ARTICLE

High acceptance of an early dyslexia screening test involving genetic analyses in Germany

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Dyslexia is a developmental disorder characterized by severe problems in the acquisition of reading and writing skills. It has a strong neurobiological basis. Genetic influence is estimated at 50–70%. One of the central problems with dyslexia is its late diagnosis, normally not before the end of the 2nd grade, resulting in the loss of several years for early therapy. Currently, research is focusing on the development of early tests for dyslexia, which may be based on EEG and genetics. Our aim was to determine the acceptance of such a future test among parents. We conducted a representative survey in Germany with 1000 parents of children aged 3–7 years, with and without experience of dyslexia. 88.7% of the parents supported the introduction of an early test for dyslexia based on EEG and genetics; 82.8% would have their own children tested, and 57.9% were willing to pay for the test if health insurance did not cover the costs. Test acceptance was significantly higher if parents had prior experience with dyslexia. The perceived benefits of such a test were early recognition and remediation and, preventing deficits. Concerns regarded the precision of the test, its potentially stigmatizing effect and its costs. The high overall support for the test leads to the conclusion that parents would accept a test for dyslexia based on EEG and genetics.

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INTRODUCTION

Dyslexia, a severe disorder in the acquisition of reading and writing skills, is one of the most common childhood disorders. Its prevalence varies depending on the transparency of the studied language,^{1,2} with a prevalence estimated at ~5% in Germany.³ One major problem related to dyslexia is its late diagnosis. Since current diagnosis is based on psychometric testing, a reliable assessment of dyslexia can only be achieved after the child has acquired sufficient reading and writing skills, normally at the end of 2nd grade in Germany. At this point, language development has already progressed significantly. Therefore, therapeutic interventions are commonly conducted at a very late stage. Although psychometrically based diagnostic tools to identify children at risk for dyslexia before school enrollment exist in Germany,^{4,5} and other countries,⁶ these lack appropriate robustness, sensitivity, and specificity.⁷ This discloses two eminent problems of diagnostic procedures for dyslexia: (i) at best,⁵ they start <1 year before school enrollment and (ii) their predictive quality is not sufficient.

Consequently, current research aims to assist in filling this diagnostic gap (details can be found at <http://www.legascreen.de>) by considering additional pathophysiological aspects of dyslexia. Indeed, neurobiological mechanisms seem to have an important role for dyslexia, and certain EEG signatures show differences between children at risk for dyslexia and children with normal reading and writing skills already shortly after birth.^{8,9} Accordingly, the disorder

has a strong genetic component with heritability ranging between 50–70%.¹⁰ These aspects may be utilized diagnostically to overcome limitations of purely psychometric diagnostic tools and thus, could provide a thorough risk assessment at much earlier stages. This may ultimately allow therapeutic interventions before school enrollment.

Despite the potential benefits of such an approach, it is not clear if a risk estimation of developing dyslexia based on genetics and EEG would be accepted among parents. To address this question, we conducted a representative survey among German parents.

MATERIALS AND METHODS

Participants

Thousand parents of children between 3 and 7 years old were given a set of 11 closed and 2 open questions. The questions regarded the attitude toward an early test for assessing the risk of developing dyslexia based on genetics and EEG as well as potential advantages and disadvantages (see Supplementary Methods). The age range of the parent's children corresponded to the age range a potential test would address. Questions concerning the personal experience with dyslexia, for example, affected children or relatives were included to obtain information about the potential differences between parents with/without such prior experience. Furthermore, questions regarding their sociodemographic status, federal state of residence, and educational level were added. Due to language specificity of dyslexia, only individuals with German as their mother tongue were included, with an equal percentage of male and female participants.

