

Selective Mutism and Social Anxiety Disorder: All in the Family?

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ABSTRACT

Objective: To examine the history of lifetime psychiatric disorders in the parents of children with selective mutism (SM) compared to parents of children in a control group. **Method:** Seventy parent dyads ($n = 140$) of children with lifetime SM and 31 parent dyads ($n = 62$) of children without SM were interviewed with the Structured Clinical Interview for *DSM-IV* (IV and II) anxiety disorders, mood disorders, avoidant personality disorder, and schizoid personality disorder modules via telephone. Interviewers were blind to proband status. The NEO Personality Inventory was also administered. **Results:** Lifetime generalized social phobia was present in 37.0% of SM parents compared to 14.1% of control parents ($\chi^2 = 10.98$; $p < .001$; odds ratio 3.6, 95% confidence interval 1.6–7.9). Avoidant personality disorder was present in 17.5% of the SM parents compared to 4.7% of control parents ($\chi^2 = 6.18$; $p < .05$; odds ratio 4.3, 95% confidence interval 1.3–14.9). The proportion of parents with other psychiatric disorders was not different between groups. SM parents had higher neuroticism and lower openness scores on the NEO Personality Inventory than control parents. **Conclusions:** These results support earlier uncontrolled findings of a familial relationship between generalized social phobia and SM. *J. Am. Acad. Child Adolesc. Psychiatry*, 2007;46(11):1464–1472. **Key Words:** selective mutism, child anxiety, social anxiety, genetics.

Selective mutism (SM) is defined as a consistent failure to speak in specific social situations, in which children are required to speak (e.g., school), despite speaking in other situations (American Psychiatric Association, 1994). According to *DSM-IV* criteria, SM is associated with significant impairment, has a duration of at least 1 month and is not due to a lack of knowledge or comfort with speaking a language or accounted for by the presence of a communication, psychotic, or pervasive

developmental disorder. Extant data suggest that SM usually begins in early childhood, often during the preschool years when a child is first required to speak in formal settings such as school and day care. Little is known about the naturalistic course of SM. The few studies that do exist suggest that even though mutism may frequently remit over time (Steinhausen et al., 2006), rates of “talking” behaviors remain lower than average (Bergman et al., 2002) and residual psychopathology such as social phobia and other anxiety disorders often persist (Steinhausen et al., 2006).

The etiology of SM is not well understood. Previous explanations suggest that overcontrolling or hostile parenting, intrapsychic conflicts, or past trauma contribute to the onset of SM; however, limited data exist to support these positions (Anstendig, 1999; Black and Uhde, 1995). Other studies suggest that child oppositionality may contribute to the refusal to speak, yet data are mixed in this regard (Cunningham et al., 2006; Yeganeh et al., 2003). To date, most research supports that SM is related to social phobia (SP) and that they share common etiologies.

Cross-sectional comorbidity rates between SM and SP range from 97% to 100% (Black and Uhde, 1995;

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Dummit et al., 1997), and characteristics such as shy, anxious, withdrawn, and serious are used to describe both those with SM and social anxiety alike (Kumpulainen et al., 1998; Steinhausen and Juzi, 1996). Findings from family history studies further support this relationship. In a study of personality characteristics of parents of 50 SM children and control parents (Kristensen and Torgersen, 2001) assessed with the Millon Clinical Multiaxial Inventory II (Millon, 1987), 39% of mothers and 32% of fathers of SM children were classified as shy/socially anxious versus 4% of mothers and 1% of fathers of controls. The avoidant and schizoid scales also predicted membership in the SM index group for mothers and fathers, respectively. Using a different assessment of temperament, parents of children with SM ($n = 38$) reported greater taciturnity in first-, second-, and third-degree relatives when compared to parents of control children ($n = 31$) (Steinhausen and Adamek, 1997). In the only family study that included a diagnostic assessment ($N = 30$ families with a child diagnosed with SM), 37% of the first-degree relatives had SM and 70% had SP (Black and Uhde, 1995). In that study, information was initially gathered by checklist format and then followed up by unstructured clinical interviews; a control group was not included.

