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Children born before 32 weeks of gestation displayed impaired reading fluency, comprehension, and spelling skills at nine years of age

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Short Title: Literacy deficits and very low gestational age

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Abstract

Aim: Our aim was to study whether prematurity, associated with prenatal and neonatal risk factors, affects specific literacy skills among schoolchildren born at a very low gestational age (VLGA) of <32 weeks.

Methods: The study group comprised 76 prospectively followed VLGA children born between November 1998 and November 2002 at Oulu University Hospital, Finland, and 51 term controls. The median gestational age of the VLGA children was 29.0 (24.1 – 31.9) weeks. All children were examined at a median age of 8.9 (8.0 – 9.9) years in Oulu between November 2007 and November 2011. Reading fluency, comprehension and spelling skills were evaluated using standardised tests for Finnish-speaking children.

Results: VLGA children had significantly poorer test results in reading comprehension (median 6.9 versus 8.3, *p=0.014*) and spelling (median 35.7 versus 38.0, *p=0.013*) than term children. Furthermore, VLGA children more often performed below the 10th percentile normal values in spelling (*p=0.012*) compared to term controls. Foetal growth restriction was associated with lower scoring in reading fluency *(p= 0.023*) and spelling (*p= 0.004*) among VLGA children.

Conclusion: VLGA schoolchildren performed poorer in reading comprehension and spelling than term children. In addition, poor foetal growth in VLGA children was associated with literacy problems.

Keywords: foetal growth restriction, literacy skills, spelling, very low gestational age

Key notes

- Children born at a very low gestational age showed significantly poorer performance in reading comprehension and spelling compared to term children at nine years of age.
- Very low gestational age children scored below the 10th percentile in spelling more often than term controls.
- Poor foetal growth was an independent risk factor for literacy problems in children born at a very low gestational age.

1. INTRODUCTION

It has been demonstrated that preterm children need more educational support compared to term children, even before starting school. [1,2] Difficulties among school-aged children have been reported, e.g. in naming skills [3,4] as well as in grammar, vocabulary and speech production.[5] Problems may also exist in literacy skills such as reading fluency and spelling, [6] as well as in learning mathematics.[7-9] In addition, children born at a very low gestational age (VLGA) have been found to have difficulties in identifying phonemes (phonological awareness) at 8 years of age.[6] On the other hand, some studies have found no differences between VLGA and term children in language and reading tasks.[10]

Both antenatal and neonatal factors may affect delicate processes of brain development among this high-risk population, thus contributing to deficits in language, memory and learning abilities.[11-13] White matter damage may disrupt myelination and axonal integrity in developing brain, which further may constitute a basis for problems in language development among children born preterm.[14] Preterm children also maintain atypical bilateral language organisation longer than term born controls, which might reflect a delay of neural language organisation due to very premature birth.[15] Foetal growth restriction (FGR) has been associated with differential effects on brain development [16] as well as poor language functions in preterm children.[17] FGR is caused by impaired transplacental nutrition and oxygen supply due to placental insufficiency. Hypoxia induces redistribution of the blood flow in favour of vital foetal organs, particularly the heart and brain.[18]

In this study, we hypothesised that preterm birth and poor intrauterine growth would be related to literacy problems during children's early school years. Our specific aims were to study whether reading fluency, comprehension and spelling differ between Finnish-speaking VLGA and term children at nine years of age. We especially wanted to investigate whether different dimensions of reading, i.e. fluency, accuracy (including number of letter substitutions and omissions), comprehension and correct spelling would differ between VLGA and term children in Finnish literacy tests. Finnish language is an example of transparent orthography in which reading fluency is an important indicator of potential reading difficulty or even dyslexia. Furthermore, Finnish VLGA children might differ from their preterm peers who, when studying literacy skills, are exposed to opaque orthographies in languages such as English. A further aim was to evaluate whether FGR and other ante- and neonatal factors were associated with reading and spelling skills among VLGA children.

