4

Comorbidity of Speech-Language Disorder

Implications for a Phenotype Marker for Speech Delay

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COMORBIDITY, AS DEFINED IN AN epidemiological context, refers to "disease(s) that coexist in a study participant in addition to the index condition that is the subject of the study" (Last, 1988, p. 28). Information on the comorbidity of diseases within individuals is used by public health researchers and health care providers. For research purposes, comorbidity data can provide unique, descriptive, explanatory perspectives on the nature and origins of diseases, possibly leading to hypotheses about etiologic processes and alternative forms of treatment. For example, insights about the nature of a newly identified disease might be gained if comorbidity data indicated that it frequently co-occurred in individuals with a related and well-described disease. For applied concerns, comorbidity data guide health care providers in allocating the resources needed to identify and serve people with the co-occurring disorders in question. Thus, as a special type of epidemiological data, comorbidity information has the potential to significantly contribute to the study, treatment, and prevention of disease.

This chapter examines data on the comorbidity of two relatively prevalent public health concerns: child speech disorders and child language disorders. The focus is on measurement issues in child speech disorders, specifically on the potential contributions of comorbidity data to the goal of identifying a phe-

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notype marker for a genetically transmitted form of child speech disorders. Do comorbidity data suggest strong speech–language associations, supporting the likelihood of a common phenotype marker at a processing level that does not involve productive speech? Alternatively, do they suggest significant independence of the two disorders, which would support the need for a phenotype involving speech production? These issues relate to the general focus of this volume, that is, ways in which speech and language converge or diverge in development and disorders.

To appreciate historical, theoretical, and methodological perspectives on this issue, this chapter begins with a brief overview of research in child speech disorders and some background in contemporary speech-genetics research. A sample of available comorbidity data on speech-language disorders is then examined. The third section reviews a classification system for child speech disorders developed specifically for etiology studies, including genetics research. The fourth section is a presentation of results from four comorbidity studies using the new classification system. Findings suggest that the comorbidity of speech-language disorders is lower than previously reported, supporting a perspective that an autonomous phenotype for speech disorders is a valid need in speech-genetics research. As is discussed, findings also support a greater divergence of at least the productive aspects of phonological development and disorder relative to language development and disorder.

DEVELOPMENTAL PHONOLOGICAL DISORDERS

This section provides an overview of nosology and research trends in child speech disorders. Also discussed are relevant issues in speech-genetics research.

Nosology

Child speech disorders is the suggested cover term for all speech-sound disorders occurring during the developmental period for speech acquisition, nominally from birth to 9 years. The most prevalent form of child speech disorders is referenced by a number of labels, including functional articulation disorder; developmental phonological disorder; hybrids such as articulation/phonological disorder; and less theoretically committed terms such as multiple phoneme disorder, speech delay, or intelligibility impairments. The discipline of communicative disorders continues to accommodate this multiplicity of terms at least in part because of the lack of theoretical clarity on the role of cognitive-linguistic, sensorimotor, and psychosocial substrates as original and/or maintaining causes of speech disorder. Whichever is the preferred term, this form of child speech disorder is distinguished from those forms for which causal origins have been identified (e.g., speech disorders associated with craniofacial dysmorphologies, sensorimotor impairments, pervasive developmental disabilities). As described in this chapter, the term *special populations* subsumes all developmental phonological disorders associated with known etiologies. The authors of this chapter use the term *developmental phonological disorder* (DPD) for the form of child speech disorder characterized by mild to severe speech involvement of unknown origin.

The terminological situation described is familiar to those involved in the study and treatment of language disorders in children, where a parallel literature on classification systems and alternative clinical-research labels includes equally lively dialogue (e.g., Aram, Morris, & Hall, 1993). It would be efficient to use *child language disorders* as the companion cover term to child speech disorders and, in turn, the term *developmental language disorder* (DLD) for children with language disorders of unknown origin. However, to avoid adding another term to the mix, we use the well-entrenched, albeit contentious, term *specific language impairment* (SLI) for children with DLD of unknown origin. In cases where researchers may not adhere to or provide information on inclusionary and exclusionary criteria associated with SLI, DLD is a useful cover and companion term to DPD.

Research Trends

A brief review of research in developmental phonological disorders suggests why there are few reliable data on the comorbidity of DPD and SLI (viz., DLD). For this purpose, the 60 years of research in developmental phonological disorders between 1930 and 1990 can be divided into two 30-year periods.

1930–1960 From approximately the 1930s through the 1950s, there was considerable research activity attempting to associate the origins of all forms of child speech disorders to clinical or subclinical (i.e., suspect) differences in children's speech-hearing mechanisms, cognitive-linguistic functions, or psychosocial processes. Although impressive gains were made in understanding why and how children in special populations had difficulties producing intelligible speech, syntheses of the classic studies of this period conclude that no single cause for DPD was identified (Bernthal & Bankson, 1993; Creaghead, Newman, & Secord, 1989; Shriberg, 1980; Stoel-Gammon & Dunn, 1985; Weiss, Gordon, & Lillywhite, 1987; Winitz, 1969; Winitz & Darley, 1980). A crucial methodological observation is that studies undertaken during this period treated DPD as a unitary disorder that varied in severity of involvement. That is, compared with theoretical perspectives and measurement approaches to phonological comprehension, phonological organization, and speech production in the 1990s, the measurement of both speech and language status in the studies of 1930–1960 reflected a limited sector of the relevant variables.

1960–1990 A second period of research, from approximately the 1960s to the late 1980s, was characterized by the absence of widespread programmatic explanatory studies of child speech disorders, both within special populations and in DPD. This period included paradigmatic research shifts to alternative methods for linguistic description of the speech of children with all forms of child speech disorders, regardless of etiology, and acquisition and treatment strategies stressing associationist psychology. Some theoretical evolutions from these linguistic and behaviorist lineages, respectively, are emerging nonlinear phonological theories that describe developmental phonological disorders (e.g., Bernhardt & Stemberger, 1997; Goldsmith, 1995; Schwartz, 1992) and emerging neural network models of speech-sound acquisition and performance (e.g., Menn, Markey, Mozer, & Lewis, 1993; Stemberger, 1992). Consistent with the explanatory levels in such frameworks, individual differences in distal causes (i.e., etiology) are not represented as relevant independent variables.

1990s The 1990s mark the onset of a third period of research in child speech disorders in which there is a notable return to studies directed at pathogenesis, including both distal and proximal causes, and to clinical subtyping. Among other putative etiologic origins of speech-language disorders, perhaps most promising is the possibility that there may be a familial form expressed as a genetically transmitted trait. As introduced previously, a crucial need in pursuit of this hypothesis is the development of one or more phenotype markers for use in behavioral and molecular genetics designs. The following discussion is an overview of the relevant issues.

Speech-Genetics Research

The hypothesis that genetic transmission is the distal origin of the most common form of a speech-language disorder has gained a significant base of support in classic and emerging findings using twin and familial aggregation designs (e.g., Arnold, 1961; Beitchman, Hood, & Inglis, 1992; Bishop, 1992; Bishop, North, & Donlan, 1995; Borges-Orsorio & Salzano, 1985; Felsenfeld, Broen, & McGue, 1992; Gopnik & Crago, 1991; Hurst, Baraitser, Auger, Grahm, & Norell, 1990; Ingram, 1959; Lahey & Edwards, 1995; Lewis, Cox, & Byard, 1993; Lewis, Ekelman, & Aram, 1989; Lewis & Thompson, 1992; Locke & Mather, 1989; Matheny & Bruggeman, 1973; Neils & Aram, 1986; Samples & Lane, 1985; Schuele & Rice, 1996; Shriberg & Kwiatkowski, 1994; Stromswold & Rifkin, 1996; Tallal, Ross, & Curtiss, 1989; Tallal, Townsend, Curtiss, & Wulfeck, 1991; Tomblin, 1989). The data on familial aggregation, for example, indicate that more than half of the children with SLI have at least one other family member with a history of speech-language and/or learning disorders (usefully termed SLLD in Lahey & Edwards, 1995), including the widely cited highest proportion of .77 reported by Tallal et al. (1989). Whitehurst et al. (1991) did not find higher proportions of family members with SLLD in the families of the children they studied; but, as these authors pointed out, the probands were young children whose language involvements were limited to expressive language disorder. Perhaps the most significant finding in behavioral genetic studies was reported by Felsenfeld and Plomin (1997), who tested children followed in the Colorado Adoption Project. These authors examined speech outcomes in 156 adopted and nonadopted children at varying risk for speech disorder based on self-reported parental speech history. Children with an affected biological parent were significantly more likely to have a speech disorder (approximately three times the risk) compared with children raised by an affected parent and children with no parental history. Rice (1996) provides excellent coverage of the scope of genetic research in SLI, and Gilger (1995) provides an informative tutorial on genetic methods applied to communicative disorders.

As emphasized in all other areas of human genetics, the major methodological problem in speech-genetics research is the development of an appropriate phenotype (cf. Brzustowicz, 1996). The techniques for behavioral analysis of family and twin data and molecular analysis of tissue samples are increasingly accessible, including procedures to assess how the disorder is genetically transmitted and eventually the genetic coding for the disorder. It is the identification of a phenotype marker for speech-genetics research that presents the formidable challenge. Candidate markers are likely to arise from one of three sources:

1. Clinical diagnostic marker
2. Research diagnostic marker
3. Autonomous speech marker

Clinical Diagnostic Marker It might turn out that one of the common diagnostic markers used to classify a child as having a speech disorder in educational or clinical environments, for example, a score on a standardized articulation test, will have the sensitivity and specificity required for a phenotype marker in genetics studies. Most behavioral genetics studies have used articulation test scores to identify affected children. When classification of probands or relatives is based on presence or absence of a disorder, the diagnostic marker provides only a qualitative index. Several of the diagnostic classifications in the Speech Disorders Classification System (SDCS) (Shriberg, 1993), described later in this chapter, could also serve as a qualitative phenotype marker. When a score or standardized score is used, a common practice in twin concordance studies, the diagnostic marker is a quantitative index (cf. Pennington, 1986). If the history of research in medical and psychiatric genetics is instructive for speech-language genetics, diagnostic markers taken directly from the clinic are not likely to have the sensitivity and specificity needed for

genetics research (cf. Smith, Pennington, & DeFries, 1996; Tomblin, Freese, & Records, 1992).

Research Diagnostic Marker A second possibility is that speech and language measures will need to be developed or modified to identify a phenotype marker that meets behavioral and molecular genetics research needs. Some specific linguistic and psychometric needs for speech testing in multigenerational, multidialectal, and multicultural populations have been considered elsewhere (Shriberg, 1993). Potential phenotype markers may come from levels of speech processing other than natural speech production, especially processing levels that might claim closer association with developmental neurobiological processes (cf. Pennington, 1986). Thus, in addition to phenotypes based on productive speech errors on traditional citation form tasks (e.g., Felsenfeld et al., 1992) and conversational speech samples (e.g., Shriberg, 1993; Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997a, 1997b), the speech-genetics and related literature includes potential phenotype markers based on auditory-processing tasks (cf. Farmer & Klein, 1995), speech perception (e.g., Boada et al., 1998), multisyllabic and nonsense word repetition tasks (e.g., Bishop, 1994; Lewis & Freebairn, 1992), and phonological awareness tasks (e.g., Bird, Bishop, & Freeman, 1995).

