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# **PREDICTIVE FACTORS OF PERSISTENCE AND RECOVERY: PATHWAYS OF CHILDHOOD STUTTERING**

EHUD YAIRI, NICOLINE G. AMBROSE, ELAINE P. PADEN, and REBECCA N. THRONEBURG

This article presents broad preliminary findings from a longitudinal study of stuttering pertaining to differentiation of developmental paths of childhood stuttering, as well as possible early prediction of High Risk, Low Risk, and No Risk for chronic stuttering. More than 100 preschool children who stutter have been closely followed for several years from near the onset of stuttering using a multiple data collection system, with 45 nonstuttering children serving as controls. Thirty-two stuttering and 32 control subjects who have progressed through several stages of the investigation were identified for the present indepth analyses. They represent four subgroups: I. Persistent Stuttering; II. Late Recovery; III. Early Recovery; IV. Control. Comparative data for the groups with special reference to differences in frequency of disfluency, acoustic features, phonologic skills, language development, nonverbal skills, and genetics are presented. The results suggest several promising predictors of recovery and chronicity.

## **INTRODUCTION**

One of the most compelling reasons for thorough understanding of the pathognomonic course of stuttering is the potential to make subgroup differentiation, determine risk, state prognoses, and provide optimal treatment at each stage of the disorder. For nearly 60 years since Bryngelson's (1938) estimate that approximately 40% of young children who stutter outgrow the disorder, the clinical and research literature has suggested many estimates of spontaneous recovery. With the high end of this range at 79% (Andrews & Harris, 1964) and 89% (Yairi & Ambrose, 1992), reported by investigators who conducted longitudinal studies that are more reliable than the commonly reported retrospective methods, the theoretical and practical clinical importance of early prediction of the developmental paths of stuttering becomes apparent.

Indeed, differentiating between beginning stutterers who are at high risk of developing a chronic disorder and those who are likely to recover has become a central objective of investigators who study childhood stuttering (Yairi, 1990; 1993). Cooper and Cooper (1985), Curlee (1980), Riley (1981), and Van Riper (1971) listed various characteristics that should be taken as danger signs for stuttering chronicity. Gender differences have been an obvious target for consideration due to the substantial increase in the male-to-female ratio from about 2:1 near onset (Yairi & Ambrose, 1992b) to 4: or 5:1 in older children and adults (Bloodstein, 1995). Such changes may be attributed to higher recovery rates among girls, an assumption supported by recent findings of Yairi and Ambrose (1992a). The possibility that overt speech characteristics provide a predictive means has been explored both clinically and experimentally. According to Van Riper's (1971)

differentiation system, early symptomatology dominated by repetitions has a favorable prognosis for recovery but if blocks and prolongations dominate, chronic stuttering is likely. Couture (1990), Curlee (1993), and Riley (1981) also have commented that the presence of a substantial proportion of sound prolongations in the child's total disfluency is a danger sign of chronicity. Their impressions, however, were not verified by documented longitudinal data. Yairi and Ambrose's (1992a) longitudinal study of 27 preschool stuttering children found that changes in the developmental curves of Stuttering-Like Disfluencies (SLDs) reveal a divergence, in terms of frequency of occurrence, between persistent and recovering stutterers during the period of 14 to 18 months after onset.

Two investigations have focused on acoustic properties, particularly second formant dynamics, of young stutterers' disfluent utterances. Stromsta (1965) reported that stuttering children whose disfluencies contained abnormal formant transitions and terminations of phonation were more likely to exhibit stuttering 10 years later than were children whose stuttering did not evidence these characteristics. Missing or vague descriptions of subject selection, measurements, and follow-up procedures, raise questions about the strength of his findings. Recently, Yaruss and Couture (1993) assessed the prognostic power of formant transitions by comparing patterns in high-risk and low-risk groups of young stutterers who were so identified by the Stuttering Prediction Instrument (Riley, 1981). They reported negative findings; however, their classification of subjects and conclusions were not verified by longitudinal observations of the children.

The relations between stuttering and articulation/phonology, language disorders, non-verbal skills and intelligence factors, has been discussed in past (Andrews & Harris, 1964; Berry, 1938) as well as more recent literature (Louko, Edwards, & Couture, 1990; Nippold, 1990; Throneburg, Yairi, & Paden, 1994; Wall, Starkweather, & Cairns, 1981). Similarly, there has been growing recognition of the role of genetics in stuttering (Kidd, 1980; Kidd, 1984). Unfortunately, however, none of these factors has been investigated in relation to its potential value in differentiating risk subgroups of individuals who stutter. Thus, early prediction of chronic stuttering has remained largely in the realm of general clinical impression and speculation. Relevant scientific data are sparse and often are not based on valid verification through longitudinal follow-ups, the most accurate scientific method for the study of the developmental course of any disorder.

For the past six years, the Stuttering Research Program at the University of Illinois has been conducting its second longitudinal investigation of early childhood stuttering. This large-scale study has been sponsored by the National Institutes of Health. The overall objective of the investigation has been to follow a substantial number of children from close to the onset of stuttering through a period of several years. Through systematic periodic observation, recording, and testing, variations in stuttering symptomatology over time can be objectively tracked and subgroups of children who recover and those

whose stuttering becomes chronic can be prospectively identified. Once such subgroups are identified, it should be possible, through analyses of data obtained at early testing periods, to isolate factors that could distinguish the groups at that time. One of the main objectives of this project has been to identify factors that will predict persistence and recovery in stuttering. Specifically, we set out to explore whether the parameters of frequency of disfluency, acoustic characteristics of disfluency, secondary head and facial movement, phonological and language skills, nonverbal skills, and genetics can be used for this purpose. As the study will continue actively for several years, this report presents our preliminary findings. Because of the diverse nature of our investigation and the large amount of data for each of the listed parameters, we deemed it useful to present here an overview of the current results, emphasizing general trends.

## **METHOD**

### **Subjects**

A large, unbiased pool of subjects was required. A multiple-criteria scheme for admitting stuttering children into the study was employed: (a) under six years of age, (b) regarded by parents as having a stuttering problem, (c) regarded by two investigators (Yairi and Ambrose) as having a stuttering problem, (d) stuttering history of no longer than 12 months, (e) stuttering severity rated by parents as at least 2 on an 8-point scale: 0 = normal; 1 = borderline; 2 = mild; 7 = very severe, (f) severity rating of 2 or higher assigned by the investigators, (g) exhibit at least 3 Stuttering-Like Disfluencies (SLDs) per 100 syllables, (h) no obvious neurologic disorders or abnormalities. Because of previous indications that significant changes in stuttering, including complete recovery, occur during the first few months of the disorder (Andrews & Harris, 1964; Yairi, Ambrose, & Niermann, 1993), an all-out effort was made to secure subjects close to onset.

Criteria for recovery were as follows: (a) clinician general judgment that the child did not exhibit a stuttering problem, (b) parental general judgment that the child did not exhibit a stuttering problem, (c) parent rating of stuttering severity as less than 1, (d) clinician rating of stuttering severity as less than 1, and (e) SLD frequency of fewer than 3 per 100 syllables.

The nonstuttering control subjects were recruited primarily from daycare centers. They were (a) regarded by parents as not having a history of stuttering, (b) regarded by the investigators as not exhibiting stuttering, and (c) not regarded as having histories of neurologic disorders or abnormalities.

