Fetal growth restriction has negative influence on narrative skills in 8–10-year-old children

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Running head: Fetal growth restriction and narrative skills

KEYWORDS

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Fetal growth restriction

Narrative skills

Placental insufficiency

Premature birth

ABSTRACT

Aim: The risk for neurocognitive difficulties is increased in children born with fetal growth restriction (FGR), but no data exists yet on their narrative skills. The narrative skills of 8–10-year-old children born with FGR between 24–40 weeks were compared with those of children born with appropriate growth for gestational age (AGA).

Methods: A prospectively collected cohort of 36 children with FGR were recruited prenatally at a Finnish tertiary hospital from 1998–2001, and 31 children with AGA served as controls. Narrative skills were assessed using a standardised test, and correlations between narrative, communication, reading and spelling skills were studied.

Results: Children born with FGR produced significantly less information and shorter utterances in their narratives than the AGA group. Children born preterm with FGR performed significantly more poorly in their narratives than the preterm AGA group. Poor narrative skills correlated with poor communication, reading and spelling skills.

Conclusion: Children born with FGR had poorer narrative skills compared to their AGA peers at the age of 8–10 years, and narrative skills were linked to other language-based skills, which underlines the importance of early detection and preventive measures to optimise the educational outcome of children born with FGR.

KEY NOTES

- There are no studies on narrative skills of children born with fetal growth restriction.

- Children born with fetal growth restriction, especially those born preterm, have poorer narrative skills than their peers born at appropriate growth for gestational age.

- Close follow-up of narrative skills at early age with timely interventions may help to support children born with fetal growth restriction to optimise their educational outcome.

ABBREVIATIONS*:*

AGA, Appropriate growth for gestational age;

ERRNI, The Expression, Reception and Recall of Narrative Instrument test;

FGR, Fetal growth restriction;

MLUw, mean length utterance in words;

SD, standard deviation.

INTRODUCTION

Fetal growth restriction (FGR) with placental insufficiency, indicated by birth weight < 10th percentile and/or umbilical artery pulsatility index > 2 SD in Doppler ultrasound (1,2), has been associated with multiple neurodevelopmental risks. Impaired transplacental oxygen and nutritional supply may cause redistribution of the blood flow in favour of vital fetal organs, especially the heart and the brain (3). In most studies, the combination of prematurity and FGR has been reported to increase the risk of cognitive problems (4,5), including language and academic and communication skills in 5–10-year-old children. In a study by Kallankari et al (6), FGR predicted independently poor language, memory and learning skills. Low academic achievement (4,7), poor language skills (8), deficiency in verbal short-term memory (9), lower executive cognitive functions (5,7,10) and poor performance in communication (11), reading and spelling skills (12) have also been reported in children born with FGR. A narrative skill is defined as a written or spoken, fictive or real life–based description of causally and temporally related events, and it focuses on a certain theme to form a complete story (13). It requires the integration of cognitive, linguistic and social abilities (14). Narratives compose approximately 80% of our everyday language, and narrative skills develop along with other language skills (15) already present in early childhood (14,16). Children's ability to tell stories and describe everyday events develops from first words to more syntactically and episodically complex language (14). Fluent narrative demands good working memory (17), a sense of metalinguistics (18) and pragmatic skills (15). Narrative skills are connected with the development of communication skills (16), which together form the basis for academic abilities (14,16). FGR is known to increase the risk of poor communication (11), reading and spelling skills (12). However, narrative skills of children born with FGR have not yet been studied.

In this study, we hypothesised that 8–10-year-old children born with FGR, especially those born preterm, have poorer narrative skills than children with appropriate fetal growth for gestational age (AGA). Our secondary hypothesis was that the influence of FGR on language-based skills becomes more evident with advancing age (11,12). Specifically, we evaluated oral storytelling using a wordless, illustrated story book to assess how the children perceived coherence in its story, how well they retold the story

after a short break and how they related the purposes, intentions and emotions of the characters. In addition, we compared the children's narrative skills to our previous findings on their communication skills, as described by their parents, as well as their reading and spelling skills.

