
A Subtype of Speech Delay Associated With Developmental Psychosocial Involvement

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This report presents findings supporting the hypothesis of a clinically relevant subtype of childhood speech sound disorder, provisionally titled speech delay—developmental psychosocial involvement (SD–DPI). Conversational speech samples from 29 children who met inclusionary criteria for SD–DPI were selected from a case record archive at a university speech clinic for children. Participants with SD–DPI had been characterized by speech clinicians and caregivers as having speech delay with psychosocial issues that required attention in the course of at least 1 semester of speech treatment. The 29 participants were divided into 2 subgroups, based on clinicians’ and parents’ records indicating either *approach-related negative affect* ($n = 23$) or *withdrawal-related negative affect* ($n = 6$). Each participant with SD–DPI was matched by age, gender, and type of speech involvement to 3 comparison speakers with speech delay of unknown origin ($n = 87$). Analyses of the conversational speech samples indicated that in comparison with participants in the control group, those with SD–DPI had significantly more severe speech delay, averaging approximately 7% to 10% lowered speech competence in conversation. The clinical prevalence of SD–DPI was estimated at approximately 12% of children referred to the university speech clinic in the present study. The authors interpret the present findings to indicate that approach-related or withdrawal-related negative affect, negative emotionality or mood, and decreased task persistence or attention are risk factors for increased severity of expression of speech delay.

KEY WORDS: articulation, classification, diagnosis, etiology, phonology, speech disorder, temperament

Individual differences in genetic as well as environmental risk and protective factors contribute to the origin, severity, and normalization rates of speech sound disorders (SSD) of currently unknown origin. Shriberg et al. (2004) have described seven putative etiologic subtypes of SSD, including five types of speech delay (SD) suspected to be associated with (a) genetic transmission, (b) early recurrent otitis media with effusion, (c) apraxia of speech, (d) dysarthria, and (e) developmental psychosocial involvement (SD–DPI). The remaining two subtypes of SSD constitute the most common types of speech errors, including children whose speech disorder is limited to clinically significant distortions of sibilants and other fricatives and/or distortions of rhotic consonants and vowels. Reports have proposed perceptual and acoustic speech and prosody–voice markers that may eventually have the sensitivity and specificity to differentiate among six of the seven subtypes of SSD. The present article reports the first attempt to identify possible speech markers that differentiate SD–DPI from the other six proposed subtypes

of speech delay of currently unknown origin. Following an overview of relevant concepts and literature, we provide additional rationale for the present study and a statement of the problem.

Overview Perspectives

From its earliest beginnings as a clinical and research discipline, the study of communicative disorders has sought explanatory accounts of the interaction between psychosocial variables and disorders of speech, hearing, language, voice, and fluency. The ability to communicate effectively plays a principal role in the development of appropriate psychosocial skills and behaviors. Studies have documented that persistent misarticulation of even one frequently occurring phoneme can have significant negative impacts on a speaker's self-concept and may have lasting consequences for social development and vocational choices (e.g., Felsenfeld, Broen, & McGue, 1992; Silverman & Paulus, 1989).

Prevalence estimates of clinically relevant psychosocial involvement in children with speech–language disorders have reported comorbidity rates as high as 50% (Baker & Cantwell, 1982, 1987a, 1987b; Beitchman, Hood, & Inglis, 1990; Beitchman, Nair, Clegg, Ferguson, & Patel, 1986; Benasich, Curtiss, & Tallal, 1993; Cantwell & Baker, 1985, 1987; Cantwell, Baker, & Mattison, 1979; Paul, Cohen, & Caparulo, 1983; Stevenson & Richman, 1978; Tomblin, Zhang, Buckwalter, & Catts, 2000). The most frequent indications of psychosocial involvement have included symptoms associated with attention-deficit and disruptive behavior disorders (e.g., attention-deficit/hyperactivity disorder [ADHD], conduct disorder, oppositional defiant disorder). Among children diagnosed with psychiatric disorders, the prevalence of speech–language disorders has been estimated to range from approximately 40% to 80% (Baltaxe & Simmons, 1988; Camarata, Hughes, & Ruhl, 1988; Chess & Rosenberg, 1974; N. J. Cohen, Davine, Horodezky, Lipsett, & Isaacson, 1993; N. J. Cohen, Davine, & Meloche-Kelly, 1989; Gualtieri, Koriath, van Bourgondien, & Saleeby, 1983; Javorsky, 1995; Love & Thompson, 1988; Mack & Warr-Leeper, 1992; Tirosh & Cohen, 1998). The most frequently reported associations have been among attention-deficit and language disorders, although it is believed that a significant proportion of children with ADHD have undiagnosed language deficits. It is currently unclear whether the psychiatric disorders reported in this literature are consequences of the communicative disorder or whether communicative disorders are secondary symptoms of the psychiatric disorders.

Specific to the present context, a number of studies have reported increased behavioral problems in chil-

dren with speech delay of unknown origin. Studies from this primarily older literature report that children with speech delay were significantly more likely to experience (a) attention and concentration deficits (Baumgartner, 1980; Wylie, Franchack, & McWilliams, 1965); (b) anxiety and fearfulness (Baumgartner, 1980; Fitzsimons, 1958; Solomon, 1961); (c) depressive mood and low self-esteem (Barrett & Hoops, 1974; Baumgartner, 1980; Davis, 1937; Wylie et al., 1965); (d) eating, elimination, and sleeping difficulties (Fitzsimons, 1958; Wylie et al., 1965); (e) poor academic performance and disruptive behavior, including hyperactivity, aggression, destructiveness, tantrums, and fights with parents (Calnan & Richardson, 1976; Fitzsimons, 1958; Wylie et al., 1965); (f) poor peer relations, due to shyness, isolation, and/or lack of acceptance (Baumgartner, 1980; N. R. Butler, Peckham, & Sheridan, 1973; Davis, 1937; Fitzsimons, 1958; Lerea & Ward, 1966; Perrin, 1954; Sheridan & Peckham, 1973; Solomon, 1961; Wylie et al., 1965); (g) poor overall adjustment and emotional disturbance (K. G. Butler, 1965; N. R. Butler et al., 1973; Sheridan & Peckham, 1973; Solomon, 1961); and (h) symptoms of personality and psychotic disorders (Lindholm & Touliatos, 1979). Early studies also have suggested that parents of children with speech delay may have been overly critical of their children's speech (Andersland, 1961; Beckey, 1942; Moll & Darley, 1960; Peckarsky, 1953; Wood, 1946). In a comprehensive literature review, Winitz (1969) has concluded that children's articulation disorders are more likely influenced by parenting style than by individual differences in the children's personalities. Later reviews of the earlier studies on personality variables in childhood speech disorders have interpreted the findings as inconclusive, due to the use of unreliable assessment instruments, biased sampling procedures, lack of protections against examiner bias, and inadequate research designs (cf. Baker & Cantwell, 1985; Bloch & Goodstein, 1971). More recent studies have explored associations between speech–language disorders and constructs such as withdrawal and anxiety, social immaturity, and peer acceptance (cf. Fujiki, Brinton, Morgan, & Hart, 1999; Fujiki, Brinton, & Todd, 1996; Gertner, Rice, & Hadley, 1994; Hadley & Rice, 1991; Redmond & Rice, 1998; Rice, Sell, & Hadley, 1991); see recent review by Fujiki and Brinton (2004).

