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Subtyping Stuttering II:

Contributions from Language and Temperament

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Abstract

This paper is the second in a series of two articles exploring subtypes of stuttering, and it addresses the question of whether and how language ability and temperament variables may be relevant to the study of subtypes within the larger population of children who stutter. Despite observations of varied profiles among young children who stutter, efforts to identify and characterize subtypes of stuttering have had limited influence on theoretical or clinical understanding of the disorder. This manuscript briefly highlights research on language and temperament in young children who stutter, and considers whether the results can provide guidance for efforts to more effectively investigate and elucidate subtypes in childhood stuttering. Issues from the literature that appear relevant to research on stuttering subtypes include: (a) the question of whether stuttering is best characterized as categorical or continuous; (b) interpretation of individual differences in skills and profiles; and (c) the fact that, during the preschool years, the interaction among domains such as language and temperament are changing very rapidly, resulting in large differences in developmental profiles within relatively brief chronological age periods.

Keywords

Language; Stuttering; Subtyping; Temperament

One of the central aims of the Stuttering Research Program at the University of Illinois is to delineate subtypes among young children who stutter. We have approached this aim through the integration of data from multiple domains that are relevant in speech and language

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development, namely linguistic, motoric, genetic, and psychosocial areas. The broad objective is to examine developmental trends and pathways, as various patterns of risk factors and/or clusters of behaviors associated with persistent or recovered stuttering may emerge. Underlying this research are several hypotheses, 1) stuttering arises from a complex, dynamic, interaction of multiple factors (Smith, 1990; Smith & Kelly, 1997), 2) the predominant preschool-age onset of stuttering suggests that the relevant multiple factors are rooted in developmental processes (Watkins, Yairi, & Ambrose, 1999; Yairi & Ambrose, 2005), and 3) the alignment of multiple factors lasting for a period (or periods) during their varied pace of development may contribute to the emergence of stuttering (Watkins, Yairi, & Ambrose, 1999; Yairi & Ambrose, 2005).

Several researchers have proposed that the heterogeneous characteristics and symptoms of those who stutter highlight the need for a delineation of subtypes (Blood, 1985; Riley & Riley, 2000; Watson & Freeman, 1997; Yairi, in press). Even among young children near the time of stuttering onset, heterogeneity is evident with respect to predominant disfluencies (e.g., prolongations vs. repetitions), onset characteristics (e.g., sudden vs. gradual), language skills (e.g., precocious vs. delayed), and relative profiles of strength/weakness (e.g., weak phonology/ weak language skills vs. weak phonology/strong language skills) (Yairi & Ambrose, 2005; Yaruss, LaSalle, & Conture, 1998). This heterogeneity may reflect the random variation of individual characteristics among the vast population of children as a whole with no relevance to stuttering. Or, when examined more carefully, the data may reveal several predominant patterns of characteristics (i.e., denoting subtypes) evidenced among children who stutter.

The crucial question is how to begin the careful examination of these variables in order to determine whether subtypes exist, and if they exist, their ultimate nature. As reviewed in the first article in this series (Yairi, in press), there are multiple variables of potential interest to be examined, and they span essentially all the diagnostic skill domains relevant to communicative disorders (i.e., phonology, articulation, voice, semantics, syntax, pragmatics, etc.), as well as many feature domains associated with the inherent condition of the communicator (i.e., biological, physiological, psychological, etc.). Broadly, the domains of interest include both skill (or ability) domains and feature (or attribute) domains. A comprehensive analysis of the contributions of research from all these areas would be a massive undertaking beyond the scope of this paper. An increased understanding with potential to enhance our subtyping endeavors is possible however, from the examination of relevant research from one domain from each area. In this case, the particular ability domain will be language, and from the attribute domain, temperament. We will begin by highlighting selected studies of each of these domains in young children who stutter, and proceed with a discussion of previous efforts to investigate subtypes in these domains. Finally, we will integrate these insights to recommend directions for future research.

The domains of language and temperament will receive intensive focus because: 1) significant changes take place in these domains during the time frame when stuttering onset is most likely, 2) both offer a long research tradition with contradictory and complex findings, but also potential implications for subtyping research, and finally, 3) underlying commonalities are evident for these domains particularly in potentially fruitful methodological strategies that, to date, have not been applied to developmental stuttering.

In addition to the firmly established stuttering-language links related to the loci of stuttering both in adults and children (Bloodstein & Grossman, 1981; Brown, 1945), several scholars have investigated potential links between developmental stuttering and *language* (see Nippold, 1990; Ratner, 1997; for reviews, see Bloodstein, 2002, and Bosshardt, 2002; and for example, see Logan & Conture, 1995, 1997, and Watkins, Yairi & Ambrose, 1999) and between developmental stuttering and *temperament* (Anderson, Pellowski, Conture, & Kelly, 2003;

Karrass et al., 2006; Lewis & Golberg, 1997). In general, this research has identified associations between variables in these domains and stuttering behaviors, rather than suggesting causal relations. The stuttering-related literature suggests the possibility of several potential relationships: 1) some characteristics may make a child vulnerable or predisposed to the development of stuttering, 2) regardless of its role in initial onset, the characteristic may place a child at risk for persistence of stuttering once it has begun, and finally, 3) the developmental pattern of the characteristic may be influenced itself, from the other direction, by the stuttering. The subtyping literature from these domains also reveals several common themes: (a) concern with whether a particular variable is most accurately characterized as dimensional (i.e., described by a continuum) or categorical (i.e., consisting of discrete patterns that differ in predictable, coherent and systematic ways from others); (b) concern with how to interpret the relevance of individual differences; and (c) concern with how to adequately probe rapidly changing profiles during the course of their development.

