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ORIGINAL CONTRIBUTION

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The family history of children with elective mutism: a research report

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Abstract The family history was studied in children with elective mutism. The samples comprised a series of N=38 children with elective mutism and a control group of N=31 children with a similar behavioural phenotype, i.e., the combination of an emotional disorder and a developmental disorder of articulation or expressive language. Interviews were performed with the respective mothers. There was a clear excess of the personality trait of taciturnity in first-, second-, and third-degree relatives. Although mutism was reported almost exclusively in the group of relatives of children that manifested elective mutism, the differences between the two samples were not significant probably due to low frequencies. Disorders of speech and language were quite common in the relatives of subjects in both samples. Psychiatric disorders were more frequently reported in the families with an electively mute child. The study lends some evidence for the assumption that genetic factors may play a role in the etiology of elective mutism.

Key words Elective mutism – family history – etiology

Introduction

Elective mutism is a relatively rare disorder in children. The term was coined by the Swiss pioneer of child psychiatry, Moritz Tramer, in 1934 (7). Since that time, various reports largely based on single cases and only very few analyses of more extended series of patients appeared in the German and Anglo-Saxon literature. Recently, the scientific interest in elective mutism has increased considerably, as indicated by various publications. A review of the Anglo-Saxon literature, including practical guidelines for the assessment and treatment of this relatively rare clinical phenomena, was provided by Dow et al. (3). The psychiatric characteristics of the affected patients were described by Black and Uhde (1) and an evaluation of a specific treatment approach was provided by Krohn et al. (1994). In addition, we reported on the most extended series of one hundred patients whom we were able to collect by co-operation of a parents' self help group and by analyzing the case files of two institutions that serve in the field of child and adolescent psychiatry (6).

Our interest in this issue arose out of the rather unclear etiology of elective mutism. Tramer (7) was the first to point to the familial trait of speech avoidance when he analyzed his three cases. However, the issue of familial or genetic factors has been almost entirely ignored by other researchers who later contributed to the literature. Brown and Lloyd (2) reported that 51% of the children who did not speak at school had at least one shy parent and 32% had siblings with at least a transient form of mutism. In a series of eleven children, Wergeland (8) noted that nine of the children came from homes with a strong familial shyness and reservation. Similarly, Wright (10) noted the possibility of a

ECAP 044

European Child & Adolescent Psychiatry, Vol. 6, No. 2 (1997)

familial predisposition. Kolvin and Fundudis (4) found a high rate of personality disorders and an excess of psychiatric disturbance in the parents of children with elective mutism as compared to controls.

In their study on the effectiveness of a specific treatment for 20 children with elective mutism, Krohn et al. (5) noted that only one parent had a history of documented mental illness whereas five of the parents described themselves as currently being pathologically shy or anxious or having been so as children. Wright (9), in a recent letter to the editor, described a family history positive for anxiety disorder in three of the four children. Finally, in another recent report on 30 non-clinical children with elective mutism, Black and Uhde (1) found a surprisingly high rate of social phobia (70%) as well as elective mutism (37%) in the family history. However, their study was restricted to first-degree family members only. Given this rather limited knowledge on the role of familial factors in childhood elective mutism, we decided to study this issue in a more systematic way.

Method

Subjects

Family data were collected in 38 patients with elective mutism diagnosed in accordance with ICD-10 criteria. This sample contained 19 children whose parents had joined a self-help group and who were personally assessed by the first author in 1992 and 1993. Another 19 children were originally seen between 1988 and 1992 at the Child and Adolescent Psychiatric Service of the Canton of Zürich, Switzerland.

The clinical control group was selected from the same institution. In order to come up with a relatively similar behavioural phenotype, it was decided to identify those children who were assessed as fulfilling the following two criteria: an emotional disorder (according to F 93 in the ICD-10 scheme of diagnoses) and a specific developmental disorder of speech and language, i.e., either an articulation disorder (F 80.0) or an expressive language disorder (F 80.1). By including the component of the developmental delay of speech and language, the issue was taken into consideration that elective mutism in a large number of children develops out of retarded speech development. A total of N=34 children who attended the service between 1988 and 1992 and who fulfilled the two criteria were identified in the files. Three parents refused to co-operate, leaving a total of N=31 children as a control group.

