Neonatal brachial plexus palsy : impact throughout the lifespan

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CHAPTER SIX

Neonatal brachial plexus palsy in children aged 0 to 2.5 years; parent-perceived family impact, quality of life, and upper extremity functioning

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ABSTRACT

Objective

To investigate whether parents perceive impact of neonatal brachial plexus palsy on family and quality of life and upper extremity functioning in children less than 2.5 years old.

Methods

This cross-sectional study used the PedsQL Family Impact Module (36 items/one total/four scales/scores 0 to 100), TNO-AZL (Dutch Organisation of Applied Natural Science and Academic Hospital Leiden) Preschool Children's Quality of Life (43 items/12 scales/scores 0 to 100) and 21 upper extremity functioning questions. Associations between neonatal brachial plexus palsy/patient characteristics and family impact, perceived quality of life, and upper extremity functioning were investigated using regression analysis.

Results

Parents of 59 children (median age, 18 months) participated, 49 with C5-C6/C5-C7 lesions. Median Family Impact Module and TNO-AZL Preschool Children's Quality of Life scores were 81.3 to 100.0/100.0 and 78.6 to 100.0/100.0. TNO-AZL Preschool Children Quality of Life scores did not differ significantly to healthy references except for stomach, skin, communication, and motor functioning problems. Parents reported around three upper extremity functioning problems. Greater lesion extent, lower age, still being in follow-up, and right-sided lesions were associated with greater family impact ($P < 0.01$ to $P < 0.1$). No clinically relevant associations were found for perceived quality of life. Greater lesion extent and nerve surgery history were associated with more upper extremity functioning problems ($P < 0.01$). Problems were associated with parental worrying ($P < 0.05$).

Conclusions

Parents perceive having a child with neonatal brachial plexus palsy as impacting on their family depending on the side and severity of the lesion, treatment history, still being in follow-up, and age. They perceive the child's quality of life as relatively normal and not significantly different to healthy peers. However, parents noticed upper extremity functioning problems which increased parental worrying. Healthcare specialists should take these findings into account to better inform or counsel parents in an early stage during treatment.

INTRODUCTION

Neonatal brachial plexus palsy (NBPP) is the result of a stretch injury to the plexus during delivery; its incidence ranges from 1.3 to 2.9/1000.[1,2] Most injuries are mild, but 20% to 30% of the children are left with diminished upper arm function.[2-4] Severe lesions can be treated with nerve surgery at a young age (3 to 9 months). Depending on the clinical course over time, secondary surgery (muscle tendon transfers/osteotomies) may be indicated later on.[5-7] When a child is diagnosed with NBPP, parents face an uncertain future.[8,9] Over time, it will become apparent to which extent recovery can be expected and if nerve surgery will be indicated. Depending on neurological recovery, a better prediction can be made of future arm function. This period is often stressful and worrying for parents and their families. The prognostic uncertainty and consequences for the child's quality of life (QoL) might have impact on families and their QoL.[8-10]

Despite these observations, little research has been done on the impact of NBPP on family and parental QoL in the first years of a child's life. One study found that impact on family was not age dependent.[11] Another study found that having a younger child with NBPP (age 0 to 2 years) had more impact on maternal QoL.[12] Some studies reported impact on the family in terms of finances, personal strain, social and mastery problems, increased risk of psychological problems or distress, and lower maternal QoL.[10-14] Another study found that condition severity was associated with paternal stress and psychological adjustment, both affecting family functioning.[15]

Little is also known about the parent-perceived QoL of young children (less than 2.5 years old) with NBPP. Studies in children with NBPP who are more than two years of age showed that these children have a poorer QoL and limited upper extremity functioning (UEF).[13,16,17] To fully understand the impact of NBPP in young children on the family, it is important to know how parents perceive their child's functioning. However, this has not been studied before. Insight into family impact, QoL, and UEF and possible influential factors is important to be able to provide adequate care, which may help reduce the impact of having a child with NBPP.

Therefore, the goal of our study was to assess the impact of NBPP on family (including parental QoL), perceived QoL, and UEF of young children (less than 2.5 years old). In addition, we explored possible factors associated with family impact, parent-perceived QoL, and UEF and compared the parent-perceived children's QoL with that in the general Dutch population.

PATIENTS AND METHODS

Study design and patients

This study had a cross-sectional design and was part of a larger study on functioning and QoL of patients of all ages with NBPP. That study was conducted between October 2014 and March 2015 at the multidisciplinary, supraregional NBPP care unit of the Leiden University Medical Center and was approved by its medical ethics committee (P14.071). All patients who visited the NBPP care unit, for whom an electronic medical record was available and who were diagnosed with NBPP, were eligible to participate. Patients with concurrent other medical diagnoses that might influence arm functioning (e.g., cerebral palsy, reduction defects) were excluded.

