Aus der Klinik für Kinder- und Jugendpsychiatrie
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DISSERTATION

Kommunikative Kompetenz bei Eltern von Kindern mit Autismus, Spezifischer Sprachentwicklungsstörung und Down Syndrom

Communicative Competence in parents of children with autism, Specific Language Impairment and Down Syndrome

zur Erlangung des akademischen Grades
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TO THE CHILDREN WHO PARTICIPATED IN THIS STUDY

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INTRODUCTION

Definition of autism

Autism is a developmental disorder that presents with severe qualitative impairment in social interactions and communication and with restricted, repetitive, or stereotyped behaviors and interests before the age of 3 (APA, 2000; WHO, 2004).

Communication deficits are a hallmark of autism. Communication deficits include delayed and abnormal speech and language development or lack thereof and impairment in pragmatic language.

Language impairment in autism

Most children with autism begin to speak late and develop speech and language at a significantly slower rate than controls (LeCouteur et al., 1989). According to one estimate, only about 50% of individuals with autism develop useful speech (Lord & Paul, 1997). When language does develop, comprehension as well as expression is usually affected (Paul et al., 1983). Semantic impairments vary from milder difficulties in understanding word play in metaphor, irony and jokes (Happe, 1994) to more severe problems, like using words and phrases in narrow, context-bound or idiosyncratic ways (Eskes et al., 1990)**.** Development of syntax proceeds at a slower pace than in normal children and is more likely related to the autistic child's developmental level then its chronological age (Tager-Flusberg, 1981). In a language-impaired subgroup of children with autism, deficits in higher-order syntax and grammar were found to be similar to those seen in children with Specific Language Impairment (SLI) (Kjelgaard & Tager-Flusberg, 2001).

Pragmatic language, the use of language appropriate to social context (Bates, 1976), is impaired even in children with good language skills, including children with Asperger's Syndrome. In conversations and play situations, verbal children with autism initiate conversations less often, give fewer responses to questions, take fewer turns, chat less and are less able to maintain a topic of conversation compared with children with Down syndrome, SLI and normal development (Bartak et al., 1975; Eales, 1993; Loveland et al., 1988; Tager-Flusberg & Anderson, 1991). Another striking feature of autistic children's use of language is their reversal of pronouns - referring to themselves as "you" and their conversational partner as "I" (Baltaxe, 1977; Bartak et al., 1975). Although reversing personal pronouns is not unique to autism, it does occur more

frequently in this group than in any other (Lee et al., 1994). Children with autism also often fail to adapt their account to the conversational context; for example 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 et al., 1990; Tager-Flusberg, 2000).

Social impairment in autism

Impairment in social interaction is another central feature of autism that partly overlaps with deficits in pragmatic language. Children with autism have been observed to show less facial expression and are less likely to share enjoyment, have eye contact and use gestures less often than mentally handicapped children with similar verbal IQ (Lord et al., 1989).

Etiology of autism

Autism is etiologically heterogeneous. It has been associated with congenital rubella (Chess, 1977), tuberous sclerosis (Smalley, 1998), and fragile X syndrome (Gillberg & Wahlstroem, 1985). Twin and family studies and more recently studies of molecular genetics imply a strong genetic basis for the disorder. Twin studies found a large disparity between monozygotic and dizygotic pairs (Folstein & Rutter, 1977), with 60% vs. 5% of the siblings being affected, respectively (Bailey et al., 1995). This large disparity suggests a synergistic interaction of several genes rather than a single-gene Mendelian pattern of inheritance. Family studies found that the rate of autism in siblings of autistic individuals reached 3% (Bolton et al., 1994). This is a 30- to 100-fold increase in relative risk, depending on the assumption about the base rate in the general population.

Broader autism phenotype – a milder phenotypic expression in family members

Non-affected family members of autistic individuals show mild impairments that are qualitatively similar to those seen in narrowly defined autism (Bolton et al., 1994; LeCouteur et al., 1996). These include milder deficits in social interaction and communication as well as repetitive and stereotypic behaviors. Most commonly reported are social or communication impairments or a combination of both, whereas repetitive and stereotyped behaviors occur at lower rates among biological autism relatives (Bailey et al., 1998; Bolton et al., 1994; Szatmari et al., 2000). Certain personality traits,

such as rigidity and aloofness were also found more commonly in parents from families with at least two autistic children than in parents of children with Down syndrome (Piven et al., 1997). It is thought that these familial characteristics represent a milder phenotypic expression of, or genetic liability to, autism and autism spectrum disorders. They have been subsumed under the term “broader autism phenotype” (LeCouteur et al., 1996).

Mild communication impairments are common among relatives of autistic individuals. Several studies have reported language delay in first degree relatives of children with autism (DeLong & Dwyer, 1988; Piven et al., 1990), some of them with significantly increased incidence compared to relatives of controls (August et al., 1981) (Bolton et al., 1994; Folstein et al., 1999). Bolton et al. asked about childhood histories of 332 first-degree autism relatives and 136 first-degree relatives of probands with Down syndrome. Autism relatives significantly more often reported early communication deficits such as language delay defined as no words by the age of 2 years or no phrases by the age of 33 months, reading retardation, articulation disorder and spelling difficulties in childhood (Bolton et al., 1994). Folstein et al studied 162 autism parents and 73 parents of children with Down syndrome and found that autism parents significantly more likely reported a history of late onset of phrase speech, articulation deficits and early difficulties with reading and spelling (Folstein et al., 1999). Four studies were unable to detect significant differences between autism and control relatives. Boutin and colleagues compared 156 first-degree relatives of 46 children with autism or pervasive developmental disorder (PDD) with 55 first-degree relatives of children with mental retardation using the family history method (Boutin et al., 1997). 3.2 % of the autism relatives and none of the relatives of the mentally retarded patients had reportedly language delay, a difference that was not significant. A larger number of relatives may have resulted in a statistically significant difference. Another study compared relatives of 35 children with autism with relatives of 42 children with attentional, motor or perceptional deficits and relatives of 51 normally developing children and found no differences in the incidence of language impairment between these groups (Gillberg et al., 1992). Twelve of the autistic children (34%) had a significant medical condition, such as fragile X syndrome, trisomy 13, tuberous sclerosis and hydrocephalus. This sample of autistic children appears to have included a high percentage of non-idiopathic cases, and may therefore not be best suited for genetic studies (Folstein & Rosen-Sheidley, 2001). Szatmari et al. studied 52 families of

children with PDD and 33 families of children with Down syndrome or very low birth weight (Szatmari et al., 1995). By history, there were no differences between the groups in regard to parental speech delay, reading, writing or spelling problems. In a similar study that was diagnostically more thoroughly designed, Szatmari et al. found a significantly higher incidence of communication deficits in biological relatives compared with non-biological relatives in autism families (Szatmari et al., 2000). Overall, the evidence is stronger for an increased risk of language impairment in autism families.

Milder social and pragmatic deficits are part of the broader phenotype often found in relatives of individuals with autism. Using informant-based family history data, social-pragmatic deficits are found more frequently among autism relatives than Down syndrome relatives and among biological than non-biological relatives of individuals with pervasive developmental disorder, an autism spectrum disorder (Bolton et al., 1994) (Szatmari et al., 2000). Three studies have directly examined communication in autism parents. Wolff, Narayan and Moyes 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, tended not to smile much and to regard others with suspiciousness (Wolff et al., 1988). Landa and colleagues developed the Pragmatic Rating Scale (PRS), an interviewer-based instrument that assesses the more subtle deficits in pragmatic language, to compare 43 autism parents and 21 control parents of both children with Down syndrome and normal children (Landa et al., 1992). The total PRS scores were significantly higher, reflecting greater impairment, for the autism parents than the control parents. Piven and colleagues replicated these results in families in which at least 2 children carried the diagnosis of autism also using the PRS (Piven et al., 1997). They compared 19 pragmatic language and 6 speech items of the original PRS in 48 autism parents and 60 Down syndrome parents. Composite scores for both, the pragmatic language and the speech items, were significantly higher among autism parents than parents of children with Down syndrome.

Definition of Specific Language Impairment (SLI)

Specific Language Impairment (SLI) is a developmental disorder. It is defined by low language test scores in the absence of another language-related deficit or disorder

such as mental retardation, hearing loss, motor disorder, neurological disorder and overt impairment in social interaction (Plante, 1998).

The core feature of SLI is delay in language development. SLI infants and toddlers experience a significant delay in acquisition of first words and word combinations (Leonard, 1998). During the school years, lexical learning remains impaired (Oetting et al., 1995) and word-finding difficulties occur (McGregor & Leonard, 1995). In the English language, naming errors include semantic ("shoes" for "pants") and phonological ("wrangler" for "ankle") substitutions (McGregor, 1994). Children with SLI form less complex sentences than normally developing children. Their syntactic structure consists of basic categories (subject-verb-object), and they often omit a category (Leonard, 1998). Errors in grammatical morphology are made frequently. Articles, pronouns, regular past tense of verbs (-ed) and third person singular (-s) as well as the plural of nouns (-s) are omitted, replaced or mistakenly used (Johnston & Kamhi, 1984). Phonological development is delayed as well. Children with SLI acquire phonemes in the same order as normal children do, but at a slower rate (Leonard, 1998). Children with SLI are at high risk of developing a reading disability or have difficulty in reading comprehension (Tallal et al., 1988). Overall, language production or expression is more affected in SLI than language comprehension or reception (Leonard, 1998).

