

# Communicative Competence in Parents of Children with Autism and Parents of Children with Specific Language Impairment

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**Abstract** While the primary language deficit in autism has been thought to be pragmatic, and in specific language impairment (SLI) structural, recent research suggests phenomenological and possibly genetic overlap between the two syndromes. To compare communicative competence in parents of children with autism, SLI, and down syndrome (DS), we used a modified pragmatic rating scale (PRS-M). Videotapes of conversational interviews with 47 autism, 47 SLI, and 21 DS parents were scored blind to group membership. Autism and SLI parents had significantly lower communication abilities than DS parents. Fifteen percent of the autism and SLI parents showed severe deficits. Our results suggest that

impaired communication is part of the broader autism phenotype and a broader SLI phenotype, especially among male family members.

**Keywords** Autism · Specific language impairment · Communication · Pragmatics · Family study

Communication deficits are hallmark features of autism, even among the highest functioning individuals. In conversations and play situations, verbal children with autism initiate less often, give fewer responses to questions, take fewer turns, chat less

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and are less able to maintain a topic of conversation compared with children with down syndrome (DS), specific language impairment (SLI) or typical development (Bartak, Rutter, & Cox, 1975; Eales, 1993; Loveland, Landry, Hughes, Hall, & McEvoy, 1988; Tager-Flusberg & Anderson, 1991). Children with autism also often fail to adapt their account to the conversational context; e.g., they use technical jargon, fail to make clear references, give inadequate background information, and make more socially inappropriate remarks than controls (Baltaxe, 1977; Bartak et al., 1975; Loveland, McEvoy, Tunali, & Kelly, 1990; Tager-Flusberg, 2000).

Milder social and communication impairments have been reported consistently among relatives of autistic individuals as a part of the broader autism phenotype. Using informant-based family history data, social-pragmatic deficits are found more frequently among autism relatives than DS relatives (Bolton et al., 1994) and among biological than non-biological autism relatives (Szatmari et al., 2000). Three studies have directly examined communication in autism parents. Wolff, Narayan, and Moyes (1988) observed more frequent communication impairments, labeled schizoid traits, in autism parents compared with parents of children with mental handicap. Autism parents were noted to have lack of empathy, lack of emotional responsiveness, impaired rapport, too little smiling, and suspiciousness (Wolff et al., 1988). Landa et al. (1992) developed the Pragmatic rating scale (PRS), an interviewer-based instrument that assesses the more subtle deficits in pragmatic language among autism relatives. They found that autism parents had higher and therefore more abnormal scores on the PRS than parents of children with DS (Landa et al., 1992). Piven et al. (1997) replicated these results in multiple-incidence autism families using the PRS. They also found speech abnormalities more frequently among autism parents than parents of children with DS. In all three studies, ratings were made blind to family membership.

While a hallmark of autism is a primary deficit in the use of language for communication, parents, and siblings of autistic probands were also more likely to report delay in onset of speech, articulation defects, and reading and spelling difficulties in family history studies (Bartak et al., 1975; Bolton et al., 1994; Wzorek et al., 1989). When directly tested, autism parents who had reported a history of these language-related difficulties scored significantly lower on verbal intelligence, spelling, the nonsense reading test from the Woodcock–Johnson battery, and higher on the PRS in comparison with autism parents without such difficulties (Folstein et al., 1999).

Individuals with SLI have occasionally been included as a comparison group in autism studies to control for language abnormalities. It has been thought that in SLI mainly structural language is impaired. However, some children with SLI show pragmatic and social deficits (Bishop & Norbury, 2002; Bishop, North, & Donlan, 1995). Their verbal expressions are less cohesive, and their messages less specific, accurate and intelligible than those of unaffected children. They were also impaired in their ability to revise and clarify messages (Prutting & Kirchner, 1987). Adults with SLI have prosodic oddities, problems sustaining a conversation and difficulties reporting events (Mawhood & Howlin, 2000).

Among children with SLI, a subgroup was found with both structural and pragmatic language impairment (PLI). For these children, who had fluent and complex expressive language but used language in an abnormal way, the terms ‘semantic-pragmatic disorder’ and ‘pragmatic language impairment’ were coined (Bishop, 2000; Conti-Ramsden & Botting, 1999; Rapin & Allen, 1983). Bishop (2000) argues that PLI is intermediate between SLI and autism. Bishop proposed that PLI be diagnosed only in children who do not meet stringent criteria for autism. These non-autistic children with PLI who were characterized as sociable and talkative, with verbal and non-verbal communication skills but who used stereotyped language with abnormal, often exaggerated prosody, also scored significantly lower on expressive and receptive language composite scores than typically developing controls (Bishop & Norbury, 2002).

A family study has revealed a higher rate of autism in siblings of SLI probands than in the general population (Tomblin, Hafeman, & O’Brien, 2003). However, communication has not yet been studied in family members of children with SLI. Given the partial overlap of language deficits found in autism and SLI probands and the existence of the broader autism phenotype in autism family members, we hypothesized that SLI parents would also endorse communication deficits, albeit milder than autism parents.

Communicative competence covers a wide range of skills that include elements of social communication, pragmatic language and speech, and expressive fluency. Social communication and pragmatic language are related and overlapping concepts that have been used to describe communicative behaviors in autism and SLI. Social communication refers to the ability to convey abstract and emotional information using facial expression, gesture and prosody, and “implies knowledge of social rules of communication and the implicit ability to deduce the thoughts and motives of others” (Tanguay, Robertson, & Derrick, 1998). Pragmatic

language is generally referred to as the use of language appropriate to social context (Bates, 1976). In the strict linguistic sense, conversational pragmatic abilities include initiation, turn-taking/conversational to and fro, cohesion/appropriate use of references, coherence, topic maintenance, and social appropriateness (Adams & Bishop, 1989; Baltaxe, 1977; Bishop, 1998; Craig & Evans, 1993; Landa et al., 1992; Prutting & Kirchner, 1987; Roth & Spekman, 1984). In addition, non-verbal communicative behaviors such as eye contact, facial expressions, gestures, and body posture and paralinguistic aspects of speech such as prosody, fluency, and intelligibility (Bishop, 1998; Prutting & Kirchner, 1987) are subsumed under a broader definition of pragmatics.