Recruitment

Eligible patients and/or their parents were sent an invitation (including information) to participate. They were asked whether they wanted to participate online or on paper. All participating patients aged greater than 18 years and parents of patients aged less than 18 years provided written informed consent. Questionnaires were sent via regular mail, or patients were invited by e-mail to the online questionnaire. Patients and/or parents who had not responded to the invitation or did not complete the questionnaires received a reminder.

The present analysis only used data on children aged 0 to 2.5 years.

NBPP and patient characteristics

Age, gender, lesion extent (C5-C6/C5-C7/C5-C8/C5-T1/C8-T1), affected side (right/left), and treatment history (conservative/nerve and/or orthopedic surgery) were extracted from the medical records, and current status regarding discharge from follow-up (yes/no) was noted. Parents were asked whether NBPP was present in their families, what kind of household they had (single-parent/two-parent), and whether the child with NBPP was their firstborn (yes/no). Parents were also asked to state whether they had contact with specific health care professionals (apart from the NBPP care unit) or patient organizations and whether their child had been admitted to hospital for NBPP in the past 12 months.

Parent-reported family impact

The 36-item PedsQL Family Impact Module (FIM) measures the impact of a child's chronic condition on their family and yields a Total Scale score, a parental QoL Summary score (Physical/Emotional/Social/ Cognitive Functioning subscales; 20 items), a Family Functioning Summary score (Daily Activities/Family Relationships; eight items), a Worry score (five items), and a Communication score (three items). It uses a Likert-type response scale (0: never to 4: almost always), and scores are transformed to a 0 to 100 scale (0 = 100/ 1 = 75/ 2 = 50/ 3 = 25/ 4 = 0). Scores are computed as the sum of items divided by the number of items answered. Higher FIM scale scores indicate lower impact. If more than 50% of the items in a scale were missing, no score was computed. The FIM was found to be reliable and valid and is available in Dutch.[18]

Quality of life

The TNO-AZL (Dutch Organisation of Applied Natural Science and Academic Hospital Leiden) Preschool children's QoL (TAPQOL) was developed to measure QoL in children aged six months to five years. It is a parent-reported, 43-item generic questionnaire, with 12 scales (three to seven items/scale) covering the domains of physical, social, cognitive, and emotional functioning. Questions relate to the past three months and are scored on a three-point scale (complaint/limitation present: never/occasionally/often). In addition, in seven of the 12 scales (stomach/skin/lung/sleeping/appetite/motor functioning/communication), the child's well-being is also measured in relation to these complaints/ limitations, on a four-point scale (fine/ not so good/quite bad/bad). Scale scores are transformed to a 0 to 100 scale, with higher scores indicating better QoL. No missing values are allowed in three-item scales, one in scales with four items, and two in scales with seven items. The social functioning/motor functioning/ communication scales are only relevant for children aged over 1.5 years.[19]

TAPQOL scores were compared with those of healthy, age-matched references, using a sample from the publicly available reference database. The reference data were derrived from 340 Dutch babies visiting youth health care centers (consultatiebureaus, visited by all Dutch children regularly in the first four years).[20] The sample was selected based on age (six to 30 months) and the absence of health problems, resulting in a reference group consisting of 118 children (median age, 21.0 months; range, ten to 30 months), 45 of whom were male.

Upper extremity functioning

To further understand the QoL issues in NBPP, parent-perceived children's UEF was assessed. No NBPP-specific questionnaires on UEF are available for very young children. Therefore, we developed a set of questions regarding activities (15 items), bodily appearance (three items), and development (three items; Table III). The questions were developed by a group of experts from the NBPP care unit bearing in mind the recommendations for measurement properties of health status questionnaires.[21] The measurement aim is discriminative for the upper extremity physical functioning and evaluative for the bodily appearance and developmental aspects. The questions were pilot tested in the present study. Internal consistency was measured by computing Cronbach's α for the different question parts and were 0.92, 0.81, and 0.91 for activities, bodily appearance, and development, respectively. Because there is no gold standard available, criterion validity could not be determined. Because of the design of the present study, reproducibility and responsiveness were not tested.

Regarding UEF activities, parents could state whether they had observed their child perform certain activities using their affected arm/ hand and if so, whether their child had difficulties with them. Scores were (1) "not observed," (2) "has difficulty," (3) "has no difficulty."

The number of problematic activities was counted and divided into three groups: one to three, four to six, and seven or more problems (i.e., mild, moderately, and severely affected UEF). As regards bodily appearance and development, statements were presented which could be rated as (1) “disagree,” (2) “agree,” (3) “not applicable/no opinion.”

Statistical analysis

Descriptive statistics were used for patient characteristics and all outcome measures according to their distribution (Kolmogorov-Smirnov). TAPQOL scores were compared with those of age-matched, healthy references using analysis of covariance (covariates: age/ gender; significance level, $P < 0.05$).