Communication and social deficits in Specific Language Impairment

It has been thought that in SLI, mainly structural language is impaired, while a hallmark of autism is the deficit in the use of language for communication (Bishop, 2000; Bishop et al., 1995). However, many children with SLI show some pragmatic language and social deficits. Children with SLI use speech acts such as naming, requesting, and thanking less often than age-matched controls, although they are able to express requestive and declarative functions through gestures (Snyder, 1978). Speech acts most often resemble those of younger control children (Prinz, 1982). In judging which kind of requests would be most appropriate to certain conversational contexts, children with SLI did most poorly in comparison to age-matched controls and language-level matched controls (Messik & Newhoff, 1979). Children with SLI show less participation in conversations than same-age peers. They are less likely to initiate a conversation, and often restrict their answers to acknowledging the prior message or indicating that they understood (Siegel et al., 1979). Moreover, children with SLI have great difficulty

entering ongoing conversations among groups of normal controls (Craig & Washington, 1993). At the same time, they show more initiative in conversations with children of similar language level (Fey et al., 1981). In answering requests for clarification, children with SLI are less likely to revise and clarify messages than same-age peers (Brinton & Fujiki, 1982; Prutting & Kirchner, 1987). Furthermore, children with SLI have difficulty understanding language beyond its literal meaning such as in metaphors (Nippold & Fey, 1983). Adults with SLI have been reported to have prosodic oddities, problems sustaining a conversation and difficulties reporting events (Mawhood *et al.*, 2000).

Individuals with SLI are heterogeneous in regard to their pragmatic skills (Bishop & Adams, 1989; Bishop et al., 1995; Fey & Leonard, 1983). The diagnostic category "semantic-pragmatic disorder"/"pragmatic language impairment" has been introduced to describe a subgroup of children with SLI who have early language delay with fluent expressive language in middle childhood and significant deficits in their use of language in social context (Bishop & Norbury, 2002a; Bishop & Rosenbloom, 1987; Conti-Ramsden & Botting, 1999; Rapin & Allen, 1983). The term pragmatic language impairment (PLI) will be used here, since semantic errors can be but are not necessarily associated with the pragmatic impairments in this group (Bishop, 1998). Among children with SLI, 40% of those found to be pragmatically impaired were not distinguishable in their pragmatic skills from children with an autism spectrum disorder (Bishop & Norbury, 2002a).

Family studies in Specific Language Impairment

Significant familial aggregation of language impairment and findings from molecular genetic studies indicate that SLI has a strong genetic component. While the prevalence among English-speaking school children is estimated to be 2%-7% (Tomblin et al., 1997), family studies report prevalence rates of 24%-78% for SLI family members compared with 3%-46% in control family members (Stromswold, 1998). Twin studies found consistently higher concordance rates for monozygotic compared with dizygotic twins (Bishop et al., 1995; Lewis & Thompson, 1992; Tomblin & Buckwalter, 1998). One family study has revealed a higher rate of autism in siblings of SLI probands than in the general population (Tomblin et al., 2003). Despite evidence of impairment in pragmatic language and social interaction in children with SLI, and a higher incidence of autism among SLI relatives, pragmatic deficits have not yet been studied in SLI relatives.

Genetic linkage studies in autism and Specific Language Impairment

With the development of molecular genetic techniques and improved characterization of both autism and SLI, genetic linkage studies have been employed in the search for autism and SLI susceptibility loci. For autism, only a few chromosomal regions detected through linkage analyses seem to be supported by independent studies (Folstein & Rosen-Sheidley, 2001). The most consistent finding has been linkage to chromosome 7q (Alarcon et al., 2002; Auranen et al., 2002; Barrett et al., 1999; CLSA, 2001; IMGSAC, 2001a, 2001b; Shao et al., 2002). One study also points to a marker on chromosome 13q (CLSA, 2001). To date, no study has looked at a phenotype of impaired communication as a trait marker for linkage analysis. This is in part due to the difficulty of defining and quantifying such a phenotype.

The first gene found to be implicated in speech and language development, FOXP2, was discovered in a large family suffering from a severe autosomal dominant speech and language disorder; it is located at the chromosomal region 7q31 (Fisher et al., 1998; Lai et al., 2000). In genetic linkage studies of SLI, a strong association was found between the language phenotype and markers on 7q31, adjacent to or within FOXP2 (Newbury et al., 2002; O'Brien et al., 2003). Of the two published genome wide scans for SLI, one study detected a significant LOD score on chromosome 13q21 (Bartlett et al., 2002).

Defining communicative competence

Communicative competence covers a wide range of communication skills that include elements of social communication, social interaction, pragmatic language, speech and expressive fluency. Social communication, social interaction and pragmatic language are related and partly 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 et al., 1998). Social interaction includes social communication, and in addition, specifies areas of potential weakness such as age-appropriate development of peer relationships, spontaneous sharing of emotions and interests, and social and emotional reciprocity (DSM IV; APA, 2000). 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 (Baltaxe, 1977; Bishop, 1998; Bishop & Adams, 1989; Craig & Washington, 1993; Landa et al., 1992; McTear, 1985; Prutting & Kirchner, 1987; Roth & Spekman, 1984). In addition, non-verbal communicative behaviors such as eye contact, facial expressions, gestures and body posture (Bishop, 1998; Prutting & Kirchner, 1987) and paralinguistic aspects of speech such as prosody, fluency and intelligibility (Prutting & Kirchner, 1987) are subsumed under a broader definition of pragmatics.

DERIVING THE TASK

The developmental disorders autism and SLI overlap in their phenomenology. Both, individuals with autism and individuals with SLI, show impairments in their communication. Delay in language acquisition is a hallmark of both disorders (LeCouteur et al., 1989; Leonard, 1998; Lord & Paul, 1997). Impairment in structural language defines SLI (Stark & Tallal, 1981), and can be part of autism (Eskes et al., 1990; Happe, 1994; Paul et al., 1983; Tager-Flusberg, 1981). Higher syntactic and grammatical errors were found to be similar in autism and SLI children (Kjelgaard & Tager-Flusberg, 2001). Impairment in pragmatic language and social interaction appears to occur along a continuum, with children with autism and pragmatic language disorder, a subgroup of SLI, being most severely affected (Bishop & Norbury, 2002a; Bishop & Rosenbloom, 1987; Conti-Ramsden & Botting, 1999; Rapin & Allen, 1983). Children with SLI other than pragmatic language disorder are mildly to moderately impaired in this domain (Bishop & Adams, 1989).

Given the considerable phenomenological similarities in communication impairment seen in individuals with autism and SLI, the question arises whether these similarities may also be found in their family members. Autism families are known to have a higher incidence of communication deficits, including deficits in language acquisition, structural and pragmatic language and social interaction compared with control families (Bolton et al., 1994; Folstein et al., 1999; Landa et al., 1992; Piven et al., 1997; Szatmari et al., 2000). In SLI families, language delay and impairment of structural language are highly prevalent (Tallal et al., 2001). However, nothing is known about the level of communicative competence in these families.

Therefore, the goal of this project was to assess and compare a broad range of verbal and non-verbal communication skills in parents of autistic children and parents of SLI children. The parents were matched for verbal IQ to eliminate potential between-group differences in verbal IQ as a confounding factor for potential differences in communication skills.

Existing measures of pragmatics and social communication either did not cover all the aspects of communication that are potentially impaired in autism or SLI or they were used in a different 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). Therefore, the PRS was modified (PRS-M) to include additional aspects of non-verbal communication and formal language.

The PRS-M was used to compare communication impairments in conversational speech of parents of children with autism, SLI and Down syndrome (DS). Parents of children with Down syndrome were chosen as control group because they do not carry an increased genetic liability for communication disorders and to control for the effect of caring for a handicapped child.

Hypotheses

At the beginning of this study, the following hypotheses were formulated. First, parents of children with autism and parents of children with SLI have communication impairments when compared with parents of children with Down syndrome. Second, the communication impairment measured using the PRS-M is greater in parents of children with autism than in parents of children with SLI. Third, there will be differences in the pattern of communication deficits between the autism and the SLI parents.

METHODS

Sample

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 (Tufts-NEMC) and the University of Iowa. SLI families from the Iowa site were members of a longitudinal cohort (Tomblin et al., 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. Sampling bias toward ascertaining SLI families who were concerned that their child may have symptoms of autism was avoided by telling SLI families only that the project was about language and reading in family members of children with SLI. The autism recruitment was carried out 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.

The Down syndrome 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 had been the same (Piven et al., 1997).

For this investigation of parents' communicative competence, 47 parents of autistic probands ("autism parents") and 47 parents of SLI probands ("SLI parents") who matched pair-wise on verbal IQ were selected. To maximize the number of matched pairs, parents were 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 Down syndrome ("DS parents") were not matched on verbal IQ. The 21 DS parents came from 12 families, each of which 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 et al., 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 (CELF-III) (Semel et al., 1995) or at or below the 9th percentile on the Nonword Repetition subtest of the Comprehensive Test of Phonological Processing (Wagner et al., 1999). The Nonword repetition task has been shown to be a sensitive and specific psycholinguistic marker for SLI (Conti-Ramsden et al., 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 Down syndrome 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. Families were also excluded when 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.

Data analysis

For comparison of autism, SLI and DS parents, the paired t-test was used for autism - SLI parent pairs matched by verbal IQ for continuous variables. The independent samples t-test was applied for non-matched comparisons. The chi-square statistic was utilized to compare autism parents with SLI parents and fathers with mothers and autism and SLI parents with DS parents on the individual items of the

PRS-M. The gender distribution between the diagnostic groups was also compared using the chi-square statistic for both parents and probands. The Bonferroni correction was used for multiple comparisons. Reliability procedures included the kappa statistic for the individual items of the PRS-M and the intra-class correlation coefficient for the total PRS-M score and its subscales. To test the consistency of the PRS-M as a scale, the intra-class correlation coefficient was used. The subscales of the PRS-M were derived using the SAS-procedure VARCLUS. The Pearson correlation coefficient was used to validate the subscales. To predict group membership for autism and SLI parents, individual PRS-M items were entered into logistic regression analyses.

