Health-care use and information needs of children with neonatal brachial plexus palsy: a cross-sectional survey among 465 Dutch patients

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Abstract

To investigate health-care use (HCU) and information needs of children aged 0–18 years with neonatal brachial plexus palsy (NBPP), a cross-sectional study was performed. Patients and/or parents seen in our NBPP clinic were invited to complete a survey comprising questions on HCU due to NBPP and current information needs. Outcomes were described for three age-groups (0–1/2–9/10–18 years), based on follow-up status (early/late/no-discharge). Four hundred sixty-five parents/patients participated (59 in the 0–1, 226 in the 2–9, and 180 in the 10–18-year group). Two hundred ninety-three patients had C5–C6 lesions, 193 were discharged from follow-up, 83 of whom categorized as 'early discharged' (<1 year of age). Over the past year, 198 patients had contact with the expert team (49 in the 0–1, 81 in the 2–9, and 68 in the 10–18-year group) and 288 with at least one other health-care professional (53 in the 0–1, 133 in the 2–9, and 102 in the 10–18-year group). Of the 83 patients discharged early, 34 reported health-care use. Two hundred twenty-eight participants reported current information needs of whom 23 were discharged early. HCU and information needs of Dutch children with NBPP remains considerable even in children who were discharged. Stricter follow-up and information provision for these patients is needed.

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Introduction

Neonatal brachial plexus palsy (NBPP) occurs in about one in three children per 1000 births in Western countries (Chauhan et al., 2014; Pondaag et al., 2004). Seventy to ninety percent of these children recover spontaneously, while the remaining 10–30% are left with neurological damage, possibly resulting in functional impairments (Chauhan et al., 2014; Evans-Jones et al., 2003; Lagerkvist et al., 2010; Waters, 1999). Children with severe NBPP in, for example, Canada, Sweden, and the Netherlands are usually referred to a tertiary NBPP expert center for further diagnostics and follow-up (Coroneos et al., 2015a; Pondaag et al., 2004; Strombeck et al., 2000). If these children do not show sufficient spontaneous recovery around the age of three months, primary, nerve, surgery may be indicated (Pondaag and Malessy, 2014; Tse et al., 2015).

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 (Hale et al., 2010; Louden et al., 2013; Waters, 2005).

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 (Coroneos et al., 2015a; Malessy et al., 2011). The Leiden Nerve Center has successfully promoted early referral, that is, at the age of one month (Malessy et al., 2011). Infants are assessed and treated by a multidisciplinary expert team involving a variety of medical and allied health-care professionals using an interdisciplinary approach (Coroneos et al., 2015a; Coroneos et al., 2015b; Hale et al., 2010; Pondaag et al., 2004; Pondaag and Malessy, 2014; Strombeck et al., 2007; Waters, 2005). In addition, most children are treated by health-care professionals in primary care in their place of residence (e.g. allied health care or psychosocial) and, if insufficient, interdisciplinary rehabilitation care is provided (Ramos and Zell, 2000; Smania et al., 2012). 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 (Bain et al., 2009; Bialocerkowski et al., 2005; Bialocerkowski and Gelding, 2006; Coroneos et al., 2015b; Duff and DeMatteo, 2015; Pondaag and Malessy, 2014; Socolovsky et al., 2015). 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 no literature on the health-care use (HCU) of children with NBPP and factors related to it/associated with it. Furthermore, 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 (Shah et al., 2006). To date, it remains unclear whether patients and/or their parents/caregivers, whether in clinical follow-up or not, have unmet information needs.

Aim

The aim of this study was to quantify the HCU (defined in this study as the number of professionals involved in the care) of children with NBPP due to their condition, to determine which factors influenced health-care use, and to identify the information needs of patients and/or their parents/caregivers at different ages and in various follow-up categories.

Methods

Study design and patients

To investigate the above objectives, a cross-sectional study was designed 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.

The present study used only the data of patients who were 18 years and younger.

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, orthopedic, surgery, 4; primary and secondary surgery).

Three age-groups were distinguished based on the age requirements of the used outcome questionnaires (TNO-AZL (the Netherlands Organization for Applied Scientific Research and Leiden University Hospital) Preschool children's Quality of Life (QoL) (TAPQOL) and the Pediatric Outcome Data Collecting Instrument (PODCI), see section QoL and physical functioning). Age-groups were as follows: 0–1 (0–1 years old), 2–9 (2–9 years old), and 10–18 (10–18 years old). For these groups, outcomes were described separately.

