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Non-cardiac Surgery in Neonates with Major Congenital Anomalies: a Pilot Study on the Developmental and Psychosocial Effects in Toddlers

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Non-cardiac Surgery in Neonates with Major Congenital Anomalies: a Pilot Study on the Developmental and Psychosocial Effects in Toddlers

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ABSTRACT – *Past studies have shown that infants who underwent surgery in the neonatal period because of major congenital anomalies, often show mental and psychomotor developmental delays in later life. Furthermore, less optimal parent-child interaction among these families have been found. The purpose of this pilot study was to gain a first insight in behavior problems, developmental delays and parent-child interaction in non-cardiac surgery requiring CA infants as a group, and outcomes and implications will be considered in the set up of a longitudinal research in the Wilhelmina Children Hospital (WCH) in Utrecht. The current study investigated parents of 36 toddlers (M = 43.6 months, SD = 11.0), who underwent surgery in the WCH in Utrecht because of major non-cardiac congenital anomalies. Being part of the non-cardiac CA group, infants with developmental intrusive syndromes were included. Parents completed questionnaires through which developmental delays (Ages and Stages Questionnaire), behavior problems (Child Behavior Checklist) and parent-child interaction (Nijmeegse Ouderlijke Stress Index) were measured. Results were compared to the tests' normative groups, except for developmental delays (percentages were given). Contrary to prior research, significantly less behavior problems and better parent-child interaction were found. In addition, non-optimal parent-child interaction was found to be predictive for developmental delays. Lastly, analyses were run for the non-syndromic infants solely and similar outcomes on behavior problems and parent-child interaction were found. Despite the fact that comparison between non-syndromic children and the whole group of participants on developmental delays was not tested for significance, developmental delays were found to be noticeable lower in the non-syndromic children.*

Niet-cardiale Operaties bij Pasgeborenen met Grote Aangeboren Afwijkingen: een Pilot Studie naar de Ontwikkeling en Psychosociale Effecten in Peuters

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Samenvatting – *Onderzoeksresultaten naar de gevolgen van een operatie ten gevolge van een grote aangeboren afwijking bij pasgeborenen, duiden op mentale en psychomotorische ontwikkelingsachterstanden op latere leeftijd. Ook zijn er aanwijzingen voor een minder optimale ouder-kind interactie. Het doel van deze pilot studie was om een eerste inzicht te krijgen in de gedragsproblemen, ontwikkelingsachterstanden en ouder-kind interactie in kinderen met een niet-cardiale aangeboren afwijking waarvoor een operatie kort na de geboorte vereist is. Resultaten en implicaties zullen worden overwogen en meegenomen bij de opzet en start van een longitudinaal onderzoek in het Wilhelmina Kinderziekenhuis (WKZ) in Utrecht. In huidig onderzoek vulden ouders van 32 peuters (M = 43,7 maanden, SD = 11,1) die een operatie ondergingen in het WKZ ten gevolge van een grote aangeboren niet-cardiale afwijking, van 2 tot 5 jaar oud, vragenlijsten in. Omdat kinderen met syndromen onderdeel zijn van de groep kinderen met niet-cardiale operatie vereisende aangeboren afwijkingen, werden kinderen met ontwikkelings-beïnvloedende syndromen geïncludeerd. Op basis van de door ouders ingevulde vragenlijsten werden de eventuele ontwikkelingachterstanden (Ages and Stages Questionnaire), gedragsproblemen (Child Behavior Checklist) en mate van ouder-kind interactie (Nijmeegse Ouderlijke Stress Index) van hun kind gemeten. Resultaten werden vergeleken met de scores van de normgroepen op de vragenlijsten, met uitzondering van ontwikkelingsachterstanden (alleen percentages werden beschreven). In tegenstelling tot eerdere onderzoeksresultaten werden minder gedragsproblemen en een betere ouder-kind interactie gevonden. De ouder-kind interactie bleek bovendien een voorspeller voor ontwikkelingsachterstanden. Tenslotte werden dezelfde analyses uitgevoerd voor de participanten zonder syndromen en ook bij deze groep werden minder gedragsproblemen en een betere ouder-kind interactie gevonden, die daarnaast voorspellend bleef voor ontwikkelingsachterstanden. Hoewel een vergelijking tussen niet-syndromale kinderen en de totale groep participanten op ontwikkelingsachterstanden niet op significantie werd getest, werden zichtbaar minder ontwikkelingsachterstanden bij niet-syndromale kinderen gevonden.*

Introduction

Approximately 183,000 babies a year are born in the Netherlands, of which roughly 7000 are born with anatomical congenital anomalies (CA) (Mohangoo & Buitendijk, 2009). These malformations consist of physical defects involving many different parts of the body, including the brain, heart, lungs, liver, bones, and intestinal tract. Some of these defects have a life threatening impact requiring immediate surgical interventions in the neonatal period. The most well-known and researched CA which require surgery, are the congenital heart defects (structural anomalies of the heart). Other less examined examples of major surgery needing non-cardiac CA are: abdominal wall defects (e.g. intra abdominal organs situated outside of the body), congenital diaphragmatic hernia's (malformation of the diaphragm allows the abdominal organs to push into the lungs, leading to obstruction), esophageal atresia's (obstruction of the oesophagus), anorectal malformations), Hirschprung's diseases (absence of nerves in the rectum and part of the last part of the colon, causing obstruction), urogenital defects (e.g. ambiguous) and ear- nose- and throat anomalies (e.g. choanal atresia).

