish and can vary (see Table 1). While some studies have used *DSM-IV* (APA, 1994) criteria, others followed the diagnostic scheme proposed in the *International Classification of Diseases* (10th edition; World Health Organization, 1992). Studies also included samples of children from different settings (e.g., schools, clinics), ages, and countries of origin. Overall, with the exception of two studies (Kopp & Gillberg, 1997; Kumpulainen, Räsänen, Raaska, & Somppi, 1998), prevalence rates oscillate in the .47%–.76% range (see Table 1). The difficulty in establishing prevalence rates is also compounded by lack of a comprehensive and uniform theory about the etiology, assessment, and treatment of SM. Many studies originally relied on chart reviews to derive diagnoses; standardized structured assessments were rare. Most importantly, few attempts (Cohan, Price, & Stein, 2006) have been made to understand SM from a developmental psychopathology perspective, despite accumulating findings pointing to a complex and multidetermined etiology. Developmental psychopathology represents a useful heuristic for conceptualizing SM and serves as an integrative framework for the sometimes disparate findings that permeate the literature.

This review summarizes the literature on SM with the main goals of clarifying its clinical presentation, offering a theoretical understanding of SM from a developmental psychopathology perspective, and highlighting the existing research and practice deficiencies. Our goal includes making recommendations to stimulate needed research on a disorder not yet well understood. Although initial investigations and case studies provide rich clinical detail, the present review centers on the last 15 years of research, which we consider to be representative of the period when the methodologically strongest – albeit not ideal – studies were published. The search for articles was conducted through extensive electronic database search on PsycINFO, PubMed (Medline), and Web of Science using relevant keywords (e.g., selective mutism, elective mutism, social phobia, mutism, lack of speech, muteness) and these abstracts were reviewed for their relevance. Selected books and chapters were also consulted for relevant information, as well as reference lists. Articles were excluded if they were (a) not published in peer-reviewed journals, (b) published before 1992, (c) written in a language other than English, and (d) case studies. Because the majority of the SM treatment literature consists of case studies, they did not qualify for inclusion in this review. Thus, the reader will note that the treatment section aims specifically at highlighting the dire need for larger treatment studies (i.e., randomized clinical trials) that may better shed light on the efficacy of the currently proposed interventions for SM. The review begins with a brief description of developmental psychopathology and its main tenets. This is followed by a comprehensive review of the available literature on SM. A final summary section integrates the findings of the review from a developmental psychopathology perspective, highlighting how this organizing framework may provide theoretical and practical guidance to the understanding of this rare and sometimes puzzling disorder.

Table 1
Recent prevalence estimates of SM and associated study characteristics

Study	Prevalence[a]	Country	*N*	Age	Sample	Diagnostic criteria
Steinhausen and Juzi (1996)	.47	Switzerland	12,438	7.81[b]	Clinic	ICD-10
	.54	Germany	4093	9.74[b]	Clinic	ICD-10
Kopp and Gillberg (1997)	.18	Sweden	2793	7–15	School-based	DSM-IV
Kumpulainen et al. (1998)	1.90	Finland	2009	7–10	School-based	DSM-III-R
Bergman et al. (2002)	.71	USA	2256	K-2[c]	School-based	DSM-IV
Elizur and Perednik (2003)	.76[d]	Israel	8475	4–6	School-based	DSM-IV
Chavira, Stein, Bailey, and Stein (2004)	.50	USA	190	8–17	Clinic	DSM-IV

Note. ICD-10 = International Classification of Diseases, 10th edition; DSM-III-R = Diagnostic and Statistical Manual of Mental Disorders, 3rd edition, revised; DSM-IV = Diagnostic and Statistical Manual of Mental Disorders, 4th edition.

[a] In percentage.

[b] Mean age at assessment.

[c] Grade in school.

[d] Overall prevalence rate. Prevalence rate for children of immigrant families (n = 1441) was 2.2%; prevalence rate for native families was .47%.

1. Previous literature on SM

Our decision to restrict this review to the last 15 years does not imply that the earlier literature was not informative and worthwhile. Indeed, we are cognizant that the earlier studies and reviews (e.g., Kratochwill, 1981) were key in elucidating important aspects related to the phenomenology, assessment, and treatment of SM. At the same time, our focus on contemporary research consolidates the literature utilizing stronger methodologies and assessments. Indeed, by restricting our focus, we are able to review a literature based on shared definitions and ideas, allowing a stronger scientific analysis based on diagnostic samples that are quite similar in psychopathology.

2. Developmental psychopathology: an overview

The developmental psychopathology perspective (Cicchetti, 1984) conceptualizes pathology as a dynamic process resulting from multilevel, complex transactions (see Sameroff, 1975) between the individual and the environment over time. These inherent complexities require multiple levels of analysis to gain a comprehensive understanding of human development, both normal and abnormal (Cicchetti & Toth, 1997). Genetic, biological, neurological, cognitive, and interpersonal domains that operate within the individual as well as the effects of the various ecological layers in which the developing organism functions (Bronfenbrenner, 1979; see also Kuperminc & Brookmeyer, 2006) are purported as essential in the process of understanding the etiological mechanisms implicated in the development of childhood psychopathology.

Developmental psychopathologists view psychopathology as *deviations* from normal developmental trajectories, and as a longitudinal *process* continuously being shaped by intra-and-extra-individual influences. Thus, adaptive and maladaptive outcomes can only be understood relative to the context in which they take place (Cummings, Davies, & Campbell, 2000). This complex, ongoing interplay of transactional forces implies that no single risk factor can accurately and fully predict a particular psychopathological pathway. Children or individuals may display the same behaviors or be subjected to the same set of circumstances, yet follow very different developmental trajectories, a phenomenon known as *multifinality*. On the other hand, individuals may start developmentally at very different places and end up on the same trajectory, a phenomenon known as *equifinality* (see Cicchetti & Toth, 1997).

As noted above, developmental psychopathology integrates concepts from multiple theoretical perspectives (e.g., biological, genetic, developmental, psychodynamic, behavioral, family systems, ecological). Based on this conceptual model, multiple methods are needed to

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understand psychopathological phenomena, as no single methodology accurately captures the complexity of ontogenic development. Methods germane to each theoretical perspective are required to gain the specificity needed to inform the complex questions that developmental psychopathology attempts to answer: *why* and *how* psychopathology originates, rather than the mere identification of symptoms (Achenbach, 1979).

3. SM: a brief history

First described by German physician Adolf Kussmaul in 1877, SM was called *aphasia voluntaria*, highlighting the conception that children *voluntarily* withheld speech in certain settings. In 1934, the term *elective mutism* was coined by Swiss child psychiatrist Moritz Tramer (Tramer, 1934), with the new term still suggesting that children elected to remain quiet. In newer diagnostic schemas such as the *DSM-IV* (APA, 1994) and *DSM-IV-TR* (APA, 2000) the term *elective* was changed to *selective* (note that the term elective is still used in other diagnostic nosologies such as the ICD-10; World Health Organization, 1992), to avoid previous conceptualizations of the behavior as inherently volitional (i.e., the child actively refusing to talk).

As the reader may notice, previous definitions operated under the assumption that SM was characterized by oppositionality and manipulative withholding of speech. On the other hand, *DSM-IV* highlights the fact that the child withholds speech when confronted with 'select' contexts. Thus, whereas normal speech occurs at home, the child may be completely silent at school. This latter definition inherently broadens the focus of SM beyond oppositional and defiant tendencies to simply withhold speech. It also includes the possibility that children are reacting anxiously in response to a threatening environment. Speech is not possible due to anxiety (i.e., "scared speechless") (see, e.g., Krysanski, 2003; Sharp, Sherman, & Gross, 2007).

3.1. Age of onset

SM is a "young child" disorder. Overall, mean age of onset ranges from 2.7 to 4.1 years (Cunningham, McHolm, Boyle, & Patel, 2004; Garcia, Freeman, Francis, Miller, & Leonard, 2004; Kristensen, 2000). Often, there is a considerable lag between onset of the disorder and the time of referral. In some instances, SM may go unrecognized until the child enters elementary school (approximately age 5) where (s)he is consistently confronted with the challenge of speaking in a novel social setting. In one sample (Ford, Sladeczek, Carlson, & Kratochwill, 1998) the mean age at diagnosis was 6.5 years. Other samples report a mean lag time between symptom onset and diagnosis of 14.1 months (Kristensen, 2000). Similarly, in their review of a multidisciplinary intervention for SM (Giddan, Ross, Sechler, & Becker, 1997) a girl who had not spoken to anyone outside the home since she was 3-years-old was not referred for services until age 8. One reason that the condition may go undiagnosed for some time is that the child's mutism does not occur at home. Similarly, relative to children who are disruptive, the behavior of a child with SM may be unnoticed by teachers and school personnel — which may also delay appropriate referral for services. This lag period, with its possibility of entrenchment, has important assessment, treatment, and service delivery implications. Earlier identification could lead to earlier intervention thereby perhaps preventing or limiting functional impairment (Schwartz, Freedy, & Sheridan, 2006).