Autonomous Speech Marker A third possibility is that whichever the level of assessment, the phenotype for developmental phonological disorders might need to differ from markers associated with other verbal trait disorders. Such a marker might be called an autonomous speech phenotype to differentiate it from all processing markers that do not directly assess the speech signal. There are many places to look for such markers in relation to the developmental neurobiology of central and peripheral speech systems (Christman, 1995). Although academic and clinical disciplines treat developmental speech disorders and developmental language disorders as distinct entities for the purposes of diagnostic classification (cf. American Psychiatric Association, 1994), the distinction between speech processing and language processing is often blurred in genetics research.

Our own research focuses on the utility of identifying a phenotype marker of DPD in conversational speech. The primary constraint on such a marker is that it may no longer be available in people (i.e., relatives of probands) whose speech has normalized, that is, those who no longer have speech production errors. We are currently using acoustic analysis techniques to determine if markers for prior or continuing disorder might be retrievable at the level of subphonemic description. The magnitude of comorbidity estimates for co-occurring speech-language disorders should inform research among the three possibilities previously described. At least for a first approximation, the higher the comorbidity rates, the less likely the need for an autonomous speech phenotype.

Summary

The 1990s mark the onset of an exciting period in speech-language disorders in which there is a real possibility to learn why some children have significant difficulty acquiring speech and language. Toward that end, data on the comorbidity of child speech and language disorders might provide important insights. Useful comorbidity research requires classification systems for the disorders under study and consensus on diagnostic criteria for each of the disorders. In a widely cited review and study of the prevalence of speech and language disorders, Beitchman, Nair, Clegg, and Patel stated, "The lack of a comprehensive classification system for speech and language disorders is . . . a serious barrier to developing useful and accurate prevalence estimates" (1986, p. 98). The following section describes a classification system for speech-sound disorders that is used in four new comorbidity studies described later in this chapter.

THE SPEECH DISORDERS CLASSIFICATION SYSTEM

The SDCS shown in Figure 1 was designed to classify speech disorders throughout the life span. It includes primary classification categories that account for number and type of speech errors, suspected etiological subtypes, and course of the disorder. The latter is particularly important in genetics research, as gene regulation issues affecting the onset of disorder may also underlie the relative time course of normalization and topographic changes within different time periods. The SDCS was first described in Shriberg (1993), updated in Shriberg (1994), and finalized in Shriberg et al. (1997b). Formally, the system is structured to reflect both descriptive classification (solid lines) and etiologic explanatory classification (dashed lines; cf. Shriberg, 1982). The eight boxes in the top two rows of Figure 1 indicate the major typological classifications in the SDCS system. Developmental phonological disorders subsumes two forms of child speech disorders: speech delay and residual errors. Speech delay subsumes four etiological subtypes, shown in dashed lines, as well as speech delays of known origin in special populations. Residual errors, the other form of a developmental phonological disorder, subsumes six descriptive subtypes.

The discussion in this section, which closely follows another description of the SDCS (Shriberg, 1997), highlights emerging research findings for the 20 classification categories that compose the SDCS portrayed in Figure 1. Preliminary prevalence estimates are included for each disorder classification, data that bear on comorbidity and phenotype issues. Especially for the reader less familiar with the speech side of the speech–language connection, this review provides a perspective on what was described prior to the 1990s as a monolithic functional articulation problem.

Normal and Normalized Speech

The upper-left-most box in Figure 1 includes two descriptive classifications, which accounts for the 20 SDCS classifications in 19 boxes. Speakers with normal speech for their age at assessment and no history of nonnormal speech are classified as having normal speech acquisition (NSA). The SDCS program uses tables of reference data and a set of inclusionary and exclusionary criteria to determine if a conversational speech sample from a speaker meets criteria for NSA (Shriberg, 1993; Shriberg et al., 1997b). People with documented histories of earlier speech delay are classified as having normalized speech acquisition. Longitudinal studies estimate 9 years as the approximate end point of the typical developmental period, with additional productive speech development at allophonic and prosodic levels occurring up to 12 years of age (cf. Shriberg, Gruber, & Kwiatkowski, 1994; Shriberg, Kwiatkowski, & Gruber, 1994). Another important point in time for biobehavioral development observed in our longitudinal studies appears to be 6 years of age, when 75% of children with speech delay normalize their speech disorders (i.e., are classified as NSA by the SDCS program) (Shriberg, Gruber, et al., 1994).

Nondevelopmental Speech Disorders and Speech Differences

The two right-most boxes in Figure 1 represent two categories of speech patterns in children or adults that either are not developmental in origin or are not appropriately classified as a speech disorder. Nondevelopmental speech disorders is an appropriate classification for all speech disorders that occur after the developmental period for speech, nominally 9 years of age. This classification includes the full scope of adult disorders, including those due to trauma, disease, and various physical and emotional illnesses. The study and treatment of adult-onset speech disorders is a complex topic, but differential diagnosis is not complicated by those speech-hearing mechanism, cognitive-linguistic, and psychosocial processes that are associated with children's growth and development.

The right-most classification in the first row of Figure 1 is a category for child and adult speakers with speech differences. This category includes all speech and prosody-voice differences that speakers might elect to modify, such as those associated with mastery of English as a second language or the speech and prosody-voice patterns of children or adults speaking an English vernacular. As indicated by the dashed line connecting this box to the others, the discipline of speech-language pathology clearly differentiates speech differences from speech disorders. However, because of the similarity in assessment and instructional techniques for differences and disorders, it is useful to tie this classification category to the categories of speech disorder in the SDCS. Service providers increasingly need information on the comorbidity of

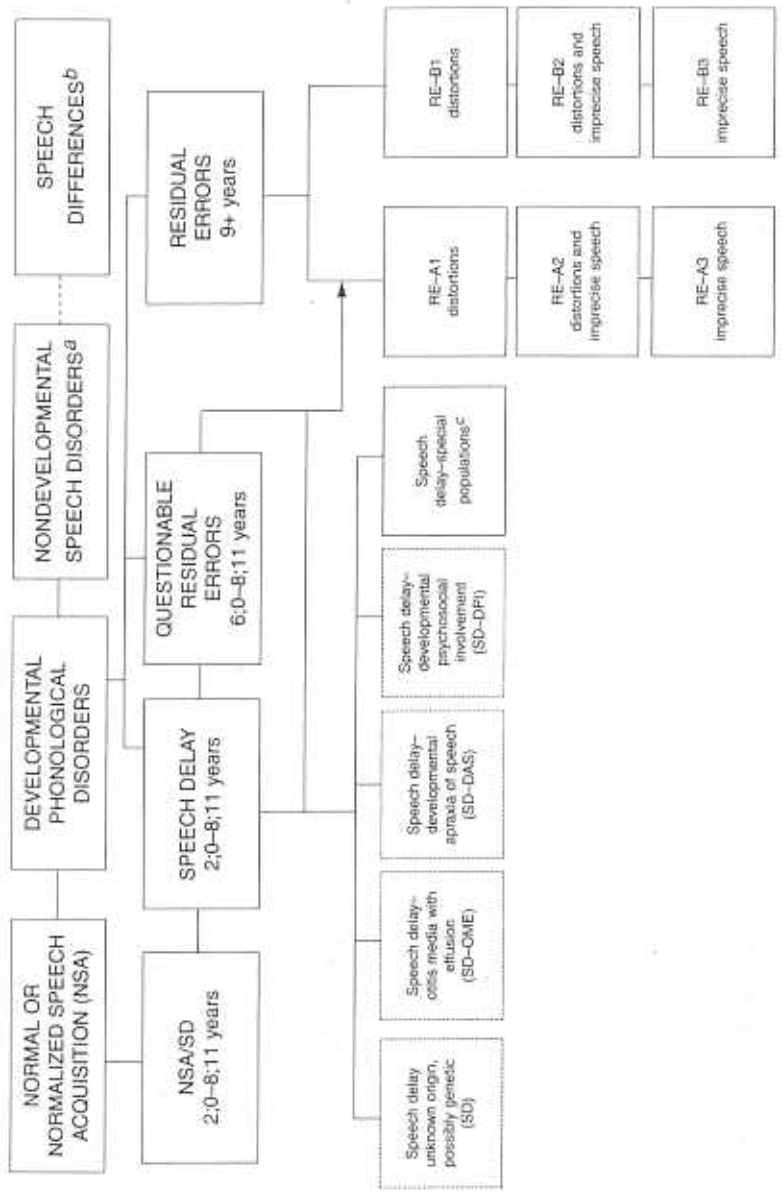

Figure 1. The Speech Disorders Classification System (SDCS). [a]Speech disorders first occurring after 9 years of age; [b]Nondevelopmental speech differences such as those associated with cultural diversity and accent reduction; [c]Speech delay in special populations such as those associated with craniofacial and sensorimotor involvements.)

speech-language disorders in children from diverse linguistic and multicultural backgrounds.

Normal Speech Acquisition/Speech Delay

The need for a hybrid classification termed *normal* (or *normalized*) *speech acquisition/speech delay* (NSA/SD) is a familiar issue in epidemiologic research. Although accounting needs in service delivery systems typically require dichotomous classifications of people as either normal or affected, few diseases or disorders divide in this fashion. Especially if the underlying trait or disorder is continuously distributed in a population, arbitrary cutoff points yield children whose status is actually intermediate between normal and affected. An example is the procedure used to classify children in the Collaborative Perinatal Project of the National Institute of Neurological and Communicative Disorders and Stroke (NINCDS) (Lassman, Fisch, Vetter, & La Benz, 1980), discussed later in this chapter. Using statistical criteria not relevant here, researchers classified all speech, language, and hearing scores into three categories: normal, suspect, and abnormal. An intermediate category was needed for both initial identification of disorder and for classification preceding complete normalization of many disorders. Similarly, in addition to NSA and SD, the SDCS classification NSA/SD indicates status between normal or normalized speech acquisition and speech delay. As shown in Figure 1, children classified as NSA/SD may meet criteria for questionable residual errors (QREs) at 6 years of age, may normalize completely at any age up to 8 years, or may meet criteria for residual errors (REs) at 9 years of age if they continue to have speech errors.

The data in Figure 2 support the validity of NSA/SD as a classification category. These data are the SDCS classifications of 586 children whose conversational speech was sampled in several different studies (Shriberg, 1997). The metric used to assess speech in conversation, the Percentage of Consonants Correct–Revised (PCC–R), scores all correct sounds and distortion errors as phonemically correct (Shriberg, Austin, et al., 1997a), with incorrect sounds including all phoneme deletions and substitutions. The means and standard deviation bars in Figure 2 indicate that the PCC–R scores differ for 3- to 6-year-old children classified as NSA, NSA/SD, and SD. The mean PCC–R scores for children classified as NSA/SD fall generally between the trends for NSA and SD at each age. The standard deviation bars also indicate good separation among the distributions for each classification group, suggesting that the scores of most children in this group fall in between the scores of most children in the other two groups. As described in the next section, we use the term *subclinical* as the diagnostic label for children classified as NSA/SD by the SDCS program. This classification is important in the comorbidity studies reported later in this chapter.

