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Familial Aggregation of Spelling Disability

Gerd Schulte-Körne, Wolfgang Deimel, Katarina Müller, Cornelius Gutenbrunner and Helmut Remschmidt

Department of Child and Adolescent Psychiatry, Philipps-University Marburg, Germany.

This study examined the familial aggregation of spelling disability in a sample of 32 German school-aged children and their relatives. The influence of two different diagnostic criteria (low-achievement criterion, and regression-based IQ-discrepancy criterion) on the rate of affectedness was investigated. Results revealed that 52.3–61.9% of the sibs and 26–34% of the parents were spelling disabled. Little evidence was found for an influence of the diagnostic criterion on the rate of affectedness.

Keywords: Spelling disability, familial aggregation, diagnostic criteria, polygenic threshold model

Introduction

Ever since reading disability was first described by Morgan (1896) and Kerr (1897), a familial aggregation has been evident. Analysis of single and partly extended pedigrees showed a transmission of reading and spelling disability over three generations and provide evidence for an autosomal-dominant transmission with sex-dependant penetrance (Stephenson, 1907; Fisher, 1905; Hinshelwood, 1907; Plate, 1909; Op't Hof & Güldenpfennig, 1972; Schulte-Körne & Remschmidt, 1994). The comparison of family samples of reading disabled and non-reading disabled probands revealed higher rates of reading difficulties in parents and siblings of reading disabled children than in siblings and parents of controls (Owen, Adams, Forrest, Stolz, & Fisher, 1971; Foch, DeFries, McClearn, & Singer, 1977; DeFries, Singer, Foch, & Lewitter, 1978).

Although different diagnostic procedures (interviews, questionnaires, and psychometric tests), different diagnostic criteria (e.g. low reading achievement, discrepancy between IQ and reading achievement), and ascertainment strategies (clinic samples, high selected samples, high loading pedigrees) were used, the estimated familial risk for reading disability was found to be higher than the population risk. The overall rate of 30–70% of affected first-degree relatives strongly supports a familial aggregation and gives evidence for a major gene effect (Hallgren, 1950; Zahalkova, Vrzal, & Klobouka, 1972; Finucci, Guthrie, Childs, Abbey, & Childs, 1976; Vogler, DeFries, & Decker, 1985; Gilger, Pennington, & DeFries, 1991; Pennington et al., 1991; Wolff & Melngailis, 1994). Although single pedigree analyses give valuable information, only complex segregation analyses, preferably based on a large unselected family sample, allow statistical testing of the hypothesis on the mode of transmission. Recent segregation analyses of 204 families provided evidence for a sex-influenced, additive, or dominant transmission on the assumption of a prevalence of 7.5% and a male to female ratio of 1.8:1 in three of four samples (Pennington et al., 1991). Results obtained from the fourth sample gave evidence for a polygenic transmission.

The higher prevalence of reading plus spelling (Critchley, 1970) and pure spelling disability (Allred, 1990) in males gave support for a gender-influenced polygenic threshold model. This model (Carter, 1969) assumes that an underlying liability to spelling and reading disability is a function of genetic and environmental influences. An individual becomes reading and spelling disabled when the liability exceeds a certain threshold. To the extent that spelling and reading disability is heritable, relatives of affected individuals will have a higher average liability and thus will be at greater risk for spelling and reading disability than members of the general population. For spelling and reading disability, in which the prevalence rate is higher in males than females, females must have a higher threshold. One consequence would be that females on average must have a greater predisposition (e.g. genetic predisposition) than affected males for becoming reading and spelling disabled. A further consequence would be that the rate of spelling and reading disabled relatives of affected girls should be higher than the rate of spelling and reading disabled relatives of affected boys. The empirical data of DeFries and Decker (1982) and DeFries (1989) gave some support for the polygenic threshold model. They found a higher rate of affected

Requests for reprints to: Dr Gerd Schulte-Körne, Department of Child and Adolescent Psychiatry, Philipps-University Marburg, Hans-Sachs-Straße 6, D-35039 Marburg, Germany.

first-degree relatives of reading disabled girls than of reading disabled boys. Further support for a sex-influenced genetic model came from the reported family studies. The rate of reading disability for parents of male probands was 35%, and 41% for female probands (Gilger et al., 1991).