To determine which factors are associated with family impact, perceived QoL, and UEF, univariate regression analyses were performed for all FIM and TAPQOL scales and for UEF (activities only; significance level, $P < 0.1$). Factors entered independently, one at a time, were lesion extent (C5-C6/C5-C7 and C5-C8/C5-T1/C8-T1), discharged from follow-up (yes/ no), treatment history (nerve surgery/conservative), affected side (right/left), household (single parent/two parent), firstborn (yes/no), responding parent (father/mother), age in years (<1/1 to 2/>2), and UEF activities (1 to 3/ 4 to 6/ >7 problems). Subsequently, a multiple regression analysis was performed with only those factors that were significant in the univariate analyses ($P < 0.1$). All analyses were executed using SPSS 20.0 software (IBM SPSS Sta- tistics for Windows, Armonk, NY: IBM Corp).

RESULTS

From the total cohort of 1142 patients, 104 were eligible for the present study. Parents of 59 patients participated in the present study. Figure 1 shows the flow of these patients, and Table I presents the patient characteristics including healthcare use. Twenty-eight patients (48%) were boys; the median age was 18 months (range, 6 to 30); 26 (44%) had their right side affected and 21 (36%) had been discharged from follow-up. The majority (88%) received physiotherapy.

Table II provides the FIM and TAPQOL outcomes. Median FIM total score was 87.9 (interquartile range [IQR], 74.6 to 96.6), and median FIM scale scores ranged from 81.3 to 100.0 (IQR, 58.3 to 100.0). Figure 2 shows that there is a wide variety in how parents perceive NBPP as impacting on their families. Median TAPQOL scores ranged from 78.6 to 100.0 (IQR, 64.3 to 100.0). About 66% of the TAPQOL scores were not significantly different from the scores of the reference group. However, stomach, skin, motor functioning, and communication scores were lower in the NBPP study population ($P < 0.05$).

Table III provides the outcomes regarding UEF. Parents reported around three problematic activities (IQR, 0.0 to 5.3) and 13 parents report more than seven problems. The most frequently reported problems were “playing with construction materials,” “colouring/

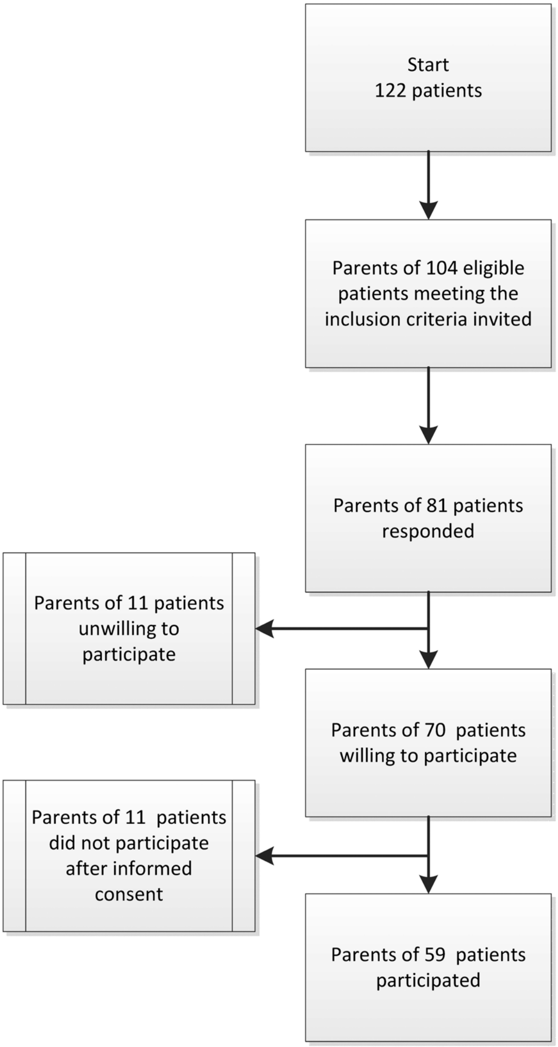


Figure 1 Flowchart of participating parents of patients (0-2.5 years)

painting," "throwing/ rolling a ball," "grasping something located above the head with two hands," "breaking his/her fall," and "drinking from a mug without ears."

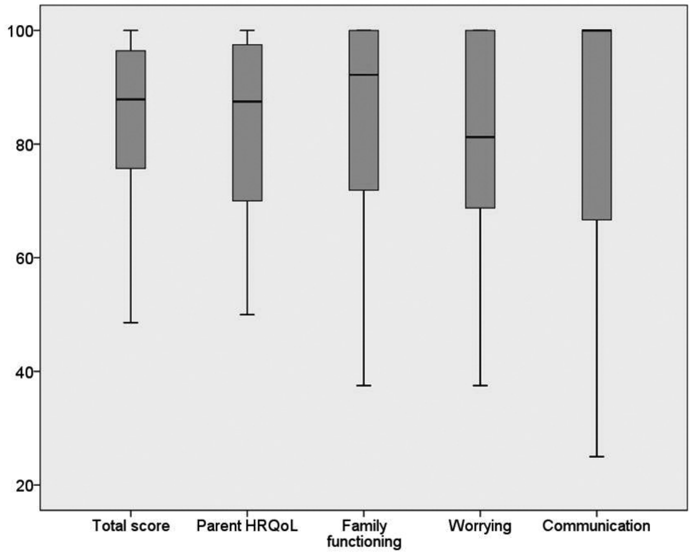
With respect to bodily appearance, 16 parents (27%) thought their child's arm looked different; nine (15%) thought the affected arm was shorter; and seven parents (12%), whose child had undergone nerve surgery, felt bad about the visible scars.

