

# Economic evaluation of dyslexia intervention

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In many countries, intervention costs are not covered by public health care. A critical basis for deciding whether an intervention is covered or not is to analyse the relation between benefits and costs of the intervention, and to quantify the consequential costs. In this study, a cost-utility analysis was computed to investigate the costs of individualized dyslexia intervention while quantifying the benefit in terms of health-related quality of life in a sample of 36 individuals with dyslexia. In addition, educational outcomes and costs of untreated dyslexia for the society were estimated using information for class repetition, school success, and unemployment rates from previous studies and official statistics. A significant increase in quality of life with medium effect sizes was found across all quality-of-life measures. Increases in quality of life were domain-specific, thus occurring specifically in those domains that are affected by learning disorders. The cost-utility ratio was 9,782 Euros per quality adjusted life years (QALYs), which is in line with similar therapy forms, such as speech therapy. The loss of productivity for untreated dyslexia in the German population was estimated for class repetition and reduced income due to lower school degrees. The cost-utility analysis and the calculation of consequential costs suggest that the dyslexia intervention is cost-effective.

Kristina Moll and Beatrice J. Georgii shared first authorship.

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**KEYWORDS**

cost-utility analysis, dyslexia, intervention benefits, intervention costs, quality of life

**Practitioner Points**

- Dyslexia intervention increases quality of life across different measurements.
- Improvements were found for physical and emotional well-being, self-esteem, and in daily activities in different settings (family, friends, and school).
- Cost-utility analyses and the calculation of consequential costs suggest that dyslexia intervention is cost-effective.
- Covering intervention costs would induce social equality and cohesion, and at the same time reduce loss of productivity.

## 1 | INTRODUCTION

Dyslexia is characterized by difficulties in word reading (i.e., deficits in reading accuracy, fluency and/or reading comprehension) and spelling, and is one of the most prevalent neurodevelopmental disorders, affecting 4–9% of the population worldwide (Rutter et al., 2004; Shaywitz, Shaywitz, Fletcher, & Escobar, 1990; for overview see, Hasselhorn & Schuchardt, 2006). Dyslexia is a heritable and life-long condition, and is supposed to affect brain development and cognitive processing (i.e., phonological processing). Dyslexia is listed as specific learning disorder in standard diagnostic manuals (DSM-5 and ICD-10: American Psychiatric Association, 2013; World Health Organization, 1992), and by national health systems (e.g., by the NHS in the United Kingdom).

In addition to severe problems in literacy skills, children with dyslexia are also more frequently affected by co-occurring mental disorders like anxiety, depression, and ADHD, compared to children without dyslexia (Arnold et al., 2005; Esser, Wyschkon, & Schmidt, 2002; Maughan, Rowe, Loeber, & Stouthamer-Loeber, 2003; Visser et al., 2020). Without adequate support and intervention, these problems negatively affect social and academic development. Common problems are school anxiety, refusing to go to school, or even dropping out of school (Daniel et al., 2006). As a result, individuals with dyslexia often receive lower school degrees and are more often unemployed than individuals without dyslexia, despite comparable levels of intelligence (Esser et al., 2002; Schulte-Körne, 2007). From a socio-political viewpoint, the consequence will likely be a loss of productivity for the society. Indeed, the few existing studies, which assessed educational outcomes, such as college dropouts and loss of productivity due to dyslexia, suggest a negative prognosis regarding professional life and mental development (Carroll, Maughan, Goodman, & Meltzer, 2005; Daniel et al., 2006; Esser et al., 2002; Esser & Schmidt, 1993), inducing high costs to education and health systems (Bynner & Parsons, 2006).

In order to foster successful education and psychological well-being and to decrease the risk of co-occurring mental disorders for individuals with dyslexia, it is important to provide adequate support and intervention as early as possible. This is especially important for children with more severe reading and spelling difficulties, where intensified instruction at school does not improve literacy skills. In these cases, one-on-one training lessons with specialized therapists are needed. Ideally, individual training lessons should be provided in close collaboration with the school-teacher so that transfer into everyday life at school is guaranteed. Effective dyslexia interventions show significant improvements with regard to literacy development (Galuschka, Ise, Krick, & Schulte-Körne, 2014; Goodwin & Ahn, 2010; Ise & Schulte-Körne, 2010; Schulte-Körne, Deimel, Jungermann, & Remschmidt, 2003), as well as

psychosocial well-being (Beer, 2013; Giovagnoli et al., 2020; Schulte-Körne et al., 2003). Meta-analyses suggest that an effective intervention should be symptom-specific, thus targeting directly the affected literacy skills (Galuschka et al., 2014, 2020). With respect to the duration of the intervention, Ise, Engel, & Schulte-Körne (2012) showed in a meta-analysis that intervention effects differ depending on the duration of the intervention. The effect size of the reading intervention effect was only reliable for studies with at least 20 weeks of intervention, suggesting that a minimum of 20 weeks of individual symptom-specific intervention is necessary for intervention effects to emerge.

Countries differ in how exactly interventions for children with dyslexia are implemented in or outside school. What is common across countries is that access to diagnostic assessments based on ICD-10 or DSM-5, including the diagnosis of frequently co-occurring disorders (e.g., ADHD, anxiety, or depression), as well as individual training is restricted and/or cost-intensive. In some countries, *diagnostic costs* are at least partly covered by the educational, health care or child and youth act systems (e.g., in Germany, Austria, and The Netherlands). However, *costs for individualized dyslexia interventions* are in most countries not covered at all (e.g., Germany, Austria, United Kingdom; for an exception see The Netherlands, where costs are covered by the Child and Youth Act), or only covered when psychosocial problems co-occur (e.g., in Germany and Austria).

In Germany, there are different institutions, such as child and adolescent psychiatrists, public educational and counselling services, and the social service of the public youth welfare, which provide support, professional advice, and diagnostic assessments for individuals with dyslexia (Schulte-Körne, 2009). In addition, psychological services at school offer assessments of reading and spelling skills for children with poor literacy skills, and support children with dyslexia in the classroom, in form of extra lessons, and in form of compensation for disadvantages (such as extra time). Although schools support children with dyslexia, they do not offer individualized dyslexia therapy. In Germany and in many other countries, individualized dyslexia therapy is mainly offered by specialized dyslexia therapists outside of school. In Germany, dyslexia therapists undergo an intensive professional training (usually over 2 years), which is certified by the German Dyslexia and Dyscalculia Association (Bundesverband Legasthenie & Dyskalkulie e.V. [BVL]: <https://www.bvl-legasthenie.de/>). This certificate stands for a high-quality education as dyslexia therapist. It guarantees high standard intervention that follows the clinical guidelines for diagnosis and intervention of dyslexia, which have been developed based on research evidence (Galuschka & Schulte-Körne, 2016; S3-Leitlinie Diagnostik und Behandlung bei der Lese- und/oder Rechtschreibstörung [Clinical guideline for diagnosis and intervention of dyslexia]: <https://www.awmf.org/leitlinien/detail/II/028-044.html>).