Purpose of the Study

The present study builds on past findings in its assessment of personality traits and psychiatric disorders among parents of children with and without an SM diagnosis. In this study, we address some of the methodological limitations of previous research in this area by including a control group to provide appropriate comparisons; well-established semistructured diagnostic interviews rather than informal assessments; and multiple clinicians, blind to proband status to minimize diagnostic bias. In addition, this study also differs from previous research in its inclusion of SP subtypes. The generalized subtype of has been defined as the fear of most social situations (American Psychiatric Association, 1994), whereas the nongeneralized type refers to the fear of circumscribed situations, usually performance in nature. Data support differences in severity, comorbidity, and etiologies across the subtypes (Chavira et al., 2004; Wittchen et al., 1998), with some data suggesting that the generalized type may have a more temperamental and perhaps heritable basis

(Stein et al., 1998a,b; Stemberger et al., 1995). Given these differences, we believed it necessary to delineate SP subtypes in this study.

Hypotheses

On the basis of previous findings suggesting a relationship between social anxiety and SM, higher rates of SP among parents of children with SM were expected. In particular, higher rates of the generalized type of SP were hypothesized. On a self-report personality assessment, higher scores on Neuroticism and lower scores on the Extraversion subscales were expected among the parents of children with SM when compared to controls.

METHOD

Design and Procedures

This study is part of a larger project that includes the collection of DNA samples from families of children with SM. A nationwide sample was recruited by means of two sources: a Web site sponsored by a nonprofit organization for children with SM (the Selective Mutism Group-Child Anxiety Network), and parent-oriented conferences organized by this same nonprofit group. The Selective Mutism Group Web site receives approximately 500,000 hits per month from parents, professionals, and educators worldwide. Parents who were interested in participating in the project completed a consent-to-contact form and were thereafter sent study consents/child assents and contacted by telephone or consented in person if recruited from the conferences. Control families were recruited through community advertisements posted in San Diego County and a university Web site advertising participation in research studies.

Families who returned their consent forms were screened over the telephone with the SM module of the Anxiety Disorders Interview Schedule for Children-Parent Report (ADIS-P/C; Silverman and Albano, 1996) and the Selective Mutism Questionnaire (SMQ; Bergman et al., 2001). Families assigned to the SM group had to endorse symptoms consistent with a lifetime diagnosis of SM for the child and at least a moderate amount of impairment in one of the domains assessed by the SMQ. Screening questions to exclude children with psychosis, developmental, or communication diagnosis were also taken from the ADIS-P/C and augmented by supplemental questions to adequately rule out these conditions. The first and last authors (D.A.C. and M.B.S.) discussed all of the cases that were questionable or excluded from the study. In those instances in which diagnoses remained questionable, the second author (E.S.B.) was asked to review a videotape of the child speaking at home to rule out the presence of a pervasive developmental disorder or significant communication disorder; this was necessary for five cases and resulted in the exclusion of two cases. Control families were eligible if their child did not screen positive for lifetime SM or the above-mentioned exclusionary diagnoses.

Appointments for telephone interviews with the parents were scheduled by a study coordinator who was not blind to proband diagnostic status. All of the interviewers were blind to proband status and as part of the introduction to the interview, it was requested that parents not reveal whether their child had previously

been diagnosed with SM. Self-report questionnaires were returned by mail. All of the study procedures were approved by the institutional review board at our institution.