Regarding development, 18 parents (31%) felt their child was not able to do what other children were able to, nine (15%) felt their child developed differently, and nine (15%) thought their child more easily became frustrated trying to perform bimanual tasks.

Table I NBPP Patient characteristics and their healthcare use in the past 12 months

Patients (n=59)		
Gender		
Male (%)	28	(47.5)
Age in months Median (IQR) Range	18 (13/24)	6-30
Affected side		
Right (%)	26	(44.1)
Lesion extent		
Group 1: Upper plexus lesions (%)		
C5-C6	40	(67.8)
C5-C7	9	(15.3)
Group2: Total and lower plexus lesions (%)		
C5-C8	7	(11.9)
C5-T1	2	(3.4)
C8-T1	1	(1.7)
Surgical intervention (%)		
Nerve surgery	23	(39.0)
Conservative treatment	36	(61.0)
Discharged from follow-up		
Yes	21	(35.6)
NBPP in family		
Yes (No. of family members with NBPP, range)	4	(1-3)
Questionnaire completed by		
Father (%)	19	(32.2)
Family situation		
Single parent household (%)	4	(6.8)
Firstborn		
Yes (%)	21	(35.6)
Care received from professionals outside NBPP care unit (%)		
Physical therapy	52	(88.1)
Occupational therapy	4	(6.8)
Contact with professionals within NBPP care unit (%)		
Physical therapist	45	(76.3)
Occupational therapist	30	(50.8)
Neurosurgeon	40	(67.8)
Orthopaedic surgeon	12	(20.3)
Rehabilitation specialist	20	(33.9)
Contact with professionals apart from NBPP care unit (%)		
General practitioner	13	(22.0)
Neurosurgeon	17	(28.8)
Orthopaedic surgeon	6	(10.2)
Rehabilitation specialist	8	(13.6)
Paediatrician	30	(50.8)
Plastic surgeon	1	(1.7)
Psychologist	6	(10.2)
Psychiatrist	1	(1.7)
Social Worker	4	(6.8)
NBPP Patient Organisation	8	(13.6)
Hospital admission		
Yes (%)	15	(25.4)

IQR = interquartile ranges (25th percentile -75th percentile)



Scores 0-100, higher scores indicate lower impact

Figure 2 Boxplots showing Family Impact Measure (FIM) scores of 59 children with NBPP

Univariate regression analyses showed that lesion extent, not having been discharged from follow-up, age less than one year, and affected side (right) were independently associated with lower scores on almost all FIM scales (all P < 0.01 to P < 0.1 [Table IV]). Scores on the Worry and Communication scales showed that nerve surgery treatment was associated with lower scores (P < 0.01). Having more UEF problems was associated with more parental worrying (P < 0.05).

Multiple regression analysis was done for all FIM scales, with only factors entered which were significantly associated with all FIM scales in the univariate analysis as described previously. This showed that the combination of lesion extent, affected side (right), and lower age (less than one year) was associated with worse outcome on all FIM scales (all P < 0.01 to P < 0.1 except age in the Worry and Communication scales).

Hardly any significant associations were found for the TAPQOL scales (Table IV). Affected side (right) was associated with a lower score on the “appetite” scale (P < 0.01). No multiple regression was performed for the TAPQOL scales because no clinically relevant associations were found in the univariate regression analyses.
Lesion extent (C5-C8/C5-T1/C8-T1), not having been discharged from follow-up, and treatment history (nerve surgery) were independently associated with more reported UEF problems (P < 0.01). Multiple regression analysis for UEF showed that lesion extent and nerve surgery history were associated with more reported problems (all P < 0.01).