Temperament

A developmental perspective that may be useful as a conceptual framework for the present concerns is the construct of *temperament*, generally defined as a distinctive behavioral style that is relatively stable across an individual's life span (Buss & Plomin, 1984; Rothbart & Derryberry, 1981; Thomas & Chess, 1977). The core

dimensions of temperament are not currently agreed on by all researchers but may include variables such as *mood*, *negative emotionality*, *approach-withdrawal*, *intensity*, *threshold*, *rhythmicity*, *distractibility*, *attention span*, *task persistence*, and *adaptability*. Most researchers view temperament as a stable attribute that may be modified by the child's later experiences (Bouchard & Loehlin, 2001; Goldsmith, 1988; Rothbart & Bates, 1998). Genetic studies of temperament have reported relatively high heritability coefficients in the range of .50 to .60 (e.g., Buss & Plomin, 1984; Kagan, 1989; Rothbart & Derryberry, 1981).

Because individual differences in temperament are associated with fundamental differences in human functioning, temperament may be an early and significant precursor of typical and atypical development (Teglasi, 1995). For example, developmental studies have reported significant associations between aspects of temperament and cognitive development (Miceli, Whitman, Borkowski, Braungart-Rieker, & Mitchell, 1998; Singer & Fagen, 1992); language acquisition (Dixon & Shore, 1997; Dixon & Smith, 2000); adaptation skills and academic achievement (Martin, Drew, Gaddis, & Moseley, 1988; Martin, Olejnik, & Gaddis, 1994); and behavioral adjustment (Kyrios & Prior, 1990; Windle, 1989). School-based studies have described how the three temperamental characteristics of *activity*, *distractibility*, and *persistence* (Martin et al., 1988) may contribute positively or negatively to children's success in school (cf. Keogh, 2003). In the domain of communicative disorders, studies have assessed individual differences in temperament as a risk factor or correlate of delayed language development (Caulfield, Fischel, DeBaryshe, & Whitehurst, 1989; Paul & James, 1990; Paul & Kellogg, 1997) and stuttering (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts, Ebben, Franke, & van de Poel, 2000; Lewis & Goldberg, 1997). Such studies report that in comparison with typically speaking controls, children with speech and language disorders have significantly increased scores on measures of sensitivity, anxiety, distractibility, neuroticism, withdrawal, and difficulty in adaptability.

Temperament variables representing negative affect have been classified into two domains (Goldsmith, Lemery, & Essex, 2004): *approach-related negative affect* (temperament associated with specific constructs of anger) and *withdrawal-related negative affect* (temperament associated with specific constructs of fear and sadness). These two domains have been assessed using both direct methods (e.g., behavioral observation) and indirect methods (e.g., parent/examiner interview/questionnaire). Although information provided via direct observation is limited, it may reduce some of the bias associated with parent interview or questionnaire methods (Martin, 1988). However, parental reports

often yield a more comprehensive view of temperament characteristics in the child's most natural environment (cf. Anderson et al., 2003). Sajaniemi, Salokorpi, Rita, and von Wendt (1997) reported high concurrent validities for scores on temperament and behavior questionnaires and the risk for delayed speech development.

Rationale and Statement of Purpose

Notwithstanding the breadth of findings reviewed above, many questions remain in regard to the nature of psychosocial involvements in children with speech delay of unknown origin. In addition to the lack of prevalence, comorbidity, and explanatory data using contemporary classifications of speech sound and psychosocial disorders, there are no available data describing the speech error profiles of children with comorbid speech delay and psychosocial involvement. Such information is deemed important for both research and practice, in efforts to develop explanatory models that may lead to more targeted treatment approaches.

The purpose of the present study was to compare the speech and prosody-voice profiles of children with suspected SD-DPI to the profiles of children with other forms of speech delay of unknown origin. To address possible differences in the types of psychosocial involvement, participants with suspected SD-DPI were classified into two subgroups based on parents' and clinician's assessments of approach-related or withdrawal-related negative affect. A primary goal of the present study was to determine if one or more speech or prosody characteristics might have sufficient accuracy to be used as a diagnostic marker of children meeting criteria for SD-DPI.

Method

Participants

Group 1: SD-DPI Participants

A subset of 21 male and 8 female participants with suspected DPI was assembled from a database of conversational speech samples collected at a university phonology clinic. As described in a prior report using this archive (Shriberg & Kwiatkowski, 1994), these speech samples were collected from 3-6-year-old children whose speech errors were severe enough to interfere with intelligibility and to warrant speech services. Exclusionary criteria included significant deficits in cognitive, sensory, structural, or speech motor processes, as well as significant affective deficits that would constitute a diagnostic classification within the autism spectrum.

Table 1. Descriptive information for participants with suspected developmental psychosocial involvement (Group 1) and matched comparison participants (Group 2).

| Group | n | Age (in months) | | AWU ^a | |
|----------------|----|-----------------|------|------------------|------|
| | | M | SD | M | SD |
| 1 | | | | | |
| Boys | 21 | 57.2 | 12.9 | 3.56 | 1.19 |
| Girls | 8 | 51.4 | 10.9 | 3.37 | 0.89 |
| Total | 29 | 55.6 | 12.5 | 3.51 | 1.10 |
| 1 _A | | | | | |
| Boys | 18 | 55.1 | 12.6 | 3.69 | 1.22 |
| Girls | 5 | 46.6 | 10.9 | 3.05 | 0.83 |
| Total | 23 | 53.2 | 12.5 | 3.55 | 1.16 |
| 1 _B | | | | | |
| Boys | 3 | 70.0 | 5.6 | 2.84 | 0.84 |
| Girls | 3 | 59.3 | 5.0 | 3.90 | 0.85 |
| Total | 6 | 64.7 | 7.5 | 3.37 | 0.95 |
| 2 | | | | | |
| Boys | 63 | 57.2 | 12.3 | 3.95 | 1.13 |
| Girls | 24 | 53.7 | 8.4 | 4.33 | 1.40 |
| Total | 87 | 56.3 | 11.4 | 4.05 | 1.22 |
| 2 _A | | | | | |
| Boys | 54 | 55.2 | 12.1 | 3.94 | 1.16 |
| Girls | 15 | 50.1 | 8.2 | 4.05 | 0.69 |
| Total | 69 | 54.1 | 11.5 | 3.96 | 1.07 |
| 2 _B | | | | | |
| Boys | 9 | 69.1 | 4.2 | 4.00 | 1.02 |
| Girls | 9 | 59.8 | 4.6 | 4.79 | 2.10 |
| Total | 18 | 64.4 | 6.4 | 4.40 | 1.66 |

Note. AWU = average words per utterance (Shriberg, Allen, McSweeney, & Wilson, 2001). Subscript A = approach-related negative affect; subscript B = withdrawal-related negative affect.

