Neonatal brachial plexus palsy : impact throughout the lifespan

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CHAPTER SEVEN

Healthcare use and information needs in children with neonatal brachial plexus palsy: a cross-sectional survey among 465 patients

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ABSTRACT

Objective

To investigate healthcare use and information needs of children aged 0-18 years with neonatal brachial plexus palsy (NBPP).

Patients and methods

For this cross-sectional study, all patients and/or their parents seen in our multidisciplinary NBPP clinic over the last 18 years were invited to complete a survey. The survey comprised questions on healthcare use due to NBPP in the past year (contact with the expert team and/or 11 other types of healthcare professionals) and on their current information needs (12 NBPP-related topics). Outcomes were described for 3 age groups (0-1, 2-9 and 10-18 years), and based on follow-up status (early/late/no discharge).

Results

465 parents/patients participated (59, 226 and 180 patients in the 0-1, 2-9 and 10-18 age groups, respectively). 293 (63%) had C5-C6 lesions, 193 (42%) had been discharged from follow-up, 83 of whom were categorized as ‘early discharge’ (defined as <1 year of age) due to spontaneous lesion recovery (19/59, 50/226 and 14/180).

Over the past year, 198 patients had had contact with the expert team (49/59, 81/226 and 68/180) and 288 with at least 1 other healthcare professional (53/59, 133/226 and 102/180). Of the 83 patients discharged early, 34 reported healthcare use. 228 participants (49%) reported current information needs regarding at least one topic and 23 of these patients were discharged early.

Conclusions

Healthcare use and information needs of children due to NBPP remain considerable even in children who were early and late discharged. Stricter longitudinal follow-up and information provision for all patients with NBPP throughout life is needed.

INTRODUCTION

Neonatal brachial plexus palsy (NBPP) occurs in about 1-3 children per 1000 births in western countries.[1,2] Seventy to ninety percent of these children recover spontaneously, while the remaining 10-30% are left with neurological damage, possibly resulting in functional impairments.[2-5] Children with severe NBPP are usually referred to a tertiary NBPP expert center for further diagnostics and follow-up.[1,6,7] If these children do not show sufficient spontaneous recovery around the age of 3 months, primary, nerve, surgery may be indicated.[8,9] Children with persistent functional limitations can be treated with secondary surgery (e.g. osteotomies, tendon transfers) to improve the functionality of the affected arm/hand and prevent bone and joint deformities.[10-12]

In the Netherlands, most children with NBPP are referred to a specialized NBPP clinic (e.g. the Leiden Nerve Center located at Leiden University Medical Center) by their family doctor, or a pediatrician or pediatric neurologist at a local hospital.[6,13] The Leiden Nerve Center has successfully promoted early referral, i.e. at the age of one month.[13] Infants are assessed and treated by a multidisciplinary expert team involving a variety of medical and allied health care professionals using an interdisciplinary approach.[1,6,8,11,12,14,15] In addition, most children are treated by healthcare professionals in primary care in their place of residence (e.g. allied healthcare or psycho-social) and, if insufficient, interdisciplinary rehabilitation care is provided.[16,17] A considerable proportion of patients are discharged from clinical follow-up at the Leiden Nerve Center, either in their first year of life because of spontaneous recovery without indications for any interventions, or later on in their care trajectory if good functional recovery takes place after conservative or surgical treatment.[8,14,18-22]

Discharge from follow-up necessitates a low threshold for renewed consultation but also satisfactory information for both parents and patients. The need for, and specific content of, this information may change over time and differs for each age group.

At present, there is virtually no literature on the healthcare use of children with NBPP. Furthermore, factors influencing healthcare use by patients with NBPP are largely unknown. No literature is available on the information needs of the NBPP population (whether in clinical follow-up or not), even though decision making regarding NBPP is influenced by the information that is sought or provided.[23] To date, it remains unclear whether patients and/or their parents/caregivers, whether in clinical follow-up or not, have unmet information needs. In order to improve medical decision making, it is important to understand with how many and which healthcare professionals children and their parents have contact throughout their lives due to NBPP. In what way is healthcare use determined by patient characteristics, quality of life (QoL) and physical functioning parameters. Furthermore, what information do patients and/or their parents/caregivers need in order to feel provided with the right information throughout the NBPP treatment phase.

The aim of the present study was therefore to quantify the healthcare use of children with NBPP due to their condition, defined in the present study as the number of professionals involved in their care, and to specify the information needs of patients and/or their parents/caregivers at different ages and in various follow-up categories.

PATIENTS AND METHODS

Study design and patients

This study had a cross-sectional design and was part of a larger study on the functioning and quality of life of patients with NBPP. It was conducted between October 2014 and March 2015 at the Leiden Nerve Center, and was approved by the university's medical ethics committee (P14.071). All patients who visited the Leiden Nerve Center and were diagnosed with NBPP, and for whom an electronic medical record was available, 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 >18 years and parents of patients <18 years of age provided written informed consent. Questionnaires were sent via regular mail, or patients were invited by e-mail to complete the online questionnaire. Patients and/or parents who had not responded to the invitation, or did not complete the questionnaires, received a reminder.
A total of 1142 patients were invited to participate in the overall study of whom 508 patients and/or their parents participated. The present study used the data of 465 patients from this sample who were 18 years or younger. The flow of these patients is presented in Figure 1.