Currently, the project includes more than 100 preschool-age children who have stuttered (Experimental Group) and 45 normally-fluent children (Control Group). The two groups present similar gender and age distributions. Although many of the children are still progressing through the investigation, we have identified a number of subjects

whose problems have taken different paths over the period of the study, leading to the formation of three groups. They have been classified as follows:

- I. Persistent Stutterers: Continued stuttering for 36 months or more after stuttering onset (**n** = 12)
- II. Late Recovered: Recovered between 18 and 36 months post onset (**n** = 10)
- III. Early Recovered: Recovered within 18 months post onset (**n** = 10).

The formation of two recovered groups was based on documentation that a number of children recover very quickly (Yairi, Ambrose, & Niermann, 1993; Andrews & Harris, 1964), while others do not recover until considerably later. Furthermore, without such grouping, data for those who already recovered would be combined with that for children still stuttering, which could confound interpretation. The subgrouping may eventually address the possible existence of differences among children who recover.

Ten of the 12 subjects in the Persistent group received fluency therapy for various time periods. The parents of the remaining two subjects chose not to enroll their children in treatment. None of the subjects in the Recovered groups received any therapy for fluency.

Three control groups, one for each of the stuttering groups, were formed from the nonstuttering control subject pool by matching each of the children in the three experimental **groups** with a nonstuttering subject of the same gender and similar age in months. Thirty of the resulting 32 pairs differed in age by no more than one month; the remaining two pairs differed by 2 and 3 months and were among the oldest pairs. Because speech/ language development is rapid between ages 2 and 4 years, the age-range of most subjects at entry to the study, matching according to the factor age as well as gender insured that any differences in language skills between stuttering and control groups were not accounted for by these two variables.

### **General Plan of Study**

Each child received a comprehensive initial evaluation that generated case history and familial pedigree information. Additionally, a 30- to 40-minute conversational speech sample was audio- and video-recorded over two days approximately one week apart, and a large battery of speech, language, hearing, motor, psychological, and other tests was administered. Among others, the Assessment of Phonological Processes-Revised (APP-R) (Hodson, 1986) and the Preschool Language Scale (PLS) (Zimmerman et al., 1979) were administered at entry into the study and one year later. Measures of number of different words and mean length of utterance (MLU) were obtained from the recorded samples of conversation taken at these same time periods. In addition, the Arthur adaptation of the Leiter International Performance Scale (Arthur, 1952) was given at entry to the study. Follow-up testing and recording have been conducted every six months for all the subjects who stutter and every year for the control subjects.

The large body of data collected in these test sessions lends itself to several types of analysis and produces findings on a number of different parameters. This preliminary report will present general findings from separate but related projects that involved the same subjects. More comprehensive reports of each of these projects will be forthcoming which together will provide broad, detailed descriptions of those subjects. Any background or procedures specific for a given study will be included under the report of that study. Because the intent of this report is to provide a broad overview, we will emphasize general trends and will defer more definite conclusions and detailed statistics to upcoming final reports. Thus, significant statistical differences are mentioned with little elaboration. For all statistical analyses, alpha was set at .05. In all cases, standard deviations allowed for meaningful results but are not reported here.

## **SELECTED PARAMETERS: BACKGROUND AND RESULTS**

### **Ages of Subject Groups**

Unlike studies in which subjects are assigned to groups before the experiment or observation begins, a unique feature of the present investigation was the fact that subject classification into groups was possible only after many months of observation and testing. In other words, subject classification was a basic finding permitting all other comparisons.

Table 1 displays information concerning the distribution and mean ages of the subjects when they were first tested and their ages at the time of onset of stuttering. As can be seen, at the outset there were significant age differences in the groups. Children in the Persistent group were older by almost 11 months when they entered the study than were children in the two Recovered groups. More important, the Persistent subjects began stuttering five to eight months later than those who recovered. Analysis of variance showed that this difference was statistically significant. Thus, a substantial part of the age difference at entry can be attributed to later stuttering onsets. The mean interval between stuttering onset and the initial evaluation of the Persistent group was 8.58 months. Both Recovered groups had statistically significantly shorter intervals (2 to 5 months) between onset and first interview.

### **Disfluency**

Recorded speech samples of 1,000 words per visit were subjected to careful, double-checked analyses to identify and classify disfluencies. Seven disfluency types were identified and grouped as SLDs (part-word repetition, monosyllabic word repetition, disrhythmic phonation) or Other Disfluencies (interjection, revision-incomplete phrase, multisyllabic word repetition, phrase repetition). Average interjudge, point-by-point agreement for disfluency identification has already been reported at .84 (Ambrose & Yairi, 1995; Yairi, Ambrose, & Niermann, 1993).

Group	Mean Age in Months (SD)	Range in Months	Onset Age (SD)	Months Post-Onset (SD)
Persistent, <i>n</i> = 1	47.33 (8.91)	30–65	38.67 (7.49)	8.58 (2.68)
Control for Persistent, <i>n</i> = 12	46.83 (8.24)	31–63		
Late Recovered, <i>n</i> = 10	36.60 (5.91)	28–49	30.20 (5.55)	6.40 (3.72)
Control for Late Recovered, <i>n</i> = 10	36.40 (5.44)	29–48		
Early Recovered, <i>n</i> = 10	36.70 (7.30)	27–48	33.60 (6.24)	3.10 (2.03)
Control for Early Recovered, <i>n</i> = 10	35.75 (6.74)	27–47		

Table 1. Mean Age and Standard Deviation (in Parentheses) for Experimental and Control Subjects at Entry into the Study, and Age at Onset and Months Post Onset of Stuttering for Experimental Subjects

The longitudinal trends for the three Experimental and the combined Control groups are shown in Figure 1 which displays the frequency of SLDs per 100 syllables across 4 test periods. We make four observations. First, contrary to our expectations, the initial level of SLDs is appreciably higher in the two Recovered groups than in the Persistent group. Second, the Persistent group is relatively stable. Third, while there is little change over time in the Persistent group, there is a slow drop of SLDs in the Late Recovered group, but a quicker decline in the Early Recovered group. Fourth, and most important, the data indicate divergence between the developmental course of the Persistent and the Recovered groups as early as 7 to 12 months after stuttering onset. Analysis of variance indicated a statistically significant difference between the Persistent and each Recovered group at this time interval. While Group I (Persistent Stuttering) subjects maintained consistent levels of SLDs, those who recovered exhibited sharp declines in SLDs no later than 7 to 12 months post-onset and continued to gradually decline to normal levels thereafter. The important conclusion is that a child who has stuttered for more than 12 months has an increasing chance of continuing stuttering although some spontaneous recovery continues to occur. At this preliminary stage of the investigation, keeping in mind the objective of identifying a group of markers indicating possible chronic stuttering, these clear trends in the results should be viewed as important leads that may have important clinical implications.

The analysis of group means for Other Disfluencies indicated that they remained fairly stable over time for all groups. These data provide little helpful developmental information and, therefore, will not be reported here.

