Reading comprehension difficulties in children with rolandic epilepsy

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ABSTRACT

Aim Difficulties in reading comprehension can arise from either word reading or listening comprehension difficulties, or a combination of the two. We sought to determine whether children with rolandic epilepsy (RE) had poor reading comprehension relative to controls, and whether such difficulties were associated with word reading and/or general language comprehension difficulties.

Method In this cross-sectional study, children with RE (*n*= 25; 16 males; mean age 9 years:1 month, SD 1 year :7 months) and controls (*n*= 39; 25 males; mean age 9:1, SD 1:3) completed assessments of reading comprehension, listening comprehension, word/non-word reading, speech articulation and non-verbal IQ.

Results Reading comprehension and word reading were worse in children with RE, $F(1, 61) = 6.89$, $p = .01$, $\eta_p^2 = .10$ and $F(1, 61) = 6.84$, $p = .01$, $\eta_p^2 = .10$, with listening comprehension being marginal, $F(1, 61) = 3.81$, $p = .06$, $\eta_p^2 = .06$. Word reading and listening comprehension made large and independent contributions to reading comprehension, explaining 70% of the variance.

Interpretation Children with RE may be at risk of reading comprehension difficulties. Thorough assessment of individual children is required to ascertain whether the difficulties lie with decoding text, and/or with general comprehension skills.

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What this paper adds:

- Children with RE may be at risk of poor reading comprehension.
- For some children, this was related to poor word-reading skills, whereas for others it was related to poor listening comprehension abilities, or both of these combined.
- Educators and speech therapists need to conduct assessments of word reading and listening comprehension, in addition to reading comprehension, to tailor interventions to the profile of difficulties presented by individual children with RE, when remediating poor reading comprehension.

RUNNING FOOTER: Rolandic epilepsy and reading comprehension

Reading comprehension difficulties in children with rolandic epilepsy

Rolandic epilepsy (RE: or benign epilepsy with centrotemporal spikes, BECTS) is reported to account for 8 to 25% of all cases of childhood epilepsy in the 5 to 14 year range. [1,2] The onset occurs between 3 and 13 years and the condition resolves by adulthood. [1] Children with RE have an increased risk of reading problems in school [3] where reading comprehension is essential for learning across the curriculum.[4] More broadly, reading comprehension in school is a critical determinant of academic and professional success. [5] Two sets of skills enable reading comprehension: word recognition skills and listening comprehension.[4] These can fail independently, or together, resulting in poor reading comprehension. Poor word recognition can result in poor reading comprehension because slow or inaccurate word retrieval processes can lead to an information processing bottleneck.[6] In contrast, specific reading comprehension difficulties have been documented in children who have age-appropriate word reading but weak listening comprehension. [7]

A recent meta-analytic review confirms poor single word reading in children with RE [1], and language difficulties at the word, sentence and discourse level have been found [8] in addition to teacher reports of difficulties specifically with reading comprehension. [9] However, studies reporting standardised measures of discourse-level reading or listening comprehension for children with RE are lacking. Our primary aim was to determine if discourse-level reading comprehension is an additional area of weakness in this population. To do this, we examined reading comprehension in a group of RE children relative to age and gender matched controls. A second main aim was to examine the contributions made by word reading and listening comprehension in the prediction of reading comprehension. It is important to understand the source(s) of any reading comprehension difficulties associated with RE, to target appropriate remediation.[6]

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Pal and colleagues have shown that children with RE have an increased risk of speech sound disorder (SSD), a developmental condition that resolves around 5-6 years [3] and is associated with reading difficulties in non-RE populations.[10] Children with RE can also show more generalised dyspraxic difficulties [11] which have been related to literacy difficulties.[12] Thus, we measured SSD and Developmental Coordination Disorder (DCD) in our sample (through parental questionnaire) to ascertain if any reading difficulties were associated with these comorbidities. We also measured children’s ability to pronounce different speech sounds to determine the extent of current articulation difficulties, given the early resolution of SSD.

