The Formative Years of Stuttering: A Changing Portrait

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All speech-language pathologists who have practiced in the field as generalists are likely to be well aware that the disorder of stuttering can be devastating in its impact on an individual’s ability to communicate in countless situations. They know that sometimes stuttering is so severe that the individual is physically, and vigorously, straining when attempting to utter a word, and that in some cases, speech is so affected as to render it unintelligible. Interpersonal communication can literally come to a complete halt that may last as long as a full minute or two. After nearly 40 years in the field of speech pathology, I still keep running once in a while into perplexing stuttering cases the likes of which I have never seen.

Speech-language clinicians who have specialized in treating stuttering and have accumulated more direct experience know that speech is quite unpleasant for the person who stutters, as well as for his or her listeners, not only in extreme circumstances, but also during more common, shorter, and less severe stuttering events. These, too, are associated with various degrees of effort and physical tension that disrupt communication, and with many unpleasant emotional reactions, such as fear, anxiety, shame, and so on. Furthermore, the effects of stuttering may extend far beyond difficulties in verbal communication, leaving deep traces in the psychological, educational, occupational, and social spheres of the affected individuals. At times, the negative experiences of people who stutter (PWS) can be even traumatic. The author acutely remembers a real panic attack he experienced many years ago, an hour before delivering a paper at the American Speech-Language-Hearing Association (ASHA) convention, for fear of expected stuttering. For these reasons, in addition to stuttering being definitely a disorder of speech, it has been construed as eventually becoming “a serious personal disorder” (Bloodstein, 1960, p. 374).

Inasmuch as the current essay is focused on the formative years of stuttering, which is the early childhood period, one central issue is the extent to which the aforementioned characterizations also apply to very young children who stutter or have ever stuttered. We raise the question because traditional concepts would let us believe that many of these characteristics typically emerge in later years: late childhood, adolescence, or even young adulthood. Several influential developmental models were among the principal promoters of this concept. Froeschels (1921, 1943), for example, described early stuttering as (a) gradual in appearance, (b) consisting of effortless repetitions of syllables or words, (c) lacking in physical tension, and (d)
lacking in awareness and affective reactions related to the stuttering. Bloodstein (1960), although allowing for individual variations and some age overlaps, suggested that several overt characteristics, such as repetitions, hard contact, or associated mannerism, can be the dominating symptoms in the second stage in his development sequence of the disorder, most typical of school-age children, whereas certain cardinal covert emotional and social reactions, such as avoidance, were assigned to the fourth stage that typifies young adults. Perhaps the most extreme view was expressed by Bluemel (1932), who made a clear distinction between the very nature of “primary stuttering” and “secondary stuttering.”

As my research and clinical work became more focused on stuttering in preschool-age children, questions arose in my mind whether differences between whatever is referred to as “early childhood stuttering” and “advanced stuttering” are indeed as large as have been traditionally asserted. Are they large enough to be construed as different forms of the disorder? In this respect, until recently, there appears to be a good measure of confusion that pervades the field. Overall, I sense that a tendency for responding positively to the question dominates. It is reflected in Bloodstein’s (1987) notion that stuttering can, or should, be viewed as a disorder of early childhood, with the rationale being the fact that stuttering typically begins between ages 2 and 5. This notion was reinforced by Yairi (1993), adding that early childhood is also the period when many of the affected children exhibit natural remission from stuttering. In other words, it is amply clear that, in the great majority of people who have ever stuttered, the disorder begins and ends within the relatively narrow time window of the preschool years. Conture (1991, p. 367) emphasized the distinction between childhood stuttering and advanced stuttering, stating that “children who stutter are not small adults who stutter,” apparently implying that he too sees major distinctions in the respective age-related stuttering disorder.

Against such a background of opinions, we have always wondered why, for many years, a very large portion of basic research and clinical studies in stuttering, as high as 85% according to Adams (1986), was conducted on adults who stutter to the neglect of young children who stutter. Because of past dearth of research dealing with early childhood stuttering, limited credible data were available concerning the disorder’s epidemiology and characteristics, including onset, early overt and covert symptomatology, identification and differentiation from normal speech, developmental courses, and subgroup differentiation. The scarcity of information has led to two undesirable consequences.

First, there were not sufficient opportunities to test pervasive, and important, assumptions about the characteristics of early stuttering and its variance from advanced stuttering. For example, the belief that young children who stutter have no, or only little, awareness of their stuttering has been accepted, even recently (e.g., Silverman, 2004), without a serious challenge, and had enormous, lasting influence on what treatment modes were acceptable. Accordingly, to prevent the development of the child’s awareness of his or her speaking difficulties that, supposedly, would only worsen the stuttering, therapy had to be conducted indirectly, through parent counseling. Direct therapy with the child was shunned if at all possible. Similarly, it was widely accepted that secondary physical characteristics occur typically late into the stuttering problem and are not a common feature of stuttering in preschool-age children near onset of stuttering.

Second, in rather peculiar, if not absurd, ways, while still holding to the position of major differences between early and advanced stuttering, we have allowed the lopsided focus on advanced stuttering to spawn inferences about the etiology and nature of stuttering in children based on data or models derived from adults who stutter (Yairi, 1990). For example, beliefs and theories about the role of anxiety in the formation and maintenance of stuttering (Brutton & Shoemaker, 1967; Wischner, 1952) were significantly influenced by strong manifestations of such emotions in adults who stutter. In spite of the position that children who stutter are not just small adults who stutter, in either proposing or accepting such theories, minimal considerations were given to the common observation, albeit not sufficiently tested, that anxiety is not clearly detected in most young children when they begin stuttering between ages 2 and 4.