Existing measures of pragmatics and social communication either do not cover all the aspects of communication that we wished to assess or they are not used in the same context. The PRS includes few aspects of non-verbal communication and formal language. The Children's communication checklist (CCC) is comprehensive, but is scored by therapists who are familiar with a child's communication abilities across a range of contexts, and is not validated for use with adults (Bishop, 1998, 2003). We therefore modified the PRS (PRS-M) to include additional aspects of non-verbal communication and formal language. We used it to compare communication impairments in conversational speech of parents of children with autism, SLI, and DS. We chose parents of children with DS as control group because they do not carry an increased genetic liability for communication disorders and have been used to control for the effect of caring for a handicapped child.

## Methods

### Participants

#### *Ascertainment of Families*

For the ascertainment of autism and SLI families, the project drew on language samples collected for a family study of the language phenotype in autism and SLI. Two sites participated, Tufts-New England Medical Center in Boston and the University of Iowa. SLI families from the Iowa site were members of a longitudinal cohort (Tomblin, Zhang, Buckwalter, & Catts, 2000) that had been sampled from a cross-sectional population sample of kindergarten children (Tomblin et al., 1997). SLI families at the Boston site were recruited through classes and services specifically for children with language impairment. We wished to

avoid a sampling bias toward ascertaining SLI families who were concerned that their child may have symptoms of autism. Therefore, the SLI families at both sites were told that we were studying language and reading in family members of children with SLI. The autism recruitment was carried through services for children with autism spectrum disorders at both the Iowa and the Boston sites. These families were told only that the study was an investigation of language and reading in families of children with autism.

DS parents had been ascertained as the control group for an earlier study of personality and language characteristics in autism parents at the University of Iowa. In this earlier study autism parents were compared with DS parents on multiple measures, including the PRS (Piven et al., 1997). Videotaped language samples from 21 of 55 DS parents were randomly selected for the current study. For this set of tapes, language samples from both autism and DS parents were scored to maintain rater blindness. The conditions for obtaining the language sample were the same in both studies (Piven et al., 1997).

For this investigation of parents' communicative competence, we selected 47 parents of autistic probands ("autism parents") and 47 parents of SLI probands ("SLI parents") who could be individually matched on verbal IQ in order to avoid possible influences of verbal IQ on communication. To maximize the number of matched pairs, we matched without reference to family membership, so that one or both parents of 27 probands with autism and 29 probands with SLI were included in these analyses. Due to power constraints, the parents of children with DS ("DS parents") were not matched on verbal IQ. The 21 DS parents came from 12 families who had one child with DS.

#### *Entry Criteria/Proband Definition*

The autism and SLI probands were between the ages of 6 and 16, had a verbal IQ of 60 or above as measured on the Wechsler intelligence scale for children vocabulary and similarities subtests (WISC-III) (Wechsler, 1991b) and had at least one sibling in the same age and IQ range. Both parents agreed to participate, and the family's first language was English. Probands with autism met criteria for autism according to the Autism diagnostic interview-revised (Lord, Rutter, & Le Couteur, 1994) and had sufficient language ability to be tested on the full battery. Probands were defined as having SLI if they performed at or below the 13th percentile on the total language score of the Clinical evaluation of language fundamentals (Semel, Wiig, & Secord, 1995) or at or below the ninth percentile on the non-word repetition subtest of the Comprehensive test of

phonological processing (Wagner, Torgesen, & Rashotte, 1999). The non-word repetition task has been shown to be a sensitive and specific psycholinguistic marker for SLI (Conti-Ramsden, Botting, & Faragher, 2001; Tager-Flusberg & Cooper 1999), and it detects a history of SLI in over 50% of school-aged probands who, by that time, often score above threshold on standardized language tests (Conti-Ramsden et al., 2001). The probands with DS had a non-disjunction of chromosome 21 and were between the ages of 3 and 25.

### Exclusion Criteria

Exclusion criteria included diagnosis of fragile-X syndrome, congenital rubella, phenylketonuria, neurofibromatosis, tuberous sclerosis, familial mental retardation, severe birth trauma, or brain injury. We also excluded families where probands had no specific medical diagnosis but had dysmorphic features or serious illness in early life that could have caused their disorder. Families who had more than one child with autism were included only if there was also a non-autistic sibling in the required age range.

### Proband and Parent Characteristics

Table 1 presents the characteristics for the probands and parents. The probands differed only in their gender

**Table 1** Demographic characteristics of the probands and parents

|                        | Mean (SD)   |  |   |
|------------------------|---|--|---|
|                        | Autism<br>(proband<br>N = 27)<br>(parent<br>N = 47) | SLI<br>(proband<br>N = 29)<br>(parent<br>N = 47) | DS<br>(proband<br>N = 12)<br>(parent<br>N = 21) |
| <b>Probands</b>        |   |  |   |
| Age                    | 10.71 (2.80)  | 11.41 (1.55)                                     | 10.02 (6.63)                                    |
| Grade                  | 4.53 (2.80)   | 5.02 (1.51)                                      |   |
| Performance IQ         | 88.98 (22.75)                                       | 91.38 (14.22)                                    |   |
| Verbal IQ              | 87.44 (18.75)                                       | 87.04 (10.27)                                    |   |
| Fullscale IQ           | 86.63 (19.60)                                       | 87.69 (12.07)                                    |   |
| <b>Parents</b>         |   |  |   |
| Age                    | 40.89 (4.50)  | 39.49 (5.38)                                     | 39.20 (7.63)                                    |
| Education <sup>a</sup> | 3.23 (0.73)   | 3.15 (0.75)                                      | 2.76 (0.94)                                     |
| Performance IQ         | 105.01 (12.84)                                      | 106.82 (11.93)                                   | 112.22 (18.10)                                  |
| Verbal IQ              | 105.38 (10.43)                                      | 104.42 (10.51)                                   | 108.47 (14.73)                                  |