### **Secondary Physical Characteristics**

The literature on stuttering reflected for many years the assumption that secondary characteristics are a late developing phenomenon (Bluemel, 1932; Froeschels, 1921; Van Riper, 1971). More recently, however, there has been a growing indication that such physical components of stuttering may be present early on (Conture & Kelly, 1991; Yairi, Ambrose, & Niermann, 1993). Interestingly, Cooper and Cooper (1985), Curlee (1980), and Van Riper (1971) considered the presence of secondary characteristics as a prognostic sign for chronicity. This line of research has been continued in group comparisons for this study.

Ten instances of SLDs were selected from recordings of each of two speech samples of each subject, one taken at 7 to 12 months post stuttering onset and the second sample at 13 to 18 months post onset. These instances of disfluency were selected by dividing the sample into 10 approximately equal segments and choosing the perceptually longest SLD instance in each segment. As children recovered and disfluencies were few, the majority of the SLDs produced were analyzed. A simplified version of the Facial Action Coding System (Ekman & Friesen, 1978) was used, and the 19 of 44 head and facial movements (Yairi, Ambrose, & Niermann, 1993) implemented are listed in the Appendix. Each of the 10 disfluent events per subject per sample was viewed several times using a frame-by-frame progression. The first viewing determined the initiation and termination points of each disfluency. The second, and sometimes third and fourth viewings, tallied the number of head and facial movements that occurred. For each disfluent event, a fluent segment of the same time length was identified in the sample to control for extraneous movements that 2- to 4-year-old children normally make when talking. Fluent segments were chosen that were similar in grammatical function and sentence position. Often two to three words were spoken in equal amounts of time. The number of facial and head movements was tallied in the time-matched fluent segment and then subtracted from the total number of actions identified in the disfluent segment, resulting in a net score. The mean of the net scores was calculated for the 10 disfluent events selected from the speech sample taken at each of the two test periods.

Figure 2 presents group means for the net number of movements observed during disfluent events. Means were less than one movement per disfluency for all groups at both visits and no statistically significant differences were found. At the 6 to 12 months post-onset visit, the three groups of children had similar numbers of movements, 0.6, 0.7, and 0.7. By the 13 to 18 months post-onset visit, the number of movements decreased slightly for the Early Recovered group, remained about the same for the Late Recovered group, and increased slightly for the Persistent group. Although these data appear to evidence a time-related trend toward group differences, the discrepancy at



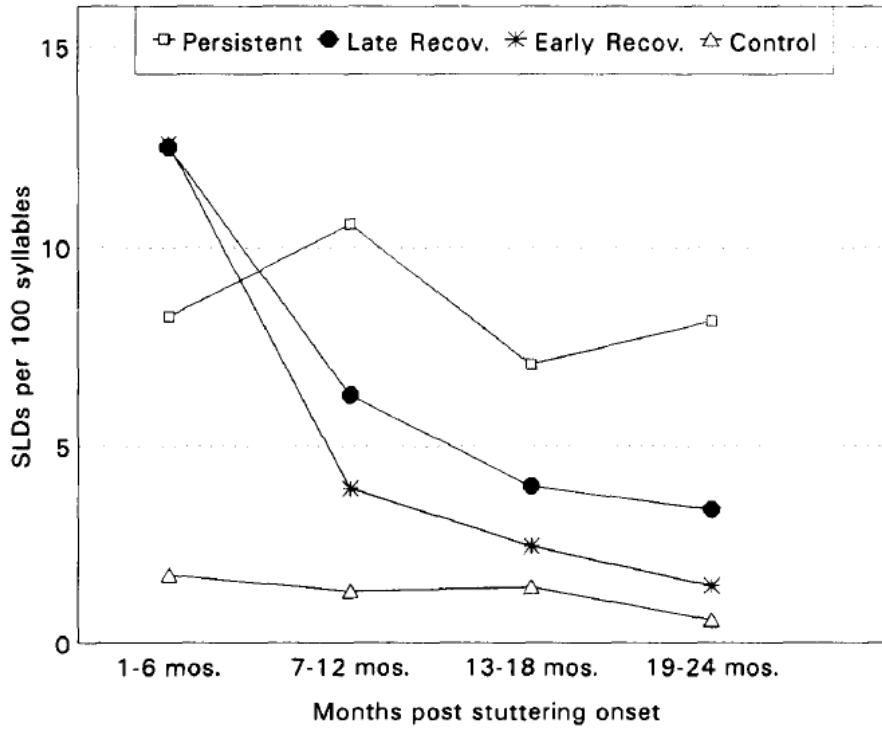


Figure 1. Longitudinal variations in group means of frequency of Stuttering-Like Disfluencies for Persistent, Late Recovered, Early Recovered, and Control subjects.

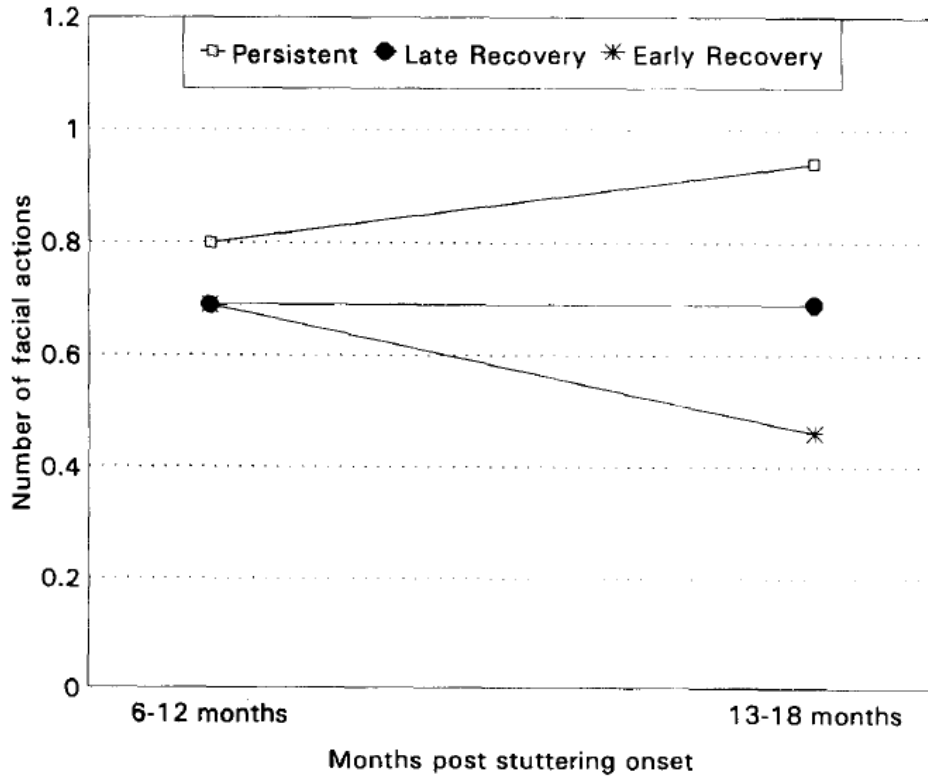


Figure 2. Longitudinal variations in group means for the net number of head and facial movements for Persistent, Late Recovered, and Early Recovered subjects.

the 13- to 18-months post-onset visit is only one-half movement per disfluency and analysis of variance indicated that the difference was not statistically significant. Secondary physical characteristics do not, therefore, appear to contribute to predictability of persistent stuttering.