In the past 25 years, however, there has been a significant shift in the target population of both basic and clinical research, effecting an intensely sharper focus on early childhood stuttering; for example, the research of Edward Conture on the physiology of stuttering (Conture, Rothenberg, & Molitor, 1986), and the research of Onslow and his associates on the treatment of stuttering (Onslow, Andrews, & Lincoln, 1994). In addition to the overwhelming evidence that stuttering typically begins and ends during the preschool years, the significance of childhood stuttering for the overall perspective of the disorder stems from a number of critical observations. Among them is the extensive overlap between the timing of the onset of the disorder and the critical period of speech and language development and the evidence that stuttering is associated with language functions. It is also a period of rapid neuromotor development; indeed, the disorder appears to involve problems in motor control. Not less important, it is the only time when the enormous influences of the home environment on shaping of the disorder can be studied closely. Lastly, different developmental pathways emerge during formative stages of the disorder. Adding realization of past neglect to the apparent great potential of turning the field into a new direction, I, with the generous contributions of several colleagues and many students, have become increasingly involved in studying stuttering during its formative years. Naturally, the question of how much the characteristics of stuttering in children who have just begun stuttering differ from advanced stuttering has continued to guide my curiosity. As a result of refocusing the research spotlight, noticeable transformations have taken place in current knowledge and theoretical thinking about early childhood stuttering. An attempt will be made here to review some of the changes in current knowledge that have emerged, mostly by presenting the work that has been accomplished at the University of Illinois where I have been a professor of speech and hearing science for the past
27 years. (Much more extensive detailed information can be found in Yairi & Ambrose, 2004.) Obviously, this kind of review bears heavily my point of view, perhaps bias, so readers, especially students, should be extra critical in assessing the information and conclusions.

THE ILLINOIS INTERNATIONAL STUTTERING RESEARCH PROGRAM

For more than a quarter of a century, the University of Illinois Stuttering Research Program has focused on stuttering in preschool-age children, although not to the exclusion of other age groups. Over the years, it has been supported by several major grants awarded to us by the National Institutes of Health, National Institute on Deafness and Other Communication Disorders (NIDCD). With time, several faculty members in our department (Professors Nicoline Ambrose, Elaine Paden, Adele Proctor, and Ruth Watkins), as well as my former doctoral students (e.g., Professor Kelly Hall from Northern Illinois University and Professor Rebecca Throneburg from Eastern Illinois University) and colleagues in other universities (Professor Nancy Cox from the University of Chicago), joined the team with active participation, providing expertise in several domains, such as language development, phonology, speech acoustics, personality, multicultural aspects, genetics, and more. Furthermore, scholars from other countries, including Israel, Sweden, and the United Kingdom, have joined too, making the program a truly international venture. I believe that this is the largest team that has been researching stuttering. More recently, the Illinois Stuttering Research Program became an important component in a large research project that includes several other universities in the United States, including the University of Iowa and the University of Wisconsin at Milwaukee.

Objectives

The overall goal of the part of the Illinois Stuttering Research Program reported herein has been to study the epidemiology and pathognomonic development of stuttering in early childhood with a focus on changes that occur with time in the characteristics of stuttering and the differentiation of persistent and naturally recovered subtypes of the disorder. Thus, we sought to:

- study the onset of stuttering: time, circumstances, types
- delineate and quantify a wide range of characteristics of stuttering and other related domains
- study the developmental course of these characteristics during the early years of the disorder’s existence
- determine the incidence and timing of natural recovery as well as the occurrence of persistent stuttering
- identify risk factors that differentiate persistent and recovered stuttering
- isolate the best means for early prediction of recovery and risk for persistency
- identify genetic bases of stuttering in general and its two subgroups: persons with persistent stuttering and persons with recovered stuttering
- expand research of early childhood stuttering to minority children, especially African Americans

A central component of the project was construed as a hypothesis-driven investigation, testing the assumption that persistency and recovery can be differentiated (a) behaviorally, on the basis of stuttering characteristics and other developmental domains, including phonology, language, and motor speech control; and (b) etiologically, on the basis of the type of underlying genetic liability.

METHOD

Procedure and Subjects

The project was designed as a longitudinal investigation where a group of preschool-age stuttering children were first seen close to the onset of their stuttering. Only children younger than 6 years who had stuttered for less than 1 year were admitted. (A few seen later than 1 year post onset were dealt with separately). Multiple objective and subjective criteria ensured that the children exhibited stuttering (see Yairi & Ambrose, 1999, for details). A group of normally fluent control children was also employed. By the end of the project, 190 children who stutter, 50 nonstuttering control children, and approximately 450 parents of the two groups took part in the investigation.

Each child underwent comprehensive initial observations, testing, and recordings that typically required several hours to complete. Lengthy speech samples were audio- and video-recorded and tests of language development, phonological skills, motor skills, hearing, awareness of stuttering, anxiety, and others, were administered. Parents were the main source of information pertaining to the detailed circumstances and characteristics of the onset, as well as the characteristics of early stuttering, before the child was examined by us for the first time. The child’s progress from onset to entry into the study, his or her health and physical development, and the family’s background and its history of stuttering were also obtained from parents through detailed and repeated interviews.

Follow-ups were conducted every 6 months, although some children were seen at the beginning at 3-month intervals. The follow-ups continued for at least 4 years and up to a period of 8 years, repeating the various observations, tests, tasks, and recording of speech samples. Several tasks and tests were administered only once a year. Parents also provided progress reports and rated the child’s stuttering at home. In many cases, especially when parents noted that the child stopped stuttering and no, or low level, disfluency was observed in the laboratory as well, speech samples were also recorded at home for verification.

In addition to the main investigation, a special grant awarded by the NIDCD enabled us to conduct an extensive survey of the incidence of stuttering in preschool-age children, especially African Americans. In this part of the
project, 3,414 children between 2 and 6 years of age participated, the majority of whom were African Americans and the rest represented other minorities and European Americans. They were identified and recruited in 49 different preschools in urban, rural, and suburban regions of Illinois.