To summarize, dyslexia has a high prevalence rate and is recognized in standard classification systems (ICD-10 and DSM-5). In many cases, dyslexia negatively affects education and psychological well-being. Even though effective intervention is available to reduce the negative long-term effects, the costs for individual dyslexia intervention are not covered by health insurance in Germany and in many other countries (e.g., United Kingdom: Government of the United Kingdom, 2017; The Netherlands: CZ, 2021). In Germany, parents usually have to pay the intervention costs themselves (Schulte-Körne, 2009). Only exception is if the child not only suffers from dyslexia but also shows symptoms of psychosocial problems, such as anxiety, depression, or ADHD. In this case, parents can apply by the youth welfare office to cover the intervention costs with the aim to avoid a negative developmental trajectory and to prevent children from emotional and mental illness. This application requires a special medical expert report according to Section 27 of the German Social Code VIII (the German "SGB VIII") or §35 a SGB VIII.

Even though effective interventions for dyslexia are available, an important question for health insurance to decide whether an intervention is covered by insurance or not is whether the benefits of the intervention justify the costs of the intervention. The reason for this is that it is important to allocate health care resources efficiently because of the continuous increase in health care costs. In Germany, the health care sector represents more than 10% of the whole national economic turnover (Hajen, Paetow, & Schumacher, 2000). The costs for mental diseases spent in 2015 in Germany make up 13% of the whole health care costs (Statistisches Bundesamt [Federal Statistical office], 2017). As a consequence, there has been a considerable increase over the last few years in performing health economic analyses in many paediatric domains, in order to calculate the relation between the benefits and the costs of an intervention/treatment (Gerber & Lauterbach, 2006). However, there are hardly any studies examining the

benefits together with the costs of dyslexia intervention. One exception is a Dutch study (Hakkaart-van Roijen, Goetsch, Ekkebus, Gerretsen, & Stolk, 2011), where the authors used a modelling approach to assess long-term costs and benefits in terms of quality of life by comparing an evidence-based dyslexia training with care as usual (i.e., unspecific training or no training). Successful intervention showed a quality-of-life gain of about 11%, while the costs per quality adjusted life years (QALYs) decreased from primary to secondary school and after secondary school. In sum, the authors argue that intervention of dyslexia is cost-effective.

One reason for the low number of health economic analyses in dyslexia research is that health economic evaluation in this field faces a number of challenges, specifically concerning the way, how intervention benefits are best operationalized and measured. Compared to studies assessing physical impairments such as cancer, in which benefits can be measured by physiological values (e.g., decrease of tumour cells), the most frequently used measure of intervention benefit in psychiatric disorders (including dyslexia) is an improvement in quality of life (Ravens-Sieberer et al., 2013; Ravens-Sieberer, Voss, Reiss, Wüstner, & Otto, 2019). However, the paediatric population comprises a wide age range (Ungar, 2009, 2011) and there is currently no instrument available to measure the quality of life across all age groups. In the current study, we, therefore, focused on children aged between 12 and 18 years. For this age group, quality of life can be reliably and directly assessed via self-reports, rather than indirectly via parental report.

Another problem of health economic analysis in dyslexia research is that, in order to compute a cost-utility analysis, a general health-index needs to be calculated (as for example measured by the EQ-5D questionnaire: EuroQol Group, 1990). The biggest advantage of such a cost-utility analysis is that it provides one common unit of measure (i.e., the costs per QALY). This common unit allows to either directly compare two alternative interventions by calculating the incremental cost-effectiveness ratio (ICER), or to classify the cost-effectiveness of an intervention and compare it with other disorders. In the current study, we focus on the latter approach rather than calculating the ICER, because data for benefits and costs for no intervention that are necessary for calculating the ICER do not exist.

The flip side of assessing the general health-index, which is needed to calculate the cost-utility analysis, is that the general health-index does not capture all relevant domains that might be affected by the disorder of interest (here dyslexia), and that are likely to be modified by the intervention. In the case of dyslexia, relevant domains that are not sufficiently covered by the general health-index are, for example, changes in self-esteem and in emotional well-being in relevant settings, such as school, family, and peers. Previous research provided evidence that dyslexia is frequently associated with these domains (Giovagnoli et al., 2020; Livingston, Siegel, & Ribary, 2018; Terras, Thompson, & Minnis, 2009), and that problems in these domains are related to the severity of literacy difficulties (Mugnaini, Lassi, La Malfa, & Albertini, 2009). Thus, reducing literacy problems is likely to affect the mentioned quality-of-life dimensions. In the current study, we, therefore, added another measure of quality of life (i.e., the Kiddo-KINDL<sup>®</sup>), that allowed us to assess these dimensions (i.e., “Physical,” “Emotional,” “Self-esteem,” “Family,” “Friends,” and “School”).

Furthermore, other intervention outcomes such as a reduction in health care costs or educational outcomes that affect productivity should also be considered when analysing the monetary benefit of dyslexia intervention. In addition to the cost-utility analysis calculated based on the current sample, we, therefore, computed analyses based on information from previous large-scale studies, in order to calculate consequential costs related to dyslexia (i.e., repetition of classes; school education; unemployment).

## 1.1 | Aims of the study

The overall aim of the study was to quantify changes in quality of life by comparing the quality of life before and after state-of-the-art individual dyslexia intervention using different self-report questionnaires (EQ-5D and Kiddo-KINDL<sup>®</sup>), and to compare the benefit of the intervention in terms of improvement in quality of life with the costs of the intervention by computing a cost-utility analysis.

More specifically, we first assessed the intervention effect for the total health-index score measured by Part 1 of the EQ-5D questionnaire (EuroQol Group, 1990), which is one of the most commonly used measures to assess the health-index (aim 1a). Next, we analysed the specificity of the intervention effect by analysing the five different health dimensions of the EQ-5D separately (aim 1b). We expected to find greater changes in the two dimensions that are related to dyslexia and dyslexia intervention (“usual activities” and “anxiety/depression”) compared to the three dimensions that are not related to dyslexia and, therefore, unlikely to be affected by the intervention (“mobility,” “self-care,” and “pain”).