METHODS

Participants

This study is part of a prospective cohort study of 73 prenatally diagnosed children born with FGR at 24–40 gestational weeks from 1998–2001 at the Oulu University Hospital, Oulu, Finland. FGR was defined as a birth weight below the 10th percentile for estimated gestational age and/or an abnormal umbilical artery pulsatility index > 2 SD (2). In all cases, gestational age was confirmed by ultrasound prior to 20 gestational weeks. Fetuses with major structural and chromosomal anomalies were excluded (8). The control group, which comprised 31 children with AGA matched with their gestational age and delivery within two weeks to the index neonate with FGR, were selected from the delivery room records. Recruitment procedure for postnatal examinations of these native Finnish-speaking children has been described in detail in our previous studies (11,12). After an 8–10-year follow-up period, the participants were approached by email and phone, and informed, written parental consent to a follow-up visit was obtained. The study groups are presented in Figure 1. At the time of the assessment, 21 children with FGR and 17 children with AGA were eight years old, 13 children with FGR and 14 children with AGA were nine years old, and three children with FGR had just turned 10 years old. Of the children with FGR, 58% were born preterm (< 37 gestational weeks) and 42% full term. In the AGA group, 61% of children were born preterm and 39% full term. Information on socioeconomic factors, speech and language therapy received prior to or at preschool age, and special education given at school were collected using a parental questionnaire.

Procedures

The Expression, Reception and Recall of Narrative Instrument (ERRNI) test (19) was used to measure children's expressive language and story comprehension. The test

measures children's ability to narrate, retell and understand the idea of an illustrated story (19). Ideas in the children's initial storytelling, and their retelling of the story without looking at the illustrations, reflects their ability to interpret the illustrated story and describe it verbally. The forgetting index, which may expose deficiencies in verbal long-term memory, was calculated by subtracting the scores of their retelling of the story from the scores of their initial storytelling. Comprehension questions about the story's essential episodes were asked after their retelling of the story to evaluate if children had understood the main ideas. Their narrative productivity was measured by mean length of utterances in words (MLUw), which was calculated by dividing the total number of words by the total number of utterances in both storytelling tasks (19). The ERRNI test material and procedure are presented in Appendix 1.

Analyses of the children's stories included their ideas in the initial storytelling, their ideas in its retelling, forgetting index, comprehension and MLUw. The researcher (LP) who performed the test was blinded on FGR or AGA data. The original norms of the ERRNI test were used as there are no standardized norms for Finnish-speaking children.

Statistical analyses

SPSS version 21.0 for Windows (IBM Corporation, Armonk, New York, USA) was used to analyse the data. The medians, quartiles, and minimum and maximum values of ERRNI standard scores were calculated. For average performance, ERRNI standard scores are expected to be close to 100, with a standard deviation (SD) of 15 (19). In this study, 1 SD (standard scores ≤ 85) was considered to indicate a possible language problem, and 2 SD (standard scores ≤ 70) was considered to indicate a significant impairment. As ERRNI test results are standardized according to age, two age groups were formed of children less than nine years and children nine years and older (19). Due to a skewed score distribution, the differences between FGR and AGA groups in both age groups were analysed using a nonparametric Mann-Whitney U test. In addition, as prematurity is recognized as an independent risk factor for impaired cognitive development, narrative performances were evaluated in children with FGR and AGA born either at term (≥ 37 gestational weeks) or preterm (< 37 gestational weeks). Characteristics of the FGR and the AGA groups were tested using Fisher's exact test and Mann-Whitney U test.

Correlations between narrative, communication, and reading and spelling skills were analysed using the Spearman rank-order correlation coefficient with a two-tailed signification. Detailed information on the measurements of communication, reading and spelling skills is available in our earlier publications (11,12).

RESULTS

The characteristics of the FGR and AGA groups (n = 67) are presented in Table 1. In the FGR group, more than half of the children were first-borns, while in the AGA group, around one third were first-borns. The children born with FGR had received significantly more special assistance at school compared to the children with AGA. They were also more frequently referred to speech and language therapy at the age of five to six years than the children with AGA. Indications for speech and language therapy were difficulties with language skills (9/20 children with FGR and 2/10 children with AGA) and articulation disorders (11/20 children with FGR and 8/10 children with AGA). Of the children who received speech and language therapy, approximately 65% in both groups received therapy at the age of five to six years once a week or once every two weeks, and 55% of the children received it for 6–12 months.