This paper is designed to build on the conceptual framework and literature review of subtyping research that is provided by the initial paper in this series (Yairi, in press). Although these papers neither present nor evaluate a particular theoretical model, they share a general conceptual framework, summarized by the following principles: (a) stuttering is a complex, multifaceted condition that may be better understood through subtyping efforts, (b) certain developmental domains should be considered in subtyping efforts, particularly language ability and temperament attribute variables, and (c) developmental pathways and abilities in domains related to speech production may provide critical clues to more complete understanding of stuttering, particularly in young children. The collective goal of this paper and its companion article is to explore whether and how investigations of subtypes among people who stutter may inform our understanding of the disability, and treatment of the condition. The approach is a constructivist one, insofar as instead of hypothesizing a specific model, data and insights from existing research are used to identify relevant variables, construct conclusions based on extant findings, and outline critical next steps for research in this area.

The broad aim of subtyping is the valid and reliable characterization of individuals (according to specified variables) such that those within their subtype have more in common with each other than with those outside that subtype. There is significant clinical and theoretical motivation to elucidate subtypes of stuttering, particularly in terms of developing trajectories of the disability in young children. From a clinical perspective, recent research indicates that three of every four youngsters who begin to stutter as preschoolers will recover within a relatively brief time period without formal intervention (Yairi & Ambrose, 2005). Clear recognition of associated risk factors and/or subtypes could optimize delivery of clinical resources to the children most likely to persist as stutterers, and would be likely to alleviate parental stress and anxiety in many cases. Furthermore, better knowledge can also inform development of effective therapeutic approaches. In terms of research and theoretical implications, the presence of discrete subtypes should simplify the task of illuminating etiology, and promote accurate phenotype and genotype characterizations. In sum, more clear understanding of stuttering subtypes, if indeed such subtypes exist, should enable considerable sharpening of diagnostic processes and enhance the focus of subsequent research endeavors.

Review of Selected Literature: Stuttering and Child Language

The connection between language and stuttering in young children is intuitive. Yairi (1983), Ratner (1997), as well as other scholars, observed that stuttering onset coincides with a time of rapid expansion in expressive and receptive language ability (i.e., stuttering most typically begins in 24- to 48-month-old children). During this period children are acquiring new words at a rate of 9–10 per day, expanding and refining their grammatical structures, and learning social uses of language forms. It is in the process of verbal communication, namely in producing

language, that the repetitions and prolongations that characterize stuttering are observed. A variety of theoretical accounts have been proposed that link language production and stuttering (e.g., the covert-repair hypothesis, the neuropsycholinguistic account, and a range of other viewpoints that emphasize language processing factors in stuttering, cf. Logan & Conture, 1995, 1997; Packman, Onslow, Richard, & Van Doorn, 1996; Perkins, Kent, & Curlee, 1991; Postma & Kolk, 1993; Weber-Fox, 2001; Wingate, 1988).

Focusing specifically on language abilities in young children who stutter, findings are varied and somewhat controversial (see Yairi, Watkins, Ambrose & Paden, 2001; Watkins & Johnson, 2004; Wingate, 2001). For example, research conducted by the Illinois Stuttering Research Project has not revealed language learning difficulties in young children who stutter; in contrast, several investigations revealed expressive language abilities at and above expectations for age in preschoolers near the onset of stuttering (Watkins, Yairi & Ambrose, 1999). Recent research in other labs supports this conclusion (Anderson & Conture, 2000; Bonelli, Dixon, Ratner, & Onslow, 2000; Miles & Ratner, 2001; Ratner & Silverman, 2000; Rommel, Hage, Kalehne, & Johannsen, 1999). Among several studies however (Anderson & Conture, 2000; Bonelli, Dixon, Ratner, & Onslow, 2000; Ratner & Silverman, 2000), it was noted that although most participants displayed language abilities in the typical range or above, the children who stuttered demonstrated lower language proficiency than matched peer controls. Several years earlier, Nippold (1990) reviewed numerous studies of language ability in children who stutter, and similarly noted: 1) the remarkable variability, especially in semantic skills, among children who stutter, 2) no tendency for language deficits among children who stutter; as a group, their language skills are usually not significantly different from children who do not stutter; however, 3) some children who stutter have particular language difficulties contributing to their overall communication problems.

Two recent studies with relatively large sample populations offer estimates of the percentage of children who stutter with concomitant language disorders. Arndt and Healey (2001) explored the extent to which school-age children who stutter were reported to have concomitant difficulties in other areas of communication. They gathered survey responses from speech-language pathologists working with 241 school-age children who stuttered. Of these, slightly more than half (56%) were reported to have disorders confined to fluency, whereas 44% had developmental difficulties in the areas of language and/or phonology. Similarly, Blood, Ridenour, Qualls, and Hammer (2003) conducted a mail survey that invited responses from 2,000 speech-language pathologists (SLPs). The investigators asked SLPs to report on their caseloads of children who stuttered, specifically to indicate the number with concomitant speech, language, speech-language, and non-speech-language disabilities. The responding clinicians reported that 9–13% of the children on their caseloads who stuttered had difficulties with aspects of receptive or expressive language, and some 33.5% had articulation problems. In both the Arndt and Healy and Blood et al. investigations, it is important to recognize the primary focus on school-age youngsters; that is, we must consider the potential influence of a history of stuttering on language performance. Nevertheless, the number of children with reported concomitant difficulties is noteworthy. One must also keep in mind that school-age children tend to represent, at most, only 25% of the original stuttering population among young children. The concomitant language abilities for 75% of young children who stutter, those who experience natural recovery, would not be represented in these studies.

Yaruss, LaSalle, and Conture (1998) examined language abilities among the many domains assessed in their large diagnostic study of 100 children who stutter. Results on the *Peabody Picture Vocabulary Test (PPVT)* (N=86) revealed approximately 15% with below-normal receptive vocabulary. Their (in)formal [sic] assessment of expressive language (N=83) found 25% with above-normal, 46% with normal, and 29% with below-normal measures in this domain. If valid, these percentages suggest notable differences for children who stutter

compared to the typical sample population. Sample populations typically reflect a normal curve where approximately 32% (16% below + 16% above) perform outside of normal range on either end of the scale. In this case however, 54% (29% below + 25% above) of children who stutter were found with atypically low or high language abilities. Results such as these suggest the possibility that being outside the normal range of the performance scale at either end could pose some risk factor for stuttering. These findings leave an impression that the heterogeneity among children who stutter may even exceed what is typical of the general population of children who do not. If so, it strengthens the need to identify the subgroups among children who stutter for both research and clinical purposes, rather than handling them as though they represent one similar group. In their brain imaging research with adults, Frances Freeman and Ben Watson (1994, 1997) have recognized the need for dividing the sample population of individuals who stutter into subgroups with linguistic impairment and with average language ability. Their investigations of regional cerebral blood flow revealing differences between these two groups, suggests a need to test the hypothesis that associated risk factors may differ between such subgroups.