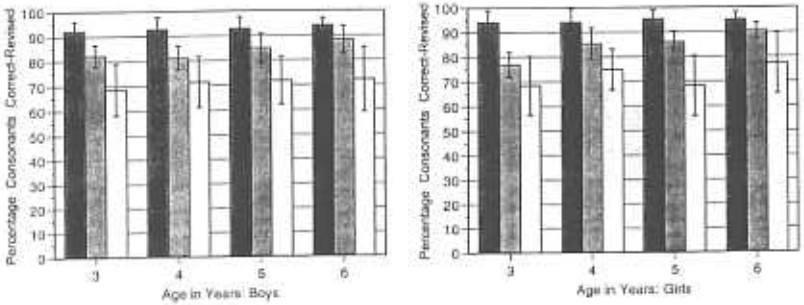

Figure 2. Age trends (means, standard deviations) for 3- to 6-year-old boys and girls classified by the SDCS as having normal or normalized speech acquisition (NSA), intermediate status (NSA/SD), or speech delay (SD). (■, normal speech acquisition; ■, normal speech acquisition/speech delay; □, speech delay.)

Speech Delay

Speech delay, or *SD*, has been proposed as the appropriate nosological term for the estimated 2%–3% of preschool children with a developmental phonological disorder of unknown origin (Leske, 1981). The SDCS classifies a child as having SD when age-inappropriate speech-sound deletions and/or substitutions occur in a conversational speech sample. As described, some children normalize SD by 6 years of age, some completely normalize between 6 and 8 years of age, some retain QRE from 6 to 8 years, and some retain residual errors after 9 years of age. Such different normalization histories might be important to the development of a behavioral phenotype for speech disorders, with gene-to-behavior pathways being a possible governing source for mechanisms underlying different normalization histories. In a study of language acquisition in which trends for heritability coefficients were higher in older monozygotic twin pairs, Stromswold and Rifkin (1996) suggested that the role of genetic factors in language acquisition may increase as children get older. From a methodological perspective, as considered in the studies reported later in this chapter, normalization rates play an important role in the interpretation of prevalence and comorbidity estimates.

The boxes subsumed under speech delay in Figure 1 comprise four proposed etiological subtypes of developmental phonological disorders and a classification for children in special populations with known causes. Information on the biolinguistic and sociolinguistic correlates of each disorder will inform research in the others, toward an eventual account of the origin and course of all child speech disorders. This section continues with brief comments on each of the proposed etiological subtypes; rationale and preliminary estimates of epidemiological and other nonspeech data (e.g., prevalence, gender ratio, language status, speech-sound normalization history, residual error status, familial aggregation) are provided in Shriberg (1994).

Speech Delay The most prevalent type of SD is simply termed *speech delay*. Based solely on a clinical database of approximately 350 children re-

ferred to a university phonology clinic for assessment or treatment of a phonological problem of unknown origin, a preliminary estimate is that this form of SD occurs in perhaps 60% of preschool children identified by speech-language pathologists as having a developmental phonological disorder (Shriberg, 1994). These are essentially the children whose case data and assessment results suggest no significant involvement in speech-mechanism, cognitive-linguistic, or psychosocial processes. That is, there is no one area of involvement or information in the case data that could be posited as a sufficient explanation for the presenting speech problem.

Emerging support for the possibility of a genetic origin for this form of a child speech disorder was cited previously. The primary source of evidence for genetic transmission of SD is the high prevalence of verbal trait impairments in nuclear and extended families of children with speech-language disorders. Studies have reported familial aggregation in 20% to nearly 60% of children (i.e., probands) ascertained on the basis of a speech-language disorder (cf. Felsenfeld, McGue, & Broen, 1995; Lahey & Edwards, 1995). The authors' data indicate that 56% of a sample of 84 children with SD had one or more family members with (or who previously had) the same speech problem (Shriberg & Kwiatkowski, 1994). For this proposed subtype of speech delay, the research challenges are to explicate the underlying neurolinguistic processing deficits and eventually the sociobiological origins of those deficits. In the context of this chapter, the focus is on the crucial importance for genetics studies of identifying the correct phenotype markers for SD.

Speech Delay–Otitis Media with Effusion A second possible more proximal origin of speech delay is linguistic processing deficits associated with fluctuant conductive hearing loss due to early recurrent otitis media with effusion (i.e., speech delay–otitis media with effusion [SD–OME]). Many children referred to our speech clinic for suspected speech delay have had nearly continuous OME during some period of the first 3 years of life and beyond. The case histories of such children typically include ample evidence of fluctuant hearing loss in one or both ears. Analysis of two cohorts of children, one group followed in a university pediatrics clinic and another group followed in a Native American tribal health clinic, indicated that early recurrent OME is a significant risk factor for speech disorder (Shriberg, Flipsen, et al., 1997). Based on the percentage of children with SD in a university phonology clinic who have suspected SD–OME, a preliminary prevalence estimate is that this form of SD constitutes approximately 30% of preschool children with speech delay of unknown origin.

The language characteristics of children with SD–OME have yet to be compared with the language profiles of children with other forms of SD as well as with profiles for children with other forms of SLI. The auditory-perceptual consequences of fluctuant conductive hearing loss can be used to generate hypotheses about effects of OME and the acquisition of certain lexical and

morphosyntactic forms. The primary finding is that SD–OME appears to be associated with reduced intelligibility, lower than expected for a child's speech status (Shriberg, Flipsen, et al., 1997). The intelligibility deficit is likely to reflect an interaction with language variables. Neither the prevalence nor the potentially unique speech-language consequences of this proposed subtype of SD (which, like the other suspected forms of SD, could also have unique distal genetic origins) can be estimated until one or more diagnostic markers for SD–OME are identified.

Speech Delay–Developmental Apraxia of Speech A third possible subtype of SD shown in Figure 1 is a form suspected to resemble acquired apraxia of speech in adults. The acquired form of apraxia (adult apraxia of speech) meets criteria for one of the SDCS classifications aggregated within the category termed nondevelopmental speech disorder (see Figure 1). To be consistent with the most widely used term for this form of speech delay, the SDCS classification for this subtype is *speech delay–developmental apraxia of speech* (SD–DAS).

In a study series, we reported that approximately half of a total of 53 children with suspected DAS have inappropriate sentential stress (Shriberg, Aram, & Kwiatkowski, 1997). The specific pattern observed in this work and assessed in subsequent linguistic analyses (e.g., Velleman & Shriberg, 1997) is one of excessive-equal stress. This prosodic difference is proposed as a diagnostic marker for at least a subgroup of children with suspected SD–DAS. Inappropriate stress of the type coded in these studies is seldom observed in children acquiring speech typically or with any of the other suspected forms of SD shown in Figure 1. The reported high familial aggregation in suspected SD–DAS (Hall, Jordan, & Robin, 1993) suggests that this prosodic diagnostic marker might also serve as a phenotype marker. Using the same clinical population as previously cited, we estimate that children with this suspected subtype of SD compose 3%–5% of all children referred for a developmental speech disorder of unknown origin.

Speech Delay–Developmental Psychosocial Involvement The fourth type of speech delay shown in Figure 1, speech delay–developmental psychosocial involvement (SD–DPI), is the most speculative of the SD subtypes in the SDCS. The proposal that psychosocial issues can be causally sufficient for speech delay is predicated on the observations of pioneer speech pathologists such as Charles Van Riper, Wendall Johnson, and Muriel Morley. Each of these perceptive clinical researchers has reported psychosocial needs in some children with speech delay. These observations are supported in several surveys (e.g., Baker & Cantwell, 1982; Beitchman, Nair, Clegg, & Patel, 1986) and follow-up studies (e.g., Shriberg & Kwiatkowski, 1988) documenting psychosocial needs in some children with suspected SD–DPI. Based solely on the authors' local clinical referral estimates, children with SD–DPI and/or some other form of SD compose approximately 7% of the remaining children

referred for speech delay of unknown origin (i.e., SD: 60%; SD–OME: 30%; SD–DAS: 3%; SD–DPI and others: 7%).

There are only preliminary proposals for diagnostic markers for children with SD–DPI. A leading candidate is in the prosody-voice domain during discourse, the feature of speech that reflects affective states and traits. Children with suspected SD-DPI exhibit moment-to-moment variability in prosody-voice domains as well as retest variability on indices of segmental competence. Relative to language characteristics, some children with suspected SD–DPI seem to have considerable difficulty with the types of dyadic activities that most children like and from which they learn. As with each of the other proposed etiological subtypes, eventual validation of a subtype of speech delay associated with psychosocial issues would be presumed to have useful implications for the form and content of treatment.

Speech Delay–Special Populations The many special populations within the current scope of practice in speech pathology demand ever-increasing clinical competencies. Inclusion of this cover term for children with exceptional needs in addition to their speech needs emphasizes two considerations: 1) These children have developmentally related speech needs in addition to their unique speech involvements, and 2) clinical decision making for these children is informed by findings in other proposed subtypes of speech delay. This perspective also emphasizes the need for integrated speech-language research, as discussed in several chapters in this volume. However, much of the large-scale research in special populations since the 1970s has focused on either the speech domain or the language domain but seldom on both using well-developed methods in the same project. Thus, the inclusion of special populations in the SDCS encourages clinical–research rapprochement among all types of speech disorders and among speech-language domains in the same individual that too often are studied and treated as independent systems.

Questionable Residual Errors

During the period from ages 6 to 8 years, there is one other type of developmental speech disorder that a child can have in addition to SD or NSA/SD. The classification QRE is used for children who have one or more speech-sound distortions or common substitutions (e.g., distortions of fricatives or liquids, substitutions of /θ/ for /s/ or /w/ for /r/ and others), errors that do not meet SDCS criteria for SD or NSA/SD (which would require significant age-inappropriate deletions and substitutions). Some children with QRE could be children who normalized from SD or NSA/SD status, or, as shown in Figure 1, they could have no such history. The importance of this distinction in the time course of the disorder is discussed in the following section.

Residual Errors

As noted previously, speech errors may continue as the residuals of a speech delay, or they may occur without such histories. Residual errors is the classification for children and adults with speech-sound errors that are developmental in onset and persistent beyond 9 years of age, whereas QRE is the appropriate classification for children with articulatory errors occurring from the 6- to 8-year-old period when they may still normalize.

The SDCS has six subtypes of RE based on a speaker's history and error pattern. Speakers ages 9 years and older with histories of SD are classified as RE-A. Children or adults with speech errors but no history of speech delay are classified as RE-B. The three numbered RE subtypes in Figure 1 can be used to further classify error patterns first occurring during the developmental period. Those following a history of some form of SD are included in the A alternatives; those not associated with SD are included in the B alternatives. RE-A1 or RE-B1 is for residual common distortion errors (see Shriberg, 1993; Appendix). RE-A2 and RE-B2 are for residual common distortions and imprecise speech (omissions and substitutions). RE-A3 and RE-B3 are for imprecise speech. Although there are no reliable prevalence data, preliminary findings indicate that each of these error types occurs in adult speech (Shriberg et al., 1997b). In one study of 112 adult relatives of children identified as having speech errors, there were some notable gender trends for distortion errors (e.g., dentalized fricatives versus derhotacized /r/, /ɝ/, and /ə/) that persisted in all age groups, including speakers older than 50 years of age (Lewis & Shriberg, 1994). As observed elsewhere in our database studies, fricative distortions may be more prevalent in females and derhotacized /r/ and rhotic vowels more prevalent in males. For phenotype issues as well as more general questions about the origin of speech-sound distortions, large-scale studies of RE could be productive. Decreased articulatory precision, as it may first occur in some adult populations, would be classified in the first row in Figure 1 as a nondevelopmental speech disorder if associated with a disorder or specific event (e.g., stroke, traumatic brain injury); it would be classified as a speech difference if associated with typical aging.

