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The Cost-Effectiveness of an Intensive Treatment Protocol for Severe Dyslexia in Children

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Studies of interventions for dyslexia have focused entirely on outcomes related to literacy. In this study, we considered a broader picture assessing improved quality of life compared with costs. A model served as a tool to compare costs and effects of treatment according to a new protocol and care as usual. Quality of life was measured and valued by proxies using a general quality-of-life instrument (EQ-5D). We considered medical cost and non-medical cost (e.g. remedial teaching). The model computed cost per successful treatment and cost per quality adjusted life year (QALY) in time. About 75% of the total costs was related to diagnostic tests to distinguish between children with severe dyslexia and children who have reading difficulties for other reasons. The costs per successful treatment of severe dyslexia were €36366. Successful treatment showed a quality-of-life gain of about 11%. At primary school, the average cost per QALY for severe dyslexia amounted to €58647. In the long term, the cost per QALY decreased to €26386 at secondary school and €17663 thereafter. The results of this study provide evidence that treatment of severe dyslexia is cost-effective when the investigated protocol is followed. Copyright © 2011 John Wiley & Sons, Ltd.

Keywords: dyslexia; remediation; cost-effectiveness; quality of life; model

INTRODUCTION

A study on the clinical effectiveness of a new protocol for the treatment of severe uncomplicated dyslexia indicated that this treatment seemed effective (Gerretsen, Hasselman, & Ekkebus, 2006). However, uncertainty remains about whether the added value of this treatment to society is large enough to justify its cost. As the healthcare system is subject to resource constraints, policymakers have become aware that it may not be possible to meet all the healthcare needs of patients. Therefore, in many countries, policymakers now require that those who request funding for a new treatment submit evidence not only about the effectiveness of the interventions but also about cost-effectiveness to inform their decisions. Such is also the case in the Netherlands, where the funding decision about dyslexia treatment was to be informed by information on clinical and economic effects. So that it can be determined if allocation of scarce

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healthcare resource to dyslexia treatment represents social value, a framework of economic analysis that can estimate costs and effects on the basis of all the available evidence is needed, over a relevant time horizon and for specific patient groups. It must also enable the accumulated evidence to be synthesized in an explicit and transparent way, while also accounting for uncertainty in the parameters used to calculate cost-effectiveness (Drummond, Sculpher, & Torrance, 2005). Against this background, we undertook a cost-effectiveness analysis of the new protocol for treatment of severe uncomplicated dyslexia.

Dyslexia is defined as a failure to learn to read at an age-appropriate level despite adequate intelligence and appropriate opportunity. It is widely accepted that the core deficit underlying dyslexia is a phonological deficit (Lyon, Shaywitz, & Shaywitz, 2003; Vellutino, Fletcher, Snowling, & Scanlon, 2004). Around 10% of school-age children fail to read efficiently, approximately 4% are assumed to suffer from dyslexia (Blomert, 2005). Literacy problems in dyslexia do not limit themselves to reading. In a nationwide prevalence study of dyslexia, it was shown that 95% of the children with severe reading problems also exhibit spelling problems (Blomert, 2002). The academic and later vocational success of these children is likely to be reduced if they do not receive extra support (Maughan, 1995). Dyslexia also has been found to have a negative effect on children's social and emotional development (Edwards, 2003). A Norwegian study concluded that preadolescent dyslectic children showed a wide range of behaviour problems that could not be attributed to social or developmental background variables (Heiervang, Stevenson, Lund, & Hugdahl, 2001). Another study showed that children's aggressive behaviour and reading difficulties during early elementary school are risk factors for adolescent problem behaviours such as delinquency, academic failure and substance use (Barrera *et al*., 2002). Hence, no or ineffective treatment of dyslexia will generate extra cost to society, for example increased healthcare costs and costs due to special education and/or remedial teaching. Furthermore, dyslexia will decrease quality of life.

Reviews of international dyslexia intervention studies explored the outcomes of treatment programmes and identified the basic ingredients for an effective treatment programme for dyslexia: training phoneme awareness, relating spoken and written language at the subword level and explicit tutored instruction (Foorman, Breier, & Fletcher, 2003; Goetry, Nossent, & Hecke van, 2006; Lin *et al*., 1999; National Reading Panel, 2000; Torgesen, 2005). These ingredients are combined in the Dutch National Protocols for Dyslexia Diagnostics and Treatment (Blomert, 2006) including training of phoneme awareness in combination with grapheme–phoneme association as the basis for a highly structured reading and spelling training at subword and word level. Two Dutch effect studies revealed that progress in clinical settings towards normalization of reading and spelling performance was possible (Gerretsen, Vaessen, & Ekkebus, 2003; Tijms, Hoeks, Paulussen-Hoogeboom, & Smolenaars, 2003).