Table 1 includes descriptive information for this group. Participants met the following inclusionary criteria for the purposes of the present study: (a) speech status classified as SD, questionable SD (QSD), or normal speech acquisition/SD (NSA/SD), as defined in the Speech Disorders Classification System (Shriberg, Austin, Lewis, McSweeney, & Wilson, 1997b); (b) normal cognitive-linguistic, structural, and sensorimotor function, as determined by additional screening protocols used in the database; (c) notable evidence of consistent psychosocial involvement, as confirmed by a consensus between the clinician who supervised the children's treatment (third author) and an additional examiner who reviewed their case history and treatment records (first author); and (d) minimally, participation in more than one dynamic assessment session and at least one session of diagnostic teaching. The latter criteria were included to provide detailed information on participants' behaviors during treatment sessions, as annotated by their clinicians, in addition to case records that included relevant notes from participants' parents,

teachers, and physicians. Formal documentation was not obtained regarding past or current evaluation of psychological status. All children were native speakers of American English and had no significant dialectal differences from General American English.

As shown in Table 1, the percentage of boys in Group 1 (72%) is consistent with the percentage of boys typically reported in studies of young children with undifferentiated speech delay of unknown origin (70%; Shriberg, Aram, & Kwiatkowski, 1997; Shriberg, Tomblin, & McSweeney, 1999). The mean age for the 29 Group 1 participants was 4;8 (years;months) ($SD = 1;0$), which was consistent with the mean age of 4;3 for children with speech delay of unknown origin referred to the university speech clinic as reported in a prior study (Shriberg & Kwiatkowski, 1994). Average words per utterance (AWU; Shriberg, Allen, McSweeney, & Wilson, 2001) was the only language measure available for all participants in the present study; for children in this age group, AWU values correlated in the high .90s with mean length of utterance (Shriberg, Allen, et al., 2001). Statistical comparisons indicated that there were no statistically significant between-group differences in age, $t(14) = 1.22, p = .242$, or AWU, $t(17) = 0.47, p = .643$.

Group 1 Subgroups

The group of 29 participants with suspected SD-DPI was divided into two subsamples based on case records data supporting either *approach-related negative affect* or *withdrawal-related negative affect* (cf. Goldsmith, Lemery, & Essex, 2004). Subgroup 1_A comprised 18 male and 5 female participants who had regularly demonstrated behavior during treatment sessions that was interpreted by one or more clinicians as "aggressive," "angry," or "manipulative/control-seeking." For inclusion in Subgroup 1_A, clinicians' observations had to be confirmed by case records indicating that the child's parent(s) had also documented these behaviors as regular and persistent outside the speech therapy environment. A second subsample, Subgroup 1_B, comprised 3 male and 3 female participants who had regularly demonstrated behavior during treatment sessions that was perceived by their clinician(s) as "socially withdrawn," "shy/fearful," and "extremely taciturn" and had not demonstrated behavior that was "aggressive," "angry," or "manipulative/control-seeking." For all such children, inclusionary criteria for Subgroup 1_B also required confirmation by the parent(s) that the above behavior patterns were regular and persistent outside the speech therapy environment. Table 1 includes descriptive age and AWU information for the participants in the 1_A and 1_B subgroups. Statistical analyses indicated no significant differences in the AWU scores of participants in the two SD-DPI

subgroups, $t(9) = 0.39$, $p = .709$, although the 6 participants in Subgroup 1_B were significantly older, $t(13) = -2.84$, $p = .014$. A test of proportions comparison yielded a nonsignificant difference in gender distribution between the two subgroups ($z = -1.28$, $p = .202$).

Group 2: Comparison Participants

For each of the 29 participants with suspected SD–DPI, 3 speakers with speech delay of unknown origin were individually selected to constitute a comparison group (Group 2; $n = 87$). Their conversational speech samples were assembled using a database of samples ($n = 600$) both from speakers treated at the same university clinic and speakers included in collaborative projects with other investigators. Inclusionary criteria for comparison participants in Group 2 were as follows: (a) matched gender with Group 1 counterpart participant; (b) matched age (within 8 months, if necessary) with Group 1 counterpart participant; (c) matched classification of SD, QSD, or NSA/SD with the classification of the Group 1 counterpart participant; and (d) normal cognitive, sensorimotor, and psychosocial function, as determined by protocols used in the database. If more than three potential comparison samples from the database of candidate samples equally matched their respective Group 1 participant, three comparison samples were randomly selected from the available pool. If a comparison candidate could not be matched for exact age in months, the candidate meeting inclusionary criteria with the next closest age was selected. Of the final group of 87 comparison participants in Group 2, 55 (63%) were age matched within 1 month, 72 (83%) were age matched within 2 months, and 80 (92%) were age matched within 3 months of their Group 1 counterpart. The remaining 7 comparison participants were age matched within 4 to 8 months of their Group 1 counterparts. A series of t tests indicated no significant differences between the ages of participants in Group 1 and Group 2, $t(44) = -0.25$, $p = .800$; although the participants in Group 2 had significantly higher AWU scores, $t(52) = -2.23$, $p = .030$.

The comparison subgroups were termed *Subgroup 2_A* (matched with SD–DPI subgroup 1_A) and *Subgroup 2_B* (matched with SD–DPI subgroup 1_B). As shown in Table 1, Subgroups 2_A and 2_B included conversational speech samples from 63 (72%) male speakers whose mean age was 4;9 ($SD = 1;0$) and 24 (28%) female speakers whose mean age was 4;6 ($SD = 0;8$). The mean age for the 87 speakers in Group 2 was 4;8 ($SD = 0;11$).

Temperament Information

Table 2 contains individual information on the 12 temperament variables that were used to classify the 29 Group 1 participants as suspected SD–DPI. The first

6 variables are subsumed under the two primary dimensions of approach- and withdrawal-related negative affect. Participant data for the remaining 6 variables are subsumed under two additional dimensions reflecting related constructs in the temperament literature (cf. Keogh, 2003). Tabular entries of 1 indicate unequivocal clinician and parental support for a variable in the case histories. A 0.5 value was assigned to behaviors that were only occasionally observed or inconsistent. Missing data are indicated by a dash. Subgroup assignments are noted in column 1. Participants with one or more 1 entries in the variables for approach-related negative affect were assigned to Subgroup 1_A, and participants with entries in all three variables for withdrawal-related negative affect (but no entries in the approach-related negative affect columns) were assigned to Subgroup 1_B. These decisions, which were made after the data for Table 2 had been assembled, reflected our best attempt to differentiate the two groups based on participants' individual profiles. As indicated by the totals in the rightmost column, clinician and parent observations attested from 27% to 92% of the 12 variables for each participant, averaging 62% ($SD = 18%$) temperament variables per participant.