4. Etiology of SM

There is no single identified cause of SM and the disorder may be better conceptualized as arising from the interplay of various environmental and genetic factors (see Cohan et al., 2006). Various etiological theories exist. For example, the psychodynamic perspective (e.g., Yanof, 1996) highlights unresolved internal conflicts as potential causes of SM. Behavioral theorists (Leonard & Topol, 1993) suggest maladaptive reinforcement patterns, whereas family systems theorists (Meyers, 1984) suggest maladaptive family dynamics. A genetic predisposition has been suggested by findings that children with SM often have parents who meet criteria for an anxiety disorder. In a sample of 30 children with SM (Black & Uhde, 1995), a family history of social phobia and of SM was present in 70.0% and 37.0% of first-degree relatives, respectively.

Traumatic experiences have also been suggested as an etiological factor (Andersson & Thomsen, 1998), although recent evidence does not support this view (Black & Uhde, 1995; Dummit, Klein, Tancer, & Asche, 1997; Kopp & Gillberg, 1997; Steinhausen & Juzi, 1996). Using a structured interview (Black & Uhde, 1995), there was significant early trauma in only 4 of 30 children with SM and, among these cases, there was no convincing evidence of a causal relation between trauma and onset of SM. Unlike the Black and Uhde (1995) study, not all research investigations used structured diagnostic procedures and thus, the outcome often is difficult to interpret.

These proposed theoretical models (i.e., unresolved psychic conflicts, genetics, trauma, maladaptive family dynamics, dysfunctional reinforcement) suggest that a developmental psychopathology perspective may be particularly useful to account for these multiple etiological factors. Because developmental psychopathology does not adhere to any one theory (Cicchetti, 1984), multiple pathways are assumed under this integrative framework (i.e., equifinality). Previous theories of SM, when taken together, support this hypothesis with regards to etiology, course, and maintenance. It is thus pertinent to review in more detail the hypothesized pathways.

4.1. Genetic vulnerabilities

Several studies have found that SM and associated anxiety-based conditions (e.g., social phobia) occur in families at a disproportionately high rate (Black & Uhde, 1995). For example, among 45 children with *DSM-III-R* (APA, 1987) (s)elective mutism who were referred for treatment between 1964 and 1979 (Remschmidt, Poller, Herpertz-Dahlmann, Hennighausen, & Gutenbrunner, 2001), 18.0%, 9.0%, and 18.0% of the mothers, fathers, and siblings, respectively, also had a history of mutism. Taciturnity (minimal speech) was found in 51.0%, and 44.0% of the fathers and mothers, respectively. Speech disorders (e.g., stuttering, cluttering) were found in 9.0% of the families ($n=4$).

Other studies (e.g., Andersson & Thomsen, 1998) identified other forms of psychopathology, such as personality disorders and depression, that were elevated in families of children with SM. These data are limited, however, because information on parental psychopathology was not collected for the comparison group. Remschmidt et al. (2001) also found substantial psychopathology among parents of children with SM; 60.0% of mothers had some symptoms of psychopathology, including depression, lack of drive, neurotic disorders, and personality disorders. Among fathers, only 9.0% was free of psychopathology and problems included alcoholism, depression, personality disorders, withdrawal, shyness, and irritability.

In contrast to evaluating these disorders in children, other investigators have examined personality and symptom traits in parents of children with SM (Kristensen & Torgersen, 2001). In comparison to control families, a greater proportion of mothers of children with SM reported a history of shyness and social anxiety (3.7% vs. 38.9%). The same was true for fathers (.9% vs. 31.5%). Parents of children with SM also endorsed more features of schizoid personality than did the control group. Additionally, mothers endorsed more avoidant and schizotypal behaviors, and fathers reported more anxiety in comparison to control families. These findings coupled with those of Remschmidt et al. (2001) suggest that psychopathology, in general, and avoidance, anxiety, and a preference for being alone, in particular, may characterize families of children with SM.

Rates of familial psychopathology may vary with the presence of comorbid communication disorders (see below). Interestingly, when

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parents of children with SM with and without a comorbid communication disorder (50.0%, respectively) were examined separately (Kristensen & Torgersen, 2001), only mothers of SM children without a communication disorder scored higher on the schizoid, avoidant, schizotypal, and dysthymia scales even though both groups were different from controls in histories of shyness and social anxiety. Thus, when children have SM and communication disorders, the family component may be less related to personality variables than to communication and speech difficulties. In contrast, higher rates of psychopathology may exist among the families of children with SM without communication disorders. Further research is clearly warranted.

Overall, the relation between SM and family history of psychopathology has been equivocal. In a recent long-term follow-up of 33 clinic-referred children with SM (Steinhausen, Wachter, Laimböck, & Winkler Metzke, 2006), family histories of psychopathology (assessed via ICD-10 criteria in interviews with the mothers) were positive in 39.4% of cases (see also Steinhausen & Adamek, 1997), which was also the rate of taciturnity in the family. Other researchers (Elizur & Perednik, 2003) found no significant differences on self-reported scores in anxiety, depression, or lack of emotional/behavioral control between mothers of children with SM and mothers of controls. Furthermore, scores were in the normal range for both groups (Elizur & Perednik, 2003). These differences, however, may result from the very different method of assessment used in this study.

Finally, one recent study of family psychopathology (Chavira, Shipon-Blum, Hitchcock, Cohan, & Stein, 2007) found that, relative to controls, parents of children with SM had higher rates of lifetime generalized social phobia (37.0% versus 14.1% in control parents) and avoidant personality disorder (17.5% versus 4.7% in control parents) as assessed by the Structured Clinical Interview for *DSM-IV* Axis I Disorders — Clinician Version (SCID-CV; First, Spitzer, Gibbon, & Williams, 1997) and the Structured Clinical Interview for *DSM-IV* Axis II Personality Disorders (SCID-II; First, Gibbon, Spitzer, Williams, & Benjamin, 1997), respectively. However, these group differences only held for fathers. There were no other significant differences in parental psychopathology. Because these families were recruited via a website (i.e., Selective Mutism Group — Child Anxiety Network) and clinical interviews were conducted over the phone, it is possible that the sample may have been skewed in several ways, thereby influencing the findings. Despite its limitations, these data are important as they support a clear connection between familial social anxiety and SM in children.

4.2. Neurological/neurodevelopmental vulnerabilities

Typically conceptualized in childhood as an index of neurodevelopmental delay, communication disorders are found among children with SM. Premorbid ICD-10 speech and language disorders were present in 30.3% of one SM sample (Steinhausen et al., 2006) and in another (Steinhausen & Juzi, 1996), 38.0% of children with SM had speech or language disorders, most commonly expressive language disorders (28.0%) and articulation disorders (20.0%). Among Norwegian children with SM and controls, 50.0% and 11.5%, respectively, met *DSM-IV* criteria for an additional communication disorder (Kristensen, 2000), most commonly phonological disorder (42.6%), mixed receptive–expressive language disorder (17.3%), and expressive language disorder (11.5%). In the Ford et al. (1998) study described earlier, 19% of participants reported language and speech problems.

Other studies (Manassis et al., 2003) have directly compared language abilities of children with SM and children with social phobia in an attempt to investigate potential differences between these two groups. After examining parents' reports of overall communication ability, phonemic awareness via the Lindamood Auditory Conceptualization Test (LACT; Lindamood & Lindamood, 1971), and receptive language using the Peabody Picture Vocabulary Test-III (Dunn & Dunn, 1997) and the concepts and directions subtest of the Clinical Evaluation of Language Fundamentals (CELF-3; Semel, Wiig, & Secord, 1995), SM children scored significantly lower on only one task (discrimination of speech sounds) than socially phobic children. Although overall group means were in the average range, a subgroup (42.9%) of the children with SM scored in the clinical range on at least one language measure. However, the abnormalities were inconsistent, suggesting that they may be normally distributed and not indicative of an etiological role. No differences were found in parental reports of overall communication ability or in standardized measures of receptive language. Although these findings suggest that there may be deficits in phonetic awareness in children with SM, the study did not include a non-disordered control group and did not correct for multiple comparisons as the findings were considered largely exploratory.