Limits on healthcare resources mandate that resource allocation decisions be guided by considerations of cost in relation to expected benefits. Although cost-effectiveness analyses do not reflect every element of importance in healthcare decisions, the information it provides is critical to inform decision makers about the allocation of healthcare resources. Because dyslexia treatment belongs to the healthcare domain, many policy decision makers would like to be informed how the cost and benefits of these programmes compare with their alternatives as well as with other interventions in health care. From the viewpoint of cost-effectiveness in health, the educational outcomes are perceived as intermediate outcomes that can be translated into main outcomes: cost and compared with gain in health-related quality of life. In a cost-effectiveness analysis, we explore if the extra

cost for the best programme is justified by the extra benefits. Unfortunately, the outcomes in current international studies on the effectiveness are mainly educational. The associated effects on quality of life have not yet been established.

Although effective interventions for dyslexia are available, until now, information about the cost-effectiveness of these interventions is lacking, nationally and internationally. In the current study, we assessed the cost-effectiveness of application of an intervention for severe dyslexic young children in the Netherlands. The analysis was conducted from a societal perspective including all relevant costs and effects. We applied a modelling technique to assess (long-term) costs and effects in terms of quality adjusted life years (QALYs) of treatment of severe dyslexia according to the protocol compared with care as usual.

METHODS

We compared the cost-effectiveness of the dyslexia protocol with that of care as usual. We applied a cost utility analysis, a form of cost-effectiveness analysis in which clinical outcomes are expressed in the form of QALYs gained. Cost and effects were measured from the societal perspective. Cost and effects were analysed over various time horizons: 6, 12 and 18 years, which are generally similar to start of primary school, secondary school and post-secondary school, respectively.

Intervention

The dyslexia protocol (Blomert, 2006) is a practice-based guideline for the diagnostics and treatment of severe uncomplicated dyslexia. The dyslexia protocol is based on Gramma. Gramma is a computer-aided treatment programme developed at the Regional Institute for Developmental Dyslexia (RID, the Netherlands). This integrated reading and spelling programme has three main training components. The phonological component focuses on explicit phonics skills with a special emphasis on learning to classify phonemes into different phoneme classes or types that have a direct relevance for the application of spelling rules. The algorithms employed in the programme, follow the following form: if you hear phoneme type X at a particular position in the word, apply rule Y and write Z (Tijms *et al*., 2003). The phonological structure of words and the consequences this has for grapheme–phoneme connections is the basis for word building. During all relevant steps, auditory feedback is given and although the programme is computer-aided, it is never the case that the child works at the computer without individual supervision and feedback. The weekly training sessions by psychologists or special educationalists last 50 min, augmented with homework exercises of about 15 min during the other workdays of the week supervised by the parents. On average, children are enrolled in the programme for one and a half year. The aim of the treatment is a functional level of reading and writing. Treatment according to the protocol should be provided by mental health psychologist and special educationalists.

Care as usual for severe dyslexia included non-evidence-based treatment by speech therapists, general practitioner (GP), school doctor or no treatment.

Effectiveness

A study assessed the short-term and long-term effectiveness of Gramma, a treatment programme based on the guidelines of the protocol (Gerretsen *et al*., 2006). We used the data of this study on effectiveness in our study on the cost-effectiveness, controlling

for uncertainty. The intervention study consisted of 193 dyslexic children, between 7 and 14 years of age. Informed consent for anonymous use of the test data was obtained from the parents. Children were referred for diagnosis after extra remedial help at school had proven ineffective. The children were all diagnosed with developmental dyslexia using an extensive cognitive psycho-diagnostic procedure. With very few exceptions, all children performed within the lowest 10% of the grade-level norms for reading. They all attended regular schools and had normal intelligence (IQ > 85), normal or corrected to normal vision, no (history of) hearing, emotional or social problems and no manifest co-morbidity. Children were entered in the study consecutively according to the end date of treatment if they fulfilled the above described criteria. The children were divided in three grade levels at pre-test: level 1 = children (second half) grades 1 and 2; level 2 = children grades 3 and 4; level 3 = grade 5, 6 or 7. At the pre-test, the whole group had a mean *z*-score of −1.98 (SD = 0.74) for reading words and a mean *z*-score of −1.91 (SD = 0.72) for spelling.