Procedure

An interview schedule that assessed sociodemographic and family history data was designed for the present study. Interviews were performed with the mothers of the subjects. The assessment started with a genogram of the respective family. Besides parents and siblings, data on all aunts and uncles, cousins, and grandparents were systematically recorded. After drawing the family genogram, the following items were addressed in the interview by pointing to each individual member of the extended family tree: taciturn behaviour or speech avoidance, mutism, disorders of speech and language, and psychiatric disorders. In each instance the informant was asked whether or not the individual member of the large family tree fulfilled any of these four items. For the present study a 'taciturn' person was defined in the interview as a person with very little if any spontaneous speech, a restricted length of spoken sentences, and long periods of no verbal communication at all. 'Taciturnity' was defined as a personality trait and not as a passing behavioural phenomena. The other three categories were conceptualized in terms of the diagnostic criteria of the ICD-10 schema.

Nineteen mothers of a child with elective mutism who belonged to the self-help group were interviewed by the senior author in 1992. The rest of the index-sample mothers were interviewed again in 1993 by the respective clinicians who were responsible for treating the individual child at the time that he or she attended the service. The interviews with the mothers of the control group were performed by the co-author.

Data analyses consisted of cross-tabulation, including Chi^2 or Fishers Exact Test.

Results

A comparison of sociodemographic data is given in Table 1. The two samples differ significantly concerning the distribution of sex and of socioeconomic status (SES). Elective mutism favoured girls and a middle class background. In addition, the subjects of the target group were significantly older at presentation (M=107.7, SD=34.2 months) than the control group (M=81.0, SD=30.6 months; t=3.37, df=67, p=0.001).

Table 1 Sex and socioeconomic status

	Children with elective mutism		Controls		Chi^2	df	p
	N	%	N	%			
Sex							
Male	12	31.6	20	64.5	7.44	1	0.006
Female	26	68.4	11	35.5			
Socioeconomic status							
Lower class	15	39.5	19	61.3	4.06	1	0.04
Middle class	22	57.9	10	32.3			
Missing data	1	2.6	2	6.4			

Table 2 Family history findings (life-time prevalence rates in %)