2. METHOD

2.1. Participants

This study was part of a longitudinal study of 163 VLGA children (gestational age <32 weeks at birth) born at Oulu University Hospital between November 1998 and November 2002.[11] The follow-up cohort was recruited during a four-year period between November 2007 and November 2011. The recruitment of the VLGA population is presented in Figure 1. There were no differences in basal prenatal or neonatal data between VLGA children who participated and those who did not.[11] The exclusion criteria for the current study were severe neurosensory disabilities defined as cerebral palsy (CP), blindness, a need for a hearing aid, and intellectual disability. In addition, all children were supposed to be Finnish-speaking (Figure 1). Altogether, 76 VLGA children underwent the assessments of language and literacy skills at a median age of 8.9 years (range 8.6 – 9.5 years).

The control group consisted of 51 term children, (born ≥ 37+0 gestational weeks and matched for calendar age and gender) who were recruited both from the Oulu University Hospital birth register (n=27) and the elementary schools located in Oulu (n=24). Term children underwent the same assessments as VLGA children at a median age of 8.9 years (range 8.0 – 9.9 years).

In total, 50 VLGA children and 26 term children were second graders. The corresponding numbers of third graders were 26 and 25. All children attended main-stream school, although 3 of 76 (4%) VLGA children needed a special education class. This study received approval from the ethical committee of Oulu University Hospital. Written informed consent was required prior to study entry from children and parents.

2.2. Clinical characteristics

The definition of FGR in the present study was based on birth weight below two standard deviations from the mean of gestation-adjusted birth weight,[19] documented evidence of growth restriction due to placental insufficiency defined by Doppler ultrasound and histological placental perfusion defect. [11, 18] Intraventricular haemorrhage after birth was defined according to Papile.[11] CP was confirmed at the age of 5 years by a child neurologist. The definition provided by the Surveillance of Cerebral Palsy in Europe network was used: CP is a permanent but not unchanging disorder of movement and/or posture; and of motor function; causing activity limitation that is attributed to a non-progressive abnormality of the developing brain. All participants of the current study also underwent a structured neurological assessment at nine years of age. None of the term children were diagnosed as having CP. Intellectual disability was defined by an intelligence quotient under 70, based on previous psychological evaluations and

the diagnosis was set on clinical bases by a child neurologist. Hearing impairment was verified as a need for a hearing aid and the diagnosis was set on clinical bases by an audiologist. None of the prospective cohort participants were blind nor did they have congenital infections or malformations.[11]

The mothers` educational level was classified into two categories: low level referred to a nine-year compulsory comprehensive school with or without vocational education; high level referred to a 12-year education of comprehensive and upper secondary school with vocational education, polytechnic or university degree.

2.3. Procedure

The language and literacy tests were performed at Oulu University Hospital by two speech therapists (M.H., L.P.), each test session lasting approximately 2½ hours. All assessments were video-recorded for subsequent analyses. In the present study, two standardised tests for Finnish-speaking primary school-aged children were used (Table 2). The YTTE-test [20] was used to measure reading skills such as fluency, accuracy, and comprehension and Lukilasse[21] to check spelling.

In the YTTE-test children are asked to read a text about a cat. Second graders read about a cat called Moris who wants to awaken its family and third graders read about a cat called Vilpe who climbs down ladders after trying to catch a bird. After reading the text, certain questions were asked to find out if the child understood and remembered the main points of the content. The participants were informed that they would be asked about the content after reading the text. Reading fluency was estimated by dividing the time spent on reading by the numbers of words. In this test, appropriate reading fluency time was less than 1.79 seconds/word for second graders (range 1.3–1.78 seconds/words for average performance).10 th percentile cut-off score was >2.6 seconds/word. Appropriate reading fluency time was less than 1.20 seconds/word (range 1.0–1.19 seconds/word) for third graders, >1.6 seconds/word being a cut-off score. In addition, the number of reading errors was calculated. No more than six errors were accepted as a normal performance among second graders (range 4–6 errors) and four (range 2–4 errors) among third graders. After reading, children were asked five questions about the key issues in the text. The answers are scored from zero (no, wrong or inappropriate) to three (precise, adequate answer). The scores are calculated and divided by maximum scores and multiplied by 100. For second graders, the scores between 35–59 were considered as average and the scores between 60–100 as a good performance. The corresponding scores for third graders were 43–67 (average) and

68–100 (good performance). The 10th percentile cut-off score was 34 for second graders and 42 for third graders. Performance below these cut-off points was considered weak. The test also included six true or false items related to text content to measure recognition; scores between 4 to 5 were considered normal (/average) for children in both grades.