Data were analysed at group level and in terms of individual growth. In the group analyses, the raw scores were analysed with a repeated-measures ANOVA, with time of measurement (pre-treatment–post-treatment) as the within-subjects factor. Partial eta squared was used as a measure of effect size. An effect size of $d = 0.2$ is considered a small effect size, a medium effect size is around $d = 0.5$ and a large effect size is $d = 0.8$ or higher (Cohen, 1988). To investigate whether the gain made in raw scores exceeds the gain a normal population makes within the same time frame, we conducted an additional ANOVA in which the post-test score of the subject was corrected for expected 'natural' growth. That is, the expected gain between pre-test and post-test in the normal population at the lower end of the normal range (within −0.75 SD) was subtracted from the post-test score of each subject. This is an especially stringent criterion for treatment effectiveness because all subjects had an established history of *delayed* growth in reading and spelling. Next, pre-test–post-test differences in standard scores were analysed by means of a repeated-measures ANOVA with time of measurement (pre–post) as the within-subjects factor. This provided insight into whether subjects, besides showing clinically significant gain in raw scores, had been able to significantly improve their position in relation to the relevant norm group. In conclusion, it was investigated what these treatment effects at group level meant in terms of individual growth. To this aim, the growth in standardized scores at an individual level was investigated as well as the percentage of subjects post-treatment that performed within the normal reading range.

For reading, the main effects of time of measurement for the raw scores, the standard scores and the 'post-test scores corrected for normal gain' were significant ($p < 0.0005$) at all grade levels. The effect sizes for the three grade levels were respectively $d = 0.89$, $d = 0.87$ and $d = 0.81$. For spelling, the main effects of time of measurement for the raw scores, the standardized scores and the 'post-test scores corrected for normal gain' were significant for all grade levels ($p < 0.0005$). The effect sizes for the three grade levels were respectively $d = 0.93$, $d = 0.93$ and $d = 0.82$, indicating strong treatment effects for all grades.

The children were able to close the gap between their performance and the performance of age mates. Forty per cent performed at a level that can be considered average for their age. A 1-year follow-up of these children showed that they were able to maintain this level a year after treatment had finished (Gerretsen *et al*., 2006). Additionally, the study assessed the long-term effectiveness in a group of adults who were treated 10 years ago (Gerretsen *et al*., 2006). This study showed that they had been able to maintain their level of reading and writing in the long run, in contrast with the untreated adults with dyslexia, whom on average performed at grade 3 level. These results were in line with the result of a study of Tijms *et al*. (2003) on long-term effects of psycholinguistic treatment for dyslexia.

Unfortunately, the effectiveness study did not include controls, care as usual. However, all children included in the study received a phonics-based reading curriculum at school, had a history of reading and spelling problems and had received reading and spelling remediation at school and in many cases additional support. A considerable group of children included in the study were from grade 5 and higher. These children may thus be considered to have received care as usual from grade 1 to at least grade 4. Their average reading score when they entered the study was 4.0 (SD 1.9) on a reading test with a mean of 10 and SD of 3, and their average for spelling lay at the 10th percentile (SD 13.4). Both means demonstrate that they seriously lagged behind their classmates. Hence, such care as usual had not been sufficient for these severely dyslexic children. In the current study, we therefore assumed care as usual to be ineffective for severely dyslexic children in the base-case analysis.

Costs

In line with the societal perspective of this study, all relevant cost categories were considered. That is, direct medical costs and indirect non-medical cost, for example remedial teaching. The cost estimates were based on different sources, for example patient files, literature and expert opinion in case of lack of empirical data. Direct medical costs due to diagnostics were based on data files of an outpatient institute for dyslexia (RID) and expert opinion. The valuation of the health costs was based on the unit prices according to the reference prices for the Netherlands (Oostenbrink, Bouwmans, Koopmanschap, & Rutten, 2004).