The results of family studies indicate a high familiarity and probable heritability of reading disorder. However, for spelling disability, little research has been done to examine the familiarity of this disorder. Using family history information, Naidoo (1972) found significantly more spelling disabled first-degree relatives of spelling disabled probands than of non-spelling disabled probands. Wolff and Melngailis (1994) examined the reading and spelling disability of first and second degree relatives of spelling and reading disabled probands. This study suggests that these relatives scored significantly lower on spelling than reading tests. Further support for a genetic aetiology for spelling disorder came from twin studies. A large-scale London twin study found a high heritability of $h^2g = .75$ for spelling disability after controlling for the individual differences in IQ, and a moderate heritability for reading disability (Stevenson, Graham, Fredman, & McLoughlin, 1987). The high rate of heritability for spelling ability was confirmed by DeFries, Stevenson, Gillis, and Wadsworth (1991), who found a heritability of $h^2g = .62$ for spelling in the Colorado Twin Study. The authors concluded that the similar high heritability estimates obtained in the two independent samples strengthen the evidence for a substantial genetic aetiology of spelling deficits.

One major interest of the present study was to examine the incidence of spelling disorder in first-degree relatives (parents and sibs) of spelling disabled probands. The results of family studies could be mainly influenced by different diagnostic criteria as was pointed out for twin studies by Pennington, Gilger, Olson, & DeFries (1992). The diagnostic procedures varied from interviews and questionnaires on the one hand to psychometric tests on the other hand, the former being preferred for adults in most family studies (Pennington et al., 1991; Gilger et al., 1991). There is also ongoing discussion about whether to use a low achievement or IQ discrepancy as criterion for reading disability (Rutter & Yule, 1975; Rispens & Van Yperen, 1990; Pennington et al., 1992; Shaywitz, Fletcher, Holahan, & Shaywitz, 1992; Stanovich, 1994). The question is whether there are specific characteristics of the IQ-discrepant or low achievement defined groups. Rutter and Yule (1975) differentiate between a specific reading retarded group, in which reading achievement was significantly below the expected level on the basis of IQ and age, and a low achievement group (reading backwardness), whose reading achievement was below age. The specific reading retarded group had more affected males than females, fewer neurological abnormalities, and more abnormalities in speech and language development in comparison to the reading backward group. However, Rutter and Yule did not find differences between the two groups regarding the percentage of reading disabled first-degree relatives. A large number of studies examined whether there were differences between two distinctive subgroups, an IQ-discrepant and an achievement discrepant-defined reading disabled group (Aaron, 1987; Shaywitz et al., 1992; Pennington et al., 1992; Fletcher et al., 1994). The results were equivocal. Some authors found small but statistically significant differences between the two groups (Aaron, 1987; Silva, McGee, & Williams, 1985; Jorm, Share, Maclean, & Matthews, 1986; Das, Mensink, & Mishra, 1990; Pennington et al., 1992), whereas others did not find group differences regarding neuropsychological test profiles and cognitive functions (Siegel, 1992; Shaywitz et al., 1992; Fletcher et al., 1994). These two criteria might define different subgroups of dyslexia for which different aetiologies can be assumed. Therefore, we are interested in finding out the influence of adopting the IQ-discrepant and low achievement criteria on the rate of spelling disability in first-degree relatives of spelling disabled probands.

Materials and Methods

Sample

The probands were selected from our outpatient clinic at the Department of Child and Adolescent Psychiatry of the Marburg University. All probands consulting the clinic for the first time and having a history of learning disability underwent our diagnostic procedure containing the non-verbal Culture Fair Intelligence Test (CFT) (Weiß, 1987; Weiß & Osterland, 1977), and a grade-appropriate German spelling test (Brickenkamp, 1975). The CFT was chosen for sample selection in order to reduce the influence of verbal abilities and cultural and educational influences on IQ test performance. Inclusion criteria were a spelling ability below the tenth percentile (low achievement criterion) and below IQ (IQ-discrepancy regression criterion: z value of difference between expected and actual spelling achievement >1).