Proband and parent characteristics

Table 1 presents the characteristics for the probands and parents. The probands differed only in their gender distribution: 85% of the probands with autism, 59% of the probands with SLI, and 46% of the probands with DS were male (χ^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 level of education than the DS parents (t (66) =2.25, p=0.028). 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.

Table 1: Demographic characteristics of the probands and parents

	Mean (SD)		
	Autism	SLI	DS
	(Proband N=27)	(Proband N=29)	(Proband N=12)
	(Parent N=47)	(Parent N=47)	(Parent N=21)
PROBANDS			
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)	
PARENTS			
Age	40.89 (4.50)	39.49 (5.38)	39.20 (7.63)
Education*	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)

*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.

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-IIIR); the parallel abbreviated WISC-III was administered to the probands (Wechsler, 1991a, 1991b).

A modified version of the investigator-based Family History Interview of Developmental Disorders of Cognition and Social Functioning (short FHI) was used to assess traits characteristic of autism in the autism and SLI parent groups (1991,

developed by Rutter and Folstein; 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, information was obtained directly from each parent when possible, or in some cases from the spouse.

Language sample

A 20-minute 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, for example by asking the participant to describe her occupation and hobbies. This was 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

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 6 speech items were later published and shown to be typical in autism parents (Piven et al., 1997). Items from the original unpublished and published PRS were incorporated in the PRS-M. Some items and codes were refined by making them more specific and thus easier to code reliably. Several items that were not mutually exclusive were combined. "Verbal emotional expressions" and "grammatical errors" were added, since they have been found to be abnormal in children with autism (Lord et al., 1989; Pearlman-Avnion & 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 0-30. The 15-item PRS-M is attached.

Psychometrics

Inter-rater reliability: Two raters (Tilla Ruser and Sara Putnam, research technician) blindly and independently watched and rated 47 videos that were randomly selected from the larger sample. Formal inter-rater reliability for these 47 cases was calculated. Items for which there was disagreement between the two raters were resolved by discussing each individual rating. In 9 of the 94 cases in this study, blind ratings could not be made because the parent mentioned the proband's diagnosis at some point during the language sample. Later this was avoided by having identifying information removed through prescreening by a third person.

The intra-class correlation coefficient for the sum of all 15 PRS-M items was 0.72, indicating an overall good reliability. Inter-rater reliability for the subscales was good as well, with intra-class correlation coefficients ranging from 0.74 to 0.85 (Table 2). The kappa values for the individual items ranged from 0.31 – 0.80 with the percent agreement ranging from 66% to 96% (Table 3). The two items 'Indirect verbal emotional response' and 'Mispronunciation' were only seldom endorsed with positive ratings in 3 to 7 of the 47 reliability cases. Therefore, the kappa values of these items were below 0.30. Because the inter-rater agreement was at least 78%, these items were retained.

Table 2: Reliability for the four subscales (intra-class correlation coefficient)

Subscales	**Intra-class correlation coefficient**
Emotional Expressiveness and Awareness of the Other	0.85
Communicative Performance	0.83
Over-talkativeness	0.74
Language	0.79

Table 3: Inter-rater reliability for 47 cases by item

PRS-M item	Kappa	Percent agreement
Grammatical errors	0.65	83
Prosody	0.44	74
Direct verbal emotional expression	0.50	79
Indirect verbal emotional expression	0.19	85
Confusing account	0.59	83
Dominating	0.45	77
Descriptive gestures	0.60	85
Emphatic gestures	0.38	89
Overly detailed	0.55	91
Failure to reference	0.66	87
Reformulation	0.61	80
Mispronunciation	0.06	78
Eye contact	0.60	85
Facial expressions	0.39	78
Empathy	0.31	79

Scale construction: The intra-class correlation coefficient of 15 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, the 15 items were entered 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, only variables that were correlated with the other variables in their own cluster at R>.30 or R^2>.09 were included. To improve item discriminant validity, variables were only included 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 (Table 4). 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.

Table 4: Subscales of the Pragmatic Rating Scale - Modified

1: Emotional Expressiveness and Awareness of Other	***R^2 with own subscale***	**2: Communicative Performance**	***R^2 with own subscale***
Direct emotional verbal expression	.54	Prosody	.38
Indirect emotional verbal expression	.51	Descriptive Gestures	.61
Failure to reference	.25	Emphatic Gestures	.67
Facial expressions	.28	Eye contact	.11
Empathy	.59		

3: Over-talkativeness		**4: Language**	
Dominating Conversation	.82	Grammatical errors	.33
Overly detailed	.82	Confusing accounts	.51
		Reformulation	.46
		Mispronunciation	.36

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, the data obtained from Family History Interview for the autism and SLI parents were chosen to validate the subscales of the PRS-M. Single items and three of the factors derived from the FHI by Zwaigenbaum and colleagues were used in this study (Zwaigenbaum et al., 2000). Zwaigenbaum and colleagues had obtained family history data for 1327 biological and 326 non-biological relatives. A factor analysis revealed seven factors, all of which occurred with an increased relative risk in biological relatives compared to non-biological relatives. The factors relevant to this study appeared to be academic learning problems, social-pragmatic impairment, odd behavior and speech delay. Since Zwaigenbaum had included in his analysis all items from the original FHI as well as additional items covering characteristics of high functioning autism/Aspergers syndrome, and an abbreviated version was used in the current study, the factors were modified slightly. Furthermore, many parents did not know their own or their spouses' age at which they spoke their first single words and phrases and whether they had speech delay beyond 24 months. Therefore, speech delay could not be included in this validation analysis.

Predictions were made as to which FHI factors and items would correlate with each subscale, as shown in Table 5. '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'. It was further assumed that 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 were 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 5.

Family history information was missing for two autism and one SLI parent. Therefore, only 91 cases were used in the validation analyses.

Table 5: Validation of the Pragmatic Rating Scale – Modified

	Predicted Correlation	Significant Correlation
Emotional Expressiveness and Awareness of the Other	Social-pragmatic impairment** Friendships in childhood Friendships in adulthood Adult conversation	Friendships in adulthood r = .21; p = .046
Communicative Performance	Social-pragmatic impairment** Shyness Adult conversation	Adult conversation r = .31; p = .002 Social-pragmatic impairment r = .21; p = .041
Over- talkativeness	Friendships in childhood Friendships in adulthood Adult conversation	Friendships in childhood r = .21; p = .047
Language	Academic learning problems*** Difficulties with reading Difficulties with writing	Difficulties with reading r = .37; p < .001 Academic learning problems r = .30; p = .003

**Social-pragmatic impairment = lack of affection, social play in childhood, adult conversation

***Academic learning problems = difficulties with reading, spelling or mathematics

RESULTS

Total PRS-M score and subscale scores

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 Figures 1 and 2, 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, see Figure 2). Parents who scored ≥ 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. A total PRS-M score of 7 was used as the cut-off to define a more severely impaired subgroup.

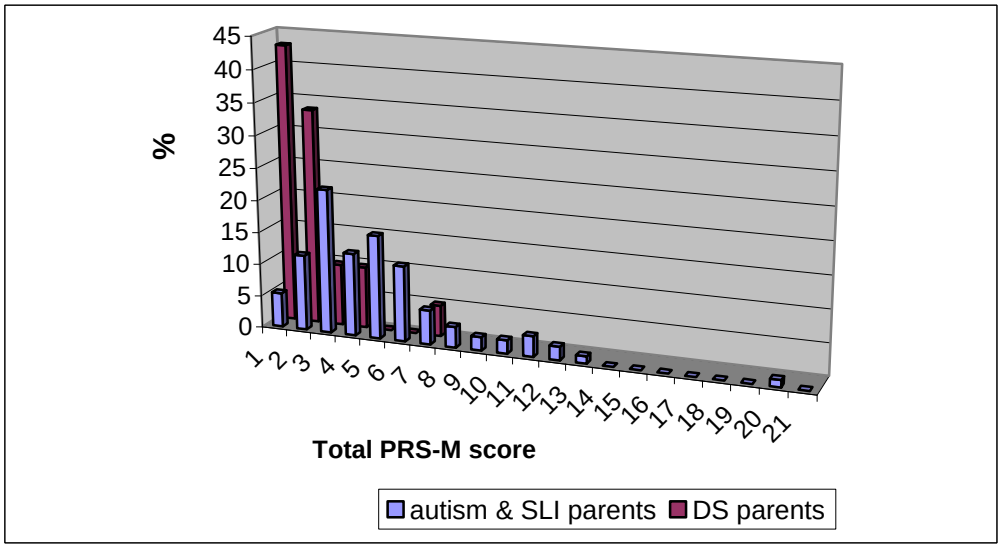

Figure 1: Distribution of the total PRS-M scores

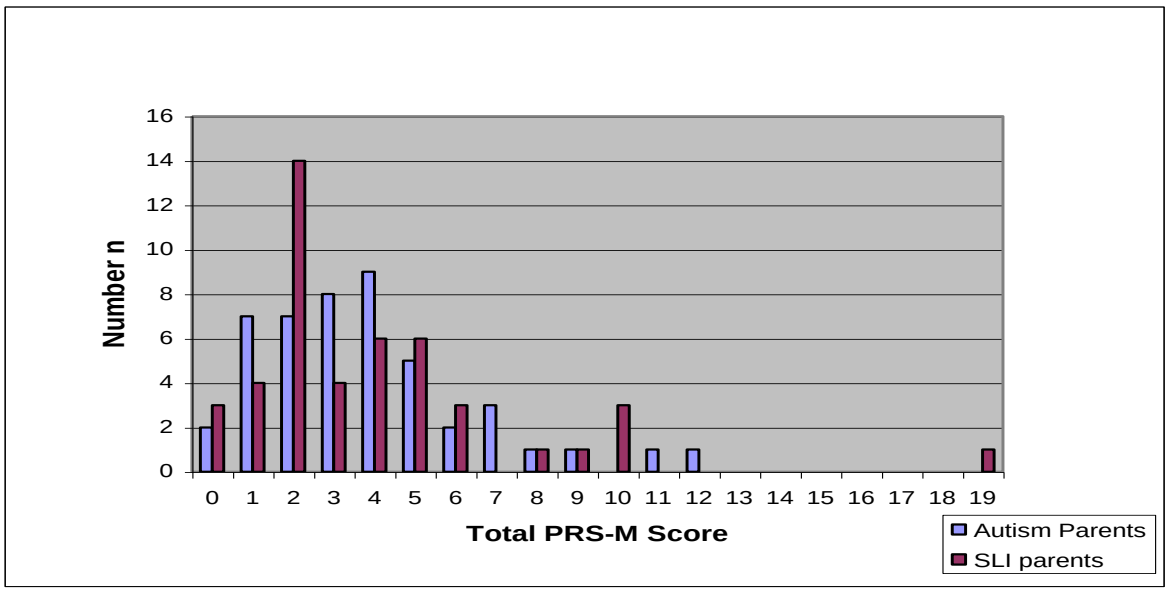

Figure 2: Distribution of the total PRS-M score for autism and SLI parents

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 3 of the 4 subscales and for the total PRS-M score as shown in Table 6. Autism parents scored also significantly higher than DS parents on the individual items reformulation (χ^2 (1) = 10.97, p = .0008) and confusing accounts (χ^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 (Figure 3).

