Reading disability spectrum, subtreshold, full comorbidity and quality of life

PhD theses

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1. Introduction

1.1. Reading disability spectrum

Reading disability (RD) belongs under Neurodevelopmental Disorders in the group of Specific Learning Disorders (SpLD) in the Diagnostic and Statistical Manual of Mental Disorders, 5th edition (DSM-5). It manifests in childhood and is not caused by impairments in general intelligence or in hearing and vision, inadequate schooling, or motivation and poor socio-cultural status. Children with RD face with several difficulties in their academic performance due to this impairment, which can cause anxiety, low self-esteem, behavior problems and difficulties in social interactions as well. Several studies have described the high comorbidity of RD with both externalizing and internalizing disorders.

1.2. RD early/late recognation, subthreshold psychopathology and comorbidity

Early contact to specialists, i.e. before the age of 7 years (in preschool or first grade) and prompt, effective interventions are critical for achieving good outcomes in children with RD. Early intervention depends on recognition of early signs of RD. Several recent studies suggested that the prevalence of psychiatric disorders was higher in individuals with RD than those without RD. Recently subthreshold psychopathology –

symptoms which are insufficient to fulfill diagnostic criteria of classification systems – have attracted considerable research attention. Studies have shown that even subthreshold psychopathology has a negative impact on functioning and is associated with increased risk of developing a full symptom disorder.

To date, according to our knowledge, no study has focused on comorbid subthreshold psychopathology in children with RD, moreover we have not found any published research comparing full syndrome or subthreshold psychopathology in children whose RD was recognized early and late.

1.3. Quality of Life

The concept of Quality of Life (QoL) of patients with mental health disorders gained increasing importance in the last decades. Assessing the functional status of the patients had been important previously, only it has been included into the diagnoses according to the classification systems. The concept QoL is much broader. The definitions of QoL consist the somatic, social and psychological areas. Rating QoL, subjective aspects come ahead, however recent studies highlighted, that in surveying the QoL of children, we cannot solely rely on the self-report judgment of the children, a proxy

report is also needed. It might be interesting, how much the child's and parent's report are the same.

1.4. RD and QoL

There is very little research on the QoL in children with RD. Additionally all these studies examined children with SpLD and not only children with RD. These data show that several aspects (e.g., emotional well-being, self-esteem and satisfaction in their relationships with family and friends) of self-perceived and parent-reported QoL for children with newly-diagnosed SpLD are poorer than the QoL of typically developed children. Comparing the QoL of children with SpLD and typically developed peers, a significant differences was found between the parent-child proxy ratings: mothers of children with SpLD reported significantly lower scores regarding the child's functioning in school, but significantly higher scores in their physical and emotional well-being. However, between typically developed children and their mothers' proxy ratings, significant differences were found in physical well-being and self-esteem; in both cases parents reported higher scores.

2. Objectives

2.1.First part of the PhD work

- We aimed to examine the prevalence of comorbid psychiatric disorders and subthreshold psychiatric disorders with RD.
- We aimed to compare the prevalence of comorbid psychopatology in children diagnosed / or recognized the risk of RD at early and late (= 7 years of age).

2.2. Second part of the PhD work

- We aimed to examine the QoL of children in both RD and healthy control groups both self- and parent-reports.
- We aimed to examine comorbidity of RD and the QoL of children with RD.
- We aimed to examine age and gender differences in the above questions.

3. Methods

3.1. Participants

The study was approved by the Ethical Committee of the Ministry of Human Capacities, Hungary. The parents of each child and children older than 14 years included into this study provided written informed consent after being informed of the nature of the study. Children under 14 years received both written and oral information about the project.

Study participants were recruited from Vecsés City Local Government Department of Children and Family Services, Vecsés, Hungary. Inclusion criteria were children had to be <18 years and been diagnosed with an RD, according to a computer-based dyslexia test. A control population from the same age group was recruited from the local community through word of mouth; here, the inclusion criterion was not having an RD, according to the dyslexia test. For both study groups, the exclusion criterion was having had a diagnosis of mental retardation in the medical history. Additionally, in the control group, exclusion criterion was any current or previous psychological or psychiatric treatment in the medical history.