Thus, the literature regarding language abilities in young children who stutter is mixed. While some research suggests that language deficiency may be a risk factor for a subgroup of young children who stutter, other research, such as our own, has supported the possibility that precocious language abilities may be a risk factor for stuttering for another subgroup of young children who stutter. Following 23 young preschool children longitudinally for at least 4 years, we found that expressive language skills remained at or above normative expectations for the 8 children whose stuttering persisted, while the 15 children who recovered exhibited a deceleration of language development toward the range that would be expected for their age (see Yairi & Ambrose, 2005, pp.244–247). It is intriguing to consider whether a trade-off of linguistic resources (advanced language at the expense of speech motor skill) may be contributing to the risk of persistent stuttering for a subgroup of these young children. In support of this possibility, Anderson, Pellowski and Conture (2005) found that a subgroup of children who stutter appear to have greater disassociations across various speech and language domains.

Competence in the area of language development among a number of preschool children who stutter has been one of the most interesting and unexpected findings of the Illinois Stuttering Research Project. Expressive language ability at or above expectations for age has been documented in several investigations, particularly near stuttering onset (Watkins, Yairi & Ambrose, 1999; see also Anderson & Conture, 2000; Miles & Ratner, 2001; Silverman & Ratner, 2002). This finding is intriguing, particularly in contrast with the speech production difficulty of stuttering. Current investigations are exploring this finding in greater detail, teasing out performance in semantic and syntactic domains, which have consistently been at least at average in the Illinois sample, and with phonological and fluency abilities, over developmental time (cf. Watkins, Seery, Throneburg, & Yairi, 2004). It is hoped that this type of analysis will foster greater understanding of individual developmental pathways, and larger profiles that characterize subgroups of participants.

Thus, insights from the discipline of child language may shed light on both the literature in terms of pathways of early childhood stuttering, and useful directions for future explorations of stuttering subtypes. Associations and co-influences between the domains of speech and language make it logical to hypothesize that disorders affecting the process of their development are not as apt to occur in isolation as they are across multiple domains and/or subdomains (Watkins & Yairi, 1997). Given the strong potential for an association between developmental language processes and stuttering, the consideration of language ability and disability in research on subtypes and risk factors is important. As Yairi (in press) has noted, attempts in this direction have already been made by Riley and Riley (1972, 1979, 1980). The most fruitful approach for this purpose has not been determined yet. Because several subtyping

systems have been applied to disabilities within the language domain, a review of the relevant research should serve as a useful guide in the pursuit of stuttering-related subtypes.

Subtypes in Language Disability

In the broad field of language disability, there are several widely-used descriptive subtyping systems. In the adult aphasia literature, the distinction between expressive language problems and global difficulties with both expressive and receptive language is often linked to site of lesion and well-established in clinical application. In early childhood language, research and clinical literature in childhood language disability distinguishes between children with receptive language limitations, children with expressive language problems, and children whose difficulties include both receptive and expressive language disabilities (Dale, Price, Bishop, & Plomin, 2003; Leonard, 1998; Paul, 2000; Rescorla, 2002). Although findings in this area are not entirely straightforward, there is some coherence in terms of severity and likely trajectory of persistence versus recovery (Dale et al., 2003; Paul, 2000).

The issue of discrete categories versus a continuous, dimensional disability has long been a focus of research in the domain of language disorders. For each of these general examples of subtypes in the area of language, there have been numerous investigations of the validity of these characterizations as discrete or categorical subtypes of disability, with varied patterns of development, etiologies, and/or outcomes. The precise nature of these investigations has varied. For example, in terms of early language disability, researchers have documented that prognosis varies in relation to the subdomains of language that are involved. Two- and three-year-old children with disabilities confined to expressive language have better linguistic and academic outcomes during the early school years than do children who had both expressive and receptive difficulties as toddlers (Dale et al., 2003; Paul, 2000). For children who appear to recover from an initial profile as a late-talker, however, research suggests that difficulties may reappear later as reading skills advance and expectations for reading proficiency increase (see Rescorla, 2002). Thus, one approach to evaluating the validity of a descriptive or empirical subgrouping or subtyping system that is particularly relevant for young children is to track whether groups diverge in outcome and pathway and/or whether groups differ in neurological and/or genetic correlates.

This research may relate to early childhood stuttering. Specifically, the work of Yairi and his colleagues has illuminated the basic pathways of persistent versus recovered stuttering (Yairi & Ambrose, 1999, 2005). Ambrose, Cox, and Yairi (1997) have revealed that family history of stuttering is perhaps the most relevant predictor of whether a child is likely to follow a persistent or recovered trajectory in the development of their stuttering. A team of the Illinois Stuttering Research Program recently reported findings regarding specific chromosomal regions likely to be the locations of genes underlying stuttering, also showing differences for persistent stuttering (Suresh et al., 2006). Yet, Ambrose et al. (1997) also noted that family history alone does not fully account for their developmental pathways. Therefore, the disability is more complex and multifaceted than suggested by genetic factors alone.