In another comparative study (McInnes, Fung, Manassis, Fiksenbaum, & Tannock, 2004) children with SM had normal receptive language and cognitive abilities but produced shorter, simpler, and less detailed narratives than children with social phobia. As the authors pointed out, these findings suggest that despite similar emotional presentations (e.g., shy, withdrawn, anxious), children with SM may show subtle expressive language deficits not seen in children with social phobia. However, it is important to note this study's small sample size (7 children with social phobia, 7 children with SM) and the lack of inclusion of measures specifically assessing social phobia, which limit the extent to which these groups may be appropriately compared.

In a replication and extension of the Manassis et al. (2003) study, Manassis et al. (2007) examined language abilities (using nonverbal tests), cognition, and anxiety levels across children with SM, children with anxiety disorders, and controls. Children with SM scored significantly lower than the other two groups on language measures of phonological awareness, receptive vocabulary, and grammar. Consistently, percentage of children in the clinical range for language abilities was also greater in the SM group (ranging from 18.0% with respect to receptive vocabulary to 43.0% with respect to grammar). Children with SM also had significant deficits in visual memory in comparison to the other two groups, and deficits in some nonverbal working memory tests in comparison to controls but not to children with anxiety disorders. However, deficits in nonverbal working memory were not entirely consistent across measures and may have been influenced by the motor requirements of some of the tests. The investigators also examined predictors of mutism severity and found that (younger) age of the child, (greater) mother-reported social anxiety, and (poorer) receptive grammar accounted for 38.0% of the variance.

Language problems are only one sign of neurodevelopmental delay. Other indicators include fine and gross motor problems, physical deformities, and delays in socioemotional milestones. These, however, have been far less studied in children with SM than language delay. One study examining these broad markers (Kristensen, 2002) found that children with SM, irrespective of comorbid communication disorders, were overrepresented in comparison to controls on both parental and medical reports of gross (SM=42.6% vs. Controls=7.4%) and fine (SM=25.9% vs. Controls=.9%) motor development delay. They also scored significantly lower than controls on assessment of motor skills and higher than controls in the number of minor physical anomalies present and the number of pre-and perinatal risks factors reported by parents. Thus, neurodevelopmental deficits and delays may play a role in the development of SM. However, as the authors pointed out, most deficits were considered moderate, not severe, and the manner in which motor performance was assessed has not been properly validated (see Kristensen, 2002). Also, all motor assessments were made by one clinician who knew the diagnostic status of the children — likely because, for obvious reasons, a blinded study is difficult in children with SM.

Auditory processing deficits may also play a role in SM. In a study of abnormal auditory efferent activity (AEA) (i.e., deficits in the ability to

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mask the influence of your own voice during vocalization, which increases sensitivity to external noise; Arie et al., 2006), children with SM and abnormal AEA showed significant deficits in the processing of auditory input relative to children with SM but normal AEA and controls. Thus, there may be a subgroup of children with SM who experience significant difficulty processing auditory input during vocalization, which, coupled with high anxiety, may result in speech avoidance. Findings of this study are consistent with those of Bar-Haim et al. (2004), but do not account for the etiology of all cases of SM.

4.3. Psychological vulnerabilities

4.3.1. SM and internalizing pathology: evidence for SM as an anxiety disorder variant

A number of investigators (Beidel & Turner, 2005; Black, 1996; Black & Uhde, 1995; Dow, Sonies, Scheib, Moss, & Leonard, 1995; Garcia et al., 2004) have argued that SM is characterized primarily by anxiety, as evidenced by its clinical presentation (e.g., high anxiety, avoidance) and high rates of comorbidity with other anxiety disorders, in particular social phobia (Anstendig, 1999). Kristensen (2000) found that 74.1% of children with SM had a comorbid anxiety disorder, most commonly social phobia (67.9%) and separation anxiety disorder (31.5%). Manassis et al. (2003) found that 50.0% of children with SM had a comorbid diagnosis of simple phobia. In a recent study (Manassis et al., 2007), 61.4% of children with SM also met criteria for social phobia, consistent with, although higher than, Arie et al.'s (2006) 44.4% comorbidity rate between SM and social phobia. Some authors have actually argued that SM may be a severe variant of SP (Black & Uhde, 1992). The relative success of cognitive–behavioral and behavioral therapies in the treatment of SM further provides support for this view.

In one study (Black & Uhde, 1992), 97.0% of children with SM met *DSM-IV* criteria for social phobia, avoidant disorder, or both. Thirty percent also met criteria for comorbid simple phobia and 17.0% for separation anxiety. Mutism severity was significantly positively correlated with parent ratings of overall anxiety, separation anxiety, and social anxiety. Twenty percent of the children were described as moderately shy and 57.0% as extremely shy. Based on these results, the authors argued that SM may be better conceptualized as a symptom of extreme social anxiety rather than as a distinct disorder.

Other data beyond diagnostic comorbidity confirm presence of anxiety in SM. One study (Steinhausen & Juzi, 1996) found that 85.0% and 66.0% of a sample of children with SM – some of whom were directly assessed and some whose data were obtained by reviewing data banks of clinics at Switzerland and Germany – were rated as shy and anxious, respectively. Among a subset for whom normed data was available (i.e., Child Behavior Checklist [CBCL]; Achenbach, 1991a), children with SM scored significantly higher and in the 'at-risk' range for internalizing and withdrawal problems relative to same-age peers. Another study (Ford et al., 1998) established SM diagnoses according to parental responses on a survey that included *DSM-III-R* and *DSM-IV* criteria for SM. In this sample, 72.2% met DSM-IV criteria. There were significantly higher scores on the CBCL internalizing problems scale relative the externalizing problems scale. This study, however, is limited by its methodology and the finding that 34.0% of the sample had SM in the past, not at the time of the survey. This may explain why, despite significant differences between internalizing and externalizing scales, all *T*-scores were in the normal range. Nevertheless, 84.3%, 78.0%, and 74.8% of participants endorsed the items "shy or timid," "refuses to talk," and "self-conscious or easily embarrassed," respectively, as either "sometimes true" or "often true" on the CBCL. Other studies have found internalizing and withdrawn scores on a teacher-reported measure (Teacher Report Form [TRF]; Achenbach, 1991b) clinically elevated (Bergman, Piacentini, & McCracken, 2002; Vecchio & Kearney, 2005).

More recently, using a multimethod approach, fifteen children with SM were compared to children with anxiety disorders without SM, and a control group (87.0% diagnosis free, 2 children with externalizing disorders; Vecchio & Kearney, 2005). All children with SM received a diagnosis of social anxiety disorder and 53.0% received an additional anxiety disorder diagnosis. There were no differences in parent and teacher ratings of internalizing problems between the SM group and the anxiety disorders group, suggesting a high degree of behavioral similarity between these groups.

Although limited by the lack of a control group, Manassis et al. (2003) also found that children with social phobia and children with SM obtained similar scores on a number of standardized measures of general anxiety and social anxiety. Although not statistically significant, there was a general trend towards greater child-reported separation and physiological anxiety, and parent-reported social anxiety for the social phobia group. The fact that the SM group scored lower than the social phobia group on these measures runs counter to the argument that SM may be a severe variant of social phobia (see, e.g., Anstendig, 1999). Alternatively, it may be that as a result of their behavioral avoidance, children with SM may underreport anxiety symptoms, consistent with the findings of Kristensen (2001).

Using a nationwide sample of 54 of treatment seeking Norwegian children with SM (Kristensen, 2001), parent- and teacher-reported internalizing symptoms were significantly higher for the SM group when compared to controls. Interestingly, the groups did not differ on self-report of internalizing symptoms, suggesting that children with SM may underreport anxiety.

Other studies have found significant differences between children with SM and controls on parent and teacher ratings of anxiety. In comparison to controls, parents and teachers rated children with SM significantly higher on symptoms of anxiety (Cunningham et al., 2004) and higher on obsessive compulsive disorder symptoms. Symptoms of depression did not differentiate between children with SM and controls. Bergman et al. (2002) also found that children with SM were rated significantly higher than controls by teachers on the internalizing, withdrawn, and anxious/depressed subscales, consistent with the conceptualization of SM as an anxiety-based disorder.