The direct medical costs for treatment of dyslexia according to the protocol consisted of diagnostics, face-to-face contacts with clients, mutual contacts within the multidisciplinary treatment team and consultations with the school. To measure medical cost of care as usual, we investigated to what extent patients used other (ineffective) health care because of their severe reading and writing problems in the absence of treatment according to the protocol. During intake, patients at the RID were asked to indicate the healthcare use before start of the treatment. For 2005, we assessed 121 patient files to estimate healthcare use in the absence of treatment according to the protocol as proxy for care as usual for severe dyslexia.

The valuation of the health costs was based on the unit prices according to the reference prices for the Netherlands (Oostenbrink *et al*., 2004). Costs were adjusted to 2006 using the general price index from the Dutch Central Bureau of Statistics. Additionally, we conducted a micro-costing study to estimate the unit incremental costs per contact for treatment according to the protocol. Unit costs of labour were based on normative incomes and time spent on direct and indirect patient care. Indirect costs accounted for 35% of the costs per contact. According to the general guidelines of economic evaluations, the discount rate for future costs was 4% (Oostenbrink *et al*., 2004).

Utilities

We estimated utility values for different states of dyslexia, namely severe and mild dyslexia. These utilities were applied to the clinical outcomes before and after treatment as observed in the aforementioned clinical trial published by Gerretsen *et al.* (2006). Utility weights were assessed with a proxy version of the EQ-5D, which is a validated tool for measuring general health-related quality of life. It consists of five items (mobility, self-care, usual activities, pain/discomfort and anxiety/depression), each of which is rated as causing 'no problems', 'some problems' and 'extreme problems'. The 243 health descriptions can be directly linked to empirical values for health states of the general public, which allows utilities to

be computed (Essink-Bot, Stouthard, & Bonsel, 1993). The EQ-5D was administered by a panel consisting of people with a completed medical education, for example medicine, nursing, psychology, pharmacy. These persons were recruited internally (Erasmus MC) as well as externally (national consumer panel). We provided the panel with a description of hypothetical patients with severe and mild dyslexia. The description assumed dyslexia patients with no complications. A Web-based version of the EQ-5D was used; a paper version was also available. We valued the quality of life on the basis of the Dutch tariff (Lamers, 2007). In accordance to Dutch guidelines for economic evaluations, future QALYs were discounted at 1.5% (Brouwer, Niessen, Postma, & Rutten, 2005).

Model

Each year 200 000 children enter primary school in the Netherlands (Blomert, 2006). Of these children, about 6% will present serious reading and writing difficulties that meet the criteria for diagnostic testing of dyslexia (Blomert, 2002). About 1.8% will finally meet the criteria of severe dyslexia (Blomert, 2002). The cost utility was evaluated by relating the difference between average costs per child receiving treatment for dyslexia according to the protocol compared with care as usual to the difference in effects measured in QALYs, which yield an incremental costs per QALY gained (ICER). For estimating the ICER, we applied a simple Markov model divided by four stages: diagnostics, primary school, secondary and post-secondary education. The latter refers to education after secondary school, for example vocational education, higher professional education or university (Figure 1). The model describes the probabilities of successful treatment and simulates the costs and effects of the incidence of children with severe dyslexic problems in children who enter primary school. Table 1 presents the input parameters for the model.

Sensitivity analyses

To account for uncertainty in the parameters, we performed sensitivity analyses. The assumptions applied to calculate the costs effectiveness according to several scenarios are displayed in the second section of Table 1. The effectiveness of the protocol was 40% in the base-case scenario. In our calculations of the cost-effectiveness, we subsequently assumed that the quality of life of the other 60% of the children would stay comparable to children with severe dyslexia. This was a conservative estimate as the study showed that 70% of the children significantly improved (Gerretsen *et al*., 2006).

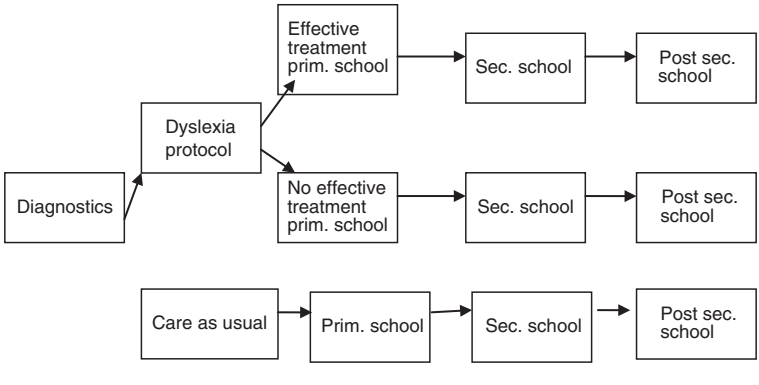

Figure 1. Structure of the Markov model.