3.2. Measures

To verify RD, we used the shortened version of the Dyslexia Differential Diagnosis, Maastricht, Hungarian standard Test (3 DM-H). Psychopathology was evaluated using the Hungarian version of the Mini International Neuropsychiatric Interview Kid (M.I.N.I. Kid) ant the Strengths and Difficulties Questionnaire (SDQ). In this study, we used the parent-reported version of the scale. QoL was measured by the Hungarian version of the ILK scale. A structured interview was also performed to determine demographic data characteristics. The characteristics upon on which we collected data included

e.g. parents' education, parents' economic activity, family structure, early adaptation, the age when the risk/early warning signs of RD or RD had been diagnosed and the intervention had been started for it.' We assessed psychiatric pathology with the modified version of the Hungarian Mini International Neuropsychiatric Interview Kid (M.I.N.I. Kid).

3.3. Statistics

3.3.1. First part of the PhD work

The prevalence of full and subthreshold internalizing and externalizing disorders is reported. Group comparisons (RD group vs. control group; early vs. late RD) of pathologies were carried out using chi-squared tests. To explore the effect of RD severity on comorbidity within the RD group, we compared groups of children categorized as below average, poor, and very poor 3DM-H CRS-index scores.

3.3.2. Second part of the PhD work

In a bivariate analysis, we explored group differences in self- and parent-reported QoL and psychopathology in the total sample, separately for girls and boys, and their associations with age. Because of deviations of normality of the distributions of study variables, we used non-parametric tests (Mann-Whitney U-tests and Spearman's correlations).

In the multivariate analyses we investigated the relationships between RD, comorbid psychopathology, and QoL as dependent on gender. We tested moderated mediational models in which comorbid psychopathology, as measured by SDQ Difficulties scales, mediated the relationship between group membership (RD = 1, control = 0) and self- and parent-rated QoL, which was dependent on gender (boys = 0, girls = 1). Logarithmic transformations were used to assure normality. Bootstrapping with a resample procedure of 1000 bootstrap samples (bias corrected and accelerated (BCa) estimates and 95% CI) was used for significance testing, because this method does not impose the assumption of normality of the sampling distribution. Child's age and parents' level of education were included as covariates.

4. Results

4.1. Participans

There were 130 children in the RD group and 82 in the control group. There were no differences between the control group and RD group in terms of mean age (RD: 10.23 years, SD=2.12; control: 9.66 years, SD=2.12) or in gender distribution (RD: girls=40.0%; control: girls=39.0%).

4.2. First part of the PhD work

4.2.1. Subthreshold and full psychiatric comorbidity of RD

Older children had more subthreshold (rho=.196, p=.004) and full diagnoses (rho=.219, p=.001) than younger children. There was no gender difference for any of the mood or anxiety disorders, but boys were more frequently diagnosed with externalizing disorders (boys N=32, 25.0%; girls N=10, 12.0%; $\chi 2$ (1)=5.298, p=.021, ϕ =.158).

There was a difference in M.I.N.I. Kid diagnoses of internalizing and externalizing disorders between the RD and control groups ($\chi 2$ (2)=10.894, p=.004, Cramer's V=.227). Eighty-two children (63.6%) in the RD had no diagnosis, 16 (12.4%) had one diagnosis and 31 (24.0%) had two or more M.I.N.I. Kid diagnoses. Sixty-nine children (84.1%) in the control group had no M.I.N.I. Kid diagnosis, 6 (7.3%) had one diagnosis and 7 (8.5%) had two or more diagnoses. Anxiety, internalizing and externalizing disorders were more prevalent in the RD group.

When subthreshold and full diagnoses were considered together, anxiety and internalizing diagnoses, but externalizing diagnoses were more frequent in the RD group than in the control group. To explore the effect of RD severity on comorbidity within the RD group, we compared groups of children categorized as below average, poor, and very poor 3DM-H CRS-index scores. There were no significant

differences in the three groups in M.I.N.I. Kid diagnoses of internalizing and externalizing disorders ($\chi 2$ (2)=.025, p=.988, Cramer's V=.014 and $\chi 2$ (2)=.956, p=.620, Cramer's V=.086, respectively). Similarly, no differences among the three groups were found, when subthreshold conditions were also included (any full or subthreshold internalizing and externalizing disorders: $\chi 2$ (2)=.242, p=.886, Cramer's V=.042 and $\chi 2$ (2)=3.019, p=.221, Cramer's V=.153, respectively).

4.2.2. Early and late recognition RD sub-groups

The early recognition RD sub-group was younger than the late recognition RD sub-group (M=9.93, SD=2.23 and M=10.98, SD=1.87, respectively; t(110)=2.575 p=.011), but the two sub-groups did not differ in terms of gender, father's or mother's level of education or economic activity or family structure. There were also no sub-group differences in complications during pregnancy and birth, prematurity or early development.