The Lukilasse test is a standardised spelling test, where a child writes dictated words. For each correctly written word a child gets two points. The maximum score was 40 for the second graders and 42 for the third graders, while average score was 37 for children in both grades. The 10th percentile cut-off scores were 28 and 29, respectively. Scores below these levels were considered as weak performance.

2.4. Statistical analysis

The statistical analyses were performed using the Statistical Package for Social Science (SPSS), version 25.0 (IBM Corp). Categorical data between VLGA and term groups were analysed using cross tabulation with Fisher`s exact test and Pearson's chi square test. The distributions of the raw test scores were skewed, and the variances of the VLGA and term study groups were unequal, so the Mann-Whitney *U* test was employed to analyse differences between the two groups (Table 2). The Kruskal-Wallis test was used to compare three groups (Table 3). A factorial ANOVA was used to study the associations between FGR and each dependent variable adjusting for the following potential confounding variables: gender, gestational age and mother's education. All tests were two-tailed. The level of significance was set at $p < 0.05$.

3. RESULTS

The clinical and background factors of VLGA and term children are shown in Table 1. Term children did not differ significantly from VLGA children in terms of gender distribution or maternal education (data not shown). In the present study, VLGA children performed significantly poorer than term children in reading comprehension and spelling (Table 2).VLGA children scored below the 10th percentile significantly more often in spelling compared to term children (30.3% versus 11.8%; *p=0.012*). Otherwise, there were no statistical differences in reading fluency (33.8% versus 22.4%; *p=0.125*), reading accuracy (23% versus 18.4%; *p=0.353*), recognition (1.4% versus 2%; *p=0.640*) or reading comprehension (28.4% versus 16.7%; *p=0.101*) between the two groups. We found no association between gender and literacy tests among VLGA children (data not shown). However, term-born girls performed significantly better than term-born boys in YTTE fluency (median 0.82 seconds/word versus 1.19 seconds/word*, p=0.02)* and spelling (median

38.6 points versus 36.8 points, *p=0.02*). VLGA children had received speech therapy (p<0.001), physiotherapy and occupational therapy (p<0.001) more frequently compared to term children.

VLGA children with FGR, VLGA children without FGR and term children differed significantly from each other in reading fluency (*p=0.012*), comprehension (*p=0.042*) and spelling skills (*p=0.001*) (Table 3). The pairwise comparisons were further performed. In the YTTE reading fluency task, VLGA children with FGR scored significantly poorer compared to VLGA children without FGR (*p=0.023*) and compared to the term group (*p=0.003*). In the YTTE reading comprehension task, no significant differences were found when VLGA children with FGR were compared to VLGA children without FGR (p=0.717) or to term group (*p=0.050*) . In the Lukilasse test VLGA children with FGR showed significantly poorer spelling skills compared to VLGA children without FGR (p=0.004) and compared to term children (*p<0.001*). Gender, mother's educational level or gestational age did not associate with literacy skills among VLGA children evaluated using the factorial ANOVA (data not shown).

4. DISCUSSION

In the present study, VLGA children without severe neurosensory disabilities had significantly more difficulties in reading comprehension and spelling skills than term children at nine years of age. Also, VLGA children more often scored below the 10th percentile in spelling tasks compared to controls. In addition, FGR independently predicted impaired literacy outcome in VLGA children.