Children with RE completed assessments of reading comprehension, single word and non-word reading, and listening comprehension. We also collected information about their speech and motor skills. The following questions were explored:

1. What is the level of reading ability in children with RE in comparison to typically developing controls? Based on previous research, we predicted that children with RE would do worse on measures of real word and nonword reading, and reading and listening comprehension.

2. Are reading comprehension outcomes in RE predicted by word reading and listening comprehension, over and above age and general cognitive ability, to the same degree as found for typically developing controls, or is one component skill the more critical determinant, as in populations with predominantly word reading difficulties or listening comprehension problems (e.g., children with dyslexia or poor comprehension)?

3. Is there any evidence that SSD or DCD are more likely associated with reading difficulties in children with RE than in controls?

4. Are clinical variables (age of onset, medication status) associated with reading difficulties?

METHOD

Participants

Children with RE were identified between October 2013 and December 2014 at 11 participating hospital trusts in Northern England. The inclusion criteria were children aged between 6 and 12 years with at least two observed seizures and confirmatory EEG, as assessed by a paediatric neurologist (either CdeG, HB or AI). All were English-speaking. In terms of sample size, there was no prior basis for estimating the effect size of the reading comprehension difference between RE children and controls. Therefore using the average effect size of 0.72 reported in a recent meta-analysis for single word reading[1], an alpha level of 5% and power of 80%, a sample size of 32 RE children and 32 controls is indicated.

A research nurse from the Medicines for Children Research Network together with local site Principal Investigators identified potential cases and made initial contact. Interested families gave consent to be contacted by the research team. Following testing (January 2014 to July 2015) the medical notes of the final sample of children were reviewed by one of three Consultant Paediatric Neurologists (CdeG, HB, AI) for full clinical data. Four children were excluded at this stage due to insufficient evidence for rolandic epilepsy (see Figure 1). Our final sample was 25 children.

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The control sample was recruited from three mainstream primary schools in the North West of England and via research study advertisement at Lancaster University. They had no known neurological or neurodevelopmental conditions, or diagnosed reading difficulties (based on parental report), and spoke English. From sixty initial recruits, only those with a birthday within six months of a child with epilepsy and the same gender and school year group were included in the data analyses reported here. As a result all RE children had at least one match but some had several matches, one child did not have a gender match. This gave an overall control sample of 39. See Table I for participant characteristics.

A National Health Service Research Ethics Committee (North West – Liverpool East) and a University Research Ethics Committee approved the study. Parents of the RE and control sample gave written consent and children gave verbal consent prior to commencement of testing.

Measures

Children completed standardised tests assessing reading and listening comprehension, word reading, non-verbal IQ and speech articulation. Re-test reliability (unless otherwise stated) and test validity are referenced for each from the test manual or papers assessing validity. Parents completed questionnaires on their child's language and motor coordination.

The York Assessment of Reading Comprehension [13] (YARC) was completed. Children read aloud two short stories and answered eight open-ended comprehension questions after each one. The time taken to read each text, the number of word reading errors made, and the number of correct responses to the comprehension questions were used to calculate separate scores for reading rate (reliability = .90 - .95, depending on age), word reading accuracy (.75 - .93) and reading comprehension

(Cronbach's = .71 - .93), respectively. [13] The test has validity relative to an alternative standardised measure of comprehension. [14]

Children completed measures of sight word efficiency and phonemic decoding efficiency from the Test of Word Reading Efficiency (TOWRE) Second Edition[15]. The number of correctly read words/non-words in 45 seconds was recorded. This test has good reliability (.86 to .93) and validity, demonstrated by correlations with other assessments of word reading.[15]

The Understanding Spoken Paragraphs subtest of the Clinical Evaluation of Language Fundamentals (CELF) 4th edition assessed listening comprehension.[16] Children heard three stories and answered open-ended comprehension questions after each one. Reliability is adequate to good (range of .51 to .87 depending on age). Validity is good, evidenced by correlation with other subtests and discrimination in terms of performance by typical and clinical samples.[16]

Non-verbal IQ was assessed using the matrix reasoning subtest from the Wechsler Intelligence Scale for Children IV-UK. [17] This subtest has good reliability (.71 to .85, dependent on age) and validity (correlation of .84 with overall performance on WISC perceptual reasoning index).[17]

