Early Human Development 106–107 (2017) 47–52

Contents lists available at ScienceDirect

Early Human Development

journal homepage: www.elsevier.com/locate/earlhumdev

Functional outcome at school age of children born with gastroschisis

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ARTICLE INFO

Article history:
Received 3 November 2016
Accepted 8 January 2017
Available online xxxx

Keywords:
Gastroschisis
Functional outcome
Physical
Motor
Cognitive
Behavioural outcome
School age
Long-term follow-up

ABSTRACT

Objective: We aimed to determine motor, cognitive and behavioural outcomes of school aged children born with gastroschisis compared to matched controls.
Study design: We compared outcomes of 16 children born with gastroschisis treated at the University Medical Center Groningen, the Netherlands, between 1999 and 2006 with 32 controls matched for gender, gestational age, birth weight, and corrected for small for gestational age (SGA) and parental socioeconomic status (SES). Intelligence, auditory-verbal memory, attention, response inhibition, visual perception, motor skills, visuomotor integration, problem behaviour and executive functioning were evaluated.
Results: Median verbal intelligence quotient and global executive functioning scores of children born with gastroschisis were poorer than of controls (95 (inter quartile range (IQR) 88–100) vs. 104 (IQR 98–113), $P = 0.001$, and 29 (IQR 6.8–63.8) vs. 5.0 (IQR 2.8–19.8), $P = 0.03$, respectively). Children with gastroschisis were more often classified as borderline or abnormal than controls regarding response inhibition (odds ratio (OR) 20.4; 95%-confidence interval (95%-CI); 2.4–171.5), selective visual attention (OR 40.4; 95%-CI 5.9–275.4), sustained auditory attention (OR 88.1; 95%-CI 5.8–1342.8), and fine motor skills (50% vs. 0%). Grade retention was more prevalent in gastroschisis children (OR 6.07; 95%-CI 1.42–25.9). These associations persisted after adjustment for SGA and SES. The auditory-verbal memory, visuomotor integration and behavioural problems did not significantly differ from the controls.
Conclusions: Gastroschisis is associated with poorer verbal intelligence, and with an increased risk for poor performance on several aspects of attention, response inhibition and fine motor skills at school age. The follow-up of children born with gastroschisis deserves attention regarding these specific domains, to improve their functional outcomes.

1. Introduction

Gastroschisis is a congenital disorder with evisceration of the bowels trough an abdominal wall defect. It is often an isolated finding, with good survival (>90%) [1,2]. Gastroschisis needs surgical intervention within 24 h of life to protect the extra-abdominal intestine. Intensive care is necessary for several days to weeks and total parenteral nutrition (TPN) for weeks to months. Repeated surgical procedures are often required. Such events in early life may affect neurodevelopment [3]. The majority of gastroschisis children are born preterm (60%) [4] and up to 61% is born small for gestational age (SGA) [5–7]. Both conditions increase the vulnerability for impaired neurodevelopment [8–11].

Studies on neurodevelopmental outcome in gastroschisis are sparse. The current study aimed to determine motor, cognitive and behavioural outcome at school age of children with gastroschisis. Comparing children with gastroschisis with a control group matched for gender, gestational age, and birth weight enabled us to explore whether the

Abbreviations: AGA, Average for gestational age; AVLT, Rey Auditory Verbal Learning Test; BRIEF, Behaviour Rating Inventory of Executive Function, Dutch version; BSID-III, Bayley Scales of Infant and Toddler Development 3rd Edition; CBCL, Child Behavior Checklist; CI, confidence interval; GA, gestational age; IQ, intelligence quotient; Lollipop, Longitudinal Preterm Outcome Project; M-ABC, Movement Assessment Battery for Children; Nepsy-2-NL, Developmental Neuropsychological Assessment Battery, Second Edition Dutch version; NICU, Neonatal Intensive Care Unit; OR, odds ratio; PIQ, performance IQ; RR, relative risk; SES, socioeconomic status; SGA, small for gestational age; TEA-Ch NL, Test of Everyday Attention for Children, Dutch version; TPN, total parenteral nutrition; TIQ, total IQ; UMCG, University Medical Center Groningen; WISC-III-NL, Wechsler Intelligence Scale, Third Edition, Dutch Version; VIQ, verbal IQ.

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http://dx.doi.org/10.1016/j.earlhumdev.2017.01.005
0378-3782/

gastroschisis in early life impacted the children's development beyond other risk factors often seen in children with gastroschisis. We hypothesized that gastroschisis hampers all aspects of development.

2. Methods

2.1. Participants

We selected all infants with gastroschisis, treated at the University Medical Center Groningen (UMCG) between 1999 and 2006. Children with additional major nonintestinal abnormalities were excluded. For every gastroschisis case we included two controls, matched, in order of importance, on gestational age (GA), gender, and birth weight.

We derived the children of the control group from two cohorts that covered similar populations and centres as we derived our patients from. The first was derived from the LOLLIPOP cohort, a large community-based prospective follow-up study on growth, development and general health in moderately preterm (GA 32–35 weeks) born children and a full-term control group, without major congenital malformations, infections or syndromes, born in 2002–2003 in the Northern provinces of the Netherlands [10]. The second cohort consisted of children included in a prospective follow-up study with very preterm (<32 weeks of gestation) SGA children as the clinical group and with very preterm average for GA (AGA) children as the control group, admitted at the NICU of the UMCG [9].