What is important about children with RE and QRE is whether they should be included in a tabulation of people with speech disorders in prevalence or comorbidity studies. If they do not have an earlier history of SD, are such behavioral differences consistent with the genotype posited to underlie speech delay? Specifically, does RE reflect simply a less severe expression of the genes that code for SD, or do common distortions and substitutions arise from sources different than those posited to underlie SD? If considered the same disorder, are children who persist from QRE to RE more severely involved than children who normalize QRE, or do they differ in some qualitative way from children who normalize QRE during the developmental period?

These questions and the extended review of the SDCS were provided to highlight both the complexity and the importance of such detail for the speech side of the speech-language comorbidity research reviewed in the next section of this chapter.

COMORBIDITY OF SPEECH AND LANGUAGE DISORDERS

It is useful to begin a discussion of comorbidity (also termed *concomitant* [Paul, 1995] or *coexisting* [St. Louis, Ruscello, & Lundeen, 1992] disorders) with a brief review of some alternative ways to express the prevalence and comorbidity of diseases or disorders.

Prevalence Estimates

In genetics research, prevalence estimates for a disorder provide the unconditional probabilities or liabilities against which the prevalence of a disorder in a specific target group (e.g., an extended family) is compared. The prevalence percentage of a disorder or disease is computed by dividing the number of people positive for the disorder (i.e., affected) by the number of people in the population or population sample at that period of time, multiplying the result by 100 (e.g., 6 affected, 1,000 people: prevalence = .6%). A prevalence rate expresses a similar concept, but it is referenced to a standard unit (e.g., 60 affected people per 10,000 people: prevalence rate = .60). Prevalence percentages are typically used to describe a given sample or population, whereas prevalence rates are useful to estimate or compare health service needs in relation to population size. Burd, Hammes, Bornhoeft, and Fisher (1988) provided an example of an epidemiological study in communicative disorders that uses prevalence rates to estimate the number of people expected to be affected in a population of interest.

Comorbidity Estimates

Estimates of the comorbidity of speech disorders and language disorders are obtained in two ways, a possible source of confusion to the casual reader. One type of comorbidity percentage or comorbidity rate reflects the same information as a prevalence estimate obtained for a single disorder. The difference is that the statistic reflects people in the sample who are positive for two or more disorders. Thus, in addition to prevalence percentages for each disorder, an epidemiological study may provide a separate percentage or rate for the joint occurrence of disorders in the sample. Comorbidity percentages estimated in this way typically are relatively small in absolute magnitude and not too discrepant from the individual prevalence of each disorder.

The second and more frequently reported type of comorbidity estimate reflects the percentage of children identified as positive for one disorder who

are also identified as positive for one or more other disorders. This is the definition of *comorbidity* given at the outset of this chapter and is likely to be of greater relevance for the issues raised in this chapter. The denominator for this second type of comorbidity estimate is the number of individuals affected with the first or index disorder, and the numerator is the number of those people positive for the second or other disorders. Continuing the previous examples, if 15 of 60 people positive for the index disorder or condition were also positive for another disorder, the comorbidity percentage would be 25%. For the reader interested in epidemiological method, Kahn and Sempos (1989), Khoury, Beaty, and Cohen (1993), and Last (1988) are useful references; for a brief explanation, see Streiner, Norman, and Munroe Blum (1989).

The generalizability and utility of both types of comorbidity estimates depend on the validity and reliability of the methods used to obtain the sample. Comorbidity estimates of the first type typically are undertaken on large samples, which requires testing protocols that are time and cost efficient. Comorbidity estimates of the second type are generally based on considerably smaller samples, typically convenience samples ascertained from available clinical sources or databases. Thus, they do not have the external validity associated with epidemiologic data. However, smaller sample sizes typically permit more detailed assessment data on either or both the index disorder and the other disorder, depending on the focus of the study. Even when based on a relatively small number of cases, well-developed comorbidity data of the second type can be quite useful to generate and test hypotheses.

A set of comorbidity estimates for speech and language disorders ideally should include both types of comorbidity estimates, with estimates of the second type being based on both language and speech as the index group. Although there are many available prevalence estimates individually for speech disorders and language disorders (see reviews in Beitchman, et al., 1986; Silva, 1987; Tomblin et al., in press; Tomblin, Records, & Zhang, 1996), there are few reported comorbidity data of either type.

Epidemiology Studies in the United States

It is important for future research planning to understand why detailed comorbidity data of the first type have not been obtained in each of the three large-scale epidemiologic projects on communicative disorders conducted in the United States.

The National Speech and Hearing Survey The National Speech and Hearing Survey (NSHS; Hull, Mielke, Timmons, & Welleford, 1971) conducted in the 1960s randomly selected and assessed 38,884 children in first through twelfth grades in 100 school districts in nine U.S. census regions. Although spontaneous and imitative language samples were obtained on audiotape, there was no standardized measure of language considered appropriate

for inclusion in the protocol at that time. St. Louis et al. (1992) provided a thorough historical and methodological description of the NSHS. Moreover, based on retranscription of a small sample of the original audiotapes, St. Louis and colleagues have completed comorbidity studies in several target disorders, including speech and language (see Table 1). The files and audiotapes have been archived, and interested researchers are invited to make arrangements to gain access to this substantial resource.

The NINCDS Collaborative Perinatal Project The massive nationwide NINCDS Collaborative Perinatal Project conducted in the 1970s (Lassman et al., 1980) assessed approximately 20,000 children at 3 years of age and again at age 8 years. Although the published volume of findings provides percentages for children classified as normal, suspect, and abnormal on many speech and language variables, the tables and figures do not provide or allow the reader to derive per-subject data on the comorbidity of speech-language involvement. D. Vetter (personal communication, February 1997) suggested that cross-tabulation output allowing for these comorbidity comparisons may still be accessible to the interested researcher.

Table 1. Estimates of the comorbidity of speech and language disorders

Study	*n*	Mean age[a]	Comorbidity estimates by index disorder: Speech index	Comorbidity estimates by index disorder: Language index
Paul (1993)	37	3[b]		37%
Whitehurst et al. (1991)	22	3		35
Connell, Elbert, and Dinnsen (1991)	23	4	43%	
Shriberg, Kwiatkowski, Best, Hengst, and Terselic-Weber (1986)	33	4	60	
Shriberg and Kwiatkowski (1994)	64	4	66	
Bishop and Edmundson (1987)	66	4[c]		74
Paul (1993)	37	4[b]		16
Tallal, Ross, and Curtiss (1989)	76	4		60
Bishop and Edmundson (1987)	67	4.5[c]		55
Shriberg et al. (1986)	38	5	50	
Bishop and Edmundson (1987)	68	5[c]		34
Tomblin (1996a)	862	5		25
Paul and Shriberg (1982)	30	6	66	
Shriberg and Kwiatkowski (1982)	43	6	77	
Schery (1985)	718	7	75[d]	75[d]
St. Louis, Ruscello, Grafton, and Hershman (1994)	20	7[e]	45	
St. Louis et al. (1994)	20	7[e]		65
Ruscello, St. Louis, and Mason (1991)	24	12.5	54[f]	
Ruscello et al. (1991)	24	12.5	21[g]	

[a] Ages rounded up at .5, unless mean age was provided within narrow age range
[b,c] Same children followed longitudinally
[d] No index disorder in this survey
[e] Estimated
[f] Classified as having delayed articulation
[g] Classified as having residual errors

Epidemiology of Specific Language Impairment Project The Epidemiology of Specific Language Impairment (EpiSLI) study conducted in the 1990s (Tomblin, 1996a; Tomblin et al., in press) used stratified cluster sampling procedures to assess more than 7,000 kindergartners living in three midwestern areas. Estimates of the prevalence of speech disorder and comorbidity of speech and language disorders were not goals of this project. However, because the diagnostic protocol in the Iowa studies did include the Word Articulation subtest of the Test of Language Development–2: Primary (TOLD–P; Newcomer & Hammill, 1988), a 20-item single-word articulation test administered by imitation, it was possible for Tomblin and colleagues to provide a preliminary estimate of the comorbidity of language and speech disorders using a subsample of children in the EpiSLI study (comorbidity of the second type; see Table 1). Additional data from this project are presented in the final section of this chapter, including a description of the criteria used to define SLI.

Comorbidity Data: Studies and Findings

Table 1 is a sample of estimates of the comorbidity of speech and language disorders arranged by increasing age and the index disorder. Not all studies used the inclusionary and exclusionary criteria typically associated with SLI. Each of the studies in Table 1 used diagnostic markers based on natural or evoked speech production to classify children's speech status, rather than phonological processing tasks or challenging speech production tasks. The studies in Table 1 undoubtedly only sample the available estimates, but they do include those located in a reasonably wide literature search. Several are discussed in the monograph by St. Louis et al. (1992). Evaluative review of the studies generating the entries in Table 1 would extend beyond the scope of this chapter. The focus here is only on the implications of trends in Table 1 for phenotype issues in speech disorders. Three observations are of interest.

First, in comparison with studies that ascertained children by language disorder, studies in which the index disorder is speech appear to report somewhat higher comorbidity estimates. The average comorbidity of the 10 estimates in which children were ascertained by speech was 55.7%, whereas the average comorbidity of the 10 estimates based on language as the index disorder was 47.6%. Second, our own previous estimate of the percentage of preschool children with speech delays who also have a language disorder was 50%–75% (Shriberg & Kwiatkowski, 1994). Based on the studies in Table 1, using just the six studies of children ages 3–6 years with speech as the index disorder, the average is approximately 60%. The third and most important observation about the entries in Table 1 underscores the variability in estimates, even for children of the same age ascertained by the same index disorder and obtained in studies

conducted by the same research group. Given the similarities in the subjects and speech-testing procedures in the five independent estimates from our own studies, ranging from 50% to 77%, it is likely that the sources for these differences are in the language domain, including differences in the language measures used in each study and in the criteria used to classify children as having language disorders.

Comorbidity Data: Some Preliminary Observations

It is challenging to speculate on what the comorbidity estimates in Table 1 might suggest about associations between speech and language disorders and their implications for a phenotype marker for speech disorders. After a presentation of findings from four comorbidity estimates in the next section of the chapter, we provide some summary observations. This section comments on three possible methodological explanations for the previous observations and makes one preliminary observation on implications for the phenotype issue.

First, on the issue of differences in comorbidity estimates based on index disorder, one explanation could invoke the concept of ascertainment bias. Shaywitz, Shaywitz, Fletcher, and Escobar (1990) provided a signal call for the possibility that the social-behavioral characteristics of children with dyslexia might bias prevalence rates and gender ratios when ascertainment was by clinical referral as opposed to population surveys. On similar grounds, comorbidity data obtained by clinical referral could also be biased by a number of social factors affecting sampling.