Participants

The participation of both biological parents and having a child between the ages of 3 and 11 were initial requirements for this study. Telephone screens were conducted with 132 parents of SM children and 43 parents of control children. Of the potential SM participants, 27 SM families lost interest after completing the screen, and another 26 were excluded because the child was outside the age range ($n = 2$), had significant speech problems ($n = 6$), had symptoms consistent with a pervasive developmental disorder diagnosis ($n = 5$), had subthreshold SM symptoms ($n = 6$), and only one parent was willing to participate ($n = 7$). In the control group, six families lost interest after completing the screen and three others were excluded for the following reasons: significant pervasive developmental disorder symptoms ($n = 1$), an SM diagnosis ($n = 1$), and only one parent was willing to participate ($n = 1$). Complete data were collected for 70 mother-father dyads ($n = 140$) with a proband child who had an SM diagnosis (either current or past) and 31 control mother-father dyads ($n = 62$) with a proband child for whom an SM diagnosis was never present. The majority of children in the SM group had a current diagnosis (70%).

Measures

Screening. The ADIS-C/P (Silverman and Albano, 1996) is a semistructured diagnostic interview designed to assess *DSM-IV* childhood anxiety disorders as well as depressive and behavioral disorders. Published κ coefficients for the ADIS-C/P disorders are .88 for separation anxiety, .86 for SP, .65 for specific phobia, .72 for generalized anxiety disorder, and 1.00 for attention-deficit/hyperactivity disorder (Silverman et al., 2001). Supplemental questions were added to the interview to assess for the presence of lifetime diagnoses.

The SMQ (Bergman et al., 2001) is a 17-item parent report measure of child SM behaviors and SM-related impairment. The SMQ queries speaking behaviors in three domains: school, home/family, and public settings. A 4-point Likert scale is used, and total scores range from 17 to 68. Higher scores represent greater SM severity. Data from 576 parents have revealed a meaningful factor structure with adequate psychometric properties (Bergman et al., 2001).

Family History. The Structured Clinical Interview for *DSM-IV* Disorders (SCID-IV) (First et al., 1997a) was used to assess Axis I disorders. For the present study, we included the depressive, manic, psychotic, and anxiety disorders modules. Questions were phrased in terms of "ever in your life" and therefore prevalence rates are lifetime. The SCID is a widely used semistructured diagnostic interview and its reliability and validity have usually been in the fair to good range (First and Gibbon, 2004; Williams et al., 1992). A generalized SP (GSP) diagnosis was applied if significant fear and avoidance were endorsed for at least four social situations and at least two situations were interactional (Stein et al., 1998a,b). Individuals who had fewer than four fears or only performance-related fears were assigned to the non-GSP category. The any SP group included individuals with either the generalized or non-generalized subtype.

The SCID for Axis II Personality Disorders (SCID-II; First et al., 1997b) is a semistructured interview used to obtain diagnoses for the

Axis II disorders of the *DSM-IV*. In this study, only the avoidant personality disorder and schizoid personality disorder modules were administered. Fair to good median interrater kappas have been found for the more commonly occurring Axis II disorders (Renneberg et al., 1992; Zanarini et al., 2000), and test-retest kappas have also been found to be in the fair to good range (Zanarini et al., 2000).

The NEO Personality Inventory-Revised (Costa and McCrae, 1992) is a widely used measure of personality with well-established psychometric properties (Costa et al., 1991). It includes 240 items that assess personality domains that are consistent with a five-factor model of personality: neuroticism, extraversion, openness, agreeableness, and conscientiousness. Each of the five domains also has six lower level facets. According to sex-specific norms developed by Costa and McCrae (1992), *T* scores ranging from 45 to 54 are average, scores between 55 and 65 are high, and scores above 65 are considered very high.

Diagnostic Reliability

Interviewers listened to audiotapes of the other raters and provided diagnostic summaries for 15% of the SCID interviews. Kappa statistics were .65 for GSP, 0.50 for non-GSP, and .75 for any SP. Kappas were .60 for major depression, .65 for dysthymia, .60 for generalized anxiety disorder, .88 for specific phobia, .65 for a history of SM, and 1.00 for childhood separation anxiety disorder, post-traumatic stress disorder, and panic with agoraphobia.