Follow-up status

The follow-up status of all patients of the Leiden Nerve Center was extracted from the medical records. Based on this, three subgroups were defined: (1) early discharge, that is, discharged from follow-up within a year after birth; (2) late discharge, that is, discharged from follow-up at a later age; and (3) no discharge, that is, 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 satis-factory spontaneous recovery, not needing further treatment. This was verified by checking the medical records for the reason of discharge.

Health-care use (HCU)

The proxy for HCU by children with NBPP in this study was defined as the number of health-care 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 health-care 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 one of the five members of the NBPP expert team (i.e. neurosurgeon, orthopedic surgeon, rehabilitation specialist (physia-trist), physical therapist, occupational therapist). Furthermore, one point was allocated for each of the 11 types of health-care professionals contacted outside the expert team. In addition, one 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. homeop-athy, 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.

QoL and physical functioning

Perceived QoL and physical functioning were examined using the TAPQOL and PODCI (Bunge et al., 2005; Holst et al., 2015).

The TAPQOL was developed to measure QoL in children aged six months to five years (Bunge et al., 2005). 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. 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, because only these scales were found to provide some insight into the QoL of young children with NBPP (van der Holst et al., 2016).

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 (Bae et al., 2008; Christakou and Laiou, 2014; Holst et al., 2015; Huffman et al., 2005). The instrument consists of five subscales and one total score. PODCI scale scores range from 0 to 100, with higher scores indicating better functioning/QoL. The present study used the 2–9 years and 10–18 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 whether 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 health-care 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 (IQRs) or means with standard deviations (SDs) based on the distribution of the data (Kolmogorov–Smirnoff test)) were used for patient characteristics and 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 independent samples Mann–Whitney U test, and PODCI scores were compared using a one-way analysis of variance with Fisher's least significant difference post hoc test (significance level, $p < .05$).

To determine which factors were associated with HCU-12, univariate regression analyses were performed for all age-groups (significance level, $p < .05$). Factors entered independently, one at a time, include 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 $p < .2$ in the univariate analyses. Differences in HCU based on the factors entered in the univariate and multiple regression analyses are presented as β estimates with 95% confidence intervals.