Second, we aimed to assess changes in quality of life in more detail by analysing (a) the self-estimated health status measured by the visual analogue scale (= Part 2 of the EQ-5D questionnaire) (aim 2a), and (b) the Kiddo-KINDL<sup>®</sup> self-report questionnaire (Ravens-Sieberer & Bullinger, 2000). The Kiddo-KINDL<sup>®</sup> includes six life domains that are strongly related to dyslexia and dyslexia intervention (i.e., “physical well-being,” “emotional well-being,” “self-esteem,” “family,” “friends,” and “school”). We, therefore, expected to find changes related to intervention in all six life domains (aim 2b).

Third, we aimed to compute a cost-utility analysis in order to compare the benefit of high standard dyslexia intervention delivered by certified dyslexia therapists in terms of improvement in quality of life (as measured by the health-index of the EQ-5D) with the costs of the intervention (aim 3).

Finally, we aimed to estimate consequential costs for untreated dyslexia by calculating productivity loss due to school failure (i.e., repetition of classes; school education; unemployment) (aim 4). We, therefore, used data from official statistics (i.e., from the Statistisches Bundesamt [Federal Statistic Office of Germany], Destatis: <https://www.destatis.de>) as well as information based on previous studies comparing school failure in German samples with and without dyslexia (Beer, 2013; Esser et al., 2002; Esser & Wyszchkon, 2008).

## 2 | METHOD

### 2.1 | Participants

Ethical approval for the study was obtained from the Clinical Ethics Committee of the LMU Hospital Munich, Germany. Participants included in the study were aged between 12 and 18 years old, and had a clinical diagnosis of dyslexia based on ICD-10 criteria and the German clinical guidelines for diagnosis and intervention of dyslexia. In order to ensure comparable and high-quality interventions, all participants were recruited via BVL<sup>®</sup>-certified therapists, who were contacted via email or phone. Although, we did not systematically assess the material used in the interventions, the recruiting procedure guaranteed that all participants were diagnosed according to the German clinical guideline for diagnosis. In order to receive a diagnosis of dyslexia, performance in reading and/or spelling has to be at least 1.5 standard deviations (SD) below the age- or grade-specific mean on standardized reading and spelling tests. A less stringent criterion of at least 1 SD can be applied if additional information (e.g., school reports) is supporting that the child's literacy problems are persistent. With respect to intervention, all participants received an individualized symptom-specific intervention that follows the guidelines for intervention of dyslexia. According to these guidelines, interventions are based on systematic phonics instruction, which has been shown to improve reading skills in children with dyslexia across orthographies. For spelling, intervention programs are based on phonics instruction together with morpheme-based approaches and teaching of orthographic rules, which showed the strongest effects according to meta-analyses (Galuschka et al., 2014, 2020).

All study documents, including the study information for children/adolescents and their parents, the consent forms, and the questionnaires for assessing the quality of life (EQ-5D and Kiddo-KINDL<sup>®</sup>) before and after the intervention were sent by email or post to the participating therapists ( $N = 286$ ). Therapists then asked their therapy children/adolescents and the parents to participate in the study. All participants had received a minimum of 20 weeks of intervention when filling-in the two quality-of-life questionnaires (EQ-5D and Kiddo-KINDL<sup>®</sup>). Children/adolescents

had to rate their quality of life before the start of the intervention and after the intervention, while parents were asked to provide information about the direct and indirect costs of the intervention. Thirty-six children/adolescents (9 females and 27 males); average age 13.28 years (SD 1.16) fulfilling inclusion criteria (aged between 12 and 16 years and clinical diagnosis of dyslexia based on ICD-10 criteria) returned the completed questionnaires.

At the time of the study, 14 of the 36 children had not yet finished their intervention, but had attended a minimum of 20 h. The average amount of hours of lesson that the participants had attended until the start of the survey was 80.46 h (SD = 41.71). According to the meta-analysis by Ise et al. (2012), intervention effects emerge after 20 weeks of support. Thus, all participants included in the study had reached the duration of the intervention that is necessary to reliably observe intervention effects.

In addition to the diagnosis of dyslexia and in line with the high comorbidity rates observed for dyslexia, 12 patients (33%) also showed comorbid disorders, including attention-deficit-hyperactivity disorder, somatic symptoms such as stomach pain and headache, sleep disorder, anxiety, social withdrawal, low self-esteem, or depressive symptoms.

## 2.2 | Quality-of-life measures

### 2.2.1 | EQ-5D

The EQ-5D (EuroQol Group, 1990: <https://euroqol.org/>) is the most widely used standardized instrument for measuring health-related quality of life in cost-effectiveness analysis. The EQ-5D can be used from 12 years of age onwards. The EQ-5D questionnaire consists of two parts: *Part 1* comprises five questions which assess five different health dimensions at a three-point Likert-scale (no problems, few problems, many problems). The five dimensions are: “mobility,” “self-care,” “usual activities,” “pain/discomfort,” and “anxiety/depression.” The different possible combinations of response patterns represent 243 health states (Greiner, Claes, Busschbach, & von der Schulenburg, 2005), which are then transferred to a *health-index score* based on the manual (EuroQol Research Foundation, 2018). The health-index score of the EQ-5D is needed to compute the cost-utility analysis (aim 3). Based on the health-index score, the QALYs can be calculated using two dimensions, namely changes in quality of life and the life expectancy or critical life period. These can be combined as a compounded new unit (Schöffski & Graf von der Schulenburg, 2012), resulting in a one-dimensional unit (for more details see description of the cost-utility analysis).

In addition to the analysis based on the total health-index score of the EQ-5D (aim 1a), the five different dimensions of *Part 1* were analysed separately, in order to assess whether changes in quality of life related to the dyslexia intervention are domain-specific; thus, occurring only in domains that are related to the disorder of interest (here dyslexia) but not in other domains of quality of life (aim 1b).

Finally, *Part 2* of the EQ-5D, the *visual analogue scale*, describes the self-estimated health status on a vertical scale from 0 to 100 (Oemar & Janssen, 2013). Participants are asked to judge their health status on the day of the assessment by ticking the corresponding number on a scale from 0 (worst imaginable health state) to 100 (best imaginable health state) (aim 2a).