SD standard deviation, SLI specific language impairment, DS down syndrome, IQ intelligence quotient

<sup>a</sup> Parents education is given in four educational attainment categories: 1 = without H.S. diploma, 2 = H.S. graduate without college education, 3 = some college education, 4 = degree from 4-year college or higher

distribution: 85% of the probands with autism, 59% of the probands with SLI, and 46% of the probands with DS were male ( $\chi^2(2) = 7.29, P = .026$ ). For the DS probands school grade and IQ data were not available. The gender of the parents was equally distributed with 49% of the autism, 45% of the SLI, and 48% of the DS parents being fathers. The autism parents had a significantly higher education than the DS parents ( $t(66) = 2.25, P = .028$ ). The parents' ethnicity revealed no significant differences. Most autism and SLI parents and all DS parents were Caucasian. Three autism parents were Hispanic and one of the SLI parents fell in the "other ethnicity" category. There were no other significant differences between the autism, SLI, and DS parents on the demographic variables.

### Measures

#### IQ and Family History

The parents' IQ scores were estimated using two verbal subtests (vocabulary and similarities) and two performance subtests (block design and picture arrangement) of the Wechsler adult intelligence scale (WAIS-III); the parallel abbreviated WISC-III was administered to the probands (Wechsler, 1991a, b).

A modified version of the investigator-based Family history interview of developmental disorders of cognition and social functioning (FHI) was used to assess traits characteristic of autism in the autism and SLI parent groups (Bolton et al., 1994). These traits include developmental disorders of speech, reading and spelling, indices of social-pragmatic functioning in childhood and adulthood, and obsessive-compulsive phenomena. For this study, we obtained information directly from each parent when possible, or in some cases from the spouse.

#### Language Sample

A 20-min language sample was recorded on video. The interviewer fostered a situation that is thought to best reveal social pragmatic deficits (Landa et al., 1992). The interviewer first familiarized the participant with the goal of creating a conversation without defining "conversation", but encouraged the participant to be conversational partner, i.e., to ask questions him/herself since the language sample followed a highly structured interview. The interviewer initiated the conversation, e.g., asking the participant to describe her occupation and hobbies. This provided a prompt for the participant to use and define terminology, provide references, and to express preferences and feelings. The interviewer also related personal accounts appropriate to the context to

show understanding and encourage empathy. The interviewer occasionally indicated misunderstanding of a word or fact by saying “What do you mean by that?” or “What is that?” in order to observe whether and how the issue was clarified. All interviewers were female.

Fifteen minutes of the language sample was scored blindly using the PRS-M.

### Development of the PRS-M

#### *Item and Coding Development and Training of the Raters*

The PRS (Landa et al., 1992) is a rater-based instrument developed to evaluate the pragmatic deficits in the social use of language in relatives of autistic children. The original, unpublished PRS includes 31 items (Landa, 1991). For 19 of these items interrater reliability was obtained, and they were initially published as the PRS (Landa et al., 1992). An additional six speech items were later published and shown to be atypical in autism parents (Piven et al., 1997). For the PRS-M we incorporated items from the original unpublished and published PRS. We refined some items and codes by making them more specific and thus easier to code reliably and combined several items that were not mutually exclusive. We added verbal emotional expressions and grammatical errors, since they have been found to be abnormal in children with autism (Lord et al., 1989; Pearlman-Avni & Eviatar, 2002; Tager-Flusberg & Sullivan, 1995), autism parents (Folstein et al., 1999) and in probands with SLI (Leonard, 1998). Each pragmatic behavior was rated on a 3-point scale with 0 indicating typical behavior, 1 indicating some abnormal behavior, but limited in quantity, and 2 indicating frequently abnormal behavior. Possible overall scores ranged from 0 to 30. The 15-item PRS-M is attached in the appendix.

One author (D.A., speech and language pathologist) had been trained to score the published PRS. Three authors who became the raters for the PRS-M (D.A., T.R., and S.P.) developed the PRS-M and applied it blindly to 20 randomly selected language samples. Through discussion of each individual score a consensus was reached and qualitative and quantitative abnormalities were clearly defined.

### Psychometric Properties of the PRS-M

#### *Inter-rater Reliability*

Two raters (T.R. and S.P.) blindly and independently watched and rated 47 videos that were randomly

selected from the larger sample. The intra-class correlation coefficient for the sum of all 15 PRS-M items was .72, indicating an overall good reliability. The  $\kappa$  values for the individual items ranged from .31 to .80 with percent agreement ranging from 66 to 96%. The four items ‘Indirect verbal emotional expression’, ‘Emphatic gestures’, ‘Mispronunciation’ and ‘Empathy’ were only seldom endorsed with positive ratings in 3–7 of the 47 reliability cases. Therefore, the  $\kappa$  values of these items were below .30. Because the inter-rater agreement was at least 72%, these items were retained.