Improved care and new surgical techniques have caused a significant decrease of mortality rates in babies who require surgery. As a result of this, morbidity rates have increased and the focus of attention has shifted to the long-term consequences of these anomalies on normative development. In other words, given that surgery enhances the survival rates in newborns, the question of whether there are developmental consequences in these children becomes more important. As mentioned, a great deal of research on these consequences and subsequent adapted care has been focused on children with congenital heart defects, although there are roughly as many children with non-cardiac CA (Mohangoo & Buitendijk, 2009). There is a great lack of studies aimed at the specifically non-cardiac children as a group for various reasons. Firstly, different underlying factors in infants can cause non-cardiac CA. For example underlying development limiting syndromes (e.g. Down-syndrome) are associated with CA. In most studies in children with non-cardiac CA, children with syndromes are excluded from analyses. In addition, the various congenital defects are heterogeneous and numbers of grouped non-cardiac CA are relatively small. Nevertheless, these children have all endured bodily defects which required immediate surgery and aftercare.

In this research, the focus of attention is aimed at the possible consequences in these underdeveloped children with major non-cardiac CA, who have undergone surgery in the neonatal period. The purpose of this study is to examine the long-term consequences on development, behavior and parent-child interaction in all of these children, as a group. To gain an insight into the

consequences in this group, and as infants with developmental intrusive syndromes are part of the non-cardiac CA children, children with syndromes will be included.

Outcomes of studies on (non-cardiac) CA thus far

Growing evidence suggests that major surgery in neonates may be associated with (neuro)developmental impairment (Walker, Holland, Winlaw, Sherwood & Badawi, 2006). Outcomes of studies aimed at the specific CA individually support this proposition of developmental impairment, including such behaviors as emotional and behavioral difficulties in children born with congenital diaphragmatic hernia's and oesophageal atresia's (Bouman, Koot, & Hazebroek, 1999).

As mentioned before, within the CA disease-specific studies the majority is aimed at the (neuro)developmental and psychosocial outcomes in infants who suffer from congenital heart defects. Test results reveal cognitive and motor delay in early infancy in these children (Snookes et al., 2010). In the little research aimed at the long-term outcome in infants with major non-cardiac CA surgery as a group, three types of studies have been conducted: firstly, research in which children with congenital heart defects are included; secondly, studies in which cardiac children are compared to children with non-cardiac CA surgery; and thirdly, studies focused at the non-cardiac CA infants.

Outcomes including cardiac defects

An example of a study in which the cardiac surgery group was included, is a study of Van Der Cammen-Van Zijp et al. (2010). In this study 5 year olds (*N* = 102) were tested, who were born with major CA and therefore underwent (multiple) surgical interventions. Besides congenital heart defects, children with congenital diaphragmatic hernia's, esophageal atresia's, small intestinal anomalies and abdominal wall defects were included. Patients with syndromic abnormalities were excluded. The aim of the study was to determine whether or not these CA children were at risk for impaired motor-function and reduced exercise capacity. Scores of participants were compared to the tests' normative group and a significant delayed motor-function performance and disturbed exercise capacity were found. Since cardiac children are at significant risk for cognitive and motor delay, inclusion of these children might have affected the outcome negatively in the CA children as a group.

Cardiac CA compared to non-cardiac CA

In the study of Walker et al. (2012), a distinction between cardiac CA and non-cardiac CA infants was made. The aim of their study was to describe developmental outcomes of infants in both cardiac (*n* = 128) and non-cardiac (*n* = 172) groups compared to a group of healthy controls (*n* = 239), and in addition compare both groups to each other. Infants were assessed at 1 year of age using the Bayley Scales of Infant and Toddler Development (BSID), and a significant developmental delay on 4 out of 5

subscales (cognition, receptive language, fine motor, gross motor) in both groups was found, compared to the healthy control group. On the fifth subscale, 'expressive language', no significant developmental delay was found for the non-cardiac children. Lastly, the outcomes of both tested groups were compared to each other, and a significantly greater developmental delay was found in the cardiac group compared to the non-cardiac group on all named subscales, except for fine motor skills.

A second study found developmental delays in both cardiac and non-cardiac CA infants as well. In an Australian sample of infants with major CA requiring cardiac (*n* = 50) and non-cardiac (*n* = 45) surgical interventions, Laing, Walker, Ungerer, Badawi and Spence (2011) compared the groups on mental- and motor development using the BSID test. Here, a developmental delay at 24 months of age was found in the cardiac group and in the non-cardiac group as well: compared to the normal population scores, infants in both groups had significantly greater mental (Mental Developmental Index: MDI) and motor delays (Psychomotor Developmental Index: PDI). Yet, contrary to the significantly greater delay in cardiac children found by Walker et al. (2010), in this study no differences were found between the cardiac and non-cardiac CA children.