### 2.2.2 | Kiddo-KINDL<sup>®</sup>

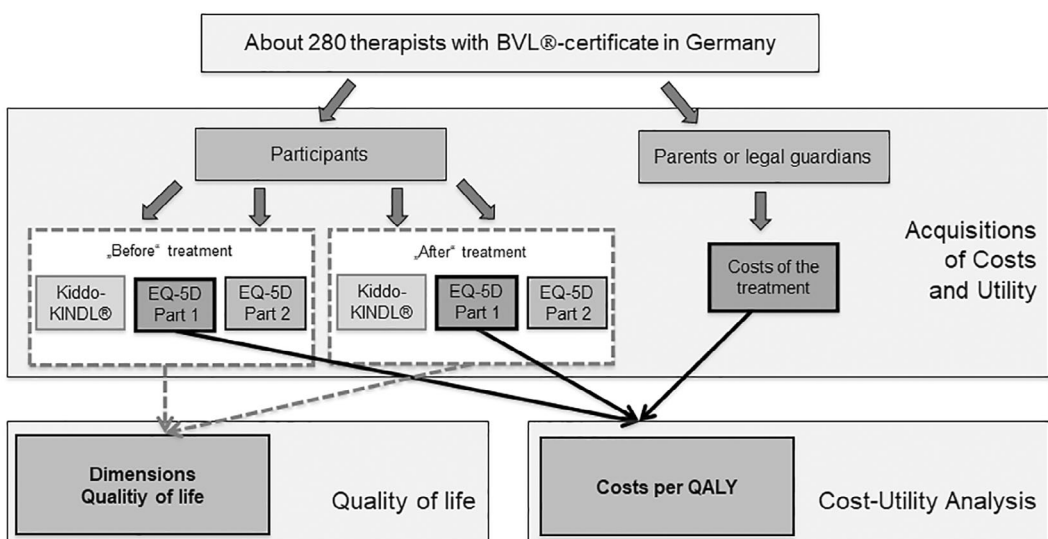
In order to measure changes of quality of life as outcome of the intervention in more detail (aim 2b), the Kiddo-KINDL<sup>®</sup> questionnaire (Ravens-Sieberer & Bullinger, 2000) was applied. The Kiddo-KINDL<sup>®</sup> allows to assess the quality of life in several domains that have been shown to be related to learning disorders. This questionnaire can be used in primary- and secondary-school-aged children/adolescents. The questionnaire contains 24 items that assess

the following six dimensions (four items per dimension): (a) physical well-being, (b) emotional well-being, (c) self-esteem, (d) family, (e) friends, and (f) school. Participants score each statement on a five-point Likert scale. For example, “last week I felt lonely” “never,” “rarely,” “sometimes,” “often,” and “all the time.” To analyse the questionnaire, a syntax was established according to the manual provided by Ravens-Sieberer and Bullinger (2000): First, items with reversed polarity were recoded so that all items were on the same scale, and missing values were counted for each subscale. Next, the six dimension-subscale scores were calculated by summing the four items in each subscale. Subscale scores were only calculated when at least three out of four items in a scale were available. Finally, the total score was generated based on the mean score of all six dimensions, and was then transformed into a standard score ranging between 0 and 100, with higher values reflecting a higher quality of life.

### 2.3 | Cost-utility analysis

The cost-utility analysis (aim 3) calculates the ratio between the costs of the intervention and the benefit of the intervention (Schöffski & Graf von der Schulenburg, 2012). The costs are measured in monetary units. Measurement of the benefit is the quality of life as assessed by the health-index score of the EQ-5D (see description Part 1 of the EQ-5D above). In cost-utility analyses assessing the benefit of a medical treatment (e.g., operation), the quality of life is often measured in the years the patient is gaining in full health through the treatment, the so-called QALYs. Obviously, this concept cannot be directly transferred to dyslexia, given that dyslexia does not affect overall life expectancy. Instead, a meaningful procedure is to calculate the QALYs for critical time periods (Gerber & Lauterbach, 2006). In the current study, we calculated the QALYs for the six years between the age of 12 and 18 years which are critical for education and personal well-being, because adolescents in Germany normally decide at the age of 18 on their next educational steps.

The cost-utility analysis was computed by setting the costs of the dyslexia intervention against the benefits of the intervention. Both, direct and indirect costs of the intervention were considered. The direct costs are tangible costs in terms of intervention expenses and other costs, such as transportation costs. Indirect costs are costs resulting from, for example, loss of family income due to reduced working hours of the parents during lessons.



**FIGURE 1** Structure of the study design



The benefit was measured by the health-index score of the EQ-5D, which assesses the health status before and after intervention and allows to compute the cost-utility analysis (EuroQol Group, 1990). The structure of the study design is provided in Figure 1.

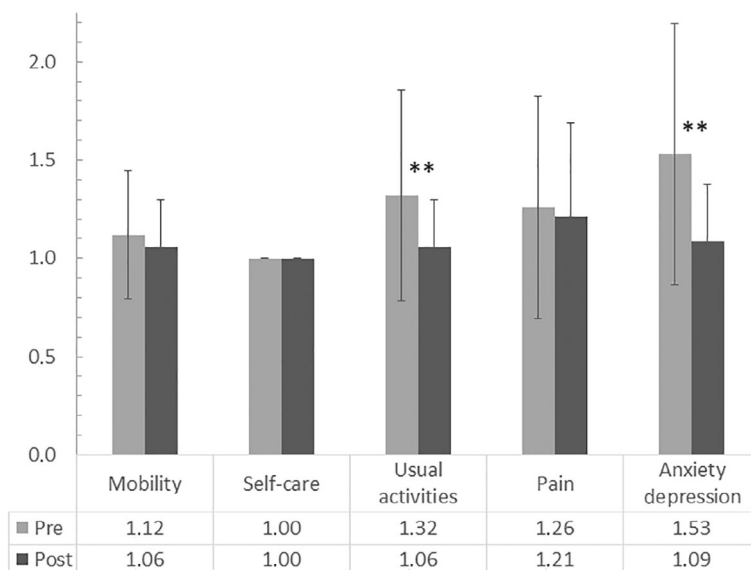
### 3 | RESULTS

#### 3.1 | Quality-of-life measures

For the EQ-5D questionnaire, complete datasets for pre- and post-intervention were available for 34 participants for *Part 1* (health-index score and five dimensions) and for 35 participants for *Part 2* (visual analogue scale) of the questionnaire. The health-index score, which was calculated based on the total score across the five dimensions of *Part 1* (aim 1a), increased significantly (Wilcoxon-Test,  $Z = -2.317$ ,  $p = .020$ ; effect size  $r = 0.281$ ) from pre-intervention (index score = 0.8996 [SD 0.206]) to post-intervention (index score = 0.9587 [SD 0.133]).