There were sub-group differences in number of M.I.N.I. Kid internalizing and externalizing diagnoses ($\chi 2$ (2)=6.861, p=.032, Cramer's V=.248). Fifty children (72.5%) in the early RD sub-group had no diagnosis, 8 (11.6%) had one diagnosis and 11 (15.9%) had two or more. Twenty-one children (48.8%) in the late RD group had no diagnosis, 7 (16.3%) had one

diagnosis and 15 (34.9%) had two or more. Frequencies of M.I.N.I. Kid subthreshold and full diagnoses in the early and late RD sub-groups. The prevalence of anxiety and mood disorders was similar in the two sub-groups, but the prevalence of externalizing disorders was higher in the late RD sub-group. When subthreshold and full diagnoses were considered together the prevalence of mood and externalizing diagnoses was higher in the late RD sub-group than the early RD sub-group.

4.3. Second part of the PhD work

Self- and parent-reported QoL ratings were significantly and positively related; the effect size was at a medium level, however. We found significant positive correlations of medium effect sizes among child's age and both self- and parent-reported ILK scores, indicating lower levels of QoL with increasing age. SDQ scores were not related to age. High levels of psychopathology were associated with low levels of QoL according to both self- and parent-reports .In the total sample, after a Bonferroni correction ($\alpha' = 0.05/7 = 0.007$), the RD group showed higher ILK scores in the school ($U = 3674.0$, $p < 0.001$), family ($U = 3659.5$, $p < 0.001$), time spent alone ($U = 3972.5$, $p = 0.003$), and mental health ($U = 3472.5$, $p < 0.001$) domains and general QoL ($U = 3662.5$, $p < 0.001$), according

to the parent-reports, while no differences between RD and control groups were found, according to the self-reports. According to parent reports, the ILK total score was also higher in the RD group than in the control group ($U = 3375.5$, $p < 0.001$), but no differences were found between study groups, according to self-reports. For boys, the RD group showed higher ILK scores in the family ($U = 1299.0$, $p < 0.001$) and mental health ($U = 1127.0$, $p < 0.001$) domains regarding general QoL ($U = 1165.5$, $p < 0.001$) as well as ILK total scores ($U = 1155.0$, $p < 0.001$), according to parent-reports, and again, no differences between the two groups were found, according to self-reports. For girls, the RD group showed higher ILK score in the school domain ($U = 486.5$, $p = 0.004$), according to parent ratings, but no differences were found between the two groups, according to self-reports. In the total sample, the two study groups did not differ in the total SDQ difficulties score. For boys, the RD group showed significantly higher scores in SDQ's total difficulties score than controls ($U = 1407.5$, $p = 0.007$), while in girls, no differences were found. Group status and gender, as well as the interaction term of these variables were significantly related to SDQ scores, when controlling for child's age and parents' level of education. A post-hoc analysis revealed that for boys, having

an RD diagnosis was associated with higher SDQ scores, while in girls SDQ scores did not differ between the RD and control groups. In the first model, self-rated ILK scores were related to SDQ total difficulties score, gender, and the interaction term of these variables. Group membership and the interaction term of group membership by gender were not associated with self-reported ILK scores, again, when controlling for child's age and parents' level of education. The post-hoc analysis revealed that, for boys, higher SDQ scores were related to higher ILK scores, but no relationship was found for girls .The conditional direct effects of group membership on self-rated ILK scores were non-significant for both boys (= −0.046, BCa 95%, CI: −0.135 to 0.043) and girls (= −0.050, BCa 95%, CI: −0.161 to 0.061). The conditional indirect effect of group membership on self-rated ILK scores through SDQ total difficulties score was significant for boys (= 0.064, BCa 95%, CI: 0.024–0.111), but non-significant for girls (= −0.004, BCa 95%, CI: −0.041 to 0.008), and the difference between the conditional indirect effects was significant (= −0.068, BCa 95%, CI: −0.120 to −0.022). In the second model, parent-rated ILK scores were related to SDQ total difficulties score and group status. Gender and the interaction terms of group membership by gender as well as SDQ scores by gender were not associated with parent-

reported ILK scores, again, when controlling for child's age and parents' level of education. For both boys and girls, higher SDQ scores were related to higher ILK scores. The conditional direct effect of group membership on parent-rated ILK scores was significant for boys (= −0.098, BCa 95%, CI: 0.012–0.184), but not for girls (= 0.079, BCa 95%, CI: −0.028 to 0.186). The conditional indirect effect of group membership on self-rated ILK scores through SDQ total difficulties score was significant for boys (= 0.073, BCa 95%, CI: 0.029–0.127), but non-significant for girls (= −0.020, BCa 95%, CI:−073 to 0.013), and the difference between the conditional indirect effects was significant (= −0.093, BCa 95%, CI: −0.164 to −0.034).