Data Analysis

Thousands of audio- and video-recorded speech samples obtained during the course of the longitudinal investigation were carefully analyzed to classify and quantify all disfluent speech events in order to describe the early characteristics of stuttering and the changes that occur over time (Yairi & Ambrose, 2004; Yairi & Ambrose, 1999). These have been the core data of the investigation. Videotapes of selected children were also analyzed to study secondary head and neck movement associated with disfluency. Using acoustical analyses, various temporal parameters of the children’s fluent and disfluent speech, such as speaking rate, vocal fold fundamental frequency, and second formant transition, were studied for selected subsamples of children. The speech samples were also used to derive expressive language data while scores on formal language, phonology, motor, nonverbal skills, and psychological reactions tests were derived. Data from parent interviews were used to determine the time of onset, classify the type of onset, describe the early disfluency and its physical characteristics, and assess possible emotional reactions. Data from familial pedigrees were processed with special statistical software programs for genetic analyses. Data on the frequency of stuttering were used to track children over time and to determine those children who exhibited natural recovery and those who developed chronic/persistent stuttering. Once members of these two important subgroups were classified, after several years of follow-ups, data taken from the initial and later visits were used to study the possibility that the two groups can be identified and predicted from early on.

FINDINGS CONCERNING EARLY STUTTERING

Prevalence and Incidence

The incidence of stuttering, that is, how many people have ever stuttered for various lengths of time, has been estimated by only a few investigators at approximately 5% for the period between early childhood to young adulthood (e.g., Andrews & Harris, 1964; Mansson, 2000). On the other hand, numerous surveys have led to the conclusion that the prevalence of stuttering, that is, the percentage of people who are identified as active stutterers at a given point of time when the survey is conducted, is either a little more or a little less than 1% (Bloodstein, 1995). The figure of 5% incidence is quite impressive in projecting the magnitude of the disorder of stuttering and the number of affected individuals who may require at least some clinical attention, limited as it may be in many cases to initial evaluation, counseling, and periodic monitoring. It appears to me, however, that the smaller figure of 1% prevalence has become deeply ingrained in the literature and common knowledge and is typically perceived as “small” compared to other disorders of communication. The larger figure, the 5% incidence, is scarcely recognized by our scientific and professional discipline, thus affecting academic curricula and clinical training in the area of fluency disorders, typically by de-emphasizing clinicians’ training in it. Even more worrisome is the lack of understanding or clear appreciation of the simple fact that the 1% prevalence figure is an average for the population at large, covering a large age range. But, because stuttering is, to a significant extent, a disorder that prevails during early childhood, an uneven age distribution of its prevalence should be expected, with a much larger case concentration at the low end of the preschool-age range.

Although our longitudinal project did not originally include a prevalence study based on a representative sample of the preschool population in the United States, our focus on the disorder in African American children provided some relevant data (Proctor, Duff, & Yairi, 2002). A fairly good number, 3,414, of preschool children participated. Of these, 2,223 were classified as African Americans and, 1,191 represented other minorities and European Americans. Inasmuch as possible, identification of stuttering was done through three methods: investigator screening of individual children, staff identification, and parent identification. The key results were that the prevalence of stuttering in the African American children was 2.6%, and there were no statistically significant differences in the prevalence from same-aged European American or other children. These findings have important implications in shattering the common notion that stuttering is a small problem in terms of its numbers. In preschool-age children, it is presented quite often. Those who are educated and trained to become practicing clinicians should have adequate knowledge and experience with the disorder in this age group. Additionally, our findings negate previous reports that the prevalence of stuttering among African Americans is higher than that among European Americans. Incidentally, for all groups, there was a statistically significant difference for gender, with males exhibiting a higher prevalence of stuttering than females.

Onset

Results of the University of Illinois Stuttering Research Program provide much new information on how stuttering begins—information that significantly contradicts the traditional views, such as those expressed by Froeschels (1943), Bluemel (1932), and especially Johnson et al. and their diagnosogenic theory (1959). On the most general level, it can be stated that our studies negate past ideas that emphasized uniformity in stuttering onset. We have observed substantial variability in many aspects of the disorder’s onset.

Age. A new, and potentially important, finding of our project is that the age at which stuttering begins is younger...
than what has been thought. The mean age at onset, 33 months, is approximately 9 months earlier than what we calculated from 11 prior reports. Although the study was open to children just under the sixth birthday, 68% of onsetstook place before the children reached age 3 or 36 months of age (59% between 25 to 36 months of age), with 95% of all cases occurring by age 4 years. Put in other words, we currently estimate that the risk for stuttering could be more than 2 years earlier than what Andrews (1984) estimated based on his studies in the United Kingdom. If our findings are valid, the extensive overlap (1984) estimated based on his studies in the United Kingdom. If our findings are valid, the extensive overlap constitutes a different subgroup.

**Type and severity.** Whereas in the past, there were strongly held views that stuttering onset is uniformly mild and gradual, our findings show great variations. Nearly 30% of the children in our project were reported to have sudden onsets that occurred over 1 day. This figure rose to 40% by adding those for whom onset was reported to occur over 2–3 days. Intermediate onsets (a week or two) comprised 33% of the cases, whereas gradual (more than 2 weeks) onsets constituted the smallest group—only 25%. As for stuttering severity at onset, most parents rated the initial stuttering as moderate (45%) or severe (20%); only 35% rated it as mild.

**Conditions.** Traditional views have depicted stuttering beginning in uneventful circumstances. Once again, our data reveal great variability. Although a little more than 50% of the children began stuttering in uneventful circumstances, in an almost equally large proportion, onset was associated with various stresses: physical, emotional, and language expression. For example, approximately 14% of the children experienced illness or physical fatigue just before onset, more than 40% experienced emotionally upsetting events, and approximately 50% were reportedly undergoing language “stress” such as fast developing vocabulary when they began stuttering. Although, as will be discussed later, genetic factors appear to have a large role in stuttering, these findings emphasize the importance of environmental factors and the need to continue investigating them.

### Speech Characteristics

**Disfluency features and stuttering severity.** Traditional beliefs suggest that stuttering is almost always mild and begins with easy repetitions of sounds and syllables that are hardly distinguishable from normal. In contrast, our data show diverse symptomatology from the very beginning, including multiple repetitions (in one case, we counted 22 repetitions of a syllable), tense sound prolongations, complete silent blocks, respiratory abnormality, and complex secondary characteristics. Just about every “advanced” form of stuttering can be detected at the early stages in some children. Although Bloodstein (1960) also mentioned that complex symptomatology can be found in a few children within the age range of his “Stage I” of stuttering, we have observed such complexity in a good number of children, and very close to the time of onset.