Table II Family Impact Measure (FIM) and TAPQOL scores of 59 children with NBPP; TAPQOL scores compared with healthy, age-matched references

	NBPP group (n=59)			**Healthy reference (HR) group for TAPQOL (n=118)**		
	Median	**IQR**	**(% ceiling score)**			
FIM scores				-	-	-
Total score	87.9	74.6-96.6	(12.1)			
Parent health-related quality of life	87.5	69.7-97.5	(19.0)			
Family functioning	92.2	71.9-100.0	(41.4)			
Worrying subscale	81.3	67.2-95.3	(24.1)			
Communication subscale	100.0	58.3-100.0	(53.4)			
				Median	**IQR**	**(% ceiling score)**
TAPQOL scales,						
Stomach problems scale	**91.7***	81.3-100.0	(48.1)	100.0	95.8-100.0	(75.2)
Skin problems scale	**83.3***	75.0-100.0	(32.7)	100.0	91.7-100.0	(51.7)
Lung problems scale	100.0	97.9-100.0	(75.9)	100.0	100.0-100.0	(86.4)
Sleeping scale	81.3	56.3-93.8	(20.0)	81.3	75.0-100.0	(25.4)
Appetite scale	100.0	83.3-100.0	(52.7)	91.7	75.0-100.0	(39.8)
Liveliness scale	100.0	100.0-100.0	(96.4)	100.0	100.0-100.0	(79.9)
Positive mood scale	100.0	100.0-100.0	(89.1)	100.0	100.0-100.0	(96.6)
Problem behaviour scale	78.6	64.3-92.9	(16.4)	71.4	64.3-80.4	(2.5)
Anxiety scale	83.3	66.7-100.0	(49.1)	83.3	66.7-100.0	(41.5)
Social functioning scale† (NBPP: n=32, HR: n=83)	100.0	83.3-100.0	(68.8)	100.0	83.3-100.0	(61.4)
Motor functioning scale† (NBPP: n=32, HR: n=83)	**87.5***	81.3-100.0	(31.3)	100. 0	93.8-100.0	(74.7)
Communication scale† (NBPP: n=32, HR: n=79)	**81.3***	75.0-93.8	(19.4)	93.8	81.3-100.0	(40.5)

IQR = Interquartile ranges (25th percentile -75th percentile). For all outcomes 0-100, higher scores indicate lower impact/ better functioning. * $p<0.05$ difference between TAPQOL NBPP group and TAPQOL healthy reference group. † Only for children aged ≥1.5 years.

Table III Upper extremity functioning (UEF) of 59 children with NBPP

UEF activities **My child has difficulty:**	**Not observed**	**Has no difficulty using the affected arm/hand No. (%)**	**Has difficulty using the affected arm/hand No. (%)**
1. Picking up toys (n=59)	0	40 (68%)	19 (32%)
2. Passing toys from one hand to the other (n=59)	0	44 (75%)	15 (25%)
3. Playing with construction materials (Duplo etc.) (n=44)	15	30 (68%)	14 (32%)
4. Colouring/painting (n=29)	30	17 (59%)	12 (41%)
5. Throwing/rolling a ball (n=45)	14	25 (56%)	20 (44%)
6. Carrying big things (big toys etc.) (n=47)	12	35 (74%)	12 (26%)
7. Grasping something above the head with two hands (n=45)	14	29 (64%)	16 (36%)
8. Crawling (n=51)	8	39 (77%)	12 (23%)
9. Raising him/herself to standing position (n=52)	7	39 (75%)	13 (25%)
10. Getting up from the floor (n=47)	12	36 (77%)	11 (23%)
11. Climbing during play (n=46)	13	33 (72%)	13 (28%)
12. Climbing onto a chair or couch (n=42)	17	32 (76%)	10 (24%)
13. Breaking his/her fall (n=47)	12	27 (52%)	20 (38%)
14. Drinking from a mug without ears (n=40)	19	24 (60%)	16 (43%)
15. Putting something to eat in his/her mouth (cake, bread etc) (n=51)	8	37 (73%)	14 (27%)

No. of reported problems of Upper Extremity Functioning activities	
Median (IQR)	3.0 (0.0-5.3)
1-3 problems (no.)	11
4-6 problems (no.)	14
>7 problems (no.)	13

UEF cosmetics and development	**Agree No. (%)**	**Disagree No. (%)**	**Not applicable/ No opinion No. (%)**
Cosmetics:			
My child's arm looks different	16 (27%)	24 (41%)	19 (32%)
My child's arm is shorter than his/her other arm	9 (15%)	22 (37%)	28 (48%)
I feel bad about scars of the operation being visible	7 (12%)	28 (48%)	24 (40%)
Development:			
My child is not able to do what other children are able to	18 (31%)	19 (32%)	22 (37%)
My child does not develop the same as other children	9 (15%)	31 (53%)	19 (32%)
My child is more easily frustrated than other children when trying to perform bimanual tasks	9 (15%)	26 (45%)	24 (40%)

Table IV Regression analyses for all PedsQL™ FIM and TAPQOL scales, and UEF activities for 59 children with NBPP