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Survey characteristics

The survey was done by the survey research company Lightspeed Research GmbH (Hamburg, Germany) according to the BVM ('Bundesverband Deutscher Markt- und Sozialforscher e.v.'): that is, 'Federal association of German market and social researchers') standards. An actively managed online access panel was used. Participants made a conscious decision to participate in the online survey through a double opt-in registration process. Quotas to ensure representativeness for all German federal states were based on official statistics by the German Federal Bureau of Statistics (Statistisches Bundesamt), including place of residence (according to the populations of the German federal states), income, and education of the parents. In order to reduce a potential bias toward persons answering without carefully reading the questions and giving illogical answers, a minimum completion time of 100 s (time between appearance of first question and last answer in the questionnaire) was set. Furthermore, a set of logical filters was applied to avoid accidental mistakes (Supplementary Methods). Participants dropping below the minimal survey completion time were excluded. In cases of apparent violations of logic, participants were asked to check the respective answer. The survey was closed after 1000 data sets had been obtained, fulfilling all the above-mentioned quotas and criteria.

Since the study was based on an online panel, reality checkpoints, such as verifying IP, email and postal address, were included to ensure that the panel consisted of real people.

For the open questions regarding the advantages and disadvantages of the test, content categories were created, which were defined by the number of similar answers, that is, repetitions. All answers with a total number of repetitions below 10 were grouped in the category 'Other'.

Statistics

Statistical analysis was conducted by applying standard procedures of data evaluation and by using the PSP software (Free Software Foundation Inc, Boston, MA, USA) for calculation. Differences between the groups were calculated using χ^2 statistics and *t*-tests. In case of non-normally distributed data, a Mann–Whitney *U*-test was performed. According to standard definition, significance levels are indicated as $P \leq 0.05$, $P \leq 0.01$, and $P \leq 0.001$. Non-significant differences were described as trends if $0.05 < P < 0.1$. Given a sample size of 1000, 777, and 223 respondents and $\alpha \leq 0.05$, we had a power of 80% to detect a difference of 6.2%, 7.1%, and 13.1% compared with a proportion of 50% with statistical significance.

RESULTS

Study population

Mean age $\pm$ SD of the participating parents was 37 ± 7.25 years. Their average number of children aged 3–7 years old was 1.25, being very close to the general birth rate of 1.36 in Germany.¹¹ 27.7% had graduated from university or a university of applied science. 21.7% had finished grammar school (Gymnasium, 12–13 years, depending on federal state) without subsequent university education. 41.7% had visited middle school (Realschule, 10 years), while 9% of the parents received only secondary education (Hauptschule, ~8 years of schooling). 5.4% reported to be dyslexic, which is close to the disorder's

estimated prevalence of 5% in Germany,³ and 22.3% reported to have a child or relatives with dyslexia.

General acceptance and financial aspects

The main questions for general acceptance was if the test is considered useful, and if parents would have their own children tested. Here we found high levels of endorsement among all the groups (with and without former dyslexia experience), ranging from 80.1 to 93.3%. Willingness to pay for the test was lower, but did not fall below 52.6% in any group (Table 1). We assessed factors potentially affecting decisions by checking for former dyslexia experience, level of education, sex, and federal state of residence. Among all the three above-mentioned questions, acceptance was higher if participants had former experience with dyslexia. Individuals with such former experience showed higher general acceptance ($P < 0.05$), higher willingness to have their children tested ($P < 0.001$), and higher acceptance to have their children tested at own expense ($P < 0.001$).

Test acceptance among all levels of education was very high. The level of education had a role nonetheless as parents with the lowest level of education showed the lowest level of acceptance among all the questions. Differences in the distribution of school types reached significance concerning the question of whether parents would have their own child tested at their own expense, that is, individuals with higher education agreed significantly more often ($P \leq 0.01$; Figure 1a; Supplementary Table 1). Among all the three questions, there were no significant sex differences and no interaction between sex and experience.

Furthermore, we analyzed potential regional influences on acceptance by looking at the regional distribution between Germany's 16 separate federal states. The rationale was that the former division of Germany, which most parents may have actively experienced (average age at unification: 14 years old), might still shape different attitudes. Results showed differences between federal states (Figure 1b), but their impact on test acceptance was only marginal and not statistically significant.

Financial aspects represent an important point of test acceptance. As in other countries, not all available tests are covered by health insurance in Germany. Generally, participants with higher income showed a trend ($P < 0.1$) toward a higher acceptance in all the three questions. Higher acceptance was statistically significant if asked about testing their own child ($P \leq 0.05$, Supplementary Table 2). All participants agreeing to have their own children tested were asked about the fee they were willing to pay, if their health insurance did not cover the costs (Figure 2). Participants with former dyslexia experience showed a higher willingness to spend larger sums ($P \leq 0.001$), but the absolute majority of parents preferred overall test costs below €150.