Next, we assessed the specificity of the intervention effects (aim 1b), by analysing the five dimensions of the EQ-5D separately ((a) mobility, (b) self-care, (c) usual activities (including school, family and friends), (d) pain/discomfort, and (e) anxiety/depression). Therefore, we compared the values of each dimension before and after the intervention. In line with our expectations, we found significant improvements in quality of life only in those domains that are related to symptoms of dyslexia and therefore likely to be affected by the intervention, namely in the dimension “usual activities” (Wilcoxon-Test,  $Z = -2.714$ ,  $p = .007$ ; effect size  $r = 0.329$ ) and in the dimension “anxiety/depression” ( $Z = -3.260$ ,  $p = .001$ ; effect size  $r = 0.395$ ). In contrast, no changes in quality of life were observed for the other three dimensions (“mobility,” “self-care,” and “pain/discomfort”), which are not directly related to dyslexia ( $Z_s \leq -1.414$ ,  $p_s \geq .157$ ). The results for the five dimensions of the EQ-5D are presented in Figure 2.

Next (aim 2), we assessed changes in quality of life in more detail by analysing changes through intervention in the visual analogue scale (*Part 2* of the EQ-5D) and in the six different domains of the Kiddo-KINDL® questionnaire. The results from the visual analogue scale of the EQ-5D (aim 2a) showed that the self-estimated quality-of-life status



**FIGURE 2** Means and standard deviations of the five EQ-5D subscales. \*\* $p < .01$



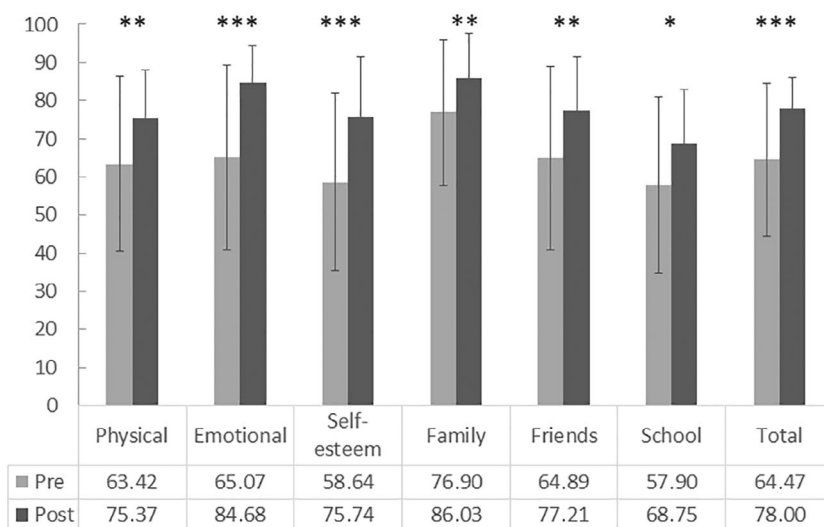
on the 0–100 scale increased significantly by 15.37 points (Wilcoxon-Test,  $Z = -3.963$ ,  $p < .001$ ; effect size  $r = 0.474$ ) from 74.86 (SD 22.19) before the intervention to 90.23 (SD 9.90) after intervention.

For the Kiddo-KINDL<sup>®</sup> (aim 2b), all 36 patients filled out the questionnaires, but two participants did not complete both, pre- and post-intervention questionnaires. Therefore, the pre-post analysis is based on the 34 participants with complete data sets. Before intervention, participants achieved a mean of 64.47 (SD 20.08) for the total score of the Kiddo-KINDL<sup>®</sup>. After intervention, the total mean score was 78.00 (SD 7.99). Thus, the health-related quality of life increased by 13.53 points, which represents a significant improvement (Wilcoxon-Test,  $Z = -3.509$ ,  $p < .001$ ; effect size  $r = 0.426$ ). A significant increase was not only found for the total score of the Kiddo-KINDL<sup>®</sup>, but also for each of the six subscales (“Physical,” “Emotional,” “Self-esteem,” “Family,” “Friends,” and “School”):  $Z$ s between  $-2.509$  and  $-3.961$ ; all  $p$ s  $\leq .012$ ; effect sizes  $r$ s between .304 and .480). The results for the six subscales and for the total score of the Kiddo-KINDL<sup>®</sup> are presented in Figure 3. The strongest effect sizes were found for the subscales “emotional well-being” and “self-esteem” ( $r$ s = .480 and .435, respectively).

## 3.2 | Cost-utility analysis (aim 3)

### 3.2.1 | Costs

In order to compute the cost-utility analysis, we first analysed the direct and indirect costs of the intervention based on the information provided by parents (intervention costs and indirect costs) and by child and adolescent psychiatrics (diagnostic costs). The direct costs were divided into costs for the diagnostic assessment and intervention costs. The diagnostic costs were calculated based on the average diagnostic costs over three quarters allowances reported by child and adolescent psychiatrics from two different federal states in Germany: Cologne €92.17 and Munich €105.08 (Association of Statutory Health Insurance Physicians Bavaria, 2012; Association of Statutory Health Insurance Physicians North Rhine, 2012). Based on these reports, the diagnostic costs were calculated with €100 per subject.



**FIGURE 3** Means and standard deviations of the six EQ-5D subscales and the total score. \* $p < .05$ ; \*\* $p < .01$ ; \*\*\* $p < .001$

With regards to intervention costs, parents filled in a questionnaire and reported that the costs per lesson ranged between €20 and €55. The average costs per lesson in the current sample were €39.49 (SD 7.73). Based on an average number of 80.46 lessons and average costs per lesson of €39.49, the costs of the dyslexia intervention amount to €3,177 per patient. Twenty of the 36 patients (55.6%) paid for the lessons themselves. In 12 cases (33.3%), the youth welfare office covered the costs and in one case, the private health insurance. Two other participants had the social paediatric centre cover their costs. For one patient, this information was missing.