Table 1. Input variables for the cost-effectiveness model and sensitivity analyses

Input parameters base-case scenario	Protocol	Care as usual
% of children diagnosed with severe dyslexia	1.8	1.8
% of children with normal reading and writing skills after treatment	40	0
Quality of life of children with severe dyslexia	0.77	0.77
% of children with diagnostic testing dyslexia	6	6
Sensitivity analyses		
% of children with normal reading and writing skills after treatment	70	0
% of children with normal reading and writing skills after treatment	40	10
Quality of life of children with severe dyslexia	0.80	0.80
% of children with additional healthcare costs and remedial teaching	60	100
% of children with diagnostic testing dyslexia	3	3

Hence, we performed sensitivity analysis assuming that the effectiveness of treatment according to the protocol was 70% instead of 40%. Generally, it was assumed that care as usual was ineffective in this group of children. However, it could be argued that a percentage of the children would recover. Hence, we estimated the cost-effectiveness assuming 10% of the children with severe dyslexia using care as usual would recover to the normal writing and reading level. Additionally, we performed a sensitivity analysis for the utility score of severe dyslexia. We assumed the mean quality-of-life score for severe dyslexia was 0.80 instead of 0.77 compared with 0.88 for the general population (see Result section), so that utility gain associated with treatment decreased from 0.11 to 0.08. Furthermore, we performed a sensitivity analysis for the percentage of children that enter primary school exhibiting reading difficulties that meet the criteria for diagnostic testing of dyslexia. Furthermore, we assessed the assumption that children treated according to the protocol would still use additional health care and remedial teaching. We assessed this assumption by adding these costs for the group of children in which the treatment protocol was not effective. In the base case, this was true for 60% of the children treated according to the protocol. Obviously, this type of care was provided to all children in the care-as-usual arm. Finally, we assumed the percentage cut down by half to 3% of the total number of children entering primary school that meet the criteria for diagnostic testing of dyslexia.

RESULTS

Costs

The average number of hours for diagnostics was 10h. Treatment according to the protocol is 50h on average. According to a micro-costing study, the unit costs for treatment was estimated at €72 per hour. Additionally, the costs for the diagnostic instrument were €125. Hence, the total average costs for diagnostics and treatment per child according to the protocol were €4445.

From the study of patient files (n=121), 35% of the children used other health care for their severe dyslexia before treatment according the protocol. We used this information as a proxy for care as usual. About 20% of these children visited a speech therapist, nearly 15% of the children went to a school doctor and a similar percentage visited a GP for their reading and writing problems. Less than 10% of the children visited psychologists, physiotherapists and other medical care specialists (e.g. eye specialist). Combined with

the number of visits for the corresponding healthcare providers, the average cost per patient was about €200. Additionally, at primary school, 96% the children with severe reading and writing problems received remedial teaching (1 h/week for 3 months/year during 4 years) (Blomert, 2005). The unit cost per hour was estimated at €40, making a total cost of €520 per year.

Effects

The EQ-5D was administered by 125 people, including 59 nurses (46.5%) and 18 physicians (14.2%). The mean quality-of-life score that was attributed by the EQ-5D to the health state of severe dyslexia was 0.77 (SD 0.20). In comparison with the population norm of 0.88 (SD 0.16), we calculated that severe dyslexia reduced quality of life by 11%. Mild dyslexia showed no significant difference compared with the general population.

Cost utility

The costs per successful treatment of severe dyslexia were €36 366. This study analysed cost-effectiveness of protocolized diagnosis of a treatment for severe dyslexia compared with care as usual in terms of cost per QALY. The total costs when the protocol would be introduced included the costs of the diagnostics (38.9 million euros) and the costs of treatment according to the protocol (13.5 million euros). In total, these costs were over 52 million euros. The costs of care as usual contained the direct medical costs (720 000 euros) and non-medical costs (remedial teaching) (over seven million euros). In total, the costs of care as usual was about 7.8 million euros, Table 2. The cumulative utility scores were higher for the group treated according to the protocol because in 40% of the children the quality of life was improved to the normal level. At the start of primary school, the difference in costs was €44 537 030 against 759 QALY gained. So we estimated the average ICER for severe dyslexia at €58 647 per QALY (€44 537 030/759), Table 2. The cumulative number of QALYs gained in time was more favourable for the intervention group because of the higher effectiveness. In contrast, the cumulative costs did not change after primary school. Consequently, the cost-effectiveness improved with longer time frames and was estimated at €26 386 per QALY at the start of secondary school and €17 663 per QALY thereafter. Table 2 showed that the cumulative cost does not increase after primary school; however, the cumulative gain in utilities increased in the long run because in 40% of the children the quality of life was improved because of the intervention.