Consistent with parent and teacher ratings, children with comorbid SM and SP were rated higher by behavioral observers and clinicians on measures of social distress than children with SP alone (Yeganeh, Beidel, Turner, Pina, & Silverman, 2003). The groups were indistinguishable in terms of self-report measures assessing social anxiety, trait anxiety, and general fears. It is difficult to draw firm conclusions regarding the inconsistency between clinician ratings and self-report measures of social anxiety found in this study. However, the findings do suggest that although children with SM have social anxiety, their experience of it in comparison to the perception of their anxiety by others may differ in important ways.

Recently, social phobia symptoms and parenting styles were compared in children with SM (all of whom met criteria for comorbid SP), children with SP-only, and controls (Yeganeh, Beidel, & Turner, 2006). No significant differences were found between the two diagnostic groups on social phobia symptoms, but clinician severity ratings for the diagnosis of SP were higher for children with SM. This is consistent with previous investigations (Manassis et al., 2007; Yeganeh et al., 2003). Finally, children with SP reported significantly lower parental acceptance than controls, but no differences were found on parenting style dimensions between children with SM and children with SP.

Contrary to other studies (Yeganeh et al., 2003, 2006), Manassis et al. (2007) found that children with SM self-reported significantly higher social anxiety than children with anxiety disorders and controls. However, children with anxiety disorders reported greater overall anxiety than children with SM and controls. Maternal reports of social anxiety did not differ for children with SM and children with anxiety disorders, with both groups scoring higher in comparison to controls. On the other hand, maternal reports of overall child anxiety

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were higher for children with anxiety disorders in comparison to children with SM and controls.

Another investigation (Cunningham, McHolm, & Boyle, 2006) further dichotomized children with SM according to whether their mutism was specific (e.g., did not speak to teachers but spoke to friends at school) or generalized (e.g., spoke only to parents at home), following the diagnostic distinction made in children with social phobia. Children with generalized SM and those with specific SM had similar parent-reported social phobia, generalized anxiety, and obsessive compulsive disorder (OCD) symptoms, despite the fact that children with specific SM spoke across a wider range of social situations (e.g., playground, hallways). Verbal and nonverbal social skills were significantly deficient in both SM groups in comparison to controls as rated by parents and teachers. Despite this, children with SM did not see themselves as less accepted by peers. Overall, these studies suggest that children with SM suffer from internalizing symptoms in general and anxiety and social anxiety in particular.

4.3.2. SM and externalizing pathology: mixed evidence

Although there is substantial evidence of an association between SM and anxiety-based conditions, some studies (e.g., Kumpulainen et al., 1998; Steinhausen & Juzi, 1996) have also reported that a very small proportion of children with SM display controlling, demanding, oppositional, and aggressive behaviors. Black and Uhde (1995) found that only 10% of children with SM met criteria for oppositional defiant disorder and that parent and teacher ratings of conduct disorder and immaturity did not correlate with mutism severity. Similarly, at the item level, scores on teacher-rated items describing oppositional behavior were low and did not correlate with mutism severity. In one recent study (Manassis et al., 2007) 6.8% of children with SM met criteria for oppositional defiant disorder and in another (Arie et al., 2006), 11.1% were comorbid for SM and attention deficit/hyperactivity disorder (ADHD). Thus, at the diagnostic level comorbidity between SM and disruptive disorders, when present, appears to range anywhere between 6 and 10%, somewhat elevated in comparison to rates of these disorders found in the general child population (see Barkley, 2003; Lahey, Miller, Gordon, & Riley, 1999).

One study (Steinhausen & Juzi, 1996) found that 21% and 17% of an SM sample showed oppositionality and hyperactivity, respectively. These rates, however, were not based on formal diagnostic assessments but were determined by clinician judgment. For a subset for whom normed data were available (i.e., CBCL) externalizing problems were in the normal range and not significantly different from their same-age peers. Others (Vecchio & Kearney, 2005) also found low parent and teacher ratings of externalizing problems in children with SM; ratings that were not significantly different from children with anxiety disorders (without SM) and controls. This study provides evidence that SM might be better conceptualized as an anxiety-based condition (Black, 1996).

In fact, only one study (Kristensen, 2001) found that parent-reported externalizing and aggressive behaviors were higher for the SM group relative to controls. Analyses at the item level indicated that the two items “stubborn” and “screams a lot” best differentiated the groups. It is difficult to determine whether these behaviors represent a truly acting-out tendency in SM children or may be instead a reaction when asked to confront fearful situations. In fact, others (Beidel, Turner, & Morris, 1999) described similar reactions in children with social phobia, suggesting that these behaviors are not restricted to children with externalizing pathologies but may represent anxious or fearful responses to a situation. In another study, children with comorbid SM and SP were rated higher by parents on the CBCL (Achenbach, 1991a) Delinquency subscale than children with SP alone (Yeganeh et al., 2003), although scores were in the normal range for both groups (see also Ford et al., 1998). Yeganeh et al. (2006) also compared oppositionality (as measured by the Eyberg Child Behavior Inventory [ECBI; Eyberg & Pincus, 1999]) in children with SM, children with SP-only, and controls. Symptoms were in the normal range for all children regardless of diagnostic status. Contrary to findings of self-report measures, however, children with SM were diagnosed with comorbid ODD at a significantly higher rate than children with SP. As the authors suggest, these latter findings should be interpreted with caution as it may be that (a) the diagnostic criteria for ODD is less specific (and therefore more easily endorsed by parents during clinical interview) than the behaviors included in the ECBI or (b) that in fact a subset of children with SM do present with disruptive disorders.

However, other studies dispute presence of externalizing symptoms among children with SM. In comparison to controls, teachers rated children with SM significantly lower on ADHD and oppositional defiant disorder (ODD) subscales (Cunningham et al., 2004). Furthermore, there were no significant differences in parent-reported externalizing problems between SM children and controls. Another study (Bergman et al., 2002) did not find significant differences on teacher ratings of externalizing problems between children with SM and controls. Teacher ratings of children with generalized SM supported more generalized anxiety and *fewer* ODD symptoms in the classroom relative to controls (Cunningham et al., 2006). Children with specific SM did not differ from controls on teacher-rated generalized anxiety or ODD symptoms relative to controls.

Overall, there is mixed evidence for presence of externalizing or oppositional symptoms in children with SM. Some children with SM may present with mild oppositional symptoms (see Beidel & Turner, 2005, chapter 10). Furthermore, oppositional behaviors have also been described in children with anxiety disorders (e.g., SP; Beidel et al., 1999), making it difficult to disentangle the true role of oppositional behaviors sometimes seen in children with SM. Other studies have found that children with SM were *lower* in ADHD and ODD symptoms as rated by teachers (Cunningham et al., 2004, 2006), suggesting that children with SM may react disruptively in those settings where they are asked to speak (i.e., school), but do not present with a general overall pattern of defiant behavior.

4.3.3. SM and other types of comorbidity

SM appears to be associated with numerous conditions, the most prominent being communication and developmental disorders or delays, and elimination disorders (e.g., Black & Uhde, 1995; Kristensen, 2000). However, SM has also been associated with mild mental retardation and Asperger's Syndrome (Kristensen, 2000). Seventeen percent of children with SM met criteria for enuresis, and 7.0% for encopresis and tic disorder, respectively (Black & Uhde, 1995). Similarly, Arie et al. (2006) found a 16.7% comorbidity rate for enuresis. Thirty one percent of Kristensen's (2000) sample also had a comorbid elimination disorder, enuresis being the most common (29.6%), and 42.6% had a comorbid phonological disorder. As mentioned earlier, mixed receptive–expressive language disorder and expressive language disorder were less common (17.3% and 11.5% respectively), but still much higher than the rates observed among controls (1.0%). Fifty five percent of Kristensen's (2000) sample met criteria for at least 3 *DSM-IV* disorders. These findings suggest that comorbidity between SM and disorders characterized by delays in developmental milestones (e.g., communication skills, bladder control) is not uncommon.

4.4. Family and environmental vulnerabilities

Families of children with SM have been described as conflictual, isolated, and refraining from social contact (e.g., Remschmidt et al., 2001), with parent–child relationships described as overprotective, especially between mother and child. Additionally, among children with SM, 47% had parents with marriage problems and 84% had families where parenting style was rated as “deviant or insufficient” (p. 288). Difficult marital relationships also have been reported in other samples, including significantly higher rates of marital conflict in the presence of the child (Elizur & Perednik, 2003) when compared

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to controls. Vecchio and Kearney (2005) found that in comparison to controls, family environments of children with anxiety disorders and children with SM were rated by parents as significantly less socially active and involved in recreational activities. However, no significant differences were found between the two disordered groups.