#### *Scale Formation*

The intra-class correlation coefficient of the 15 PRS-M items for the autism and SLI parents combined was .08, indicating that the PRS-M does not form a single scale. To explore possible subscales, we entered the 15 items into VARCLUS (SAS, 2000). The VARCLUS procedure uses oblique principal component analysis (Harman, 1976). For any group of variables, the principal components are the “directions” (each given by some linear combination of the variables) in which most of the variation of the data is explained. The first principal component is the one that accounts for more variation than any other linear combination of the items. VARCLUS splits the variables into clusters or subscales to maximize the amount of variation explained by the totality of all the first principal components of the clusters. Variables loaded on to a cluster tend to be correlated, while variables in distinct clusters tend to be uncorrelated. In order to maximize internal consistency, we included only variables that were correlated with the other variables in their own cluster at  $R > .30$  or  $R^2 > .09$ . To improve item discriminant validity we included variables only when their correlation with variables of their own cluster was large relative to their correlation with the next closest cluster. Four clusters or ‘subscales’ emerged that accounted for about 46% of the variation, as shown in Table 2.

Under subscale 1, verbal emotional and facial expression were evaluated as an indication of a person’s expressiveness, whereas empathy and referencing skills tap understanding and awareness of the conversational partner. Under subscale 2, the items prosody, descriptive and emphatic gestures and eye contact influence the immediacy of the conversational contact. The core characteristic of subscale 3 is overproduction of two different aspects of speech: dominating the conversation and providing unnecessary details. Formal speech and language items

comprise subscale 4: grammatical errors, mispronunciation, confusing accounts and frequent reformulations.

### Validation Procedures

Since there does not exist a gold standard for the interview-based assessment of communication abilities, and the PRS has not been validated, we chose to validate the subscales of the PRS-M against the data obtained from the FHI for the autism and SLI parents. Single items and three of the factors derived from the FHI by Zwaigenbaum et al. (2000): academic learning problems, social-pragmatic impairment, and odd behavior were employed. Predictions were made as to which FHI factors and items would correlate with each subscale, as shown in Table 3. ‘Adult conversation’ is an FHI item that covers a broad range of conversational skills. It was predicted to correlate with all PRS-M subscales except for ‘Language’. We further thought the FHI factor ‘Social-pragmatic impairment’ would be associated with high scores on the subscales ‘Emotional expressiveness and awareness of the other’ and ‘Communicative performance’. Lack of friendships in childhood and adulthood are expected to be indirectly linked to poor performance on the PRS-M subscales ‘Emotional expressiveness and awareness of the other’ and ‘Over-talkativeness’. Correlations between the FHI items and factors and the PRS-M subscale scores were obtained, and significant correlations are displayed in Table 3.

**Table 2** Items and subscales of the Pragmatic rating scale—modified

| Items  | R <sup>2</sup> (own subscale) |
|--|-------------------------------|
| <b>Emotional expressiveness and awareness of the other</b> |                               |
| Direct emotional verbal expression                         | .54                           |
| Indirect emotional verbal expression                       | .51                           |
| Failure to reference                                       | .25                           |
| Facial expressions   | .28                           |
| Empathy  | .59                           |
| <b>Communicative performance</b>                           |                               |
| Prosody  | .38                           |
| Descriptive gestures                                       | .61                           |
| Emphatic gestures  | .67                           |
| Eye contact  | .11                           |
| <b>Over-talkativeness</b>                                  |                               |
| Dominating Conversation                                    | .82                           |
| Overly detailed  | .82                           |
| <b>Language</b>  |                               |
| Grammatical errors   | .33                           |
| Confusing accounts   | .51                           |
| Reformulation  | .46                           |
| Mispronunciation   | .36                           |

### Results

#### Total PRS-M score, Subscale Scores and Individual Items

The mean total PRS-M score for the autism and SLI parents combined was 3.94 ( $SD = 3.13$ ) with a range from 0 to 19, and for the DS parents 1.09 ( $SD = 1.48$ ) with a range from 0 to 6. As shown in Fig. 1, the distribution of the PRS-M score for the autism and SLI parents is skewed to the left, describing a large subset of the sample with normally distributed scores and a smaller subset with high scores. About 15% of the autism and SLI parents scored 7 or higher (14.9% of the autism parents and 14.9% of the SLI parents). Parents who scored  $\geq 7$  conversed with great difficulty. An autism parent with a score of 7 and higher tended to be a man who had no or poor eye contact, gave confusing accounts with many reformulations, made no empathic statements, produced several grammatical errors and had a flat intonation. The SLI parent in this group in addition tended to have no or minimal facial expression and leave out explanations for references. One autism father with the score of 7 had a possible autism spectrum disorder according to the FHI.

The autism parents did not score differently in their communicative competence from the SLI parents on either the total PRS-M score or the four subscales. When the individual items of the PRS-M were compared, no differences remained significant after Bonferroni correction. When the autism and the SLI parents were each compared with the DS parents, significant differences resulted for three of the four subscales and for the total PRS-M score as shown in Table 4. Autism parents scored also significantly higher than DS parents on the individual items reformulation ( $\chi^2(1) = 10.97, P = .0008$ ) and confusing accounts ( $\chi^2(1) = 7.88, P = .003$ ). These were the only differences that remained significant after Bonferroni correction. When the SLI parents and the DS parents were compared on individual items, no differences remained significant after adjusting for multiple comparisons.