Parents completed a questionnaire incorporating items used in previous research [3, 10] to assess the presence of SSD early in development. Children with a reported history of speech and language problems, who had received speech/language therapy, and had delayed production of one- and two-word utterances are considered likely to have experienced this disorder. [3, 10] On this basis, we coded children who met all of these criteria as possible SSD cases (see Table I). Children's articulation of consonant sounds in English was assessed with the Goldman-Fristoe Test of Articulation (G-F)[18]. Reliability is high (.98) and validity has been demonstrated by comparison with a Canadian English-speaking typical sample. [18]

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Parents completed a DCD questionnaire, (DCDQ'07)[19] with good reliability (.89, Cronbach's alpha) and clinical validity.[19] Cut-off scores for possible DCD are provided in relation to age.

Procedure

Children were tested individually at their home, school or the university, depending upon parental preference. All assessors were experienced testers with PhD level training. The measures were part of a larger battery of language, literacy and memory assessments and were administered in the same order for each child, with appropriate breaks. Twenty children were assessed in several sessions split over the course of a day; the remainder in sessions spread over several weeks.

Statistical analyses

Given the wide age range being studied we used raw scores in all analyses, entering age as a predictor or covariate where appropriate. To examine group differences, we conducted Analysis of Covariance (ANCOVA) for the measures of reading comprehension (YARC), listening comprehension (CELF) and non-verbal IQ (WISC), with age entered as a covariate. All assumptions of the test were met (normal distribution of measures and homogeneity of variances). For word reading, we formed a composite score averaging word reading accuracy and reading rate from the YARC, and word and non-word reading from the TOWRE-2, as these were highly inter-correlated (*r*s =.87-.95). An ANCOVA with age as a covariate was conducted on this score. Fisher's exact tests were conducted to compare the number of children in the RE and control sample with significant impairment on the measures (excluding non-verbal IQ and listening comprehension, which are designed to be used as part of a composite).

To determine whether word reading and listening comprehension make independent contributions to reading comprehension, as found in prior research,[4] a general linear modelling (GLM) forward-fitting approach was adopted to model the effects of age, non-verbal IQ, group (RE and Controls), word reading ability (composite), and listening comprehension, on the YARC reading comprehension score. The Bayesian Information Criterion Index was used to ascertain the best-fitting model. All assumptions of this test were met and the residuals were checked on the model of best fit (reported in Table II).

Several clinical variables were examined in relation to the study measures, using raw scores. Age of onset was analysed using partial correlations controlling for age. Separate ANCOVAs of sub-groups were conducted for medication status (AED, none, control) with age as a covariate. Planned analyses of sub-groups based on seizure frequency and occurrence of SSD were not conducted due to low numbers (see Table II). Partial correlations controlling for age examined whether DCD scores correlated with any reading measures. G-F articulation measures were assessed by ANCOVA (group differences) and partial correlation (relation to reading and comprehension measures).

RESULTS

Differences between rolandic epilepsy and control groups

Figure 2 provides the mean differences and 95% confidence intervals (z-scores) for each literacy measure. Single factor ANCOVAs with group (RE, Control) and age entered as a covariate were conducted on the raw ability scores. Reading comprehension and word reading composite scores were significantly lower in RE children than controls, $F(1, 61) = 6.89$, $p = .01$, $\eta_p^2 = .10$, and $F(1, 61) = 6.84$, $p = .01$, $\eta_p^2 = .10$, respectively. Although children with RE performed worse on the test of listening comprehension this was marginal $F(1, 61) = 3.81$, $p = .06$, $\eta_p^2 = .06$. Non-verbal IQ was significantly worse in the RE sample, $F(1, 61) = 10.53$, $p = .002$, $\eta_p^2 = .15$.

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Table I reports the number of children in the RE and control samples with significant impairment (one standard deviation below standard score mean) on each measure (excluding non-verbal IQ and listening comprehension). A significantly greater number of children in the RE sample fell into this category for YARC reading accuracy ($p = .02$, Fisher's exact test) and TOWRE sight word reading ($p = .04$), and marginally more for YARC reading comprehension ($p = .06$). The p values on all other measures were greater than $p = .19$.