NBPP and Patient characteristics

Medical records were used to extract information on age, gender, lesion-extent (1; upper plexus lesions: C5/C5-C6/C5-C7/C7 and 2; total and lower plexus lesions: C5-C8/C5-T1/C8-T1), affected side (right/left/both) and treatment history (1; conservative, 2; primary, nerve, surgery, 3; secondary, orthopaedic, surgery, 4; primary and secondary surgery).
Three age groups were distinguished, whose outcomes were described separately: 0-1 (0-1 years old), 2-9 (2-9 years old) and 10-18 (10-18 years old).

Follow-up status

The follow-up status of all patients of the Leiden Nerve Center was extracted from the medical records. Based on this, 3 subgroups were defined: (1) Early discharge, i.e. discharged from follow-up within a year after birth; (2) Late discharge, i.e. discharged from follow-up at a later age; and (3) No discharge, i.e. still in follow-up at the Leiden Nerve Center.
For patients in the early discharge subgroup, the reason for discharge had to be full or satisfactory spontaneous recovery, not needing further treatment. This was verified by checking the medical records for the reason for discharge.

Healthcare use (HCU)

The proxy for healthcare use by children with NBPP in this study was defined as the number of healthcare professionals involved in the care for NBPP, within or outside the

Leiden Nerve Center. HCU due to NBPP was measured by asking parents and/or patients whether they had been in contact with specific healthcare professionals, due to the NBPP of their child, since birth (HCU-ever) and whether this contact had taken place in the past 12 months (HCU-12) due to the consequences of NBPP. They were also asked whether they had ever been admitted to hospital for NBPP and whether this had happened in the past 12 months.
One point was allocated when there had been contact with at least 1 of the 5 members of the NBPP expert team (i.e. neurosurgeon, orthopedic surgeon, rehabilitation specialist (physiatrist), physical therapist, occupational therapist). Furthermore, 1 point was allocated for each of the 11 types of healthcare professionals contacted outside the expert team. In addition, 1 point was allocated when the patient had been admitted to hospital. Total HCU scores (range 0-13) since birth (HCU-ever) and with respect to the past 12 months (HCU-12) were calculated.

In addition, the questionnaire asked about any use of complementary medicine (e.g. homeopathy, alternative healers) and contact with the patient organization (Erbse Parese Vereniging Nederland; EPVN, a nationwide patient organization for children and adults with NBPP), since birth and/or in the past 12 months.

Quality of Life (QoL) and physical functioning
Perceived QoL was examined using the TNO-AZL (Netherlands Organization for Applied Scientific Research and Leiden University Hospital) Preschool children's QoL (TAPQOL) and the Pediatric Outcome Data Collecting Instrument (PODCI).

The TAPQOL was developed to measure QoL in children aged 6 months to 5 years. It is a parent-reported, 43-item generic questionnaire, with 12 scales (3-7 items/scale). Questions relate to the past three months and scale scores are transformed to a 0–100 scale, with higher scores indicating better QoL.[24] For the present study, only the TAPQOL scales for Positive mood, Problem behavior, Anxiety, Social functioning and Motor functioning were used for children <2 years of age, since only these scales were found to provide some insight into the QoL of young children with NBPP.[25]

The PODCI was designed to assess different aspects of daily living, including upper extremity functioning, in children with musculoskeletal disorders (including NBPP) and is available in Dutch.[26-28] The instrument consists of 5 subscales and one total score. PODCI scale scores range from 0-100, with higher scores indicating better functioning/QoL. The present study used the 2-10 years and 11-18 years parent-reported versions.

Information need
To determine whether respondents felt a need for information, the first question asked was whether respondents had ever searched for information about NBPP, and if so, whether they had found the information they were looking for. Secondly, we asked if they currently

felt the need for more information (yes/no) regarding: NBPP in general, physical consequences of NBPP, medical treatment of NBPP, assistive devices and government social support, physical activity and sports, pediatric or general physical therapy, occupational therapy, primary surgery, secondary surgery, rehabilitation medicine, social work and patient organizations/peer contact. Thirdly, we asked what the preferred mode of information delivery would be and whether they would use the opportunity to e-mail with a specialized NBPP consultant regarding possible questions and information needs.

We were also interested to find out whether parents or patients had ever received contradictory information from different healthcare professionals (yes/no), to check whether there is a need to further promote uniformity of information provision regarding NBPP.

Statistical analysis

Descriptive statistics (medians with interquartile ranges [IQR] or means with standard deviations [SD]) were used for patient characteristics and all outcome measures. All outcomes are reported separately for all age groups, based on follow-up status.

TAPQOL scores for all follow-up subgroups were compared using an unpaired t-test, and PODCI scores were compared using a one-way analysis of variance with Fischer's Least Significant Difference post-hoc test (significance level, $p<0.05$).

To determine which factors were associated with HCU-12, univariate regression analyses were performed for all age groups (significance level, $p<0.1$). Factors entered independently, one at a time, were: gender (male/female), age, affected side (right/left/both), lesion-extent (1/2), treatment history (1/2/3/4), follow-up status (1/2/3), TAPQOL motor functioning (only for 0-1 year age group) and PODCI Upper Extremity (UE) and Global Functioning (GF) scales (only for 2-9 and 10-18 year age groups). Subsequently, a multiple regression analysis was performed with only those factors that had a significance level of $p<0.2$ in the univariate analyses. Differences in healthcare use based on the factors entered in the univariate and multiple regression analyses are presented as β-estimates with 95% confidence intervals.