Data on the overall severity rating of the early stuttering are quite illustrative. Instead of the expected high concentration of children having mild stuttering, the result of the severity rating made by us (the investigators) reveals a distribution that is skewed the other way. The curve shows 27% of the children rated as having mild stuttering, 45% as moderate, and 28% as severe. Thus, when the disorder was initially assessed, most children were rated as having moderate to severe stuttering. Note the close similarity to the severity ratings given by parents that were presented above.

**Disfluency norms.** Until recently, normative disfluency data for preschool children in the early stage of stuttering did not exist. Old data reported by Johnson et al. (1959) were acquired by pooling speech samples of children who spanned the ages from 2 to nearly 9 years. Furthermore, many of the children were recorded long after stuttering onset. Any speech-language norms obtained by mixing together data for 2-year-olds all the way to 9-year-olds make little sense. Therefore, an important contribution of our project was the publication (Ambrose & Yairi, 1999) of an essential normative reference for children who stutter and for normally fluent controls, limited to the narrow range of 2½ to 5 years and based on speech recorded within 6 months after onset. Furthermore, data for subsets for each year within the range, as well as for subsets of mild, moderate, and severe stuttering, are provided in the Ambrose and Yairi study.

### Identification and Differentiation of Stuttering

Contrary to traditional concepts of substantial overlap between speech disfluency of young stuttering children and normally fluent children (e.g., Johnson et al., 1959), our findings indicate marked differences in several dimensions, even at the earliest stages of stuttering. One of these differentiating dimensions is the frequency count of three disfluency types: part-word repetition, single-syllable word repetition, and dysrhythmic phonation, collectively referred to as stuttering-like disfluency, or SLD. Children who stutter exhibit, on average, 5 to 8 times more SLDs than their normally fluent peers (Ambrose & Yairi, 1999; Yairi & Lewis, 1984). Our most recent data show that children near onset of stuttering had 11.30 SLDs per 100 syllables in comparison with 1.41 for normally fluent children (Yairi & Ambrose, 2004). We found that disfluency types such as interjections, phrase repetitions, and revision, referred to collectively as other disfluencies, or ODs, that have been included in diagnostic instruments of other clinicians, do not contribute to the differentiation of the children who stutter and those with normal disfluency. Our respective figures for ODs are 5.79 and 4.48 per 100 syllables. Thus, some narrowing of relevant symptomatology has occurred.

The second dimension is the length of disfluency as reflected in the number of iterations, sometimes referred to as repetition units (RUs), per disfluent event. This is a powerful discriminator. Children who stutter repeat a syllable, or a single-syllable word, more times than do
focused our research on the temporal differences, leaving a very small zone of overlap (Ambrose & Yairi, 1999).

Beyond frequency and number of iterations, we also focused our research on the temporal characteristics of disfluency, especially the speed, or rate, of the repetitions, as a third differentiating dimension. The question was whether or not children who stutter repeat syllables or words faster or slower than normally fluent children. To answer, it was necessary to know whether the duration of each of the iterations in, for example, “on-on-on-on” and/or the three intervals between the four productions of “on” differ for the two groups of children. As reported by Yairi and Hall (1993) and Throneburg and Yairi (1994), the results showed that in children who stutter, such intervals between the repeated productions were only one half, or even one third, the duration of the intervals produced by normally fluent children. It means that the repetitions of children who stutter are much faster in tempo than normal. The findings negate earlier beliefs that repetitions are initially of normal tempo, becoming faster at later stages of the disorder (Froeschels, 1921; Van Riper, 1971). Interval duration alone was sufficient to differentiate children who stutter from children who do not stutter with 72%-87% accuracy, depending on disfluency type.

These findings offer an important clue as to why parents (and other listeners) perceive their children’s disfluencies as stuttering. To further investigate the effect that specific segment durations within disfluency have on the perception of “stuttering” or “normal,” we digitally lengthened and shortened vowels within repeated syllables or words and intervals between their iterations. Both factors, interval between repetitions and vowel duration of the repeated segment, affected listeners’ perception. As intervals and vowels durations were shortened, the disfluent event tended to be judged as stuttering. When they were lengthened, it tended to be judged as normal (Amir & Yairi, 2002).

Here too, an important finding of the Illinois Stuttering Research Program was the contradiction of this traditional view by demonstrating with hard data, speech and otherwise, that stuttering presents divergent developmental paths, evidenced especially by a strong phenomenon of natural recovery. We concluded that natural recovery is a powerful factor in early childhood stuttering (Yairi & Ambrose, 1999, 2004). The findings are convincing that the rate of natural recovery is at least 75% of all children who begin to stutter, whereas only 25% persist in what eventually becomes chronic stuttering. These are conservative estimates, reflecting close agreement among several measures, including frequency of SLD obtained from speech samples, clinician rating of stuttering, and parent rating of stuttering.

Complete recovery occurs in approximately 12% of children during the first year after onset, with 62% recovery within 3 years after onset and 75% within 4 years. Additional cases still recover up to 5 years following the onset of stuttering. Thus, although natural recovery appears to accelerate from the first to the third year after onset, it tapers off beyond that point. Whereas a child who has just begun stuttering has a 75% (or more) chance of natural recovery, after 3 years of stuttering, the chance diminishes to only about 15%. The rate (percentage) of natural recovery is higher among girls than among boys. Girls also recover sooner than boys. An important finding is that by about 12 months following onset, the disfluency profiles of children who eventually recover, as a group, and those of children who eventually persist, become significantly diverged.

Interestingly, independent listeners were unable to distinguish the speech samples of children who recovered naturally from the speech of normally fluent controls (Finn, Ingham, Ambrose, & Yairi, 1997). Furthermore, there are good reasons to believe that the real figure of natural recovery is higher by at least several percentage points. Obviously, these findings have major implications to theories and models of stuttering and for the conduct of all research with young children in terms of the need to consider population heterogeneity in subject selection. There are also critical clinical implications for assessing the risk for chronic stuttering at each stage after onset and for making decisions concerning treatment. Finally, clinical efficacy studies must employ control groups to minimize confusing the effect of treatment with the effect of natural recovery.