A second related possibility concerns possible differences in the impact of speech versus language disorders on the likelihood of clinical referral, an issue discussed by Tomblin (1996a). Tomblin noted that in comparison to the 7.4% prevalence of SLI found for kindergarten children in the EpiSLI Project, the generally lower SLI prevalence estimates from clinical referral studies could be due to the social salience of speech versus language disorders. The premise is that if a speech disorder is more noticeable than a language disorder, milder forms of language disorder might go unidentified unless co-occurring with a speech disorder. This hypothesis might be invoked to directly explain differences in prevalence estimates based on clinical referral, but it predicts the opposite of the present findings, that is, that comorbidity estimates were higher for those indexed by speech disorder. If severity of language disorder is at least moderately associated with severity of speech disorder, comorbidity estimates for children ascertained by language disorder should be higher than when ascertained by speech disorder. That is, if language disorder is less salient than speech disorder, children who are identified as having a language disorder should be more severely involved and hence more likely to have a speech disorder.

A third possible explanation for the differences in estimates associated with index disorder could relate to differences in the normalization (i.e., recovery) rates of speech versus language disorders. This explanation is conditional on the validity of two assumptions. The first assumption is the same as for the second possibility—that the more severely involved a child is in one disorder, speech or language, the more likely the child is to be clinically involved at least to some degree in the other. The second assumption, based on data on typical speech and language acquisition, is that speech disorders normalize sooner than language disorders. If these two assumptions are valid, it would follow that the comorbidity rates for children with persistent speech disorder should be higher than for children with persisting language disorder, the trends observed in Table 1. That is, comorbidity is higher in children ascertained by speech disorder because only those who have not normalized are eligible for inclusion; such children have more severe speech involvement and hence are more likely also to retain language involvement.

Finally, measurement issues and possible biases notwithstanding, the data in Table 1 appear to support more independence between speech and language disorder than is generally reported, including discussions elsewhere in this volume. Comorbidity for speech-language disorders was less than 50% in 8 of the 20 comorbidity estimates in Table 1. Thus, a substantial number of children appear to have one disorder but not the other, at least at some time during the developmental period. As suggested at the outset of this chapter, if these findings are cross-validated in well-developed epidemiological studies, comorbidity data can generate useful hypotheses about shared and nonshared substrates of developmental speech disorder and developmental language disorder.

COMORBIDITY OF DPD AND SLI IN FOUR STUDY SAMPLES

The following is a preliminary report of four studies that address some of the methodological differences among studies noted in Table 1. First, the SDCS system is used in all four studies; therefore, speech status across studies is determined by the same criteria. Second, although language measures vary across studies, we provide detailed rationale for the definition of subclinical and clinical involvement for each measure, based primarily on the work of Lahey and Edwards (1995) and Tomblin et al. (1996). Third, we calculated comorbidity at the level of both clinical and subclinical involvement for both language and speech to allow a look at possible differences associated with severity of involvement. Fourth, we address possible contributions of gender to the magnitude of comorbidity estimates. Finally, to provide more detail on language variables, findings are arranged by language modality (i.e., receptive, expressive) and language domain (i.e., vocabulary, grammar, vocabulary/grammar).

Participants and Ascertainment Procedures

Table 2 is a summary of the gender, age, and speech status of 219 children in four study samples drawn from several projects. The number of children in each study ranged from 40 to 79, percentage of boys from approximately 62% to 68%, and mean age from approximately 4 to 6 years. In Studies 1, 2, and 3, children were ascertained by referral from speech-language pathologists asked to identify children with developmental speech disorders or intelligibility problems, regardless of language status. Children in Study 4 were identified as SLI in an epidemiological study of language disorders in kindergarten children. In each of the four studies, associated measures and case history data ensured that children were free of significant involvements of the speech and hearing mechanism, cognitive-linguistic function, or psychosocial processes. In keeping with the previous discussion on the importance of method in prevalence and comorbidity research, the following sections provide detailed information on subject ascertainment and measurement procedures.

Study 1 Study 1 included 58 of 64 children obtained by referral from speech-language pathologists in the Madison, Wisconsin, Metropolitan School District (cf. Shriberg & Kwiatkowski, 1994) for a descriptive study conducted in the mid-1980s. The six children not included in Study 1 did not meet criteria for NSA/SD or SD in the revised SDCS system.

Study 2 Study 2 included two cohorts of children (25 children in Cohort 1, 20 in Cohort 2) whose speech was assessed at 6-month intervals in the early 1990s until normalization (cf. Kwiatkowski & Shriberg, 1997; Shriberg, Gruber, & Kwiatkowski, 1994). Data are reported from the initial assessment of these children, who were also recruited by referral from clinicians in the Madison, Wisconsin, area. All 25 of the children in Cohort 1 and 17 of the children in Cohort 2 met revised SDCS criteria for NSA/SD or SD.

Study 3 Study 3 included 40 children from a collaborative study of familial aggregation of speech disorders initiated in the early 1990s (cf. Lewis & Shriberg, 1994). Children with speech delay were recruited through referrals from speech-language pathologists in the Cleveland, Ohio, area. Probands and nuclear family members received a speech and language assessment appropriate for their age. Of the original 53 probands whose speech was transcribed in our laboratory, 16 had speech errors that did not meet criteria for NSA/SD or SD at the time of conversational speech sampling. Five probands were excluded from this report because they did not meet age criteria for the TOLD–P (two were younger than age 4 years, one was older than age 7 years) or did not have subscores available. The 40 children reported here include 32 of the original 53 probands and eight 4- to 7-year-old siblings of probands meeting criteria for NSA/SD or SD in the revised SDCS system.

Study 4 Study 4 included 79 children classified as SLI in an epidemiological study of language disorders in kindergarten children conducted in Iowa and western Illinois (Tomblin et al., 1996). Conversational speech samples

Table 2. Descriptive statistics for children in four study samples, including age (in months), gender, and speech characteristics (subclinical, clinical)

	Speech Status																															
	Subclinical												Clinical												Total							
	Male				Female				Both				Male				Female				Both				Male				Female				Both	
	Age				Age				Age				Age				Age				Age				Age				Age				Age	
Study	*n*	%	*M*	*SD*	*n*	%	*M*	*SD*	*n*	*M*	*SD*	*n*	%	*M*	*SD*	*n*	%	*M*	*SD*	*n*	*M*	*SD*	*n*	%	*M*	*SD*	*n*	%	*M*	*SD*	*n*	*M*	*SD*
1	9	64.3	53.0	8.7	5	35.7	54.0	4.5	14	53.4	7.3	28	63.6	50.8	9.2	16	36.4	49.0	7.8	44	50.1	8.7	37	63.8	51.3	9.0	21	36.2	50.2	7.4	58	50.9	8.4
2[a]	5	71.4	42.8	4.5	2	28.6	43.0	2.8	7	47.0	4.1	23	65.7	48.3	7.6	12	34.3	48.9	5.9	35	48.5	7.0	28	66.7	48.2	7.1	14	33.3	48.4	5.6	42	48.1	6.6
3	10	58.8	65.3	16.7	7	41.2	74.3	15.8	17	69.0	16.5	17	73.9	60.5	11.2	6	26.1	59.0	9.0	23	60.1	10.5	27	67.5	62.3	13.4	13	32.5	67.2	14.9	40	63.9	13.9
4[b]	—				—				—			—				—				—			49	62.0	71.8	3.6	30	38.0	70.8	4.3	79	71.4	3.9
All	24	64.8	53.4		14	35.2	57.6		38	56.5		68	67.7	53.2		34	32.3	52.3		102	52.9		141	65.0	58.4		78	35.0	59.2		219	58.6	

M, mean; *SD*, standard deviation.
[a] Study 2 includes two cohorts of children followed prospectively (see text).
[b] Study 4 subjects were selected on the basis of language disorder rather than speech (see text). The numbers of children with speech involvement are reported in Figure 4.

were obtained from a randomly selected subsample of children from the epidemiology project: 6 speech samples from the pilot phase of this study and 73 speech samples from two study cohorts collected in successive years of the project. Five additional speech samples were excluded because transcripts could not be classified reliably (too few usable words) by the SDCS program.

Conversational Speech Sampling and Classification of Speech Status

Conversational speech samples were obtained from each child in each study using a standard protocol followed by all research collaborators to maximize technical quality and linguistic productivity (Shriberg, 1993). High-quality monaural audiocassette recorders, matching external microphones, and high-quality audiocassette tapes were used, with microphone-to-lip distance monitored at 15 centimeters. Children were asked to discuss favorite home and school activities and recent events. For some children, pictures were used to suggest topics. Examiners verbally glossed speech that was likely to be unintelligible to transcribers, following a protocol that was minimally intrusive to the child. The average number of utterances per sample was 68 (Study 1, 78; Study 2, 147; Study 3, 77; Study 4, 46).

All speech samples were transcribed by one of three research transcriptionists trained in systems for narrow phonetic transcription (Shriberg & Kent, 1995), consensus transcription (Shriberg, Kwiatkowski, & Hoffmann, 1984), and formatting transcripts for computer analysis (Shriberg, 1986). Research assistants entered transcripts in the PEPPER program (Shriberg, 1986) running in a VAX environment. The SDCS program, which was included in the suite of PEPPER analyses, classified the sample as a variant of NSA, QRE, NSA/SD, or SD. For our purposes, additional information from the SDCS system (i.e., subclassifications not described in the previous review) was collapsed to yield two clinical classification groups: subclinical speech disorder and clinical speech disorder. The subclinical group included children classified as NSA/SD, and the clinical group included children classified as SD. Children classified as NSA or QRE were included only in Study 4.

Classification of Language Status

As previously discussed, definitions of language involvement have included varying measures of language domains and modalities and varying cutoff scores to index disorder. Commonly used cutoff criteria for clinical involvement on standardized language tests include a range of scores from 1 to 1.3 standard deviation units below the mean. For example, Lahey and Edwards (1995) used a score 1.3 standard deviation units below the mean to define clinical involvement, whereas Lewis et al. (1993) used 1 standard deviation below the mean to define clinical involvement. In Tomblin et al. (1996), z scores below -1.25 in two of five areas of language performance were found to yield the greatest sensitivity and specificity based on concurrent validity assessment using the Iowa Severity Rating Scales (Jeffrey & Freilinger, 1986).

Children in these four study samples were given tests of both receptive and expressive language modalities, with vocabulary and grammar domains evaluated in separate subtests or combined in a single test or score. The children in Study 4 also were given a measure of narrative comprehension and production. Table 3 includes a list of the measures used in the four studies and the criteria used to code language status. Each study used a different set of language measures, including standardized measures and analysis of the conversational speech sample to assign structural stage. Standardized measures differed in the way scores were calculated and reported, with most measures using normal curve equivalents (i.e., z scores, standard scores, percentile scores), others using age-equivalent scores, and structural stage expressed as a difference between the expected and emerging stage.

For each of the language measures in Table 3, a score of 1.25 standard deviation units below the mean (whether expressed as a z score, standard score, or percentile score) was adopted as the scoring criteria for clinical involvement for all measures that provided a score of that type, which was consistent with the scoring criteria for Study 4. For measures whose reference data provide only age-level scores, a 1-year or greater delay was considered clinical involvement, with a two-stage gap between the emerging and expected structural stage also considered clinical involvement. To retain maximum sensitivity to language problems, scores that were greater than 1 and less than 1.25 standard deviation units below the mean were classified as reflecting subclinical involvement for a measure. For measures providing age-level scores, greater than 6-month and less than 1-year delays were considered subclinical involvement, with a one-stage gap between emerging and expected structural stage also considered subclinical involvement. To present information consistently across studies, an ordinal coding system was developed to index normal language, subclinical involvement, or clinical involvement on each language measure. (See the Appendix to this chapter for additional subject and language classification information for each of the four studies.)