When all individual PRS-M variables for the autism and DS parents were entered into a logistic regression analysis, the items “failure to reference” and “reformulations” best predicted group membership for the autism parents (model:  $\chi^2(15) = 42.48, P = .0002$ ; group membership for 89% of the autism parents and 71% of the DS parents was predicted correctly; for “failure to reference”:  $B$  coefficient =  $-2.45$ , S.E. = 1.14, Wald statistic 4.61,  $df = 1, P = .032$ ; for “reformulations”:  $B$  coefficient =  $-2.1$ , S.E. = .98,

**Table 3** Validation of the pragmatic rating scale—modified<sup>a</sup>

|   | Predicted correlation  | Significant correlation   |
|---|--|---|
| Emotional expressiveness and awareness of the other | Social-pragmatic impairment <sup>b</sup><br>Friendships in childhood<br>Friendships in adulthood<br>Adult conversation | Friendships in adulthood<br>$r = .21; P = .046$   |
| Communicative performance                           | Social-pragmatic impairment <sup>b</sup><br>Shyness<br>Adult conversation  | Adult conversation<br>$r = .31; P = .002$<br>Social-pragmatic impairment<br>$r = .21; P = .041$       |
| Over-talkativeness                                  | Friendships in childhood<br>Friendships in adulthood<br>Adult conversation   | Friendships in childhood<br>$r = .21; P = .047$   |
| Language  | Academic learning problems <sup>c</sup><br>Difficulties with reading<br>Difficulties with writing                      | Difficulties with reading<br>$r = .37; P < .001$<br>Academic learning problems<br>$r = .30; P = .003$ |

<sup>a</sup> Family history was missing for three participants

<sup>b</sup> Social-pragmatic impairment = lack of affection, social play in childhood, adult conversation

<sup>c</sup> Academic learning problems = difficulties with reading, spelling or mathematics

Wald statistic 4.66,  $df = 1, P = .031$ ). For the model that included the SLI and DS data and was designed to predict group membership for SLI parents, the items “grammatical errors” and “dominating conversation” were significant (model:  $\chi^2(15) = 40.64, P = .0004$ ; group membership for 91% of the SLI parents and 67% of the DS parents was predicted correctly; for “grammatical errors”:  $B$  coefficient =  $-2.07, S.E. = .97$ , Wald statistic 4.52,  $df = 1, P = .034$ ; for “dominating conversation”:  $B$  coefficient =  $-2.97, S.E. = 1.04$ , Wald statistic 3.85,  $df = 1, P = .049$ ).

Comparison of Parents by Gender

Autism and SLI Parents Combined

Table 5 presents the significant results for the comparison of fathers and mothers for the combined autism and SLI parent group. Fathers had significantly higher scores on the total PRS-M and the communicative performance subscale. Fathers also more often endorsed poor eye contact than mothers ( $\chi^2(1) = 12.7,$

$P = .002$ ); the only result that remained significant for a single item after Bonferroni correction.

We then compared eye contact for fathers and mothers in each group. Autism mothers more often had poor eye contact than SLI mothers (20.8% of autism mothers vs 3.8% of SLI mothers), although this was only a trend statistically ( $\chi^2(1) = 3.41, P = .065$ ). Autism and SLI fathers had equally poor eye contact.

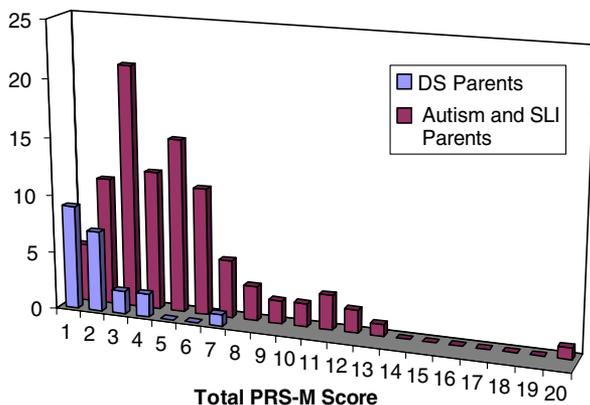
Gender Differences Within Groups

Autism fathers scored higher than autism mothers on the total PRS-M score ( $t = (45) 2.75, P = .008$ , Cohen’s  $d = 0.820, r = .38$ ) and on the communicative performance subscale ( $t = (45) 2.59, P = .013$ , Cohen’s  $d = 0.772, r = .36$ ). These differences remained significant after Bonferroni correction. For eye contact, autism fathers scored significantly higher than autism mothers (20.8% of mothers vs 52.2% of fathers,  $\chi^2(1) = 5.00, P = .025$ ).

Differences between SLI fathers and mothers on the PRS-M score and subscale scores did not remain significant after Bonferroni correction. Fathers of children with SLI had significantly poorer eye contact than did SLI mothers (3.8% of mothers vs 42.7% of fathers,  $\chi^2(1) = 10.55, P = .001$ ).

Discussion

This is the first comparison of communicative abilities in parents of autism, SLI and DS probands, and the first report on communicative competence in SLI parents. Four main results emerged. First, the autism and SLI parents had significantly lower communication abilities than the DS parents. About 15% of both, the autism and SLI parents had serious communication problems. Second, “failure to reference” and “reformulations”