RESULTS

Of the 465 included patients, 59 belonged to the 0-1 year age group (median age 1 year), 226 to the 2-9 years group (median age 6 years) and 180 to the 10-18 years group (median age 14 years). The flow of patients is presented in Figure 1. A total of 83 patients belonged to the early discharge subgroup: 19 from the 0-1 age group, 50 from the 2-9 group and 14 from the 10-18 group. All patient characteristics are shown in Table I.

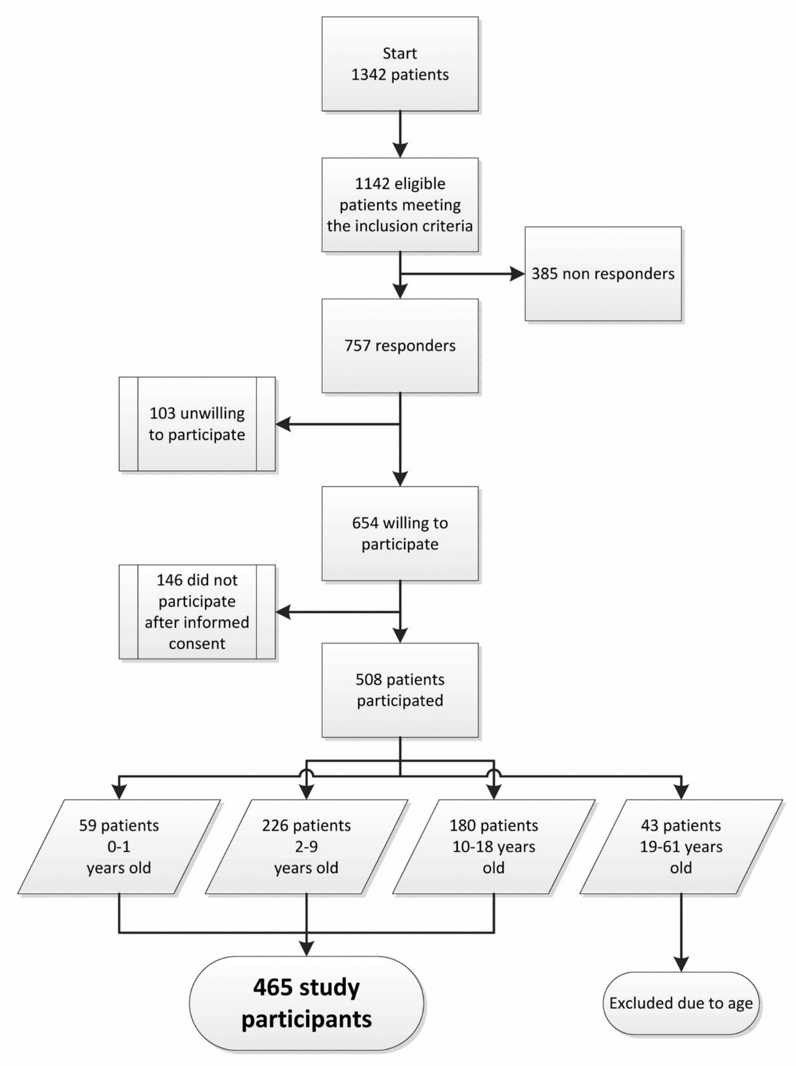

Figure 1. Flowchart showing the formation of the study sample (n=465)

Table I also shows QoL and physical functioning scores (TAPQOL and PODCI). In the 0-1 year age group there was no difference in QoL between the early discharge subgroup and the subgroup still in follow-up. In the 2-9 years age group, however, the subgroup still in follow-up had significantly lower scores on all PODCI scales than the early and late discharge subgroups. Moreover, the subgroup still in follow-up reported significantly lower scores for pain and comfort than the late discharge subgroup. In the 10-18 years age group, statistically significant differences between the subgroup still in follow-up and the two other subgroups were only found for the UE and GF scales. The early discharge subgroup reported problems of upper extremity functioning as well as with sports and physical functioning, resulting in lower QoL scores (GF scale).

Table II presents the healthcare professionals involved in the care of children with NBPP, and these children's median healthcare use (HCU-ever/HCU-12) for all age groups and follow-up subgroups. Since birth, all patients had had contacts with at least 1 (range 1-11) healthcare professional in addition to the NBPP expert team. Hospital admissions due to NBPP were reported by 278 patients (60%) since birth. The most frequently mentioned healthcare professionals contacted since birth were: pediatric or general physical therapist, neurosurgeon, rehabilitation specialist (physiatrist), orthopedic surgeon and pediatrician, but other professionals were mentioned as well, including psychologists (n=39) and psychiatrists (n=21). In the past 12 months 198 patients had had contact with the expert team (divided over the 3 age groups as follows: 49 (83%), 81 (36%) and 68 (38%), respectively). At least 1 (additional) healthcare professional (range 1-7) had been contacted by 288 patients (divided over the 3 age-groups: 53 (90%), 133 (59%) and 102 (57%)). The physical therapist was again the most frequently mentioned healthcare professional contacted.
In the early discharge subgroup, 34 patients (41%) had contacted at least 1 healthcare professional during the past 12 months for their NBPP. In this subgroup, physical therapists were mentioned 23 times.