Outcomes in non-cardiac CA exclusively

Lastly, there is a dearth of studies that focused exclusively on non-cardiac CA infants. The longitudinal cohort study of Gischler et al. (2009) focused on physical, mental and psychomotor developmental outcomes, and participants (*N* = 101) were children with major anatomical non-cardiac CA admitted to the pediatric surgical intensive care within 7 days after birth. Patients with syndromal abnormalities were excluded. All children were tested at 6, 12, 18 and 24 months of age by a professional pediatrician and psychologist. Overall birth weight and head circumference were found to be within the norm, but the CA survivors did show significantly impaired growth. In addition, psychomotor and mental developmental outcomes were assessed using the BSID and compared to Dutch population norms. A significantly psychomotor developmental delay (PDI total score) in the CA group was found. Contrary to what they expected, no significant mental delay (MDI total score) was found. Both psychomotor and mental development proved to be influenced by the numbers of CA and number of surgical interventions needed to correct the bodily defects.

Non-cardiac CA infants & parent-child interaction

Taking all these outcomes into account, results imply a great risk for developmental mental and psychomotor developmental delays in later life in infants who are born with major non-cardiac surgery requiring CA. Given that these children are faced with immediate surgery and bodily impairments from the start of their lives, and moreover are confronted with possible developmental

delays, the focus of attention shifts to the impact of these worrisome children on parents. To be more specific, as parent-child relationships are transactional (Belsky, 1984), the question of reciprocal impact of these children on their relationship with parents becomes important.

Smith, Oliver, and Innocenti (2001) examined the relationship between toddlers' disabilities and parenting stress. Participants were parents (*N* = 880) of children who were moderately or severely developmentally delayed (as measured by the Battelle Developmental Inventory). Parenting stress was measured by means of the Parenting Stress Index/Short that provides a total score and three subscale scores: parental distress, parent-child dysfunctional relationship, and difficult child. Although the severity of the child's disability had minimal impact on parental stress, it was found to have a noteworthy impact on stress related to the parent-child relationship.

Another study that highlights the impact of the child's impairments on parent-child interaction is a study of Laing et al. (2010). Given that parents behaviors towards children play a crucial role in child development during the toddler years, they examined in addition to developmental outcomes the mother-child interaction in children with major CA (including cardiac CA infants) requiring surgery. Children (*N* = 93) were between 18 and 36 months of age and were assessed developmentally with the BSID. Quality of mother-child interaction was assessed using an observational measure (the National Institute of Child Health and Human Development Qualitative Scales of the Observational Ratings of Mother-Child Interaction), based on videotaped mother-child free-play interactions. Results indicate significantly more non-optimal mother-child interactive behaviors compared to a normal population. In addition, a strong relationship between the child's developmental capacity and the mother's interactive behaviors was found: compared to children developing normally (MDI and PDI score > 85), those with developmental delay (MDI and PDI score < 85) showed significantly more non-optimal interactive behaviors with their mothers compared to the normal population. Hence, Smith, Oliver and Innocenti, and Laing et al. add another important matter to the outcomes in children with major CA: possible reciprocal effects in children with delays (because of major CA) and their parents.

Current study

In the current study we were interested in further examining the developmental outcomes in the group of children with non-cardiac CA. We investigated the outcomes on developmental delays, behavioral problems and parent-child interaction in toddlers ranging from 2 to 5 years of age. This age span was chosen, given that assessment in this period is proven to be highly predictive of cognitive and motor outcomes in later life (Ludman, Spitz & Wade, 2001; Mazer et al., 2010). Outcomes were measured through questionnaires, completed by parents. Potential developmental

delays were measured through the Ages and Stages Questionnaire (Squires, Potter & Brickler, 1999), social-emotional development was measured through the Child Behavior Checklist (Verhulst, Koot, Akkerhuis, & Veerman, 1990) and parental stress was measured through the Nijmegen Parental Stress Index (De Brock, Vermulst, Gerris, & Abidin, 1992).

The current study had three aims. Firstly, the developmental outcomes of 2-5 year old children who had undergone non-cardiac CA surgery were compared to a normative group of children who had not had surgery. Based on previous research we hypothesize behavior problems (Bouman, Koot, & Hazebroek, 1999), developmental delays and a non-optimal parent-child interaction (Laing et al., 2010) (compared to the tests' normative groups) in these toddlers. Secondly, the relationship between parent-child interaction and developmental delays was examined. As the parent-child relationship is known to be important for parent-child attachment, and social-emotional, cognitive and language development, our second hypothesis is that we expect a relationship between non-optimal parent-child interaction and developmental delays. Lastly, more research is needed to specify the outcomes within the relative heterogenic non-cardiac CA group. Since possible underlying syndrome defects are at the origin of CA, the third aim of this study was to make a distinction between outcomes in children with syndromic and non-syndromic CA. Because of the fact that in most previous research infants suffering from syndromes were excluded, and still developmental delays were found in those studies, we expect developmental delays (compared to the tests' normative group) in the non-syndromic group as well.

Method

Participants

Participants were 36 toddlers (67 % boys, 33 % girls) who ranged in age from 29 to 64 months (mean age = 43.6 months, *SD* = 11.0). They were born from March 2007 to March 2009, and admitted to the Neonatal Intensive Care Unit (NICU) and Pediatric Intensive Care Unit (PICU) in the WCH in Utrecht. Additional study inclusion criteria were infants born with major non-cardiac anomalies, who underwent surgery in the first 52 weeks after conception. Major non-cardiac anomalies were subdivided into subsequent groups: abdominal wall defects (AWD; *n* = 7), congenital diaphragmatic hernia's (CDH; *n* = 1), small intestinal anomalies (SIA's; *n* = 7) esophageal atresia's (EA; *n* = 7), anorectal malformations (ARM's; *n* = 7), urogenital defects (*n* = 2), ear- nose- and throat anomalies (ENT; *n* = 4) and additional (*n* = 1). Furthermore, 5 children were suffering from a developmental intrusive syndrome (14 %: Down syndrome (*n* = 2), CHARGE syndrome, Pierre Robin Sequence, Smith Magenis Syndrome).