**Fig. 1** Distribution of the total PRS-M scores

**Table 4** Paired and independent *t* tests for the four subscales and the PRS-M by diagnosis

|                           | Mean (SD)   |             |             | AU vs SLI parents <sup>a</sup> |          |                  | AU vs DS parents <sup>b</sup> |          |                  | SLI vs DS parents <sup>b</sup> |          |                  |
|---------------------------|---|-------------|-------------|--------------------------------|----------|------------------|-------------------------------|----------|------------------|--------------------------------|----------|------------------|
|                           | Autism parents                                      | SLI parents | DS parents  | <i>t</i> -value                | <i>P</i> | Cohen's <i>d</i> | <i>t</i> -value               | <i>P</i> | Cohen's <i>d</i> | <i>t</i> -value                | <i>P</i> | Cohen's <i>d</i> |
|                           | Emotional expressiveness and awareness of the other | 0.97 (1.34) | 1.38 (1.85) | 0.29 (0.56)                    | 1.17     | 0.247            | 0.25                          | 3        | .004*            | 0.66                           | 3.71     | >.001**          |
| Communicative performance | 0.83 (1.18)   | 0.91 (1.30) | 0.38 (0.97) | 0.34                           | 0.734    | 0.06             | 1.52                          | 0.133    | 0.42             | 1.68                           | 0.097    | 0.46             |
| Over-talkativeness        | 0.53 (0.97)   | 0.53 (1.01) | 0.09 (0.30) | 0                              | 1        | 0                | 2.79                          | .007*    | 0.61             | 2.69                           | .009*    | 0.59             |
| Language                  | 1.47 (1.40)   | 1.28 (1.36) | 0.33 (0.66) | 0.63                           | 0.529    | 0.14             | 4.55                          | <.001**  | 1.04             | 3.85                           | <.001**  | 0.89             |
| Total PRS-M score         | 3.81 (2.65)   | 4.10 (3.56) | 1.09 (1.48) | 0.44                           | 0.66     | 0.09             | 5.39                          | <.001**  | 1.27             | 4.92                           | <.001**  | 1.1              |

SD standard deviation, AU autism, SLI specific language impairment, DS down syndrome, PRS-M modified pragmatic rating scale

\* *P* < .05 after Bonferroni correction; \*\* *P* < 0.005 after Bonferroni correction

<sup>a</sup> Paired *t*-test

<sup>b</sup> Independent *t*-test

best predicted group membership for autism parents while “grammatical errors” and “dominating conversation” predicted whether the parent had a child with SLI. Third, autism and SLI fathers had overall lower communication abilities and scored higher on eye contact than autism and SLI mothers. Fourth, there was a trend for autism mothers to have poorer eye contact than SLI mothers, and to be more like the autism fathers on this aspect of non-verbal communication.

Contrary to our hypothesis, we did not find differences in the overall communication skills between the autism and SLI parents. All previous studies are consistent in finding more frequent communication deficits in parents of autistic children than in parents of typically developing children, children with DS or mental handicap (Landa et al., 1992; Piven et al., 1997; Wolff et al., 1988). Results from the studies that used the PRS can be compared with ours. Using the 19-item PRS, Landa et al. (1992) reported a mean total score for autism parents and parents of children with DS of 4.41 and 0.45, respectively. Likewise, Piven et al. (1997) found in their sample of autism parents a mean PRS score of 3.9 compared to a score of 0.8 for the DS control parents, as well as significantly higher scores among the autism parents for six additional speech items. These mean PRS scores are similar to the ones we obtained with the 15-item PRS-M for the autism, SLI and DS parents, 3.80, 4.10, and 1.09, respectively. Thus, we conclude that SLI parents, like autism parents, more often have significant communication problems than controls. This is a striking and somewhat unexpected result.

Several earlier studies reported higher scores in fathers of autistic children than in mothers. Male autism relatives more frequently endorse social-pragmatic deficits and the broader autism phenotype according to family history studies (Bolton et al., 1994; Szatmari et al., 2000). However, neither Landa nor Piven found a gender difference in their autism and control parents, although their sample sizes were comparable with ours (Landa et al., 1992; Piven et al., 1997). One explanation for the discrepancy may be that the PRS does not tap certain non-verbal characteristics, such as eye contact, which yielded a gender difference in our study. In addition, there may have been gender differences for single items in these studies that were obscured in the overall score.

The finding that fathers have higher scores than mothers on some items of the PRS-M is consistent with the superior performance of females compared with males on pragmatic, social, and language measures that has been reported in studies of typically developing children and adults. Preschool-aged girls performed better than age-matched boys on six out of eight

**Table 5** Comparison of Mothers and Fathers on the PRS-M - combined Autism and SLI group

|   | Mean ( <i>SD</i> ) |             | <i>t</i> | <i>df</i> | <i>P</i>      | Cohen's <i>d</i> | <i>r</i> |
|---|--------------------|-------------|----------|-----------|---------------|------------------|----------|
|   | Mothers            | Fathers     |          |           |               |                  |          |
| Emotional expressiveness and awareness of other | 0.86 (1.26)        | 1.54 (1.90) | 2.03     | 73        | .046          | 0.475            | .23      |
| Communicative performance                       | 0.46 (0.84)        | 1.34 (1.44) | 3.55     | 67        | <b>.001**</b> | 0.867            | .40      |
| PRS-M   | 2.94 (2.03)        | 5.11 (3.72) | 3.45     | 65        | <b>.001**</b> | 0.856            | .39      |

*SD* standard deviation, *PRS-M* modified pragmatic rating scale

\*\* Significant at the .005 level after Bonferroni correction

pragmatic language variables (Klecan-Aker & Swank, 1988). In a study of college students, males spent significantly less time engaged in mutual eye contact during a 10 min interview compared with females, independent of the interviewer's gender (Exline, Gray, & Schuette, 1965). Therefore, the fact that all interviewers in this study were female has likely not influenced the participant's eye contact significantly, although it may be difficult to apply findings from typically developing adults to our parents. Differences between males and females in communicative abilities are consistent with Baron-Cohen's theory that autism can be viewed as an exaggeration or extreme form of some aspects of maleness (Baron-Cohen, 2002; Baron-Cohen, Knickmeyer, & Belmonte, 2005).

Eye contact is the only item that differentiated fathers from mothers in both diagnostic groups combined and in each separately. In our sample, the gender difference was more striking among the SLI parents than among the autism parents, but with similarly poor eye contact between autism and SLI fathers. In contrast, autism mothers were more likely to have poor eye contact compared with SLI mothers. Poor eye contact is an important feature in autism. Focus on the person's mouth and body as well as on objects, rather than on the person's eyes, has been found among autistic males using a visual tracking method. This abnormality predicted social competence and was proposed to serve as a criterion for a social phenotype in autism (Klin, Jones, Schultz, Volkmar, & Cohen, 2002).