Ethics approval

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Results

A total of 1142 patients were invited to participate in the overall study, of whom 508 patients and/or their parents participated. Of these 508 patients, 465 were aged 18 years and younger, of whom 59 (13%) belonged to the 0 to 1-year age-group, 226 (48%) to the 2 to 9-year group, and 180 (39%) to the 10 to 18-year group. A total of 83 (18%) patients belonged to the early discharge subgroup. The flow of patients and their characteristics are presented in Figure 1 and Table 1, respectively.
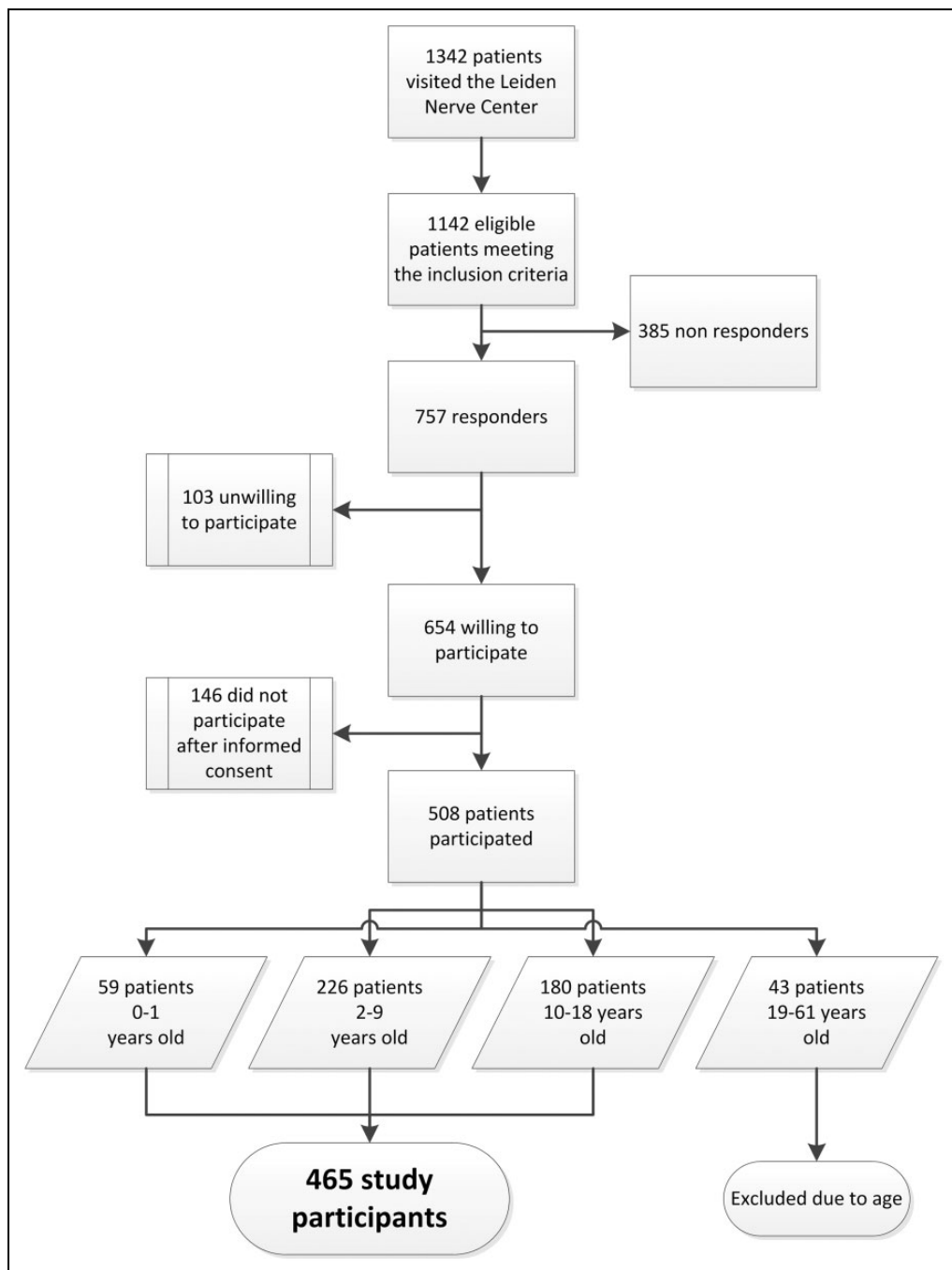
Figure 1. Flowchart showing the formation of the study sample (*n* = 465).

Table 1. 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[a] (n = 19)	No discharge[b] (n = 40)	Early discharge[a] (n = 50)	Late discharge[c] (n = 44)	No discharge[b] (n = 132)	Early discharge[a] (n = 14)	Late discharge[c] (n = 66)	No discharge[b] (n = 100)
Gender								
-Male	12 (63%)	16 (40%)	28 (56%)	16 (36%)	70 (53%)	9 (64%)	26 (39%)	43 (43%)
Median age (IQR 25;75)	1 (1;1)	1 (1;2)	6 (4;7)	6 (4;8)	6 (5;8)	14 (11;15)	15 (12;17)	13 (11;15)
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	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, median (IQR)	.2 (.0;.7)	1.0 (.4;1.6)	.4 (.0;.7)	2 (2;5)	6 (3;7)	.3 (.0;.6)	10 (5;13)	10 (5;12)
TAPQOL scales, median (IQR)								
Positive Mood Scale	100 (100;100)	100 (100;100)	x	x	x	x	x	x
Problem Behavior Scale	86 (73;98)	71 (64;93)						
Anxiety Scale	100 (71;100)	83 (67;100)						
Social Functioning Scale	100 (83;100)	100 (83;100)						
Motor Functioning Scale	91 (83;98)	84 (72;100)						

(continued)

Table 1. (continued)