The outcomes of the regression analyses are presented in Table III. Factors independently associated with healthcare use were lesion-extent, treatment history, follow-up status and QoL and physical functioning (all $p<0.05$). Male gender was associated with higher healthcare use in the 2-9 years age group.
Multiple regression analysis showed that for the 2-9 years age group, greater extent of the lesion, treatment history (primary and secondary surgery), being in follow-up and lower QoL (lower PODCI GF scale-scores) were associated with higher healthcare use. For the 10-18 years age group, only greater extent of the lesion and QoL (lower PODCI GF scale scores) were associated with higher healthcare use (all $p<0.001$-$p<0.05$).

All age groups and all follow-up subgroups reported information needs (Table IV). Sixty-eight percent of the respondents had ever sought information regarding NBPP, but only 49% had found what they were looking for. Furthermore, 18% of the respondents had received/found contradicting information regarding NBPP. A need for information regarding a variety of

NBPP-related topics was reported by 228 patients/parents (49%). In the early and late discharge subgroups, information need was reported by 23/83 patients (28%) and by 42/110 patients (40%), respectively. Information on consequences of NBPP, physical activities/sports and assistive devices and government social support were the most commonly reported topics. The most frequently mentioned preferred modes of information delivery were: internet, the treating physician and the pediatric or general physical therapist.

DISCUSSION

This cross-sectional study in a large sample showed that healthcare use (HCU) since birth by children due to NBPP in the Netherlands is considerable, with up to 11 healthcare professionals involved in care in addition to the expert team, and with possible hospital admissions. However, healthcare use did decrease over time: respondents reported that over the past 12 months, up to 7 healthcare professionals had been involved in addition to the possible involvement of the expert team and hospital admissions. HCU was associated with the children's follow-up status at the tertiary Leiden Nerve Center, as well as with lesion-extent, treatment history, quality of life and physical functioning. A large proportion of patients (42%) discharged from follow-up by the Leiden Nerve Center still had contact with regional healthcare professionals for their NBPP. They included a relatively large proportion of patients (34/83, 41%) discharged at a young age due to supposedly satisfactory spontaneous recovery; this indicates that, against the expectation of the Leiden Nerve Center team, these patients may still perceive functional limitations due to their NBPP. Furthermore, a large proportion (228/465, 49%), including children discharged from follow-up (either early discharge: 23/83, 28%, or late discharge: 42/110, 40%), reported information needs regarding a variety of NBPP-related topics (treatment, sports and physical functioning, assistive devices etc.).

Healthcare use

No study of the healthcare use by patients with NBPP has been performed before, so no comparisons with other countries or centers can be made. Although studies of healthcare use in pediatric populations have been performed, they mainly focused on hospitalization and/or healthcare costs.[29-32] One study among children with various musculoskeletal disorders (e.g. bone, spine, and soft tissue conditions) showed that on average these children had had 1.7 contacts/visits with healthcare professionals in the past 12 months.[33] In contrast, our study found up to a median of 6.0 contacts (range for medians 0-9 depending on age and follow-up status, Table II). However, we only counted the number of healthcare professionals contacted, but not the number of visits. In addition, we took contacts with other healthcare professionals besides the Leiden Nerve Center team into account. Our study showed that allied health professionals, especially (pediatric) physical therapists, were frequently contacted.

Table I Characteristics of 465 patients with neonatal brachial plexus palsy and their quality of life/physical functioning in relation to current follow-up status at the Leiden Nerve Center

	0-1 years (n=59)		**2-9 years (n=226)**			**10-18 years (n=180)**		
	Early discharge* (n=19)	**No discharge*** (n=40)**	**Early discharge* (n=50)**	**Late discharge** (n=44)**	**No discharge*** (n=132)**	**Early discharge* (n=14)**	**Late discharge** (n=66)**	**No discharge*** (n=100)**
Gender								
Male	12 (63%)	16 (40%)	28 (56%)	16 (36%)	70 (53%)	9 (64%)	26 (39%)	43 (43%)
Median age (Range)	1 (0-1)	1 (0-1)	6 (2-9)	6 (2-9)	6 (2-9)	14 (10-18)	15 (10-18)	13 (10-18)
Affected side:								
Right	7 (37%)	19 (48%)	20 (40%)	23 (52%)	61 (46%)	8 (57%)	32 (48%)	53 (53%)
Both	0 (0%)	0 (0%)	1 (2%)	0 (0%)	3 (2%)	1 (7%)	2 (3%)	3 (3%)
Lesion extent:								
Group 1: Upper plexus lesions								
C5	0 (0%)	0 (0%)	1 (2%)	0 (0%)	1 (1%)	1 (7%)	1 (2%)	2 (2%)
C5-C6	16 (84%)	24 (60%)	45 (90%)	39 (89%)	67 (51%)	10 (71%)	34 (51%)	46 (46%)
C5-C7	3 (16%)	6 (15%)	3 (6%)	5 (11%)	33 (24%)	2 (15%)	26 (39%)	30 (30%)
C7	0 (0%)	6 (15%)	0 (0%)	0 (0%)	0 (0%)	1 (7%)	0 (0%)	0 (0%)
Group 2: Total and lower plexus lesions								
C5-C8	0 (0%)	3 (8%)	0 (0%)	0 (0%)	14 (11%)	0 (0%)	0 (0%)	7 (7%)
C5-T1	0 (0%)	1 (2%)	0 (0%)	0 (0%)	17 (13%)	0 (0%)	5 (8%)	15 (15%)
C8-T1	0 (0%)	0 (0%)	1 (2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Surgical intervention								
Primary, nerve, surgery	0 (0%)	23 (58%)	0 (0%)	15 (33%)	92 (70%)	0 (0%)	45 (68%)	79 (79%)
Secondary, orthopedic, surgery	0 (0%)	0 (0%)	0 (0%)	2 (5%)	20 (15%)	0 (0%)	17 (26%)	38 (38%)