Procedure

The study was approved by the Medical Ethical Committee of the WCH. Participants were selected from the electronic medical record system applied in the WCH. To participate, parents of selected infants were approached by telephone by one of the pediatrician (*N* = 50, 100 % of parents consented), after which the questionnaires and informed consent forms were sent (72 % was sent back, 100 % written consent of parents).

Measures

Parents of participants completed questionnaires at home and were asked to send it back by way of self-addressed envelopes (72 % was returned, *N* = 36). Furthermore they were asked to hand over an envelope to the teacher/babysitter of their infant, including a questionnaire (to evaluate children's emotional and behavioral problems: the Teacher Report Form (TRF; Verhulst et al., 1997b) and an informed consent form (78 % was sent back, of which 14 % did not return their informed consent forms). Because of the little amount of returned questionnaires and missing informed consents by teachers/babysitters (*n* = 24), the TRF questionnaires were not included in analyses.

Neonatal data

Neonatal data were collected during hospitalization and were extracted from the electronic medical record system applied in the WKH.

Child development

An indication of children's development was measured using a Dutch adaption of the Ages and Stages Questionnaire (ASQ; Squires, Potter, & Bricker, 1999). Developmental delays were calculated using ten age assorted questionnaires in designated timed intervals (4 to 60 months). All questionnaires contained 38 questions suitable for the age interval, concerning five developmental domains: communication (Cronbach's α was .90), fine motor skills (Cronbach's α was .85), gross motor skills (Cronbach's α was .89), problem solving (Cronbach's α was .86) and personal-social development (Cronbach's α was .86). Parents were required to assess their children's abilities by examining the questions (e.g., *"Does your child stand up in the middle of the floor by himself and take several steps forward?", "Does your child help turn the pages of a book?"*). Items were rated on a 3-point scale (*"Yes":* 10 points, *"Sometimes":* 5 points, *"Not yet":* 0 points). For the five developmental domains, total scores (high scores (≥ 40) indicated no developmental delays) and clinical scores (which stands for a notable low score compared to an average child of their age: total scores ≤ 40)

were calculated. Total developmental delays were measured by adding up clinical scores of the five domains (total clinical score > 2 was determined as total developmental delay).

Child behavior

The Dutch adaption of the Child Behavior Checklist (CBCL; Verhulst, Koot, Akkerhuis & Veerman, 1990) for children aged 1.5 to 5 years, was used to evaluate children's emotional and behavioral problems, as experienced by parents. With this reliable scale, internalizing (i.e., anxious, depressive, and overcontrolled; Cronbach's α was .76) and externalizing (i.e., aggressive, hyperactive, noncompliant, and undercontrolled; Cronbach's α was .93) and total behavior problems (Cronbach's α was .95) were assessed. Items were rated along a 3-point scale (0 = not at all true/never, 1 = somewhat true/sometimes, 2 = completely true/frequently). Questionnaires raw scores and *t*-scores (≥ 65 = subclinical, ≥ 70 = clinical) were calculated, where high raw scores and (sub)clinical *t*-scores on all three scales stand for above average behavior problems.

Parent-child interaction

The degree of parent-child interaction was measured using the Nijmeegse Ouderlijke Stress Index (NOSI; De Brock et al, 1992). This test assesses two domains of stress, as experienced by parents: the parent- and child domain. The questionnaire contained 123 items spread among 13 subscales, of which seven subscales in the parent domain (e.g. Competence, Attachment, Depression) and six subscales in the child domain (e.g. Mood, Distractibility, Acceptation). Items were rated along a 6-point scale (0 = not at all, 6 = completely true). To measure parent-child interaction, three subscales regarding parent-child interaction were selected: attachment (the degree of created emotional bounds between parent and child; Cronbach's α was .74), positive reinforcement (the degree of by parents experienced positive reinforcement from their child; Cronbach's α was .45) and acceptance (the degree in which children fall short of expectations of parents on physical, intellectual and emotional level; Cronbach's α was .86). Raw total scores on the three subscales were calculated, where high scores stand for less positive parent-child interaction.

Analyses

Data were entered into a computer for editing and analysis, using IBM Statistical Package for the Social Sciences (SPSS) 20 for Windows. Scores on the CBCL and NOSI were compared to tests normative groups by way of independent *t*-tests. Regression analyses were used to interpret the predictive value of parent-child interaction on developmental delays. Lastly, a comparison in outcomes between syndromic and non-syndromic infants was made, and the *t*-tests and regression analyses were run for the non-syndromic infants only.

Results

Preliminary analyses

Missing data on the NOSI (*n* = 14, total of 23 questions) and ASQ (*n* = 3, total of 5 questions) were manually calculated, as prescribed and approved in both of the questionnaires manuals. After data were entered into SPSS, 4 participants were excluded because they fell out of the designated ASQ age categories. Table 1 presents the descriptive statistics for participants, after exclusion.

