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GENETIC FACTORS IN STUTTERING: A REPLICATION OF AMBROSE, YAIRI, AND COX'S (1993) STUDY WITH ADULT PROBANDS

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The relationship between the gender of stuttering probands and the incidence of stuttering among their relatives was investigated by obtaining detailed pedigrees from 106 adult stutterers. We found, as Ambrose, Yairi, and Cox (1993) did in a study of preschool probands, that no evidence existed for the contention that relatives of female probands are more likely to stutter than those of male probands.

INTRODUCTION

In a recent study Ambrose, Yairi, and Cox (1993) presented data on the incidence of stuttering among relatives of preschool children who stutter. Using these young children as probands, Ambrose et al. (1993) reported discrepancies between their results and those of Kidd, Heimbuch, and Records (1981), whose sample was composed primarily of adult stutterers. More specifically, they did not find that relatives of the female probands were more likely to stutter than relatives of the male probands. In other words, their results were not consistent with those who reported a relationship between the gender of the

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stuttering proband and the incidence of stuttering among their relatives (Kidd et al., 1981).

Ambrose et al. (1993) studied young stuttering children as probands because, in their opinion, data on familial incidence obtained from adults who stutter may be biased. In this regard they pointed out that in studies of adult probands, females may be undersampled because of their greater tendency to recover. Thus, it is likely that the use of unrecovered females will result in samples of female stutterers who have an especially strong propensity for stuttering. In addition, Ambrose and colleagues argued that data on stuttering among relatives of probands who are children, rather than adults, may be more reliable because their recollection will likely be fresher. Moreover, they pointed out that the pool of respondents would be wider in range because aunts, uncles, and grandparents may still be living and available for questioning.

It can be argued that the discrepancy between the Kidd et al. (1981) data and that of Ambrose et al. (1993) may well be a result of the relatively small number of probands used in the latter study rather than a reflection of population differences. It follows from this that family data from a relatively large sample of adult probands would help shed light on the reported discrepancy. Therefore, as part of a longitudinal study on factors that contribute to the development of incipient stuttering in high-risk children (Kloth, Janssen, Kraaimaat, & Brutten, 1995), family data of 77 adult male probands and 29 adult female probands were collected. These data were used to investigate the gender of proband effect suggested by Kidd et al. (1981).

METHOD

Subjects

The 106 stutterers, whose families were the subject of study, were the parents of the "high-risk" children who participated in a longitudinal study on factors that contribute to the development of incipient stuttering. The parents (77 stuttering fathers and 29 stuttering mothers) were brought to the authors' attention

Table 1. Frequency of Stuttering Among Male and Female Relatives of Male and Female Probands, Represented by the Number of Male and Female Relatives Who Stutter Divided by the Total Number of Male and Female Relatives, and Male-to-Female (M/F) Ratios

	Male relatives	Female relatives	M/F ratio
Male probands ($n = 77$)	127/846 = 0.150	51/833 = 0.061	2.46
Female probands ($n = 29$)	39/311 = 0.125	19/306 = 0.062	2.02

by speech-language pathologists (41%), the Dutch Association of Stutterers "Demosthenes" (23%), and area pediatricians (36%).

Procedure

A family pedigree of stuttering was obtained from each adult proband by means of a self-report questionnaire. The questionnaire, which was developed for the purpose of the present study, asked for the presence of stuttering at any time in each first-, second-, and third-degree relative. Each proband had two weeks to gather the data from their own family and to complete the questionnaire. Subsequently, the data were checked by the second author in an intensive interview with the proband.

RESULTS AND DISCUSSION

As the data in Table 1 and Table 2 make evident, our findings tend to be consistent with those of Ambrose et al. (1993). We, like Ambrose et al. (1993), did not observe the hypothesized gender-of-proband effect. For, as the data in Table 1 show, the relatives of female probands were not more likely to stutter than were those of male probands (Chi square = 0.66; $p > .70$). As can be seen in Table 2, our female probands had only a slightly higher frequency of affected sons and sisters than did our male probands. The frequency of all of the other affected relatives, however, was about the same or lower among the female probands than among male probands. Noteworthy, also, is the fact that though we found that among female probands there was a somewhat higher frequency of female relatives who stutter than among male probands, the gender effect was even less pronounced than those reported by Ambrose et al. (1993) (see Table 1). Whereas in their study the male-to-female ratio was 1.50

Table 2. Frequency of Stuttering Among Relatives of Male and Female Probands, Represented by the Number of Relatives Who Stutter Divided by the Total Number of Relatives

Relatives	Male probands	Female probands	Total
Fathers	17/77 = 0.221	7/29 = 0.241	0.226
Mothers	4/77 = 0.052	0/29 = 0	0.038
Sons	28/102 = 0.275	10/32 = 0.313	0.284
Daughters	15/83 = 0.181	6/32 = 0.188	0.183
Brothers	34/146 = 0.233	12/59 = 0.203	0.224
Sisters	12/124 = 0.097	9/46 = 0.196	0.124
Uncles	34/330 = 0.103	6/132 = 0.045	0.087
Aunts	11/334 = 0.033	2/148 = 0.014	0.027
Male cousins	14/191 = 0.073	4/59 = 0.068	0.072
Female cousins	9/215 = 0.042	2/51 = 0.039	0.041

for female probands, we found a male-to-female ratio of 2.02. Thus, for the female probands the distribution of affected relatives was less evenly distributed by gender than in the Ambrose et al. study (1993).

Regarding our findings, attention should be given to the fact that our subjects were adult stutterers. Nevertheless, no support could be found for Kidd's model of vertical transmission with sex-modified expression. A Mendelian major locus model of genetic transmission in stuttering is still open to question.

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