FINDINGS CONCERNING THE DEVELOPMENT OF STUTTERING

Once stuttering exists, the question is, What happens next? What changes does it undergo? Or, does it remain static? Is there a single course or diverse development? Does it become a lifetime disorder or is it only a temporary condition? Historically, the answers were fairly clear and widely accepted. In spite of observations by many clinicians that a good number of children stop stuttering on their own accord without treatment (Bryngelson, 1938), and in spite of several research studies that supported these clinical observations (Sheehan & Martyn, 1970), stuttering continued to be portrayed as a generally unitary disorder that progresses in a relatively uniform, linear, step-wise succession, gradually increasing in severity and abnormality.

In addition to data specific to the features of stuttering, our research program has yielded a wealth of information about several domains, both speech and nonspeech, that were investigated in the participating children and parents. These domains were selected because of past indications of possible relations to the disorder of stuttering. Pertinent findings concerning these domains, reflecting the children’s initial performance near stuttering onset, and, when appropriate, changes that occurred over time, are reported below.

OTHER SIGNIFICANT DOMAINS

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Awareness of Disfluency and Stuttering

Most 2- to 4-year-olds do not talk about their stuttering. Does this mean that they are not aware of it? In general, traditional views of early childhood provided a positive answer, holding that most, if not all, children who begin stuttering have no, or very little, awareness of their stuttering and no, or only little, emotional reaction to it until years into the problem (e.g., Luper & Mulder, 1964). Awareness, it was believed, comes at a later stage (e.g., Blumel, 1932). This position has had far-reaching consequences on the conceptualization of childhood stuttering, especially its treatment. Specifically, it was a major factor that gave prominence to indirect treatment in the form of parent counseling. It was reasoned that direct speech therapy would lead to, or enhance, the child’s awareness of the stuttering, causing emotional reactions and more severe tension. The data we obtained from the parent interviews (Yairi, 1983; Yairi & Ambrose, 2004), however, revealed that more than 20% of the parents felt that their children behaved in ways that, directly or indirectly, reflected awareness of their speaking difficulties at, or soon after, onset. For example, the children turned their heads and looked down; others frowned or looked frustrated. Most (12%), however, reported a high level of awareness, citing children’s specific comments such as “I can’t talk.”

Our research concerning awareness was further pursued, employing more direct methods where the child had to identify his or her speech with that of either of two puppets shown on a television screen, each saying the same sentence. One puppet spoke fluently and one spoke with disfluencies. Several sentences were used, and the order of the fluent and disfluent puppet, as well as its place on the screen, was controlled. Correct identification above chance was taken as an indication of awareness.

In an early study (Ambrose & Yairi, 1994), it was shown that early on, approximately 10% of children who stutter showed indications of possible awareness; an additional 10% indicated clearer and even highly consistent awareness. In a much larger set of children (Yairi & Ambrose, 2004), 20% showed indications of possible awareness and an additional 10% indicated clearer, more consistent indications. Likewise, normally fluent children show signs of being aware of the differences between fluent and disfluent speech. It is a challenging task, however, to assess awareness over time among young children who stutter because so many of them greatly improve or stop stuttering and begin to identify themselves with the fluent puppet. In any event, as age progressed, the agreement between the children’s fluency status and a puppet with whom they identified increased. Awareness grows rapidly between ages 4 and 5. Our cooperative research in Israel with a large number of normally speaking children (Ezrati-Vinacour, Platzky, & Yairi, 2001) confirmed the conclusions regarding the rapid growth of awareness of disfluency between ages 4 and 5. This information, concerning both children who stutter and their normally fluent peers, is extremely relevant to understanding the development of emotional reactions in children who stutter, and the reactions of their social environment to their stuttering. The data also seem to justify more direct approach in treatment.

Secondary Characteristics

Until recently, the phenomenon of physical tension and body movement associated with stuttering events was assumed to be a characteristic of advanced, or “secondary,” stuttering, a term suggested many years ago by Blumel (1932). Only a few authors reported occasional parents’ comment regarding its presence in early stuttering (Johnson et al., 1959; Yairi, 1983). Conture and his coworkers (e.g., Conture & Kelly, 1991; Schwartz & Conture, 1988) were among the first to conduct direct analyses of secondary characteristics in children who stutter using frame-by-frame videotape measurements. All of the children in their studies exhibited some or many head and neck, torso, and limb movements while stuttering, and in numbers that were significantly larger than that for normally fluent children. However, the age range of the children, 3 to 9 years, suggests that many of them were tested long after stuttering onset.

Using videotapes, secondary characteristics were also investigated in our project. One study of 16 very young children, all within 3 months of onset (Yairi, Ambrose, & Niermann, 1993), showed that the mean number of movements per instance of disfluency was 3.18, twice as many as reported by Conture and Kelly (1991). Later, Throneburg and Yairi (2003) reported a second study of head and neck movement in 32 stuttering children. Here, each disfluent instance was matched with a fluent segment in the child’s speech, and the difference in the number of movements between the two matched segments was calculated. The group data showed that near stuttering onset, the mean number of movements in disfluent segments was 4.8 as compared with 3.9 for the fluent segments. Head, eyeball, and eyelid movements were the most common.

We also conducted a perceptual rating of secondary characteristics for 103 stuttering children (Yairi & Ambrose, 2004). It showed that near stuttering onset, approximately 75% of the children had some degree of physical behaviors present during moments of stuttering. These were observed without the aid of slowed down video. On a severity rating scale from 0 (none) to 1.00, the secondary characteristics of approximately 37% of the children were rated between .25 and .49; for another 37%, the secondary characteristics were rated between .50 and 1.00.