Follow-up in years Mean (SD)	0.2 (0.3)	1.0 (0.7)	0.4 (0.3)	3.1 (2.5)	5.1 (2.5)	0.3 (0.3)	9.3 (4.6)	8.9 (4.7)
TAPQOL scales Mean (SD)			x	x	x	x	x	x
Positive Mood scale	100.0 (0.0)	95.7 (10.9)						
Problem Behavior scale	80.4 (23.1)	74.2 (19.4)						
Anxiety scale	86.5 (21.3)	81.6 (20.7)						
Social Functioning scale	95.2 (8.1)	91.1 (15.8)						
Motor Functioning scale	90.6 (7.5)	82.4 (18.0)						
PODCI scales Mean (SD)	x	x						
Upper Extremity			89.9 (12.5)	90.1 (14.1)†	75.3 (18.8) β	92.7 (13.3)	83.2 (15.7)	79.7 (17.6)[α]
Transfer and Basic Mobility			99.0 (2.4)	99.4 (1.6)†	96.7 (5.4) β	98.9 (2.0)	98.3 (2.8)	98.8 (2.6)
Sports and Physical Functioning			94.4 (7.9)	95.7 (6.9)†	90.9 (9.6) β	94.4 (10.1)	91.3 (10.5)	91.1 (9.4)
Pain and Comfort			94.3 (11.6)	97.9 (7.2)	92.6 (14.7)‡	99.0 (3.3)	87.2 (18.6)[α]	90.9 (16.3)
Happiness scale			97.5 (7.2)	96.7 (10.0)†	91.6 (13.1) β	90.1 (23.1)	84.6 (17.4)	86.2 (18.6)
Global Functioning scale			94.3 (5.9)	95.9 (5.9)†	88.8 (9.4) β	96.2 (6.6)	89.4 (10.7)	90.1 (8.8) β

*Discharged from follow-up within 1 year after birth, **Discharged from follow-up later in life, ***Still in follow-up.
TAPQOL: TNO-AZL (Netherlands Organization for Applied Scientific Research and Leiden University Hospital) Preschool children's Quality of Life questionnaire
PODCI: Pediatric Outcome Data Collecting Instrument
β significantly different from the early discharge and late discharge groups ($p<0.05$); ‡ significantly different from the late discharge group ($p<0.05$),
α significantly different from the early discharge group($p<0.05$); †not significantly different from the early discharge group.

Table II Healthcare use by 465 patients due to neonatal brachial plexus palsy, since birth and in the past 12 months, in relation to current follow-up status at the Leiden Nerve Center.

	0-1 years (n=59)		**2-9 years (n=226)**			**10-18 years (n=180)**		
	Early discharge* (n=19)	**No discharge*** (n=40)**	**Early discharge* (n=50)**	**Late discharge** (n=44)**	**No discharge*** (n=132)**	**Early discharge* (n=14)**	**Late discharge** (n=66)**	**No discharge*** (n=100)**
	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months	Ever/past 12 months
Number of patients having had contact with NBPP expert team N	19/12	40/37	50/0	44/5	132/76	14/0	66/17	100/51
Physical therapist	13/11	34/34	33/0	26/4	110/66	3/0	41/12	73/39
Occupational therapist	8/5	25/25	8/0	13/1	57/27	0/0	13/3	31/12
Neurosurgeon	10/7	36/33	24/0	33/3	115/44	4/0	54/12	85/27
Orthopedic surgeon	4/1	13/11	12/0	11/1	64/31	2/0	36/1	68/21
Rehabilitation specialist (physiatrist)	7/4	19/16	14/0	18/1	83/43	2/0	37/9	65/33
Number of patients having had contact with at least 1 professional outside the NBPP expert team N	19/16	40/37	50/14	44/15	132/104	14/4	66/31	100/67
Pediatric/General Physical therapist	15/15	38/37	47/5	37/11	126/86	8/3	65/17	89/53
Occupational therapist	0/0	4/4	1/1	1/0	39/23	1/1	13/3	23/6
Family doctor	7/2	20/11	32/1	15/2	65/14	6/1	39/8	57/8
Neurosurgeon	5/3	20/14	7/0	14/0	78/13	2/1	36/3	54/3
Orthopedic surgeon	4/2	4/4	1/0	2/0	33/14	2/0	14/1	32/4
Rehabilitation specialist (physiatrist)	3/1	10/7	2/0	5/0	51/27	2/1	23/8	52/18
Pediatrician	15/9	32/21	39/2	31/0	94/9	6/0	46/0	70/8
Plastic surgeon	1/0	1/1	1/1	0/0	12/3	0/0	6/0	6/0
Psychologist	0/0	6/6	5/3	2/1	13/6	1/1	4/1	8/1
Psychiatrist	0/0	2/1	2/2	0/0	7/3	0/0	3/0	7/2
Social Worker	1/0	4/4	4/1	0/0	9/2	0/0	5/2	7/3