Statistical Analyses

Speech and prosody–voice analyses were completed using a suite of software programs developed for research in typical and atypical speech acquisition (Shriberg et al., 2001). For the theoretical and clinical goals of this study, t tests (unpooled variance) and effect sizes (Hedges-corrected) were calculated to assess the magnitudes of between-group differences. To obtain more sensitivity from the effect-size metric, J. Cohen's (1988) conventional recommendation for a large effect size (i.e., >0.79) was further subdivided into three subclasses: large (0.80–0.99), very large (1.00–1.99), and extremely large (>2.00).

Results

Clinical Prevalence of SD–DPI

The procedures used to assemble a sample of children meeting criteria for SD–DPI allowed for a preliminary estimate of the clinical prevalence of this proposed subtype of child speech sound disorders of currently unknown origin. As indicated in the Method section, the convenience sample for the present study was composed of children treated at a university clinic specializing in the treatment of children's intelligibility problems of unknown origin (cf. Shriberg & Kwiatkowski, 1994). A total of 245 children, the parent group for the present study, were treated at the clinic

Table 2. SD–DPI participant information on four dimensions of temperament.

| Participant no. | SD–DPI subgroup | Approach-related negative affect | | | Withdrawal-related negative affect | | | Negative emotionality/mood | | | Task persistence/attention | | | Total ^a |
|-----------------|-----------------|----------------------------------|-------|---------------------------------|------------------------------------|----------------|--------------------|----------------------------|-------------------|------------------------|---|---|----------------------|--------------------|
| | | Aggressive | Angry | Manipulative or control-seeking | Socially withdrawn | Shy or Fearful | Extremely taciturn | Easily upset | Easily frustrated | Sensitive about speech | Unmotivated to work on general speech tasks | Unwilling to try difficult speech tasks | Short attention span | |
| 1 | 1 _A | 1 | | 1 | | | | | 1 | – | 0.5 | 0.5 | 1 | 45 |
| 2 | 1 _A | 1 | | 1 | | | | | 1 | – | 1 | 1 | 1 | 55 |
| 3 | 1 _A | 1 | 1 | 1 | 1 | | | | 1 | – | 1 | 1 | – | 70 |
| 4 | 1 _A | 1 | | 1 | | | | | | 1 | 0.5 | 0.5 | 1 | 42 |
| 5 | 1 _A | 1 | | 1 | | | 0.5 | 1 | 1 | 1 | 0.5 | 1 | 1 | 67 |
| 6 | 1 _A | | | 1 | | | | 1 | 1 | – | | 1 | | 36 |
| 7 | 1 _A | 1 | 1 | 1 | – | | | 1 | 1 | – | 1 | 1 | 1 | 80 |
| 8 | 1 _A | 1 | 1 | 1 | – | – | | 1 | 1 | – | 0.5 | 0.5 | 1 | 78 |
| 9 | 1 _A | 1 | | 1 | – | | | | 1 | – | 0.5 | 0.5 | 1 | 50 |
| 10 | 1 _A | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 92 |
| 11 | 1 _A | 1 | 1 | 1 | | | | 1 | 1 | – | 0.5 | 1 | 1 | 68 |
| 12 | 1 _A | 1 | 1 | 1 | 1 | | | 1 | 1 | – | 1 | 1 | 1 | 82 |
| 13 | 1 _A | 1 | 1 | 1 | | | 0.5 | 1 | 1 | 1 | 1 | 1 | 1 | 79 |
| 14 | 1 _A | 1 | 1 | 1 | | | 1 | 1 | 1 | 1 | | 1 | | 67 |
| 15 | 1 _A | 1 | 1 | 1 | 1 | | | 1 | 1 | – | 1 | 1 | 1 | 82 |
| 16 | 1 _A | 1 | 1 | 1 | | | | 1 | 1 | – | 1 | 1 | 1 | 73 |
| 17 | 1 _A | | | 1 | | | | 1 | 1 | – | 0.5 | 1 | 1 | 50 |
| 18 | 1 _A | 1 | 1 | 1 | 1 | | | 1 | 1 | 1 | | | | 58 |
| 19 | 1 _A | | | 1 | 1 | 1 | | 1 | 1 | – | 0.5 | 1 | 1 | 68 |
| 20 | 1 _A | 1 | | 1 | 1 | 1 | | 1 | 1 | – | 0.5 | 0.5 | 1 | 73 |
| 21 | 1 _A | | | 1 | 1 | 1 | | | 1 | – | 1 | 1 | 1 | 64 |
| 22 | 1 _A | 1 | 1 | 1 | 1 | | | 1 | 1 | 1 | 1 | 1 | 1 | 83 |
| 23 | 1 _A | 1 | 1 | 1 | – | | | 1 | 1 | – | 1 | 1 | 1 | 80 |
| 24 | 1 _B | | | | 1 | 1 | 1 | – | 1 | – | – | | | 44 |
| 25 | 1 _B | | | | 1 | 1 | 1 | – | – | – | | 0.5 | | 39 |
| 26 | 1 _B | | | | 1 | 1 | 1 | | | 1 | | | | 33 |
| 27 | 1 _B | | | | 1 | 1 | 0.5 | 1 | 1 | 1 | – | – | 0.5 | 60 |
| 28 | 1 _B | | | | 0.5 | 1 | 0.5 | 1 | | – | | | | 27 |
| 29 | 1 _B | | | | 1 | 1 | 1 | – | – | – | 0.5 | 0.5 | | 44 |
| Subtotal | 1 _A | 83 | 57 | 100 | 47 | 27 | 4 | 74 | 96 | 100 | 67 | 85 | 82 | |
| Subtotal | 1 _B | 0 | 0 | 0 | 92 | 100 | 83 | 67 | 50 | 100 | 13 | 20 | 8 | |
| Total | 1 | 66 | 45 | 79 | 58 | 43 | 21 | 73 | 89 | 100 | 59 | 73 | 66 | |

Note. SD–DPI = speech delay—developmental psychosocial involvement. Entries in the *Total* column are the sum of values on all temperament variables per participant, divided by the number of variables for which the participant had available data (according to clinical records and confirmed by parental report). Variables attested as characteristic of a participant were given 1 point. Variables considered “varied” were given 0.5 point. Variables for which data were not available are indicated by a dash. All totals and subtotals are percentages.

for at least 1 semester during the 18-year period from 1984 to 2001. Additional children seen during this period were excluded from the prevalence-estimate denominator for one or more of three reasons: (a) They had associated developmental deficits, including cognitive disorder (e.g., Down syndrome), craniofacial disorder (e.g., cleft palate), sensorineural or conductive hearing impairment, or primary affective disorder (autism, Asperger syndrome); (b) they were referred only for intransigent distortion errors; or (c) they were over 9 years of age when assessed and treated.