Table 1 Test acceptance

Question	All (n = 1000)	Former experiences with dyslexia (n = 223)	No former experiences with dyslexia (n = 777)
Is an early test for dyslexia, based on genetics and EEG, generally useful?	Yes: n = 887 88.7% (86.5–90.6%)	Yes: n = 208 93.3% (88.9–96.1%)	Yes: n = 679 87.4% (84.8–89.6%)
Would you have your own child tested?	Yes: n = 828 82.8% (80.3–85.1%)	Yes: n = 206 92.4% (87.9–95.4%)	Yes: n = 622 80.1% (77.0–82.8%)
Would you have your own child tested if health insurance does not cover the costs?	Yes: n = 579 57.9% (54.8–61%)	Yes: n = 170 76.2% (70–81.5%)	Yes: n = 409 52.6% (49.1–56.2%)

Provides the results of the survey's closed questions, contrasted between parents with and without former dyslexia experience, in absolute and numbers and percentages. Percentage ranges in brackets represent the 95% confidence interval.

Figure 1 Distribution of acceptance in relation to level of education and region. (a) Acceptance and education: Green indicates agreement. Red indicates disagreement. The level of education is marked by shades. (b) Regional distribution of acceptance. General acceptance level (left), acceptance to have own child tested (middle), and acceptance to have own child tested at own expense (right) are shown. Maps were taken from www.stepmap.de and edited.

Notion regarding test advantages and disadvantages, and required support after diagnosis

Arguments mentioned in the open question about advantages of testing could be grouped in eight categories, the frequency of which is presented in Figure 3a. The category 'precision of test' was mentioned 48 times. All other categories were mentioned more often. The major advantage of a potential genetics/EEG test for the early diagnosis of the risk for dyslexia was seen in the possibility of early remediation, followed by the viable prevention of deficits, and early recognition itself (Figure 3a and Supplementary Table 3). The only significant difference between parents with and without previous dyslexia experience was found in the category 'Improved understanding for children', which was more frequently mentioned by people with former experience (10.8% vs 5.3%, $P \leq 0.05$).

For disadvantages, 12 categories were defined. The category 'diagnosis as excuse', that is, dyslexia as an apology for laziness, was mentioned 14 times. All other categories were mentioned more often (Figure 3b and Supplementary Table 4). The major disadvantages seen for the test were concerns of its potential imprecision, the concern that the diagnosis could stigmatize the children, and the testing costs. Despite the comparatively wide range of concerns, 303 (30.3%) persons saw no disadvantages at all, compared with only 6.9% who could see no advantages. The percentage of parents who did not answer the question for disadvantages or did not know an answer is 24.3%, while only 16.5% did not answer the question for advantages. This suggests an overall positive reception of the test's benefits. In the areas of 'Over-reaction/Abundance of support', 'Data security', and 'Diagnosis as an excuse', people with former dyslexia experiences showed nominally higher concerns than people without such

Figure 2 Acceptable test fees among parents with and without former experience with dyslexia. The graph shows the sum that people with (black) and without (gray) former dyslexia experience would be willing to pay for a test.

experience, without the difference reaching significance but showing a statistical trend for 'Over-reaction/Abundance of support' (7.2% vs 4.1%, $P < 0.1$).

Beside the actual acceptance of the test, we were interested in the parents' opinion on the subsequent support they would like to receive

beyond test result reports. Given the option to choose between different kinds of support together with the test results, the overwhelming majority of parents (87.4%) was interested in being advised on home support options for their children and/or recommendations of professional therapists in their vicinity (Figure 4). There were no significant differences between parents with and without former experience with dyslexia.

DISCUSSION

Our research aim was to study the attitudes toward an early screening test for dyslexia, based on genetics and EEG, currently under development. We asked a representative sample of 1000 parents with and without experience with dyslexia about different test aspects.