Subtypes of Language Disorders

Subtypes of language disorders (i.e., receptive only versus expressive only versus both receptive and expressive) are of interest in most studies of developmental language disorders. A variety of criteria have been used to define subtypes. For example, Lahey and Edwards (1995) defined an *expressive only disorder* by requiring all scores from receptive measures to be within 1 standard deviation of the mean, two measures of expressive language to be -1.3 standard deviation units or more below the mean, and the difference between the expressive and receptive language scores to be greater than 2 standard errors of measurement of the difference. Although the rationale for this model is

Table 3. Language measures reported for the four study samples and criteria used to code language status

Measure	Study				Modality		Domain		Coding criteria[a]			
	1	2	3	4	Receptive	Expressive	Vocabulary	Grammar	A	B	C	D
Peabody Picture Vocabulary Test–Revised (PPVT–R) (Dunn & Dunn, 1981)	X	X			X		X		X			
Preschool Language Scale (PLS) (Zimmerman, Steiner, & Pond, 1979)	X				X		X	X		X		
Structural Stage (StruSt)[b] (Miller, 1981; Miller & Chapman, 1986; Paul & Shriberg, 1982)	X	X				X		X			X	
Test for Auditory Comprehension of Language–Revised (TACL–R)[c] (Carrow-Woolfolk, 1985)												
Elaborated Sentences (TALC–ES)		X			X			X	X			
Grammatical Morphemes (TACL–GM)		X			X			X	X			
Word Classes and Relations (TACL–WC)		X			X		X		X			
Sequenced Inventory of Communication Development (SICD)[d] (Hedrick, Prather, & Tobin, 1975)		X			X		X	X			X	
Test of Language Development–2: Primary (TOLD–2) (Newcomer & Hammill, 1988)												
Subtest[e]												
Grammatic Completion (TOLD–GC)		X				X		X	X			
Grammatic Understanding (TOLD–GU)		X			X			X	X			
Picture Vocabulary (TOLD–PV)		X			X		X		X			
Sentence Imitation (TOLD–SI)		X				X		X				
Quotients												
Listening Quotient (TOLD–LiQ)			X	X	X		X	X	X			
Speaking Quotient (TOLD–SpQ)			X	X		X	X	X	X			
Spoken Language Quotient (TOLD–SLQ)			X		X	X	X	X	X			

[a] Coding criteria:

A *Normal:* *z* score −1.0 or higher, standard score 85 or higher, or 16th percentile or higher
Subclinical: *z* score between −1.25 and −1.0 (exclusive), standard score of 82–84, or 11th–15th percentile
Clinical: *z* score −1.25 and lower, standard score 81 and lower, or 10th percentile or lower

B *Normal:* Scored 6 months below age level or higher
Subclinical: Scored more than 6 months but less than 1 year below age level
Clinical: Scored 1 year or more below age level

C *Normal:* Emerging stage at or above expected stage
Subclinical: Emerging stage one stage below expected stage
Clinical: Emerging stage two or more stages below expected stage

[b] Structural stage was evaluated for all children in Cohort 1 and all 3-year-old children in Cohort 2 (N = 33).

[c] TACL–R subtests were given to subjects ages 3 and older in Cohort 1 (N = 23).

[d] SICD was given to subjects younger than age 3 years in Cohort 1 (N = 2).

[e] TOLD was given to subjects ages 4 years and older in Cohort 2 (N = 10). The TOLD Oral Vocabulary subtest was also given but is not reported here, because it was the only independent measure of expressive vocabulary in the four studies.

attractive, the variety of measures used in Studies 1–3 did not allow us to use differences between receptive and expressive scores. Instead of focusing on subtypes, we have focused on Modality × Domain performance on individual measures. In Table 3, each measure is classified by modality (i.e., receptive, expressive, both) and domain (i.e., vocabulary, grammar, both). In the following discussion of findings, subclinical or clinical involvement in a Modality × Domain category is defined by the most severe involvement (i.e., normal, subclinical, clinical) on any measure in that category. For example, some children in Study 2 received both the Peabody Picture Vocabulary Test–Revised (PPVT–R) (Dunn & Dunn, 1981) and the Word Classes and Relations subtest of the Test for Auditory Comprehension of Language–Revised (TACL–WC) (Carrow-Woolfolk, 1985), which are both measures of receptive vocabulary. A child coded normal language for the PPVT–R and subclinical language for the TACL–WC would be coded subclinical language for receptive vocabulary.

Results: Comorbidity of Language Disorder in Children with Speech Disorder

Figures 3 and 4 are summaries of the comorbidity findings for children in Studies 1–3. The children in these three studies were ascertained by clinical referral on the basis of their speech problems of unknown origin and by their classification as NSA/SD or SD on the SDCS. Figure 3 includes percentages for children with co-occuring impairments on receptive language measures, and Figure 4 includes percentages for children with co-occurring impairments on expressive language measures. Comorbidity estimates are provided by gender, language modality, and level of involvement (i.e., clinical, subclinical) in each domain. The number of children contributing to each percentage calculation is provided below each bar. The information in Figures 3 and 4, which bears on three questions, is now examined.

What Is the Comorbidity of Receptive and Expressive Language Disorder in Children with Speech Disorder? The right-most percentage in each panel containing data reflects the comorbidity percentage collapsed by gender and level of clinical involvement. There are five such estimates available in Figure 3 and three in Figure 4, totaling eight estimates of the comorbidity of speech and language disorder. For receptive involvement (Figure 3), the estimated comorbidity of speech-language disorder, in increasing order, is 6% (Study 1, vocabulary and grammar), 15% (Study 1, vocabulary), 17% (Study 2, vocabulary), 20% (Study 3, vocabulary and grammar), and 21% (Study 2, grammar). Thus, the range of comorbidity estimates is 6%–21%, with most (5/6) between 15% and 21%. For expressive involvement (Figure 4), the three summary comorbidity statistics in increasing order are 38% (Study 3, vocabulary and grammar), 43% (Study 2, grammar), and 62% (Study 1, grammar). Thus, estimates range from 38% to 62%, with two of the three estimates being less than 45%.

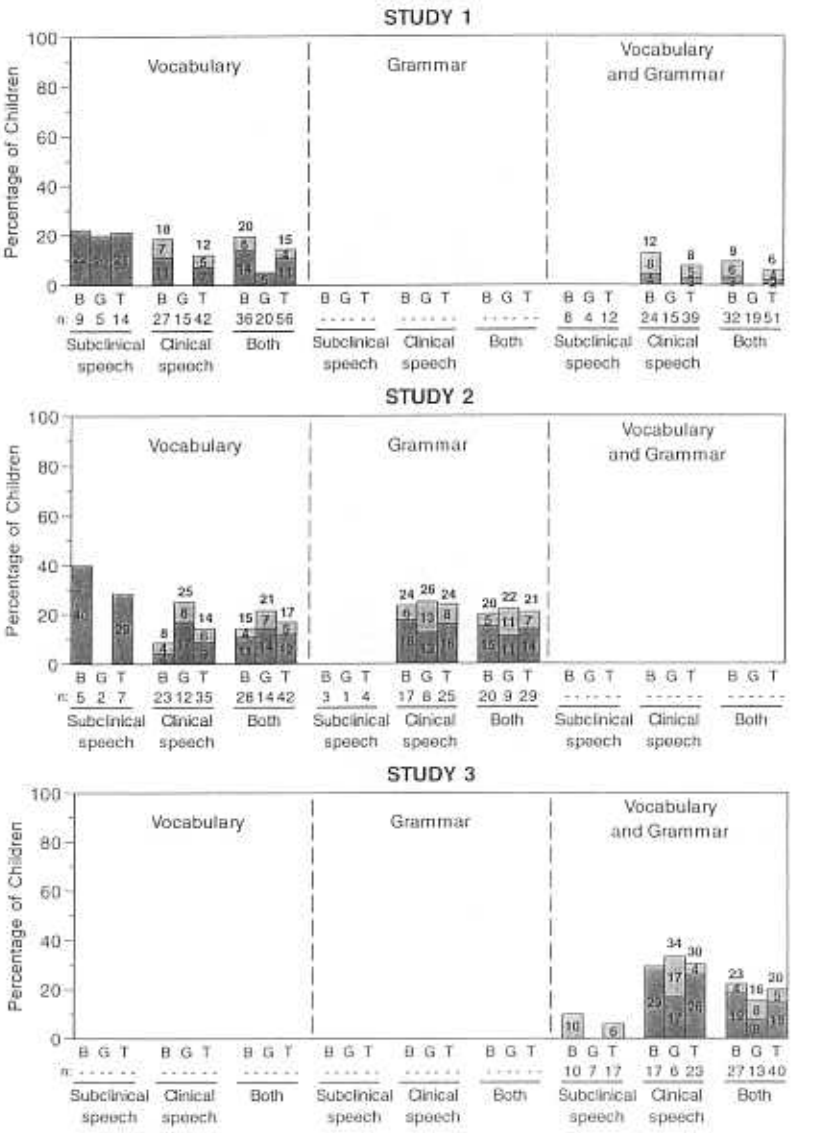

Figure 3. Estimates of the comorbidity of speech disorder and receptive language disorder in Studies 1, 2, and 3. The index disorder is speech disorder. (B, boys; G, girls; T, total; ■, subclinical language; ■, clinical language.)

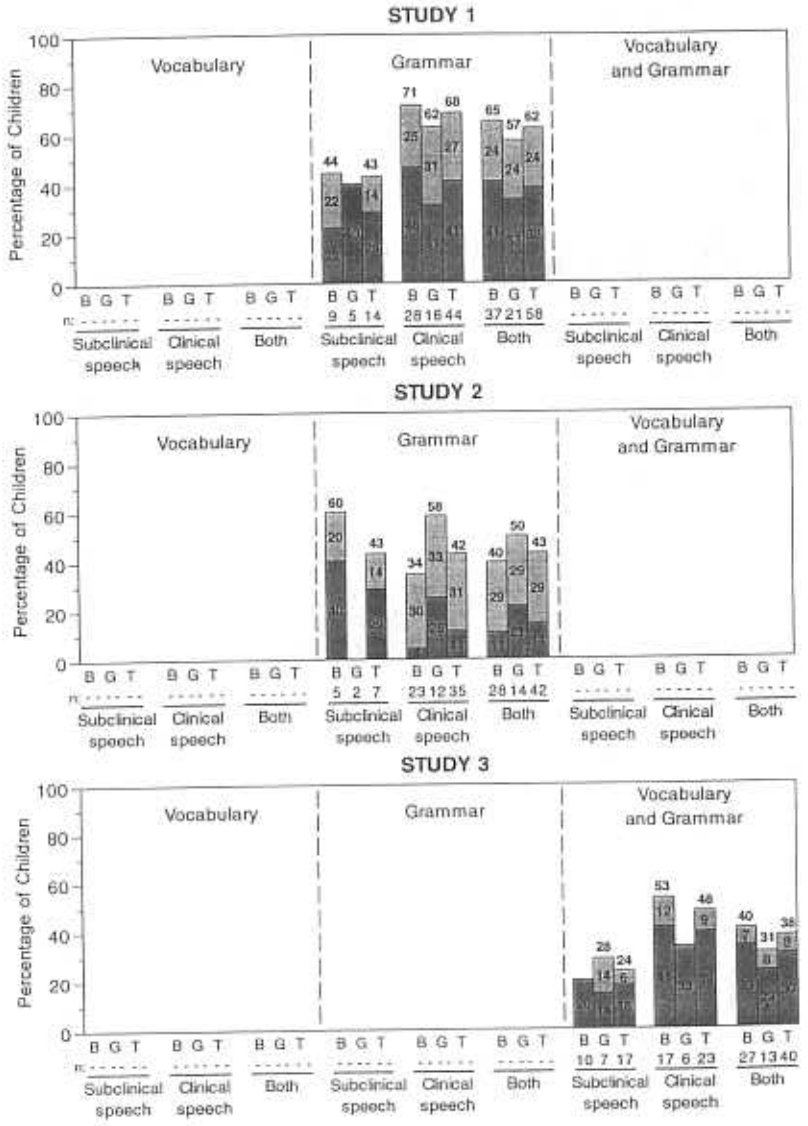

Figure 4. Estimates of the comorbidity of speech disorder and expressive language disorder in Studies 1, 2, and 3. The index disorder is speech disorder. (B, boys; G, girls; T, total; ■, subclinical language; ■, clinical language.)