As constrained by the demographics of the referral region and ascertainment procedures in the university speech clinic (and by the inclusionary criteria used in the present study), the preliminary prevalence estimate of the proportion of children with speech sound disorders who meet criteria for SD–DPI is 11.8% (29/245). Shriberg and Kwiatkowski (1994) provided a clinical profile of children with speech sound disorders of unknown origin, including clinical prevalence estimates for other proposed subtypes. Based on a total sample of 178 children from several prior studies, the clinical prevalence estimate for disorders meeting criteria for either childhood apraxia of speech or developmental psychosocial involvement was estimated at 14%. This percentage was the remainder from the sum of the estimates for two other putative subtypes (86%). That is, 56% of the children with speech delay in the prior study were estimated to be at risk for genetically transmitted speech disorder (because they were positive for familial aggregation), and 30% of this sample of children were classified as positive for histories of early recurrent otitis media with effusion. Thus, because childhood apraxia of speech is likely a rare disorder (cf. Shriberg & Campbell, 2002), the present estimate of the percentage of children with speech delay who meet criteria for SD–DPI (11.8%) is quite consistent with prior preliminary estimates.

Preliminary Analyses of Group I Subgroups

A series of preliminary analyses was completed to determine whether the speech or prosody–voice findings for the two SD–DPI subgroups (1_A and 1_B) differed significantly from their comparison subgroups or from each other. The purpose was to determine whether the two subgroups should be treated separately in the primary analyses or whether they could be combined to increase statistical power.

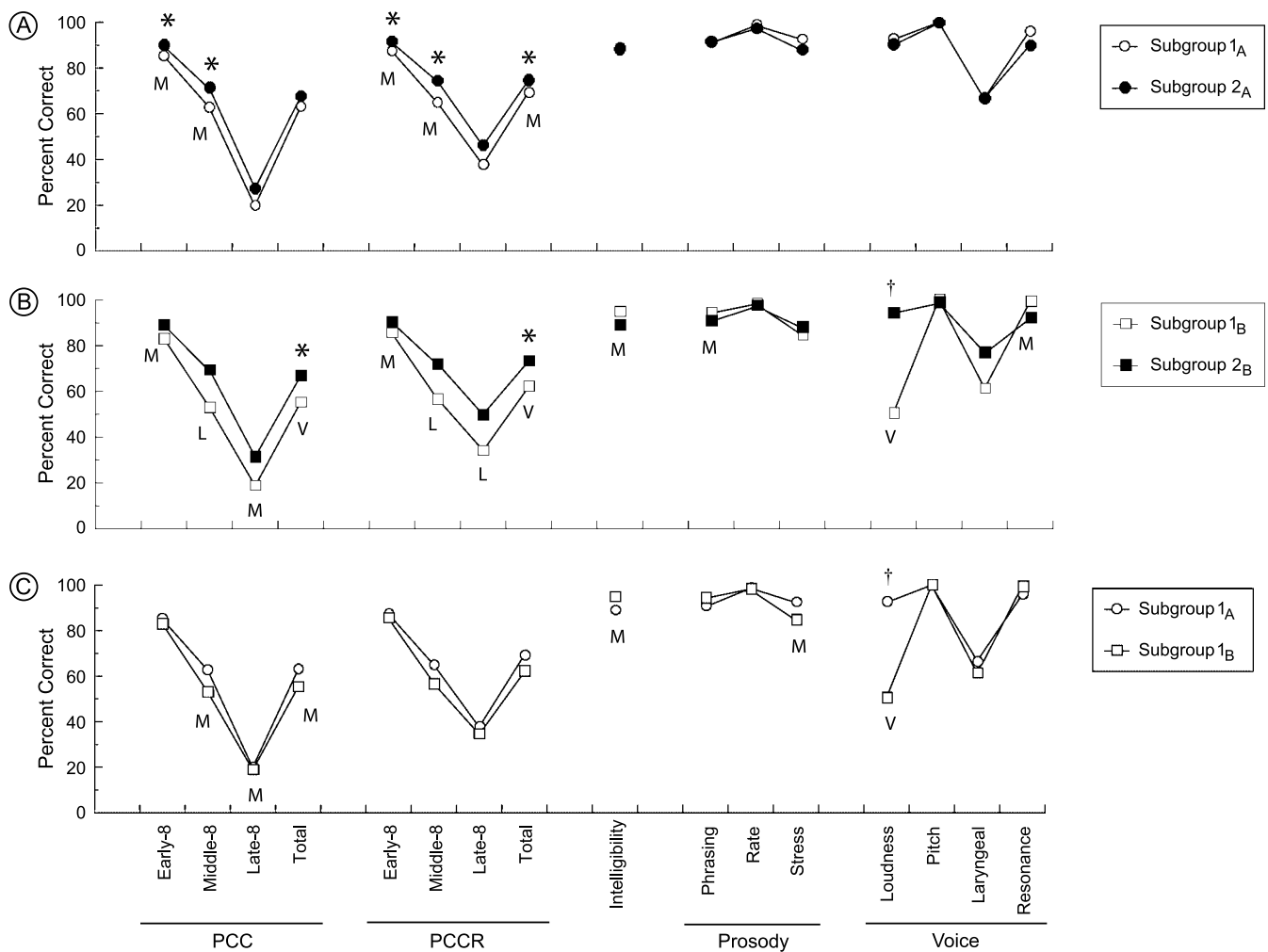
Figure 1 provides a summary of three comparisons: Subgroup 1_A ($n = 23$) with comparison subgroup 2_A ($n = 69$) in (Figure 1A), Subgroup 1_B ($n = 6$) with comparison subgroup 2_B ($n = 18$) (Figure 1B), and Subgroup 1_A with Subgroup 1_B (Figure 1C). Within each panel are

comparative data on several indices of speech competence, including (a) Early-8, Middle-8, Late-8, and Total values for the Percentage of Consonants Correct (PCC) and Percentage of Consonants Correct—Revised (PCCR) metrics (Shriberg, Austin, Lewis, McSweeney, & Wilson, 1997a); (b) the Intelligibility Index (II; Shriberg et al., 1997b); and (c) the percentage of utterances coded as appropriate for each of seven prosody–voice variables (Phrasing, Rate, Stress, Loudness, Pitch, Laryngeal Quality, and Resonance) included in the Prosody–Voice Screening Profile (PVSP; Shriberg, Kwiatkowski, & Rasmussen, 1990; Shriberg, Kwiatkowski, Rasmussen, Lof, & Miller, 1992). The PCC scores clinical distortions (e.g., dentalized /s/, derhotacized /r/, word-initial labialized /l/) as errors, whereas the PCCR scores all such subphonemic-level errors as correct (cf. Shriberg, 1993). The II metric is based on the number of words the transcriber is able to gloss from a conversational sample (Shriberg, 1993). Statistically significant *t*-test comparisons at an alpha level of .05 or greater are indicated by the conventional symbols placed above each such comparison in Figure 1, and effect sizes reaching at least moderate level are noted below each comparison (see key in Figure 1 caption). As summarized in the following three observations regarding the findings in Figure 1 and additional analyses, results from the preliminary analyses supported the pooling of Subgroups 1_A and 1_B (as well as the pooling of comparison subgroups 2_A and 2_B) for the primary analyses.