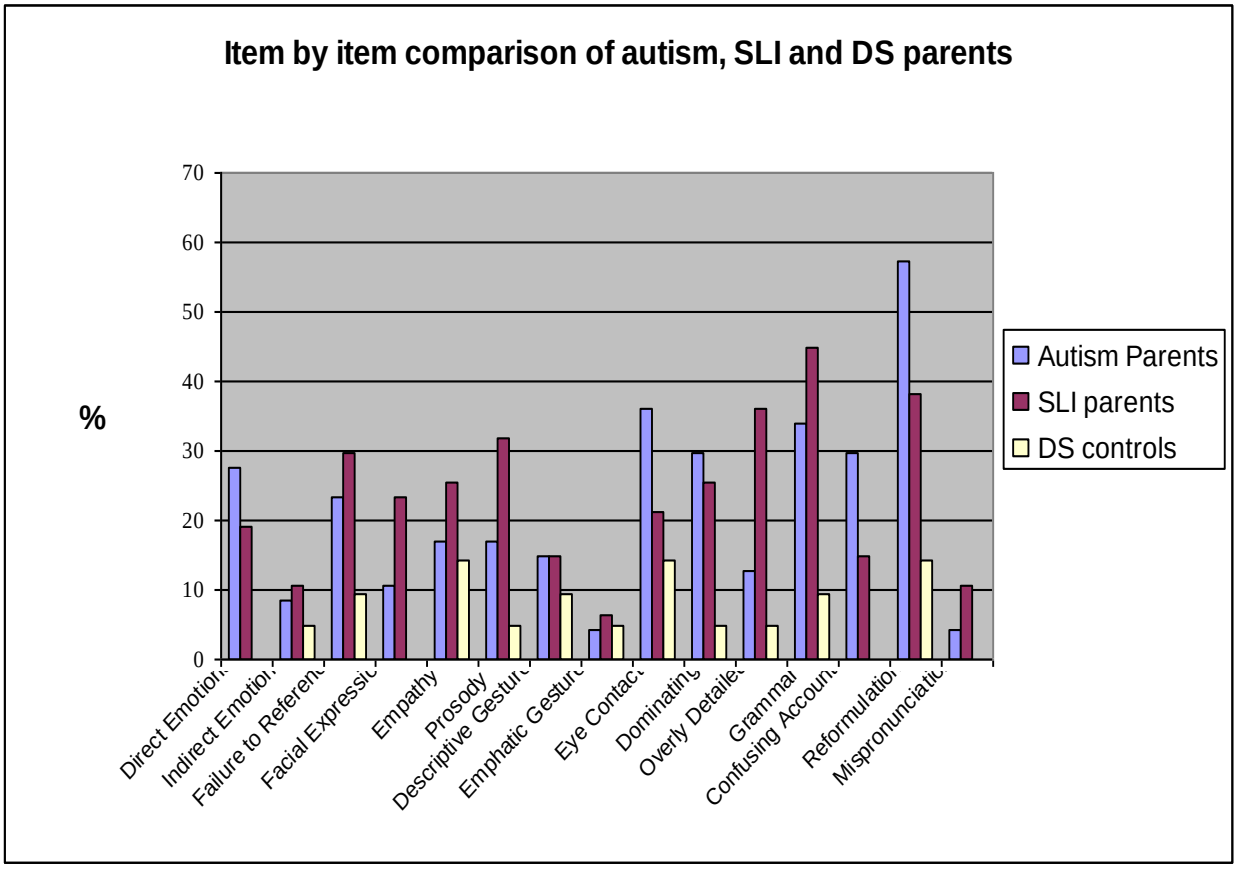

Figure 3: Item by item comparison of 47 autism, 47 SLI and 21 DS parents

Table 6: Paired and independent t-tests for the 4 subscales and the PRS-M score by diagnosis

				AU vs. SLI parents*			AU vs. DS parents**			SLI vs. DS parents**		
	Autism Parents	SLI Parents	DS Parents	t-value	p	Cohen's d	t-value	p	Cohen's d	t-value	p	Cohen's d
	Mean (SD)	Mean (SD)	Mean (SD)									
Emotional Expressiveness and Awareness of the Other	.97 (1.34)	1.38 (1.85)	.29 (.56)	1.17	.247	.25	3.00	**.004+**	.66	3.71	**<.001++**	.80
Communicative Performance	.83 (1.18)	.91 (1.30)	.38 (.97)	.34	.734	.06	1.52	.133	.42	1.68	.097	.46
Over-talkativeness	.53 (0.97)	.53 (1.01)	.09 (.30)	.0	1.000	.00	2.79	**.007+**	.61	2.69	**.009+**	.59
Language	1.47 (1.40)	1.28 (1.36)	.33 (.66)	.63	.529	.14	4.55	**<.001++**	1.04	3.85	**<.001++**	.89
Total PRS-M Score	3.81 (2.65)	4.10 (3.56)	1.09 (1.48)	.44	.660	.09	5.39	**<.001++**	1.27	4.92	**<.001++**	1.10

*Paired T-Test, ** Independent T-Test, + p<.05 after Bonferroni correction, ++ p<.005 after Bonferroni correction

When all individual PRS-M variables were entered into a logistic regression analysis to predict group membership for the autism parents, the items “failure to reference” and “reformulations” were significant for the model including the autism and DS data (model: χ^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, 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: χ^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 autism parents and SLI parents with impaired communication (Total PRS-M Score >= 7)

Seven autism and seven SLI parents scored seven or higher on the PRS-M total score. The gender ratio was six fathers to one mother for both groups. The mean total PRS-M score for this impaired autism parents group was 8.7 compared with 11.0 for the impaired SLI parents.

When the PRS-M subscale scores were compared, none of the differences remained significant after Bonferroni correction.

Item by item comparison showed that autism parents made significantly more grammatical errors (X^2 = 4.67, p = 0.05) than SLI parents, while SLI parents had significantly less facial expressiveness than the autism parents did (X^2 = 10.50, p = 0.002). After Bonferroni correction, the difference remained significant at the 0.05 level only for “facial expressiveness” (Figure 4).

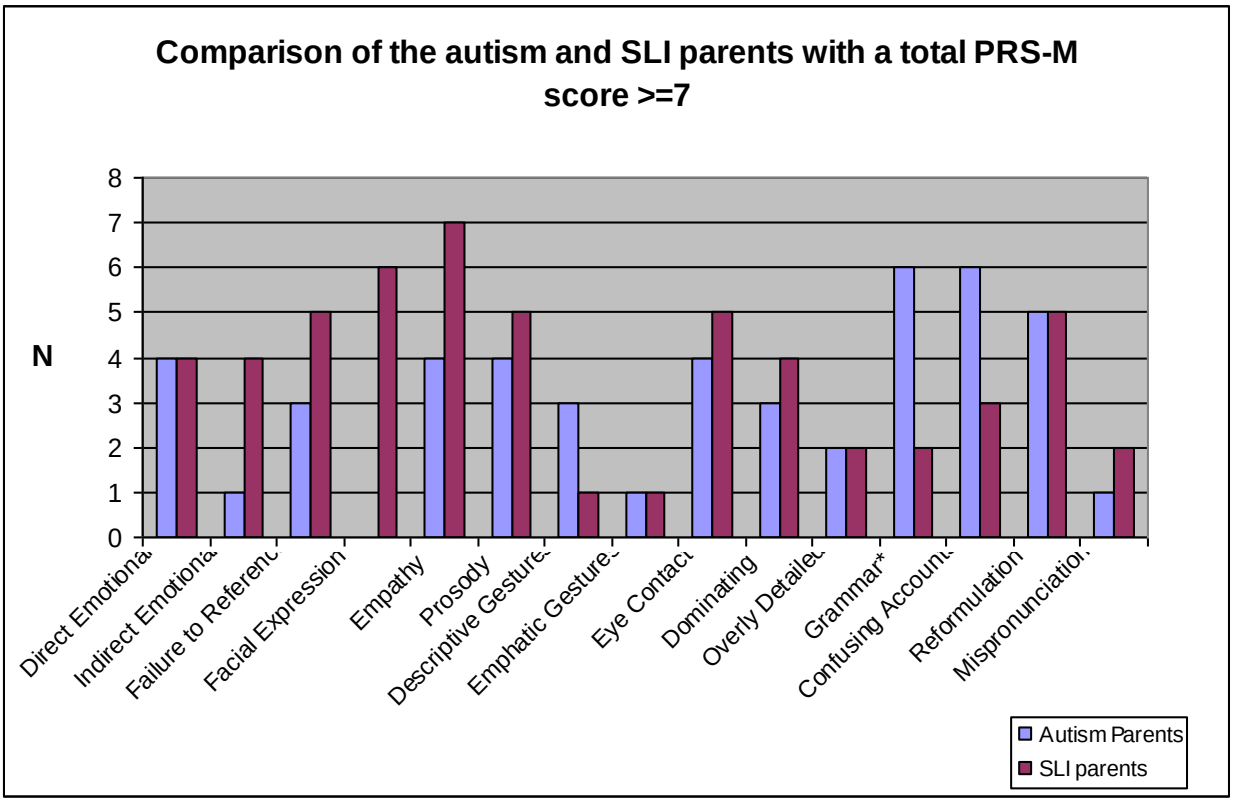

Figure 4: Item by item comparison of 7 autism parents and 7 SLI parents with a total PRS-M Score of 7 or higher

When looking at Figure 4, it appears that the patterns of communication deficits for the parents with a high total PRS-M score were different for the autism and SLI parents. Six out of seven autism parents made significant grammatical errors and endorsed at least one of the two variables “confusing accounts” and “reformulations”, all of which are language variables (Table 7). One parent with this pattern was a mother; the other five were fathers.

