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RELATIONSHIP BETWEEN STUTTERERS' GENETIC HISTORY AND SPEECH-ASSOCIATED VARIABLES

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The symptomatology, reading ability, anxiety levels, responsiveness to therapy, and speech-motor behavior of elementary- and high-school children who stutter and had a positive family history of stuttering were compared with those of stutterers with a negative kinship history of stuttering. The stutterers with a positive family history of stuttering differed significantly from those whose family history was negative with respect to the frequency of silent and oral prolongations and speech-motor behavior. These findings suggest that their neuromotor functioning is related to their genetic susceptibility to stuttering.

INTRODUCTION

Several studies have shown that the risk of stuttering among relatives of an individual who stutters is greater than that present in the general population (Andrews and Harris, 1964; Kidd, 1977). The data of these investigations also have made it evident that the risk of stuttering among relatives of the female stutterer is greater than the risk among relatives of the male stutterer. Yet, females are less likely than males to stutter. This suggests that females who stutter have a greater genetic loading than male stutterers. These risk data provide a strong argument for the importance of heredity in the etiology of stuttering

An assumption fundamental to many current theories is that stuttering results from a genetic predisposition that is triggered by environmental factors (Andrews et al., 1983). Also, it is generally posited that the relative contribution of genetic and environmental factors varies among stutterers. This posture is consistent with Kidd's (1977) data, and his conclusion that there is heterogeneity among stutterers in the degree of genetic loading. In this respect, the data from the risk studies suggest that the relative

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contribution of genetic predisposition is greater for stutterers who have a family history of stuttering than for those who do not.

Another possibility is that the heterogeneity among stutterers observed may be a reflection of etiological differences. Those with a family history of stuttering may be individuals whose disruptions are a manifestation of an inherited neuromotor disorder. In contrast, those without a family history may display fluency failures as a result of an acquired or learned impairment that is stress induced. This notion corresponds with the distinction that has been made over the years between so-called neurogenic and psychogenic stutterers (e.g., Van Riper, 1971).

Few studies have been conducted that have compared the behavioral, cognitive, and physiological characteristics of diagnosed stutterers who do or do not have a family history of stuttering. In one such investigation, Andrews and Harris (1964) found that the presence or absence of a family history of stuttering was unrelated to any of the variables (e.g., severity of stuttering) that they studied. More recently, Kidd et al. (1980) reinvestigated the importance of the presence or absence of a history of stuttering. Although they more precisely determined whether or not stuttering occurred among their subjects' relatives, they, too, did not find a relationship between genetic history and stuttering severity. Still, the relationship between heredity, as reflected in family history, and speech-associated variables warrants further and more complete study. This is both because so few investigators have explored the issue involved and because the criterion variables investigated have been relatively limited in number.

PROCEDURE

The responses of stutterers in a series of four studies, previously conducted by the experimenters, were reanalyzed to meet the purpose of the present investigation (Janssen et al., 1983; Janssen and Kraaimaat, 1980; Janssen and Wieneke, 1987; Kraaimaat et al., 1988). The available data include both information relative to heredity and information specific to the stuttering subjects' symptomatology, reading ability, speech-associated anxiety, responsiveness to therapy, and speeech-motor behavior.

The subjects in each of the aforementioned studies (or their parents) completed a self-report questionnaire, which provided information relative to the presence or absence of stuttering among family members. On the basis of these data, and those specific to the separate investigations, two groups were formed: a group of stutterers with a positive family history of stuttering and a group in which there was no such history. Those assigned to the positive history group had at least one parent and/or one sibling who stutters or was reported to have stuttered. Those in the negative history group did not report the presence of stuttering in the family.

That is, no one having first-, second-, or third-degree kinship to the subject was said to have ever stuttered. Those who did not meet the criteria for inclusion in either group were not subjects of analysis.

RESULTS

Symptomatology

The fluency failures and nonverbal concomitants of elementary- and high-school male stutterers who did and did not have a positive family history were investigated. Thirteen elementary-school stutterers met the criteria for inclusion in the positive family history group. They ranged in age from 8 to 12, and their mean age was 10.6 years. Twenty-five children did not have a family history of stuttering. These youngsters also ranged in age from 8 to 12, and their mean age was 10.1. Eighteen of the high-school children diagnosed as stutterers had a family history of stuttering. Their age ranged from 13 to 16, and their mean age was 14.8. Twenty-two of the children had a negative history. They ranged in age from 13 to 17, and their mean age was 14.9 years.