Kappas were also calculated for 25% of the ADIS-P/C interviews. Kappas were 1.0 for major depression and attention-deficit/hyperactivity disorder, .80 for any SP, .75 for enuresis, .67 for separation anxiety disorder, .60 for generalized anxiety disorder, and .47 for obsessive compulsive disorder. Kappas were not computed for dysthymia, oppositional defiant disorder, and conduct disorder because of the low frequency of these diagnoses in the reliability sample.

All of our reliabilities are based on agreement between diagnoses derived from raters listening to audiotapes of interviews (done by other interviewers) and consensus diagnoses given after interviews were discussed at meetings. Using the consensus diagnoses as the comparison rather than the original diagnoses given by the interviewers may have reduced kappa estimates but most were still in the acceptable to good range.

Training and Consensus Meetings

The interviewers in this study included advanced doctoral students, postdoctoral fellows, and psychologists. Interviewers received approximately 8 weeks of training on the diagnostic instruments and were required to meet gold standard criteria (at least 80% agreement on three mock cases) before conducting actual interviews. Weekly consensus meetings, at which each and every case was reviewed in detail, were held for the purposes of determining final diagnoses. *DSM-IV* guidelines, clinical judgment, review of audiotapes, and ultimately majority votes (when necessary) were used to establish final diagnoses.

Data Analysis

Chi-square analyses and one-way analyses of variance were conducted to compare the demographic characteristics across groups. Omnibus χ^2 analyses were used to compare the distribution of *DSM-IV* disorders (lifetime) across SM and control groups.

Analyses were also run separately for mothers and fathers and logistic regressions were used to test for an interaction between proband status (SM or control parents) and parent sex for each psychiatric disorder derived from the SCID. Multivariate analyses of variance (MANOVA) were conducted with personality dimensions and facets as the dependent variables and proband status (SM or control) as the independent variable. Analyses including interaction terms for parent sex and proband status were also conducted.

RESULTS

Parent and Child Demographics

The geographic distribution of the SM families was as follows: 24% were from the west, 26% were from the midwest, 20% were from the south, and 40% were from the northeast. The majority of controls were from the west (84%). As shown in Table 1, there were no significant differences in age of parent, education, or ethnicity across the SM and control groups. Among children, there were also no sex or age differences across groups: There were 9 boys and 22 girls in the control sample and 26 boys and 44 girls in the SM sample ($\chi^2_{1,100} = 0.51$; $p = .48$). The mean age of children with SM was 6.37 years old (SD 2.51) and 7.12 years old (SD 2.99) for controls ($F_{1,96} = 1.73$; $p = .19$).

Child Psychiatric Disorders and SMQ Severity

The distribution of lifetime Axis I psychiatric disorders is presented in Table 2. Children with lifetime SM (i.e., had a current or past diagnosis) had significantly

higher lifetime rates of SP, specifically the generalized type, than control children. Rates of separation anxiety disorder were also significantly higher among the children with SM when compared to controls. The same findings emerged when only children with current SM diagnoses were included. On the SMQ, which assesses SM severity, the child SM group had a mean total score of 53.55 (SD 6.65) and the control group had a mean score of 21.15 (SD 3.34; $F_{1,91} = 578.83$; $p < .001$). For indirect comparison purposes, the score of the original psychometric sample of children with SM was 50.47 (Bergman et al., 2001). In a series of univariate logistic regressions, child SM severity was found to be associated with an increased odds of parent SP (odds ratio [OR] 1.06, 95% confidence interval [CI] 1.02–1.09, $\chi^2 = 12.22$; $p < .001$) for each 1-point increase in total SMQ score. There was no significant relationship between childhood SM severity and any other parent psychiatric disorder as measured by the SCID (data not shown).