First, the directional trends were similar for the comparison of Subgroup 1_A with Subgroup 2_A (Figure 1A) and Subgroup 1_B with Subgroup 2_B (Figure 1B). In both sets of comparisons, participants with SD–DPI tended to have lower speech and prosody–voice competence, as assessed by the two speech metrics and their subscales, by the intelligibility metric, and by the seven prosody–voice variables. Owing to the differences in statistical power, more of the comparisons were statistically significant for the Subgroup 1_A comparisons (5 of 16) compared with Subgroup 1_B comparisons (3 of 16); however, the mean differences and associated effect sizes (which are less influenced by cell size) were generally greater for the comparisons between the 6 Subgroup 1_B participants and their comparison subgroup. The statistically significant difference (and *very large* effect size) associated with the number of utterances coded as “inappropriate in loudness” for Subgroup 1_B (50.4%) is consistent with the inclusionary criteria for this subgroup, as each of these utterances was judged “too soft.”

Second, the data in Panel C indicate that with the exception of the comparison in loudness, there were no statistically significant between-group differences on these measures. Subgroup 1_B participants generally

Figure 1. Perceptual support for pooling speech delay—developmental psychosocial involvement subgroups 1_A and 1_B (as well as the pooling of comparison subgroups 2_A and 2_B) for the primary analyses. M = moderate effect size; L = large effect size; V = very large; PCC = Percentage of Consonants Correct; PCCR = Percentage of Consonants Correct—Revised. Statistically significant comparisons are indicated by the conventional symbols located above each pair of findings: **p* < .05; †*p* < .01.



had lower speech competence as assessed by the two measures, but none of the comparisons were associated with an effect size greater than moderate.

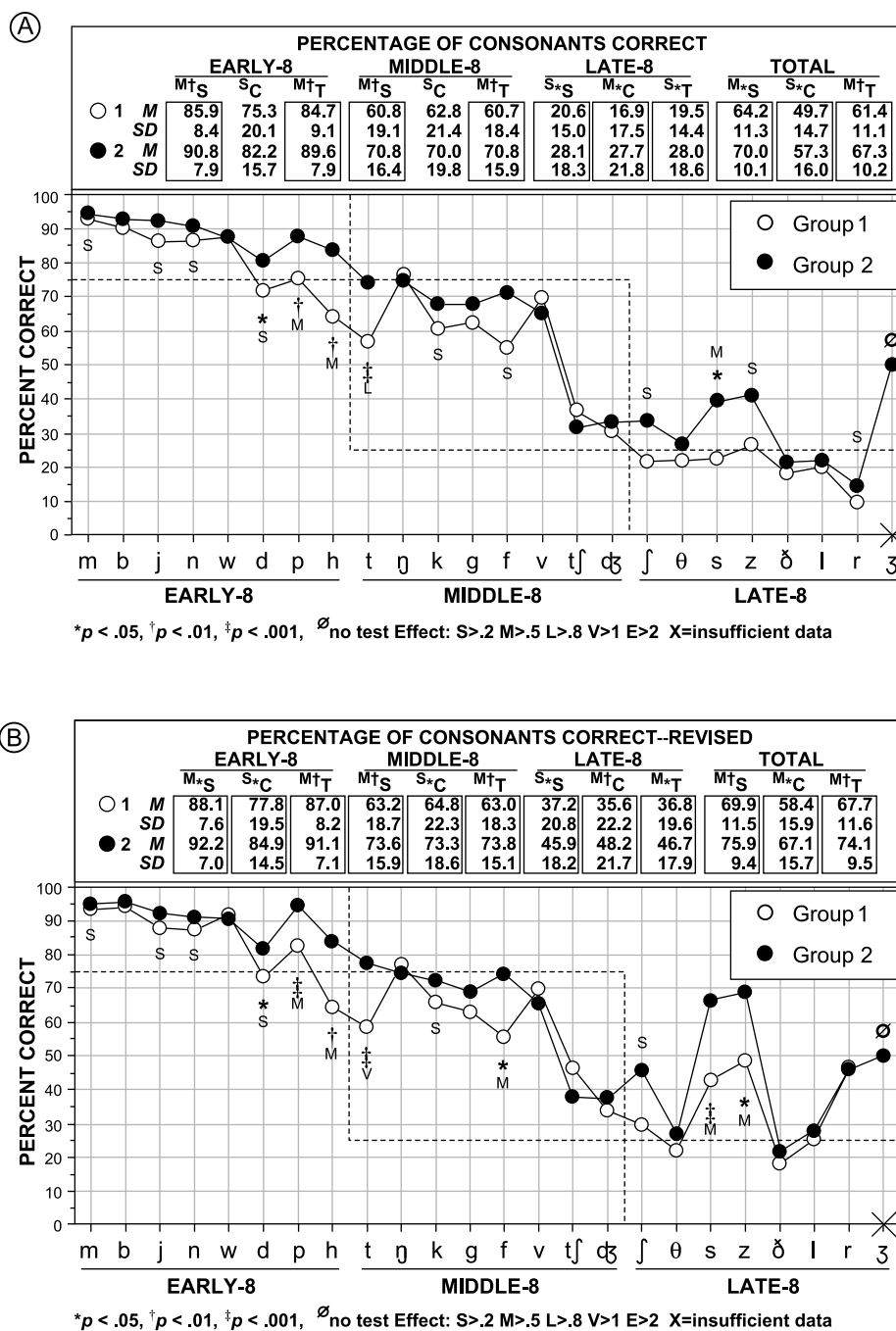
Third, supplemental analyses also supported pooling the two subgroups in the primary analyses. To provide a cross-check on the validity of the comparisons in Figure 1, participant scores on each of the nine speech variables were converted to *z* scores, using comparison scores on these variables obtained from typical speakers matched by age and gender (Austin & Shriberg, 1996). Findings were wholly consistent with those described above for the raw score comparisons in Figure 1, indicating no significant differences between Subgroups 1_A and 1_B when scores were adjusted for age and gender. Analyses were also completed for a number of other speech metrics, including (a) vowels and

diphthongs, grouped by place and height; (b) speech sounds aggregated by place, manner, and voicing features; and (c) individual consonants and vowels/diphthongs. Although the 6 participants in Subgroup 1_B tended to average somewhat lower scores than Subgroup 1_A participants, findings indicated that the error targets and error types were similar for the two SD–DPI subgroups.

Analyses of the Aggregated Group of Participants With SD–DPI Speech Comparisons

Indices of speech competence. Figure 2 contains summary descriptive and inferential statistics comparing the total group of children meeting criteria for SD–

Figure 2. Perceptual findings for the Percentage of Consonants Correct and the Percentage of Consonants Correct—Revised of participants with suspected speech delay—developmental psychosocial involvement (Group 1) versus comparison participants (Group 2). M = moderate; S = small; S = singleton; C = cluster; T = total.