In addition to family environment, other investigators have examined the potential role of singular traumatic events as precursors to development of SM. Black and Uhde (1995) reported that 13% had experienced significant trauma, but only in two cases had the trauma preceded the onset of the disorder. Kumpulainen et al. (1998) found that 47% of their SM sample had been exposed to a stressful event (e.g., death in the family, change of schools), but only in 16% was the event prior to the onset of the mutism. In Steinhausen and Juzi's (1996) study, 24% of children with SM had experienced a stressful life event prior to its onset. These studies suggest that for a small proportion of children with SM, a traumatic experience may be related to its onset. Furthermore, marital problems may also be present in some of these families.

4.4.1. SM, bilingualism, and immigration

Environmental factors such as immigration and the need to speak a second language may be implicated in the development of SM. One study (Elizur & Perednik, 2003) found that the prevalence rate of SM among immigrant children in Israel was 2.2%, compared to .5% among native children. Additionally, immigrant children with SM had significantly higher social anxiety scores (on a composite based on CBCL anxiety-related items) than native children with SM, but lower neurodevelopmental delay scores (also based on a CBCL item composite) and higher social competence scores. Considering that immigrant families in the study "usually did not speak Hebrew at home," and that 90% of the immigrant children were Israeli-born, this finding suggests that second language acquisition and another language being the primary tongue at home may be important variables in development of SM. Elizur and Perednik's (2003) findings point to a group of children with SM where immigrant backgrounds but also high social anxiety (even relative to appropriate comparison groups, such as native children with SM) were implicated in above-average prevalence rates of SM. What is not clear is how minority status, acculturation processes, and fewer opportunities for socialization as a result of high social anxiety – within the context of the second language acquisition process (Tabors, 1997) – interact to produce and/or maintain SM in immigrant children (see also Toppelberg, Tabors, Coggins, Lum, & Burger, 2005). Furthermore, significantly higher rates of neurodevelopmental delays and younger age of onset among native children with SM suggest to clinicians and researchers alike that SM may "look different" in native versus immigrant children. Immigration status – and the processes attached to it, such as acculturation, learning a second language, potential discrimination and peer ostracism, to name a few – may deserve unique attention in our efforts to understand SM. Developmental psychopathology may serve as a useful framework to try to understand the respective influences of these processes.

5. Assessment of SM

With its multi-faceted clinical presentation, it is clear that a multimodal, multi-trait assessment strategy is necessary (Dow et al., 1995). Obviously, lack of speech towards strangers precludes the child as the primary source of information; thus, a clinical interview with parents is essential. The Anxiety Disorders Interview Schedule for Children and Parents (ADIS-C/P: Silverman & Albano, 1996) is a semi-structured interview that assesses SM as well as many other disorders. It is important to gather precise information about symptom history, including onset and conditions under which mutism occurs (e.g., places, specific persons). Additionally, despite being completely silent in certain situations, children with SM may still communicate nonverbally and demonstrate prosocial communicative behaviors (e.g., nodding, smiling, giggling). Formal functional analysis under a variety of conditions may prove helpful in specifically determining maintaining variables (see, e.g., Schill, Kratochwill, & Gardner, 1996).

Because other conditions may also be characterized by lack of speech (e.g., autism, aphasia, mental retardation), a thorough developmental history is necessary. Potential pre-natal and perinatal complications suggestive of neurological insults may help explain language difficulties and delays. Ruling out other conditions that may better account for SM is an essential step in assessment.

Teachers are a valuable source of information in the assessment of SM (Cline & Baldwin, 2004). They can describe verbal and nonverbal inhibition in school settings. They may identify children with whom the child with SM speaks to so that peers may later assist during specific phases of treatment. Teachers may also describe situations in which the child is more or less likely to talk (e.g., reading activities versus free-play). Finally, teachers may have insight regarding previously used strategies that were successful in remediating the mutism.

The child's mutism complicates direct assessment. Despite this, it is still important to interview and observe the child directly to examine the nature and extent of impairment (Dow et al., 1995; Yeganeh et al., 2003). For example, does the child attempt to communicate nonverbally? The assessor may also be able to observe the child's temperament and the degree to which he or she is able to warm up to a stranger. This may inform treatment decisions.

A speech and language assessment is an important element for the diagnosis of SM (Dow et al., 1995; Manassis et al., 2007). For example, the PPVT-III (Dunn & Dunn, 1997) is a nonverbal test of receptive language successfully used with children with SM. The research by Manassis et al. (2007) has been important in this respect, highlighting potentially significant deficits in phonetic awareness, receptive language and grammar ability. Additional useful strategies (e.g., Dow et al., 1995) may involve having the parents audiotape the child speaking normally at home and use the tape to evaluate phonetics, length of utterances, tone, rhythm, and quality of response. This assessment strategy may allow identification of potential speech problems (Cleator & Hand, 2001) contributing to the presence of mute behavior in other settings.

6. Treatment of SM

Three excellent reviews of the scant literature on SM treatment are currently available (Anstendig, 1998; Cohan, Chavira, & Stein, 2006; Pionek Stone, Kratochwill, Sladezcek, & Serlin, 2002). As noted in these reviews, methodological weaknesses abound, and most investigations are in the form of case studies. A statistically stronger variation of this methodology – the single-case experimental design – has also been used to evaluate treatment outcome. Emerging from these reviews is the fact that there are no controlled trials of SM treatment, limiting the utility of the current treatment literature.

6.1. Behavioral interventions

Despite the above limitations, extant studies using single-case experimental designs suggest that behavioral interventions are efficacious (Cohan, Chavira, et al., 2006). These interventions often consist of a combination of behavioral strategies, such as contingency management, shaping, stimulus fading, systematic desensitization, and self-modeling (see, e.g., Watson & Kramer, 1992). Contingency management involves the administration of positive reinforcement contingent upon verbalization. Oftentimes reinforcement is provided for initial approximations to communicative behavior, like pointing or nodding, and continued until 'shaped' into the desired outcome (i.e., verbalization). Stimulus fading techniques have also been used and consist of sequentially increasing (i.e., fading in) the number of people present when the child is speaking. Initial sessions may consist of rewarding the child when speaking in the presence of only one stranger. Upon mastery, a second stranger may stand at the door while

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the child continues to be reinforced for speaking. The person gradually comes closer and closer to the child until seated at the same table with the child and the initial stranger. The process continues until the child is speaking in front of a larger number of people.

Combination of contingency management and shaping procedures has proven useful at increasing and maintaining speech during a 13 week follow-up (Amari, Slifer, Gerson, Schenck, & Kane, 1999), although it should be noted that contingent reinforcement continued during the follow-up period. Other investigators (Blum et al., 1998; Kehle, Madaus, Baratta, & Bray, 1998) combined contingency management, stimulus fading and self-modeling, which consists of audio/or video recording the child and editing the tape to show him/her speaking in settings where the child does not speak (e.g., the classroom). The tape serves as the "feared stimuli" in an exposure paradigm. During treatment sessions, the child, in the company of others, watches and listens to him/herself speaking. Across sessions, habituation to the fear of others hearing the child speak occurs thus allowing the child to speak normally in front of others. The three children involved in Kehle et al.'s (1998) study showed improvement in speech and at 7–9 month follow-ups, all of the children were speaking freely and appropriately with peers. Self-modeling appears promising in the treatment of SM, but its use in combination with a wide other behavioral strategies precludes conclusions regarding its unique efficacy.

6.2. Cognitive behavioral interventions

Cognitive behavioral interventions typically used for treating anxiety disorders appear to be successful for SM although the evidence is less clear (see Cohan, Chavira, et al., 2006). Considering that (a) cognitive interventions may overtax the cognitive abilities of very young children and (b) that SM is a 'young child' disorder, their efficacy certainly warrants further study. Multimodal interventions that combine behavioral, cognitive, family, psychodynamic and approaches also appear to have some promise, but which component(s) is responsible for change is unknown. As in the other modalities, the available evidence is based on case studies (e.g., Fung, Manassis, Kenny, & Fiskenbaum, 2002) and thus not elaborated here.