### **Durational Characteristics**

A recent series of studies by Yairi and Hall (1993), and Throneburg and Yairi (1994) reported differences between preschoolers who were beginning to stutter and normally-fluent children in the duration of repetitive units of part-words and monosyllabic words. For example, if the child repeated "that that," the length of the first and the second production of the word was measured as well as the silent pause between. These two studies reported that the length of the spoken portions of the disfluency was similar for normally-fluent and stuttering children. More important, Yairi and Hall (1993) reported a tendency toward, and Throneburg and Yairi (1994) reported significant group differences in, the length of the pause between the spoken units of disfluencies. The silent interval for the stuttering children was approximately 500µs shorter than that for the normally-fluent children and differentiated the two groups with approximately 80% chance of accuracy.

The same technique was used in the present study to obtain preliminary data for six children who stuttered at three consecutive visits. Three children were from the Persistent group and three from the Recovered groups. Transcripts of these subjects' speech samples were used to locate instances of single-unit part-word repetitions and single-unit monosyllabic wholeword repetitions. Disfluent episodes that contained only single unit part or monosyllabic whole-word repetitions<sup>2</sup> and were clear of interfering noise were low-pass filtered at 7.5 kHz, digitized at 20,000 samples per second (DT 2821 A/D convertor) and acoustic measurements were made from an FFT-based spectrogram display using the CSpeech analysis computer program (Milenkovic, 1987). Cursors were placed at the initiation and termination of spectral energy to measure the length of each repetition unit and the silent interval between them. One-hundred and two part-word repetitions and 91 whole-word repetitions were measured for the Persistent group, and 114 single-unit part-word repetitions and 106 whole-word repetitions were measured for the Recovered groups. Pearson correlation for intra judge agreement based on remeasurement of 100 segments was .993. The data indicated that the length of the spoken portions of the disfluencies were similar for both groups of children. Changes in duration of the pause from the first through the third visit differentiated the two groups. Because there were only 3 subjects for each of the two groups, statistical analysis is deferred to a more comprehensive forthcoming report.

For single-unit monosyllabic word repetitions, shown in Figure 3, pause times at the first visit for the Persistent group were slightly shorter than that for the Recovered group. As the children in the Recovered group started to improve, however, the duration of their pauses increased markedly. By the third (one year) visit, the Persistent group continued

to evidence the same short pause time between spoken units, while the pause time for the Recovered group had increased even more.

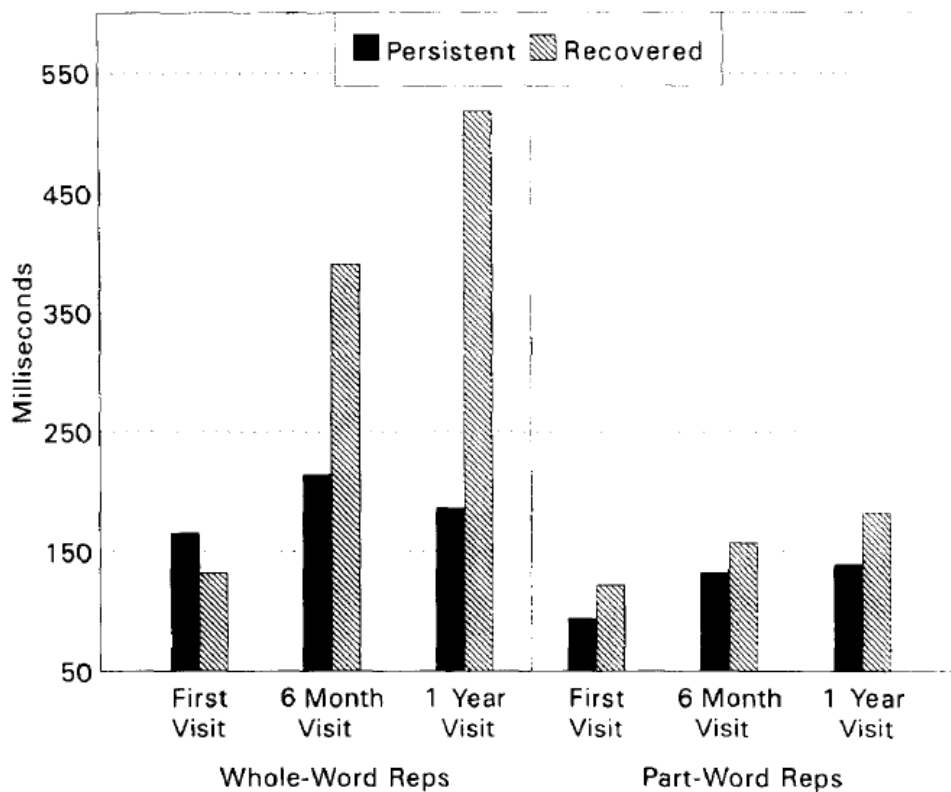


Figure 3. Longitudinal variations in the duration (msec) of silent intervals between units of whole-word and part-word repetitions for Persistent and Recovered subjects.

A similar data pattern was found for single-unit part-word repetitions. The Persistent group had shorter pause times between the repetition units at all three visits than did the Recovered children. Although the pause time for the Persistent group remained relatively static, it increased over the three visits for the Recovered group. Pause times for part-word repetitions differ, however, in that they are shorter overall in duration and the increase in duration over time was less dramatic.

Previously reported comparisons of children who stutter with normally fluent children (Throneburg & Yairi, 1994) indicated that a longer interval was typical of disfluencies of normally fluent children. Thus, increasingly longer intervals appear to be characteristic of stuttering children as they recover from stuttering and their repetitions become normalized. The pause times for whole word repetitions of recovered children reach normal levels (494 ms reported by Throneburg & Yairi, 1994). While the trend of increasing pause times is similar for part-word repetitions, they have not yet reached durations comparable to those of normally fluent children (418 ms).

### Language and Non-Verbal Skills

Data for the APP-R, PLS, MLU, number of different words and Leiter were analyzed together using multivariate analysis of variance. All results except number of different words and MLU reached statistical significance.

**Phonology.** Many investigators, beginning as early as McDowell (1928), have offered evidence that children who stutter have more articulation errors or phonological impairments than normally-fluent children. (See Nippold, 1990, for a review.) Estimates of the proportion of children who stutter and who also have phonological problems have ranged widely, however, from 160Jo (Blood & Seider, 1981) to 720Jo (Williams & Silverman, 1968). As Nippold pointed out, because these surveys differed considerably in the methods used to collect data, in subjects' ages, and in criteria defining an articulation or phonological problem, it is virtually impossible to draw an overall conclusion from them. Moreover, each of these studies was based on one-time assessments, so that changes over time were not reported.

Our research project evaluates children a short time after their stuttering is first identified and reassesses them over a period of several years. Thus, it can observe not only the extent of any early phonological impairment, but also the course of a child's phonological development in the presence of fluency problems.

The APP-R is scored by checking each time a targeted phonological pattern is absent in the child's naming response. The percentage of opportunities on which a child failed to produce each of 10 basic patterns is determined, and the mean of these 10 scores is computed. Thus, lower scores represent better phonological acquisition (i.e., fewer inadequacies).