Figure 2 shows that children with FGR produced significantly less relevant story content both in initial storytelling (p = 0.001) and in retelling the story (p = 0.01) when compared to the AGA group. In initial storytelling, 44% of the children born with FGR performed -1 SD, and in retelling 47% performed -1 SD, compared to 25% and 29% respectively in the AGA group. Furthermore, 28% of the children with FGR performed -2 SD in initial storytelling and 25% in retelling, while the corresponding percentages were 13% and 16% in the AGA group. The children born with FGR had significantly shorter MLUw (p = 0.001) in their narratives compared to the children with AGA, and 44% of the children born with FGR scored ≤2 SD in MLUw compared to 10% in the AGA group. There were no significant difference between the study groups in comprehension tasks or forgetting index.

Table 2 presents the subscale scores in the ERRNI test. Children less than nine years old born with FGR performed significantly more poorly in initial storytelling, retelling,

comprehension and MLUw than the children with AGA. At the age of nine years and older, children born with FGR had poorer MLUw compared to the AGA group.

Children with FGR born preterm (n = 21) performed significantly more poorly in initial storytelling (U = 314.5, p = 0.001), retelling (U = 285.0, p = 0.020), story comprehension (U = 202.5, p = 0.015) and MLUw (U = 280.5, p = 0.027) compared to 19 preterm children with AGA. In the full-term group, MLUw was poorer in 15 children with FGR than in 12 children with AGA (U = 135.0, p = 0.028). In the FGR group, children born preterm performed significantly more poorly in story comprehension (U = 158.5, p = 0.043) than the full-term children. The performances between children born preterm and full term in the AGA group did not differ significantly.

Correlations between narrative, reading, spelling, and communication skills are presented in Table 3. In the FGR group, story content in initial storytelling and retelling, as well as story comprehension, correlated positively with General Communication Composite scores, reading comprehension and word reading skills, as well as spelling skills (coefficients between 0.360 and .0558, p values < 0.05 for all correlations). In the AGA group, ideas in initial storytelling and retelling correlated with reading comprehension (coefficients between 0.464 and .0557, p values < 0.05 for all correlations).

DISCUSSION

The purpose of this study was to evaluate the narrative skills of 8–10-year-old children born with FGR and to explore possible links between narrative abilities and communication, reading and spelling skills. In line with our study hypothesis and previous research suggesting difficulties in language-related cognitive skills, children born with FGR produced less informative and shorter narratives with simpler utterances than children with AGA. Short sentences and the lack of declarative subordinate clauses negatively influenced their ability to produce a cohesive narrative (18,20). The measures of ideas in initial storytelling and in retelling indicated that poorly performing children born with FGR had not understood the events of the illustrated story and that they had failed to describe the causal relationships between episodes.

Adequate narrative skills are important for academic performance, not only as a predictor of reading and reading comprehension skills but also because school instructions and written materials are given verbally, which requires the ability to understand the meanings of words and sentences (21). In our study, poor storytelling and story comprehension in the children with FGR was associated with problems in reading comprehension, word reading and spelling skills. Our results are in line with a recent study (22) by Mäkinen et al, in which story-generation tasks of typically developing Finnish-speaking eight-year-old children were associated with reading comprehension and orthographic skills. In addition, correlations between oral storytelling, word reading skills and reading comprehension have been reported (18,23). It seems that narrative, reading, spelling and communication skills are linked and indicate potential language difficulties and later academic problems in children born with FGR (24). These difficulties may have long-lasting effects on a child's life.

Our study found that narrative skills were poorest in children born preterm with FGR. Earlier, Crosbie et al (25) had reported that children born preterm demonstrate difficulties formulating a narrative compared to their peers born full term, but they did not assess FGR. Our literature search did not reveal any studies examining both FGR and prematurity with regard to narrative skills. In our study, children born preterm with FGR had difficulties in all ERRNI subtests except in forgetting index, in which their performance did not differ from other groups. This might be due to the validation method of this subtest, as Bishop (19) mentions in the ERRNI manual. For example Dodwell and Bavin (17) suggest that recalling the story without seeing pictures is easier when a child is retelling his or her own story.

The children born with FGR who were less than nine years old had significantly poorer narrative skills than their AGA peers, while children born with FGR who were older than nine years performed more poorly only in MLUw compared to the respective AGA group. However, short MLUw may indicate grammatical limitations and problems in processing long and complicated utterances (19).