6.3. Pharmacological interventions

Pharmacological interventions have also been used with children with SM. Black and Uhde (1992), using a sample of 16 children with SM (ages 5–16), tested efficacy of fluoxetine in a double-blind placebo-controlled study. Significant effects were found for both the fluoxetine and the placebo-controlled groups on parent, teacher, and clinician ratings of SM, social anxiety, and anxiety. However, those treated with fluoxetine were perceived by parents as significantly more improved with regard to their mutism than those who received placebo. As the authors noted, however, the majority of participants in both groups remained highly symptomatic at the end of treatment. Similarly, Dummit, Klein, Tancer, Asche, and Martin (1997) tested the efficacy of fluoxetine in an open 9 week trial of 21 children with SM and concurrent anxiety disorders. All children met *DSM-III-R* criteria for either avoidant disorder or social phobia. Physical complaints (i.e., side effects) were reported for 43% of children, although these did not persist for more than a week. Behavioral disinhibition was reported in 4 cases (19%). At the end of the 9 weeks, 76% of children were considered 'improved' by the treating psychiatrists. Significant improvement was also seen across parent-, child-, and psychiatrist-rated measures assessing social behavior, social anxiety, and avoidance. The lack of appropriate controls clearly limits generalizability, but points to fluoxetine as a potentially useful medication in the treatment of SM. As the authors point out, the mechanisms by which fluoxetine decreases social fears remain unknown, however a recent study with children with social phobia (but not SM) indicates that it reduces arousal in social settings but does not increase social skill (Beidel et al., 2007). Given the diagnostic overlap, this same mechanism of action may apply to children with SM.

7. Summary and recommendations for future research

This review uses a developmental psychopathology framework to highlight the complex, multi-pathway nature of SM. As noted earlier, consensus is building that applying a unitary cause-and-effect model may not do justice to the complexity of this rare and socially impairing disorder. Although psychodynamic, behavioral, family systems, trauma-related, and genetic explanations have been proposed, accumulating findings suggest that deterministic views of SM may be inadequate. It seems likely that SM is the result of complex individual–environment transactions occurring at multiple levels over time (Cohan, Price et al., 2006; Sameroff, 1975).

As reviewed earlier, a high percentage of children with SM have parents who themselves suffered from SM or social phobia (Black & Uhde, 1995). Further, parents often describe their children in ways that are consistent with definitions of behavioral inhibition (Ford et al., 1998; Kristensen & Torgersen, 2002). Thus, there is evidence of a familial–genetic predisposition. At the same time, not all the children have parents who suffered from SM or social fears as illustrated by this review, thus the genetic pathway represents only one of the many pathways leading to SM (i.e., equifinality). Furthermore, disorders may exist in families but still be the result of environmental influences such as modeling. Further research is needed to disentangle the relative influence of genes versus environment.

Some parents of children with SM have other types of psychopathology, particularly a general preference for avoidance and being alone (Kristensen & Torgersen, 2001). While providing further support for a genetic predisposition, if considered from a developmental psychopathology perspective of reciprocal transactions the findings also suggest the possibility of dysfunctional patterns of interaction between parents and children that reinforce avoidant behavior. Parents who prefer limited social contact may model this avoidant style to their children. This interactional pattern may predispose some children to develop SM or to maintain or worsen the mutism once it develops. However, this pathway may be relevant only for the subset of children with SM who do not have comorbid communication disorders. When communication disorders are present, mutism may be more heavily influenced by their presence, encouraging lack of speech in certain settings.

A sizable portion of the SM literature (Beidel & Turner, 2005; Black, 1996; Black & Uhde, 1995; Dow et al., 1995; Garcia et al., 2004; Vecchio & Kearney, 2005) reports a significant association between SM and anxiety disorders in general and social phobia in particular. Some of these authors have even suggested that SM should be considered an extreme symptom of children with social phobia (Black & Uhde, 1995) rather than a stand-alone disorder. This theory is supported by findings of significant positive correlations between mutism severity and parent ratings of child anxiety, separation anxiety, and social anxiety. Furthermore, children with SM are consistently rated as 'shy', 'anxious', or 'timid' (e.g., Steinhausen & Juzi, 1996). Scores on rating scales further highlight the similarity between children with SM and children with anxiety disorders in general (Vecchio & Kearney, 2005) and social phobia in particular (Manassis et al., 2003). However, these assertions are challenged by findings of a trend for higher parent-reported social anxiety in children with social phobia relative to children with SM (Manassis et al., 2003), and by evidence that children with both SM and social phobia are often indistinguishable in terms of self-report measures of social anxiety, trait anxiety, and general fears (Yeganeh et al., 2003) from children with SM only. This would be unexpected if SM represents a severe variant of social phobia. Thus, although there is an association between these two disorders, clearly longitudinal investigations within a developmental psychopathology framework are needed to elucidate the nature of this relationship.

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One promising line of research is to examine contextual factors associated with development of SM relative to other anxiety disorders. This approach could shed light on the relative similarities or differences that may exist between SM and anxiety-based conditions. Some investigations have taken a related approach (Yeganeh et al., 2003, 2006) by comparing children with SM and children with social phobia. Unfortunately, these studies have primarily looked at differences in parental reports of child characteristics. Although undoubtedly important, the field would benefit greatly from broadening the scope of analysis. Examining the pattern of contextual variables (both proximal and distal) could shed specific light into the nature of the comorbidity of SM and social phobia. For example, parental overinvolvement and overcontrol are frequently present in families of children with anxiety disorders (Bayer, Sanson, & Hemphill, 2006; Dadds & Barrett, 1996). Furthermore, overcontrol and overinvolvement are not restricted to interactions with the child with anxiety, but occur with non-disordered siblings as well (Hudson & Rapee, 2002). Similarly, parents of children with anxiety disorders often amplify their children's avoidant responses (Barrett, Rapee, Dadds, & Ryan, 1996; Dadds, Barrett, & Rapee, 1996), highlighting the role of family processes in the development of anxiety. Whether and how these contextual and family processes also operate in children with SM remain unknown. Other environmental factors (i.e., immigration, second language acquisition, and primary language spoken at home) also merit further study. Developmental psychopathology offers a framework from which to ask and study these important questions.

Also noticeable in this review is rate of comorbidity between SM and communication disorders (Kristensen, 2002; Steinhausen & Juzi, 1996; Steinhausen et al., 2006), often thought of as indexes of neurodevelopmental deficit. High rates of comorbidity with communication disorders (e.g., 50.0%; Kristensen, 2000) and the markedly high proportion of these disorders in some SM groups relative to controls (Kristensen, 2000) suggest yet another etiological pathway. The lack of longitudinal data, however, precludes drawing firm conclusions regarding the temporal emergence of these disorders. Furthermore, the significant lag between onset and diagnosis of SM exists further complicates etiological explanations. Even within the group of communication disorders comorbid with SM, there is appreciable variability, with some children having problems with phonemic expression while others with expressive and speech difficulties (see also Kristensen & Oerbeck, 2006).

One tenable hypothesis under a developmental psychopathology framework is that some children with early significant speech and language delays may not notice their speech and language deficits relative to peers until they enter school. In this new environment, these children are consistently confronted with peers whose language skills are age-appropriate. At the time of this major developmental transition (i.e., school entry) their language deficits may interact with an anxious predisposition that culminates in the development of SM. Similarly, they may get teased about unusual or incorrect pronunciation thereby setting up a pattern of avoidance in order to avoid ridicule. These hypotheses would be consistent with the time when SM is often diagnosed. Furthermore, based on findings of psychopathology in parents of children with SM, these factors may further interact with a learned familial interactional style in which avoidance of threat has been repeatedly reinforced. Clearly, these ideas await testing, but it is important that future research in SM consider a multiple-pathways approach to the conceptualization of this disorder and include measurement of key variables that may explain the relative contribution of each pathway.

Further clarification of the role of oppositionality in SM is needed. Several investigations have indeed found small percentage of children with comorbid ODD (e.g., Black & Uhde, 1995; Manassis et al., 2007) and ADHD (e.g., Arie et al., 2006). Other studies report higher frequencies of disruptive behaviors (Steinhausen & Juzi, 1996) when assessments are made via retrospective review of charts. Furthermore, these typical disruptive behaviors (e.g., stubborn, screams) also exist in children with anxiety disorders (Beidel et al., 1999). A developmental psychopathology perspective, contrary to an 'either/or' view, may be particularly helpful in this respect. Contextual pressures (e.g., school demands), perceptions of the child as oppositional due to a lack of speech, and true comorbidity between oppositional and defiant behavior and intense anxiety are all rival hypotheses that may be tested. The treatment implications of this research are tremendous, as assignation of particular diagnoses will dictate very different treatment strategies (e.g., PMT for oppositionality; CBT for anxiety).