5. Conclusion

My doctoral dissertation's central thread consists of two elements related to RD: comorbidity and QoL.

5.1. First part of the PhD work

Our findings are consistent with earlier research demonstrating the importance of recognizing and providing appropriate treatment for RD comorbidities. In our sample more than one third of children with RD had at least one psychiatric comorbidity, and almost one fifth had more than one psychiatric comorbidity. There was a higher prevalence of

psychiatric disorders in the RD group than in the control group, but surprisingly almost one-sixth of the control group - consisting by definition of children without any history of psychological or psychiatric treatment - had at least one psychiatric disorder according to a structured interview. This result suggests that introduction of a more sensitive secondary prevention system among children should be considered. Particular attention should be given to screening children with RD for potential psychiatric comorbidity. The prevalence of both subthreshold and full internalizing disorders was higher in the RD group than the control group. The prevalence of full anxiety disorders and the combined prevalence of full and subthreshold anxiety disorders were higher in the RD group than in the control group. About one quarter of RD children had at least one anxiety disorder, and if subthreshold conditions were included this proportion increased to two thirds. We conclude that children with RD experience more stress than their typically developing peers as they try to meet the expectations of parents, teachers and peers. Interestingly, similar proportions of children in the RD and control groups exhibited some externalizing symptoms, but a higher proportion of children in the RD group fulfilled the criteria for an externalizing disorder. In both RD sub-groups (early and

late recognition) the percentage of children with comorbid psychiatric disorders was very high: more than half the children late RD sub-group and more than one quarter of the children in the early RD sub-group had at least one comorbid disorder. Our data also show that the prevalence of psychiatric disorders according to DSM-IV criteria was higher in children whose RD was recognized late (after 7 years) than in children whose RD was recognized before the age of 7 years. These results emphasize the importance of early recognition of RD and reinforce the importance of training teachers in kindergarten, preschool and grade school to recognize the early symptoms of RD and persuade the parents of affected children to seek professional help.

5.2. Second part of the PhD work

Our study extends several aspects of the existing, but very limited data on the QoL of children with an RD. We measured the QoL of the children from both RD and healthy control groups, and we supported the data from population and clinical studies: QoL decreases with increasing age in childhood according to both self- and parent-reports. During adolescence, children experience a transition period, as they face several changes, including psychical changes, new social roles (e.g., being less dependent on parents yet still living at home, finding

their place among peers, having their first girlfriends or boyfriends), and experiencing psychological changes; they need time and good coping mechanism to adapt. The tension of these changes can lead to lower QoL. In the current study higher level of psychopathology was associated with lower QoL in our study population, when we included both RD and healthy control groups and boys and girls together, again, based on both self- and parent-reports. When we compared the QoL of the RD and the control groups, based on both self- and parent-reports, the parents in the RD group rated several aspects (i.e., school, family, mental health, and general QoL) of their children's QoL as worse than parents rated the QoL of their control group children and the children with an RD did not rate their QoL lower in the above mentioned domains as their healthy pears did. It is very interesting when we measured gender differences we got the same result for boys; however, in the case of girls, not only self-reported, but parents-reported QoL of children with RD did not differ from the control group, except in the school domain. Our results, especially in the case of boys, belong to the line of those studies, which highlight that there is a discrepancy between self- and parent-rated QoL of children. It is not so surprising that parents of boys with RD rated their children's QoL lower than did the parents of healthy

boys, because they know all the difficulties their children experience due to their RD. This result for boys even supports those very few previous studies on children with SpLD, where parents reported about their children QoL. However, it is interesting why parents of girls with RD did not rate their children's QoL lower in many domains, except school, than did the parents of healthy girls. However, in our study, children (both girls and boys) did not rated their QoL lower on the dimension of family and general QoL than did healthy children.

Finally, as we expected, high level of psychopathology was associated with low parent-rated QoL in the cases of boys and girls. It draws the attention of clinicians to the importance of recognizing and treating comorbidity in conjunction with an RD.

In conclusion, the results of this study indicate that QoL is a useful measure of well-being in the case of children with an RD, and they highlight the importance of measuring comorbid psychopathology, even on a subthreshold level and accounting for gender and age differences. Moreover, we would like to underline the importance of getting information on psychopathology and QoL from more source.

6. Bibliography of the candidate's publications:

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