Using the severity categories specified by the APP-R, the levels of phonological deficiency exhibited by each subject group can be seen in Table 2. The first notable observation is that none of these subjects evidenced Profound phonological deficiency (scores of 60 or higher) even at first visit. In fact, only 3 of the 32 stuttering subjects had scores within the Severe range (1 Persistent; 2 Late Recovered), as did 2 control subjects. Another observation is that most Persistent group subjects were in the Moderate category at first visit, whereas each of the Recovered groups had equal numbers in the Moderate and Mild levels. Because the Persistent subjects were older, it appears that their phonological skills are poorer, suggesting a characteristic that might contribute to early prediction of stuttering chronicity. One year later, however, the Persistent subjects, along with those in all of the other groups, had moved out of the Severe category, and most were within the Mild range, a level that does not signal need for phonological intervention.

Mean APP-R scores for all subject groups at both testings are displayed in Figure 4. On the initial test, the mean score of the Persistent group is only slightly higher (worse) than that of the Late Recovered group. Remembering, however, that the Persistent group is about 11 months older, had their phonological development been at the same

Group	First Test			1 Year Later		
	Severe (40-59)	Moderate (20-39)	Mild (<20)	Severe (40-59)	Moderate (20-39)	Mild (<20)
Persistent <sup>a</sup>	1	8	3	0	1	11
Control	0	4	8	0	0	12
Late Recovered	2	4	4	0	2	8
Control	1	4	5	0	3	7
Early Recovered	0	5	5	0	2	8
Control	1	3	6	0	0	10

<sup>a</sup> Persistent subjects are approximately 1 year older than the Late Recovered and Early Recovered groups.

Table 2. Number of Subjects at Each Level of Severity of Phonological Deficiency (Based on APP-R Scores) within Experimental and Control Groups at Entry into the Study and One Year Later.

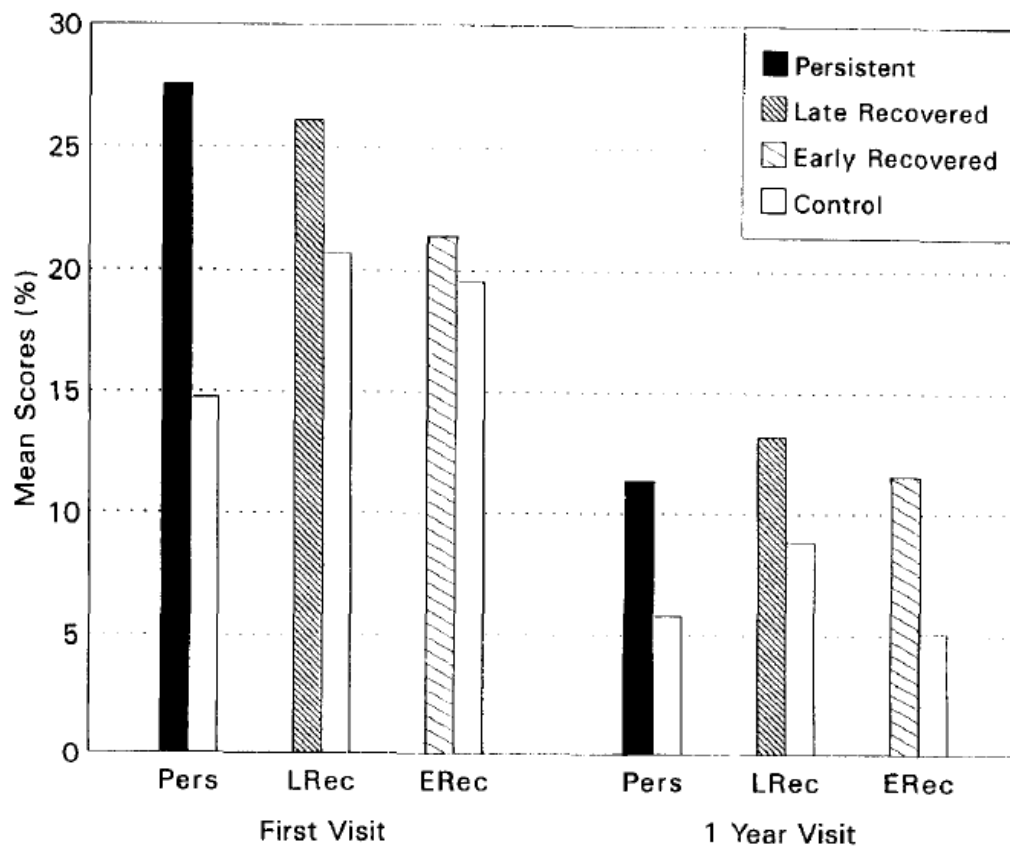


Figure 4. Differences in mean phonological error scores (from APP-R) between each Experimental group (Persistent, Late Recovered, and Early Recovered) and its nonstuttering Control group at the first and the 1-year visits.

pace as that of the Recovered groups, their scores should have been appreciably lower (better) than those of the other two groups. This assumption is further supported by the score of the Persistent Control group, which is indeed better than those of the two younger Recovered Control groups, as would be expected. Analysis of variance indicated that the difference between the Persistent group and its nonstuttering Control group at the first visit was statistically significant.

One year later, all of the stuttering groups had improved, and their mean scores are quite close. Once again, however, the Persistent group's score does not reflect their older age, when even better phonological skills would be expected. The higher phonological error scores at first testing, therefore, may contribute to early prediction of persistent stuttering.

It must also be reported that there were large individual differences within all groups. Not only was there a wide range of scores within each group (e.g., the range of scores for Persistent subjects was over 35 percentage points; Late Recovered over 40 points), there was also considerable overlap in the range of scores among groups. Thus, while mean scores of the groups suggest trends, it is clear that APP-R scores alone cannot predict whether stuttering in an individual child will be persistent. Perhaps more thorough analyses of these subjects' phonological performances in terms of types of errors and/or strategies relied upon may reveal more clear-cut differentiation between the groups. Our research continues in these areas.

**Preschool Language Scale.** To document language development, the Preschool Language Scale (Zimmerman et al., 1979) was administered at entry into the study and one year later. The test provides a general screening of receptive and expressive language development for children ages 2 to 7 years. For the Preschool Language Scale, results for the Late and Early Recovered groups were virtually identical and so the data for these two groups were combined, as were those for their corresponding Control groups, resulting in a total of four groups: Persistent, Persistent-Control, Recovered, Recovered-Control. Statistical tests yielded the same results with the recovered subjects treated as one or two groups. Figure 5 shows mean scores of the four groups for language comprehension and verbal expression one year apart. For both portions of the test, a score of 100 is considered average. As can be seen, all group means are above average although several individuals' scores were in the 80's. Mean scores for the Persistent group, however, are consistently lower on both language measures than those of the other three groups.

All comprehension scores drop similarly between the first and second testing. As shown by post-hoc testing, differences between the Persistent group and the Recovered group were statistically significant at both time periods, but the Persistent group's comprehension scores were only slightly lower than those of their control group. On the verbal expression portion of the test, scores for the Persistent group are again lower

than those of the other groups for both testing periods. Scores of the Persistent group were substantially lower than those of their control group and of the Recovered group, and post-hoc testing indicated that these differences were statistically significant.