Table 1

Baseline Characteristics

	n (%)	Mean age in months (*SD*)	Mean gestational age in weeks (*SD*)
All	32 (100.0)	43.7 (11.1)	38.2 (3.7)
Gender			
Boys	22 (68.8)	43.4 (11.2)	37.3 (4.0)
Girls	10 (31.2)	44.5 (11.3)	40.0 (1.8)
CA[a]			
1. Small intestinal anomalies	7 (21.9)		
2. Esophageal atresia	7 (21.9)		
3. Abdominal wall defects	6 (18.8)		
4. Anorectal malformations	6 (18.8)		
5. Urogenital defects	2 (6.3)		
6. Ear/nose/throat defects	2 (6.3)		
7. Diaphragmatic hernia	1 (3.1)		
8. Additional	1 (3.1)		
Syndrome			
Yes[b]	5 (15.6)		
No	27 (84.4)		

[a]Congenital anomalies requiring surgery. [b]Developmental intrusive syndromes: Down syndrome (*n* = 2), CHARGE syndrome, Pierre Robin Sequence, Smith Magenis Syndrome.

Descriptive analyses

As parent-child interaction was measured through three subscales ('Attachment', 'Positive reinforcement' and 'Acceptance'), factor extraction was performed on these subscales to reduce multicollinearity in regression. Because the numbers of variables were less than 30, and resulting communalities after extraction were all greater than .7, Kaiser's criterion was used for factor selection (factor analyses descriptives are presented in Table 2). This criterion recommends retaining all factors with *Eigenvalues* greater than 1: 'Attachment' and 'Positive reinforcement' were extracted and the subscale 'Acceptance' was used as 'Parent-child interaction' for the remaining analyses.

Table 2

Factor Analyses Descriptives (n = 32)

	Communalities		Initial Eigenvalues		
	Initial	Extraction	Total	% of Variance	Cumulative %
Subscale[a]					
Acceptance	1.00	.76	2.21	73.86	73.86
Attachment	1.00	.72	0.42	19.00	87.83
Positive reinforcement	1.00	.74	0.37	12.20	100.00

[a] On the Nijmeegse Ouderlijke Stress Index.

Primary analyses

Purpose 1

The first purpose was to compare participants to the tests' normative groups, on developmental delays (all ASQ subscales), behavior problems (CBCL subscales: internalizing, externalizing and total problems) and parent-child interaction (NOSI subscale 'Acceptance'). The ASQ contains 10 age categories, in which normative means are given per developmental domain (10 x 5 = 50 normative means). No comparable overall means were found, because of why only percentages of clinical scores in current study can be presented (Figure 1). Outcomes of two-tailed *t*-test analyses on behavior problems and parent-child interaction are presented in Table 3.

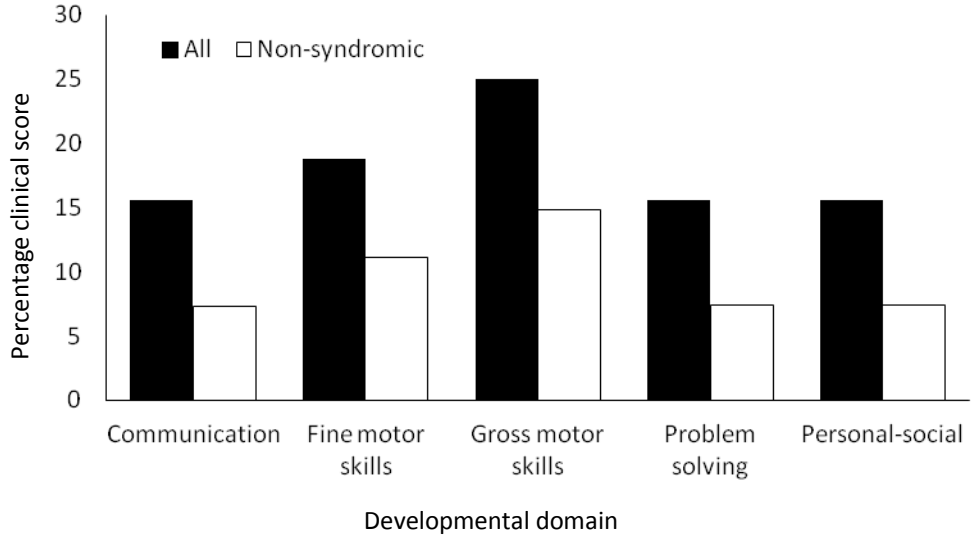

Figure 1. Developmental delays (clinical score: total score on subscale ≤ 40) on the subscales of the Ages and Stages Questionnaire.

Table 3

T-tests Analyses Comparing Participants (n = 32) to the CBCL and NOSI Normative Groups

		Normative Mean (*SD*)	Participants Mean (*SD*)	*T*-score
Behavior problems[a]				
Internalizing problems	All	8.6 (0.2)	6.4 (.8)	-2.56*
	Non-syndromic		5.4 (4.6)	-3.56**
Externalizing problems	All	12.9 (0.3)	10.2 (1.6)	-1.71
	Non-syndromic		9.1 (8.6)	-2.31*
Total problems	All	33.3 (0.4)	26.0 (3.5)	-2.08*
	Non-syndromic		22.9 (19.0)	-2.84**
Parent-child interaction[b]	All	22.6 (3.6)	18.3 (1.4)	-3.12**
	Non-syndromic		16.9 (6.8)	-4.42**

[a]Measured through the Child Behavior Checklist: total raw scores on subscales are given. [b]Total raw score on the subscale 'Acceptance', measured through the Nijmeegse Ouderlijke Stress Index.
*$p < .05$. **$p < .01$.