Finally, the SM literature needs a consistent definition and systematic assessment of SM that are applied across settings and laboratories, as well as multi-center studies that will lead to larger samples of children with SM. Assessment has often consisted of retrospective chart reviews or retrospective recall by children or parents. This approach has dramatically changed in recent years and efforts in this direction must continue. Similarly, the majority of the studies on SM consists of very small sample sizes. At the same time, single-case experimental designs may play an important role in the study of SM. While collaboration between investigators may yield larger samples, the logistics of such endeavors are often complex. Single-case designs may be key in adding methodological rigor to investigations of SM. Furthermore, studies on the etiology of SM are in dire need, as much of the literature consists of cross-sectional, descriptive studies. Two lines of research seem promising at this stage. First, the prospective study of behaviorally inhibited children, who may be at risk for developing SM. Concomitantly measuring familial as well as other contextual variables may shed light on the relative contribution of these variables and potential interactions with genetically based predispositions such as behavioral inhibition. Additionally, conceptualizing SM as an avoidant *behavior* – rather than a unique, distinct disorder – that emerges over time in the context of complex interactions between anxiety predispositions, familial patterns of inadequate reinforcement (e.g., avoidance), neurodevelopmental deficits, and other contextual pressures (e.g., second language acquisition) over time may provide an organizing framework to better understand its phenomenology. The field of developmental psychopathology provides ample opportunities for investigational frameworks of this kind.

References

Achenbach, T. M. (1979). Psychopathology of childhood: Research problems and issue. *Annual Progress in Child Psychiatry and Child Development*, 421–449.

Achenbach, T. M. (1991). *Manual for the child behavior checklist/4–18 and 1991 profile.* Burlington: University of Vermont, Department of Psychiatry.

Achenbach, T. M. (1991). *Manual for the teacher's report form and 1991 profile.* Burlington: University of Vermont, Department of Psychiatry.

Amari, A., Slifer, K. J., Gerson, A. C., Schenck, E., & Kane, A. (1999). Treating selective mutism in a paediatric rehabilitation patient by altering environmental reinforcement contingencies. *Pediatric Rehabilitation*, *3*, 59–64.

American Psychiatric Association (1987). *Diagnostic and statistical manual of mental disorders*, 3rd ed. Washington, DC: Author revised.

American Psychiatric Association (1994). *Diagnostic and statistical manual of mental disorders*, 4th ed. Washington, DC: Author.

American Psychiatric Association (2000). *Diagnostic and statistical manual of mental disorders*, 4th ed. Washington, DC: Author text revision.

Andersson, C. B., & Thomsen, P. H. (1998). Electively mute children: An analysis of 37 Danish cases. *Nordic Journal of Psychiatry*, *52*, 231–238.

Anstendig, K. (1998). Selective mutism: A review of the treatment literature by modality from 1980–1996. *Psychotherapy: Theory, research, practice, and training*, *35*, 381–391.

Anstendig, K. D. (1999). Is selective mutism an anxiety disorder? Rethinking its DSM-IV classification. *Journal of Anxiety Disorders*, *13*, 417–434.

Arie, M., Henkin, Y., Lamy, D., Tetin-Schneider, S., Apter, A., Sadeh, A., et al. (2006). Reduced auditory processing capacity during vocalization in children with selective mutism. *Biological psychiatry*, *61*, 419–421.

Barkley, R. A. (2003). Attention deficit/hyperactivity disorder. In E. J. Mash, & R. A. Barkley (Eds.), *Child psychopathology* (pp. 75–143)., 2nd ed. New York: Guilford Press.

Bar-Haim, Y., Henkin, Y., Ari-Even-Roth, D., Tetin-Schneider, S., Hildesheimer, M., & Muchnik, C. (2004). Reduced auditory efferent activity in childhood selective mutism. *Biological psychiatry*, *55*, 1061–1068.

Barrett, P. M., Rapee, R. M., Dadds, M. R., & Ryan, S. M. (1996). Family enhancement of cognitive style in anxious and aggressive children. *Journal of Abnormal Child Psychology*, *24*, 187–203.

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Bayer, J. K., Sanson, A. V., & Hemphill, S. A. (2006). Parental influences on early childhood internalizing difficulties. *Journal of Applied Developmental Psychology*, *27*, 542–559.

Beidel, D. C., & Turner, S. M. (2005). *Childhood anxiety disorders: A guide to research and treatment*. New York, NY: Taylor & Francis Group.

Beidel, D. C., Turner, S. M., & Morris, T. L. (1999). Psychopathology of childhood social phobia. *Journal of the American Academy of Child & Adolescent Psychiatry*, *38*, 643–650.

Beidel, D. C., Turner, S. M., Sallee, F. R., Ammerman, R. T., Crosbi, L. A., & Pathak, S. (2007). SET-C versus fluoxetine in the treatment of childhood social phobia. *Journal of the American Academy of Child & Adolescent Psychiatry*, *46*, 1622–1632.

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.

Black, B. (1996). Social anxiety and selective mutism. *American Psychiatric Press Review of Psychiatry*, *15*, 469–495.

Black, B., & Uhde, T. W. (1992). Elective mutism as a variant of social phobia. *Journal of the American Academy of Child & Adolescent Psychiatry*, *31*, 1090–1094.

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.

Blum, N. J., Kell, R. S., Starr, H. L., Lender, W. L., Bradley-Klug, K. L., Osbourne, M. L., et al. (1998). Case study: Audio feedforward treatment of selective mutism. *Journal of the American Academy of Child and Adolescent Psychiatry*, *37*, 40–43.

Bronfenbrenner, U. (1979). Contexts of child rearing: Problems and prospects. *American Psychologist*, *34*, 844–850.

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

Chavira, D. A., Stein, M. B., Bailey, K., & Stein, M. T. (2004). Child anxiety in primary care: Prevalent but untreated. *Depression and anxiety*, *20*, 155–164.

Cicchetti, D. (1984). The emergence of developmental psychopathology. *Child Development*, *55*, 1–7.

Cicchetti, D., & Toth, S. L. (1997). Transactional ecological systems in developmental psychopathology. In S. S. Luthar, J. A. Burack, D. Cicchetti, & J. R. Weisz (Eds.), *Developmental psychopathology: Perspectives on adjustment, risk, and disorder* (pp. 317–349). New York, NY: Cambridge University Press.

Cleator, H., & Hand, L. (2001). Selective mutism: How a successful speech and language assessment really is possible. *International Journal of Language and Communication Disorders*, *36*, 126–131.

Cline, T., & Baldwin, S. (2004). *Selective mutism in children*, 2nd ed. London, England: Whurr Publishers.

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

Cohan, S. L., Price, J. M., & Stein, M. B. (2006). Suffering in silence: Why a developmental psychopathology perspective on selective mutism is needed. *Journal of Developmental & Behavioral Pediatrics*, *27*, 341–355.

Cummings, M. E., Davies, P. T., & Campbell, S. B. (2000). *Developmental psychopathology and family process: Theory, research, and clinical implications*. New York, NY: Guilford Press.

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 and Psychiatry*, *45*, 1363–1372.

Dadds, M. R., & Barrett, P. M. (1996). Family processes in child and adolescent anxiety and depression. *Behaviour Change*, *13*, 231–239.

Dadds, M. R., Barrett, P. M., & Rapee, R. M. (1996). Family process and child anxiety and aggression: An observational analysis. *Journal of Abnormal Child Psychology*, *24*, 715–734.

Dow, S. P., Sonies, B. C., Scheib, D., Moss, S. E., & Leonard, H. L. (1995). Practical guidelines for the assessment and treatment of selective mutism. *Journal of the American Academy of Child and Adolescent Psychiatry*, *34*, 836–846.

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

Dummit, E. S., Klein, R. G., Tancer, N. K., Asche, B., & Martin, J. (1997). Fluoxetine treatment of children with selective mutism: An open trial. *Journal of the American Academy of Child & Adolescent Psychiatry*, *35*, 615–621.

Dunn, L. M., & Dunn, L. M. (1997). *Peabody picture vocabulary test-III*. Circle Pines, MN: American Guidance Services.