Exclusion criteria were an IQ < 85, an uncorrected auditory and/or visual acuity, apparent neurological, emotional or behavioural disorder or unusual educational circumstances that could account for the poor spelling ability.

From October 1993 to July 1994, 50 patients having a history of reading and spelling disability attended our outpatient clinic. From this sample 38 children fulfilled the inclusion criteria of our study (about 10% of all patients). Six probands and their first-degree relatives refused to participate in the study. The remaining 32 probands were tested in our clinic, sibs and parents were mainly tested at home by specially trained staff.

Measures

The test battery for the family members consisted of:

(i) Culture Fair Intelligence Test (CFT1 or CFT20, depending on age)
(ii) Grade-appropriate spelling tests (Brickenkamp, 1975)
(iii) Grade-appropriate reading comprehension tests (for children up to the 9th grade)

Additionally, each subject answered a questionnaire about their reading and spelling history and habits, also their self-assessment of spelling and reading skills as well as their attitudes towards reading and spelling.

The spelling tests required the spelling of 30–40 words with specific difficulties regarding the German spelling rules and German language (Brickenkamp, 1975). The total number of spelling errors (in maximum, one error for one word was counted) formed the raw scores which were then transformed

Table 1
Sample Characteristics (Age and IQ), Means ± SD

		Parents		Sibs	
	Probands	Mothers	Fathers	Sisters	Brothers
N	32	31	26	12	14
Age	11.2±2.4	36.7±4.5	39.6±5.4	12.3±3.4	11.9±3.1
IQ	98.3±8.1	109.9±14.3	103.5±14.6	104.7±10.7	101.9±11.7
Spelling	2.9±2.3	48.7±25.0	33.3±30.5	38.1±31.7	23.1±27.0
Reading	33.0±25.6			57.7±33.4	74.6±18.0

to grade adjusted percentage ranks according to large normative samples.

The reading comprehension test required the silent reading of series of short passages. Subsequently, the child had to answer several multiple choice questions concerning the contents of the passages. The total number of correct answers formed the raw scores which were then transformed to grade adjusted percentage ranks in the same manner as the spelling scores. Reading comprehension tests including norms were only available for children up to the 9th grade. Furthermore, their validity is restricted due to the 30-year-old norms. Therefore, reading comprehension tests were used as a screening instrument.

The socio-economic status (SES) was rated on the basis of the father's profession and his economic success (Kleining & Moore, 1968). The original five different classes were reduced to three classes (lower, middle, and upper).

Methods of Statistical Analysis

In order to eliminate a possible bias due to heterogeneous variances of the mean rates, for each analysis (parents and sibs) we restricted the sample to the largest homogeneous subsample with respect to that group, i.e. parental affectedness was analysed for the 25 (out of 32) families with both parents available. In four of the families, no sib could be tested. Therefore, affectedness of sibs was tested using the remaining 21 families. From the two of the 21 families with more than one sib, we used only the data of the eldest sib, again to avoid heterogeneous variances of the mean rates.

Evaluation of observed rates of affected family members requires the knowledge of expected rates of affectedness under the assumption of no familial aggregation. Firstly, we had to define affectedness. For the achievement criterion, we chose a *T*-value* of 40 as cut-off, i.e. mean $-1\,SD$, leading to an expected rate of 15.9%. For the IQ-discrepancy criterion, we selected the corresponding (i.e. leading to the same expected rate) cut-off for the regression residuals. Glogauer (1977) reported a correlation between IQ and spelling of .42 for a large normative German sample. We used this coefficient to calculate the regression equation.†

Secondly, we had to calculate the observed rates. While the observed rate of affected sibs is simply the number of affected in relation to the total number of sibs, the observed rate of affected parents had to be calculated differently. We calculated the rate of affected parents within each family. The mean of these familial rates was tested against the expected rate as defined above. The expected value of this mean is not influenced by intra-familial dependencies of the spouses which could have been caused by possible assortative mating.

For testing the observed against the expected rates, we used the binomial test for the sibs and the one-sample *t*-test for the parents. The latter was chosen because it accounts for the effect of possible assortative mating on the standard deviation of the mean rate. For this purpose, the *t*-test is more appropriate than non-parametric tests or the binomial test, which would require binomially distributed data.