Hospital admission N								
Yes	3/1	26/14	11/0	15/0	96/5	0/0	45/0	82/1
Contact with Patient Organization N								
Yes	0/0	10/8	4/1	9/4	46/20	0/0	28/5	30/9
Use of complementary medicine N								
Yes	1/0	6/6	10/3	9/0	21/7	0/0	7/3	7/2
Healthcare use[#] Median(IQR)								
Healthcare use ever	4.0 (3.0-4.5)	5.0 (4.0-6.0)	3.0 (4.0-5.0)	3.0 (4.0-5.0)	4.0 (6.0-8.0)	4.0 (1.5-5.0)	6.0 (4.0-7.0)	6.0 (4.8-8.0)
Healthcare use in past 12 months	3.0 (1.5-3.0)	6.0 (5.0-8.0)	0.0 (0.0-1.0)	0.0 (0.0-1.0)	2.0 (1.0-3.0)	0.0 (0.0-1.0)	0.0 (0.0-2.0)	2.0 (0.0-3.0)

*Discharged from follow-up within 1 year after birth. **Discharged from follow-up later in life. ***Still in follow-up.

[#] Healthcare use; number of healthcare professionals/professions involved (range 0-13): NBPP expert team (n=1), additional healthcare professionals (n=11), hospital admission (n=1)

Table III Factors associated with healthcare use in the past 12 months of 465 patients due to neonatal brachial plexus palsy, in relation to age.

	0-1 years (n=59)		2-9 years (n=226)		10-18 years (n=180)	
Factors used in the univariate and/or multiple regression analyses	**Univariate β-estimate (95% CI)**	**Multiple β-estimate (95% CI)**	**Univariate β-estimate (95% CI)**	**Multiple β-estimate (95% CI)**	**Univariate β-estimate (95% CI)**	**Multiple β-estimate (95% CI)**
Gender						
Male	-0.17 (-1.9;1.6)		0.6 (0.14;1.1)*	0.3 (-0.1;0.8)	0.2 (-0.3;0.6)	
Female	reference cat.		reference cat.		reference cat.	
Age	-1.8 (-3.2;-0.5)*	-2.0 (-3.3;-0.8) †	0.04 (-0.08;0.15)		-0.02 (-0.04;-0.01)	
Affected side:						
Right	reference cat.		reference cat.		reference cat.	
Left	-0.7 (-2.4;1.1)		-0.4 (-0.9;0.1)$^{\#}$	-0.4 (-1.1;0.2)	0.2 (-0.2;0.7)	
Both	x		0.8 (-1.0;2.6)	0.9 (-1.3;3.1)	0.4 (-0.6;1.4)	
Lesion extent:						
Upper plexus lesions	reference cat		reference cat.		reference cat.	
Total and lower plexus lesions	3.2 (1.1;5.3) †	1.8 (-0.3;3.8)	2.4 (1.7; 3.0)‡	1.3 (0.7;2.0)‡	1.2 (0.6;1.7)‡	0.9 (0.2;1.6)†
Treatment history:						
Conservative	reference cat.		reference cat.		reference cat.	
Primary,nerve, surgery	2.2 (0.5;3.9)*	1.1 (-0.7;3.0)	1.4 (0.9;1.8)‡	0.2 (-0.4;0.8)	0.4 (-0.2;0.9)	0.3 (-0.4;0.9)
Secondary, orthopedic, surgery	x		0.5 (-1.2;2.1)	-0.2 (-1.8;1.3)	0.2 (-0.5;0.9)	0.4 (-0.8;1.6)
Primary and secondary surgery	x		2.4 (1.5;3.2) ‡	1.0 (0.2;1.9)*	1.0 (0.5;1.6) ‡	0.5 (-0.3;1.2)
Follow-up status:						
Discharged aged < 1 year	-2.9 (-4.6;-1.1) †	-1.9 (-3.7;0.1)*	-1.9 (-2.4;-1.3)‡	-1.1 (-1.8;-0.4)†	-0.7(-1.6;0.2)	-0.1 (-1.0;0.8)
Discharged aged > 1 year	x		-1.7 (-2.3;-1.2)‡	-0.8 (-1.4;-0.2)†	-0.7 (-1.2;-0.3)‡	-0.3 (-0.8;0.2)
Not discharged, still in follow-up	reference cat.		reference cat.		reference cat.	
TAPQOL scales:						
Motor Functioning scale (0-100)	-0.02 (-0.1;0.05)		x		x	
PODCI scales:						
Upper Extremity (UE) scale (0-100)	x		-0.05 (-0.06;-0.04)‡	0.01 (-0.01;0.04)	-0.04 (-0.05;-0.03)‡	-0.01 (-0.02;0.02)
Global Functioning (GF) scale (0-100)	x		-0.11 (-0.13;-0.08)‡	-0.08 (-0.12;-0.03)‡	-0.07 (-0.10;-0.05)‡	-0.06 (-0.1;0.02) †

*p<0.05, †p<0.01, ‡p<0.001, $^{\#}$p<0.2. β-estimate: difference in healthcare use score compared to the reference category (reference cat.). TAPQOL: TNO-AZL (Netherlands Organization for Applied Scientific Research and Leiden University Hospital) Preschool children's Quality of Life questionnaire. PODCI: Pediatric Outcome Data Collecting Instrument. β-estimate for TAPQOL and PODCI scores: each point lower on these scales results in a β-estimate higher or lower healthcare use, e.g. in the 2-9 years age group a score of 20 points less on the PODCI GF scale results in an increase of 1 point on the healthcare use score (20 * the β-estimate of -0.05 =1).