The current study demonstrated that VLGA children had poorer literacy abilities compared to the term-born controls. These results were consistent with those of earlier studies.[22] Reading and writing deficits are often consequences of earlier language impairment and have been shown to be more common among children born preterm.[22] Preterm children have been found to have poorer vocabulary (also receptive vocabulary), grammar and phonological awareness as well as poorer skills of rapid naming compared to term children even without observable brain damage.[23] Upon entering school, letter knowledge seems to be one of the best predictors of reading and spelling accuracy in Finnish language as well as phoneme identification and pseudo word repetition.[24]

Interestingly, Finnish-speaking VLGA children did not differ significantly from term controls in reading fluency in the present study. In transparent orthographies, such as Finnish, the process of decoding accurately is rather fast, which might make the early identification of children at risk for reading difficulty quite challenging, even for professionals.[24] Approximately 30% of Finnish

children are able to decode before entering the first grade and even non-readers at school entry reach the level of early readers in reading accuracy during the first school year. Those who struggle with reading after that seem to experience persistent problems specifically in reading fluency.[24] Some studies have also shown that language problems may appear only later during follow-up or worsen with age in VLGA children.[23]

According to different studies, a range of 25 to 50% of VLGA children have various degrees of neurosensory deficiencies in cognitive as well as attentional and executive skills that may affect their abilities to read and write at school age.[8, 9,25-26] It is of note that even though we excluded all VLGA children with neurological and sensory disabilities from the current study, the remaining VLGA children still performed poorer in literacy tests compared to term children. The findings of the current study were in line with a current meta-analysis suggesting that VLGA children without neurologic or developmental sequela may still be at risk for difficulties in their future academic performance.[26] Our previous study of this cohort showed that VLGA children had compromised school achievement compared to term children. However, VLGA children did not differ significantly from the term children in reading and writing skills as graded by a teacher.[11] Therefore, we would like to emphasise the importance of systematic follow-up of VLGA children.[26,27] Additionally, the flow of communication between school and home should be fluent.

Preterm birth occurs at a time of rapid growth and development of the brain when several vulnerable processes take place, including neurogenesis, synaptogenesis, brain folding, and myelination. Advanced neuroimaging studies have brought to light the structural and functional changes in brain microstructure and connectivity in VLGA individuals.[14,28] Altered brain maturation may also lie behind aberrant functional organisation of language pathways that may further lead to atypical lingual development in VLGA children.[14]

In the current study, VLGA children with FGR scored worse in reading fluency and spelling compared to VLGA children who had a birth weight appropriate for their gestation. This result was further confirmed using factorial ANOVA, suggesting FGR as an independent risk factor for impaired literacy outcomes. The poor performance in reading fluency during early school years might indicate more permanent reading difficulties or even dyslexia in VLGA children with FGR. The association between FGR and language difficulties is supported by previous studies[17,18] and underlines this subgroup of preterm children as a candidate population for needing special literacy skills guidance during school years. The principal cause of FGR is placental insufficiency.[18] It results in chronic foetal hypoxia accompanied by deleterious haemodynamic changes, loss of nutrients, and changes in placental and foetal endocrine function, all of which

can impair foetal brain development.[18] Previously, we have shown that white matter maturation was different among VLGA children with and without FGR when compared to term controls.[28] Considering the outcomes of the current and former studies[16,17], it is possible that prematurity and FGR cause differential disruptions in brain development.[18]

In our study, gestational age had no significant effect on literacy outcome in VLGA children. However, only a few VLGA children were born before 26 weeks of gestation. This might have affected the results and might explain why the degree of prematurity failed to correlate with the literacy outcome. Environmental factors during the first hospitalisation, including abnormal auditory experience and inadequate parent–infant interaction, may also have an effect on language abilities among VLGA children.[14] Early intervention programs targeting these problems may support cognitive and language development of children born at VLGA [14] Mothers' low educational level is a known risk factor for a child's lower academic achievement.[29] However, we found no association between mothers' educational level and literacy performance among VLGA children. It is worth mentioning that, in Finland early childhood education is provided to all children equally and that might even out socioeconomical differences, also in the subgroup of VLGA children. Also, the level of education is known to be high in Finland.

Among the strengths of the current study, we consider that our VLGA population belonged to a well-defined prospective cohort.[11] Also, the study included a comparison group of term children. The inclusive reading and spelling evaluations were performed using well standardised tests for Finnish-speaking children by qualified speech therapists (M.H and L.P) and enabled us to investigate a wider range of literacy skills in VLGA children. Potential limitations of this study were the average population size and the rather high refusal rate. However, we have shown previously that the VLGA children lost due to discontinued participation before the follow-up did not differ from the VLGA participants regarding gestational age or other relevant clinical factors.[11]

5. CONCLUSIONS

Our cohort study demonstrated that VLGA children without severe neurosensory disabilities performed poorer in reading comprehension and spelling compared to term-born controls at nine years of age. Poor foetal growth in the preterm children was a risk factor for literacy problems. This study points out the importance of speech and language evaluation in VLGA children, especially for those with FGR. Parents and teachers in early education and primary schools should be offered information about potential risks for early linguistic as well as later reading and writing difficulties in these children.