PODCI scales, mean (SD)	0 –1 years (n = 59)		2–9 years (n = 226)			10–18 years (n = 180)		
	Early discharge[a] (n = 19)	No discharge[b] (n = 40)	Early discharge[a] (n = 50)	Late discharge[c] (n = 44)	No discharge[b] (n = 132)	Early discharge[a] (n = 14)	Late discharge[c] (n = 66)	No discharge[b] (n = 100)
PODCI scales, mean (SD)	x	x						
Upper Extremity			89.9 (12.5)	90.1 (14.1)[d]	75.3 (18.8)[e]	92.7 (13.3)	83.2 (15.7)	79.7 (17.6)[f]
Transfer and Basic Mobility			99.0 (2.4)	99.4 (1.6)[d]	96.7 (5.4)[e]	98.9 (2.0)	98.3 (2.8)	98.8 (2.6)
Sports and Physical Functioning			94.4 (7.9)	95.7 (6.9)[d]	90.9 (9.6)[e]	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)[g]	99.0 (3.3)	87.2 (18.6)[f]	90.9 (16.3)
Happiness Scale			97.5 (7.2)	96.7 (10.0)[d]	91.6 (13.1)[e]	90.1 (23.1)	84.6 (17.4)	86.2 (18.6)
Global Functioning Scale			94.3 (5.9)	95.9 (5.9)[d]	88.8 (9.4)[e]	96.2 (6.6)	89.4 (10.7)	90.1 (8.8)[e]

IQR 25;75: inter quartile range, 25th and 75th percentile; 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.
[a]Discharged from follow-up within one year after birth.
[b]Still in follow-up.
[c]Discharged from follow-up later in life.
[d]Not significantly different from the early discharge group.
[e]Significantly different from the early discharge and late discharge groups (p < .05).
[f]Significantly different from the late discharge group (p < .05).
[g]Significantly different from the early discharge group (p < .05).

Table 1 also shows QoL and physical functioning scores (TAPQOL and PODCI). In the 0 to 1-year age-group, there was no difference in QoL (TAPQOL) between the early discharge subgroup and the subgroup still in follow-up. In the 2 to 9-year 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 to 18-year 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 2 presents the health-care professionals involved in the care of children with NBPP, and these children's median HCU (HCU-ever/HCU-12) for all age-groups and follow-up subgroups. Since birth, all patients had contact with at least 1 (range 1–11) health-care professional in addition to the NBPP expert team. Hospital admissions due to NBPP were reported for 278 (60%) patients since birth. The most frequently mentioned health-care professionals contacted since birth were the pediatric or general physical therapist, neurosurgeon, rehabilitation specialist (physiatrist), orthopedic surgeon, and pediatrician, but other professionals were mentioned as well, including psychologists ($n = 39$; 8%) and psychiatrists ($n = 21$; 5%).

In the past 12 months, 198 (43%) patients had contact with the expert team (divided over the three age-groups as follows: 49 (83%), 81 (36%), and 68 (38%)). At least one (additional) health-care professional (range 1–7) had been contacted by 288 (62%) patients (divided over the three age-groups: 53 (90%), 133 (59%), and 102 (57%)). The physical therapist was again the most frequently mentioned health-care professional contacted.

In the early discharge subgroup, 34 (41%) patients had contacted at least one health-care professional during the past 12 months for their NBPP. In this subgroup, the physical therapist was mentioned 23 (28%) times.

Factors independently associated with HCU were lesion extent, treatment history, follow-up status and QoL and physical functioning; male gender was associated with higher HCU in the 2 to 9-year age-group (all $p < .05$, see Supplementary Material, Table S1).

Multiple regression analysis showed that for the 2 to 9-years age-group, greater lesion extent, treatment history (primary and secondary surgery), being in follow-up and lower QoL (lower PODCI GF scale-scores) were associated with higher health-care use. For the 10 to 18-year age-group, only greater lesion extent and QoL (lower PODCI GF scale scores) were associated with higher HCU (all $p < .05$, see Supplementary Material, Table S1).

All age-groups and all follow-up subgroups reported information needs (Table 3). Three hundred sixteen (68%) of the respondents had ever sought information regarding NBPP, but only 228 (49%) had found what they were looking for. Furthermore, 84 (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 (49%) patients/parents. In the early and late discharge subgroups, information need was reported by 23 (28%) patients and by 42 (40%) patients, 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.