Developmental aspects of secondary characteristics were also investigated. Throneburg and Yairi (2003) conducted an extensive longitudinal video analysis study that tracked changes in 19 different types of head and neck movement in 32 stuttering children in our project. Of these, 10 eventually persisted in stuttering and 22 experienced natural recovery. Within 6 months after onset, both groups exhibited large amounts of secondary characteristics. Thus, a large number of physical concomitants near the onset of stuttering do not appear to be a warning sign of chronic stuttering. However, beyond the first few months, the children who eventually recovered evidenced a sharp decline over time, whereas those who eventually developed
chronic stuttering evidenced more secondary concomitants and their number remained fairly stable (it did not increase over the next 3-year period). It would appear, therefore, that the absence of minimal change of physical concomitants after the first 6 months of stuttering may be indicative of persistency.

The Language Factor

That stuttering is connected in some, or many, ways to language has been known since early demonstrations that linguistic factors, such as grammatical variables, order, and length of words, appear to have a large influence on where stuttering events occur within utterances (Brown, 1945). Later investigators have shown that language complexity is a factor in the occurrence of stuttering events (Logan & Conture, 1995; Yaruss, 1999). Understandably, linguistic variables are implicated in several theoretical accounts of stuttering (Perkins, Kent, & Curlee, 1991; Wingate, 1988); some go so far as to suggest that stuttering is basically a disorder of language development (Levina, 1968). In preschool children, a link between language and stuttering in young children is a particularly inviting proposition. Yairi (1983) and other scholars (e.g., Bernstein Ratner, 1997) have pointed out that the most common age range for stuttering onset, ages 2 to 4, overlaps with a time of rapid expansion in language ability and phonological skills in young children. Several studies found that children who stutter tend to be delayed in their acquisition of language abilities. In an early review of the literature, Andrews et al. (1983) concluded that children who stutter performed more poorly than nonstutterers on tests of language development, a conclusion echoed by Wingate as late as 2001.

In the Illinois Stuttering Research Program, the status of language abilities of the young children who stutter was evaluated near onset of the disorder by employing a range of lexical, morphological, and syntactic measures derived from the recorded conversational speech, as well as from formal tests. The findings of our extensive research have questioned the traditional belief that children who stutter lag behind in language development. We reported that, as a group, preschool children near the time when they begin stuttering fall well within the normal range for language skills or even above (Watkins & Yairi, 1997; Watkins, Yairi, & Ambrose, 1999). Supported by recent findings by other scholars (e.g., Rommel, Hage, Kaledine, & Johannsen, 1999), this finding has important clinical implications. For example, it raises doubts about traditional suggestions to clinicians, as well as parents, to minimize the complexity of language stimulation for children either in therapy or at home.

Another question was whether or not children who later would exhibit natural recovery from stuttering are different in language abilities from those who would develop persistent stuttering. Within our longitudinal study, the data obtained by tracking language skills across several years indicate that there were indeed some differences. Those whose stuttering was transient tended to show a slower course of language development, with their language skills eventually approaching near normative data at about the same time that their stuttering recovered. In contrast, the children whose stuttering persisted tended to maintain above-average expressive language abilities across the time frame of the study (Watkins, in press). Additionally, in early years, several children in the persistent group exhibited unusual language developmental patterns although they were still within normal range. Thus, children who stutter, as a group, do not trail in language development, and it is even possible that accelerated language development is a risk factor for persistent stuttering.

Cognitive Intellectual Abilities

Past studies indicated slightly lower overall mean IQ scores for children who stutter as compared to their normally fluent peers (Andrews & Harris, 1964; Okasha, Bishry, Kamel, & Hassan, 1974; Schindler, 1955). In view of the structure of IQ tests, it is possible that the somewhat lower mean IQ reported for PWS reflects depressed performance in only a single skill, or a few skills, rather than overall intelligence. However, except for data showing that incidence of stuttering, or level of disfluency, is higher in populations referred to as “mentally retarded,” including in children with Down syndrome (Otto & Yairi, 1976), there are no other indications of meaningful intellectual deficit in PWS.

In our project, the Arthur adaptation of the Leiter International Performance Scale (Arthur, 1952) was given at initial evaluations to determine if nonverbal skills (e.g., recognition of basic color, shape, functions, and patterns) were within normal limits and comparable to the child’s verbal skills. Overall, scores for the stuttering children were above normal limits. Here, too, we were interested in the persistent versus recovered group’s comparisons. Children who later developed persistent stuttering scored slightly lower (but still within the upper half of the normal range) than the children who later recovered or the normally fluent controls. Thus, this factor makes only a negligible contribution to prediction of the future course of stuttering.

The Phonology Factor

The scientific literature makes frequent references to a link between childhood stuttering and phonological disorders. Most studies have agreed that the incidence of phonological difficulties is higher for children who stutter (e.g., Arndt & Healey, 2001; St. Louis & Hinzman, 1988), but the specific percentages were found to vary widely due, in large part, to experimental procedural differences. Wolk, Edwards, and Conture (1993) estimated that, on average, 30% to 40% of children who stutter also exhibit disordered articulation or phonology. A more conservative estimate suggested that the incidence is greater than the 2% to 6% expected for the general population (Beitchman, Nair, Clegg, & Patel, 1986).

There are, however, opinions that question the procedures and conclusions of past studies regarding a link between the two disorders (Nippold, 2001, 2002). The findings of the Illinois Stuttering Research Program indeed indicate that near the time when children begin stuttering, they tend to be, on average, somewhat delayed in phonological skills. Further analyses, however, show that the total group mean masks internal subgroup differences.
Specifically, here too, there is a divergence along the future developmental path. From very near onset, those children who later persist were more phonologically delayed than the children who would later recover from stuttering (Paden & Yairi, 1996). The difference, however, is diminished markedly 1 year later due to a more rapid progress by the persistent group. Two years after onset, the phonological abilities of the two groups become quite similar (Paden, Ambrose, & Yairi, 2002; Paden, Yairi, & Ambrose, 1999). The stuttering–phonology connection thus appears to be limited to a small subgroup of the population of those who stutter. We further investigated the mutual relations between these two factors. The phonological skills were further compared in two subgroups with extreme ratings of stuttering (mild-severe). Similarly, the stuttering severities of two groups with extreme levels of good and poor phonological skills were compared (Gregg & Yairi, 2001). No statistically significant differences were found for either of the two factors. Thus, we concluded that the stuttering–phonology connection is not as widespread as it has been assumed, and the nature of the relations does not seem to be linear.