Quality of Life

One of the main goals of interventions in NBPP is to improve all aspects of QoL (i.e. activities, participation) by enhancing bodily functions. The current study showed that patients with a lower QoL score used more healthcare. It is important to acknowledge the current reported QoL of patients, in order to optimize follow-up planning. Our findings regarding QoL and physical functioning are in line with those of previous studies.[25,27,28,34-37] For the more severely affected children (the group still in follow-up), QoL and physical functioning scores were comparable to those reported in other studies.[27,28,36,37] Children in the early and late discharge groups, however, also reported problems of QoL and physical functioning, with older children (the 10-18 years age group) reporting more problems (Table I).

Discharge from follow-up

Children who are discharged from follow-up by the expert team at a young age (<1 year) because of satisfactory clinical functional recovery, i.e. with no need for interventions at the Leiden Nerve Center, were expected to have no specific problems in later life and to have no need for further treatment. But contrary to the expectation of the Leiden Nerve Center team, the parents of these patients were still in need of help from healthcare professionals in their local area. This phenomenon has not been the subject of any study yet, whereas it is an important finding for both healthcare professionals and tertiary expert teams. This issue needs to be addressed, while at the same time preventing overuse of healthcare by less specialized care providers. Our study found that the reported QoL and physical functioning for some of these patients was lower than expected and that some children were still receiving active treatment for their NBPP. The expectation of full recovery at an early age was apparently incorrect, and the question arises whether this appraisal can be adequately made and whether these patients should have been discharged.

Information needs

At the Leiden Nerve Center, not only care requirements but also future information needs are among the factors used to decide whether or not to make routine follow-up appointments. As it turned out, the need for information due to sequelae of NBPP in our population was substantial. About 50% in all age groups reported to have a need for more information than they had been given regarding one or more NBPP-related topics. Since this percentage was found in all age groups, information need appears not to decrease with age.

This study also showed that 18% of the participants had received/found contradicting information regarding NBPP (Table IV). An American study found that decision making is highly influenced by the information found, so uniform, easily accessible information on all reported topics would be valuable to patients with NBPP and/or their parents.[23] Providing the opportunity for e-mail contact with a specialized NBPP consultant would also be useful to our patient population, as 63% of our participants stated that they would use such an option. Only a small proportion of the patients (n=45, 10%) had recently had contact with the patient organization. Communicating the benefits of the patient organization in providing information and peer contacts may further decrease the unmet information needs in the NBPP population.

Table IV Current information needs of 465 patients with neonatal brachial plexus palsy, in relation to age and follow-up status at the Leiden Nerve Center.

		0-1 years (n=59)		**2-9 years (n=226)**			**10-18 years (n=180)**		
		Early discharge* (n=19)	**No discharge*** (n=40)**	**Early discharge* (n=50)**	**Late discharge** (n=44)**	**No discharge*** (n=132)**	**Early discharge* (n=14)**	**Late discharge** (n=66)**	**No discharge*** (n=100)**
Information sought	yes n(%)	12 (63%)	36 (90%)	29 (58%)	30 (68%)	107 (81%)	3 (21%)	32 (49%)	65 (65%)
Information found	yes n(%)	10 (53%)	23 (58%)	23 (46%)	24 (55%)	73 (55%)	2 (14%)	26 (39%)	48 (48%)
Received contradictory information from different healthcare providers	yes n(%)	3 (16%)	9 (23%)	7 (14%)	7 (16%)	36 (27%)	2 (14%)	6 (9%)	14 (14%)
Would like more information on at least 1 of the topics below	yes n(%)	3 (16%)	28 (70%)	17 (34%)	14 (32%)	80 (61%)	3 (21%)	28 (42%)	55 (55%)
Neonatal Brachial Plexus Palsy	yes n(%)	1 (5%)	10 (25%)	9 (18%)	4 (9%)	27 (20%)	1 (7%)	6 (9%)	18 (18%)
NBPP physical consequences	yes n(%)	2 (10%)	18 (45%)	12 (24%)	11 (25%)	56 (42%)	1 (7%)	16 (24%)	36 (36%)
NBPP medical treatment	yes n(%)	1 (5%)	11 (28%)	8 (16%)	4 (9%)	36 (27%)	1 (7%)	4 (6%)	19 (19%)
Assistive devices and government social support	yes n(%)	1 (5%)	17 (43%)	7 (14%)	10 (23%)	47 (36%)	2 (14%)	12 (18%)	34 (34%)
Physical activity and sports	yes n(%)	3 (15%)	16 (40%)	16 (32%)	10 (23%)	55 (42%)	2 (14%)	13 (20%)	26 (26%)
Pediatric or General Physical therapy	yes n(%)	2 (10%)	8 (20%)	7 (14%)	5 (11%)	30 (23%)	2 (14%)	3 (5%)	15 (15%)
Occupational Therapy	yes n(%)	1 (5%)	9 (23%)	3 (6%)	3 (7%)	30 (23%)	2 (14%)	4 (6%)	14 (14%)
Primary surgery	yes n(%)	0 (0%)	3 (8%)	1 (2%)	1 (2%)	16 (12%)	0 (0%)	2 (3%)	12 (12%)
Secondary surgery	yes n(%)	0 (0%)	5 (13%)	1 (2%)	1 (2%)	24 (18%)	1 (7%)	3 (5%)	10 (10%)
Rehabilitation medicine	yes n(%)	1 (5%)	5 (13%)	3 (6%)	3 (7%)	23 (17%)	2 (14%)	4 (6%)	9 (9%)
Social work	yes n(%)	0 (0%)	4 (10%)	2 (4%)	2 (4%)	15 (11%)	0 (0%)	2 (3%)	4 (4%)
Patient organizations/ Peer contact	yes n(%)	1 (5%)	8 (20%)	1 (2%)	1 (2%)	11 (8%)	0 (0%)	4 (6%)	12 (12%)