On average, indirect costs amounted up to €1,118 (SD 1901), with a wide range from €0 to approximately €7,000 per patient. The extra costs were described as primarily transportation costs, loss of earnings of parents, and costs for special reading materials. As evident from the wide range, indirect costs are difficult to estimate. Therefore, previous cost-utility analyses often focus on the direct costs only. We, therefore, calculated the cost-utility analysis with and without including indirect costs.

In total, the average summed costs of direct costs (diagnostics and dyslexia intervention) and indirect costs per patient amount to €4,395 (with diagnostic costs of approximately €100, intervention costs of €3,177, and indirect costs of €1,118). The sum of costs for all 36 patients was €158,220. Excluding the indirect costs, the individual costs per patient amount to €3,277, thus resulting in €117,972 for all 36 patients.

**TABLE 1** Costs per quality adjusted life years (QALY) for dyslexia intervention based on the current study (bold) in comparison to other interventions/treatments in child health

Intervention	Costs (incremental costs or cost utility <sup>a</sup> ) per QALY in \$ versus € <sup>b</sup>	Study
Levofloxacin prophylaxis in hospitalized children with leukaemia	\$5,059.92 (€4,258.94)	Maser, Pelland-Marcotte, Alexander, Sung, and Gupta (2020)
Methylphenidate treatment for children and adolescents with ADHD in Brazil (children/adolescents)	\$9,103.00 (€7,661.99)/\$11,883.00 (€10,001.92)	Maia et al. (2016)
<b>Economic evaluation of dyslexia intervention</b>	<b>€13,119.00</b>	<b>Current study<sup>a</sup></b>
Wearable cardioverter-defibrillators in paediatric cardiomyopathy	\$20,103.00 (€16,920.70)	Evers et al. (2020)
Economic evaluation of stuttering treatment in preschool children	€18,617.00	de Sonnevile-Koedoot, Bouwmans, Franken, and Stolk (2015) <sup>a</sup>
Turbinoplasty with adenotonsillectomy for paediatric sleep-disordered breathing	\$27,333.00 (€23,006.19)	Baik and Brietzke (2019)
Bilateral cochlear implants for children with severe-to-profound sensorineural hearing loss in both ears (simultaneous vs. sequential implantation)	\$60,607.00 (€51,012.91) \$81,782.00 (€68,835.90)	Cheng, Soon, Wu, Ju, and Ng (2019)
Integrated care program for children with asthma	\$33,753,817.00 (€28,409,900.10)	Duenas-Meza et al. (2020)

<sup>a</sup>Cost-utility analysis, costs per QALY.

<sup>b</sup>For comparison, costs were transferred in Euro using the currency calculator retrieved from: [https://www.finanzen.net/waehrungsrechner/us-dollar\\_euro](https://www.finanzen.net/waehrungsrechner/us-dollar_euro); Exchange rate: €0.8417 = \$1.1880.

### 3.2.2 | Quality adjusted life years

As a next step for the cost-utility analysis, we calculated the QALYs. As mentioned in the method section, the calculation of the QALY was based on changes in quality of life (i.e., difference between pre- and post-intervention of the EQ-5D health-index score) and on the critical time-period of six years between the age of 12 and 18 years. Thus, the QALY is calculated by multiplying the six years with the difference score of the EQ-5D health-index before and after the intervention ( $6 \times [\text{EQ-5D pre-intervention} - \text{EQ5D post-intervention}]$ ). Next, we summed the QALYs of all participants, resulting in a summed QALY of 12.06. Thus, the costs per QALY were €9,782 when excluding indirect costs, and €13,119 when including indirect costs (sum of the costs divided by QALYs gained: €117,972/12.06 and €158,220/12.06, respectively). Table 1 shows the costs per QALY calculated for dyslexia intervention in the current study in comparison with the costs per QALY for other paediatric disorders. The results show that the costs per QALY for dyslexia intervention are in a similar range as those of similar developmental disorders, such as speech and language disorders (de Sonnevile-Koedoot et al., 2015).

### 3.3 | Consequential costs for untreated dyslexia (aim 4)

Finally, we estimated educational outcome and productivity loss based on official statistics (i.e., based on the Federal Statistic Office of Germany) and based on data from previous large-scale longitudinal studies (i.e., Kurpfalz-Studie: Esser et al., 2002; Esser & Wyszkon, 2008; Erlangen-Nürnbergiger Entwicklungs-und Präventionsstudie: Beer, 2013; Lösel, Beelmann, Jaurisch, & Stemmler, 2004), which assessed prevalence rates for *class repetition*, *school success*, and *unemployment* in individuals with and without dyslexia.

#### 3.3.1 | Class repetition

Beer and colleagues (Beer, 2013) assessed the cumulated *class repetition rate* in students aged 13–14 years with and without dyslexia, and found that students with dyslexia repeated classes 2.18 times more often than students without dyslexia (17.2 vs. 7.9%). Based on the school population in Germany of 10.9 million students (Statistisches Bundesamt [Federal Statistic Office]: Destatis, 2021), a prevalence rate for dyslexia of 5% (i.e.,  $n = 545,000$  students with dyslexia), and a class repetition rate in the general population of 2.6% in one year, the number of students within the dyslexic population repeating a class based on the general population rate would be 14,170 students. Assuming that the class repetition rate within the dyslexic population is 2.18 times higher than in the general population, 30,891 students with dyslexia are expected to repeat a class (i.e., 16,721 students more than expected based on the general population rate). According to the Federal Statistic Office of Germany (Statistisches Bundesamt: Destatis, 2020), the costs for an extra school year across all types of schools add up to 7,300 Euros on average per student per school year. Thus, the costs for the additional 16,721 students within the dyslexic population that repeat a class amount to €122,063,300 for one year.

#### 3.3.2 | School success

Esser and colleagues (Esser et al., 2002; Esser & Schmidt, 1993; Esser & Wyszkon, 2008) reported that the school outcome (as measured by a combined score of the school certificate and the final mark in the school leaving examination) in adolescents with dyslexia was significantly lower than the school outcome in those without dyslexia, and also lower than the outcome of individuals with other developmental disorders. The outcome in adolescents with dyslexia was comparable to the outcome observed in individuals with low IQ (IQ between 70 and 85); thus, the school outcome in the group with dyslexia clearly lacks behind the outcome that can be expected based on their level of intelligence. In line with these findings, Beer (2013) reported large group differences in the distribution of

**TABLE 2** Percentage of students with and without dyslexia in the different school types and average income and insurance rates according to school type

School degree	% Students without dyslexia <sup>a</sup>	% Students with dyslexia <sup>a</sup>	Income (€)	Insurance rate (€)
Gymnasium (university-entrance diploma)	76.3	27.6	4,421.34	694.15
Realschule (general certificate of secondary education)	17.0	34.5	3,060.66	480.52
Hauptschule (certificate of secondary education)	4.6	29.3	2,799.66	439.55
Grundschule/Förderschule ohne Abschluss (no school certificate)	1.1	6.9	2,246.34	352.68
Others	1.0	1.7	<sup>b</sup>	<sup>b</sup>

<sup>a</sup>Based on data from Beer (2013).