For behavior problems, independent *t*-tests were performed to compare mean raw scores of participants to mean raw scores of the CBCL American norm group on the three chosen subscales. Participants experienced significantly ($t(31) = -2.56$, $p = .016$) less internalizing problems, compared to the CBCL norm group. On externalizing problems, there were no significant differences ($t(31) = -1.71$, $p = .098$) between participants ($M = 10.2$, $SE = 1.6$) and the CBCL norm group ($M = 12.9$, $SE = .3$). On total behavior problems significantly ($t = -2.08$, $p = .046$) less behavior problems were found in participants ($M = 26.0$, $SE = 3.5$), compared to the CBCL norm group ($M = 33.3$, $SE = .4$).

For parent-child interaction, an independent *t*-test was performed to compare the mean total raw score to the mean total raw score of the Dutch NOSI norm group on the subscale 'Acceptance' (considered as parent-child interaction). As $n = 31$ mothers filled in the NOSI questionnaires, the norm group table 'for mothers' was used. Participants experienced significantly ($t(31) = -3.12$, $p = .004$) better parent-child interaction ($M = 18.3$, $SE = 1.4$), compared to the NOSI norm group ($M = 22.6$, $SE = 3.6$).

Purpose 2

The second purpose was to examine the relationship between parent-child interaction and developmental delays. Data were analyzed using a standard multiple regression analysis. The dependent variable was the total clinical score on the ASQ, defined as the number of clinical scores on the subscales (possible scores from 0 (no clinical scores at all) to 5 (clinical scores on all subscales)). The predictive variable was parent-child interaction, defined as the total raw score on

the subscale 'Acceptance'. Covariates were gender, age and syndrome.

Results for evaluation of assumptions for skewness, normality, linearity and homscedasticity of residuals were met. With the use of a $p < .05$ criterion for Mahalanobis distance, one outlier (df = 4, $\chi^2 = 10.59 \geq 9.49$) among the cases was found. Rerunning analyses without the outlier did not make a difference, and as a result the outlier was not removed from analysis. In addition, no cases had missing data and no suppressor variables were found, $N = 32$.

Table 4 displays the unstandardized regression coefficients (B) and intercept, and the standardized regression coefficients (β). The results of the regression indicated the predictors explained 67 % of the variance (R^2 = .67, (F = 11.22, p = .00). It was found that parent-child interaction significantly predicted developmental delays (ß = .49, p .00), as did syndrome (ß = -.51, p = .003).

Tabel 4

Multiple Regression Analysis Predicting Developmental Delays

	B	*SE B*	ß
Parent-child interaction			
All[a]	.11	.03	.53**
Non-syndromic[b]	.09	.03	.58**
Gender			
All	.55	.38	.16
Non-syndromic	.41	.36	.19
Age			
All	-.00	.02	.16
Non-syndromic	-.01	.02	.19
Syndrome			
All	1.77	.55	.41**

Note. Developmental delays consist of the total clinical score on the Ages and Stages Questionnaire, defined as the number of clinical scores on the subscales (possible scores from 0 (no clinical scores at all) to 5 (clinical scores on all subscales)). [a]All participants (N = 32). [b]Non-syndromic infants (n = 27).
**$p < .01$.

Purpose 3

The third and last purpose was to specify outcomes within the non-cardiac CA group. Therefore a distinction between syndromic and non-syndromic CA was made: because of the small group of infants with syndromes (n = 5), calculating percentages, the t-tests and regression analyses were run for the non-syndromic infants only (n = 27). Percentages on clinical scores on the developmental domains in the non-syndromic infants only, are presented in Figure 1. Outcomes of two-tailed t-test

analyses are presented in Table 3. For behavior problems, significantly less internalizing problems ($t(26) = -3.56$, $p = .001$), externalizing problems ($t(26) = -2.31$, $p = .029$) and total problems ($t(26) = -2.84$, $p = .009$) were found in the non-syndromic children, compared to the CBCL norm group. For parent-child interaction, significantly better parent-child interaction ($t(26) = -4.42$, $p = .000$) was experienced by mothers of the non-syndromic infants, compared to the NOSI norm group.

The data for non-syndromic infants only were analyzed using the same standard multiple regression analysis as used on the whole sample, with the exception of the covariate syndrome. Table 4 displays the unstandardized regression coefficients (*B*) and intercept, and the standardized regression coefficients (β). The dependent variable was the total clinical score on the ASQ, defined as the number of clinical scores on the subscales (possible scores from 0 (no clinical scores at all) to 5 (clinical scores on all subscales)), the predictive variable was parent-child interaction (defined as the total score on the subscale 'Acceptance') and covariates were gender and age. The results of the regression indicated that the predictors explained 40 % of the variance ($R^2 = .40$, ($F = 5.16$, $p = .007$). Lastly, it was found that parent-child interaction significantly predicted developmental delays (ß=.58, $p = .002$) in non-syndromic infants.