Table 2. Health-care 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[a] (n = 19)		No discharge[b] (n = 40)		Early discharge[a] (n = 50)		Late discharge[c] (n = 44)		No discharge[b] (n = 132)		Early discharge[a] (n = 14)		Late discharge[c] (n = 66)		No discharge[b] (n = 100)	
	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M
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 one 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

(continued)

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Table 2. (continued)

	0–1 years (n = 59)				2–9 years (n = 226)						10–18 years (n = 180)					
	Early discharge[a] (n = 19)		No discharge[b] (n = 40)		Early discharge[a] (n = 50)		Late discharge[c] (n = 44)		No discharge[b] (n = 132)		Early discharge[a] (n = 14)		Late discharge[c] (n = 66)		No discharge[b] (n = 100)	
	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M	Ever	12M
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
Health-care use[d] Median (IQR)																
Healthcare use ever	4.0 (3.0–4.5)		5.0 (4.0–6.0)		3.0 (4.0–5.0)	.0	3.0 (4.0–5.0)		4.0 (6.0–8.0)		4.0 (1.5–5.0)	.0	6.0 (4.0–7.0)		6.0 (4.8–8.0)	
Health-care use in past 12 months	3.0 (1.5–3.0)		6.0 (5.0–8.0)		.0 (.0–1.0)		.0 (.0–1.0)		2.0 (1.0–3.0)		.0 (.0–1.0)		.0 (.0–2.0)		2.0 (.0–3.0)	

Ever: patients who ever had contact with a healthcare professional and/or the expert team; 12 M: patients who had contact with a healthcare professional and/or the expert team in the past 12 months.
[a] Discharged from follow-up within one year after birth.
[b] Still in follow-up.
[c] Discharged from follow-up later in life.
[d] Health-care 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 3. 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[a] (n = 19)	No discharge[b] (n = 40)	Early discharge[a] (n = 50)	Late discharge[c] (n = 44)	No discharge[b] (n = 132)	Early discharge[a] (n = 14)	Late discharge[c] (n = 66)	No discharge[b] (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 one 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%)

[a] Discharged from follow-up within one year after birth.
[b] Still in follow-up.
[c] Discharged from follow-up later in life.

Discussion

This cross-sectional study in a large sample showed that HCU since birth by children due to NBPP in the Netherlands is considerable but decreases over time. 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. Furthermore, a large proportion ($n = 228$; 49%), reported information needs regarding a variety of NBPP-related topics (treatment, sports and physical functioning, assistive devices, etc.).

Health-care use

No study of the HCU by patients with NBPP has been performed before, so no comparisons with other countries or centers can be made. Although studies of HCU in pediatric populations have been performed, they mainly focused on hospitalization and/or health-care costs (Cohen et al., 2012; Lopez et al., 2014; Ralston et al., 2015; Zhong et al., 2015). 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 health-care professionals in the past 12 months (Gunz et al., 2012). In contrast, our study found up to a median of 6.0 contacts (depending on age and follow-up status, Table 2). However, we only counted the number of health-care professionals contacted, but not the number of visits. In addition, we took contacts with other health-care professionals besides the Leiden Nerve Center team into account. Our study showed that allied health professionals, especially (pediatric) physical therapists, were frequently contacted.

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 health-care. It is important to acknowledge the current reported QoL of patients, to optimize follow-up planning. Our findings regarding QoL and physical functioning are in line with those of previous studies (Alyanak et al., 2013; Akel et al., 2013; Bae et al., 2008; Dedini et al., 2008; Huffman et al., 2005; van der Holst et al., 2015; van der Holst et al., 2016). 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 (Bae et al., 2008; Dedini et al., 2008; Huffman et al., 2005; van der Holst et al., 2015). Children in the early and late discharge groups, however, also reported problems of QoL and physical functioning, with older children (the 10- to 18-year age-group) reporting more problems (Table 1).