Table 2. Cumulative costs, utilities and the incremental costs per QALY (ICER) of treatment according to the dyslexia protocol compared with care as usual in a cohort of 200 000 children entering primary school per year (euro, 2006)

School	Primary		Secondary		Post-secondary	
	Costs	QALY	Costs	QALY	Costs	QALY
Protocol	52 324 000	16 789	52 324 000	29 603	52 324 000	41 490
Care as usual	7 786 970	16 029	7 786 970	27 915	7 786 970	38 969
Difference	44 537 030	759	44 537 030	1 688	44 537 030	2 521
ICER	58 647		26 386		17 663	

This analysis suggests that long-term effects are an important determinant of cost-effectiveness.

To show the influence of the effectiveness of the results, we performed a sensitivity analysis that assumed the effectiveness to be 70%, which can be considered as a maximum. According to this scenario, the cost per QALY after primary school, secondary school and post-secondary school were respectively €33513, €16332 and €11558.

Assuming that 10% of the children with severe dyslexia in the usual care group would have improved to the normal level, the average ICER was estimated at €78197 per QALY at grade 1 and €29730 and €19102 per QALY, respectively, in the secondary school and post-secondary school.

If we assumed that the utility score for severe dyslexia was 0.80 instead of 0.77, the costs per QALY would be €87397, €30970 and €19606 for the primary school, secondary school and post-secondary school, respectively.

Furthermore, we tested the assumption that children treated according to the protocol would still use additional health care and remedial teaching. We assessed this assumption by adding these costs for the group of children in which treatment according to the protocol was not effective. In the base case, this was true for 60% of the children. Applying this scenario resulted in a relatively small increase of the total costs in the protocol arm and consequently only a relatively small increase of the ICER. The corresponding ICERs were respectively at primary, secondary and post-secondary school: €64231, €28898 and €19345.

The costs per QALY at the start of primary, secondary and post-secondary school decreased to respectively €49546, €22291 and €14922 if we assumed improved sensitivity of the diagnostic test of 50%. Overall, the sensitivity resulted in comparable findings to those of the base case.

DISCUSSION

The mean incremental cost-effectiveness ratio of the dyslexia protocol compared with care as usual was €58647 per QALY at the start of primary school and improved after secondary education. The long-term cost-effectiveness would generally be considered favourable when compared with current guidelines for use of cost-effectiveness in funding decisions (RVZ, 2006) and with cost-effectiveness of treatments currently included in the Dutch benefit package. Our analysis therefore suggests that the clinical effect is derived at reasonable costs.

However, our study has several limitations. We did not have clinical data on a direct comparison of Gramma with care as usual. Financial constraints did not allow applying a randomized controlled trial design evaluating the effectiveness of Gramma. Instead, the effectiveness of Gramma was compared with the effectiveness of care as usual as could be estimated on the basis of secondary data—that is data about the educational performance of children with dyslexia—about the effectiveness of care as usual. All children included in the intervention study received a phonics-based reading curriculum at school, had a history of reading and spelling problems and had received reading and spelling remediation at school and in many cases additional support (Gerretsen *et al.*, 2006). A considerable group of children included in the study were from grade 5 and higher. These children can be considered to have received care as usual from grade 1 to at least grade 4. That such care was not sufficient for these severely dyslexic children can be taken from their average reading and spelling scores when they entered the study:

mean standard score for reading for this group was 4.0 (SD 1.9) on a test with a mean of 10 and SD of 3; the mean percentile score for spelling for this group was 10.6 (SD 13.4). Both means demonstrate that they seriously lag behind their classmates. We acknowledge that this strategy does not generate a similar level of evidence as observational data collected in a randomized controlled trial.

The sensitivity analysis showed that assumptions about costs and effects have a moderate effect on ICER; thus, it would be worthwhile to collect additional data on the comparative effectiveness of the two treatment strategies.