The frequency of fluency failures of both the elementary- and high-school stutterers, those with and those without a family history of stuttering, was determined from video tape recordings taken as the stutterers read an age-appropriate passage. The reading material of the elementary-school children contained 390 syllables, and that read by the high-school children was 384 syllables in length. Relative to the number of syllables spoken, the fluency failures that were shown by the children in each of the subject groups were assigned to one of the following categories: 1) fast repetitions of an integral or interjected sound or syllable, 2) oral and silent prolongations, and 3) normal disfluencies, including word or phrase repetitions and word interjections. The frequency of nonverbal adjustive behaviors (i.e., excessive movement of the jaw, mouth, eyelid, eyebrow, forehead, or head) was also scored from the video tape recordings of the oral reading task (for more details, see Janssen et al., 1983; Janssen and Kraaimaat, 1980).

Differences in the extent to which the elementary and high-school children who did or did not have a family history of stuttering, displayed fluency failures, normal disfluencies, and nonverbal adjustive behaviors were assessed by means of univariate analyses of variance (ANOVAs). The results of these analyses, along with the means and standard deviations for the different behavioral categories, are shown in Table 1. There it can be seen that descriptively both the elementary- and high-school children with a family history of stuttering showed more oral and silent prolongations than those stutterers who had a negative history. For both groups of children, the difference was statistically significant.

Table 1. Fluency Failures, Normal Disfluencies, and Nonverbal Adjustive Behaviors of Elementary- and High-school Stutterers Having a Positive or Negative Family History of Stuttering

	Positive family history		Negative family history		Univariate
	Mean	SD	Mean	SD	*F* value
Elementary-school children					
Fast repetitions of sounds or syllables	0.52	0.56	1.22	2.74	0.84
Oral and silent prolongations	12.02	16.66	1.77	3.04	9.10[b]
Normal disfluencies	4.47	2.56	4.84	3.68	0.10
Nonverbal adjustive behaviors	14.67	12.27	6.77	8.00	5.49[a]
High-school children					
Fast repetition of sounds or syllables	2.78	2.64	4.85	9.25	0.84
Oral and silent prolongations	9.75	10.18	4.53	5.90	4.10[a]
Normal disfluencies	4.73	5.04	4.47	4.12	0.03
Nonverbal adjustive behaviors	8.66	10.39	8.61	14.22	0.00

[a] $p < 0.05$.
[b] $p < 0.01$.

The elementary-school children with a positive family history of stuttering also displayed descriptively more nonverbal behaviors than their peers in the negative control group. This difference was statistically significant. It was not for the high-school children. The latter children showed essentially the same number of nonverbal adjustive responses among both those who did and those who did not have a family history of stuttering. Possibly, this difference is a function of the fact that the elementary-school stutterers with a positive history emitted almost seven times more prolongations than those with a negative history, while among the high-school children, those with a positive history displayed only about two times more prolongations than the control subjects did.

Reading Ability

The reading skills of the aforementioned elementary school children were assessed by means of the One-Minute Word Test (Brus and Voeten, 1972), the Differential Sentence Test (Dommerholt, 1970) and the Reading Comprehension Test (Brus and Van Bergen, 1973). These standardized Dutch reading achievement tests, which were given to both groups of children, yielded six measures of reading ability: reading errors, number of words read in 1 minute, reading errors, percentage errors revised, total number of correct responses, and working time in seconds (for procedural details, see Janssen et al., 1983).

Table 2. Reading Ability of Elementary-School Stutterers with a Positive or Negative Family History of Stuttering

	Positive family history		Negative family history		Univariate
	Mean	SD	Mean	SD	*F* value
Reading errors—Word Test	3.77	3.19	3.84	3.26	0.01
Number of words—Word Test	51.92	18.51	55.48	16.42	0.37
Reading errors—Sentence Test	4.42	2.62	5.28	3.03	0.76
Errors revised—Sentence Test	18.66	14.63	19.26	12.67	0.02
Correct responses—Comprehension Test	23.61	7.11	24.28	5.57	0.10
Working time/second Comprehension Test	1220	374	1363	334	1.44

Table 2 shows the means and standard deviations of the six reading skill measures for both the group of stutterers with and those without a familial history of stuttering. The between-group differences, which were descriptively small, were assessed by means of univariate ANOVAs. The results showed that reading ability of the two groups did not differ statistically.