DPI (Group 1; *n* = 29) with the total comparison group of children with undifferentiated SD (Group 2; *n* = 87). Note that these outputs, obtained from the PEPPER speech analysis suite (Shriberg et al., 2001), are accompanied by more statistical detail than shown in the Figure 1 index scores. Figure 2A contains speech

competence information at the narrow phonetic level of transcription (i.e., distortions are scored as incorrect) for the two groups, including the total PCC metric, the developmental sound class subscales, and the 23 consonant sounds (/ʒ/ occurs infrequently in young children’s conversational speech). Figure 2B provides speech

competence information on the same variables as assessed at the broad phonemic level of transcription (i.e., distortions are scored as correct) using the PCCR metric and subscales. In the numeric sections of Figure 2, statistically significant comparisons are marked by boxes framing the comparison data, by symbols indicating conventional levels of significance, and by letters denoting the magnitude of effect sizes (see key at the bottom of each panel). Statistically significant comparisons within the graphic section of both panels are similarly indicated by conventional symbols and effect-size abbreviations.

In Figure 2A, participants in the combined SD–DPI group (Group 1) had significantly lower scores on 10 of the 12 (83%) comparisons shown in the numeric panel, as well as on 5 of the 23 (22%) consonant phonemes. Significance levels ranged from alpha values that exceeded .05, .01, and .001, and effect sizes ranged from small to large. Thus, as tabulated by the total PCC index and by subscales based on developmental sound class (Early-8, Middle-8, Late-8), singletons and clusters, and individual consonants, participants in Group 1 had significantly lower speech competence scores than their comparison speakers. The average total PCC for Group 1 speakers was approximately 6 percentage points lower than the average PCC of the control group speakers.

The PCCR data in Figure 2B indicated even stronger between-group effects than those described above. The 29 Group 1 participants averaged significantly lower speech competence scores on all 12 (100%) of the metrics in the numeric section and on 7 of the 23 (30%) individual-consonant comparisons in the graphic section. Significant alpha levels again ranged from those exceeding .05 to .001, and effect sizes ranged from small to very large. Thus, as calculated at the levels of both narrow (PCC) and broad (PCCR) transcription, children meeting criteria for SD–DPI averaged significantly more severe SD. Statistical output similar to Figure 2 but aggregated by phonetic features (i.e., by obstruent and sonorant classes, voiced and voiceless consonants, and six manner features) indicated similar and substantially stronger between-group effects, with Group 1 participants averaging 9% to 12% lower speech competence than Group 2 participants.

Error-type analyses. Additional comparative analyses on other speech metrics (including phonetic inventories, vowels and diphthongs, intelligibility, and error types at broad and narrow levels of phonetic transcription) did not yield any additional informative comparisons, with most effect sizes for these measures limited to small or moderate magnitudes. Additional error-type analyses generally indicated that the pattern of errors was similar in the two groups, when adjusted for the obtained differences in severity as described

above. The source of the lowered scores for Group 1 participants with SD–DPI was a significant increase in substitution-type errors across each of the three developmental sound classes (Early-8, Middle-8, Late-8) and across the totals for all sounds (all with moderate effect sizes). However, when adjusted for severity differences, there were no significant between-group variations in the error-type proportions. As tabulated by class and manner features, the lowered speech scores for participants with SD–DPI were associated with significantly more frequent omissions and substitutions on obstruents, on voiceless consonants, and on the manner classes of stops and fricatives (with effect sizes ranging from small to very large). Again, there were no differences in the error-type profiles aggregated by feature when participant severity scores were relativized (i.e., when error types were percentaged on each participant's number of errors rather than on the number of attempted targets). Finally, at the level of specific types of speech sound distortions, there were no between-group differences in the proportional frequencies of clinical distortions (cf. Shriberg, 1993) on sibilant fricatives (dentalized, lateralized) or on rhotic sounds (derhotacized /r/, /ʁ/, or /ʁ/).

Estimate of the magnitude of speech delay. The error-type analyses supported the general perspective that participants with SD–DPI had significantly greater across-the-board SD than their matched-comparison participants (Group 2), with no specific error-type profile that might be sensitive and specific for this proposed subtype of speech delay. To gain a sense of the severity of SD in Groups 1 and 2, participants' total PCC and PCCR scores were compared with reference data on these metrics. A reference database was consulted that used speech sampling, transcription, and analysis procedures similar to those used in the present study (Austin & Shriberg, 1996, p. 11). The reference data for 4-year-old (the average age of the present participants; see Table 1) boys and girls with typically developing speech indicated average PCC scores of approximately 80% ($SD = 7.7$) and average PCCR scores of 93% ($SD = 4.9$). As shown in Figure 2, the total PCC scores for participants in Groups 1 and 2 were 61.4% ($SD = 11.1$) and 67.3% ($SD = 10.2$), respectively. Total PCCR scores for Groups 1 and 2 averaged 67.7% ($SD = 11.6$) and 74.1% ($SD = 9.5$), respectively. When expressed as a percentage of typically developing 4-year-olds' speech competence (i.e., $[\bar{X}_{\text{refdata}} - \bar{X}_{\text{group}}] / \bar{X}_{\text{refdata}}$), the comparison children in Group 2 had approximately 85% of the speech competence of typically speaking 4-year-old children on the PCC measure, whereas children in Group 1 had approximately 75% of typical competence. For the PCCR, Group 2 participants had approximately 80% of the speech competence of typical 4-year-old children,

whereas participants in Group 1 had approximately 73% typical competence. Thus, as with the raw data shown in Figures 1 and 2, the participants with SD–DPI averaged from 7% to 10% lower scores in speech competence measures than children comparable in age and gender with SD of unknown origin.

Prosody–Voice Comparisons

None of the seven prosody–voice comparisons between Groups 1 and 2 (phrasing, rate, stress, loudness, pitch, laryngeal quality, and resonance) were statistically significant or associated with effect sizes larger than small. However, at more detailed levels of these analyses, there was one set of statistically significant comparisons with moderate to large effect sizes. Participants in Group 1 required significantly more utterances to meet the requirement for a valid Prosody–Voice Screening Profile. Whereas an average of 16.1 ($SD = 23.4$) utterances from Group 2 participants had to be excluded from prosody–voice coding, an average of 39.4 ($SD = 27.7$) utterances from Group 1 participants were excluded ($p < .0001$; effect size = 0.94 [large]). Inspection of the 31 prosody–voice exclusion codes indicated that this significant difference was due to participants with SD–DPI producing more utterances that met criteria for one or more of the following six exclusion codes: Interruption/Overtalk ($p < .0004$; effect size = 0.91 [large]); Second Repetition ($p < .035$; effect size = 0.53 [moderate]); Negative Register ($p < .048$; effect size = 0.49 [small]); Sound Effect ($p < .007$; effect size = 0.69 [moderate]); Lip Smack ($p < .04$; effect size = 0.51 [moderate]); and Body Movement ($p < .004$; effect size = 0.73 [moderate]). With the exception of Second Repetition, each of these linguistic and paralinguistic events might be viewed as consistent with the behaviors used as inclusionary criteria for SD–DPI. Specifically, each exclusion code has at least face validity as more likely to occur in children having difficulty staying on task due to psychosocial involvement. Note that due to the relatively small numbers of events involved, these between-group comparisons only marginally met distributional criteria for the parametric statistical analyses (i.e., skew and kurtosis values often exceeded 2.0), and confidence intervals for the associated effect sizes were relatively large.