Discussion

The present pilot study examined developmental delays, behavior problems and parent-child interaction in children who underwent surgery because of major non-cardiac CA in the neonatal period. Previous research indicates developmental delays, behavior problems and a non-optimal parent-child interaction in these children. Furthermore, the latter was expected to be predictive of developmental delays. Current results are used as a first insight in this research group, and outcomes and implications will be considered in the set up of a longitudinal research in the WCH (starting in the year of 2013).

Purpose 1

The first purpose was to investigate whether the non-cardiac CA infants differ from tests' normative groups on developmental delays, behavior problems and parent-child interaction or not.

Developmental delays

Because no comparable means for the group as a whole were found, only descriptive percentages of developmental delays in participants were given. Developmental delays were found on all ASQ domains, ranging from 15 % (in the subscales communication, problem solving and personal-social)

to 25 % (in gross motor skills) of participants having a delay. Percentages of fine and gross motor skills were found to be notably higher compared to the other domains. This is in line with the high psychomotor delays found in previous research (Gischler et al., 2009; Walker et al., 2010; Laing et al., 2011).

Behavior problems

On behavior problems, more internalizing, externalizing and total problems (compared to the normative group) were expected (Bouman, Koot, & Hazebroek, 1999). To the contrary, *less* internalizing and total behavior problems, and on the externalizing subscale no differences, were found. Put differently, parents experienced less behavior problems in their children compared to the reference group on the questionnaire. These previous research opposing results may be caused by the fact that the sample of Bouman, Koot and Hazebroek is possibly not representative enough for the non-cardiac CA infants as a group: the sample of Bouman et al. consisted of 10-year-old children who suffered from just esophageal atresia, as opposed to toddlers with different sorts of CA in current study.

In addition to this implication, behavior problems were measured by way of questionnaires, filled in by parents. Because of this, answers might be influenced by parents using *downward comparison* (Aronson, Wilkert & Akert, 2007): in light of this theory parents might have compared their children's behaviors to the difficult and life threatening beginning of their infants' lives. In line with this self-protective and self-enhancing theory, parents decide in a way that enhances their positive thoughts of handling the fact that they have a disabled child. In accordance with this they may reason that, compared to the difficult start, their children are doing pretty well and choose the most positive alternative in questions. This assumption is supported by the many additional comments of parents to the questionnaires: frequently parents wrote down how proud they were and how well their children were doing, compared to the life threatening situation they were born in.

Parent-child interaction

In previous research, child's disability was found to have a noteworthy impact on the parent-child relationship (Smith, Oliver, & Innocenti, 2001) and in addition, a non-optimal parent-child interaction was found in CA children (Laing et al., 2010). As opposed to these outcomes, *better* parent-child interactions were found in the current study's participants compared to the tests' normative group. First, even though the samples are comparable (toddlers suffering from non-cardiac CA in both studies), these studies differ from the current study in measuring parent-child interaction: both Smith, Oliver and Innocenti, and Laing et al. used observational measures interpreted by professionals, as opposed to parental questionnaires in the current study. As mentioned before,

parents may have used downwards social comparison to determine their answers, leading to a more rosy evaluation of their child.

In addition to this, the *perception* of parents might have interfered with the actual behavior problems and parent-child interaction too. In observing the world, people yield important clues to causal structures which is influenced by perception (Goswami, 2008). Perception does not only consist of registering and becoming aware of the physical structure of the world true the eyes, ears, mouth, nose and skin. Moreover, it is part of how people register all causal structures or events, and in addition part of how they make decisions. Biases and heuristics are known to influence these causal attributions and arise out of experiences that are learned and stored in memory, and lead to probability and logic based reasoning.

An approach in research on 'decision-making', is an approach generally referred to as *adaptive thinking* by Gigerenzer (Ashcraft, 2006). Gigerenzer not only focused on the probability and logic factors of reasoning, but reasoned that people use these heuristics because they are adaptive in the sense of leading to successful behavior or feelings. In other words, people make their decisions based on the usefulness of judgments. An example of this is "Take the best heuristic" (Gigerenzer & Goldstein, 1996) while making a decision. This process occurs when a person recognizes both alternatives, after which (based on the first useful information they retrieve) they choose based on positive evidence. Based on this "Take the best heuristic" idea, test results in the current study may be influenced by parents' perceptions of the negative and positive experiences of their child. In accordance to this theory, parents might have recognized their children in both bad and good behaviors, but in the end chose the most positive alternative.

Additionally, as parent-child interaction is part of attachment, the question whether the process of attachment develops differently in these children arises. Because of the fact that the non-cardiac CA infants need specified and more care from the beginning of their lives, a close-knit family bond could be imagined, leading to secure attachment. However, van IJzendoorn et al. (1992) conducted a meta-analysis on attachment in disabled children of 13 samples from eight studies of children with a range of disabilities including Down's syndrome, autism, cystic fibrosis and congenital heart disease. They found that rates of secure attachments were generally lower (less than 50%) compared with children without disabilities (typically around 65%). There was also a slight overrepresentation of children with disabilities classified as disorganized. Thus, based on this research, a secure attachment causing better parent-child interaction is not plausible. Therefore, an alternative explanation for the results in the current study is possible. In a study of Drotar, Baskiewicz, Irvin, Kennell & Klaus (1975), the importance of early crisis counseling in children with CA is highlighted. Outcomes suggest that early counseling in the first months of life may be particularly

crucial in parental attachment and adjustment. In the WCH, parents of infants born with CA, receive counseling via welfare institutional workers (from the WCH) in the first months of their infants' lives. This counseling of parents may account for causing the better parent-child interactions.