Speech-Associated Anxiety

The next reanalysis focused on the speech-associated anxiety of the aforementioned 18 high-school stutterers with a positive family history and the 22 with a negative history.

In this study, autonomic and cognitive measures of speech-related anxiety were assessed. The autonomic measures (heart rate and skin conductance) were continuously recorded during both a rest period and during a subsequent oral reading period. The change scores, i.e., the difference between the measures displayed in these two periods, were used to compare the anxiety shown by the subjects in the positive and negative history groups.

Cognitive anxiety was measured by means of the ABV (Wilde, 1970) and PMT (Hermans, 1976) tests, two standardized Dutch personality inventories. These test procedures provided scores for neuroticism and fear of failure. Speech-specific negative emotion was measured by the Speech Situation Checklist (Brutten, 1973). State anxiety was measured using a five-point Likert scale (for details, see Janssen and Kraaimaat, 1980).

Means and standard deviations and the univariate *F*-values for those stutterers with a positive family history of stuttering and those with a negative history are displayed in Table 3. There it can be seen that the autonomic and cognitive anxiety scores of the two groups were strikingly

Table 3. Autonomic and Cognitive Measures of Anxiety Among High-school Stutterers Having a Positive or Negative Family History of Stuttering

	Positive family history		Negative family history		Univariate
	Mean	SD	Mean	SD	*F* value
Autonomic anxiety					
Heart rate	12.73	7.32	14.61	9.82	0.45
Skin conductance	0.16	0.13	0.14	0.11	0.26
Spontaneous fluctuations	3.90	3.70	3.26	3.91	0.27
Cognitive anxiety					
Neuroticism	72.22	20.58	71.73	23.00	0.01
Fear of failure	14.11	4.95	13.41	5.64	0.17
Speech-specific negative emotion	2.46	0.59	2.25	0.55	1.38
State anxiety	3.33	0.69	3.23	0.61	0.27

similar. Univariate ANOVAs failed to reveal any significant difference between the two groups.

Responsiveness to Therapy

Thirteen of the high-school stutterers with a positive family history and 15 of those who reported the absence of stuttering in the family were investigated in a study that was originally designed to evaluate their responsiveness to therapy (Kraaimaat et al., 1988). The issue that led to the reevaluation of these data is whether or not high-school stutterers with a high genetic loading and stutterers without a family history of stuttering were differentially affected by 4 consecutive weeks of broad-spectrum therapy.

The therapy consisted of an intensive daily training in relaxation and regulated breathing, desensitization of speech-associated anxiety, cognitive restructuring, and self-control. The subjects were assessed prior to therapy and again about 7 months after the termination of therapy (for details, see Kraaimaat et al., 1988).

The pre- and posttherapy group means and standard deviations for percent stuttered syllables and number of syllables per minute for both groups are shown in Table 4.

The data were analyzed by means of a two-way ANOVA with repeated measures on one factor. Separate analyses were conducted for percent stuttered syllables and syllables per minute. For the subjects in both groups, the percent of stuttered syllables decreased significantly as a result of therapy ($F = 12.40$, $df = 1.26$, $p = 0.002$). Moreover, those with a negative family history showed a descriptively greater reduction in mea-

Table 4. Pre- and Postmeasures of Responsiveness to Therapy Among High-school Stutterers Having a Positive or Negative Family History of Stuttering

	Positive family history		Negative family history	
	Pre	Post	Pre	Post
% Stuttered syllables	17.78	10.91	15.23	3.86
	(13.57)	(11.76)	(16.37)	(6.86)
Syllables per minute	146.13	150.32	146.81	163.92
	(58.11)	(80.91)	(60.14)	(39.27)

sured stuttering than those with a positive history. However, this difference did not reach statistical significance ($F = 1.42$, $df = 1.26$, $p = 0.24$). Similarly, although the pre–post increase in the number of syllables per minute was descriptively greater for the stutterers who did not have a family history of stuttering than for those with a positive history, the groups did not differ significantly ($F = 0.12$, $df = 1.26$, $p = 0.74$). Thus, the evidence suggests that stutterers with and without a family history of stuttering do not seem to represent different clinical types with regard to responsiveness to broad-spectrum therapy.