	Children with elective mutism			Controls			p-Values		
	Total (N=38)	Males (N=12)	Females (N=26)	Total (N=31)	Males (N=20)	Females (N=11)	Total	Males	Females
Taciturnity									
Fathers	26.3	33.3	23.1	12.9	5.0	27.3	n.s.	0.05	n.s.
Mothers	21.1	16.7	23.1	6.5	10.0	–	0.09	n.s.	0.10
Parents	23.7	25.0	23.1	9.7	7.5	13.6	0.03	0.06	n.s.
Brothers	8.8	6.7	10.5	–	–	–	n.s.	n.s.	n.s.
Sisters	16.7	–	22.2	–	–	–	0.07	n.s.	n.s.
Siblings	12.1	4.8	16.2	–	–	–	0.04	n.s.	n.s.
1st-degree relatives	18.7	15.6	20.2	6.3	4.9	8.8	0.007	0.07	n.s.
2nd-degree relatives	7.8	3.6	9.9	2.0	2.1	2.0	0.001	n.s.	0.001
3rd-degree relatives	5.9	2.0	7.5	1.0	1.4	–	0.007	n.s.	0.001
Mutism									
Fathers	–	–	–	–	–	–	n.s.	n.s.	n.s.
Mothers	7.9	8.3	7.7	–	–	–	n.s.	n.s.	n.s.
Parents	3.9	4.2	3.8	–	–	–	n.s.	n.s.	n.s.
Brothers	5.9	–	10.5	–	–	–	n.s.	n.s.	n.s.
Sisters	–	–	–	–	–	–	n.s.	n.s.	n.s.
Siblings	3.4	–	5.4	–	–	–	n.s.	n.s.	n.s.
1st-degree relatives	3.7	2.2	4.5	–	–	–	0.07	n.s.	n.s.
2nd-degree relatives	0.9	0.9	0.9	–	–	–	n.s.	n.s.	n.s.
3rd-degree relatives	–	–	–	0.5	–	1.4	n.s.	n.s.	n.s.
Speech and language disorders									
Fathers	–	–	–	3.2	5.0	–	n.s.	n.s.	n.s.
Mothers	2.6	–	3.8	3.2	–	9.1	n.s.	n.s.	n.s.
Parents	1.3	–	1.9	3.2	2.5	4.5	n.s.	n.s.	n.s.
Brothers	5.9	–	10.5	–	–	–	n.s.	n.s.	n.s.
Sisters	4.2	–	5.6	4.8	–	11.1	n.s.	n.s.	n.s.
Siblings	5.2	–	8.1	3.0	–	8.3	n.s.	n.s.	n.s.
1st-degree relatives	3.0	–	4.5	3.2	1.6	5.9	n.s.	n.s.	n.s.
2nd-degree relatives	1.2	–	1.7	–	–	–	0.08	n.s.	n.s.
3rd-degree relatives	0.6	–	0.8	1.0	1.4	–	n.s.	n.s.	n.s.
Psychiatric disorders									
Fathers	5.3	8.3	3.8	3.2	–	9.1	n.s.	n.s.	n.s.
Mothers	15.8	16.7	15.4	–	–	–	0.02	n.s.	n.s.
Parents	10.5	12.5	9.6	1.6	–	4.5	0.03	0.05	n.s.
Brothers	8.8	6.7	10.5	–	–	–	n.s.	n.s.	n.s.
Sisters	4.2	–	5.6	4.8	–	11.1	n.s.	n.s.	n.s.
Siblings	6.9	4.8	8.1	3.0	–	8.3	n.s.	n.s.	n.s.
1st-degree relatives	9.0	8.9	9.0	2.1	–	5.9	0.03	0.03	n.s.
2nd-degree relatives	5.2	2.7	6.5	3.4	3.7	2.9	n.s.	n.s.	n.s.
3rd-degree relatives	3.5	2.0	4.2	–	–	–	0.008	n.s.	n.s.

Sample sizes (Elective Mutism/Controls)
Fathers: N=38/31; Mothers: N=38/31; Parents: N=76/62; Brothers: N=34/12; Sisters: N=24/21; Siblings: N=58/33; 1st-degree relatives: N=134/95; 2nd-degree relatives: N=344/293; 3rd-degree relatives: N=170/207

Family history data of the two groups are compared in Table 2. Data are shown for the total groups and for the sex of the patients. 'Taciturnity' is the item that discriminates best between the samples. In comparison to the control groups, it tends to be more frequently found in mothers and sisters of children with elective mutism. On a more aggregated level, it is significantly more frequent in the parents and in first-, second-, and third-degree relatives. It is 2.5 times more frequent in first degree relatives compared to third-degree relatives.

There are a few sex effects that indicate that fathers of boys with elective mutism more frequently manifest the personality trait of being taciturn. This also leads to a trend of parents and first-degree relatives of boys with elective mutism as more often showing this behavioural feature. In contrast, more first- and second-degree relatives of girls with elective mutism manifest taciturn behaviour than the respective relatives of the control girls.

The second item, namely 'mutism', does not significantly differentiate between the two groups, probably

European Child & Adolescent Psychiatry, Vol. 6, No. 2 (1997)

because there is a very low base rate for this item. There is only a trend for relatives of the target group to display mutism more frequently than those of the control group. Interestingly, with the exception of one single relative, all relatives with mutism are found among the target group of children.

The rate of manifestation of speech and language disorders in the relatives is similar for both groups. Finally, there is a clear preponderance of psychiatric disorders in the relatives of children with elective mutism. This is seen on various levels, i.e., in mothers, parents, and first- and third-degree relatives. In addition, there are significant sex effects insofar as parents and first-degree relatives of boys with elective mutism more frequently manifest psychiatric disorders. In terms of diagnoses, the following disorders were observed in the relatives of the index group: substance disorders, schizophrenias, affective disorders, anxiety disorders, mental retardation, speech and language disorders, emotional disorders of childhood, tics, and enuresis. The relatives of the control group is comprised of relatives who suffered from substance disorders, affective disorders, adjustment disorders, and eating disorders.