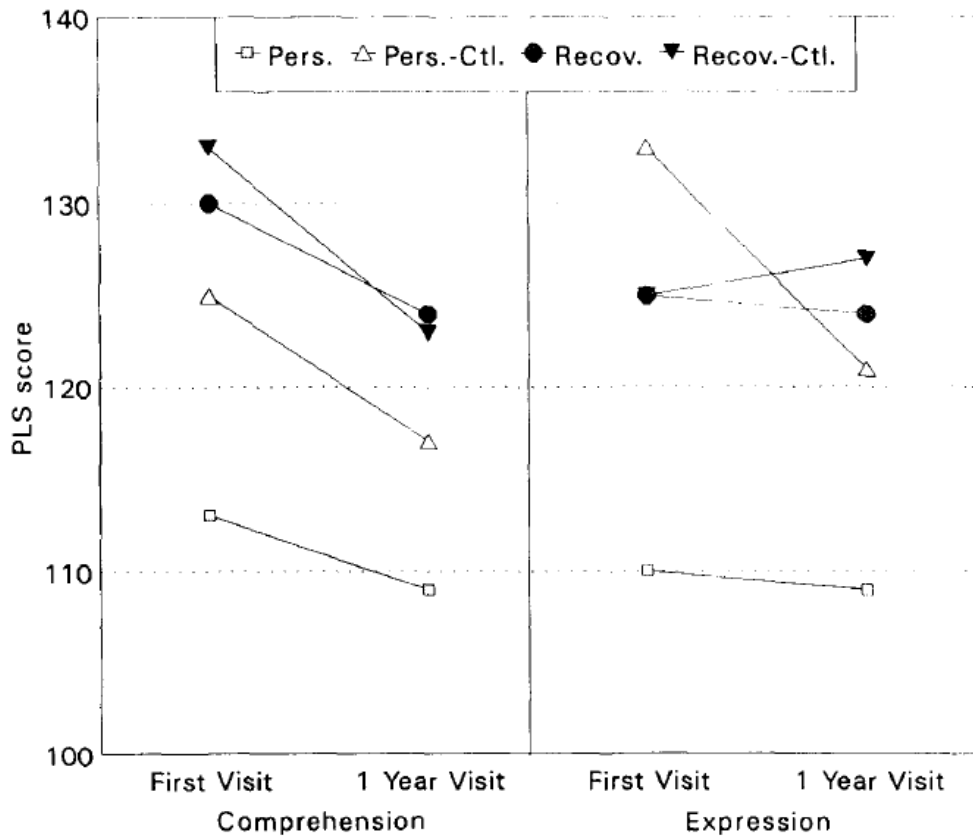


Figure 5. Differences from first visit to 1-year visit in mean PLS (comprehension and expression) scores for Persistent, Persistent-Control, Recovered, and Recovered Control groups.

**Mean Length of Utterance (MLU) and Number of Different Words.** Using SALT (Miller & Chapman, 1990), the number of different words and MLU were calculated from the speech samples of the subjects taken at entry to the study and one year later. The same samples had provided data for disfluency counts. Again, data for the Late and Early Recovered groups and their controls were combined because their separation added no new information. In contrast to the tests just discussed, there were no statistically significant differences between the Persistent group and any other group on these measures. The right-hand segment of Figure 6 shows the mean number of different words for the four groups. Each of the two stuttering groups had scores that were close to those of its control group. The scores for the Persistent group and its Control group were consistently higher than those of the Recovered group and its Control group for both measures.

This was expected because of their older age. When scores were adjusted for age, no differences were apparent. The pattern for MLU is fairly similar, as shown in the left

segment of Figure 6. As expected, the Persistent group and its Control group had higher scores than the Recovered group and its Control group at the first visit, due to the age difference, while at the one year visit, the Persistent group had the highest MLU. None of these differences were statistically significant.

**Leiter.** To determine if children were performing at comparable levels in terms of language skills and nonverbal skills, the Arthur Adaptation of the Leiter (Arthur, 1952) was administered to assess nonverbal skills at entry to the study. The Leiter involves matching blocks to pattern strips and has no verbal instructions or time limits. Because of similarity in scores, data for the two recovered groups were again combined. Mean scores for the four groups are presented in Figure 7. Scores for all four groups are above the normal mean of 100. Scores of 85 to 115 are considered to be normal limits for this test. The Persistent group has a mean considerably lower than that of the other three groups, but post-hoc testing indicated that only the difference between the Persistent group and its Control group was statistically significant.

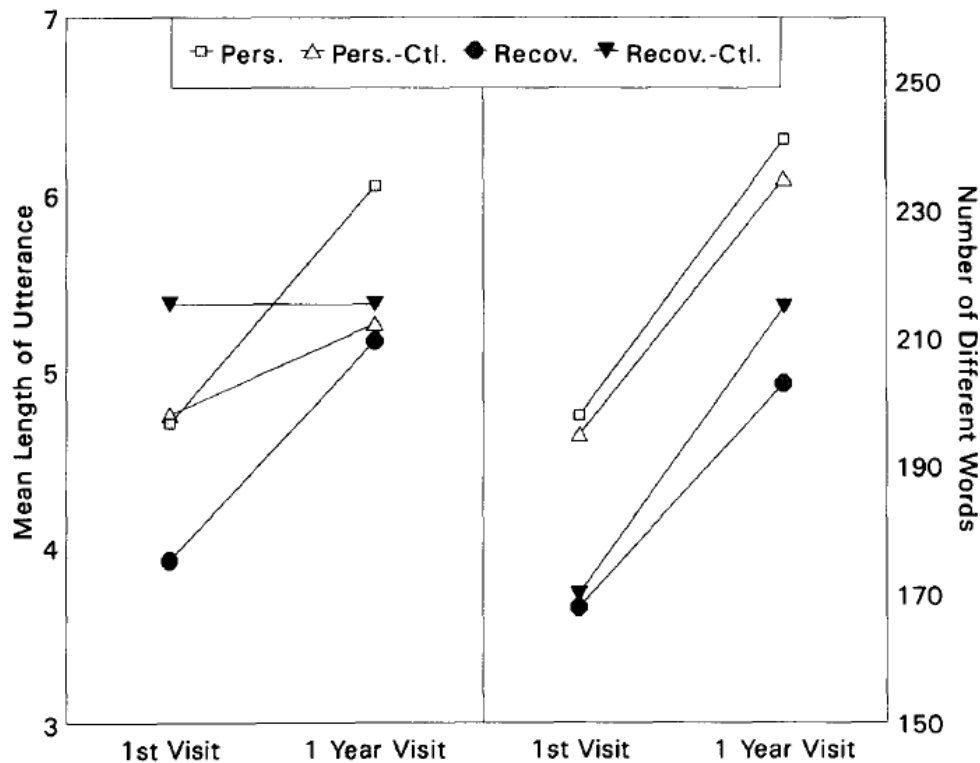


Figure 6. Differences from first visit to 1-year visit in MLU and Number of Different Words for Persistent, Persistent-Control, Recovered, and Recovered-Control groups.

**Prediction.** Information obtained on measures of phonology (APP-R), language (PLS) and nonverbal skills (Leiter) were examined for their predictive ability. Data concerning MLU and number of different words were not included. A discriminant analysis was used to determine the ability of combinations of variables to predict chronicity vs. recovery.