Purpose 2

The second purpose was to examine the predictive value of parent-child interaction on developmental delays. It was found that parent-child interaction predicted developmental delays in participants, which is in line with the finding of Laing et al. (2010): they found a strong relationship between the child's developmental capacity and the mother's interactive behaviors as well. Put differently, the more non-optimal the parent-child relationship was, the more developmental delays were found. Not surprisingly, having a developmental intrusive syndrome was found predictive for developmental delays as well.

Purpose 3

Lastly, because the overall aim of this study was to gain an insight into problems in the major non-cardiac CA children as a group, children suffering from developmental intrusive syndromes were included. Because of the developmentally intrusive character of these syndromes, a distinction in children with syndromic and non-syndromic CA was made. As expected, the non-syndromic children were still having significantly less behavior problems on all three subscales and parent-child interaction remained predictive for developmental delays. However it should be noted that a large decrease in the amount of developmental delays was observed: on all developmental subscales the percentages in non-syndromic infants were approximately half of the percentages of the whole group of non-cardiac CA participants. In other words: however developmental delays on all subscales continued to exist, percentages were notably lower in non-syndromic children compared to all participants (with no distinction in syndrome). As developmental delays on fine and gross motor skills (compared to the other subscales) were found to be high for all participants, higher motor skills delays for non-syndromic children were found as well.

These descriptive statistics imply that the developmental delays in the non-cardiac CA infants as a group were provoked to a large extent by the 5 children with developmental intrusive syndromes. The preliminary results reported here, the low percentages of developmental delays (i.e. all subscales except for motor skills) in non-syndromic CA children, are inconsistent with previous research in which non-syndromic (non)cardiac CA infants were found to have significantly more developmental delays as compared to normative groups (Gischler et al., 2009; Laing et al., 2010). Despite the fact that in previous research the samples were comparable (toddlers suffering from CA),

studies differ on measurements: the standardized BSID test (assessed by professionals) versus questionnaires filled in by parents. As mentioned before, because of the difficult situation children are born in parents answers might be influenced by heuristics as 'downwards comparison' and 'take the best' heuristic.

In addition, the opposing results might be accounted for by the optimal parent-child relationship found in participants in current study. In a sample of preterm birth toddlers, Landry, Smith and Swank (2006) found responsive parenting reduce potentially adverse impact of preterm birth. Increased maternal responsiveness facilitated greater growth in these preterm infants' social, emotional, communication, and cognitive competence, supporting a causal role for responsiveness on infant development.

Strengths, critical notes & further research

Strengths of the present study include use of a clinical group (children born in the WCH, in which specialized healthcare is provided) and a critical attitude towards the dataset (participants who exceeded the ASQ age categories were excluded). Despite the fact that the sample size was low, we were able to find verifiable less behavior problems and better parent-child interaction in participants, and a strong relationship between non-optimal parent-child interaction and developmental delays. Although the present study has identified significant differences in participants compared to the tests' normative groups on behavior problems and parent-child interactions, we were not able to compare developmental delays to a norm referenced group.

Besides the fact that the sample size was low, it is hard to draw hard conclusions out of the results of the heterogeneous group of participants. In current study the relatively broad-based research question (getting a first insight) did not contribute to the easiness of drawing conclusions. Further research is needed to further evaluate and examine developmental outcomes, behavior problems, and parent-child interaction. Firstly, as the different CA are diverse, a large sample will be required to draw hard conclusions for this relatively heterogenic group of children. In addition, including a control group of children suffering from non-cardiac CA but who did *not* undergo surgery, would diminish heterogeneous objections. Besides, longitudinal research would be useful to gain more insight in longer term outcomes, based on which further adapted care can be constructed (e.g. a multidisciplinary follow-up program in the WCH). Lastly, as discussed earlier, questionnaires may be influenced by self-protective and self-enhancing heuristics used by parents, leading to a more positive evaluation of their children. Because of this, measurements assessed by professionals are required.

Conclusion

The purpose of this pilot study was to gain a first insight in behavior problems, developmental delays and parent-child interaction in non-cardiac surgery requiring CA infants as a group. As opposed to previous research, participants were found to have less behavior problems and good parent-child interaction. Possible influence of heuristics of parents on these outcomes were discussed. In addition, developmental delays in percentages and descriptive outcomes were given (outcomes were not tested for significance). Results imply developmental delays on all tested domains, with notably higher percentages of fine and gross motor skills delays compared to the other domains, which is in line with previous research.

Lastly, a distinction was made between all participants and non-syndromic infants specifically. Less behavior problems and better parent-child interaction were found in these non-syndromic children as well and parent-child interaction remained predictive for developmental delays. Despite the fact that comparison on developmental delays was not tested for significance, given percentages of delays showed less developmental delays in non-syndromic children: percentages of delays in these children were found as well (with motor skills found to be higher), however percentages were approximately half of the percentages of delays compared to the whole group of participants. This outcome is inconsistent with previous research, in which non-syndromic (non) cardiac CA infants were found to have significantly more developmental delays as compared to normative groups, and might be caused by the optimal parent-child relationship found in participants in current study.

Concluding, despite the fact that infants who suffer from non-cardiac surgery requiring CA form a complicated and relatively heterogeneous group of participants, current study emphasizes the need of more extended research to specify outcomes.

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