Parent Psychiatric Disorders

As shown in Table 3, parents of SM children had significantly higher lifetime rates of GSP (OR 3.6, 95% CI 1.6–7.9) and avoidant personality disorder (AVPD; OR 4.3, 95% CI 1.3–14.9) than the parents of the control children. Rates of non-GSP (which consisted

TABLE 1
Parent Demographics for SM and Controls

	SM Fathers ($n = 70$)	Control Fathers ($n = 31$)	SM Mothers ($n = 70$)	Control Mothers ($n = 31$)
Parent age	Mean 40.4 SD 6.4	Mean 41.7 SD 6.6	Mean 38 SD 5.5	Mean 40.23 SD 7.4
	$F = 2.9$; $p = .09$		$F = .78$; $p = .38$	
Education				
High school	17.1%	12.9%	11.4%	3.3%
Some college	15.7%	19.4%	21.4%	13.3%
College degree	31.4%	16.1%	38.6%	40.0%
Graduate degree	35.7%	51.6%	28.6%	43.3%
	$\chi^2_3 = 3.63$; $p = .30$		$\chi^2_3 = 3.65$; $p = .31$	
Ethnicity				
Caucasian	91.4	74.2	94.3	90.3
Latino	4.3	9.7	2.9	6.5
African American	—	3.2	—	3.2
Asian	1.4	3.2	1.4	—
Filipino/Pacific Islander	0	0	1.4	—
Mixed race	2.9	9.7	—	—
	$\chi^2_4 = 6.42$; $p = .17$		$\chi^2_4 = 3.89$; $p = .42$	

Note: SM = selective mutism.

mostly of public-speaking phobias) as well as other psychiatric disorders were not significantly different across groups. The mean total of social fears for the GSP group was 8.0, 2.7 for the non-GSP group and 1.4 for the no SP group. The GSP group had significantly more fears than either the non-GSP or the no SP group.

Sex: Simple Main and Moderating Effects

Fathers of SM children had significantly higher rates of GSP ($\chi^2_1 [n = 101] = 7.9; p = .005$) and AVPD than their control counterparts ($\chi^2_1 [n = 101] = 3.97; p = .046$). The pattern was different for mothers. There was a trend toward significance for GSP ($\chi^2_1 [n = 101] = 3.62; p = .057$), but not for AVPD ($\chi^2_1 [n = 101] = 2.45; p = .13$). Logistic regression analyses were conducted to test for an interaction between sex and proband status predicting the various psychiatric disorders. Neither the effect of sex nor the proband status by sex interactions was significant.

NEO Personality Inventory-Revised Domains and Facets

MANOVAs were conducted with sex included in the model as well as the interaction between sex and SM

TABLE 2

Distributions of Lifetime Psychiatric Disorders Across Children With SM and Controls

	SM Children (n = 70)		Control Children (n = 31)		χ^2	p
	No.	%	No.	%		
SAD	28	40	2	6.5	10.0	.002
SP	70	100	1	3.2	91.8	.000
GSP	69	98.6	1	3.2	87.4	.000
NGSP	1	2.9	0	0	0.03	.86
GAD	8	11.4	2	6.5	0.17	.68
OCD	6	8.6	0	0	1.50	.22
DYS	3	4.3	0	0	0.29	.59
MDD	5	7.1	2	6.5	0	1.0
ADHD	4	5.7	3	9.7	0.09	.77
CD	0	0	1	3.2	0.18	.67
ODD	4	5.7	1	3.2	0.001	.97
TIC	2	2.9	0	0	0.03	.86
Enuresis	10	14.3	2	6.5	0.62	.43

Note: SAD = separation anxiety disorder; SP = social phobia; GSP = generalized social phobia; NGSP = nongeneralized social phobia; GAD = generalized anxiety disorder; OCD = obsessive-compulsive disorder; DYS = dysthymia; MDD = major depressive disorder; ADHD = attention-deficit/hyperactivity disorder; CD = conduct disorder; ODD = oppositional defiant disorder; TIC = tic disorder.