β -Estimates (95% CI)	**Lesion extent C5/C6-C5/C7 († C5-C8/C5-T1/C8-T1)**	**Discharged from follow-up: Yes († No)**	**Conservative treatment († nerve surgery)**	**Affected side Right († Left)**	**Firstborn: Yes († No)**	**Age <1 year († age >2)**	**Age <1 year († age 1-2)**	**Responding parent: Father († Mother)**	**Two- parent household († single parent)**	**UEF activities >7 problems († 1-3 problems)**	**UEF activities 4-6 problems († 1-3 problems)**
FIM:											
Total score	**15.3**** **(3.1, 27.5)**	**12.8***** **(3.3, 22.3)**	**9.1*** **(-0.6, 18.8)**	**-15.1***** **(-24.0, -6.1)**	1.1 (-9.1, 11.2)	**-13.3*** **(-28.8, 2.2)**	-2.2 (-13.7, 9.3)	2.9 (-7.6, 13.4)	7.8 (-14.0, 29.7)	-9.7 (-25.9, 6.5)	-9.6 (-25.2, 6.1)
Parents' health-related quality of life	**14.1**** **(0.9, 27.2)**	**12.3**** **(2.1, 22.6)**	8.2 (-2.1, 18.7)	**-17.3***** **(-26.6, -8.0)**	1.4 (-9.3, 12.2)	**-15.5*** **(-31.9, 0.9)**	-2.6 (-14.8, 9.5)	5.16 (-5.9, 16.2)	9.2 (-14.0, 32.4)	-10.5 (-27.4, 6.5)	-9.4 (-25.7, 7.0)
Family functioning	14.8** (0.8, 28.8)	**11.7**** **(0.6, 22.7)**	6.1 (-5.1, 17.3)	**-9.0*** **(-19.8, 1.8)**	-0.7 (-12.2, 10.7)	**-15.9*** **(-33.4, 1.7)**	-3.4 (-16.4, 9.5)	1.4 (-10.4, 13.4)	4.2 (-20.6, 29.1)	-6.6 (-25.6, 12.5)	-9.7 (-28.1, 8.7)
Worrying	**20.0***** **(7.3, 32.8)**	**15.1***** **(5.0, 25.2)**	**13.8***** **(3.7, 23.9)**	**-14.1***** **(-23.9, -4.3)**	3.5 (-7.4, 14.3)	2.1 (-15.0, 19.1)	5.2 (-7.5, 17.8)	-4.0 (-15.2, 7.3)	10.3 (-13.2, 33.7)	**-17.1**** **(-32.3, -2.0)**	-11.5 (-26.1, 3.1)
Communication	**18.3**** **(1.1, 35.6)**	**16.2**** **(2.8, 29.6)**	**16.3**** **(3.0, 29.5)**	**-17.51***** **(-30.3, -4.7)**	-0.2 (14.1, 14.1)	-12.6 (-34.5, 9.4)	-6.0 (-22.2, 10.3)	1.2 (-13.4, 15.9)	5.5 (-25.1, 36.0)	-3.0 (-26.8, 20.9)	-7.6 (-30.6, 15.4)
TAPQOL:											
Stomach problems	3.6 (-8.6, 15.8)	-6.5 (-16.4, 3.5)	-4.3 (-14.0, 5.4)	-6.9 (-16.4, 2.7)	-8.0 (-17.6, 1.7)	**-13.6*** **(-28.2, 1.1)**	-6.5 (-17.5, 4.5)	4.6 (-5.4, 14.7)	-11.6 (-32.2, 9.0)	-1.1 (-14.0, 11.7)	-2.1 (-14.7, 10.5)
Skin problems	0.3 (-11.3, 11.9)	3.2 (-6.2, 12.7)	-0.1 (-9.2, 9.0)	-4.3 (-13.3, 4.7)	-1.7 (-10.9, 7.6)	3.9 (-10.2, 18.0)	5.8 (-4.8, 16.4)	-2.3 (-11.8, 7.2)	-4.8 (-24.4, 14.9)	-5.2 (-19.4, 8.9)	-5.1 (-18.8, 8.5)
Lung problems	-2.2 (-11.9, 7.5)	-4.4 (-12.3, 3.6)	-4.6 (-12.3, 3.1)	-2.6 (-10.3, 5.1)	4.2 (-3.6, 11.9)	0.4 (-11.6, 12.4)	-1.6 (-10.6, 7.5)	0.5 (-7.6, 8.5)	-7.2 (-23.6, 9.2)	5.0 (-6.3, 16.3)	4.7 (-6.3, 15.8)
Sleeping	-6.4 (-22.6, 9.8)	9.4 (-3.7, 22.5)	9.9 (-2.7, 22.4)	**-14.7*** **(-26.8, -2.7)**	-7.5 (-20.4, 5.4)	8.6 (-11.3, 28.5)	5.0 (-9.9, 20.0)	7.4 (-5.9, 20.6)	-6.0 (-33.5, 21.6)	-2.9 (-22.9, 17.1)	-5.6 (-24.9, 13.6)
Appetite	6.5 (-3.8, 16.7)	5.5 (-2.9, 13.9)	2.2 (-6.0, 10.3)	**-11.4***** **(-18.9, -3.9)**	-2.4 (-10.7, 5.9)	-5.5 (-18.2, 7.2)	-3.9 (-13.4, 5.7)	4.1 (-4.4, 12.6)	-7.3 (-24.9, 10.2)	-9.3 (-23.2, 4.5)	-6.8 (-20.1, 6.6)
Liveliness	-1.1 (-4.6, 2.4)	1.4 (-1.5, 4.2)	-1.5 (-4.3, 1.2)	0.3 (-2.4, 3.1)	1.4 (-1.4, 4.2)	-2.5 (-6.7, 1.7)	1.2 (-2.0, 4.3)	-1.4 (-4.3, 1.5)	-1.0 (-7.0, 5.0)	0.0 (-4.7, 4.7)	-2.4 (-6.9, 2.1)