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Conflict of interest

None.

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TABLE 1 Characteristics of the very low gestational age (VLGA) and term-born children

	VLGA (n=76)	Term (n=51)
Child		
GA at delivery in weeks, Md, (range)	29^{+0} (24^{+1}-31^{+6})	
GA≥28 weeks, n (%)	55 (72)	
GA<28 weeks, n (%)	21 (28)	
GA<26 weeks, n (%)	3 (4)	
Birthweight in grams, Md (range)	1173 (370-2295)	
Female, n (%)	36 (51)	26 (52)
Foetal growth restriction, n (%)	18 (24)	
Respiratory distress syndrome, n (%)	48 (63)	
Bronchopulmonary dysplasia, n (%)	17 (22)	
Intraventricular haemorrhage grade 2-3, n (%) *	4 (5)	
Therapies prior study entry		
Speech therapy, n (%)	32 (42)	4 (8)
Physio/occupational therapy, n (%)	38 (50)	3 (6)
Mother		
Age at delivery in years, Md (range)	30 (18-44)	
Low education, n (%)	31 (41)	18 (35)
High education, n (%)	44 (59)	33 (65)

GA= Gestational age, VLGA = Very low gestational age

*None of the VLGA children had intraventricular haemorrhage grade 4.

The values given are as medians (Md), range, number of cases and percentages (%).

TABLE 2 Reading fluency, accuracy, comprehension, recognition (YTTE test) and spelling skills (Lukilasse) among 9-year-old VLGA and term children.

	VLGA			Term			
	Median	Range	SD	Median	Range	SD	p-value*
YTTE test	n=74			n=49			
Reading fluency, seconds[a]	1.2	0.5–4.2	0.8	1.0	0.5–4.8	0.7	*0.057*
Reading accuracy, n/errors[b]	5.1	0–32	6.8	5.3	0–29	6.0	*0.730*
Comprehension (max 12 points)[c]	6.9	0–12	2.9	8.3	0–12	2.8	*0.014*
Recognition (max 6 points)[d]	4.9	3–6	0.9	5.0	2–6	0.9	*0.535*
Lukilasse	n=76			n=51			
Spelling[e]	35.7	8–41	8.6	38.0	16–40	5.6	*0.013*

VLGA = Very low gestational age.

*Mann-Whitney U test.

The results are given as median, range (maximum and minimum scores) and standard deviation (SD).

[a]Reading fluency = Reading time spent on one word (seconds) in text, [b]Reading accuracy = Number of errors per the whole text (e.g. letter omissions or substitutions, morphological endings, etc.), [c]Comprehension = Cued questions on content of the text→the higher the scores, the better the performance, [d]Recognition = True and false propositions concerning the text, the higher the scores, the better the performance, [e]Spelling = Dictation spelling task/ a list of 42 dictated words to write.

TABLE 3 Reading fluency, accuracy, comprehension, recognition (YTTE test) and spelling skills (Lukilasse) among 9-year-old VLGA children without and with FGR and term children.

	VLGA children with FGR			VLGA children without FGR			Term children			
	Median	Range	SD	Median	Range	SD	Median	Range	SD	p-value*
YTTE test	n=17			n=57			n=49			
Reading fluency, seconds[a]	1.8	0.7–4.2	0.9	1.1	0.5–4.2	0.7	1.0	0.5–4.8	0.7	*0.012*
Reading accuracy, n/errors[b]	7.0	0–22	6.4	4.1	0–32	6.9	5.3	0–29	6.0	*0.209*
Comprehension (max 12 points)[c]	6.8	1–10	2.5	7.0	0–12	3.1	8.3	0–12	2.8	*0.042*
Recognition (max 6 points)[d]	5.1	4–6	0.7	4.9	3–6	0.9	5.0	2–6	0.9	*0.680*
Lukilasse	n=17			n=59			n=51			
Spelling skills[e]	32.5	10–38	9.0	37.2	8–41	8.3	38.0	16–40	5.6	*0.001*