Relation between word-reading ability, listening comprehension and reading comprehension

The effects of age, non-verbal IQ, group (RE and Controls), word reading (composite), and listening comprehension, on predicting the reading comprehension score were examined. Table II shows the model of best fit (accounting for 70% of variance in reading comprehension scores), which included significant main effects of word reading and listening comprehension, demonstrating that each make significant independent contributions to reading comprehension, to similar degrees in both the RE and control group.

Effects of clinical variables on reading comprehension, word-reading and listening comprehension.

Single factor ANCOVAs examining the effect of medication status (RE no medication, RE medication, controls) with age as a covariate, were conducted. Table SI reports the means and SDs (in standard scores) for the reading and comprehension measures as a function of medication status. For reading comprehension, children in the RE medication group were poorer than controls: $F(2, 60) = 6.00$, $p = .004$, $\eta_p^2 = .17$; mean difference = -10.44, 95% CI -17.87-(-3.02). The same pattern was found for word reading (composite): $F(2, 60) = 5.67$, $p = .006$, $\eta_p^2 = .16$; mean difference = -12.93, 95% CI -22.39-(-3.47), and also non-verbal IQ: $F(2, 60) = 8.13$, $p = .001$, $\eta_p^2 = .21$; mean difference = -5.66, 95% CI -9.12-(-2.20). In all comparisons, RE children not in receipt of AEDs did not differ significantly from the other two groups in the post hoc tests. There was no effect of group on listening comprehension, $F(2, 61) = 2.26$, $p = .11$, $\eta_p^2 = .07$. There were no significant (partial) correlations between age of onset and any study measures.

Are SSD or DCD more likely to be associated with reading difficulties in children with RE than in controls?

The occurrence of SSD was too low (RE: $n = 4$, TD: $n = 1$) to permit analysis. However, children with RE were more likely to have articulation difficulties than controls. A single factor ANCOVA on the raw G-F test scores with group (RE, control) as a factor and age as a covariate revealed a significant effect of group, $F(1, 62) = 5.19$, $p = .03$, $\eta_p^2 = .08$, although all children scored within age appropriate range (all scores > 85). There were no significant correlations between the G-F test scores and the literacy measures (all *rs* < -.32, all *p*s > .05). Of the four RE children with a history of SSD, two had significant word reading problems (more than one standard deviation below the standardised score mean).

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For children with RE there was a significant correlation between the overall raw score on the DCD questionnaire and the word reading composite, $r(22) = .52$, $p = .009$, CI .09 - .78. Correlations with all other literacy measures for both groups were small (all *rs* < -.35, all *p*s > .06).

DISCUSSION

In line with predictions, children with RE were significantly poorer than controls on measures of word reading, reading comprehension and non-verbal IQ. The RE group was heterogeneous; not all had poor reading comprehension and, where apparent, there was evidence that neither poor word reading nor listening comprehension was the consistent predictor. Our results support a recent meta-analysis highlighting word reading ability as a potential weakness in children with RE[1]. However, our study identifies reading comprehension as an additional area of vulnerability.

Word reading and listening comprehension made independent contributions to reading comprehension performance for both groups. The contributions of these variables were in addition to the variance explained by IQ, consistent with other work on reading in non RE populations[20, 21]. Recent work reports differences between children with RE and controls on language measures regardless of IQ, although differences were larger when IQ was low.[1] Taken together these findings support the argument that IQ is not the main independent cause of the word reading and reading comprehension difficulties children with RE may experience. Future work examining the relation between a broader range of receptive language and cognitive skills, such as working memory and executive skills, on reading comprehension outcomes is required to establish the critical pressure points in the reading system for children with RE[22].

Although the occurrence of SSD was low, there was some evidence of residual articulation difficulties for children with RE. Our study supports the proposal that word reading difficulties in children with RE may be associated with speech or more generalised dyspraxic difficulties.[3, 11, 12, 23] However, neither SSD nor DCD was evident in *all* children with RE who had reading difficulties. Whether dyspraxia is co-occurring or causally related to the reading difficulties associated with RE, at least for some children, is a question for future research.