Small for gestational age was defined as birth weight below the 10th percentile of the Dutch growth charts [12]. Neonatal data, type of gastroschisis (simple or complex, defined as atresia, volvulus, perforation or necrosis of the bowel) [13], number of operations, length of total parenteral nutrition (TPN), length of hospital stay and socioeconomic status (SES) of both parents were extracted from hospital charts and a parental questionnaire, respectively. Surgical treatment consisted of primary closure or construction of a silo.

The UMCG Medical Ethical Review Board approved the study.

2.2. Measures and procedures

After parental informed consent, children of the gastroschisis group and their parents underwent a 3-hour assessment of cognitive, behavioural and motor development by a trained investigator (SB) at the outpatient clinic. The cognitive, behavioural and motor development of the control group originating from the two cohorts were evaluated by trained investigators at the outpatient clinic or at well-child clinics [9, 10].

2.2.1. Cognitive outcomes

To test verbal, performance and total intelligence, we used a short version of the Wechsler Intelligence Scale, Third Edition, Dutch Version (WISC-III-NL) [14]. Total IQ (TIQ) was estimated based on two verbal IQ (VIQ) (i.e. Vocabulary, Similarities) and two performance IQ (PIQ) subtests (i.e. Picture arrangement, Block design), all scored according to age-scaled norms [15].

We assessed selective visual attention, sustained auditory attention, and response inhibition with the subtests Map Mission, Score!, and Opposite world of the Test of Everyday Attention for Children, Dutch version (TEA-Ch NL) [16], respectively. Selective attention refers to a child's ability to select target information from an array of distractors. Response inhibition refers to the ability to inhibit an automatic response and to replace it by another response.

We used the Dutch version of the Rey Auditory Verbal Learning Test (AVLT) to assess auditory-verbal memory [17]. This test consists of five learning trials with immediate recall of words tested after each presentation assessing auditory-verbal learning, a delayed recall trial assessing long-term memory, and a delayed recognition trial. Visuomotor integration was assessed with the Design Copying subtest of the Developmental Neuropsychological Assessment Battery, Second Edition, Dutch version (Nepsy-2-NL) [18].

Grade repetition and special or regular education were derived from information provided by parents in the Dutch version of the Child Behavior Checklist (CBCL) [19].

2.2.2. Motor outcome

To appraise motor skills required in daily life, we used the Dutch version of the Movement Assessment Battery for Children (M-ABC) [20].

2.2.3. Behavioural outcome

Parents were asked to complete two questionnaires concerning behaviour. To assess behavioural and emotional problems the Dutch version of the CBCL [19] was used. Executive functioning in daily life was assessed using the Behaviour Rating Inventory of Executive Function, Dutch version (BRIEF) [21]. Executive functioning is involved in well-organized, purposeful, goal-directed and problem-solving behaviour.

2.3. Statistical analysis

We used ANOVA and Mann-Whitney *U* test, where appropriate. IQs were classified into 'normal' (IQ > 85), 'borderline' (IQ70–85) and 'abnormal' (IQ < 70). We used percentiles from standardization samples of cognitive tests and M-ABC as described in the manual to classify raw scores into 'normal' (>p15), 'borderline' (p5–p15) and 'abnormal' (<p5). For the CBCL and the BRIEF, we used a similar classification following their manuals. Differences in categorical data were tested using Chi^2-tests.

Logistic regression analyses were used to calculate odds ratios (OR) for adverse outcomes when comparing children with gastroschisis to controls. Patient demographics that differed in the gastroschisis group compared to the control group ($P < 0.10$) were entered as potential confounders in a backward logistic regression model.

A P-value <0.05 was considered statistically significant. All statistical analyses were performed with IBM SPSS v20.0, SPSS Inc., Chicago, IL, USA.

3. Results

3.1. Neonatal outcome

Nineteen neonates with gastroschisis were treated at our centre during the study period. Two patients died early due to respiratory insufficiency, and total necrosis of the small intestine caused by antenatal volvulus, respectively. We were able to contact the parents of 16 out of the 17 survivors. All agreed to participate.

Table 1 depicts demographic and perinatal characteristics of 16 children born with gastroschisis and 32 children in the control group, matched for GA, gender and birth weight in order of importance. SGA at birth was more common in gastroschisis cases (n = 7, 44% versus n = 5, 16%, respectively) and was therefore considered a potential confounder. We found no significant differences between SES of parents and Apgar scores <7 at 5 min.

Gastroschisis defects were closed primarily in 9/16 (56%). Repeated operations were necessary in 56%. Three children (19%) had additional gastrointestinal tract disorders at birth. Median length of TPN and hospitalization were 16 (range 9–401) and 24 days (range 12–357), respectively.

3.2. Cognitive outcome

Three children (19%) in the gastroschisis group received special education versus none of controls ($P = 0.07$). Of the children with gastroschisis, 7/12 (58%) repeated a grade versus 6/32 (19%) controls ($P = 0.02$). Table 2 depicts cognitive results. One child's intelligence was assessed at school shortly before our evaluation, thus we used the