Our findings add to the evidence that autism and SLI share aspects of their etiology. Communication impairment has been observed in individuals with autism and their family members; in individuals with pragmatic language disorder (Bishop, 2000); in individuals with SLI; in siblings of SLI probands; and now in SLI parents. The inheritance of both autism and SLI are hypothesized to be oligogenic and genetically heterogeneous, which means that several genetic loci interact to cause and/or modify the disease phenotype and that not all the same loci operate in all cases. It seems likely that some genes associated with pragmatic language/communication deficits contribute to both

disorders. Indeed, linkage signals for both disorders point to the same region on chromosome 7q (CLSA, 2001; O'Brien, Zhang, Nishimura, Tomblin, & Murray, 2003) and possibly on 13q (CLSA, 2001).

Our findings support the hypothesis of a continuum of pathology between SLI and autism which ranges from SLI probands and their family members with only structural language abnormalities to SLI families with both structural and pragmatic impairments to probands with autism and their relatives with mainly pragmatic impairment and language—related difficulties. Further studies may substantiate the hypothesis that there is a broader SLI phenotype that is characterized by structural and pragmatic language deficits and partially overlaps with the broader autism phenotype. Nevertheless, there appear to be qualitative phenomenological differences between the hypothesized broader SLI phenotype and broader autism phenotype. Different language characteristics predicted group membership for autism and SLI parents. Not providing adequate references and making frequent reformulations were predictors for being an autism parent. Making grammatical errors and dominating the conversation predicted SLI group membership. Poor eye contact, while frequent among autism parents and SLI fathers, was less common in SLI mothers. Family studies using the visual tracking method would clarify whether the abnormal gaze behavior that leads to poor eye contact in autism probands is specific for autism and part of the broader autism phenotype. Furthermore, in future genetic studies the PRS-M may be applied as a quantitative phenotypic measure to assess autism and SLI family members and also other conditions with communication deficits such as fragile-X-syndrome and Prader-Willi-syndrome.

Some limitations of this study should be noted. It may be argued that the frequency of communication deficits is increased in the SLI parents because children with mainly pragmatic impairment were over-represented in our SLI sample. This is unlikely as our children with SLI had to have impaired structural language to enter the study. Furthermore, we recruited the children with autism and SLI separately, using resources that served

specifically SLI children to ascertain our cases. Our SLI sample should therefore be representative of a group with a balanced distribution of SLI subtypes. Further limitations of this study are due to power constraints. The principal component analysis was performed under the assumption that the autism and SLI parents are one sample, ignoring the diagnostic status. However, given the closely comparable scores of the two groups on individual items of the PRS-M, it is unlikely that a separate analysis would have yielded different subscales.

Furthermore, due to power constraints we were not able to include the DS parent control group in our analysis of the influence of gender on communication abilities.

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### Appendix: Pragmatic Rating Scale—Modified Version

#### 1. Grammatical Errors and Speech Complexity

Code noticeable grammatical errors (*Unacceptable expressions are* “He don’t know nothing” instead of “He does not know anything”, “Sometimes he sad” instead of “Sometimes he is sad”, “Yesterday is pretty out” instead of “Yesterday was pretty outside”, “She was wanting to go” instead of “She wanted to go”, “Do it quicker” instead of “Do it more quickly”, “The kids’es toys are broken” instead of “The kids’ toys are broken”. *Acceptable expressions are* “That’s real fun/easy” instead of “That’s really fun/easy”, “Her and her uncle went shopping” instead of “She and her uncle went shopping”, “There’s two parks” instead of “There are two parks”, “I got it good” instead of “I’ve got it good”.)

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | uses sentences in a largely grammatically correct fashion (must use some complex sentences with two or more clauses) |
|                          | 1 = | Some complex speech (Occasional utterances with two or more clauses, and two or fewer grammatical errors)            |
|                          | 2 = | some complex speech (occasional utterances with two or more clauses), but with more than two grammatical errors      |
|                          | 7 = | interviewer cannot judge the item  |

#### 2. Unusual Intonation

Code intonation abnormalities that are often seen in autism.

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | appropriately varying intonation  |
|                          | 1 = | slightly unusual intonation, slightly flat or exaggerated                 |
|                          | 2 = | little variation in pitch and tone, rather flat or exaggerated intonation |
|                          | 7 = | interviewer cannot judge the item   |

#### 3. Direct Verbal Communication of Own Emotional State

Code the participant’s spontaneous overt verbal expression of his/her own emotions (e.g. “*I feel content* just to read my book.”, “*It’s kind of calming* to watch the little boy playing.”). Code verbal expressions that contain the words “tired”, “love”, “enjoy”, “fun”. Do not code comments that only use the words “like/dislike” to express an emotional state.

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | makes spontaneous direct comments about own emotional state on at least two occasions |
|                          | 1 = | makes spontaneous direct comments about own emotional state on only one occasion      |
|                          | 2 = | never makes spontaneous direct comments about own emotional state                     |
|                          | 7 = | interviewer cannot judge the item   |

#### 4. Indirect Verbal Communication of Own Emotional State

Code spontaneous, indirect verbal expression of own emotional state (e.g. “Things haven’t been the same since he passed away.”)

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | makes spontaneous indirect comments about own emotional state on at least two occasions |
|                          | 1 = | makes spontaneous indirect comments about own emotional state on only one occasion      |
|                          | 2 = | never makes spontaneous indirect comments about own emotional state                     |
|                          | 7 = | interviewer cannot judge the item   |

5. **Confusing Accounts due to Missing Information**

Code if the examiner has difficulty following an account due to the subject's failure to reference pronouns or provide sufficient information necessary for clarity (e.g. "John and Brad went to the station He was very hungry." This is confusing as one cannot tell which boy, John or Brad, "he" is supposed to represent). Unexpected topic shifts may also be included here.