In the area of child language, one proposed subtype has received extensive research attention. Specific language impairment (SLI) is arguably the most well recognized subtype within the larger population of children with language learning difficulties. SLI is the label applied to children who have difficulties learning language in the absence of cognitive, significant social-emotional, or frank neurological limitations (Dollaghan, 2004; Johnston, 1991; Leonard, 1991, 1998; Tomblin, 1991; Tomblin, Records, Buckwalter, & Zhang, 1997). An interesting feature of SLI is that it is identified by the contrast of strength in certain domains (e.g., cognitive and social abilities) and the difficulties in other domains (e.g., aspects of expressive and/or receptive language ability). Thus, SLI is typically identified through exclusionary criteria, that

is, the presence of lower than expected language skills in the absence of cognitive, social, sensory or neurological etiologies. Tomblin et al. (1997) found that about 7% of kindergarten children displayed SLI. Scholars have addressed a wide range of issues in relationship to SLI, including the specific cognitive strengths and limitations of children with SLI (Johnston, 1991), the morphosyntactic profiles associated with the disability (Rice & Wexler, 1996), social and academic challenges faced by children with SLI (Fujiki, Brinton, & Clarke, 2002), and intervention approaches that can assist children with the disorder (Leonard, Camarata, Brown & Camarata, 2004; McCauley & Fey, 2006). In recent years, the number of disciplines addressing SLI has expanded from speech-language pathology and education to include developmental and cognitive psychology and linguistics. Research on SLI has been a funding priority of the National Institutes of Health, particularly the NIDCD, and has dominated much of the research literature in journals that publish scholarship on language disabilities in children. In fact, the majority of papers published in the journals of the American Speech-Language-Hearing Association dealing with language in children have reported on the SLI population.

Thus, the study of language disability in children has been heavily influenced by the concept of SLI. As is true in the case of stuttering, however, little attention has been paid to whether SLI is a discrete category of language ability or simply the low end of the typical continuum. A monograph published in *Language, Speech and Hearing Services in Schools* raised this question nearly fifteen years ago (see Johnston, 1991; Leonard, 1991; Tomblin, 1991). At least one of the monograph's authors suggested that SLI as a discrete category had not been empirically established. Leonard (1991) suggested that evidence supported SLI as a continuous ability (i.e., the low end of the typical language development continuum). Yet, much of research literature in the field of SLI has continued as if it is a categorical or discrete disability (see Rice & Wexler, 1996). The issue has been contentious and, to date, has not been resolved.

It may be argued that this approach is simplistic relative to the complexity presented by stuttering. Upon closer inspection, however, this is not the case. What is the rationale supporting comparison of specific language impairment (SLI) and stuttering in young children? First, both disabilities can, and do, occur in the absence of frank neurological problems or significant social-emotional challenges. Second, both disabilities are defined by behavioral characteristics relative to same-age peers: stuttering by limitations in the ability to produce fluent speech and SLI by limitations in language ability. Third, both disabilities typically appear in early childhood. The majority of individuals who will ever stutter begin to do during the preschool years, generally between the ages of 2 and 4 years. This is roughly the same time frame that specific language impairment is diagnosed. Fourth, both disabilities are thought to be linked to genetic influences (Rice & Warren, 2004, 2005; Rice, Warren, & Betz, 2005; Yairi, Ambrose, & Cox, 1996). Finally, the fundamental issue of whether the disabilities are best described as continuous, that is, low-end of typical, or categorical, fundamentally different cognitive or behavioral patterns, remains an open issue.

Although SLI is not perfectly analogous to stuttering, the SLI literature does provide stuttering researchers with a number of potentially important insights regarding possible approaches to the study of subtyping. In an innovative series of two experiments, Dollaghan (2004) explored the issue of continuous vs. categorical behavior in SLI. Dollaghan used taxometrics, an analytical tool that has rarely been applied to speech-language research. Taxometics is well-suited to addressing the issue of whether a condition is best characterized as categorical or continuous and has generally been applied in analyses of psychological conditions. In brief, taxometrics statistically evaluates the extent to which the distribution of a characteristic involves a "taxon", defined as a category (Meehl, 1992; Meehl & Yonce, 1996). One type of taxometric analysis is termed MAMBAC (representing "mean above minus below a cut", see Meehl, 1992). As described by Dollaghan (2004), the MAMBAC statistical procedure uses scores on two variables, at least one of which must be a continuous variable, from a large

sample that includes individuals with and without the disability of interest to analyze whether patterns of scores on variables of interest reveal a categorical or continuous condition. The interpretation of the MAMBAC analysis is conducted by plotting scores and reviewing and interpreting the curves associated with plots. Certainly, there may be non-statistical techniques that are also useful in delineating continuous vs. categorical conditions; nevertheless, the MAMBAC procedure offers an empirical statistical procedure that can supplement other methods.

Dollaghan (2004) used the MAMBAC taxometric procedure in two experiments with 620 preschool-aged children divided into two groups, one with SLI and one with age-matched controls with typical language skills. They were studied at two points in developmental time, at ages of three and four years. In the first experiment, four measures of language ability were used: 1) standard scores on the *Peabody Picture Vocabulary Test – Revised* (PPVT-R, Dunn & Dunn, 1981), 2) Mean Length of Utterance (MLU), 3) Number of Different Words (NDW) from a language sample, and 4) Number of Different Words reported by parents on the *Language Development Survey* (NDW-LDS, Rescorla, 1989). On all of the measures, the three-year-old children with SLI scored 1.5 or more standard deviations below the mean of their normally developing peers, demonstrating separation from typical expectations adequate to support a taxometric analysis. The MAMBAC procedure was applied twice, once with MLU as the input variable and NDW-LDS as the output variable, and once with NDW-LDS as the input variable and MLU as the output variable. The plot patterns that emerged suggested that SLI is a continuous disability (see Dollaghan, 2004). In the second experiment, the analysis was repeated with the same participants when they reached age four, using PPVT-R as the input variable and MLU as the output variable for the analysis. Again, the plots for the four-year-old participants revealed flat performance, suggesting a continuous disability rather than a categorical one.