These findings indicate that for children with speech involvement as ascertained from clinicians, the risk for co-occurring expressive language involvement is approximately two to three times greater than the risk for co-occurring receptive language involvement. The range of estimates within each modality underscores the variability across estimates observed previously in Table 1. The two estimates of receptive vocabulary are close in agreement (Study 1, 15%; Study 2, 17%), but the two estimates of receptive vocabulary and grammar (Study 1, 6%; Study 3, 20%) and expressive grammar (Study 1, 62%; Study 2, 43%) are not close in agreement. These differences are not likely to be due to differences in subjects, because children in Study 1 and Study 2 were ascertained similarly from the same population. Moreover, differences are not likely to be due to speech measurement issues, because speech status was determined in exactly the same way in each study. Other than possible differences associated with severity of involvement, the likely source of variance in comorbidity estimates within the same language domain is differences inherent in the variety of language measures, measure composites, and/or cutoff criteria used to classify language involvement (see Table 3).

Compared with previous estimates of 10%–40% comorbidity of speech disorder and language comprehension disorder (Shriberg & Kwiatkowski, 1994), the present findings (Figure 3) estimate comorbidity at 6%–20%. Furthermore, compared with previous estimates of 50%–75% comorbidity of speech disorder and expressive language disorder (Shriberg & Kwiatkowski, 1994), the present findings (Figure 4) estimate comorbidity at 38%–62%. Additional comment is deferred to summary discussion.

Does the Comorbidity of Speech and Receptive or Expressive Speech Language Disorder Vary by Gender? Of the eight summary estimates of speech-language disorder comorbidity in Figures 3 and 4 (i.e., gender estimates collapsed over severity of speech involvement), five have higher values for boys than girls. For the five panels assessing the comorbidity of speech and receptive language disorder in Figure 3, comorbidity percentages were higher for boys in Study 1, vocabulary (boys, 20%; girls, 5%); Study 1, vocabulary and grammar (boys, 9%; girls: 0%); and Study 3, vocabulary and grammar (boys, 23%; girls, 16%). Comorbidity percentages were higher for girls in Study 2, vocabulary (girls, 21%; boys, 15%); and Study 2, grammar (girls, 22%; boys, 20%). For the three estimates of the comorbidity of speech and expressive language disorder in Figure 4, estimates were higher for boys in Study 1, grammar (boys, 65%; girls, 57%); and Study 3, vocabulary and grammar (boys, 40%; girls; 31%); and higher for girls in Study 2, grammar (girls, 50%; boys, 40%).

These data suggest that there are no reliable gender trends for the comorbidity of speech and receptive or expressive language involvement. Gender differences range from only 2% to 15% across the eight estimates, and two of the three estimates of the same language domain disagree in directional trend.

For this question, the diversity of measures might be viewed as support for an interim conclusion of no differences in comorbidity associated with gender. To further assess gender and language, we completed Fisher exact tests on the proportions of boys and girls with normal versus clinical and/or subclinical language involvement in the three studies. None of the comparisons were statistically significant at .05 alpha levels.

Does the Comorbidity of Receptive or Expressive Language Disorder Increase with the Severity of the Speech Disorder? As noted previously, a number of theoretical perspectives would predict that the comorbidity of speech and language disorders would increase with increasing severity of the index disorder. In these data, support for this prediction can be assessed by comparing comorbidity questions for children with subclinical speech involvement with estimates for children with clinical speech involvement. Although the histograms in these figures provide separate percentages for subclinical and clinical language disorders (see Table 3 for per-measure criteria), only the totals (subclinical plus clinical language disorder) are used in these and all other comparisons. The reader may want to break out language severity findings for each of the three questions posed here, using the percentages provided in Figures 3 and 4. Differences in the ages and procedures for classifying language disorders in children in the first three studies compared with Study 4 prohibit a look at the other severity-related issue noted previously concerning its possible effects on ascertainment.

For the comorbidity of speech and receptive language disorder in Figure 3, there is modest support for a speech-language association based on severity of speech involvement, but only for language measures involving grammar. In comparison with children with subclinical speech disorders, children with clinical speech disorders had higher comorbidity estimates in the three comparisons involving receptive grammar: 1) Study 1, vocabulary and grammar (subclinical: 0%; clinical: 8%); 2) Study 2, grammar (subclinical, 0%; clinical, 24%); and 3) Study 3, vocabulary and grammar (subclinical, 6%; clinical, 30%). For the two measures involving receptive vocabulary, however, children with subclinical involvement had higher comorbidity rates in Study 1, vocabulary (subclinical, 21%; clinical, 12%), and Study 2, vocabulary (subclinical, 29%; clinical, 14%).

The data for expressive language disorders also support higher comorbidity rates for children with clinical compared with subclinical levels of speech involvement, including findings for both grammar and grammar and vocabulary (as shown in Figure 4, no data are reported for expressive vocabulary alone; see Table 3). Children with clinical speech involvements had higher comorbidity rates for grammar in Study 1 (subclinical, 43%; clinical, 68%), essentially equal rates for grammar in Study 2 (subclinical, 43%; clinical, 42%), and higher comorbidity rates for vocabulary and grammar in Study 3 (subclinical, 24%; clinical, 48%). Thus, although not attested in all comparisons, increased severity of speech disorder appears to be related to increased probability of receptive and expressive language disorder in the domain of grammar.

Results: Comorbidity of Speech Disorder in Children with Language Disorder

The data from Study 4 provide information on the comorbidity of speech disorder in children ascertained by language disorder. The data of Study 4 are constrained by the age of these children, which at the time of speech testing averaged 71.4 months, or nearly 6 years, of age (Table 2). Thus, for each of the questions asked in this section, the answers are limited to comorbidity estimates for speech-language disorder at an age when, as noted previously, an estimated 75% of children with earlier clinical or subclinical speech disorder have normalized. Although the sampling and measurement operations for both speech and language are well developed in Study 4, this age constraint limits the point prevalence estimate of comorbidity to this relatively late period in speech acquisition. Figure 5 includes findings for performance on measures of receptive and expressive language disorders, with separate comorbidity percentages shown by gender, level of language involvement, and level of speech involvement. The same three questions as those for comorbidity of language disorder are posed of these comorbidity estimates.

What Is the Comorbidity of Speech Disorder in Children with Receptive and Expressive Language Disorder? The data in the top panel, reflecting SLI as defined in Tomblin et al. (in press), indicate that 9% of 6-year-old children with SLI have clinical speech disorder, and an additional 29% have subclinical speech disorder, for an overall comorbidity estimate of 38%. This estimate is within 13 percentage points of the 25% estimate found by Tomblin using a larger sample of children from this study but using different procedures to classify children's speech status (see Table 1). When SLI is defined by involvement in a particular domain, as shown in the two other panels in Figure 5, the overall comorbidity estimates are fairly similar for children meeting criteria for receptive language disorder (28% subclinical speech disorder and 6% clinical speech disorder, totaling 34%) and expressive language disorder (31% subclinical and 9% clinical, totaling 40%). Unlike the differences in receptive and expressive comorbidity data observed in Figures 3 and 4, children ascertained by receptive and/or expressive language involvements in Study 4 are at approximately similar risk for continuing speech involvement, 34% and 40%, respectively. Tomblin (1996b) commented on the lack of utility of observing traditional distinctions between receptive and language domains for SLI classifications in children at this age.

Does the Comorbidity of Language and Speech Disorder Vary by Gender? Of the three summary gender comparisons in Figure 5, each indicates that boys with language disorder have slightly higher comorbidity of speech disorder than girls: for the EpiSLI system: boys, 43%; girls, 30%; for recep-

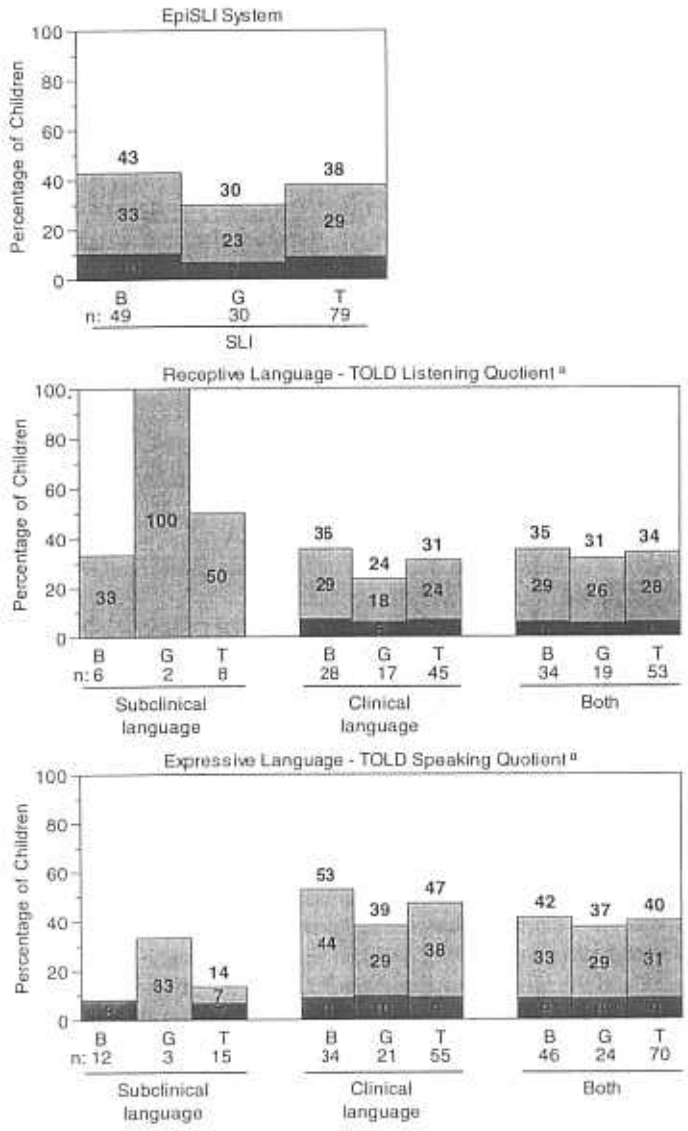

Figure 5. Estimates of the comorbidity of language disorder and speech disorder in Study 4. The index disorder is language disorder. (aOnly children with subclinical or clinical performance on this measure are reported in this panel.) (B, boys; G, girls; T, total; ■, subclinical speech; ■, clinical speech.)

tive language involvement: boys, 35%; girls, 31%; and for expressive language involvement: boys, 42%; girls, 37%. As with the gender findings for the co-occurrence of language disorders in younger speech-involved children, the small absolute differences in percentages (4%–13%) provide only the most modest support for a theoretically significant gender difference in persisting speech disorder in children with language disorder. Most of the differences were associated with subclinical speech involvement, with gender differences of only 1%–3% separating comorbidity rates for children with clinical speech involvement.