TABLE 3

Distributions of Lifetime Psychiatric Disorders Across Parents of SM Probands and Controls

	Parents-SM		Parents-Controls		χ^2	p
	No.	%	No.	%		
MDD	40	29	17	26.6	0.12	.72
Mania	1	0.7	0	0	0.47	.50
Hypomania	3	2.2	0	0	1.41	.50
Dysthymia	5	3.6	1	1.6	0.64	.42
OCD	5	3.6	2	3.1	0.03	.86
PTSD	6	4.3	3	4.7	0.01	.91
Panic/agora	4	2.9	2	3.1	0.02	.93
Specific phobia	13	9.5	6	9.4	0.001	.98
GSP	51	37	9	14.1	10.98	.001
NGSP	9	6.6	4	6.3	0.005	.94
GAD	14	10.2	2	3.1	2.99	.08
AVPD	24	17.5	3	4.7	6.18	.013
SPD	0	0	0	0	0	0
SM	6	4.4	2	3.1	0.18	.67
SEP	8	5.8	1	1.6	1.84	.18

Note: SM = selective mutism; MDD = major depressive disorder; OCD = obsessive-compulsive disorder; PTSD = posttraumatic stress disorder; Panic/agora = panic disorder with agoraphobia; GSP = generalized social phobia; NGSP = nongeneralized social phobia; GAD = generalized anxiety disorder; AVPD = avoidant personality disorder; SPD = schizoid personality disorder; SAD = separation anxiety disorder.

status (SM versus control). Neither the main effect for sex ($F_{5,163} = 1.8; p = .12$) nor the interaction between sex and SM status was significant ($F_{5,163} = 0.82; p = .71$). MANOVAs revealed a main effect of proband status on the NEO Personality Inventory-Revised domains (Wilks $F_{5,163} = 3.39; p = .006$). As shown in Figure 1, parents of SM children had significantly higher mean *T* scores on the neuroticism and lower mean *T* scores on the openness domains than parents of control children. There were no significant differences for extraversion, agreeableness, or conscientiousness. Two MANOVAs were conducted on the six lower level neuroticism facets (anxiety, angry hostility, depression, self-consciousness, impulsiveness, and vulnerability) and the six openness facets (fantasy, aesthetics, feelings, actions, ideas, values; Wilks $F_{6,164} = 2.16; p = .05$ and Wilks $F_{6,164} = 2.56; p = .02$, respectively). The parents of the SM children scored higher than control parents on the anxiety, depression, self-consciousness, and vulnerability facets of neuroticism (data not shown). Parents of the SM children scored lower than parents of control children

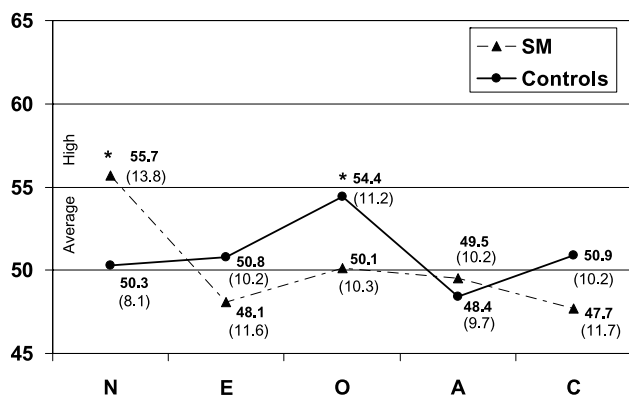


Fig. 1 NEO Personality Inventory-Revised domain mean (*T* scores) and SDs (in parentheses) across the SM and control groups. *Significant difference between groups on the given domain ($p < .05$). N = Neuroticism; E = Extraversion; O = Openness; A = Agreeableness; C = Conscientiousness; SM = selective mutism.

on the aesthetics and ideas facets of openness (data not shown).