Results

Sample

Our sample consists of 32 probands, 21 boys and 11 girls. Analysing the reading test data showed that 10 of the probands fulfilled the IQ-discrepancy criterion (two probands refused to perform the reading test). Presuming the reading comprehension test to be valid despite the 30-year-old norms, this means that 33.3% of our probands have a combined spelling and reading disability (IQ-discrepancy criterion), and the rest are purely spelling disabled. Concerning the 13 affected sibs, we found only one to be reading and spelling disabled (IQ-discrepancy criterion); the other 12 sibs were purely spelling disabled.

Because of the small sample size, separate analyses for the groups of pure spelling disabled, and for the spelling and reading disabled probands, were not carried out. In 25 families both parents could be examined, in seven cases only one parent. In the majority of our families ($N = 19$) there was one school-age sib that could be examined. Additionally, there was one family with two, one family with three, and nine families without school-age sibs. Concerning SES, 18 families were rated lower class, 12 middle class, and two upper class. The sample characteristics are presented in Table 1.

Comparison of Expected and Empirical Rates of Affected First-Degree Relatives

Table 2 shows the rates of affected parents and sibs comparing the two different criteria.

Both criteria define a higher rate of affected subjects as would be expected on the assumption of no familial aggregation for both parents and sibs. We found a significantly higher rate of affected sibs, irrespective of the diagnostic criteria used (one-tailed *p*-values, not adjusted for multiple testing). However, the empirical rates of affected parents were only significantly different from the expected rates when the IQ-discrepancy

*Refers to *T*-norm with mean = 50 and $SD = 10$.

†The *SD* of the regression residual (*T*-norm) is: $10^* \sqrt{(1 - 0.42^2)} = 9.1$. The underlying regression equation is: spelling (*T*-norm) = 0.42*(10/15)*(IQ-100) + residual.

Table 2
Empirical and Expected Rates of Parents and Sibs for Two Diagnostic Criteria

	N	Observed	Expected	Test statistic	*p*-Value (one-sided)
IQ-discrepancy criterion					
Parents	25	34.0%[a]	15.9%	2.42	.0118
Sibs	21	61.9%	15.9%	13/21	< .0001
Achievement criterion					
Parents	25	26.0%[b]	15.9%	1.41	.0850
Sibs	21	52.3%	15.9%	11/21	< .0001

[a]In four families both of the parents were affected, in nine families only one parent, and in 12 families none of the parents were affected.
[b]In three families both of the parents were affected, in seven families only one parent, and in 15 families none of the parents were affected.

Table 3
Mean Rates (%) of Affected First-Degree Relatives of Male and Female Probands

Mean Rates of Affected First-degree Relatives of:	
Male probands (12 families)	Female probands (7 families)
33.3	42.9

criterion was considered. Regarding the two different diagnostic criteria, only little evidence was found for an influence of the chosen diagnostic criterion on the observed rate of affectedness. To check if the two criteria define different subgroups of spelling disabled, cross-tabulations were carried out. All parents and sibs rated affected by the achievement criterion were rated affected, too, when the IQ-discrepancy criterion was used. That means individuals with low achievement are mainly a subgroup of those with a high difference between actual and predicted spelling based on IQ.

The Influence of Compensation on the Parents' Rate of Affectedness

Adults could have improved their spelling ability by remediation. This so-called compensation effect should be considered. For this purpose we analysed the questionnaires regarding history of spelling achievement and were able to identify a number of adults we assumed to be compensated. We regarded adults as compensated if they reported spelling difficulties at school, but did not fulfil the regression or achievement criterion. This procedure is similar to the one used by Pennington et al. (1991) for English-speaking, reading disabled adults. On the basis of these data, we found 10 adults who reported spelling difficulties during school, but having an actual spelling ability within the normal range. Including both the 17 (34%) affected adults and the 10 (20%) compensated adults, we observed a total rate of affected adults of 54%.