Subjects were included in the effect study when reading and spelling of pre-treatment and post-treatment measures were available. When children dropped out of the treatment programme, it was protocol to strive for a post-treatment measure. This was irrespective of the reason why children (parents) decided to stop treatment and irrespective of the time they attended the treatment programme. Hence, dropouts were also included in the sample. Furthermore, experience has shown that the dropout rate of the programme was low. Even children that experienced severe problems and were the most resistant to treatment did not drop out. Many of these children had fruitlessly followed 'care as usual' and considered the treatment as a last resort. Therefore, it is plausible that the study population in the effect study may be considered as a representative sample of a dyslexic population.

In estimating the cost of the care as usual, we used a conservative approach, so that if any uncertainty remained, it would impact unfavourably on the cost-effectiveness of the dyslexia protocol. The computation of healthcare cost for the care as usual was limited to the first school year of education in writing and reading. We did include the costs of remedial teaching in the subsequent years, which seems according to routine practice. Furthermore, we did not take into account the cost of children who need to repeat a grade level in school for not meeting grade-level expectations. The chance that children with severe dyslexia need to repeat a grade level is higher than the average chance for all children (Blomert, 2002). So assuming that the probability to repeat a class is higher for the care-as- usual group, including these costs would be in favour of the protocol. We also did not consider the possibility that special education or supplemental teaching is erroneously offered to children with severe dyslexia. To offer a precise estimate of cost of dyslexia, we should have calculated the marginal cost per kind per year in different school types. We did not have enough data to make these calculations. The cost of the care as usual was therefore likely underestimated.

In the absence of an effective intervention, a part of the dyslectic children will visit GP, speech therapists, psychologists, physiotherapists and other medical care specialists (e.g. eye specialist). However, according to experts, the main part of these costs would be avoided by screening children who enter the primary school and offer an effective treatment to children with severe dyslexia.

It could be hypothesized that some of the costs at school will continue when children receive Gramma treatment, for example because they cannot work on the same speed as the average child or because they need extra support by a teaching assistant. However, in the Dutch school system, this type of individual support is not exclusively for children having reading difficulties, but it is used to ensure that the needs of each child are met in class. Hence, the Dutch school system supports children with a wide range of (learning) problems and also supports children who perform better in class than average and need to be challenged. We therefore feel that the costs of this type of support per child will be limited.

A limitation of the study is that the effects of treatment for dyslexia were estimated in terms of health-related quality of life by proxies by using a health utility questionnaire, the EQ-5D. Although dyslexia is indeed considered a medical problem, it is not evident that

the symptoms are also perceived as health problems. This raises the question of whether measures of health-related quality of life sufficiently captures the social and psychological well-being effects associated with dyslexia. It could be hypothesized that the effect of dyslexia treatments on self-esteem, autonomy and daily functioning are only partially captured in the QALY. The same may be true for other interventions in youth care. So further research on the relations between health-related quality of life, overall well-being and other effectiveness measures using patient-level data in youth is warranted.

The findings should be interpreted in light of the affordability and value of new treatments for dyslexia to society. Prior to development of the dyslexia protocol, no treatment for this condition was reimbursed. The current study suggests that severe dyslexia impairs the quality of life considerably in the eyes of people with medical training and that the treatment offers value for money. Recently, from among other decision criteria, namely effectiveness and necessity, besides cost-effectiveness, the Minister of Health agreed upon reimbursement of treatment of severe dyslexia according to the protocol. This encouraged us to write this paper.

This study illustrates that a significant part of the costs was related to diagnostics, making this a relatively important part of the intervention. The cost-effectiveness highly depends on the correct identification of the group of students with severe dyslexia as well as the treatment according to the protocol. We noted that not all therapists might welcome the stringent criteria for selection of patients who will be offered the therapy (mounting into a diagnose all/treat few approach), making this an important element to consider in future protocol refinements.

Economic evaluations are still limited in this field. A modelling approach is an important tool in determining the economic value of intervention for dyslexia. This study synthesized all the currently available evidence and demonstrated decision uncertainty resulting from knowledge gaps. Further research on cost-effectiveness in this field is strongly encouraged to reduce decision uncertainty. Nevertheless, the current analyses indicated that the protocol could be funded because long-term ICER has been established in a conservative way and still was well below thresholds typically considered a maximum.

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