Does the Comorbidity of Speech Disorder Increase with the Severity of the Receptive or Expressive Language Disorder? There are few data in Figure 5 to assess severity issues because the EpiSLI definition of *language disorder* does not include a subclinical classification and does not classify involvement by modality. Only 8 children and 15 children, respectively, were classified as subclinical language on the composite receptive and expressive measures. The receptive data for the eight children with subclinical language involvement appear unruly (each of the two girls with subclinical language had subclinical speech disorder, yielding 100% comorbidity) but, taken together with data for boys, suggest no differences in comorbidity associated with severity of language involvement. For children with expressive language involvement, the data seem to point more clearly to higher comorbidity rates associated with clinical language involvement, with differences primarily involving the percentages for subclinical speech involvement. In sum, the few data available to assess severity as a moderating variable in comorbidity rates are inconclusive for receptive language involvement and supportive of a positive association for expressive language disorder, with the latter effects primarily associated with increased risk for the persistence of subclinical speech disorder.

CONCLUSIONS

This chapter has reviewed some information on the comorbidity of speech-language disorders from the perspective of emerging research in the genetics of speech and language disorders. Comorbidity findings from approximately two dozen studies indicate that developmental phonological and language disorders do not always co-occur, even when assessed in very young children. Some variables that appear to be associated with the magnitude of comorbidity estimates include the index disorder (speech disorder > language disorder), the language modality (expressive language disorder > receptive language disorder), the language domain (grammatical [i.e., syntactic, morphologic] disorder > vocabulary [i.e., semantic] disorder), and perhaps gender (boys > girls). However, although each of these variables may contribute to the magnitude of the comorbidity estimate, none of the observed combinations of in-

fluential variables were associated with estimates greater than approximately 70% in the four study samples examined. The largest comorbidity estimates in these four studies typically averaged considerably lower.

Are these findings readily explainable by measurement error, which, if removed from these estimates, would yield 100% comorbidity, especially at the earliest ages? As we concluded in response to the estimates in Table 1, the substantial gap between 100% comorbidity and the percentages obtained in these four studies suggests that measurement error is not a sufficient explanation for the lower-than-expected magnitude of the estimates. Certainly, different ways to measure SLI and different statistical criteria to define SLI have a large influence on comorbidity estimates, with the more stringent criteria for SLI associated with lower comorbidity estimates. However, comorbidity estimates were not higher in the four study samples when subclinical forms of both speech and language disorders were included in the estimates, nor were they higher when the classification of speech disorder ensured that children had what are considered phonological rather than only articulatory involvements (i.e., SD and NSA/SD rather than QRE or RE), with the latter presumably not being associated with language disorder.

Rather than attributing lower comorbidity estimates to measurement issues, our strongest suspicion is that some phonological disorders occurring without concomitant language disorder represent one of the other forms of SD described in review of the SDCS. In our preliminary estimates, only approximately 60% of children with SD may have the suspected inherited form, with the other approximately 40% being associated with one of the other three proposed etiologic origins of childhood speech disorders (i.e., SD–OME, SD–DAS, SD–DPI). As summarized previously, the comorbidity estimates in Table 1 and Figures 3–5 reflecting speech and expressive language disorder in children of preschool age are consistent with this 50%–60% prevalence estimate. We are especially interested in the estimated approximately 30% of children referred for speech delay whose disorder is suspected to be due to the fluctuant conductive hearing loss associated with early recurrent otitis media with effusion (SD–OME). Although evidence for significant language disorder as a sequela of OME remains equivocal, there is good reason to suspect earlier and more salient effects on speech (cf. Shriberg et al., 1998).

These comorbidity data illustrate the complex issues that must be addressed in the development of a phenotype for developmental phonological disorders. It may be that an inherited phonological disorder arises as a variant of an inherited language or verbal trait disorder. If so, it would not require an autonomous phenotype as proposed at the outset of this chapter, and children reflecting the diverse forms of the inherited verbal or language deficit could be treated as one undifferentiated group in molecular genetics studies to identify the genotype. However, if the relatively low reported comorbidity estimates are not explained by measurement error or by appeal to other etiologic subtypes, we are left with a sizable proportion of children—converging on 50% in the estimates in Table 1 and even higher percentages in the estimates in Figures 3–5—with only a speech disorder or only a language disorder.

A key element needed to resolve issues associated with the comorbidity findings reviewed is a set of diagnostic markers for each of the four suspected subtypes of SD. Once children with other subtypes of SD can be reliably excluded from study samples of children suspected to have the inherited form, researchers can assess whether there are differences in the speech profiles or normalization histories of children with only speech disorder compared with children with speech-language disorder. Also, performance on all proposed phenotype markers (e.g., phenotypes based on natural speech production, challenging speech production tasks, phonological awareness tasks, nonsense word repetition tasks) can be compared to determine if there are some children whose only linguistic problem is a mild to significant delay in the correct production of speech sounds. We think the existence of such children will eventually be documented, with consequent implications for effective and efficient treatment.

The data presented also highlight the questions raised in other chapters in this volume regarding relations between speech and language in development and disorders. If specific speech disorders turn out to be a distinct phenotype, separable by both behavioral and genetic markers from more pervasive disorders involving both speech and language, this finding would have important implications for understanding the mechanisms of the development of both speech-sound production and language formulation. If distinct genetic mechanisms for specific speech disorders can be identified, this would imply that discrete biochemical pathways subserve the speech acquisition system, more or less independent of the processes that enable language development. This would suggest that some aspects of speech development do not rely, or do not rely entirely, on a substrate of language, contrary to the view that a phonological orientation to speech-sound development would imply.

Finally, our views on treatment of childhood speech disorders might also be affected by the implications of these comorbidity findings. There has been a trend to see developmental speech-sound disorders as one manifestation of language disorder (cf. Bishop & Edmundson, 1987). That is, the phonological perspective, with its emphasis on underlying representations and simplification processes, has viewed child speech disorders as arising from faulty rule learning and treatable by approaches used to remediate language problems. If, however, certain child speech disorders arise from pathways distinct from those of language disorders, such an approach for these types would be called into question. Additional research to elaborate the phenotype of specific speech disorders and link it to identifiable biological markers is needed to illuminate the complex associations between child speech and child language disorders.

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Appendix: Assessment Information for the Four Comorbidity Studies

STUDY 1

A total of 56 of the 58 children in Study 1 were given the Peabody Picture Vocabulary Test–Revised (PPVT–R; Dunn & Dunn, 1981) as a measure of receptive vocabulary and the Auditory Comprehension subtests of the Preschool Language Scale (Zimmerman, Steiner, & Pond, 1979) to measure receptive language skills (both vocabulary and grammar). Structural stage (StruSt), a measure of expressive grammar, was determined by analyzing conversational speech samples for syntactic performance using procedures and reference data described in Miller (1981), Miller and Chapman (1986), and Paul and Shriberg (1982). Results for other receptive and expressive language measures administered to the original group of 64 children are reported in Shriberg and Kwiatkowski (1994) and are not considered in this chapter.

STUDY 2

All 42 children in Study 2 were given the PPVT–R to assess receptive vocabulary. A total of 23 children in Cohort 1 received the Test for Auditory Comprehension of Language (TACL–R; Carrow-Woolfolk, 1985). The TACL–R includes three subtests: Word Classes and Relations (WC), a measure of receptive vocabulary; and Grammatical Morphemes (GM) and Elaborated Sentences (ES), two measures of receptive grammar. Percentile scores for all subtests and a total score were recorded. A total of 12 children achieved baseline performance on all subtests, 22 (15 M, 7 F) achieved baseline for WC, 15 (11 M, 4 F) for GM, and 16 (11 M, 5 F) for ES. Two children who were not yet 3 years of age at initial assessment received the receptive portion of the Sequenced Inventory of Communication Development (Hedrick, Prather, & Tobin, 1975) as an overall estimate of receptive language skills. Ten children in Cohort 2 who were at least 4 years old at initial assessment received five subtests of the Test of Language Development–2: Primary (TOLD–P; Newcomer & Hammill, 1988). Subtests administered were Picture Vocabulary (PV), a measure of receptive vocabulary; Grammatic Understanding (GU), a measure of receptive grammar; Oral Vocabulary (OV), a measure of expressive vocabulary; and Grammatic Completion (GC) and Sentence Imitation (SI), two measures of expressive grammar. Two subtests of phonology (Word Discrimination and Word Articulation) were not administered. Finally, as a measure of

expressive grammar, structural stage was determined using the same procedures as used in Study 1 for all of the children in Cohort 1 and the seven 3-year-old children in Cohort 2.

STUDY 3

All children in Study 3 were given six subtests of the TOLD–P, including the five subtests listed for Study 2, and the Word Discrimination (WD) subtest, a measure of phonological awareness. We were provided with standard scores calculated from national norms in the TOLD–P manual for the Spoken Language Quotient (SLQ), which included all six subtests, and the Listening Quotient (LiQ), which included the PV, GU, and WD subtests for all 40 of the subjects reported here. We were also provided with the Speaking Quotient (SpQ), which included the OV, GC, and SI subtests, for 14 subjects. The LiQ was used as a measure of receptive language skills, and the SpQ was used as a measure of expressive skills, with both measures including subtests in the vocabulary and grammar domains. SpQ scores were not available for 26 of the children; therefore, the SLQ was used as an estimate of the child's expressive skills.

STUDY 4

All children in Study 4 were given the five subtests of the TOLD–P described for Study 2, and we were provided with LiQ and SpQ z scores as well as the child's status on the EpiSLI system (Tomblin et al., 1996). The z scores were calculated using local norms established by the Iowa project as described in Tomblin et al.

We also received each child's classification (normal language or SLI) on the EpiSLI system developed as part of the Iowa project. The EpiSLI system includes five subtests from the TOLD–P and results of a narrative comprehension and production measure based on the work of Culatta, Page, and Ellis (1983). The subtests were used to calculate composite scores in three domains (vocabulary, grammar, and narrative) and two modalities (comprehension and expression). Each domain composite included at least one receptive and one expressive measure, and the receptive and expressive composites included measures from each of the three domains: vocabulary, grammar, and narrative. As discussed previously, z scores of less than or equal to -1.25 based on local norms on two of these five composites yielded the greatest sensitivity and specificity when compared with a clinical rating system (Tomblin et al., 1996). As a result, children were classified as SLI in the EpiSLI system when they received z scores less than or equal to -1.25 for two of the five composite scores.