The results of Dollaghan's experiments are intriguing. The field of language disability has long pursued a research pathway apparently guided by a vision of SLI as a discrete condition, fundamentally different from typical language development. It is possible that Dollaghan's outcomes revealed continuity in language ability because the variables used in the two analyses were not sensitive to the condition (i.e., one could argue that more explicit morphosyntactic variables might better reveal a discrete condition). Yet, much of the research in the field suggests that this explanation is not plausible. First, many continuous disabilities are characterized by heterogeneity in behavioral patterns and multiple etiological factors and origins (Dollaghan, 2004). It is well established that children with SLI are diverse in their linguistic strengths and weaknesses, despite certain patterns that are frequently associated with the condition (Rice & Wexler, 1996). In turn, children with SLI have much in common with children with other disabilities. For example, both children with SLI and children with nonspecific language impairment show processing delays (Miller, Kail, Leonard, & Tomblin, 2001). Furthermore, children with SLI and children with other language-learning difficulties have responded in a similar way to treatment protocols (Cole, Dale, & Mills, 1991). Finally, recent epidemiological studies revealed circularity in reasoning about SLI, particularly when it is identified as the lower end of the continuum of language disability. Specifically, studies of language ability in kindergarten children in the state of Iowa reported SLI rates of about 7% (Tomblin et al., 1997). These investigations used a criterion of −1.25 SD on standard measures to identify SLI. It is not surprising that the base rate corresponded to about 7%, roughly as anticipated given the criterion established. Thus, much available evidence regarding SLI validates Dollaghan's finding that SLI is best characterized as a non-categorical disability.

There are questions to be answered in relation to stuttering that are analogous to those addressed in the language disabilities literature. At the most basic level, is stuttering a continuous (i.e., dimensional) disorder or is it categorical (i.e., subtypes)? Additionally, even if SLI represents

a non-categorical disability, when language disability co-occurs with stuttering does it also vary along a continuum or would patterns of language disability in the presence of stuttering be better interpreted categorically? Similarly in reverse, if stuttering without concomitant language disability were found to be non-categorical, might the patterns of stuttering in the presence of language disability best be understood categorically?

Just as examination of the ability domain of language in relation to subtypes sheds light on the exploration of subtypes, likewise a study of approaches related to the attribute domain of temperament may be informative. In the next section, the existing literature on child differences in stuttering and temperament will be reviewed, as will the potential relationship among these variables, and the possibility of subtypes.

Review of Selected Literature: Stuttering and Child Temperament

Attributing the disorder of stuttering to certain personality characteristics, various emotional conflicts and maladjustments, and psychopathological conditions can be traced back to ancient times. Wingate (1997, p. 14) notes that Aristotle, who lived in the 4th century B.C., wrote in his book, *Problemata*, that stuttering is provoked by nervousness, a form of fear, that creates coldness. Psychological conditions have been suspected to be contributing factors not only to stuttering onset, but also to its persistence. Tanberg (1937) expressed that the successful treatment of stuttering depended on seeing the stutterer "not as a person having difficulty with this or that particular sound or word, or as a person whose speech is merely unrhythmical," (p. 656) but instead "as a person whose total personality is subject to emotional disturbances in speaking situations…" (p.656). The nature of those emotional disturbances, however, was not fully apparent, meaning that more work was needed to characterize them.

Various psychological models of the genesis of stuttering emerged in the 1920s, and investigations of stuttering as a form of psychopathology reached a peak around the 1950s (see Bloodstein, 1995; Goodstein, 1958; Sheehan, 1958 for comprehensive reviews). Although emotional factors were still incorporated into theoretical models of stuttering (e.g., Brutten and Shoemaker, 1967), the concentrated attention to personality and stuttering declined when behaviorism became popular in the 1960s–70s and researchers focused on conditions that might bring the stuttering response under stimulus control (Prins & Hubbard, 1988). In more recent years however, there has been a resurgence of interest in the exploration of personality traits (e.g., temperament) in young children who stutter in search of risk factors for persistence.

Research of stuttering in relation to all aspects of psychopathology, behavioral response, and personality, has yielded inconsistent results both across subjects and across studies. The aforementioned observation of heterogeneity in the sample population has been noted by researchers of this domain, as well (Anderson, Pellowski, Conture, & Kelly, 2003), leading to the question of subtypes. Recently, with the renewed interest of researchers in the personalities of individuals who stutter, the possibility of subtypes involving this domain should be considered.

Whereas past interest in psychological factors has centered on personality and emotional maladjustment, recent interest has shifted to temperament. Temperament may be defined as a collection of inherited personality traits that "constitute the individuality of the person" (Goldsmith et al., 1987, p.510). As a dynamic factor, temperament "mediates and shapes the influence of the environment on the individual's psychological structure" (Goldsmith et al., 1987, p.509), and concerns the "formal and stylistic features of behavior, such as the individual's sensitivity and responsivity to environmental demands" (Caprara & Cervone, 2000, p.87). Thus, the manner with which an individual reacts and operates in his/her environment is a function of temperament. Research suggests that traits such as emotional impulse control can be identified as early as the first year of life and are

quite stable, but differ across individuals (Caprara & Cervone, 2000). Differences in temperament help to explain why people respond differently to the same environmental stimuli.

Several stuttering experts have expressed belief that temperament shares an important role in the etiology of stuttering. Douglass and Quarrington (1952) proposed that their subtype classifications of interiorized and exteriorized stutterers were emergent personalities based in child-rearing practices. Yairi (in press) listed several other past subtype-classifications of stuttering that took personality into consideration, at times as the main basis for classification, (i.e., Brill, 1923). Guitar (2006) hypothesized that a sensitive (i.e., reactive) temperament contributes to the vulnerability of a child to the development of stuttering. He suggested that children who stutter tend to be more easily aroused by stimuli, but inhibited when confronted by unfamiliar people or situations. Perkins (1992) opined similarly that children at risk for chronic stuttering are apt to be those who are more easily intimidated. He proposed that stutter events were triggered when a child's strong need to be heard and understood collided with an opposing feeling, usually induced by listener behavior, that such importunate assertions must be kept in check (Perkins, 1992). Although a number of hypotheses regarding the relationship between temperament and stuttering have been offered, there has been limited examination with actual research.