One father, subject number 7 (Table 7), was considered to have a possible diagnosis of an autism spectrum disorder according to the family history data. His wife reported that he had definite autistic-like behaviors as a child as well as in his adulthood and did not relate in a to-and-fro manner in adult conversation. He also endorsed probable socially inappropriate behaviors as well as circumscribed interests in childhood and adulthood and lack of affection as a child. His total PRS-M score was 7.

Table 7: Ratings for selected items for the autism and SLI parents with a Total PRS-M Score of >= 7

	Failure to reference	Facial expression	Empathy	Prosody	Eye contact	Grammar	Confusing accounts	Reformulations	Gender	Possible autism spectrum disorder **
AU*										
1				+	+	+	+	+	M	No
2			+	+		+	+	+	M	No
3					+	+	+	+	M	No
4			+		+	+	+	+	M	No
5	+		+	+		+	+		M	No
6	+		+			+		+	F	No
7	+			+	+		+		M	Yes
SLI										
8	+	+	+	+	+		+	+	M	No
9	+	+	+	+	+				M	No
10	+	+	+	+	+				M	No
11		+	+	+	+				M	No
12	+	+	+		+				M	No
13	+		+	+		+	+	+	M	No
14		+	+			+	+	+	F	No

* AU = Autism parents

** Autism or Autism Spectrum Disorder as per Family History Interview (FHI)

All of the SLI parents with impaired communication made no or only limited empathic statements. Five of them also scored positive on at least three of the four items “facial expressions”, “failure to reference”, “eye contact” and “prosody”. All parents with this pattern were fathers. The other two parents, a mother and a father, showed deficits in grammar, related confusing accounts and reformulated frequently. None of their family history data showed evidence of probable autism or autism spectrum disorder.

Comparison of autism parents and SLI parents with a total PRS-M Score of less than 7

The remaining 40 autism and 40 SLI parents scored six or lower on the PRS-M total score and were therefore likely to be only mildly to moderately if at all impaired in their communicative competence. The mean total PRS-M score for this group of autism parents was 2.95 (SD=1.6) compared with 2.90 (SD=1.71) for the SLI parents (t= 0.14, p=0.89, Cohen's d = 0.030, effect size r = 0.015).

For the PRS-M subscale comparisons, no significant differences were found between these two subgroups.

When they were compared on individual items, autism parents used significantly more reformulations (X^2 = 4.11, p = 0.043) and made significantly less eye contact (X^2=4.59, p=0.032) than SLI parents. SLI parents made significantly more grammatical errors (X^2=4.38, p=0.036) than this subgroup of autism parents (Figure 5). After Bonferroni correction, none of these differences remained significant.

One of the autism mothers was found to have probable autism spectrum disorder according to the family history data. She currently endorsed definite restricted, repetitive patterns of behaviors and interests and was treated for obsessive-compulsive symptoms with fluvoxamine. Her other symptoms characteristic of autism were more pronounced in childhood at which time she had impaired social play, circumscribed interests and autistic-like behavior. As adult she was reported to have probable autistic-like behaviors, rigidity and impairment in conversational reciprocity. Her total PRS-M score was one.

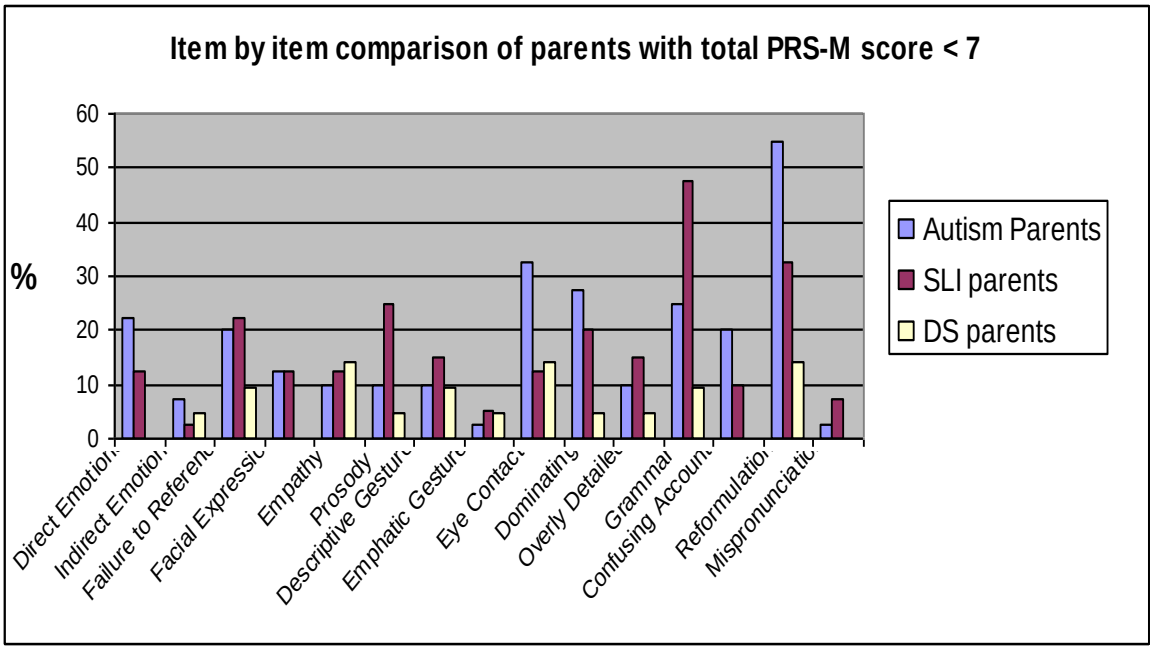

Figure 5: Item by item comparison of 40 autism parents and 40 SLI parents and 21 DS parents with a total PRS-M Score of less than 7

Comparison of parents by gender

Autism and SLI parents combined

Table 8 presents the 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 scored significantly higher on 3 of the 4 items of that subscale, including "prosody" ($X^2 = 9.6$, $p = 0.008$), "empathy" ($X^2 = 6.3$, $p = 0.04$) and "eye contact" ($X^2 = 12.7$, $p = 0.002$). After Bonferroni correction, only poor eye contact remained significant at the 0.05 level. To test whether poor eye contact was an expression of shyness, a correlation analysis was performed for poor eye contact and the FHI item shyness. The two variables were not correlated ($r = 0.0429$, $p = 0.687$).

Table 8: Independent samples t - test for the autism and SLI parents by gender

	Mothers (N=50)		**Fathers (N=44)**		**t**	**p**	**Cohen's d (r)**
	Mean	(SD)	Mean	(SD)			
Emotional Expressiveness and Awareness of Other	0.86	(1.26)	1.54	(1.90)	2.08	0.040	0.47 (0.23)
Communicative Performance	0.46	(0.84)	1.34	(1.44)	3.55	**0.001****	0.87 (0.40)
Over-Talkativeness	0.46	(0.91)	0.61	(1.08)	0.75	0.456	0.15 (0.07)
Language	1.16	(0.99)	1.61	(1.69)	1.56	0.123	0.32 (0.16)
PRS-M	2.94	(2.03)	5.11	(3.72)	3.45	**0.001****	0.86 (0.30)

** significant at the 0.005 level after Bonferroni correction

Comparison of autism, SLI and DS fathers

In the group of fathers, comparison between autism and DS fathers revealed higher scores in the autism group for the PRS-M items "direct emotional verbal expression" (X^2 (1) = 4.59, p = 0.035), "confusing accounts" (X^2 (1) = 4.59, p = 0.035), "dominating conversation" (X^2 (1) = 4.59, p = 0.035) and "reformulations" (X^2 (1) = 9.33, p = 0.002). After adjusting for multiple comparisons, only "reformulations" remained significant at the 0.05 level. Both, the total PRS-M score (mean autism = 4.83, SD = 2.89; mean DS = 1.5, SD = 1.9; t (25) = 3.91, p = 0.001, Cohen's d = 1.36, effect size r = 0.56) and subscale 4, the "Language" score (mean autism = 1.78, SD = 1.70; mean DS = 0.40, SD = 0.84; t (30) = 3.11, p = 0.004, Cohen's d = 1.03, effect size r = 0.45) reached significance which remained after adjustment for multiple comparisons (figure 6).

