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Relation of motor, linguistic and temperament factors in epidemiologic subtypes of persistent and recovered stuttering: Initial findings

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ABSTRACT

Purpose: The purpose of this study was to determine the presence of any patterns reflecting underlying subtypes of persistence and recovery across epidemiologic, motor, language, and temperament domains in the same group of children beginning to stutter and followed for several years.
Methods: Participants were 58 2–4-year-old CWS and 40 age and gender matched NFC from four different sites in the Midwest. At the end of the multi-year study, stuttering children were classified as Persistent or Recovered. The same protocol obtaining data to measure stuttering, motor, language and temperament characteristics was used at each site. They have not been included in previous reports.
Results: The Persistent group performed consistently differently from the Recovered and Control groups. They performed lower on standardized language tests and in phonological accuracy, had greater kinematic variability, and were judged by their parents to be more negative in temperament.
Conclusions: The present study provides data supporting the hypothesis that subtypes of stuttering can be identified along persistency/recovery lines, but results were not definitive.

Educational Objectives: Readers will be able to (a) describe the current state of subtypes of stuttering research; (b) summarize possible contributions of epidemiologic, motoric, linguistic and temperament to such subtyping with regard to persistency and recovery.

1. Introduction

References to stuttering as a "complex" or "multifaceted" disorder abound in the scientific literature, reflecting a general recognition that a simple characterization of stuttering is not tenable. Although speech disfluency is its cardinal feature, stuttering, as a disorder, appears to encompass more than just speech production difficulties. It is interwoven within the language, phonological, cognitive, social, emotional, and physiological domains, creating a marked heterogeneity that is

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especially apparent when stuttering persists. It is fitting to point out that more than 50 years ago St. Onge and Calvert (1964) asked: "What are we studying when we study stuttering? Whatever it is, is it one, several, or many?" (p.160). While there has been a generally accepted distinction between developmental stuttering and acquired stuttering, within developmental stuttering there is no formal recognition of subtypes.[1] In this article we echo St. Onge and Calvert's (1964) question by reporting on progress from direct testing of whether persistent and recovered stuttering are viable subtypes.

Historically, there have been proposals for subtype classification of stuttering reflecting diverse orientations. Some were based on presumed etiology (Brill, 1923; Canter, 1971), some on different phenomena of stuttering (Douglas & Quarrington, 1952; Froeschels, 1943; Schwartz & Conture, 1988), and others on the presence or absence of concomitant disorders (Blood & Seider, 1981; Riley, 1971). Biological differences have also been linked to possible subtypes (Hinkle, 1971; Poulos & Webster, 1991). Van Riper (1971) attempted to differentiate stuttering based on distinct developmental courses of the disorder. Furthermore, these classifications ranged from being based on a single domain, such as different psychological states (Brill, 1923) to multiple domains, such as St. Onge (1963) triple types: psychogenetic, organic, and speech symptoms (Yairi, 2007).

The various subtype proposals, however, have been accompanied by little research or convincing evidence. In an early study, Berlin (1954) compared 110 people who stutter (PWS) divided into seven a priori defined subtypes based on: (a) family history, (b) laterality, (c) home environment, (d) presumptive brain damage, (e) diadochokinesis, (f) maladjustment, and (g) dysphemia. They were examined in relation to 12 variables including stuttering onset, disfluency, personality, diadochokinesis, and health history. The only significant findings were that presumed brain damage was associated with a more gradual onset and higher scores on the Minnesota Multiphasic Personality Index (MMPI). Hinkle (1971) pioneered the research of subtyping in relation to brain structure and function reporting that PWS who differed in brain lateralization during dichotic listening also differed in their stuttering patterns, severity, and level of the adaptation effect. Kroll (1976) reported high accuracy in parting interiorized from exteriorized stuttering, whereas Schwartz and Conture's (1988) cluster analyses of speech samples yielded a distinction between PWS predominantly exhibiting repetitions and those predominantly exhibiting sound prolongations. Feinberg, Griffin and Levey (2000) reported that subtypes could be discerned along personality, cognitive, and intellectual dimensions.

A number of recent brain studies of PWS have explored within-population differences. For example, atypically large right planum temporale in PWS was associated with greater disfluency than that of PWS with more typical morphology (Foundas, Bollich, et al., 2004; Foundas, Corey, Hurley, & Heilman, 2004). They also responded differently to altered auditory feedback (AAF), a finding reminiscent of Hinkle (1971). The first brain structure study of children who stutter (Chang, Erickson, Ambrose, Hasegawa-Johnson, & Ludlow, 2008), conducted at the University of Illinois with 9- to 12-year-olds, revealed differences in fractional anisotropy, a measure of white matter integrity, between children who persisted in, and those who recovered from, stuttering. The latter exhibited poorer integrity of fibers connecting mostly left cortical centers. Most recently, Chang, Zhu, Choo, and Angstadt (2015) employing a larger sample of considerably younger children (down to age 3), also found that the level of fractional anisotropy in tracts interconnecting auditory-motor areas and tracts that support skilled movement control differentiated CWS. Those with low fractional anisotropy had more severe stuttering than those with high fractional anisotropy. Additionally, there were statistically significant sex differences among CWS in the patterns of white matter development.

The Illinois longitudinal studies (see a comprehensive summary in Yairi & Ambrose, 2005) contributed considerable evidence for subtypes based on diverging developmental paths during the first few years after the disorder's onset. Our findings indicate two broad categories of developmental stuttering: (1) persistent, lasting more than 3 or 4 years after onset, and (2) natural recovery, showing complete remission within 3–4 years following onset. The recovery process can be seen during the first year of stuttering, although the process for the majority of cases tends to be completed during the second and third year post onset. There is not a continuous distribution of cases as the incidence of recovery drops sharply after that.

Our longitudinal measures of observable stuttering have been reinforced by segregation analyses on the pedigrees of 66 young CWS that provided positive evidence for genetic differences between persistency and recovery (Ambrose, Cox, & Yairi, 1997). Our team's genotyping studies also yielded persistent-recovered differences in chromosomes on which genes underlying stuttering were suspected to be located (Suresh et al., 2006; Wittke-Thompson, Ambrose, Yairi, Roe, Ober, & Cox, 2007).

Other distinctions supporting persistence and recovery subtypes were also reported for language and phonology (Yairi & Ambrose, 2005; Watkins & Yairi, 1997). For example, language skills of both persistent and recovered children were found to be slightly precocious near onset, but only the recovered group returned to a normative level while the persistent group continued with higher than expected skill levels. In phonology, the development of the late 8 phonemes lagged in persistent children compared to recovered and control children, even though the pattern of development was within normal limits for all the children (Paden, Ambrose, & Yairi, 2002). The contribution of language to persistent-recovered subtypes has been reinforced by recent studies at the Purdue University Stuttering Project. They have reported that 6–8 year-old children

[1] The term "subtypes" rather than "subgroups" is employed here. Subgroups can be formed for different purposes. For example, division into arbitrary subgroups may be based upon age, or assignment to a task. Other usages include statements such as "only a small subgroup of participants responded. . ." In contrast, clinical subtypes entail assumed or observed consistent differences in symptomatology and/or etiological factors.