In terms of clinical variables, the lack of relation between our reading and comprehension measures and age of onset is consistent with other studies [8, 24]. Our findings of worse performance on reading measures relative to controls for the RE group on medication is consistent with the findings of Ay and colleagues[25]. However, in our sample, it is quite possible that medication status was confounded with a more complex epilepsy, so causal inferences cannot be drawn.

Limitations of the study include the use of only a single indicator for each of two key variables, reading and listening comprehension, resulting in a somewhat narrow assessment of these complex constructs. Additionally, our sample was smaller than expected, due to the difference between diagnostic decisions occurring at secondary (general paediatric) versus tertiary (paediatric neurology) levels, potentially resulting in a loss of power for reliably detecting differences in listening comprehension between RE and control groups. Our sample size was in line with other recent cross-sectional studies of children with RE [1], however.

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To summarise, in this first study examining the full reading profiles of children with RE, we found that reading comprehension was compromised, in addition to previous reported difficulties with word reading. Critically, we demonstrated that reading comprehension was determined by both word reading and listening comprehension. Despite the limitations noted, educators and clinicians working with children with RE should consider these children to be at risk of word reading and listening comprehension difficulties. Based upon the profile of difficulties presented by individual children with RE, intervention for one or both of these aspects of reading may be required, to support the ultimate goal of reading comprehension.

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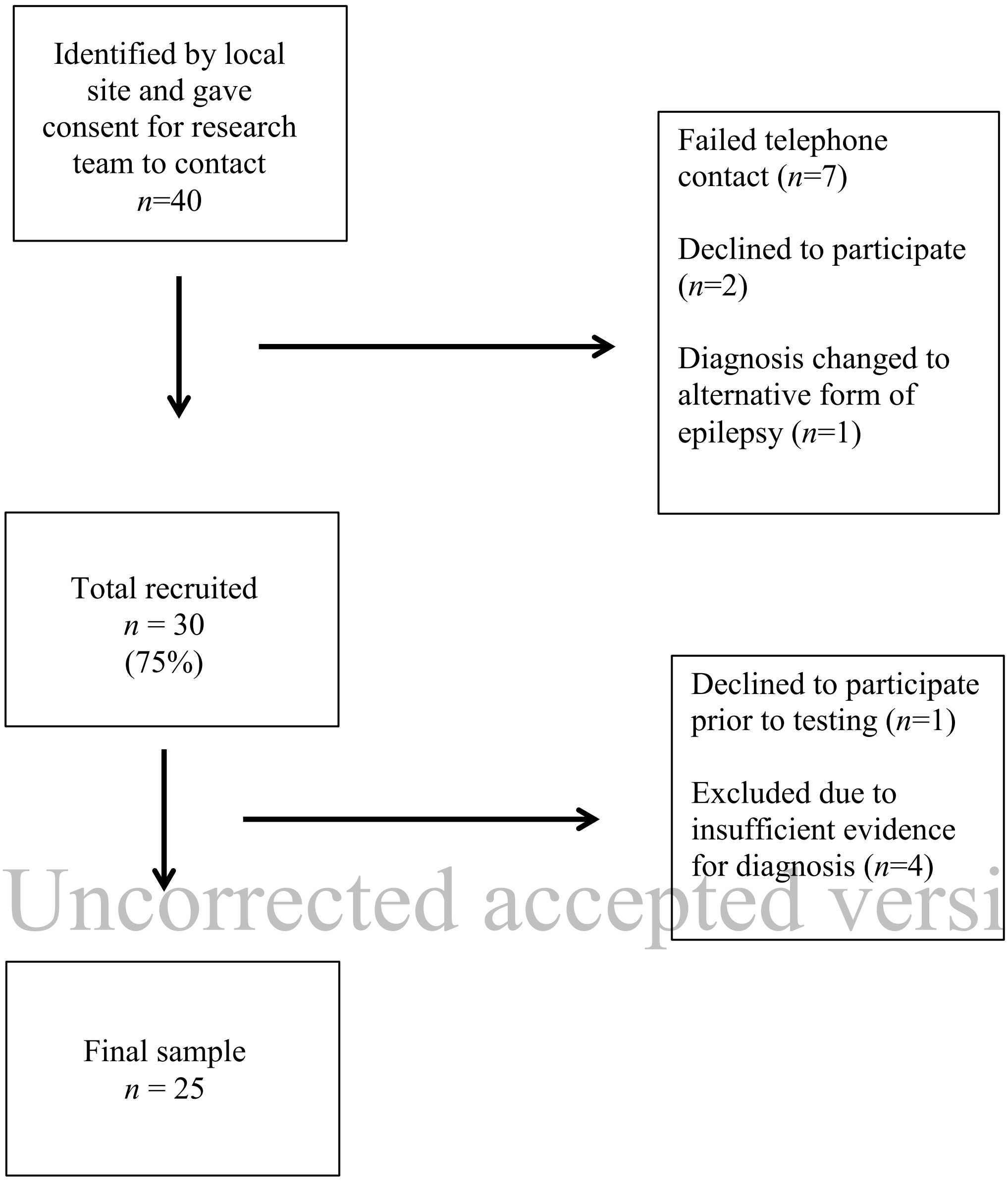