Riley and Riley (2000) evaluated 50 elementary school-age children who stutter, and noted that increased sensitivity (reactivity) and high self-expectations were more prevalent than would be expected in a random population sample. Anderson, Pellowski, Conture, and Kelly (2003) examined the role of temperament in stuttering in preschool-age children between the ages of 3 and 5. Mothers completed temperament ratings of the children using the *Behavioral Style Questionnaire (BSQ*, McDevitt & Carey, 1978). Results revealed that mothers saw children who stuttered as being slower to adapt their behavior to changes in routine, more vigilant during tasks (less distractible) and more irregular in their physiological functions. The finding of less adaptability in children who stutter was reported in other research (Embrechts, Ebben, Franke, and van de Poel, 2000; Howell et al., 2004). Howell and colleagues (2004) also observed that none of their participants' language and temperament measures were markedly correlated, and speculated this may be because language ability in young children changes over time while temperament is relatively stable.

In contrast to other research, a study by Lewis and Golberg (1997) indicated that 3- to 5- year-old children "at-risk" for stuttering were more adaptable, and more regular in their physiological functions and routines. Similarly, another investigation of *BSQ* results for 20 preschool children who stutter found a higher proportion fit the "Easy Child" classification (high adaptability) compared to those among the 20 non-stuttering controls (Williams, 2006). It was reported that a large number of the children in the study could not be classified into any of Chess and Thomas' (1996) three categories of Easy, Difficult, or Slow-to-Warm Up, which will be described later in more detail.

Oyler (1996) examined personalities of school-age children who stutter and who do not stutter, in the interest of exploring the potential construct of vulnerability, which she defined as susceptibility to adversity associated with being disadvantaged and maladroit. Vulnerability was assessed via personal and personality characteristics, self-esteem, attending, organizational and memory abilities, communication attitudes, and child behavior problems. Children who stutter were found to be significantly more vulnerable than non-stuttering children. She also found that vulnerability was positively correlated with sensitivity.

More recently, Karrass et al. (2006) examined temperament variables in 65 young stuttering children and their controls, between the ages of 3:0 and 5:11. Results of the *Behavioral Style Questionnaire (BSQ)* completed by parents found that temperament variables of emotional

reactivity and emotional regulation differed significantly between groups. The preschool children who stutter were more reactive, and showed less regulation of emotions and attention, than their normally-fluent peers. The authors suggested that these differences may represent risk factors for social development, such as difficulty separating from parents, adjusting in school, and making friends. These psychosocial adjustment issues were also deemed to place a child at risk for language development. If these children tend to be less social and/or less communicative in social interactions, then it may lead to fewer experiences essential to language learning.

The only study found to be based on actual observation of temperament-related behaviors rather than parental questionnaires was conducted by Schwenk, Conture, & Walden (2007). They examined gaze behaviors of preschool children, 13 who stuttered and 14 who did not, in response to camera movements. The latency and duration of gazes were not different between the groups, but the children who stutter looked at the camera significantly more often than the control children. Based on these results, they proposed that children who stutter may be more reactive, and less apt to habituate to environmental changes, compared to children who do not stutter. Remarkably, there was no overlap of the frequency measures between the two groups despite the result that the range in the number of gaze shifts for the children who stutter was more than twice the size of the range for the control group.

The finding of greater sensitivity among young children who stutter is relatively frequent in the literature. This may represent however, the nature of children who have a speech or language disabilities generally. Hauner, Shriberg, Kwiatkowski, & Allen (2005) cited a wide array of studies of temperament in children with a variety of communication disorders and concluded that compared to typically speaking controls, the children with speech and language disorders scored significantly higher on measures of "sensitivity, anxiety, distractibility, neuroticism, withdrawal, and difficulty in adaptability." (p.637)

Studying temperament in young children as early as possible relative to stuttering onset, is important in order to discern whether such attributes comprise etiological factors influencing the development of stuttering, or represent problems that emerge as a consequence of social reactions to stuttering (Goodstein, 1958). Karrass et al. (2006) conjectured that the emotional temperament of young children who stutter developed secondary to their experience of stuttering and negative social feedback. Because previous investigations have shown that many children who stutter have strong language and/or psychosocial abilities, the interactions of temperament, language, and stuttering are increasingly important factors to consider in the matter of stuttering subtypes.

As an attribute domain, the interaction between temperament and stuttering is apt to be different in nature from the interaction of stuttering with an ability domain such as language. Language ability is concerned with the individual's manipulation of symbols to convey meaning. Thus, to the extent that stuttering is related to interruptions in the processes of manipulating symbols to convey meaning, then stuttering (and/or its subtypes), interacts with language ability. Temperament by contrast, is about the manner with which an individual reacts and regulates responses to sensory experience. Therefore, we could expect temperament to interact with stuttering (and/or its subtypes) to the extent that speech dysfluency is a function of one's manner of reacting and responding to experiences. There is insufficient research on the topic of how temperament may be involved with the onset of stuttering or its persistence, but once it has persisted it appears obvious that reactions and self-regulation (and thus temperament) play a major role in stuttering. Uncertainty remains about how the nature of that role should be described, and whether it is best represented in terms of subtypes. Several subtyping systems and approaches to the question of subtypes have been applied in the temperament literature, so the next step is to examine these studies to gain further knowledge.

Subtypes in Temperament

Generally, research on child temperament has observed a tradition of utilizing typological terms. Several different systems of subtyping child temperament have been developed. For example, Thomas, Chess, and Birch's (1968) landmark longitudinal study of temperament was based on data rating infants on 9 aspects of temperament: mood, approach-withdrawal (reaction to novelty), adaptability (to changes in routine), intensity, rhythmicity or predictability (body rhythms: sleep-wake cycles etc.), persistence (extent to which child remains engaged in an activity), threshold (sensitivity to external stimuli), activity, and distractibility (difficulty or ease with which an ongoing activity can be interrupted). Thomas et al. identified three temperament "types" based on these 9 dimensions: Easy, Difficult, and Slow-to-Warm-Up. Easy children were described as generally positive in mood, regular in body functions and habits, adaptable to new experiences, and reacting with mild to moderate intensity. Difficult children were described as generally negative in mood, active, irregular in cycles and habits, inadaptable in new situations and to new people, and reacting with high intensity. Slow-to-warm-up children were most striking in the extent to which they withdrew from new situations, but they were also described as slow to adapt to new situations, reacting with low to moderate intensity, and low in activity. Frequently family and friends label these children as shy or inhibited. Additionally, some children in this longitudinal study were not rated highly on any dimension, and were thus described as Average.