When SLI and DS fathers were compared, SLI fathers were found to score higher on the items "prosody" (X^2 (1) = 4.19, p = 0.046), "failure to reference" (X^2 (1) = 5.13, p = 0.026) and "reformulations" (X^2 (1) = 5.13, p = 0.026). None of these differences remained significant after Bonferroni correction. The total PRS-M score (mean SLI = 5.43, SD = 4.52; mean DS = 1.5, SD = 1.9; t (29) = 2.62, p = 0.014, Cohen's d = 1.13, effect size r = 0.49), the subscale score 1, "Emotional Expressiveness and Awareness of Other" (mean SLI = 1.90, SD = 0.72; mean DS = 0.30, SD = 0.65; t (29) = 2.23, p = 0.034, Cohen's d = 2.33, effect size r = 0.76) and subscale score 4, "Language" (mean SLI = 1.43, SD = 1.69; mean DS = 0.40, SD = 0.84; t (29) = 2.26, p = 0.032, Cohen's d = 0.77, effect size = 0.36) did not remain significant after adjustment for multiple comparisons (Figure 6).

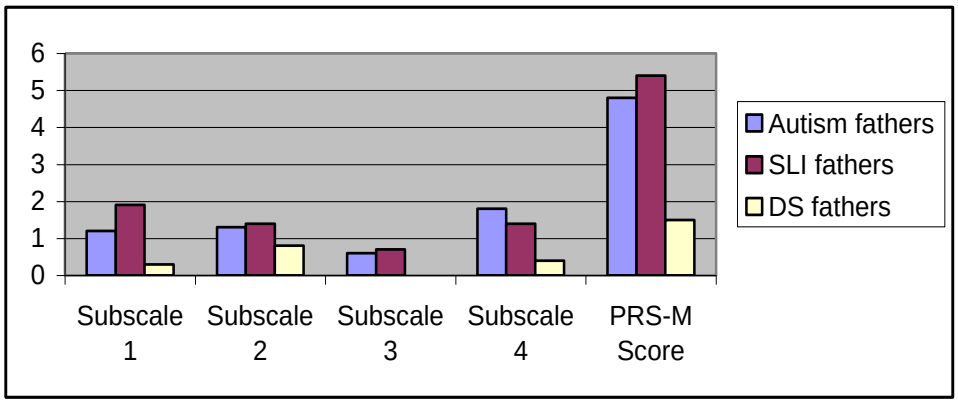

Figure 6: Mean subscale and total PRS-M scores of 23 autism, 21 SLI and 10 DS fathers

Comparison of autism, SLI and DS mothers

Comparison between autism and DS mothers showed significant differences for the total PRS-M (mean autism = 2.83, SD = 2.0; mean DS = 0.73, SD = 0.90; t (33) = 3.3, p = 0.002, Cohen's d = 1.35, effect size = 0.56) and subscale 4, the "Language" score (mean autism = 1.17, SD = 0.96; mean DS = 0.27, SD = 0.47; t (33) = 2.91, p = 0.006, Cohen's d = 1.19. effect size r = 0.51, figure 7), but not for any of the individual PRS-M items.

When SLI mothers were compared with DS mothers, SLI mothers had higher scores on the item "grammatical errors" (X^2 (1) = 6.62, p = 0.009), the total PRS-M score (mean SLI = 3.04, SD = 2.09; mean DS = 0.73, SD = 0.90; t (35) = 3.51, p = 0.001, Cohen's d = 1.44, effect size r = 0.58) and the subscale 4, the "Language" score

(mean SLI = 1.15, SD = 1.05; mean DS = 0.27, SD = 0.47; t (35) = 3.54, p = 0.001, Cohen's d = 1.08, effect size r = 0.48). After adjusting for multiple comparisons, only the PRS-M and language subscale score remained significant (figure 7).

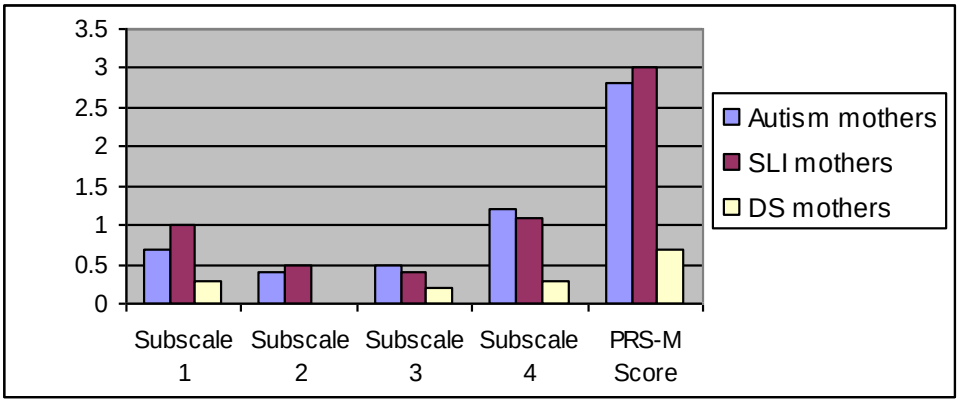

Figure 7: Mean subscale and total PRS-M scores for 24 autism, 26 SLI and 11 DS mothers

"Eye contact" was then compared within the group of fathers and within the group of mothers. Autism mothers had poorer eye contact than SLI and DS mothers. This was only a trend statistically (20.8% of autism mothers, 3.8% of SLI mothers, 0% of DS mothers; X^2 (2) = 5.52, p = 0.063). Autism, SLI and DS fathers had equally poor eye contact (52.2% of autism fathers, 42.9% of SLI fathers and 30.0% of DS fathers; X^2 (2) = 1.42, p = 0.491).

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 (Table 9). 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, X^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, X^2 (1) = 10.55, p = .001) (Table 10).

Table 9: Independent samples t-test for the 4 subscales for autism parents by gender (females N=24, males N=23)

	Autism Mothers Mean (SD)	Autism Fathers Mean (SD)	df	t-value	p 2-tailed	Cohen's d (effect size r)
Emotional Expressiveness	0.75	1.21	45	1.20	0.237	0.34
and Awareness of Other	(1.11)	(1.53)				(0.17)
Communicative	0.41	1.26	45	2.54	0.013	0.75
Performance	(0.77)	(1.39)				(0.35)
Over-Talkativeness	0.50	0.56	45	0.23	0.822	0.06
	(1.02)	(0.94)				(0.03)
Language	1.17	1.78	45	1.53	0.132	0.44
	(0.96)	(1.70)				(0.21)

Table 10: Independent samples t-test for the 4 subscales for SLI parents by gender (females N=26, males N=21)

	SLI Mothers Mean (SD)	SLI Fathers Mean (SD)	df	t-value	p 2-tailed	Cohen's d (effect size r)
Emotional Expressiveness	0.96	1.90	32	1.70	0.099	0.51
and Awareness of Other	(1.40)	(2.21)				(0.25)
Communicative	0.50	1.42	31	2.45	0.020	0.73
Performance	(0.91)	(1.53)				(0.34)
Over-Talkativeness	0.42	0.67	45	0.81	0.421	0.24
	(0.81)	(1.24)				(0.12)
Language	1.15	1.43	45	0.68	0.498	0.19
	(1.05)	(1.69)				(0.10)

Relationship between pragmatic scores and verbal IQ

Correlations between pragmatic measures and verbal IQ for all parents showed small but significant correlations for subscale 1 (Pearson's r = 0.19, p = 0.047) and the total PRS-M (Pearson's r = 0.19, p = 0.045).

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. Five main results emerged. First, the autism and SLI parents had significantly poorer communication abilities than the DS parents. About 15% of the autism and SLI parents had serious communication problems. Second, "failure to reference" and "reformulations" 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. Fifth, DS mothers and fathers had better communication abilities than their autism and SLI counterparts.

As hypothesized, autism as well as SLI parents show communication deficits in comparison with DS parents. Both, the autism and the SLI parents, have more pragmatic deficits, which are highly significant, as measured by the total PRS-M score and the subscale scores "Language", "Over-talkativeness" and "Emotional Expressiveness and Awareness of Other". Contrary to the initial hypothesis however, differences in the overall communication skills between the autism and SLI parents were not found. All previous studies are consistent in finding more frequent communication deficits in parents of autistic children than in parents of normal children, children with Down syndrome or mental handicap (Landa et al., 1992; Piven et al., 1997; Wolff et al., 1988). About 15% of the parents with autism in this study were considered to have significant communication impairments. This is comparable with the family history data by Szatmari et al. who reported that 10% of biological relatives of probands with pervasive developmental disorders had communication impairment and 14% had social impairment (Szatmari et al., 2000). Bolton et al. found a slightly lower

incidence of 8.6% combined social and communication deficits in relatives of probands with autism spectrum disorder on family history reports (Bolton et al., 1994). Results from observation studies that used the PRS can be compared with the ones from this study. Using the 19-item PRS, Landa and colleagues reported a mean total score for autism parents and parents of children with Down syndrome of 4.41 and 0.45, respectively. Likewise, Piven et al. found in their sample of autism parents a mean PRS score of 3.9 compared to a score of 0.8 for the Down syndrome 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 obtained here with the 15- item PRS-M for the autism, SLI and DS parents, 3.80, 4.10 and 1.09, respectively. Thus, it is concluded that SLI parents, like autism parents, more often have significant communication problems than controls. This is a striking and somewhat unexpected result.

While comparison between the entire autism and SLI group did not reveal differences, the division of the whole sample into a significantly impaired and a mildly impaired/unimpaired group uncovered different communication patterns for the autism and SLI parents. Most of these differences, however, did not reach significance.