Sex-Influenced Polygenic Threshold Model

A specific hypothesis that can be derived from the polygenic threshold model is that relatives of affected girls should be more often affected than relatives of affected boys. The rates of affected first-degree relatives (parents and sibs considered together) were calculated separately for male and female probands within each family. This analysis was based on families with both parents and one sib (19 families). The IQ-discrepancy criterion was chosen to define affectedness. The resulting familial rates were first transformed using the arcsin transformation (Sachs, 1973). These values underwent statistical testing (male vs female) using an exact permutation test. Table 3 shows the mean rates for male and female probands.

In the families of female probands we found a higher rate of spelling disabled first-degree relatives than in families of male probands. However, these rates did not differ significantly.

Another observation that could be best explained by a polygenic model is the relation between the number of affected parents and the affectedness of children. The risk of being affected increases with the number of affected parents (none, one, or both). Each affected parent additionally transmits a number of 'disease genes' which increases heritability. In the study we found no relation between the number of affected parents (both, one or none affected) and the affectedness of probands' sibs.

Discussion

One of the major purposes of our study was to examine the familial aggregation of spelling disability with special consideration of two different diagnostic criteria. We examined 32 probands and their 83 first-degree relatives and found significantly increased rates of affected sibs (low-achievement and IQ-discrepancy criteria) and of affected parents (IQ-discrepancy criterion). The rates defined by the achievement criterion were lower for parents and sibs. Nearly all subjects rated as affected using the achievement criterion had the same rating using the IQ-discrepancy criterion, i.e. the IQ-discrepancy criterion defined a slightly larger, but not substantially different group of individuals. We found no evidence for the hypothesis that the IQ-discrepancy and low achievement criteria define different subtypes of spelling disability regarding the familial aggregation of

the disorder. In our sample the risk of a first-degree relative being affected using the IQ-discrepancy criterion was 34–61%. This calculated risk is comparable to the risk estimated by Hallgren (1950), Zahalkova et al. (1972), Vogler et al. (1985) and Gilger et al. (1991). In all of the reported studies the estimated risks were significantly higher compared with control groups or the population risk. Results regarding estimated risks did not seem to be considerably affected by the influence of different languages, i.e. Swedish (41% for sibs, 42.4% for parents, Hallgren, 1950), Czechoslovakian (34% for sibs, 33.5% for parents, Zahalkova et al., 1972), German (our study) and English (28.5% for sibs and 27% for parents, Gilger et al., 1991).

Another relevant aspect is that the affected first-degree relatives from the reported family studies were mainly reading disabled. In our study we found a comparably high rate of spelling disabled relatives. Although we did not examine reading ability in the parents' sample, it remains open as to whether both disorders, reading and/or spelling disability segregate independently in families. Therefore, it is possible that there are pure spelling disabled families and another subgroup of families characterized by reading and spelling or pure reading disability. The high heritability estimates for spelling disability reported by Stevenson et al. (1987) and by DeFries et al. (1991) gave further support for a genetic aetiology in spelling disability. We recommend the separate examining of reading and spelling abilities in families in order to gain an impression about a common or different segregation of these abilities.

The sex ratio of probands was 1.9:1 (male:female). This is below the traditionally reported 4:1 for clinically ascertained samples. This ratio is similar to the reported sex prevalence reported by Shaywitz, Shaywitz, Fletcher, and Escobar (1990) for non-clinical sample and gives evidence for reduced bias in our ascertainment strategy by sampling probands from an outpatient clinic. Further, the gender ratio among affected relatives was 2.4:1 and suggests the hypothesis of different aetiology of spelling disability in males and females. This hypothesis was supported by the analyses of Alarcón, DeFries, and Fulker (1995), which provided evidence for different etiologies in male and female reading performance. For spelling disability, Allred (1990) showed that girls spell significantly better than boys at all grade levels (1–6).

The great influence of spelling disability in a child's development is emphasized by our findings pertaining to 54% of affected adults including compensated. As Michelsson, Byring, and Björkgren (1985) have shown in a Swedish adult sample and Strehlow, Kluge, Möller, and Haffner (1992) for a German sample, the spelling disability of dyslexic children often persists into adulthood and is the main influence on the occupational status and social and emotional development of the affected person. The adults' spelling disability could have influenced the child's phenotype in different ways.

In the ongoing study, different samples from four regions of Germany will be examined, and a segregation analysis of these data will give more exact information about the mode of inheritance of spelling disability.

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