DISCUSSION

Our data support a familial relationship between SM and SP. In this study parental GSP and AVPD were three- to fourfold more common among parents of SM children than control children. Furthermore, child SM severity predicted parent SP (generalized type). GSP has often been characterized as a more severe form of social anxiety, and it is possible that SM may be an early onset form of the disorder. Although there were not more lifetime diagnoses of SM in the parents of SM children when compared to controls, we did find that overall, 87.5% of parents with SM ($n = 8$) had a lifetime GSP diagnosis ($\chi^2 = 10.43$; $p < .001$), and in this group, the mean age at onset of SM (mean 4.3 years, SD 0.96) preceded GSP (mean 6.43 years, SD 3.15). Similarly, AVPD, which shares much in common with GSP (Ralevski et al., 2005), may also lie on this same continuum of social anxiety and represent a more severe form of the disorder during adulthood. Without additional longitudinal research, it is difficult to know whether such a continuum actually exists or whether these relationships are a product of an imperfect diagnostic classification system in which criteria overlap. At minimum, data support the conclusion that SM is related to GSP and, like GSP, has a familial and likely heritable component (Mannuzza et al., 1995; Stein et al., 1998a,b).

In this study we also found that fathers of children with SM had more SP and AVPD diagnoses than fathers of control children; however, a similar pattern was not present among mothers. Given some data suggesting that GSP and AVPD are more common in women than men (Kessler et al., 1994; Reichborn-Kjennerud et al., 2006), the finding of an increase in fathers of SM children speaks to the possibility that the fathers may have an especially severe (or penetrant) form of SP, and, thus, it may be especially likely to result in a severe form of SP (i.e., SM) in their offspring. Important to note, however, that the odds of GSP and AVPD were also increased in mothers of children with SM when compared to controls, although they failed to reach conventional levels of statistical significance; the failure to find significant associations in mothers may simply be an issue of sample size.

Quantitative assessments of dispositional characteristics may be most informative when trying to understand the heritable component of mental disorders (Eley et al., 2003; Stein et al., 1998a,b). If SM has a heritable component, then it is likely that dimensional traits such as neuroticism or temperamental characteristics (e.g., behavioral inhibition, shyness) are those variables that are transmitted. In the present study, parents of children with SM reported higher levels of neuroticism, which was further characterized as higher levels of anxiety, self-consciousness, depression, and vulnerability. In both clinical and general population studies, neuroticism has been identified as a risk factor for anxiety (Hettema et al., 2004; Jylha and Isometsa, 2006). Previous research also suggests a possible relationship between certain candidate genes, in particular, the serotonin transporter promoter polymorphism and neuroticism (Schinka et al., 2004; Sen et al., 2004; Stein and Bienvenu, 2004), and the corticotrophin-releasing factor gene and behavioral inhibition (Smoller et al., 2005). It is possible that such genetic factors and associated vulnerabilities may be present in children with SM. Given that only a portion of children with such characteristics (e.g., behavioral inhibition) develop later anxiety disorders (i.e., SP; Schwartz et al., 1999), the etiology of SM is likely complex, incorporating environmental factors (e.g., family environment, parenting style, traumatic conditioning experiences) and likely multiple genes.

In this study parents of SM children also had lower openness scores than control parents. Openness to

experience can represent a willingness to entertain novel ideas and unconventional values; those who score low on openness often prefer the familiar to the novel and tend to be more conventional in behavior and conservative in outlook (Costa and McCrae, 1992). The enrichment of these attributes in parents of SM children is understandable, particularly in the presence of elevated rates of SP in this group. Surprisingly, contrary to our hypothesis, parents of children with SM did not report significantly lower extraversion scores than control parents. Extraversion, a measure of positive emotionality, sociability, optimism, and energy (Costa and McCrae, 1992), has typically been shown to share a negative relationship with both depression and anxiety (Stein et al., 2004; Stewart et al., 2005). In a recent general population study, however, only neuroticism shared a relationship with both anxiety and depression, whereas extraversion was associated only with depression (Jylha and Isometsa, 2006). Continued investigation is warranted to more fully understand the role of extraversion in both clinical and population samples.