As narrative skills indicate academic and communication abilities, it is important to follow up with at-risk children. Our study suggests that children born with FGR should be

included in this surveillance group. It is important that health care providers and educational teams be aware of the risks related to FGR for later development. It seems that current management protocols and cooperation between health care providers, schools and parents is insufficient and that there is a lack of knowledge about the role of FGR in the development of learning skills. Appropriate assessments and preventive interventions (26) are essential to guarantee optimal outcome, and careful attention should be paid to children born with FGR, especially children born preterm with FGR.

Small sample size and differences in supportive measures provided at the age of five to six years and at school between the groups are acknowledged as limitations in our study. However, despite the speech and language therapy provided at the age of five to six years and the support provided at school, as well as the smaller number of siblings in certain families, children born with FGR had more problems in their narrative skills. According to previous studies, first- and second-born children receive more parental attention, leading to better vocabulary (27), which is an important element in the development of narrative skills (25). In addition, our results need to be interpreted cautiously, keeping in mind the variation in children's ages. On the other hand, carefully defined FGR phenotype is a significant strength of our study. Regarding assessment, the ERRNI is a structured method, tested and standardised scientifically in the United Kingdom (19). It gives quantitative information on narrative skills but an additional test that provides qualitative information about syntax, extent of the vocabulary and morphology of the story would be useful. Bishop (19) suggests that ERRNI is a good first-line test to be used prior to deeper linguistics analyses.

CONCLUSIONS

Children born with FGR, especially those born preterm, had poorer narrative skills than their respective AGA peers. The development of their narrative skills was linked to other areas of language and learning skills. This indicates that preventive measures should be targeted to children born with placental insufficiency to support their linguistic and communication capabilities in order to improve their later academic performance.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

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Appendix 1

Scoring and test procedure: The Expression, Reception and Recall of Narrative Instrument (ERRNI)

The Fish ERRNI book consists 15 wordless pictures depicting episodes as follows: A boy gets money from his mother to buy a fish from the pet shop. He goes to the shop and buys a fish, and on his way home he meets his two friends. One friend secretly swaps the fish for a doll, and the boy and the other friend go to buy ice-cream. After eating the ice-cream, the boy gets home and discovers the doll in his bag. His mother phones his friends, and they come over to switch the doll and the fish.

For data collection, each child first looked at the 15 pictures before telling the story. After a period of 10–30 minutes, each child was asked to recall the narrative with the instruction: ´You remember that story you told me about the boy and the fish? I want to see how much of it you can remember. Tell me the story again now, without looking at the pictures. Tell me as much as you can.'

Mean length utterance in words was calculated by dividing the total number of words by the total number of utterances used in both storytelling tasks, and forgetting index was calculated by subtracting the scores of ideas in retelling the story from the scores of ideas in initial storytelling. After children retold the story, they were asked a series of comprehension questions. The ERRNI test was recorded and transcribed. The number of words and sentences, as well as answers to the comprehension questions, were calculated manually using an excel calculator with formulas from the ERRNI manual.

Table 1. Characteristics of the studied groups with fetal growth restriction (FGR) and appropriate growth for gestational age (AGA). The values given are median(range), %(n)

	FGR (N=36)	AGA (N=31)	p value
Mother			
Age at delivery, years	30(18−41)	31(21−43)	0.8
Education[1],			0.5
University	34(12)	33(10)	
Vocational education	58(21)	67(21)	
Compulsory education	8(3)	0	
Child			
Female	55(18)	58(18)	0.8
Age at study entry, year:months	9:1(8:8−10:2)	9:1(8:10−9:10)	0.9
GA at delivery, weeks	35(24−40)	35(26−40)	0.9
Preterm (<37 weeks)	58(21)	61(19)	0.8
Birth weight, grams	1733(370−2940)	2700(815−4300)	0.002
Umbilical artery PI ≥2SD	69(25)	0	<0.001
Diagnosis/Therapies prior to study entry			
Disability/Handicap	9(3)	16(5)	0.5
Hearing impairment	6(2)	3(1)	0.9
Speech & language therapy	58(20)	32(10)	0.049
Special education[1]	25(9)	3(1)	0.016
Physio/occupational therapy	33(12)	32(10)	0.9
Siblings			0.003
First born child	58(21)	36(11)	
Second born child	31(11)	16(5)	
Third or later born child	11(4)	48(15)	
Speech/language/learning problems among siblings	23(8)	30(9)	0.48
Parental background			0.2
Two parents	89(30)	74(24)	
Single parent	8(3)	19(7)	
Missing value	3(1)	6(2)	

GA, gestational age; PI, pulsatility index, [1] Compulsory education refers to a compulsory nine-year primary and middle school programme for all Finnish children, and vocational education to a three-year programme, which provides practical occupational fields of study. Special education includes participation in mainstream education with special assistance or attendance in special class/school.