Another well-known study that proposed personality subtypes in young children was published by Ainsworth, Blehar, Waters and Wall (1978). Applying Bowlby's (1969, 1980, 1988) attachment theory, they documented that there were systematic individual differences in the ways in which infants organize their attachment with their parents. Ainsworth and her colleagues developed a procedure to examine individual differences in attachment behavior that came to be known as the "Strange Situation," a laboratory technique that involves a series of separations and reunions with the parent, and interactions with a stranger. On the basis of the infants' behavior in this context, children are classified into three major attachment groups: secure, avoidant, or resistant. In recent years a fourth pattern has been identified, "disorganized" (Main & Solomon, 1990). Although some researchers have suggested that a dimensional (non-categorical) approach may be more useful to characterize differences in attachment security (e.g., Fraley & Spieker, 2003), this three-, and more recently four-group taxonomy has been widely applied in the developmental literature and found to be predictive of responses in preschoolers up to 6 years of age (Main, 1996). Although attachment represents psychosocial behavior indirectly related to temperament; this line of research has contributed importantly to the broad realm of personality subtype identification in young children.

As with language, there is controversy regarding whether temperament should be described in terms of categorical or continuous dimensions. Several contemporary accounts identified clusters of characteristics to describe individual differences in temperament, but others have argued it may be advantageous to examine the various aspects of temperament (e.g., activity level, fearfulness, frustration tolerance) separately on their respective continua rather than lumping characteristics together into types. A dimensional approach to the study of individual differences in temperament has been supported by Rothbart and Bates (1998). Rothbart and her colleagues define temperament (which they regard as central to personality), in terms of individual differences in reactivity and self-regulation (Rothbart, Ahadi, & Evans, 2000; Rothbart & Bates, 1998). Biological evidence of reactivity, with regard to how easily an individual becomes aroused in terms of motor, affective, autonomic, or endocrine systems, has been noted even prenatally (DiPietro, Hodgson, Costigan, & Johnson, 1996). Self-regulation, referring to the processes of attention, cognition and behavior that modulate the individual's responses to reactivity, develop later, during infancy and early childhood.

As researchers explore potential subtypes of stuttering with respect to temperament variables, they will need to carefully consider whether a continuous or categorical approach to temperament is most appropriate, and which system/s of temperament traits or types best characterize these children and their differences. Global trait systems have sometimes been criticized because they can mask individual differences. Mischel and Shoda (1995) found that although two people score similarly on a global trait measure, this knowledge is insufficient to predict individual behavior. For example, with respect to aggression, one child may behave aggressively when threatened by peers but not when threatened by adults, while another child may behave just the opposite. This suggests that trait attributes should be interpreted cautiously taking into consideration the potential bounds of their variability. Although proponents of genetically-based temperament traits commonly regard them as context-free personality characteristics, Kagan (1994, 1998) similarly argues that inherited temperament mechanisms may be context-specific. He observed that children who tend to cry are not all alike, but differ according to the types of stimuli that evoke the crying. For example, some infants cry when physically constrained while others cry with auditory stimulation. Thus, it is apt to be important to study temperament variables in the context of observed behavioral responses, and not rely solely on parent-completed questionnaires.

Common Insights from the Domains of Language and Temperament

Can these findings offer guidance for the study of stuttering in young children, particularly the effort to characterize subtypes and risk factors? At least two general recommendations appear relevant. First, it does appear valuable to address the question of whether stuttering in young children better conforms to a categorically different developmental pathway, or whether a continuous ability is suggested. This is a fundamental question with far-reaching implications for understanding the condition, providing effective treatment, and pursuing informative research.

One useful outcome of examination of the child language and temperament subtypes literature, is the demonstration of a statistical method, taxometrics, that has the potential to address the question of the continuous versus categorical nature of stuttering. In order to undertake a taxometric study of stuttering, however, a large sample of young children would be required, with both children who stutter and typically developing peers, and selection of optimal indicator variables would be critical. Thus far, the accuracy of taxometric methods has been demonstrated with large sample sizes atypical of those that have been obtained in stuttering research (i.e., >300 cases), and even so, these methods rely on supplemental evidence of indicator validity (Haslam, 2003). In Dollaghan's (2004) research, the overall number of participants was large, but the number with SLI was not as large. From a large sample of young children, a population including participants who stutter could be identified. A more significant challenge in terms of applying the methodology to stuttering in young children may be the selection of relevant variables. The most logical candidates are measures of stuttering behavior, such as stuttering-like disfluencies, and possibly a clinical measure of stuttering severity. Even within this variable category, specific measures may be particularly sensitive. For example, in addition to frequency of SLD events, the length of the events may be crucial. Indeed Yairi and Lewis (1984) and Ambrose and Yairi (1995) found that the number of repetition units (iterations of a segment) was a powerful discriminator between children who stutter and normally speaking children. Yairi (in press) listed additional related findings. Another challenge in the application of this statistical methodology would be differentiation of young children whose stuttering will persist from those whose stuttering will abate. It would perhaps be most informative to conduct the analysis initially with a large group of young children near stuttering onset, and the follow-up analysis could be conducted when the children who stuttered could be differentiated into persistent and recovered groups, and each contrasted with typically developing peers.

Integrating Domains: Directions for Future Research

In a broad sense, future stuttering research could draw from the child language and temperament literature with particular focus on: 1) contrasting areas of relative strength and weakness, particularly in terms of illuminating differences and varied profiles of development, and 2) probing for certain linguistic skill gaps and/or temperamental response patterns that may be revealed only at developmentally sensitive time points and with appropriate measurement techniques. Both of these endeavors may be relevant to our search for subgroups.

The outcome of subtypes research has the potential to reveal relationships important to clinical practice, especially related to diagnostic, prognostic, and treatment decisions. Figure 1 illustrates the possible benefits of subtyping for clinical application based on characteristics of language and/or temperament.