**Motor Speech Aspects**

Although research regarding motor speech functions in adults who stutter has yielded mixed results, the overall trend appears to indicate the existence of some aberrancies. In contrast, for young children who stutter, the few past research reports indicated only minute differences from children who are normally fluent (Conture, 1991). In this domain, three investigations of fluent speech of preschool children who stutter conducted in our project provided evidence that they too tend to exhibit some speech aberrancies even when they do not stutter. In a study of laryngeal functions, the preschool children who stutter near the onset of stuttering had higher shimmer and smaller jitter values in vocal fold vibration than their normally fluent peers (Hall & Yairi, 1992). A study of articulatory rate (i.e., the speed of speaking during fluent speech segments) showed that preschool-age children near onset of stuttering had a slower speaking rate than normally fluent controls (Hall, Amir, & Yairi, 1999). Interestingly, longitudinal tracking comparing children who eventually persisted in stuttering, those who eventually recovered naturally, and normally fluent peers, indicated that when articulation rate was measured in phones per second, children who later recovered had the slowest speaking rate near the time of onset. Third, a study of second formant (F2) transition rate (time and frequency dimensions) showed that close to stuttering onset, children whose stuttering eventually persisted demonstrated significantly smaller frequency change during the F2 transitions in fluent speech than those exhibited by children who later recovered and by their normally fluent peers (Subramanian, Yairi, & Amir, 2003).

**Genetics**

A major change regarding the etiology of stuttering has occurred during the past three decades, shifting from the idea that the disorder is acquired through learning back to even older notions that it is rooted in genetics. The notion that stuttering is genetically based stems from the commonplace observation that the disorder runs in families. Systematic scientific surveys that began in the 1930s showed that the chance of a person who stutters to have a familial history of stuttering is much higher than that of a person who does not stutter. A review by Yairi, Ambrose, and Cox (1996) of 28 reported studies concluded that the majority of them yielded familial stuttering in 30% to 60% of PWS as compared with less than 10% for families of normally fluent controls. Our own data (Ambrose, Yairi, & Cox, 1993) also indicate a high percentage of familial incidences: 71% of stuttering children had stuttering histories in their immediate or extended family, 43% in their immediate family, and 28% had at least one parent with such history. These surveys, however, failed to consider the family size of each participant. A second source that strengthened the idea of genetic components to stuttering were studies of twins that showed a much higher concordance for stuttering in monozygotic (identical) than in dizygotic twins (e.g., Andrews, Morris-Yates, Howie, & Martin, 1991; Howie, 1981). This information was not sufficient to reach valid conclusions regarding the role of genetic transmission and stuttering. However, data from familial aggregation research (e.g., Kidd, 1980, 1984) that considered family size and degree of the relatedness revealed that specific patterns of distribution of stuttering in families concurred with genetic models. Several genetic transmission models that have been offered suggest the involvement of polygenic factors (several or many genes) as well as environmental factors, but more recent work has also indicated a major gene, or genes (major locus), in addition to polygenic and environmental factors.

In our program, statistical methods such as segregation analysis were employed to study in detail the familial pedigrees to determine the most likely mode of transmission by matching the familial distributions of stuttering in our sample against several possible genetic models. Our team was the first ever to report statistically significant evidence for the presence of a Mendelian major locus component to stuttering (Ambrose, Yairi, & Cox, 1993), originally suggested by other investigators (e.g., Kidd, 1980, 1984). We concluded, however, that the evidence favored a mixed model combining both genetics and environmental factors as the best to explain stuttering. Next, our research focused on genetics in relation to the developmental course of stuttering, comparing the genetic bases of two subpopulations: children who exhibited natural recovery and those who developed persistent stuttering. We reported that, although the same major gene(s) appears to be involved in all children who stutter, there were also significant differences between the two subpopulations. The data that emerged for the chronic group indicated additional larger genetic components as compared to the naturally recovered group. Children who stutter and have a familial history of chronic stuttering would tend to follow that same pattern and also develop chronic stuttering, whereas children who stutter but have a familial history of recovered stuttering would tend to follow that pattern. Although these tendencies are not perfect, the findings provided one
of the best early predictors of the eventual course of the disorder. In short, it was possible to conclude that both the initial expression of stuttering and its developmental course have strong genetic bases.

These findings provided justification to pursue biological genetics research of stuttering, currently in progress in our program. The first step is a large linkage study aimed at identifying the general location(s) of possible specific genes. To this end, DNA is extracted from blood samples obtained from families with multiple occurrence of stuttering in the United States, Israel, and Sweden. In the next step, forms of known marker genes are identified on every chromosome. If a marker gene form is co-inherited with stuttering, the indication is that the gene contributing to stuttering is on the same chromosome. Dr. Nancy Cox (Cox, 2000), currently on our team, reported results of the first complete genome-wide screen of DNA markers for analysis of stuttering performed on the Hutterite population in North Dakota. In this groundbreaking study, she identified three chromosomes—#1, #13, and #17—as possible locations of genes underlying stuttering. Initial genotyping results from blood samples in our current linkage study are expected at about the time that this article is published. Hopefully, with time, knowledge obtained from biological genetics can be used to improve early diagnosis and prognosis of stuttering, as well as make possible more effective counseling and treatment. One must keep in mind, however, that the available evidence has consistently assigned significant roles to nonshared environmental factors. Curiously, it appears that it will probably be more difficult to identify specific environmental than genetic factors.

**CLINICAL IMPLICATIONS**

Our findings on the onset of stuttering suggest that clinicians should be alert to diversity in age, manner, stress, and other factors at onset. With additional research, these and other factors may provide greater differentiation among types of stuttering and their clinical course. Analyses of speech samples led me to conclude that from the time when stuttering is said to begin in preschool children, their disfluent speech is markedly different from that of normally fluent children. Current data provide sufficient information for clinicians to make reliable identification of early stuttering in the vast majority of cases.