Table 2. Subscale standard scores of Expression, Reception and Recall of Narrative Instrument (ERRNI) **a)** in children <9 years of age with fetal growth restriction (FGR, n=11) and appropriate growth for gestational age (AGA, n=11) and **b)** in children ≥9 years of age (FRG, n=25 and AGA n=20). The results given are median (range) and lower-upper quartiles (Q_1/Q_3) and p values of Mann Whitney U test.

a)

Children <9 years of age, FGR (n=11) and AGA (n=11)

	Study group	Median (range)	Q_1/Q_3	p value
Ideas in initial storytelling	FGR	83(65−111)	75/103	0.011
	AGA	114(69−135)	103/119	
Ideas in recalling	FGR	94(65−108)	65/105	0.023
	AGA	113(65−123)	84/121	
Forgetting index	FGR	97(79−103)	92/102	0.478
	AGA	95(83−103)	86/100	
Comprehension	FGR	88(65−112)	67/98	0.025
	AGA	103(88−112)	98/103	
Mean length uttererance in words	FGR	74(65−93)	65/88	0.034
	AGA	90(65−123)	74/103	

Children ≥9 years of age, FGR (n=25) and AGA (n=20)

	Study group	Median (range)	Q_1/Q_3	p-value
Ideas in initial storytelling	FGR	92(65−122)	69/98	0.057
	AGA	101(65−128)	84/114	
Ideas in recalling	FGR	92(65−120)	73/108	0.084
	AGA	103(65−135)	83/118	
Forgetting index	FGR	98(65−110)	89/102	0.478
	AGA	100(78−106)	95/103	
Comprehension	FGR	96(65−113)	80/101	0.813
	AGA	96(65−117)	73/105	
Mean length utterance in words	FGR	74(65−104)	67/85	0.010
	AGA	86(65−135)	79/89	

Table 3. Summary of correlations between narrative skills (standard score), communication skills (standard score) and reading and spelling skills (percentile) in children born with fetal growth restriction (FGR, n=36) and with appropriate growth for gestational age (AGA, n=31). Spearman Rank Order Correlation Coefficient was used.

Measure	FGR (n=36)					AGA (n=31)				
	Ideas Initial	Ideas retell	forgetting index	compre hension	MLUw	Ideas Initial	Ideas retell	forgetting index	compre hension	MLUw
Communication skills:										
General communication composite	.558**	.518**	.035	.440*	.286	.045	.246	.263	-.017	.057
Reading comprehension, fluency and accuracy:										
Reading comprehension	.420*	.448*	.131	.460*	.255	.464**	.557**	.237	.327	.306
Reading fluency	.556*	.515**	.191	.408*	.061	.019	.141	-.046	.043	-.223
Reading accuracy	.298	.341*	.125	.261	.344*	.031	.051	-.019	-.089	-.231
Word reading skills:										
Word recognition	.309	.416*	.295	.405*	.321	.174	.126	-.158	.227	-.038
Word identification	.433**	.434**	.186	.498**	.291	.147	.156	.022	.025	-.058
Orthography	.378*	.360*	.220	.330	.188	-.011	.124	.087	.001	-.243
Word segmentation	.378*	.440**	.310	.345	.037	.206	.181	-.017	.152	.004
Spelling skills:										
Orthographic skills	.461**	.380*	.062	.460*	.245	.035	.123	.053	.001	-.081

Notes. Narrative skills: Expression, Reception and Recall of Narrative Instrument (19), Communication skills (11), reading comprehension, reading fluency and reading accuracy (12), word reading skills (12), spelling skills (12). Significance of correlation is marked *p <0.05, **p <0.01.