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Figure 1. *Participant recruitment for the children with rolandic epilepsy.*

Table I. *Participant characteristics and study measures expressed as standardised scores*

	Rolandic epilepsy (*N* = 25)	Control (*N* = 39)
Gender Male/Female	16/9	25/14
Mean age, y:mo (SD)	9:1 (1:7)	9:1 (1:3)
History of speech/language problems [a]	7 (29.2%)	3 (10.0%)
Received Speech Therapy	6 (25.0%)	3 (10.0%)
Indication of Speech Sound Disorder	4 (16.7%)	1 (3.3%)
Indication of possible Developmental Coordination Disorder	13 (54.2%)	3 (9.7%)
Clinical Variables n (%)		
Age at onset, y:mo (SD)	7:0 (1:10)	
Antiepileptic medication		
None	12 (48.0%)	
Monotherapy	12 (48.0%)	
Polytherapy	1 (4.0%)	
Laterality (n)[b]		
Right 9 (36.0%)	Bilateral	11 (44.0%)
Left 5 (20.0%)	Evolving	6 (24.0%, 2 left, 1 right, 3 bilateral)
Seizure type		
With motor/autonomic components	25 (100.0%)	
Sensory/psychic phenomena	10 (40.0%)	
Evolving to bilateral convulsive seizure	13 (52.0%, 4 with sensory/psychic phenomena)	
Atypical (n)		
Status Epilepticus 1 (4.0%)	Todds Paresis	4 (16.0%)
Diurnal seizures 8 (32.0%)	Leg jerking /lateral body torsion	7 (28.0%)
Screaming 2 (8.0%)	Epigastric Pain	1 (4.0%)
Seizure Frequency (n)		
Less than monthly	21 (84.0%)	
Monthly	3 (12.0%)	
Weekly	1 (4.0%)	
Comorbid diagnosis (n)[c]		
None	19 (76.0%)	
Present	6 (24.0%)	
Standardised Measures	Mean (SD)	Mean (SD)
Reading Comprehension (YARC)	100.84 (12.79)	108.23 (11.12)
Significant impairment [d]	3 (12.0%)	0
Reading Accuracy (YARC)	98.12 (12.84)	103.62 (10.80)
Significant impairment	4 (16.0%)	0
Reading Rate (YARC)[e]	95.52 (15.25)	105. 03 (15.01)
Significant impairment	5 (20.0%)	4 (10.26%)
Sight Word Reading (TOWRE)	94.32 (18.94)	103.64 (14.07)
Significant impairment	7 (28.0%)	3 (7.69%)
Phoneme Decoding (TOWRE)	95.44 (18.57)	101.69 (11.78)

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Significant impairment	7 (28.0%)	5 (19.23%)
Listening Comprehension (CELF)[f]	9.16 (3.16)	10.62 (2.71)
Nonverbal IQ[f]	7.60 (2.90%)	10.08 (2.77)
Articulation (Goldman Fristoe)	103.16 (4.02)	104.36 (2.78)
Significant impairment	0	0