The survey, conducted using an online panel, was comparable in terms of representativeness with traditional telephone surveys: the percentage of the German population over 14 years old using internet in the year of the survey (2013) is 76.5%,¹² while 86% have landline access in their home,¹³ which can be regarded a similar level. The tendency of older people preferring telephone surveys¹⁴ was not of relevance in this survey, because our focus was on parents of young children. Nevertheless, only parents with internet access could be studied in our survey. All other criteria were covered by initial quota based on German federal statistics, avoiding the selection bias commonly found in open web-based surveys.¹⁵

We found very high acceptance for an early test assessing the risk of developing dyslexia based on genetic and EEG markers. This indicates a general positive attitude toward early diagnostic testing and prevention of dyslexia among German parents, especially among those

with previous experience. These results are in accordance with studies from other countries indicating a positive^{16–18} or an increasingly positive attitude¹⁹ toward genetic testing in general and toward genetic testing in a variety of specific diseases.^{20–22}

The reasons for our finding that parents with former experience with dyslexia are more open to early diagnostic testing and also more willing to cover test-related costs cannot be conclusively answered with this survey. Personal experience with consequences of undiagnosed or late diagnosed dyslexia and a subsequent willingness to prevent such consequences for one's own children may have contributed. Furthermore, these parents may have a higher awareness of the biological component of dyslexia and the need for early diagnosis and remediation. Our findings are in contrast to other studies, where affected people showed less interest in genetic testing than non-affected ones.²³ This may be explained by the fact that in those studies adults were asked about their attitude toward a test for themselves and not parents about having their children tested. Moreover, the disease referred to is a severe condition (malign breast cancer) with encumbering therapies sometimes required and an uncertain prognosis. In turn, the positive attitude toward predictive genetic testing in our study may be linked to the availability of prophylactic or curative interventions, which exist in dyslexia but not in all other diseases. Both factors, that is, the possibility to have their children tested combined with the availability of intervention, might explain the high support for this test among parents.

Despite a very high test acceptance among all levels of education, we found the lowest level of acceptance in the least educated participants of our survey. Our sample contained only 9% of people with lower secondary education, compared with 21.9% in the general population. Therefore, we recalculated significance weighted for strata corresponding to the education levels of the general population (German Federal Statistical Office, 2013). Again, acceptance was lowest in the least educated participants, reaching significance within all questions of general acceptance. Although is in accordance with findings of other research on genetic testing, suggesting a positive correlation between educational level and interest in predictive testing,^{24,25} general acceptance rates were still notably high even among less educated survey participants.

The more divergent distribution pattern concerning answers for disadvantages than for advantages might reflect that the expected positive effects of such a test seem to be more obvious than the negative ones. Nevertheless, it should not be underestimated that the majority of parents saw no disadvantages or did not answer the question. The parents' concerns may guide improvements of an early predictive test: important issues include (a) a need for sufficient information/support delivered

Figure 3 Potential advantages and disadvantages of the test. Advantages (a) and disadvantages (b) of the test as seen by people with (black) and without (gray) former dyslexia experience. All answers with fewer than 10 repetitions were categorized as 'Other'.

Figure 4 Notion about required advice. The distribution of wishes among people with (black) and without (gray) former dyslexia experience for different kinds of support is shown.

together with the test results, (b) that a future test should be more precise than the psychometric tests currently available, (c) that exclusion or stigmatization of children should be prevented by enabling early support, and (d) to have a low to moderate price. Importantly, detailed information concerning support options such as contact addresses of local language therapists should be provided.

Future work should include similar research among the different European countries. Especially in the case of dyslexia, where language-dependent factors have an important role, such studies could provide further insight into regional differences.

In summary, the present study investigated the opinion toward early testing of dyslexia based on biological markers in a representative German sample of parents. There was significant support for introducing such a test and a positive attitude toward having their own children tested, even at their own expense, suggesting acceptance and demand once such a test is available.

CONFLICT OF INTEREST

All contributing authors are members of the LEGASCREEN consortium. The aim of this consortium is to develop of an early screening test for dyslexia based on biological markers. AW, JB, FE and HK hold a patent concerning the genetic diagnosis of dyslexia.

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MEMBERS OF THE LEGASCREEN CONSORTIUM

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