Results are presented in Table 3. Prediction of chronicity using these variables correctly identified 10 of 12 Persistent subjects, with the PLS contributing the most weight. Two were misclassified as Recovered, and only four of the 20 Recovered were misidentified as Persistent. Even though each of these tests indicated group performance within normal limits, and any single test score by itself may not indicate recovery status, the combination of these measures offers predictive information. It seems clear that there is some relation between stuttering chronicity and language capabilities, although it cannot yet be clearly defined.

PLS-COMP(2)	.643
PLS-VERBAL(2)	.516
PLS-VERBAL(1)	.509
PLS-COMP(1)	.459
APP(1)	-.437
LEITER	.379
APP(2)	-.064

Table 3. Canonical Loadings from Discriminant Analysis in Order of Magnitude.

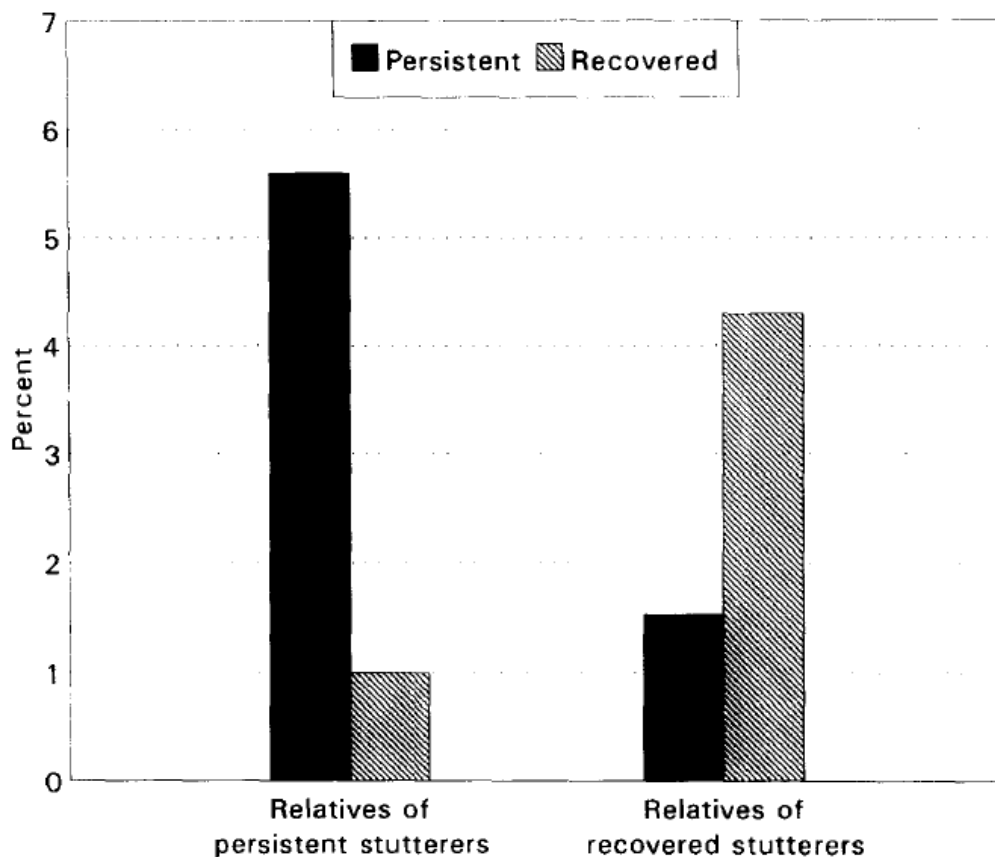


Figure 8. Percents of persistent and recovered stuttering relatives of children in the Persistent and Recovered groups.

## Genetic Factors

It is well known that stuttering tends to run in families and that the incidence of stuttering is higher among males. These observations point to a genetic factor. Research by Howie (1981) and Kidd and associates at Yale (Kidd, 1980; Kidd, 1984; Kidd, Heimbuch, & Records, 1981) added much knowledge about the role of genetics in the transmission of susceptibility to stuttering. The Yale studies' findings were consistent with a polygenic multifactorial model of inheritance with a possible major locus, and with males having a higher susceptibility. Using segregation analysis, Cox, Kramer, and Kidd (1984) found similar results. A recent study (Ambrose, Yairi, & Cox, 1993), found a higher risk of stuttering for males, but whereas Kidd's data showed the highest risk for male relatives of females who stutter, our data indicated that the highest risk is for male relatives of males who stutter. This difference may reflect the different population samples used in the two studies. Two studies using data collected at Yale dealt directly with recovery from stuttering: Cox and Kidd (1983) and Seider, Gladstien, and Kidd (1983). The first indicated that recovery is not a milder subtype of stuttering and the second that recovered and persistent stuttering are not two independent disorders. It is important to note, however, that all of the Yale data were obtained from adults who persisted in stuttering. Because our data do not have this ascertainment bias they may provide more valid information about a possible genetic role in the recovery from or persistence in stuttering.

We applied segregation analysis to our pedigree data and concluded that inheritance of stuttering is best explained with a major locus model. We have also begun relating pedigree data from the extended family to explore possible differences in incidence and in development trends in stuttering among the relatives of children who persist in stuttering and those who recover. The questions are: (a) is recovery or persistence heritable and (b) if so, is it transmitted as a milder form of stuttering or independent of stuttering?

To address these questions, we obtained pedigrees from the 12 persistent and 20 recovered subjects. Complete pedigrees are of the utmost importance in order to account for differing family sizes. Information on grandparents, aunts, uncles, cousins, parents and siblings of subjects was included. Relatives were categorized as having persistent stuttering, as having recovered from stuttering in childhood, or as having never stuttered (unaffected). Careful in-depth repeated interviewing provided a considerable degree of certainty regarding the recovery status of relatives, but further documentation is currently under way. Preliminary results indicate a clear pattern, as is shown in Figure 8. Ever-stuttered relatives of children with persistent stuttering include many more who have persisted in stuttering than who have recovered from stuttering. Children who recovered from stuttering, however, show the reverse pattern; of their relatives who have ever stuttered, many more recovered than became persistent. A chi-square test of independence indicated that this difference is statistically significant,

which means that persistence in stuttering tends to run in families.

## **DISCUSSION**

A major focus of the Stuttering Research Program at the University of Illinois has been to identify means for early prediction of the course of stuttering. The findings presented here indicate that language indexes, nonverbal performance, phonological skills, genetics and disfluency characteristics may all contribute to prediction of persistent (chronic) stuttering, emphasizing the multidimensionality of the disorder. They also suggest that a single impairment may affect several speech as well as nonspeech functions. Each of these parameters may yield important information that together may predict the outcome of childhood stuttering. One intriguing observation is that the earliest predictors of chronicity may not include disfluency, the cardinal characteristic of stuttering, but other such parameters as language skills and genetics.