Speech Motor Behavior

The last study revisited was designed to evaluate the speech-motor abilities of adult stutterers and nonstutterers during the production of fluent speech (Janssen and Wieneke, 1987). Electroglottographic measurements were made to assess their speech segment durations and durational variability. The results of this investigation indicated that there was both significantly greater variability in the duration of glottal activity and glottal inactivity among the stutterers than among the nonstutterers.

The finding of heightened variability among the stutterers was interpreted by the investigators as evidence of an instability in the timing of the phonatory system. It was seen as reflective of a general limitation in their ability to control the temporal aspects of speech. Nevertheless, the variability of the two groups overlapped; the variability of some of the stutterers was similar to that of the nonstutterers. This could possibly be a result of differences in their genetic history. That is, the stutterers with a lesser degree of genetic loading may have been motorically more capable and, thus, more like the nonstutterers. In contrast, the stutterers having greater genetic loading may have responded in ways that were less like those of nonstutterers. To explore this possibility, the data of the stutterers were reevaluated with respect to their family history. Ten of the subjects met the criteria for inclusion in the positive family group. Ten others met them for inclusion in the negative family group. Those with a

Table 5. Duration of Voiced and Unvoiced Segments and Their Variability Among Adult Stutterers with a Positive and Negative Family History of Stuttering

	Positive family history		Negative family history		Univariate
	Mean	SD	Mean	SD	*F* value
Duration of voiced segments	1,716	488	1,373	194	4.26[a]
Duration of unvoiced segments	658	482	563	212	0.32
Variability of voiced segments	298	108	276	76	0.27
Variability of unvoiced segments	463	98	315	66	15.84[b]

[a] $p < 0.05$.
[b] $p < 0.01$.

family history of stuttering ranged in age from 16 to 32, and their mean age was 23.3 years. Those with a negative history ranged in age from 17 to 40, and their mean age was 24.5 years.

The stutterers in the two subject groups were compared with respect to their average duration of voiced segments, average duration of unvoiced segments, variability of voiced segments, and variability of the unvoiced segments. The results are shown in Table 5. There it can be seen that the segment duration and variability of stutterers with a positive family history was descriptively greater than that of those with a negative history.

Multivariate ANOVA confirmed the overall difference. The multivariate difference between the two groups was statistically significant at the .01 level ($F = 4.79$, $df = 4.15$). Univariate tests indicated that the difference applied especially to the average duration of the voiced segments and to the variability of the unvoiced segments.

DISCUSSION

The reanalyzed data provided little evidence to support the hypothesis that those with a history of stuttering in the family are consistently different from those whose family history is negative. Those with a presumably greater genetic loading were not found to be less responsive to broad-spectrum therapy than those with a negative family history. Moreover, with respect to speech-related and trait-based anxiety levels, both groups showed similar responsivity. This suggests that anxiety level is not related to the same genetic factors that seemingly predispose one to stuttering.

The presence or absence of a family history of stuttering did, however, appear to be related to the extent to which certain forms of dysfluency and nonverbal adjustive behaviors are displayed. Genetic loading was associated with a greater likelihood of oral and silent prolongations, es-

pecially among elementary-school children. This finding is not consistent with those of Kidd et al. (1980). However, they measured the frequency of molar stuttering moments rather than the amount of specific types of fluency failures and nonverbal adjustments. And, as we have said before, "outcome controversy is likely to result if the analysis of . . . fluency failure is undertaken in a molar fashion" (Kraaimaat et al., 1988). The current findings suggest that molecular analysis involving specific forms of dysfluent behaviors more sensitively detects genetic influence than molar frequency counts do.

Oral and silent prolongations, the dysfluency type whose frequency was greater among those with a positive family history than those with a negative history, are generally considered to reflect speech-motor breakdown. It is interesting, in this respect, that significant between-group differences became apparent when the phonatory timing capabilities of the two groups were compared. Stutterers with a family history of stuttering were slower and more variable, particularly with respect to the duration of voiced segments and the variability of unvoiced segments, than the stutterers with a negative family history. This suggests that their neuromotor functioning is related to genetic susceptibility to stuttering.

We must view our conclusions with caution. The number of subjects in the positive and negative groups was rather small. Replication with larger subgroups, using a more precise definition of familial history of stuttering, is warranted. Nevertheless, the data suggest that in research involving stutterers, we may be losing important information by pooling data. Dividing subjects, not only on the basis of sex or stuttering severity, but also on the basis of family history, seems to be a viable strategy for identifying more homogeneous subgroups and increasing the power of our studies to detect differences.

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