Beginning with what is known about stuttering in children, available research suggests a condition that is heterogeneous in its symptoms and behavioral patterns (Yairi & Ambrose, 2005). In addition, it seems likely that stuttering in young children, even as a single disorder, is linked to a variety of causal factors. Despite these generally accepted facts, the field of stuttering research and intervention has proceeded as if stuttering is a discrete, categorical condition. With respect to temperament, an example of research to illuminate whether category or continuum is applicable could be conducted toward comparison and contrast of personality/ temperament characteristics of individuals who develop covert versus overt stuttering. This research would be useful to determine whether these two subtypes approximate a pattern that characterizes the stuttering disorder as a whole or separate subtypes, when contrasted with those who, disfluent or not, have not ever presented with any complaint of stuttering. Such research could potentially provide greater clarity for clinicians' efforts toward interpretation, prevention, and alleviation of the disorder.

Overall, it seems stuttering subtyping research could be advanced through application of recent insights from studies of child language disability and temperament. The existing knowledge base suggests several approaches and/or principles that should guide research for purpose of identifying patterns of risk factors and associated subtypes in young children who stutter. These principles are currently being applied in the studies conducted by the Illinois Stuttering Research Project.

First, patterns of risk factors in early childhood stuttering must be constructed through a multidimensional analysis that integrates varied domains and includes careful phenotypic descriptions. Single-characteristic classification systems have not been informative in studies of language disability, temperament or stuttering. Achieving a workable subtyping system will require multiple variables and a multidimensional analysis to elucidate patterns in available data on skills and abilities. One key point in multidimensional analysis is that the approach should be open to identifying strengths in relevant domains and subdomains, as well as limitations. With children with SLI, typical cognitive skills exist in conjunction with linguistic difficulties. With children who stutter, it is plausible that there are some children with typical and even above average language abilities who are, by that very characteristic, at risk for developing this communication disorder. To be fully informative, analytic and measurement approaches need to leave open the possibility of fluency problems that exist with potential trade-offs with strengths in related domains.

Second, integrating a taxometric analysis similar to Dollaghan's (2004) model may help address the issue of whether early childhood stuttering is most accurately viewed as a discrete, categorical condition or as a continuous, dimensional difficulty. This analysis is also promising in its potential to shed light on the identification of associated risk factors; it may be valuable to incorporate familial patterns and genetic findings, in concert with findings in behavioral

domains, such as language, social, and motor development, with behavioral components of actual stuttering behavior. Furthermore, given the changes in stuttering associated with age and age at onset, it will be essential in such as analysis to follow Dollaghan's model and repeat the analysis at varying points in development.

Third, longitudinal investigations of developmental trajectories in multiple domains have great potential to yield informative subtype profiles. Contrasting developmental pathways in several domains and subdomains across several years, beginning near the onset of stuttering, will be critical to evaluating initial data regarding the categorical or continuous nature of the condition. Assessments of the validity of various proposed subtypes can be aided by evaluation of converging versus diverging pathways over time, and alternative developmental trajectories that may be associated with relevant subgroupings. A longitudinal framework, beginning near stuttering onset, also enables evaluation of developmentally sensitive skills and trajectories.

Finally, expanding on the previous point, there is promise in examining connections between language ability and temperament in young children who stutter near the time of stuttering onset and across time. Language and temperament/social-emotional development share the characteristic of having significant potential to be influenced by a history of stuttering, so study beginning near onset is critical. Furthermore, associations between patterns of language development and profiles of personality/temperament may be relevant to advancing understanding of persistency and recovery in stuttering. The history of research in the domains of both stuttering and personality share several points in common. Not coincidentally, stuttering and personality both have been described with the same analogy. The mind, said Freud (1914), and stuttering, said Sheehan and Lyon (1974), are each like an iceberg, with the larger realm remaining under the surface of what people see or hear. The construct of personality has generated possibly as many models and as much controversy in the field of psychology as stuttering has generated in the field of communication disorders. Heterogeneity among individual personalities is not unlike the heterogeneity of speech characteristics among individuals who stutter. Finally, neither area has been fully understood.

In summary, this paper asserts that subtyping research in early childhood stuttering may be advanced by including both ability domains (such as language) and attribute domains (such as temperament) in efforts to develop subtypes. Examining the issue of whether stuttering is best characterized as the least fluent production of speech (continuous) or as a fundamentally different speech form of production (categorical) is relevant to theory and practice, and research specifically devoted to exploring this issue would be informative. Furthermore, subtyping efforts that recognize the developmental sensitivity of measures and domains, explicitly look for both strengths and weaknesses in skills, and seek contrasts between strengths and weaknesses, are likely to be the most promising. In order to incorporate sensitive measures and explore both strengths and limitations, informative future work needs to adopt a longitudinal time frame and utilize a multifaceted research approach.

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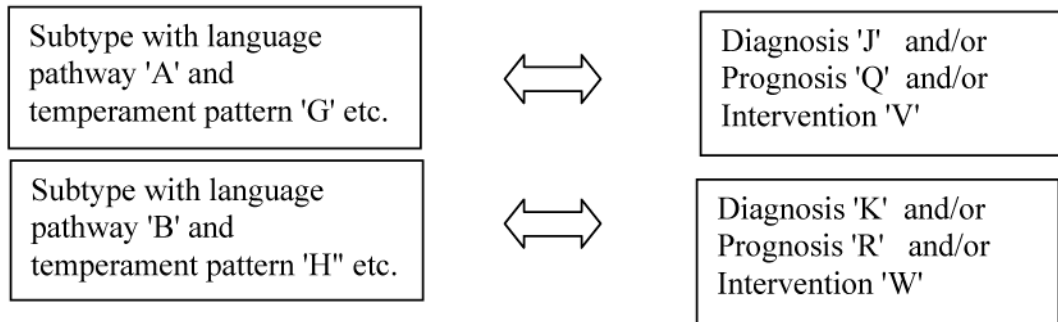
Figure 1.
Representation of potential subtype associations paired with clinical decision outcomes.