Within the impaired sample, the SLI parents presented with a wide range of both verbal and non-verbal communication impairments that so far have characterized the broader autism phenotype. The impaired SLI parents, mainly fathers, showed strikingly little and significantly less facial expression than the impaired autism parents. Facial expressiveness serves as an important communicative tool in social exchange. Furthermore, recognition of the facial expressions of one's inter-actional partner is positively correlated with social competence (Nowicki & Mitchell, 1998; Sisterhen & Gerber, 1989) and may influence one's own facial expression. There is some evidence that children with SLI have impairment in reading and producing facial expressions. Twenty six children with SLI performed significantly worse than normally developing children in identifying facial expressions from a video clip in the presence of an unfiltered vocal stimulus (Creusere et al., 2004). Literature on spontaneous facial expression in children with SLI or their relatives is not available for comparison. Research in emotional behaviors suggests that spontaneous imitation of facial expression, though subtle, plays a role in a person's expressiveness and has been found to be correlated with empathy. Electromyographic studies show that normal

subjects respond rapidly with imitation to facial expressions of others, even when they are not aware of it (Dimberg & Thunberg, 1998; Dimberg et al., 2000). Subjects who have a strong tendency to imitate are also more likely to score high on measures of empathy (Chartrand & Bargh, 1999; Sonnby-Borgstrom, 2002). All of the impaired SLI parents with decreased facial expressiveness were impaired in verbalizing empathy. The neuroanatomical link between language, SLI and facial expression may be the Broca's area. Neuroimaging data show that elicited imitation of facial expression activates both hemispheres, including the pars opercularis of the Broca's area (Carr et al., 2003). The left pars opercularis is considered one of the epicenters of language function and is activated during story listening and verb generation; both tasks are performed without word articulation (Papathanassiou et al., 2000). In children with SLI, the Broca's area lacks the normal leftward asymmetry (Gauger et al., 1997). The finding of decreased facial expression and empathy in impaired SLI parents supports a connection between deficits in language and facial expression.

About 70% of the impaired SLI parents had insufficient modulation of their intonation with mainly monotonous voices. Adults with SLI have impaired prosody (Mawhood et al., 2000) and children with SLI show decreased affective prosody (Trauner et al., 1993). The impaired SLI parents in this study may therefore have at least some characteristics of adults with SLI.

About 70% the impaired SLI parents also had poor eye contact. Poor eye contact is an unspecific finding that can occur inter-individually in different conditions and intra-individually in different situations. Individuals with autism are known to have decreased eye contact, as do patients with depression and schizoid disorder. Poor eye contact did not correlate with reported shyness in the fathers.

In the group of impaired autism parents, deficits were found most consistently for language items such as "grammatical errors", "confusing accounts" and "reformulations". This is consistent with the study by Folstein and colleagues that reports a subgroup of autism parents with language and pragmatic impairments (Folstein et al., 1999). In a group of 162 parents of 90 probands with autism, 14% had reportedly a definite history of early language-related cognitive deficits and scored significantly higher on the PRS than those parents without language-related cognitive deficits.

The non-impaired/less severely impaired SLI and autism parents had similar communication abilities. The differences for the individual items may have been stronger with a higher number of parents in each group.

Contrary to the initial third hypothesis, there have been many similarities between the autism and SLI parents. Are there features that are specific for each group? In comparison with the DS parents, different PRS-M items predicted membership for the autism or SLI parent group. The predictors for the autism group were the items “failure to reference” and “reformulations”. While “failure to reference” is a typical characteristic of individuals with autism and the broader autism phenotype (Lord & Paul, 1997; Lord et al., 1989), “reformulations” has not been identified as such. The item “reformulations” had been created to distinguish difficulty in understanding an account secondary to repeated rephrasing of an idea from difficulty in understanding secondary to missing information and unexpected topic shifts. The significance of the high frequency of reformulating ideas as seen in these autism parents remains unclear. Predictors for SLI parents were the items “grammatical errors” and “dominating conversation”. Individuals with SLI frequently make grammatical errors (Johnston & Kamhi, 1984), and family members of children with SLI are known to have language impairments (Stromswold, 1998). Dominating a conversation has not been reported as a feature of SLI in the literature. It has been described in individuals with autism and their family members (Landa et al., 1992; Lord & Paul, 1997) as part of the loss in conversational to- and fro. “Dominating the conversation” had replaced the item “Conversational to-and-fro” of the original PRS in this study because it had better inter-rater reliability. Further differences between the autism and the SLI parents emerged with the separation of the impaired and unimpaired/mildly impaired groups. The most striking result was how little information and emotion was conveyed through facial expression by the impaired SLI parents. Within the mildly impaired/unimpaired group, the SLI parents made more grammatical errors and the autism parents had poorer eye contact and made more reformulations, although these differences were relatively weak.

In the combined group of autism and SLI parents, the fathers communicated more poorly than the mothers. The mean PRS-M score for the fathers was almost twice as high as the score for the mothers. The fathers also scored almost three times higher than the mothers on the subscale “Communicative Performance”. Several earlier studies reported higher scores in fathers of autistic children than in mothers. Male

autism relatives significantly more frequently endorse social-pragmatic deficits and the broader autism phenotype according to family history studies (Bolton et al., 1994; Szatmari et al., 2000). Bolton and colleagues reported that 22% of the males and only 7% of the females among their 332 parents and siblings of probands with autism met criteria for a broadly defined autism phenotype that was characterized by either communication or social deficits or stereotypic behaviors (Bolton et al., 1994). Szatmari and colleagues found 13% of the male biological relatives versus 7% of the female biological relatives from their 92 families with one or more children with autism or pervasive developmental disorders had impairment in communication (Szatmari et al., 2000). However, neither Landa nor Piven found a gender difference between their autism and control parents, although their sample sizes were comparable with the one described here (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 this current study. In addition, there may have been gender differences for single items in these studies that were obscured in the overall score.

In general, females' performance is superior to that of males on pragmatic, social and language measures (Kring & Gordon, 1998). Preschool-aged girls performed better than age-matched boys on 6 out of 8 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-minute interview compared with female students, independent of the gender of the interviewer (Exline et al., 1965). In another study, male college students were also less likely to express their emotions through facial expressions captured by a hidden camera while watching movie sequences than female students (Kring & Gordon, 1998). On the basis of gender differences in cognition, language and social behavior, Baron-Cohen proposed that autism can be viewed as an exaggeration or extreme form of some aspects of maleness (Baron-Cohen, 2002; Baron-Cohen et al., 2005). Here it is shown that despite the small number of DS mothers and fathers and therefore low statistical power, autism and SLI mothers and fathers perform more poorly on the pragmatics measure than their DS counterparts. Therefore, it can be concluded that autism and SLI mothers also endorse communication deficits, although they are milder than in the fathers. In this study, the differences between the DS and SLI fathers did not remain significant after adjustment

for multiple comparisons due to a large standard deviation for the scores of the SLI fathers.

Eye contact is the only item that differentiated fathers from mothers in the combined autism and SLI group and in each separately. In this 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 and abnormal gaze behavior is an important feature in autism. Klin and colleagues found that while viewing naturalistic social scenes from a movie, males with autism focused on a person's mouth and body and on objects, rather than on the person's eyes (Klin et al., 2002). In the study, 15 men with autism and 15 age and verbal IQ matched typically developing males were compared in their visual fixation patterns while watching scenes from Edward Albee's "Who's Afraid of Virginia Woolf" using a newly developed visual tracking method. The best predictor for autism was significantly reduced time spent fixating on the actor's eyes, and the predictor for low social competence were long fixation times for nearby objects (Klin et al., 2002). It is unclear whether autism family members who do not have the full syndrome of autism have not only poor eye contact but also similar fixation patterns. From the sample of this current study, subject number 7 from the impaired group who had possible autism according to family history data was noticed by the interviewer to look in her direction yet not in her eyes. He therefore may have engaged in the same fixation patterns as the men with autism studied by Klin and colleagues.

The findings of this study 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 is 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 et al., 2003) and possibly on 13q (CLSA, 2001). Two of these studies found linkage between a structural language

phenotype and markers on chromosome 7q in autism families. In 75 families with at least two affected siblings, the signals obtained on chromosome 7q, and at least one signal on chromosome 13 q, were attributable to the families in which both probands had language delay (Bradford et al., 2001). In a study of 152 multiplex autism families, the linkage signal was strongest on chromosome 7q when age at first word was used as trait marker (Alarcon et al., 2002).

The 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 both 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. Poor eye contact, while frequent among autism parents and SLI fathers, was less common in SLI mothers and reduced facial expression was a frequent feature of pragmatically impaired SLI fathers.

One goal of this project was to develop a quantitative phenotypic measure to assess communication deficits in autism families which then could be used as trait marker in linkage studies. The PRS-M may now also be applied in genetic linkage studies investigating SLI and other conditions associated with autism and communication deficits such as Fragile-X-Syndrome and Prader-Willi-Syndrome. Future studies may address some of the questions that arose here, e.g.: How are the reduced eye contact and visual fixation patterns of pragmatically impaired autism family members characterized? Are they in some instances qualitatively similar to those seen in probands with autism? Further studies, including imaging studies, may substantiate and elucidate the link between language and communication impairment and lack of facial expression.

Clinical Implications

Although the PRS-M has been used to score video samples in this study, it can easily be taken into the clinical setting and applied to a conversation. For clinicians interacting with children with autism and SLI and their families, it is important to be aware of and able to assess impairments in communication not only in the patients but also in their parents and to adjust communicative and treatment approaches accordingly. The PRS-M may be useful in this process. Children attending psychiatric clinics, setting aside those with autism and SLI, have a high frequency of language problems, which are often overlooked (Giddan et al., 1996). The use of the PRS-M as part of the mental status examination could be helpful in detecting and characterizing these.

Limitations/Error analysis

There are some limitations to this study that 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 this SLI sample. This is unlikely since the children with SLI in this study had to have impaired structural language to enter the study. Furthermore, the children with autism and SLI were recruited separately, using resources that served specifically SLI children to ascertain the SLI cases. This SLI sample should therefore be representative of a group with a balanced distribution of SLI subtypes.