Positive mood	**6.5**** **(0.3, 12.7)**	**4.5*** **(-0.7, 9.7)**	0.0 (-5.1, 5.1)	**-4.8*** **(-9.6, 0.1)**	3.5 (-1.6, 8.5)	-1.5 (-9.0, 6.0)	**5.4*** **(-0.2, 11.0)**	0.4 (-4.9, 5.7)	8.6 (-2.2, 19.2)	-5.6 (-13.8, 2.7)	-4.8 (-12.7, 3.2)
Problem behaviour	0.3 (-14.3, 14.9)	3.9 (-8.1, 15.8)	6.5 (-4.9, 17.8)	-7.2 (-18.5, 4.0)	-5.0 (-16.6, 6.7)	13.3 (-4.3, 0.9)	2.3 (-10.9, 15.5)	2.7 (-9.3, 14.7)	-12.8 (-37.4, 11.7)	-10.7 (-27.5, 6.1)	-6.6 (-22.9, 9.6)
Anxiety	-6.5 (-21.2, 8.2)	6.0 (-6.1, 18.0)	3.3 (-8.3, 14.9)	-4.5 (-16.0, 7.0)	1.8 (-10.0, 13.6)	5.3 (-12.6, 23.2)	10.3 (-3.1, 23.7)	3.2 (-8.9, 15.3)	11.4 (-13.5, 36.4)	1.3 (-17.4, 19.9)	-14.6 (-32.6, 3.4)
Social functioning	-7.0 (-19.1, 5.2)	5.7 (-5.5, 17.0)	-3.1 (-13.4, 7.1)	-5.3 (-15.7, 5.2)	-5.3 (-15.7, 5.2)	x	x	6.5 (-4.3, 17.4)	-8.1 (-37.5, 21.4)	-8.3 (-24.7, 8.1)	-9.3 (-25.9, 7.4)
Motor functioning	2.1 (-12.1, 16.4)	8.6 (-3.7, 21.0)	6.8 (-4.8, 18.3)	2.3 (-10.2, 14.6)	**-9.7*** **(-21.3, 2.0)**	x	x	0.5 (-11.9, 12.9)	-15.8 (-49.2, 17.6)	-13.6 (-33.0, 5.7)	-8.8 (-28.5, 10.8)
Communication	-13.0 (-29.0, 3.1)	8.47 (-5.3, 22.3)	**-13.8**** **(-26.1, -1.6)**	6.63 (-7.3, 20.6)	-5.6 (-19.3, 8.1)	x	x	2.3 (-11.5, 16.1)	-7.2 (-44.6, 30.2)	4.2 (-16.1, 24.6)	-5.7 (-26.1, 14.6)
UEF:											
Activities	**-4.8***** **(-7.2, -2.7)**	**-3.7***** **(-5.5, -1.8)**	**-5.1***** **(-6.7, -3.5)**	0.5 (-1.6, 2.5)	-0.3 (-2.5, 1.8)	**-3.1**** **(-6.3, 0.1)**	-1.6 (-3.9, 0.7)	**1.9*** **(-0.2, 4.0)**	-2.7 (-6.7, 1.2)	xx	xx

* p<0.1, ** p<0.05, *** p<0.01. † = set as β –Estimate = 0 in the univariate regression analysis. FIM=Family Impact Module, TAPQOL= TNO-AZL Preschool children's Quality Of Life, UEF= reported problem of Upper Extremity Functioning. x= no analysis possible due to scale age limitations (no data for children <1 year), xx= no analysis possible.

DISCUSSION

This cross-sectional study on the parent-perceived family impact, QoL, and UEF of 59 children with NBPP aged 0 to 2.5 years showed that lower FIM scores were associated with younger age, lesion extent, affected side, nerve surgery treatment history, and currently being in follow-up. The parents' perception of the children's QoL was not significantly different to that of healthy references for 66% of the TAPQOL scales. Having more UEF problems was associated with lesion extent and nerve surgery treatment history. These problems were associated with more parental worrying.