Consistent with previous findings, all of the children with SM also had an SP diagnosis (Black and Uhde, 1995; Dummit et al., 1997). Such comorbidity limits some of the conclusions that can be drawn about SM in particular, given that its influence cannot be completely disentangled from childhood SP. The overlap between SM and SP is a diagnostic classification issue that warrants clarification in future revisions of the *DSM*.

Limitations

Telephone interviews were used to facilitate nationwide recruitment and are associated with the standard limitations of not conducting interviews in person. Given the low base rates of SM, however, it would have been difficult to recruit a large sample size by only ascertaining subjects from one geographic region. The use of telephone interviews as well as the young age of our probands led us to rely on parent report of their child's SM. It is possible that such reports may have been inconsistent with the child's report; however, given the visibility of such behaviors (i.e., not talking), it is likely that ascertainment of SM is less prone to parent-child discrepancies than other disorders such as depression and generalized anxiety disorder, which rely on mood symptoms or worries.

Another limitation is the fact that we did not include a formal assessment of communication disorders and

pervasive developmental disorders. An informal clinical interview, a series of questions from a screening section (taken from the ADIS-P/C) and in a small number of cases, videotape review were used to assess developmental delays, language deficits, psychotic symptoms, and psychiatric history. In-person standardized assessments to comprehensively assess for the presence of these disorders would have been optimal, yet in this case not feasible.

Last, there is the possibility of a sampling bias as well as other threats to external validity in this study. We recruited families from a Web site and from national conferences, which suggests that these families may have had more motivation to educate themselves about SM than other parents of children with SM. Perhaps this motivation may have stemmed from parents themselves having more social anxiety symptoms. It is also possible that parents with more social anxiety would have been less likely to engage such services, at least, the in-person conferences, which could have led to a bias in the opposite direction. Given that this was a substudy of a genetics project, only those families in which both biological parents were available were eligible to participate. In addition, the sample was nearly all white and the majority had at least a college education. The fact that the control sample was recruited from the San Diego region further limits the generalizability of this study.

Clinical Implications

Taken together, these findings suggest that SM has diagnostic and familial ties to GSP. At this point, it is not possible to disentangle genetic contributions from environmental forces (e.g., social learning). Future twin studies, adoption studies, and genetically informative studies (e.g., candidate and genome-wide association studies) will further inform questions of this nature. As suggested by others, SM may be an indicator of underlying psychopathology that has a more protracted course than the mere not talking symptoms that are the hallmark of SM (Steinhausen et al., 2006). More broadly, SM may be a marker for the risk of later phobic and anxiety disorders and therefore in the presence of persistent SM, early intervention is warranted. Parents who suffer from social anxiety and their children may benefit from this knowledge, particularly in the presence of validated treatments for child SP (Beidel et al., 2005; Kendall et al., 1997; Wagner et al., 2004) and new treatments for SM that are emerging.

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Beyond Munchausen Syndrome by Proxy: Identification and Treatment of Child Abuse in a Medical Setting

John Stirling Jr., MD, and the Committee on Child Abuse and Neglect

The condition widely known as Munchausen syndrome by proxy comprises both physical abuse and medical neglect and is also a form of psychological maltreatment. Although it is a relatively rare form of child abuse, pediatricians need to have a high index of suspicion when faced with seemingly inexplicable findings or treatment failures. The fabrication of a pediatric illness is a form of child abuse and not merely a mental health disorder, and there is a possibility of an extremely poor prognosis if the child is left in the home. In this statement, factors are identified that may help the physician recognize this insidious type of child abuse that occurs in a medical setting, and recommendations are provided for physicians regarding when to report a case to their state's child protective service agency. *Pediatrics* 2007;119:1026–1030.