### **Age**

The later onset of stuttering for children in the Persistent group also poses interesting questions. Their later onset may be due, in part, to the fact that this group is composed primarily of boys. Our previous data (Yairi & Ambrose, 1992b) show that boys begin stuttering, on average, 5 months later than do girls. It is possible that one predictive factor is the combined effect of gender and age at onset. A later age of onset, in turn, may also be related to slower language/phonologic development. Furthermore, the question of treatment for children who begin stuttering later is more important as they approach school age. A preschool child who stutters severely may experience little social repercussions, while a child in kindergarten or first grade may have a quite different experience. When there is less time between stuttering onset and school age, treatment decisions may need to be made sooner than with a younger child.

### **Disfluencies**

Contrary to previous assumptions (Riley, 1981; Van Riper, 1971), the initial frequency of SLDs did not seem to be a predictor of chronicity. In fact, the initial level of SLDs in the Recovered groups was higher than that of the Persistent group. Disfluencies may become more reliable predictors after 7 to 12 months post-onset. This time period is earlier than that presented in a previous report (Yairi & Ambrose, 1992a). Conclusions regarding the most appropriate time period to watch for level of disfluencies as an indicator of chronicity or recovery must be guarded at this point and will be clearer at the conclusion of this study. With additional data, it may be possible to identify the percent risk for chronic stuttering at various points in the child's course of the disorder. Considering the high rate of recovery during the early months of stuttering (Yairi, Ambrose, & Niermann, 1993) which continues until at least 15 months post-onset, there are growing indications that this point in the course of stuttering should be further investigated as the period when decision-making concerning intervention becomes

important.

### **Secondary Physical Characteristics**

Although secondary physical characteristics traditionally have been viewed as later-developing symptoms and as reactions to stuttering, their early appearance now seems unrelated to the future course of stuttering. The small between-group differences during early months of stuttering indicate that the frequency of head and facial movements do not provide a useful means of predicting stuttering chronicity. Thus, these data do not agree with past opinions on this issue (Conture & Kelly, 1991; Cooper & Cooper, 1985; Van Riper, 1971). Additional analyses are in progress attempting to identify specific movements that may serve as predictors.

### **Durational Characteristics**

Considering the limited data, any conclusion here is premature. Nevertheless, the time interval between single-unit repetitions provides a promising focus for further research and could emerge as a reliable predictive factor around 13 to 18 months post-onset of stuttering.

### **Phonology, Language, and Nonverbal Skills**

Taken together, the findings suggest that chronic stutterers perform poorer than do recovered stutterers on phonology, language, and nonverbal skills although not necessarily below average levels. Furthermore, phonologic skills may indeed be below age norms at very early stages of stuttering in children with the potential of becoming chronic. Thus, this parameter may be especially useful for children who are being evaluated soon after onset. The relation of these parameters to gender and age is not yet clear; however, they may resolve past conflicting findings about the relations among stuttering, language, phonology, and non-verbal intellectual functions. Subgrouping of subjects (e.g., Persistent vs. Recovered) may provide an important key to understanding such relations.

### **Genetic Factors**

The association of a positive familial history of chronic stuttering with similar outcomes of our subjects is an important finding of the present study. Although others (e.g., Riley, 1981) have also suggested that familial history of stuttering is a predictive factor, our findings specify the particular kind of history that one should look for. The value of this finding lies not only in its predictive power but also in its contribution to etiological research. As we add to the database of persistent and recovered probands, it may be possible to increase the precision of predictions and our understanding of the transmission of the disorder and other factors identified in this study.

## **Recovery**

As mentioned in the introduction, previous longitudinal studies reported recovery from stuttering at rates of 78% or higher. Although the present report was not planned to focus on this issue, the composition of the stuttering groups selected for the analyses here, about two-thirds recovered and one-third persistent subjects, provides a very conservative reflection of the pervasiveness of recovery. In fact, one of the main challenges in preparing this report was the identification of a sufficient number of children who have maintained stuttering for at least three years. Currently, our general assessment is that recovery exceeds 70% in the entire sample. Therefore, we disagree with recent arguments (Ramig, 1993) that recovery has been over-estimated. A careful review of Ramig's data reveals that 14 of his 17 stuttering subjects were considerably older than the children included in the present sample as well as in our previous reports. Those 14 children ranged in age from 4:9 to 8:6 years. Although no data were provided concerning the length of their stuttering histories at the time of the initial evaluation, based on their age and general information about the typical age at onset, it can be assumed to have been more than a year or two, if not several years. In other words, Ramig's sample appears to be considerably biased, consisting of children who at first assessment had already passed the period where much of the spontaneous recovery occurs. As a matter of fact, his findings appear to support our conclusions that chances for chronicity increase approximately 15 months after onset. The important point here is that accurate data on recovery can be obtained only when sampling begins close to stuttering onset, not several years later. Recovery in school-age children and in preschool children are two different issues.

## **SUMMARY**

Although no single certain predictor has yet been identified, significant progress is being made toward achieving early differentiation of a child's risk for chronic stuttering. Stuttering appears to be related to several factors, genetic and environmental, and the relationship between chronicity and factors not included in this study may prove to be pertinent. While the present findings do not add up to positive identification of persistent stuttering, they do provide predictive indicators that have implications for research and decision-making regarding treatment of early childhood stuttering. Based on these findings, researchers and clinicians may be well-advised to obtain accurate information about (a) age at onset, (b) duration of the disorder, (c) family history of persistent and recovered stuttering, and (d) scores on language/nonverbal measures. Previous reports (Ambrose, Yairi, & Cox, 1993; Seider, Gladstien, & Kidd, 1983) have indicated that sex is also an important factor in that females have a better chance for recovery. On the other hand, our present longitudinal data indicate that severity and early presence of secondary physical characteristics do not appear to provide predictive information. Although this conclusion differs from those of several previous authors (Conture, 1990; Cooper & Cooper, 1985; Curlee, 1993; Riley, 1981) we suggest that difference in age and elapsed time since stuttering onset between our subjects and

theirs can account for the disagreement. Signs of chronicity in older children (e.g., 6- or 7-year-olds) who had stuttered for two years may not be quite the same as those in 2- to 4-year-olds who have short stuttering histories.

Perhaps the most important immediate impact concerns early childhood stuttering research. Much of the conflict in past research results may be better understood in terms of the subgroups described in this study. In future basic and clinical research, including treatment efficacy research, there should be extreme care in selecting subjects, taking into account those factors that may skew findings. Accurate representation of the subject population (i.e., samples that include young children near the onset of stuttering) is clearly a paramount factor.

The ability to make accurate predictions could have a revolutionary impact on the long-term objective of cost-effective selective treatment for stuttering children. It is not practical, possible, or necessary to put every child who stutters into therapy. Economic conditions and emerging health policies, in fact, may make this option more difficult. For any child who appears likely to continue to stutter, treatment should not be delayed. But it may be advantageous to defer treatment for those with few or no risk factors and/or mild stuttering that does not cause concern for either child or parents. However, as Yairi (1995) has suggested, for children not receiving treatment, close periodic monitoring by an expert speech-language clinician aimed at detecting changes in symptomatology is a desirable strategy.

## Appendix

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Head and Facial Movements	
Brow raise	Upper lip raise
Brow lower	Lower lip depress
Lid raise	Lip corner(s) pull
Lid lower	Lip pucker
Lids tight	Lip tight/press
	Bite lip
Nose wrinkle	
Nostril movement	Jaw drop
	Jaw raise
Head movement	Jaw to side
Eye movement	Jaw clench

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