Diagnostic Marker Analyses

A series of multinomial logistic regressions, followed by artificial intelligence routines (Kiselev, Arseniev, & Flerov, 1994), was used to determine if a speech-severity metric or a combination of metrics was sufficiently sensitive and specific for SD–DPI. Scores for 25 metrics were entered into the analysis for each participant, including consonant indices (PCC, PCCR), cross-tabulated by developmental class (Early, Middle, and

Late) and context (singleton and cluster), vowel/diphthong indices and subindices, and intelligibility. None of the diagnostic marker analyses (i.e., sensitivity/specificity, positive/negative prediction, positive/negative likelihood ratios) met a minimal acceptance level (nominally, $\geq 75\%$ sensitivity and specificity). Because the pattern of target errors and error types in SD–DPI participants was best described as an across-the-board speech delay, there was no one index or combination of indices that could be used reliably to classify individual participants.

Discussion

Transactional Temperament Model

The consistency of the present data with previous findings suggests that psychosocial challenges may be a significant risk factor in speech development and in the normalization of SD. Findings suggest that a subset of preschool children with SD manifest a tendency toward either approach-related negative affect or withdrawal-related negative affect, as well as characteristics of negative emotionality/mood and decreased task persistence/attention. These findings support the hypothesis that individual differences in their temperament may mediate the severity of these children's speech delay. One possibility is that behavioral risk factors may have negatively affected typical environmental support for speech acquisition and/or the normalization of speech delay. Another alternative is that these children's psychosocial issues, although not likely a direct consequence of their speech delay, may indirectly reflect certain children's perceived pressures to communicate clearly. Many pathway models could be developed to reflect the complex of possible moderating and mediating associations among variables assessed in the current study as well as those not addressed.

The findings for task persistence/attention are viewed as especially compelling support for the hypothesis that negative affect combined with negative emotionality/mood and decreased task persistence/attention may act to increase the severity of speech delay as well as to impede normalization rates. Attention deficits were attested for 82% of participants in Subgroup 1_A (see Table 2). It was also this subgroup of participants who were most easily frustrated (96%) and least willing to work on general (67%) or difficult (85%) speech tasks. The significant correlation between the aggressive, inattentive temperaments of Subgroup 1_A participants and their lower AWU scores (although not as low as Subgroup 1_B) may reflect the fact that children who are more active and less attentive are less likely to produce long utterances. That is, AWU may be seen as a measure of talkativeness rather than language complexity.

Concerning Subgroup 1_B, children characterized as shy and withdrawn may be less motivated to communicate, which may function along with other factors to limit the development of their speech skills. The robust association between AWU scores and the tendency toward withdrawal-related negative affect (see Table 2) can be interpreted as supporting this position. The effects of social withdrawal in Subgroup 1_B participants may extend not only to these children's unwillingness to verbalize but also to their overall severity in speech delay (see Figure 1C).

Note that only 1 member of Subgroup 1_A (4%; see Table 2) was consistently willing to try difficult speech tasks, and only 3 (13%) were consistently motivated to work on general speech tasks, whereas the majority of Subgroup 1_B children consistently tried difficult speech tasks (60%) and were motivated to work on general speech tasks (75%). Subgroup 1_B participants' implicitly strong efforts to improve their intelligibility may not necessarily have met favorable results. Even though members of this subgroup were apparently more motivated to work on speech tasks than members of Subgroup 1_A, Subgroup 1_B's PCC and PCCR scores were substantially lower (despite the higher mean age of these participants). Thus, if a reliable finding, withdrawn social behavior may be a stronger causal correlate or mediator of delayed speech, compared with the risks and outcomes associated with aggressive, angry, or manipulative behaviors.

In summary, the transactional temperament model proposed in the present report addresses the mutual influence among environmental factors from a life span perspective. The behavioral characteristics a child displays at any point are rarely a function of the individual alone or of the experience itself, and viewing a child's development in this way may be misleading. It is probable that no one causal model can account for all relationships between communication disorders and psychosocial disorders. On this topic, Baltaxe and Simmons (1988) noted that the prenatal, perinatal, medical, social, and family histories of a child may all include significant variables that play a role in the development of communication and psychiatric disorders. Indeed, such risk factors were often included in the case histories of many participants in Group 1.

Implications for Treatment

Despite the present findings that speech and behavioral disorders co-occur in a clinically significant proportion of children, many of the secondary problems in each domain may go unrecognized depending on the primary diagnosis and clinical setting. The amount of overlap in developmental risk factors suggests that whether a child is eventually identified as having a speech disorder or a behavioral disorder may reflect the

salience of certain developmental symptoms to parents and professionals rather than their actual levels of severity in each domain. For example, for children whose frequent behavioral problems are most salient, intelligibility skills may not be assessed. Casby (1989) noted that in 1985 to 1986, only 9% of children in special education programs for behavioral disorders were also tested for speech–language needs. Conversely, for children who are identified on the basis of severe SD, difficulties in social interaction may be attributed to their communication problem and therefore not specifically addressed.

The present findings support a need for assessment and intervention models that integrate children's needs in speech and social interaction (e.g., Olswang, Coggins, & Timler, 2001; see conclusions in Law, Garrett, & Nye, 2003). One such model, the capability-focus treatment approach (Kwiatkowski & Shriberg, 1993, 1998), was developed expressly to support children's individual differences in ability and interactive styles. Speech–language pathologists must take into account the limitations of an assessment that does not include data (particularly from teachers and family members) on children's social and emotional uses of language in a variety of contexts (Audet & Ripich, 1992). It may be difficult to determine whether emerging behavioral deficits reflect emotional disturbances, communication impairments, or both.

Conclusion

Findings from this study are viewed as support for the hypothesis of a subtype of SD whose severity may be mediated by psychosocial variables. However, results are viewed as preliminary, with generalizations limited by the type and number of psychosocial variables assessed, the selection criteria used to classify children, the lack of formal information on psychological status of participants, and the sample size of children with suspected developmental psychosocial involvement. As constrained by these methodological limitations, both the primary finding that these children had more severe SD than children not meeting the psychosocial criteria developed for this study, as well as the finding that such children may constitute approximately 12% of this clinical population, motivate the need to cross-validate and extend the literature base on this possible subtype of SD. Additional research is needed to understand the antecedents and longer term consequences of psychosocial involvement on speech sound normalization, as such associations may differ from or interact with other proposed processes underlying subtypes of speech sound disorders (e.g., Shriberg et al., 2004). Although not identified in the present descriptive study, a major

research goal is to determine if there is a unique segmental, suprasegmental, and/or paralinguistic profile (i.e., diagnostic marker) that can be used by speech-language pathologists to discriminate these children from other children with SD of currently unknown origin. The linguistic features of such profiles are expected to contribute to theoretical frameworks toward an explanatory account of the role of psychosocial issues in atypical speech sound development.

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