Note. For all tests M = 100 and SD = 15 with the exception of the Listening Comprehension (CELF) and non-verbal IQ (WISC) where M = 10 and SD =3. [a] One child with RE and nine control children had non-returned language questionnaires. One child with RE and eight control children had non-returned DCD questionnaires. [b] In 80% of cases laterality was fully confirmed by EEG. [c] Comorbid diagnoses: 1 Anxiety, 1 ADHD and resolved hearing difficulty, 2 ASD and 2 movement disorder. [d] Number of children one standard deviation or below the standard score mean. [e] Four children had missing data for reading rate. For these children the word reading composite was calculated using the three available scores. [f] Only one subtest used therefore percentage with significant impairment not reported.

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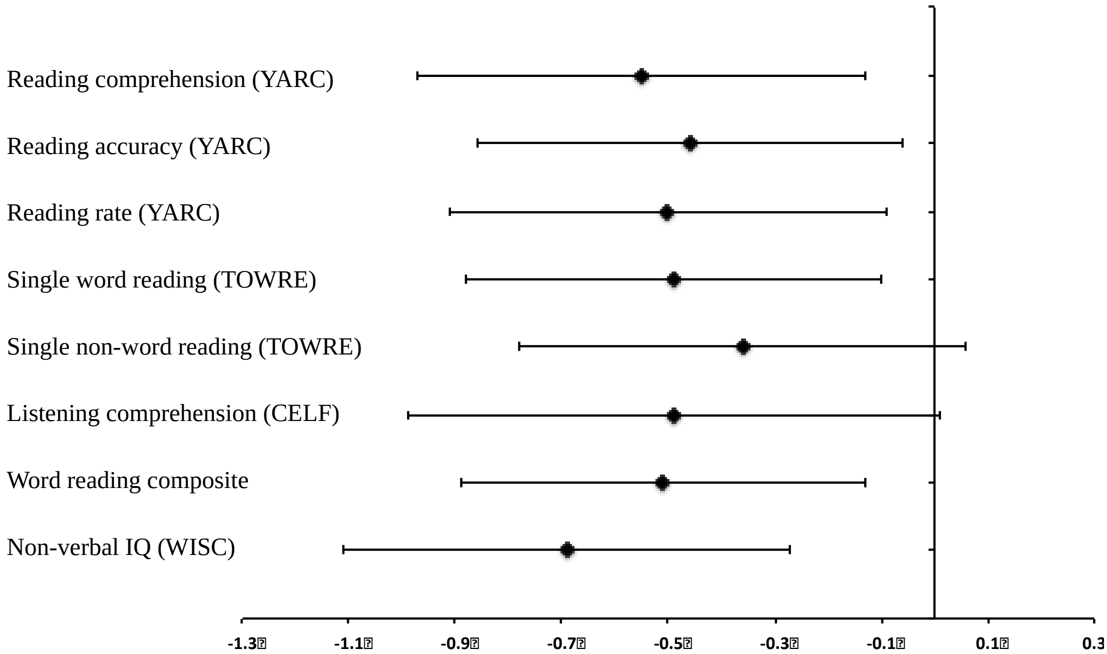

Figure 2.
Mean difference between RE and control group (95% CIs) for raw scores (expressed as z-scores) on the study measures.

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Table II. *Parameter estimates for best-fit general linear model of reading comprehension, with word reading and listening comprehension as predictors.*

Parameter (z-transformed)	Estimate (β)	SE	95% CI	*p*	R^2
Word-reading	.65	.07	.51-.79	<.0001	.70
Listening Comprehension	.39	.07	.25-.53	<.0001	

Note. The model including NVIQ was a similar fit (BIC (word reading, listening comprehension) = 120.02, BIC (word reading, listening comprehension, NVIQ) = 120.41) and accounted for a similar proportion of variance in reading comprehension (.72), thus the more parsimonious model is preferred. The residuals did not depart from normality, there was no difference between groups in residual variance and the residuals were uncorrelated, Durbin-Watson = 2.29.

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