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

Eyberg, S., & Pincus, D. (1999). *Eyberg child behavior inventory and Sutter–Eyberg student behavior inventory — Revised professional manual*. Odessa, Fl: Psychological Assessment Resources, Inc.

First, M. B., Gibbon, M., Spitzer, R. L., Williams, J. B. W., & Benjamin, L. S. (1997). *Structured clinical interview for the DSM-IV axis II personality disorders (SCID-II)*. Washington, DC: American Psychiatric Press.

First, M. B., Spitzer, R. L., Gibbon, M., & Williams, J. B. W. (1997). *User's guide for the structured clinical interview for the DSM-IV axis I disorders — Clinician version (SCID-CV)*. Washington, DC: American Psychiatric Press.

Ford, M. A., Sladeczek, I. E., Carlson, J., & Kratochwill, T. R. (1998). Selective mutism: Phenomenological characteristics. *School Psychology Quarterly*, *13*, 192–227.

Fung, D. S., Manassis, K., Kenny, A., & Fiskenbaum, L. (2002). Web-based CBT for selective mutism. *Journal of the American Academy of Child and Adolescent Psychiatry*, *41*, 112–113.

Garcia, A. M., Freeman, J. B., Francis, G., Miller, L. M., & Leonard, H. L. (2004). Selective mutism. In T. H. Ollendick, & J. S. March (Eds.), *Phobic and anxiety disorders in children and adolescents: A clinician's guide to effective psychosocial and pharmacological interventions* (pp. 433–455). New York, NY: Oxford University Press.

Giddan, J. J., Ross, G. J., Sechler, L. L., & Becker, B. R. (1997). Selective mutism in elementary school: Multidisciplinary interventions. *Language, Speech, and Hearing Services in Schools*, *28*, 127–133.

Hudson, J. L., & Rapee, R. M. (2002). Parent–child interactions in clinically anxious children and their siblings. *Journal of Clinical Child and Adolescent Psychology*, *31*, 548–555.

Kehle, T. J., Madaus, M. R., Baratta, V. S., & Bray, M. A. (1998). Augmented self-modeling as a treatment for children with selective mutism. *Journal of School Psychology*, *36*, 247–260.

Kopp, S., & Gillberg, C. (1997). Selective mutism: A population-based study: A research note. *Journal of Child Psychology and Psychiatry*, *38*, 257–262.

Kratochwill, T. R. (1981). *Selective mutism: Implications for research and treament*. Hillsdale, NJ: Erlbaum.

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.

Kristensen, H. (2001). Multiple informants report of emotional and behavioural problems in a nation-wide sample of selective mute children and controls. *European Child & Adolescent Psychiatry*, *10*, 135–142.

Kristensen, H. (2002). Non-specific markers of neurodevelopmental disorder/delay in selective mutism: A case–control study. *European Child & Adolescent Psychiatry*, *11*, 71–78.

Kristensen, H., & Oerbeck, B. (2006). Is selective mutism associated with deficits in memory span and visual memory?: An exploratory case–control study. *Depression and Anxiety*, *23*, 71–76.

Kristensen, H., & Torgersen, S. (2001). MCMI-II personality traits and symptom traits in parents of children with selective mutism: A case–control study. *Journal of Abnormal Psychology*, *110*, 648–652.

Kristensen, H., & Torgersen, S. (2002). A case–control study of EAS child and parental temperaments in selectively mute children with and without a co-morbid communication disorder. *Nordic Journal of Psychiatry*, *56*, 347–353.

Krysanski, V. L. (2003). A brief review of the selective mutism literature. *The Journal of Psychology*, *137*, 29–40.

Kumpulainen, K., Räsänen, E., Raaska, H., & Somppi, V. (1998). Selective mutism among second-graders in elementary school. *European Child & Adolescent Psychiatry*, *7*, 24–29.

Kuperminc, G. P., & Brookmeyer, K. A. (2006). Developmental psychopathology. In R. T. Ammerman (Ed.), *Comprehensive handbook of personality and psychopathology, vol. 3*. (pp. 100–113)Hoboken, NJ: John Wiley & Sons, Inc.

Kussmaul, A. (1877). *Die Stoerungen der Sprache (1st ed.) [Disturbances in linguistic function]*. Basel, Switzerland: Benno Schwabe.

Lahey, B. B., Miller, T. L., Gordon, R. A., & Riley, A. W. (1999). Developmental epidemiology of the disruptive behavior disorders. In H. C. Quay, & A. E. Hogan (Eds.), *Handbook of disruptive behavior disorders* (pp. 23–48). New York: Plenum Press.

Leonard, H. L., & Topol, D. A. (1993). Elective mutism. *Child and Adolescent Psychiatric Clinics of North America*, *2*, 695–707.

Lindamood, C., & Lindamood, P. (1971). *Lindamood auditory conceptualization test manual*. R. DLM Teaching Resources: Riverside Publishing.

Manassis, K., Fung, D., Tannock, R., Sloman, L., Fiksenbaum, L., & McInnes, A. (2003). Characterizing selective mutism: Is it more than social anxiety? *Depression and Anxiety*, *18*, 153–161.

Manassis, K., Tannock, R., Garland, E. J., Minde, K., McInnes, A., & Clark, S. (2007). The sounds of silence: Language, cognition and anxiety in selective mutism. *Journal of the American Academy of Child & Adolescent Psychiatry*, *46*, 1187–1195.

McInnes, A., Fung, D., Manassis, K., Fiksenbaum, L., & Tannock, R. (2004). Narrative skills in children with selective mutism: An exploratory study. *American Journal of Speech-Language Pathology*, *13*, 304–315.

Meyers, S. (1984). Elective mutism in children: A family systems approach. *The American Journal of Family Therapy*, *12*, 39–45.

Pionek Stone, B., Kratochwill, T. R., Sladezcek, I., & Serlin, R. C. (2002). Treatment of selective mutism: A best-evidence synthesis. *School Psychology Quarterly*, *17*, 168–190.

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

Sameroff, A. J. (1975). Transactional models in early social relations. *Human Development*, *18*, 65–79.

Schill, M. T., Kratochwill, T. R., & Gardner, W. I. (1996). An assessment protocol for selective mutism: Analogue assessment using parents as facilitators. *Journal of School Psychology*, *34*, 1–21.

Schwartz, R. H., Freedy, A. S., & Sheridan, M. J. (2006). Selective mutism: Are primary care physicians missing the silence? *Clinical Pediatrics*, *45*, 43–48.

Semel, E., Wiig, E. H., & Secord, W. A. (1995). *Clinical evaluation of language fundamentals, third edition: Examiner's manual*. San Antonio: The Psychological Corporation.

Sharp, W. G., Sherman, C., & Gross, A. M. (2007). Selective mutism and anxiety: A review of the current conceptualization of the disorder. *Journal of anxiety disorders*, *21*, 568–579.

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Silverman, W. K., & Albano, A. M. (1996). *The anxiety disorders interview schedule for children for DSM-IV: (Child and parent versions)*. San Antonio, TX: Psychological Corporation.

Steinhausen, H., & Adamek, R. (1997). The family history of children with elective mutism: A research report. *European Child and Adolescent Psychiatry*, *6*, 107–111.

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

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

Tabors, P. O. (1997). *One child, two languages: A guide for preschool educators of children learning English as a second language*. Baltimore, MD: Brookes.

Toppelberg, C. O., Tabors, P., Coggins, A., Lum, K., & Burger, C. (2005). Differential diagnosis of selective mutism in bilingual children. *Journal of the American Academy of Child and Adolescent Psychiatry*, *44*, 592–595.

Tramer, M. (1934). Elektiver mutismus bei kindern./selective mutism of children. *Zeitschrift für Kinderpsychiatrie*, *1*, 30–35.

Vecchio, J. L., & Kearney, C. A. (2005). Selective mutism in children: Comparison to youths with and without anxiety disorders. *Journal of Psychopathology and Behavioral Assessment*, *27*, 31–37.

Watson, T. S., & Kramer, J. J. (1992). Multimethod behavioral treatment of long-term selective mutism. *Psychology in the Schools*, *29*, 359–366.

World Health Organization (1992). *The ICD-10 classification of mental and behavioural disorders: Clinical descriptions and diagnostic guidelines*. Geneva: World Health Organization.

Yanof, J. A. (1996). Language, communication, and transference in child analysis: I. Selective mutism: the medium is the message; II. Is child analysis really analysis? *Journal of the American Psychoanalytic Association*, *44*, 79–116.

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*, 1069–1075.

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