Prenatally Collected FGR-cohort
N=73

Perinatal and Neonatal
Deaths n=8

Neonatal FGR survivors
Delivered at 24-40 weeks
n=65

A 8 -10 years follow-up

Refused n=10
Withdrew n=8
Only preliminary
questionnaire completed n=5
No contact information n=4

Insufficient Finnish language n=1
Missing data n=1

Assessment of narrative skills
N=36

Prenatally collected AGA-cohort
n=36

Perinatal and Neonatal
Deaths n=1

Neonatal AGA survivors
Delivered at 24-40 weeks
n=35

A 8 -10 years follow-up

Excluded for severe
handicap n=4

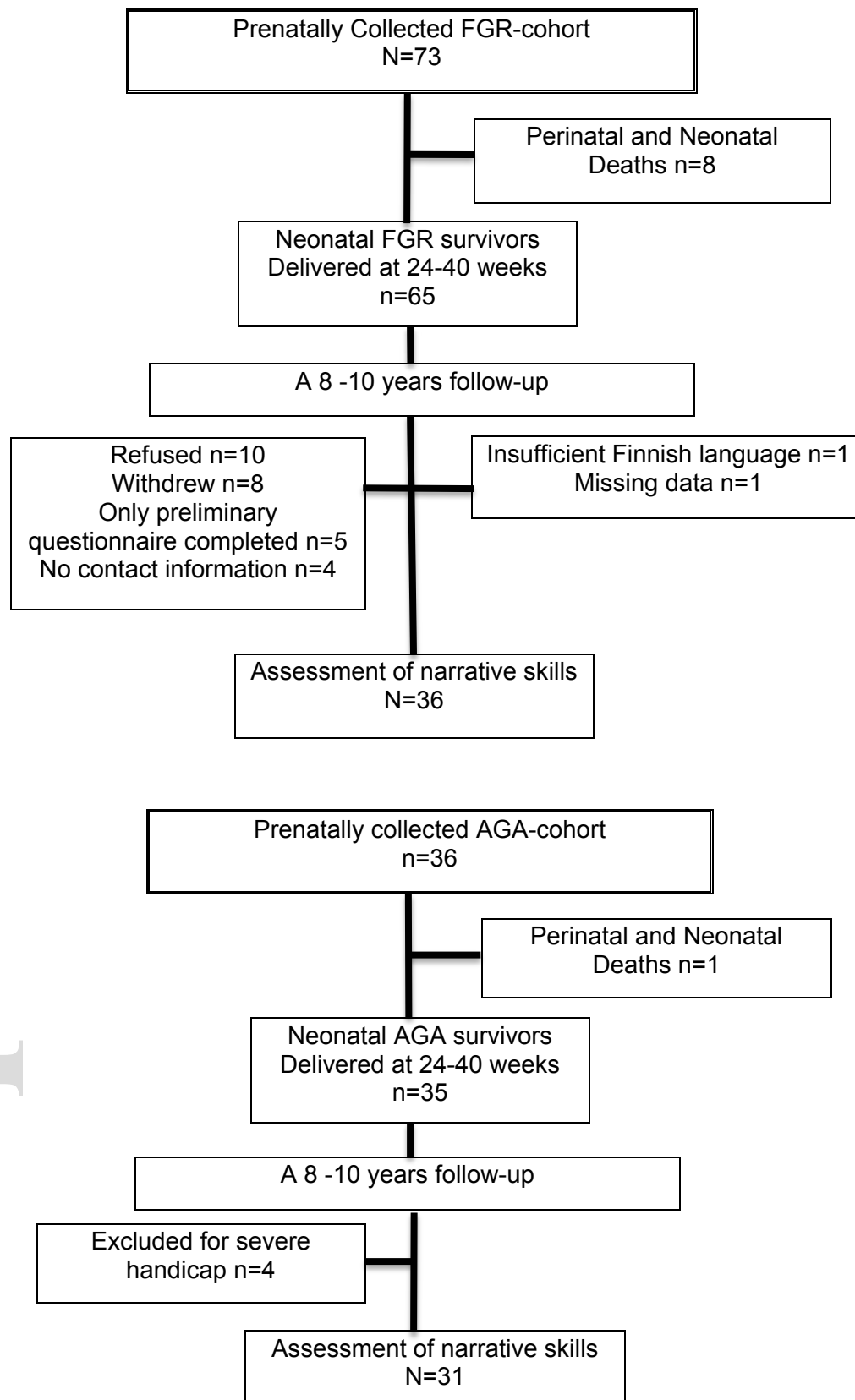Assessment of narrative skills
N=31