By matching the autism and SLI parents according to verbal IQ, differences in their PRS-M score may also have been diminished. Verbal IQ and pragmatic measures may not be entirely independent. In the group of autism parents with a history of language-related cognitive deficits Folstein and colleagues found not only significantly lower PRS scores, but also significantly lower verbal IQ scores than in the autism parents without such history (Folstein et al., 1999). In another study, the pragmatic measure in patients with schizophrenia correlated with cognitive decline as measured by the difference between pre-morbid and current verbal IQ (Linscott, 2005). Using data from all 115 parents, subscale 1 and total PRS-M score were inversely correlated with verbal IQ in this study, although the correlation coefficient was relatively low. Therefore, differences in the two pragmatic scores may have been reduced by matching the autism and SLI parents.

Since SLI family members have a high incidence of SLI (Stromswold, 1998) and autism parents may have an increased risk of SLI, some of the SLI and autism parents may have the diagnosis of SLI. Unfortunately, these diagnostic data were not available for this study.

All of the interviewers in this study were female. The question whether this has influenced communicative behavior in the fathers and mothers differently comes up especially for the item “Eye contact” because it showed a strong gender difference. As cited above, one study of male and female college students found that the male students engaged in significantly less eye contact than the female students independent of the gender of the interviewer (Exline et al., 1965). Therefore, it is likely that the fact that all interviewers in this study were female has not influenced the participant’s eye contact significantly, although it may be difficult to apply findings from typically developing adults to these parents. To address this question, a substantial number of parents would have needed to be interviewed by a female as well as a male interviewer.

Furthermore, assessing eye contact by viewing videotapes likely did not capture the typical visual fixation pattern of individuals with autism that is characterized by focusing on the person’s mouth and nearby objects. Therefore, reduced eye contact may be underreported here.

Other limitations are due to power constraints. The principal component analysis was performed under the assumption that the parents are one sample, ignoring the diagnostic status. However, given the closely comparable scores of the autism and the SLI 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 the DS mothers and fathers could not be compared to each other to further elucidate the influence of gender on communication abilities.

Finally, one of the autism mothers who was considered to have probable autism spectrum disorder by family history data had a PRS-M score of only one. As an adult, she reportedly experienced obsessive-compulsive symptoms, rigidity and probable autistic-like behavior and impairment in conversational reciprocity. The discrepancy may be due to over-reporting of impairment in communication on the Family History Interview and/or a failure to observe subtle pragmatic deficits with the PRS-M, since pragmatic behaviors were rated as probable only.

SUMMARY

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. Since both syndromes are familial neurodevelopmental disorders with language impairment, the "broader autism phenotype", a phenotypic expression of milder characteristics of autism, has been described in non-affected family members of probands with autism, and autism-like communication deficits have been found in children with "Pragmatic Language Impairment" (PLI), a subset of children with Specific Language Impairment, the goal of this study was to test the hypothesis that both, parents of children with autism and parents of children with SLI show impairments in their communication. Communicative competence was compared among parents of children with autism, parents of children with Specific Language Impairment and parents of children with Down Syndrome (DS). The Pragmatic Rating Scale was modified (modified Pragmatic Rating Scale, PRS-M) to include additional aspects of non-verbal communication and formal language that are found to be impaired in autism and SLI. VARCLUS analysis revealed four PRS-M subscales: "Emotional Expressiveness and Awareness of the Other", "Communicative Performance", "Over-talkativeness" and "Language". The PRS-M was validated against family history data. Inter-rater reliability for the subscale scores ranged from 0.74 to 0.85. Videotapes of conversational interviews with 47 autism, 47 SLI and 21 DS parents were scored blind to group membership. Five main results emerged. First, the autism and SLI parents had significantly lower communication abilities than the DS parents. About 15% of the autism and SLI parents had serious communication problems. Second, "failure to reference" and "reformulations" 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 had poorer 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. Fifth, DS mothers and fathers had better communication abilities than their autism and SLI counterparts. The results suggest that impaired communication is part of both the broader autism phenotype and a broader SLI phenotype, especially among male family members.

ZUSAMMENFASSUNG (Summary in German)

Während bisher angenommen wurde, das das primäre Sprachdefizit bei Patienten mit Autismus aus verminderten pragmatischen Fähigkeiten und bei Patienten mit Spezifischer Sprachentwicklungsstörung aus strukturellen Sprachdefiziten besteht, wird in der jüngeren Literatur eine phenomenologische und potentiell genetische Überlappung beider Syndrome beschrieben. Da beide Syndrome als heriditäre neurokognitive Entwicklungsstörungen mit Sprachdefiziten aufzufassen sind, in Familien mit autistischen Probanden regelmäßig eine milde phenotypische Ausprägung von „autistischen“ Eigenschaften als „Breiterer Autismus Phenotyp“ beschrieben wird, und zumindest beim „Pragmatischen Sprachdefizit“, einer Untergruppe der Spezifischen Sprachentwicklungsstörung, an Autismus erinnernde Kommunikationstörungen auftreten, wurde hier die Hypothese getestet, ob sowohl bei Eltern von Kindern mit Autismus als auch bei Eltern von Kindern mit Spezifischer Sprachentwicklungsstörung Kommunikationsdefizite auftreten. Die vorliegende Arbeit vergleicht pragmatische und strukturelle sprachliche Kompetenz in Konversation, kurz Kommunikative Kompetenz, bei Eltern von Kindern mit Autismus, Spezifischer Sprachentwicklungsstörung und Down Syndrom. Hierzu wurde die existierende Pragmatische Skale (Pragmatic Rating Scale - PRS) modifiziert (PRS-M) und dabei Aspekte non-verbaler Kommunikation und formaler Sprache, die bei Patienten mit Autismus und Spezifischer Sprachentwicklungsstörung beeinträchtigt sind, in die Untersuchung einbezogen. Eine statistische Analyse ergab, das sich die modifizierte Pragmatische Skale in 4 Unterskalen aufteilt: „Emotionale Expressivität und Wahrnehmung des Gegenüber“, „Kommunikative Darstellung“, „Übermäßige Sprachproduktion“ und „Formale Sprache“. Die modifizierte Pragmatische Skale wurde gegen anamnestische Daten, die Sprachentwicklung und soziale und kommunikative Fähigkeiten bewerten (Family History Interview), validiert. Interrater Reliabilität wurde erhoben und bewegte sich für die vier Unterskalen zwischen 0.74 und 0.85. Videoaufnahmen von Konversationen mit 47 Eltern von Kindern mit Autismus, 47 Eltern von Kindern mit Spezifischer Sprachentwicklungsstörung und 21 Eltern von Kindern mit Down Syndrom wurden blind, d.h. ohne Wissen der Gruppenzugehörigkeit, mit Hilfe der modifizierten Pragmatischen Skale bewertet. Es ergaben sich fünf zentrale Ergebnisse. Erstens, Eltern von Kindern mit Autismus und Eltern von Kindern mit Spezifischer Sprachentwicklungsstörung haben signifikant geringere Kommunikative Kompetenz als

Eltern von Kindern mit Down Syndrom. Etwa 15 Prozent der Eltern von Kindern mit Autismus und 15 Prozent der Eltern von Kindern mit Spezifischer Sprachentwicklungsstörung hatten schwerwiegende Kommunikationsschwierigkeiten. Zweitens, „Unfähigkeit, adequate Referenzen zu geben“ und „häufige Umformulierungen“ sagten am besten vorher, ob ein Elternteil ein autistisches Kind hatte und „Grammatikalische Fehler“ und „Dominanz in der Konversation“ sagten am besten voraus, ob ein Elterntteil ein Kind mit Spezifischer Sprachentwicklungsstörung hatte. Drittens, sowohl Väter von Kindern mit Autismus als auch Väter von Kindern mit Spezifischer Sprachentwicklungsstörung hatten insgesamt signifikant größere Kommunikationsdefizite als Mütter von Kindern mit Autismus oder Spezifischer Sprachentwicklungsstörung. Viertens, im Vergleich mit Müttern von Kindern mit Spezifischer Sprachentwicklungsstörung zeichnete sich für Mütter von Kindern mit Autismus eine Tendenz zu häufiger eingeschränktem Blickkontakt ab. Fünftens, Mütter von Kindern mit Down Syndrom hatten bessere Kommunikationsfähigkeiten als Mütter von Kindern mit Autismus oder Spezifischer Sprachentwicklungsstörung. Das gleiche galt für die Väter.

Insgesamt deuten die Ergebnisse daraufhin, daß Kommunikationsdefizite sowohl Teil des „Breiteren Autismus Phenotyp“ als auch Teil eines hypothetischen „Breiteren Phenotyp der Spezifischen Sprachentwicklungsstörung“ sind.

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Attachment

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 wet shopping", "There's two parks" instead of "There are two parks", "I got it good" instead of "I've got it good".)

☐

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.

☐

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.

☐

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.")

☐

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.

☐
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.

☐
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. 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).

☐
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").

☐
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.

☐
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.

☐
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.

☐
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.)

☐
0 = no mispronunciations
1 = mispronounces at least 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.

☐
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.

☐
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 other's 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

LIST OF ABBREVIATIONS

APA	American Psychiatric Association
CCC	Children's Communication Checklist
CELF	Clinical Evaluation of Language Fundamentals
DS	Down Syndrome
DSM	Diagnostic and Statistical Manual of Mental Disorders
FHI	Family History Interview of Developmental Disorders of Cognition and Social Functioning
ICD	International Statistical Classification of Diseases and Health related Problems
IQ	Intelligence Quotient
PLI	Pragmatic Language Impairment
PDD	Pervasive Developmental Disorders
PRS	Pragmatic Rating Scale
PRS – M	Pragmatic Rating Scale – Modified
SD	Standard Deviation
SLI	Specific Language Impairment
Tufts-NEMC	Tufts-New England Medical Center
WAIS	Wechsler Adult Intelligence Scale
WHO	World Health Organization
WISC	Wechsler Intelligence Scale for Children

LIST OF TABLES AND FIGURES

Lebenslauf

Mein Lebenslauf wird aus Datenschutzgründen in der elektronischen Version meiner Arbeit nicht mitveröffentlicht.

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