Discussion

This empirical study is based on family history data; therefore, the findings cannot be precisely interpreted as to whether genetic or environmental factors are mainly operant. Genetic studies clearly would require twin, adoption, or genetic marker designs which, however, would be difficult to perform in an area of relatively rare clinical disorders such as elective mutism. Family history studies certainly are an important first step in order to elucidate the unclear etiology of this syndrome.

In terms of sex distribution, the index group reflects the previous finding in the extended sample in which there was also an excess of female subjects (6). In contrast, the control group of children having an emotional disorder and a delay of speech and language shows the typical preponderance of boys, as is usually found in the general child psychiatric population of this age range. The SES differences are mainly due to the population differences from which the two samples are derived. Whereas the index children were recruited from a parents' self-help group, the members of which are predominantly of middle class SES, there is an excess of lower class subjects in the control group, as is usual in the population of many public health service attendees. The age difference found for the subjects of the two samples is, again, primarily due to the composition of the target group. The latter included a large group of children whose parents formed a self-help group. These children tended to be older than the children who were referred to the service at a younger age (6).

The first and most impressive result of this family history study indicates that the personality trait of being 'taciturn' is found significantly more frequently among first-, second-, and third-degree relatives of children with elective mutism than in control families. This distribution across three degrees of relatives provides some evidence for the assumption that, in addition to any intrafamilial or even cultural factor, a genetic factor is involved in the transmission. When considering the sex factors in the two samples of children, there is no clear indication for the hypotheses that a sex-linked mode of transmission plays a role. Additional analyses did not reveal any significant relation between taciturnity among first-, second-, and third-degree relatives and social class so that there is no evidence that more reservation among middle class members might account for the findings on taciturnity.

As in all family studies of this kind, criticism may be directed at the finding of increased frequencies of taciturn behaviour in the relatives of children with elective mutism, because it may be argued that these families are more sensitized to perceiving this personality trait, whereas it may be overlooked in families in which there are no children with elective mutism. This general critique, which is valid for all psychiatric family studies, cannot be entirely ignored in the present study. Theoretically, the only way to control for this factor would be to directly examine all of the family members. However, this procedure would not only imply enormous expenses but it would also suffer from not including deceased relatives and from there not being any direct assessment tool for assessing the personality trait of taciturnity. The latter argument may also be directed towards the potential critique that pertains to the methods of this study. The personality trait in question can certainly only be assessed by interviewing significant family members, and it will hardly be possible to design a more reliable assessment tool than this.

Although the second item that was examined in the present study, i.e., mutism, was not significantly more frequent in the relatives of children with the same symptom, it is noteworthy that it occurred almost exclusively in the families with an electively mute child. The relatively low rates prevented any significant differences in the two samples from being measured. Taken together with the findings on taciturnity, it may be hypothesized for further studies that the more general factor of 'speech avoidance' may be the inherited trait rather than mutism. This factor may be combined with other factors in terms of a polygenic mode of transmission where various genes and environmental factors interact. This idea of a polygenic transmission of mutism is partially supported by the significantly increased frequencies of psychiatric disorders among first- and third-

degree relatives of children with elective mutism. A common liability factor towards psychiatric disorders may be operant in these families.

In conclusion, this first, extended family history study of a considerable series of children with elective mutism revealed some evidence that genetic factors may play a significant role in the etiology of this rare disorder in childhood. Larger samples with a greater number of relatives would allow the performance of segregation analysis. In addition, future studies might further improve the methodology as compared to this first pilot study. These refinements might include blind interviewing with regard to the status of patients vs. controls in order to rule out any ascertainment bias. In general, positive genetic findings would certainly lead to a revision of traditional theories on the pathogenesis of elective mutism with a predominant emphasis on environmental and psychogenic factors.

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