Our findings regarding the family impact of having a child with NBPP are generally in line with previous studies. Most studies reported a certain degree of family impact, maternal or paternal stress, and an increased risk of psychological problems. The severity of NBPP also influenced family impact.[10-15] Parental QoL scores in the present study indicate that having a child with NBPP influences some parents' lives, which is in line with previous findings.[9,11-13,15] We found that when the right side was affected, FIM scores tended to be lower (Table IV). This might be related to the 90% right-handedness of the general population.[22] Parents may be more worried about their child not being able to fully use their right arm.

A younger age (less than one year) had a significant negative impact on the family in our study (Table IV), unlike what was found in another study.[9] In that study, however, the median age was twice as high. We also found that a lower impact on parental QoL and family functioning was reported for the older children in our study, which might be related to improving prognosis in the still growing child. When parents reported more problems on UEF, they also tended to worry more, indicating that a higher degree of functional impairment has a greater impact on the parents.

Still being in follow-up is likely to imply that the child has not fully recovered and/or is in need of additional treatment in the future which may have impact on family. Six parents (10%) reported that they had sought psychological counselling related to their child having NBPP. In a multidisciplinary NBPP unit, psychological care would probably provide added value.

The FIM has not previously been used in patients with NBPP. Parents with NBPP children scored a median of 81.3 to 100.0 points on all FIM scales, which was also found in studies among parents of children with acquired brain injury and nephrotic syndrome.[23,24] Parents of children with chronic pain had lower scores, they scored 47 to 74 points on all scales.[25] Parents in our study had better FIM scores (up to 20 points higher) compared to U.S. parents of children with a chronic condition.[26] This is most probably related to the easily accessible and well-organized healthcare system in The Netherlands, giving parents confidence that their child with NBPP is taken care of. Furthermore, there is a wide variety in FIM scores as can be seen in Table II and Figure 2. In our study, mildly and severely affected children participated which may be the reason for this variety and relative high median scores.

Nevertheless, QoL and family life scores of most parents of NBBP children are to some extent affected.

To investigate children's parent-perceived QoL, the TAPQOL has not been used before in NBPP studies. QoL outcomes in the present study are in line with the available literature on older children with respect to motor functioning.[13,16,17] TAPQOL scales refer to common problems in young children, and there were few differences in perceived QoL between our NBPP population and the healthy references (Table II). The question remains whether the TAPQOL is suitable for the young NBPP population. QoL in young children is highly dependent on care provided by the parents. Because all parents wish their child to have a good life, the perceived QoL might be biased as parents are the proxy for their own children. However, if this is true in the present study, underestimation of issues reducing QoL is more likely than overestimation.

No NBPP-specific questionnaires were available to evaluate UEF in very young children, prompting us to develop a study-specific set of questions (Table III). There are developmental tools available, but these are performance tests, not available as questionnaires, and thus were not suitable for the present study. Preliminary psychometric property analyses of the UEF-questionnaire were promising. We found that greater lesion extent and a history of nerve surgery were associated with more UEF problems. This could mean that our preliminary set of questions is disease- specific and underlines the need to further develop this NBPP-specific UEF-questionnaire for young children. In this endeavor, however, cross-cultural differences should be addressed to ensure usefulness of the questionnaire across different countries. For example, with construction materials and food, performance can vary across cultures and climbing onto a couch is dependent on its height.

This study had a number of limitations. First, a relatively small sample size was used. However, in the past two years, only 104 newborns with NBPP were seen in the NBPP care unit, 59 (57%) of whom participated. Second, patients seen at our NBPP care unit were referred to us because of a severe lesion, which might lead to confounding by indication. Third, no control group was included to compare outcomes. For family impact, only U.S. population FIM data were available, which are not comparable to our Dutch data because of differences in the health care system and society. For QoL, age-matched reference values were available, partly counteracting this limitation.[19] Fourth, this study had a cross-sectional design with no follow-up, using only self- reported questionnaires. This fact might lead to overestimation or underestimation of results as people might be influenced by unknown factors at the time of completing the questionnaires (e.g., bad mood, work-related stress, etc.). Future studies monitoring parent-perceived family impact, QoL, and UEF over time should enable further optimization of health care for children with NBPP and their parents. Individual and/or group meetings providing detailed information about NBPP, prognosis, treatment strategies, and the possibility to meet fellow parents might provide added value to reduce the impact of having a child with NBPP.

CONCLUSION

When a newborn child is diagnosed with NBPP, this may have effect on the parents and their families. Our findings confirm that parents find to some extent that having a child with NBPP has impact on their family. Although lower age (less than one year) and more severe lesions have been previously reported as being associated with more impact on the family, the present study in infants and very young children showed that right-sided lesions and having more UEF problems were also related to a greater impact on the family. No study in very young children has reported this before, even though this is an important part in the development of young NBPP children. It is essential for healthcare specialists to be aware of these findings, so they can actively provide suitable information and counselling to parents in an early stage to help reduce the possible impact on family.

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