Figure 1. Flow diagram of the included and excluded study subjects from a prenatally collected (1998-2001) cohorts with fetal growth restriction (FGR) and appropriate growht for gestational age controls (AGA). The assessment of narrative skills.
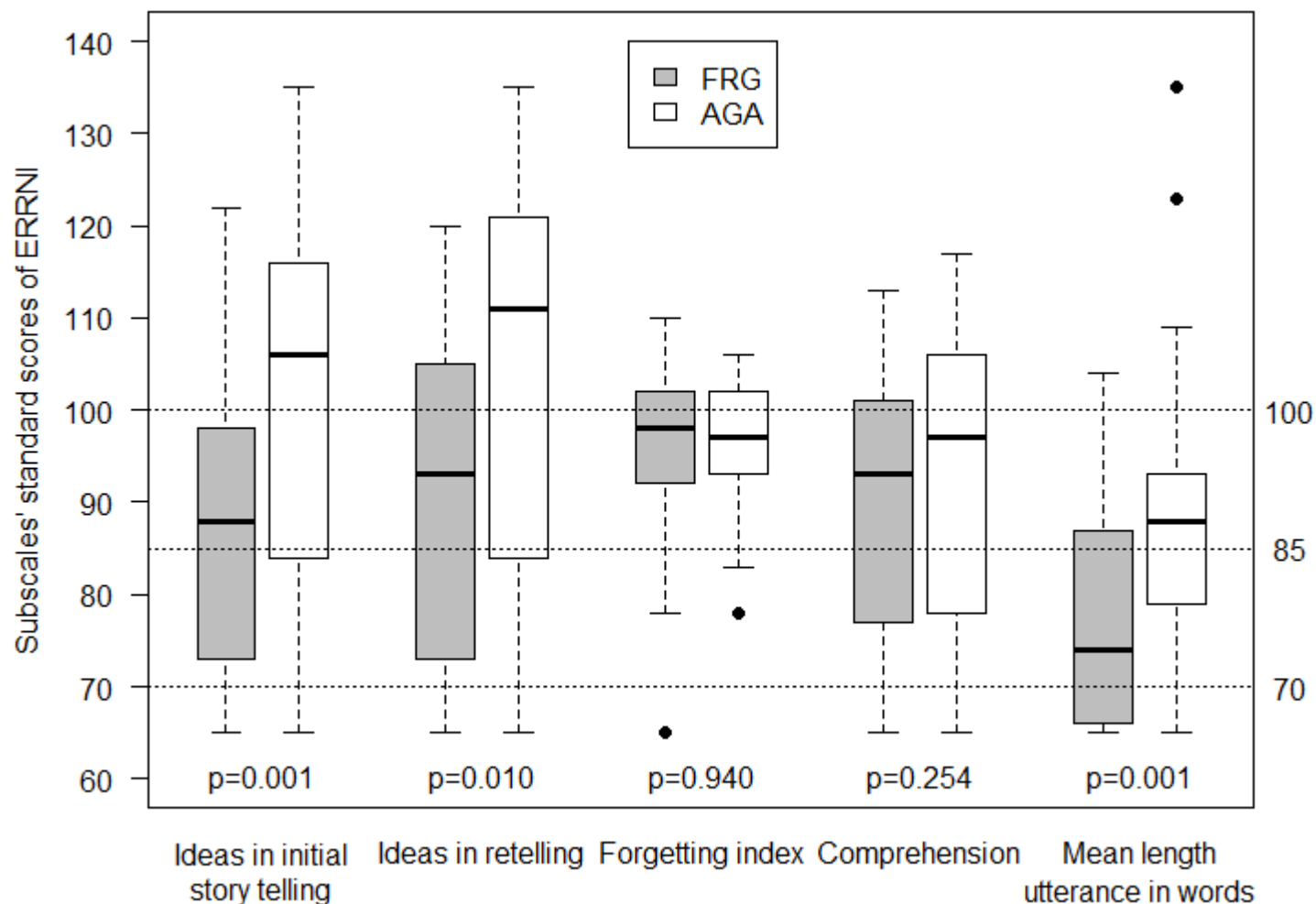
Figure 2. Standard scores of ERRNI subscales (Ideas in initial storytelling, Ideas in retelling, Forgetting index, Comprehension and Mean Length Utterance in words) in children born with fetal growth restriction (FGR) and appropriate growth for gestational age (AGA) controls. Cutting points: average level=100, average level −1SD=85, average level -2 SD = 70.