<sup>b</sup>No data available.

different school types when comparing children with and without dyslexia ( $N_{\text{total}} = 553$ ). For example, the highest school type in the German school system (i.e., Gymnasium) was attended by 76.3% of children without dyslexia, while only 27.6% of the children with dyslexia attended this school type. Table 2 reports frequencies of different school degrees in students with and without dyslexia (adapted from Beer, 2013), together with information about the average salary and insurance rate as a function of school degree, calculated using the data provided by the Federal Statistic Office of Germany (Statistisches Bundesamt, 2020). Based on these data, the average full-time pre-tax salary in the typically developing sample is €4,047 including an average insurance rate of €635, while the average salary in the dyslexic sample is €3,252 with an average insurance rate of €510. Based on the statistics published by the Federal Statistic Office of Germany in April 2021, there are 44.3 million people in employment, resulting in a subsample of 2,215,000 people in employment with dyslexia (5% prevalence rate). Applying the difference in salaries and insurance rates observed in the typically developing compared to the dyslexic sample (i.e., salary difference = €795; insurance rate difference = €125) to all 2,215,000 individuals with dyslexia in employment in the German population, the total loss in productivity amounts to €1,760,925,000 ( $2,215,000 \times €795$ ) including a loss for health insurance funds of €276,875,000 ( $2,215,000 \times €125$ ) per month.

### 3.3.3 | Unemployment

Esser and Wyschkon (2008) further reported higher unemployment rates in young adults with dyslexia compared to those without dyslexia (12 vs. 4% at the age of 18 years and 26 vs. 4% at the age of 25 years). These results suggest that unemployment rates in young adults increase with age. Consequently, the loss of productivity is likely to be substantial. To calculate the exact loss of productivity due to unemployment rates in the dyslexic population, numerous information is needed, such as the salary and length of the previous employment, which was not available for the current study.

## 4 | DISCUSSION

Dyslexia is recognized as neurodevelopmental disorder in standard classification systems (ICD-10 and DSM-5). Without adequate support, dyslexia has a negative impact on professional career and personal well-being.

Although effective interventions for dyslexia are available (Galuschka et al., 2014, 2020; Goodwin & Ahn, 2010; McArthur et al., 2012), the costs for such interventions are in most cases not covered by the German health insurance. One reason is that health care resources need to be allocated efficiently given the continuous increase in health care costs. It is, therefore, crucial to (a) quantify the benefits (utility) of the intervention, (b) calculate the costs to analyse the cost-utility-ratio, in order to judge cost-effectiveness of dyslexia intervention and to compare the cost-utility-ratio with other disorders, and (c) to estimate the consequential costs of untreated dyslexia in terms of educational outcomes. This is the first study calculating a cost-utility analysis for dyslexia intervention in Germany (for a Dutch study analysing long-term costs and benefits of dyslexia intervention in terms of quality of life see Hakkaart-van Roijen et al., 2011).

To quantify the benefits in terms of improvement in quality of life, we first compared changes in quality of life using a variety of different quality-of-life measures (EQ-5D Part 1: total health-index; EQ-5D Part 2: visual analogue scale; Kiddo-KINDL<sup>®</sup>: total score and six subscale scores). All measures consistently showed a significant increase in quality of life after intervention with medium effect sizes (i.e.,  $r$ s ranging between .281 and .480). Importantly, changes in quality of life were restricted to those domains that are associated with learning disorders, and are thus likely to be positively affected by the intervention (i.e., the dimensions “usual activities” and “anxiety/depression” of the EQ-5D and the six subscales of the Kiddo-KINDL<sup>®</sup>). In contrast, the dimensions “mobility,” “self-care,” and “pain/discomfort” of the EQ-5D, which are related to physical disorders rather than learning disorders did not change after the intervention. The specificity of this intervention effect further suggests that participants did not respond according to social expectancy (i.e., reporting a general improvement in quality of life), but provided a differentiated response reflecting the specific effects of the intervention.

Among the six subscales of the Kiddo-KINDL<sup>®</sup> ((a) physical well-being, (b) emotional well-being, (c) self-esteem, (d) family, (e) friends, and (f) school), which are all related to dyslexia and showed intervention effects, the strongest effects were found for the scales “emotional well-being” and “self-esteem” ( $r = 0.480$  and  $0.435$ , respectively). It should be noted that even the smallest effect size ( $r = 0.304$  for “school”) still represents an intermediate effect. The strong effects on “emotional well-being” and “self-esteem” highlight the fact that the individualized dyslexia intervention does not only positively affect the core-symptoms of dyslexia, namely reading and spelling skills (Galuschka et al., 2014, 2020), but also has a significant positive effect on problems frequently co-occurring with dyslexia (i.e., internalizing disorders, such as anxiety and depression). Although a control group was not included in the current study, these positive effects on emotional-wellbeing are unlikely to reflect developmental changes, given that recent findings (Giovagnoli et al., 2020) showed increased levels of self-perceived emotional problems in adolescents with dyslexia compared to typically developing controls, whereas no such group differences were observed in childhood, suggesting that emotional problems in adolescents increase rather than decrease. Thus, the increase in emotional well-being observed in our study is likely to be caused by the intervention. Giovagnoli and colleagues further showed that high levels of internalizing symptoms in adolescents with dyslexia were associated with a low level of self-esteem. Thus, individualized interventions for dyslexia which enhance self-esteem have the potential to reduce internalizing symptoms and to prevent co-occurring emotional disorders. This interpretation needs to be supported in future studies directly assessing intervention effects and how they relate to changes in emotional well-being and self-esteem, as well as by examining the role those co-occurring difficulties play in this relationship.