Preferred mode of information delivery									
Internet	yes n(%)	14 (74%)	32 (80%)	32 (64%)	27 (61%)	104 (79%)	7 (50%)	30 (46%)	74 (74%)
Brochures/Books	yes n(%)	9 (45%)	21 (53%)	20 (40%)	17 (39%)	63 (48%)	4 (28%)	18 (27%)	48 (48%)
Peer contact	yes n(%)	2 (10%)	9 (23%)	6 (12%)	5 (11%)	38 (29%)	2 (14%)	9 (14%)	30 (30%)
Patient organization meetings	yes n(%)	1 (5%)	7 (18%)	3 (6%)	2 (4%)	28 (21%)	3 (21%)	8 (12%)	22 (22%)
Information meeting in the hospital	yes n(%)	5 (26%)	13 (33%)	5 (10%)	9 (20%)	38 (29%)	3 (21%)	17 (26%)	23 (23%)
Treating physician	yes n(%)	14 (74%)	29 (73%)	15 (30%)	17 (39%)	82 (62%)	4 (28%)	22 (33%)	55 (55%)
Family doctor	yes n(%)	6 (30%)	10 (25%)	16 (32%)	6 (13%)	21 (16%)	3 (21%)	10 (15%)	24 (24%)
Pediatric) physical / occupational therapist	yes n(%)	14 (74%)	28 (70%)	27 (54%)	22 (50%)	88 (67%)	5 (35%)	25 (38%)	53 (53%)
Would use possibility to e-mail with specialized NBPP consultant	yes n(%)	9 (45%)	30 (75%)	24 (48%)	20 (46%)	102 (77%)	6 (42%)	28 (42%)	61 (61%)

*Discharged from follow-up within 1 year after birth, **Discharged from follow-up later in life, ***Still in follow-up.

Study limitations

This study has a number of limitations. Firstly, it has a cross-sectional design with no follow-up, using only self-reported questionnaires. This 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. mood, stress, etc.). Secondly, outcomes may be influenced by recall bias. Older patients and their parents may have forgotten exactly which healthcare professionals were involved at the time. We therefore only analyzed factors influencing healthcare use in the past 12 months, as recall bias for this period of time was considered minimal.

Thirdly, patients seen at our NBPP clinic were referred to us because of a severe lesion, which might lead to confounding by indication. However, since we had a relatively large group of respondents, this will reflect a good representation of the children seen at NBPP clinics in other academic settings.

The healthcare system and care at university hospitals in the Netherlands differ from those in other countries. The Netherlands has private insurance for all citizens based on a solidarity system (i.e. richer people do not receive financial government support to compensate their insurance rates). It is a small country and travelling distances between cities and to university-based centers are relatively short, which reduces the threshold for visiting a university-based center. Medical specialists in the Netherlands are diagnosis-oriented. In other countries, NBPP specialists combine performing primary and secondary surgery with rehabilitation, whereas staff at the Leiden Nerve Center are accustomed to working in interdisciplinary teams including neurosurgeons, orthopedic surgeons, physiatrists and physical and occupational therapists. This could mean that the present study may have overestimated the number of healthcare professionals involved in the care of these patients. Furthermore, parents may be emotionally attached to specific healthcare professionals, for example their local pediatric physical therapist, which may lead to more healthcare use for their child. Physical therapy for NBPP is considered a chronic indication in the Dutch healthcare system, and is reimbursed by health insurance companies. On the other hand, all patients/parents have to pay up to a maximum of €350 out of their own pocket for all healthcare used per annum, which could form a barrier to healthcare use. The number of visits to healthcare professionals, the costs of NBPP treatment and other aspects of healthcare utilization were not taken into account in the present study, and remain an interesting topic for future research.

Future research and endeavors

Future studies into clinical outcomes of NBPP should take into account the residual healthcare use by children who in the view of the expert teams had good clinical recovery. It is important to find out what patients discharged from follow-up and their parents think about care and information for NBPP, why they still have information needs and if they know how to find/contact the care providers they need.

Finally, there is a need to develop an easy and effective way to deliver information focusing on the different stages of life with NBPP (e.g. when going to school, or when choosing a sport, a subject to study or a profession etc.). Suitable options could include producing a modular informative video providing the information needed by individual patients, or information brochures.

CONCLUSION

Healthcare use and information needs of children with NBPP have not been studied before. Our study of a large NBPP sample has revealed which healthcare professionals are involved in the care for patients with NBPP and what information is needed by this population. Furthermore, it showed that children who showed satisfactory spontaneous clinical recovery at a young age, and were subsequently discharged from follow-up from our tertiary referral center, continued to seek active treatment for their NBPP, reported problems of QoL and physical functioning, and still had need for further information. As parents of children, both early and late discharged from follow-up, report healthcare use and current information needs due to their child's NBPP, stricter longitudinal follow-up on care and information needs by multidisciplinary NBPP expert teams for all patients with NBPP throughout life is needed as NBPP may result in lifelong limitations.

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