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | presents account in a clear and organized fashion  |
|                          | 1 = | leaves out information or fails to reference pronouns, so that content needs to be clarified on one occasion |
|                          | 2 = | frequently leaves out information or fails to reference pronouns, so that content needs to be clarified      |
|                          | 7 = | interviewer cannot judge the item  |

6. **Dominating Conversation**

Code if the subject tends to dominate the conversation either by interrupting or lecturing the examiner such that the experience resembles a monologue rather than a conversation.

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | does not dominate the conversation and does not frequently interrupt   |
|                          | 1 = | interrupts and/or employs lecturing style with the examiner, but reciprocity of conversation is maintained             |
|                          | 2 = | frequently interrupts and/or employs lecturing style with the examiner, and reciprocity of conversation is interrupted |
|                          | 7 = | interviewer cannot judge the item  |

7. **Descriptive Gestures**

Code descriptive gestures enact or represent an object or event (e.g. describing the size of a fish caught using your hands spread at a distance, describing a rocket shouting up by a quick, upward motion of the hands).

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | uses at least 3 spontaneous descriptive gestures, these gestures must be communicative |
|                          | 1 = | uses less than 3 spontaneous descriptive gestures                                      |
|                          | 2 = | never uses spontaneous descriptive gestures  |
|                          | 7 = | interviewer cannot judge the item  |

8. **Emphatic or Emotional Gestures**

Code emphatic gestures and emotional gestures that accompany speech. Emphatic gestures are hand movements used to emphasize a statement but without particular descriptive quality, or emotional gestures are reflexive responses to specific emotions (e.g. hands to mouth or hands up for "wow").

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | uses emphatic and/or emotional gestures (>5 occurrences)                                 |
|                          | 1 = | uses some emphatic and/or emotional gestures, but limited in frequency (< 5 occurrences) |
|                          | 2 = | never uses emphatic or emotional gestures  |
|                          | 7 = | interviewer cannot judge the item  |

9. **Overly Detailed**

Code when subject provides minute details about an event.

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | provides adequate, appropriate detail                                 |
|                          | 1 = | provides details that are unnecessary or irrelevant on one occasion   |
|                          | 2 = | frequently includes minute details that are unnecessary or irrelevant |
|                          | 7 = | interviewer cannot judge the item                                     |

10. **Failure to Reference Terminology**

Code the use of technical jargon that a lay person would not understand, such as referring to stamps as “commodities” or the Autism Society of America as “ASA” without providing the appropriate reference information.

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | appropriately references terminology                               |
|                          | 1 = | fails to reference terminology on one occasion                     |
|                          | 2 = | frequently fails to reference terminology (more than one occasion) |
|                          | 7 = | interviewer cannot judge the item                                  |

11. **Reformulation**

Code when the subject has trouble expressing or formulating an idea as indicated by frequently rephrasing the idea before it is fully expressed.

|                          |     |  |
|--------------------------|-----|--|
| <input type="checkbox"/> | 0 = | reformulates rarely; this does not interfere with the rater’s ability to follow the train of thought being expressed |
|                          | 1 = | reformulates frequently enough to be noticed, but does not interfere with the train of thought being expressed       |
|                          | 2 = | reformulates frequently which interferes with the train of thought   |
|                          | 7 = | interviewer cannot judge the item  |

12. **Mispronunciation**

Code here difficulties pronouncing specific words that are not due to the person’s difficulty articulating specific sounds. (For example, subject pronounces silent letters in words such as the “p” in “receipt” or the “e” in “comfortable”. Do not count words that are pronounced differently due to dialect.)

|                          |     |                                   |
|--------------------------|-----|-----------------------------------|
| <input type="checkbox"/> | 0 = | no mispronunciations              |
|                          | 1 = | mispronounces one word            |
|                          | 2 = | mispronounces more than one word  |
|                          | 7 = | interviewer cannot judge the item |

13. **Unusual Eye Contact**

Code gaze that is avoidant and/or limited in appropriateness.

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | does not have unusual eye contact   |
|                          | 1 = | avoids eye contact for a large part of the interview (not just in the beginning) or makes inappropriate eye contact (e.g. staring) once |
|                          | 2 = | avoids eye contact almost throughout the interview or makes inappropriate eye contact (e.g. staring) more than once                     |
|                          | 7 = | interviewer cannot judge the item   |

14. **Range of Facial Expression**

Code the participant’s range of emotions and non-verbal communications displayed through facial expression.

|                          |     |   |
|--------------------------|-----|---|
| <input type="checkbox"/> | 0 = | communicates at least 4 emotions or non-verbal communications through facial expression |
|                          | 1 = | communicates at least 1 emotion or non-verbal communication through facial expression   |
|                          | 2 = | never communicates through facial expression  |
|                          | 7 = | interviewer cannot judge the item   |

15. **Empathy/ Comments on Others Emotions**

Code the subject's communication of his/her understanding for the feelings of other people. Include any shared emotion with the examiner that the subject may express (e.g. "Boy, you work hard all day. No wonder you're exhausted"). If the subject only shows empathy in indirect ways, e.g. "We like to go hiking", code as "1". However, "He likes to go hiking" expresses a direct understanding of others' emotions.



- 0 = communicates clear understanding and shared emotion with others or the examiner on at least two occasions  
 1 = communicates clear understanding and shared emotion with others or the examiner one occasion  
 2 = never communicates emotional understanding or shared emotion  
 7 = interviewer cannot judge the item

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