FGR= Foetal growth restriction, VLGA= Very low gestational age

*Kruskal-Wallis test was used to compare the three groups. The results are given as median, range (maximum and minimum scores) and standard deviation (SD). [a]Reading fluency = Reading time spent on one word (seconds) in text, [b]Reading accuracy = Number of errors per the whole text (e.g. letter omissions or substitutions, morphological endings, etc.), [c]Comprehension = Cued questions on content of the text→the higher the scores, the better the performance, [d]Recognition=True and false propositions concerning the text, the higher the scores, the better the performance, [e]Spelling = Dictation spelling task/ a list of 42 dictated words to write.

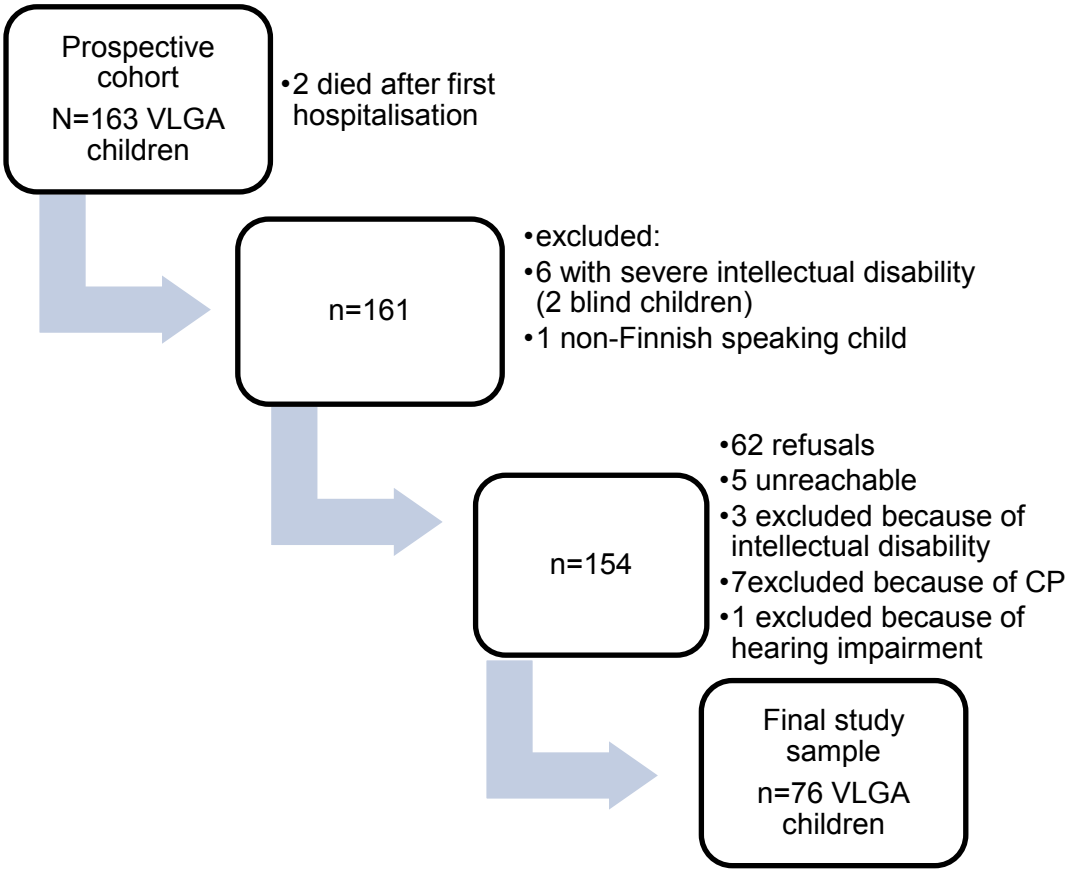

Figure 1 Flow chart of the study group of VLGA children.