After analysing the benefit of dyslexia intervention in terms of changes in quality of life across different domains and different measures, our main aim was to relate the benefits of dyslexia intervention to the costs associated with an individualized training delivered by qualified dyslexia therapists, by calculating a cost-utility analysis. We focused on the age range between 12 and 18 years to assess the effectiveness/benefits of the intervention from the patient's perspective, while the parents' perspective was used to calculate the intervention costs and the indirect costs of the intervention. The result of the cost-utility analysis showed that the costs per QALYs gained were €9,782 (excluding indirect costs) or €13,119 (including indirect costs) (cost-utility ratio). From the perspective of the society, the results can be related to other health care programmes reported in the so-called League-Table (Maynard, 1991). The table ranks the cost effectiveness for various interventions and health care programmes. The cost-utility ratios

reported in the table range between £220 (e.g., for cholesterol testing and diet therapy) and £126,290 (for Erythropoietin treatment for anaemia in dialysis patients). Thus, the dyslexia intervention with €9,782 and €13,119, respectively, is situated in the lower 10% of the League-Table, suggesting that costs are comparatively low. When comparing the cost-utility ratio of dyslexia intervention with similar interventions for developmental disorders, such as speech therapy for stuttering (see Table 1: stuttering treatment in preschool children—€18,617), the cost-utility ratios are highly comparable. While interventions such as speech therapy are covered by health insurance, this is not the case for dyslexia therapy.

Finally, the costs that result as a consequence of untreated dyslexia are likely to be substantial. In the current study sample, we did not assess the consequential costs of untreated dyslexia such as school dropout, professional career, and costs for co-occurring psychiatric disorders, such as depression and anxiety. Including these costs in future cost-utility analysis will most probably further increase the cost-effectiveness of dyslexia intervention. Nevertheless, we estimated school-related consequential costs by using information from previous research about educational outcomes, such as class repetition, school success, and unemployment rates, in adolescents/adults with and without dyslexia (see Table 2). The consequential costs for class repetition due to dyslexia amounted to €122,063,300 for one year. Reducing the class repetition rate will not only save the costs for an additional school year, but will also decrease the loss of productivity resulting from the delayed start of working life.

Costs for drop-out rates were not calculated, because we could not find reliable statistics for German school drop-out rates in individuals with dyslexia. However, in an American study the drop-out rate among poor readers was clearly increased, and reported to be as high as 30% (Daniel et al., 2006). Without a school leaving certificate, individuals with dyslexia will most likely have a lower income and worse chances with regard to social integration (Entorf & Sieger, 2010). Even if they finish school with a certificate, individuals with dyslexia often do not have a job corresponding to their general cognitive abilities because of their reading and writing problems (Esser et al., 2002; Knapp, Pfaff, & Werner, 2008). This has not only negative consequences for personal well-being but will also result in a loss of productivity for the society. When analysing the educational outcome in terms of loss of productivity based on the average income related to the different school degrees, we found that the loss amounts to €1,762,637,948 per month. It should be noted that the total loss related to school degrees and income is unlikely to be completely compensated by the positive effects of dyslexia intervention. However, even if higher school degrees can only be achieved in a certain proportion of individuals with dyslexia, this will significantly reduce the productivity loss. In addition, the loss of productivity does not only affect the individual with dyslexia and the society, but also has a negative impact on the health insurance system, given that the amount of the insurance rate depends on the income of the employees.

Finally, the loss of productivity is likely to be further increased by the higher unemployment rates observed in individuals with dyslexia compared to those without dyslexia. Future studies should assess more detailed information about the professional career in order to calculate the loss of productivity resulting from increased unemployment rates in the dyslexic population.

Last but not least there will also be costs incurred by hospitalizations, medications, or psychotherapy due to the high comorbidity rates of dyslexia. Up to 60% of individuals with dyslexia fulfil diagnostic criteria for one or more additional disorder, including severe mental disorders like depression and anxiety disorder (Daniel et al., 2006; Maughan, Hagell, Rutter, & Yule, 1994; Willcutt & Pennington, 2000). As mentioned before, individualized dyslexia therapy which positively influences self-esteem is likely to reduce internalizing symptoms (Giovagnoli et al., 2020), and in turn reduces costs for co-occurring emotional problems.

From the perspective of social equity and cohesion, the current study showed that over 55% of the parents had to cover the intervention costs themselves. Based on a recent survey by the BVL (German Dyslexia and Dyscalculia Association) with over 600 parents of children with learning disorders, the percentage of self-payment is even higher. According to the survey, 67.1% of the parents of the 309 children who received individual intervention outside school had to cover the costs themselves (BVL, 2021). In addition, some parents in our study reported horrendous extra costs, including costs for transportation, loss of earning, and for reading materials. This means that access to adequate dyslexia intervention, especially for children with severe reading and spelling problems, is at least partly

a question of parental income, thus inducing social inequality. Note that other interventions like speech therapy which are similar to dyslexia intervention in terms of therapy extent and costs are paid for by the health insurance as these intervention forms are listed in the catalogue of modalities in Germany.

The study also has some limitations. First, it was a retrospective design and participants were asked to answer the questionnaires retrospectively. This might have induced a response bias in terms of social desirability (i.e., reporting an improvement in quality of life). However, given that participants provided differentiated responses and reported increases in quality of life for specific domains only, we can largely rule out that responses were solely based on social expectancy.

Secondly, the sample might not be representative for the whole dyslexic population. Future studies should therefore replicate the current results based on larger representative samples. Future studies might also include additional outcome measures that can inform about short-term educational outcomes, such as increases in literacy skills due to intervention. This would allow to directly relate the intervention effects on educational outcomes to the changes in quality of life.

Finally, we have argued that it is generally difficult to construct life time models in the field of developmental disorders. In the current study, we have calculated the QALY based on the critical period of six years between the age of 12 and 18 years. Whether the results are the same for other life periods remains a question for further research.

## 5 | CONCLUSIONS

The objective of this study was to quantify the benefits of dyslexia intervention and to relate the benefits to the costs of dyslexia intervention by computing a cost-utility analysis. Results showed a clear benefit in terms of an increase in the quality of life across different relevant domains and measurements. The result of the cost-utility analysis was shown to be €9,782 (excluding indirect costs) per QALY. This is in line with other similar therapy forms such as speech therapy. In addition, we calculated consequential costs of educational outcomes based on official statistics and previous findings on school failure. Results showed high costs that emerge due to untreated dyslexia. Together these findings suggest that dyslexia intervention is cost-effective and the investment in dyslexia intervention is reasonably justified.

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## DATA AVAILABILITY STATEMENT

Data available on request from the authors: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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