Recent years have seen a heated discussion in the field concerning the question of whether or not all children who begin stuttering should receive some form of early intervention (e.g., Curlee & Yairi, 1997, 1998; Ingham & Cordes, 1998). In my view, not every child who begins stuttering needs to receive immediate intervention, and I do not believe that early intervention can prevent stuttering from emerging. Therapy, though, may hasten recovery for some children who have the potential for natural recovery. Considering genetic factors, some children may never recover but may still benefit from therapy. Our longitudinal research has provided a growing body of data that facilitate early prediction of risk chances for a child who begins stuttering to develop a chronic disorder or his or her chance for natural recovery. Although safe predictions are still impossible to make, the available information can be used to make a reasonable assessment of chances. Determination of risk is important, first, because those assessed to have the highest risk should be at the top of the list for immediate intervention. Establishing priority is often necessary due to limited financial and clinical resources. Second, inasmuch as possible, unnecessary treatment should be avoided both on ethical grounds and in view of possible negative effects.

Our multidimensional investigation of the onset and development of stuttering has resulted in several levels of factors that can be applied in making risk assessments, reflecting the degree of their current usefulness (Yairi & Ambrose, 2004). Those included in the first level are:

- **Disfluency patterns.** The absence or only little decrease in frequency of stuttering during the first 12 or so months after onset indicates that chances for future decline are diminished. The length (e.g., number of repetition units) and type of disfluencies are related factors. When length is not shortened during the same period, and when the percentage of prolongations and blocks does not decrease, risk for persistency is higher.
- **Duration of the stuttering problem.** Year by year, the longer the time from onset, the smaller the chance for recovery. For example, the chance for recovery is 75% at onset, 47% after 2 years, and only 16% after 3 years.
- **Family history.** A history of chronic stuttering among relatives presents a high risk of stuttering.
- **Gender.** Boys are at higher risk for chronic stuttering than girls.
- **Age at onset.** Late onset presents a risk for several reasons; among them, the growing awareness of stuttering on the part of the child and his or her peers.

Factor of lesser significance that are currently under investigation include head and neck movement, phonological skills, expressive language skills, and perhaps certain characteristics of second format transitions. No single factor is sufficient. Rather, the accumulating effect of several factors must be considered.

As far as treatment is concerned, although we have done some experimentation in this domain (e.g., Coppola & Yairi, 1982; Yairi & Ambrose, 2004), much more research has been done by our colleagues at other centers. Most of this work has shifted away from the traditional approach that focused on parent counseling. Instead, direct speech therapy with the child has been employed (e.g., Ingham & Riley, 1998; Kielstra-van der Schalk, Franken, & Boelens, 2003; Onslow, Packman, & Harrison, 2001; Ryan, 2001) with encouraging results.

**SUMMARY AND CONCLUSIONS**

Comparisons of current scientific knowledge pertaining to stuttering with what was available and acceptable 25 or 30
years ago reveal large advances in just about every major aspect of the problem, including etiology, prevalence, onset, early speech characteristics, differentiation, developmental course, motor, language, phonological skills, awareness, and more. Among the key points to remember is the fact that during the early years of life, the prevalence of stuttering is rather high, approximately 2.5%, presenting a significant problem that demands greater attention in the professional preparation of speech-language clinicians.

Within the wide range of transformations, conceptions of the onset of stuttering have shifted from being presented as a rather uniform event to that characterized by substantial diversity. With 85% of the risk appearing to be over by 3 1/2 years of age, it is considerably earlier than what has been thought, thus inviting speculations about links to phonology and language development.

In the domain of early stuttering symptomatology, there has been a considerable expansion in the recognized early repertoire of both overt and covert characteristics such as head and neck movement, awareness, and emotional reactions. This has significant implications. For example, whereas lack of awareness was an important basis for indirect therapy, the presence of awareness gives credence and support to more recent approaches that emphasize direct intervention. Generally, the greatly expanded symptomatology presents a much greater similarity to advanced forms of stuttering than what has been believed for many years. Although young children who stutter do not possess the cognitive and emotional abilities to react to, or handle, stuttering in ways that older children and adults do, the abnormality with which they deal appears to be generally analogous. Additionally, we have been able to narrow down the most relevant characteristics for the identification of stuttering in terms of both disfluency and acoustic features. Long-lasting notions of extensive overlap between stuttering and normal disfluency have been replaced by our knowledge of how to achieve a much sharper distinction.

In regards to developmental course, there has been a shift from the concept of a progressively ascending disorder to one where it is seen, for the most part, as progressively descending. Those who continue to stutter into adulthood constitute a small minority of the stuttering population. Additionally, from previous views of a homogeneous linear developmental path, there emerged a picture of subgroup differentiation. One of the most important results is the advancement of information leading to early prediction of high risk for persistent, chronic stuttering or the chance for natural recovery.

Theoretically, psychological and learning perspectives of the cause of stuttering, such as the famous idea that parental misdiagnosis of normal disfluency is the cause of stuttering, have been abandoned. In my view, stuttering is a unique disorder that is biologically based, not an exaggeration of normal behavior. Currently, the role of genetics has become a focal point to various ideas of complex etiology. Perhaps stuttering results from a relatively straightforward anatomic/physiologic abnormality, such as over-proliferation of synaptic connections and further failure of synaptic specialization to occur in some children who persist. Recent findings of brain imaging provide hope that eventually it will be possible to explain stuttering within the context of brain structure and function.

Although genetic factors are necessary, they may not be sufficient to cause stuttering. Complex environmental factors appear to be involved as well as speech, language, motor, and personality-temperament aspects. Whereas stuttering is a unique disorder, our view emphasizes its variants. Findings concerning a large factor of natural recovery, and thus the heterogeneity of this population, also have major implications to models of stuttering as well as to the conduct of all research with young children who stutter, especially studies concerning the effectiveness of treatment.

Finally, in the clinical arena, significant progress has been made in early diagnosis of, and prognosis for, early childhood stuttering. Concerning intervention, there has been a noticeable shift from indirect to direct treatment of young children who stutter with reported good success, and we have begun seeing more and better research on the effectiveness of treatment.

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