Discharge from follow-up

A large proportion of patients discharged from follow-up by the Leiden Nerve Center still had contact with regional health-care professionals for their NBPP ($n = 81$; 42%). They included a relatively large proportion of patients ($n = 34$; 41%) discharged at a young age. Children who are discharged from follow-up by the expert team at a young age (<1 year) because of satisfactory clinical functional recovery, that is, 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 expectation, these patients were still in need of help from health-care professionals

in their local area. This phenomenon has not been the subject of any study yet, whereas it is an important finding for both health-care professionals and tertiary expert teams. This issue needs to be addressed, while at the same time preventing overuse of health care by less specialized care providers. Our study found that the reported QoL and physical functioning for some of these patients were 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 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. As this percentage was found in all age-groups, information need appears not to decrease with age. Furthermore, information need was even found in the early and late discharge groups, indicating that even after discharge information need over time remains to exist.

This study also showed that 18% of the participants had received/found contradicting information regarding NBPP (Table 3). 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 (Shah et al., 2006). 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.

Study limitations

This study had a number of limitations. Firstly, it had 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). Secondly, outcomes may be influenced by recall bias. Older patients and their parents may have forgotten exactly which health-care professionals were involved at the time. We therefore only analyzed factors influencing HCU 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, as 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 health-care system and care at university hospitals in the Netherlands differ from those in other countries. The Netherlands has private insurance for all citizens (http://wetten.overheid.nl/BWBR0018450/2018-01-01#Hoofdstuk5) 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

traveling 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 may 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 this study may have overestimated the number of health-care professionals involved in the care of these patients. Furthermore, parents may be emotionally attached to specific health-care professionals, for example, their local pediatric physical therapist, which may lead to more HCU for their child. Physical therapy for NBPP is considered a chronic indication in the Dutch healthcare system and is reimbursed by health insurance companies (https://www.rijksoverheid.nl/onderwerpen/zorgverzekering/vraag-en-antwoord/is-fysiotherapie-opgenomen-in-het-basispakket). 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 HCU 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). Suitable options could include producing a modular informative video providing the information needed by individual patients, or information brochures.

Implications for practice

As parents of children, whether discharged from follow-up or not, report HCU 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.

Conclusion

HCU and information needs of children with NBPP have not been studied before. Our study of a large Dutch NBPP sample has revealed which health-care 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 have need for further information.

Declaration of conflicting interests

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Supplemental material

Supplemental material for this article is available online.

References

Alyanak B, Kilincaslan A, Kutlu L, et al. (2013) Psychological adjustment, maternal distress, and family functioning in children with obstetrical brachial plexus palsy. *The Journal of Hand Surgery* 38: 137–142.

Akel BS, Oksuz C, Oskay D, et al. (2013) Health-related quality of life in children with obstetrical brachial plexus palsy. *Quality of Life Research* 22(9): 2617–2624.

Bae DS, Waters PM and Zurakowski D (2008) Correlation of pediatric outcomes data collection instrument with measures of active movement in children with brachial plexus birth palsy. *Journal of Pediatric Orthopaedics* 28: 584–592.

Bain JR, Dematteo C, Gjertsen D, et al. (2009) Navigating the gray zone: a guideline for surgical decision making in obstetrical brachial plexus injuries. *Journal of Neurosurgery-Pediatrics* 3: 173–180.

Bialocerkowski A and Gelding B (2006) Lack of evidence of the effectiveness of primary brachial plexus surgery for infants (under the age of two years) diagnosed with obstetric brachial plexus palsy. *International Journal of Evidence-Based Healthcare* 4: 264–287.

Bialocerkowski A, Kurlowicz K, Vladusic S, et al. (2005) Effectiveness of primary conservative management for infants with obstetric brachial plexus palsy. *International Journal of Evidence-Based Healthcare* 3: 27–44.

Bunge EM, Essink-Bot ML, Kobussen MP, et al. (2005) Reliability and validity of health status measurement by the TAPQOL. *Archives of Disease in Childhood* 90: 351–358.

Chauhan SP, Blackwell SB and Ananth CV (2014) Neonatal brachial plexus palsy: incidence, prevalence, and temporal trends. *Seminars in Perinatology* 38: 210–218.

Christakou A and Laiou A (2014) Comparing the psychometric properties of the pediatric outcomes data collection instrument and the activities scales for kids: a review. *Journal of Child Health Care* 18: 207–214.

Cohen E, Berry JG, Camacho X, et al. (2012) Patterns and costs of health care use of children with medical complexity. *Pediatrics* 130: e1463–e1470.

Coroneos CJ, Voineskos SH, Coroneos MK, et al. (2015a) Obstetrical brachial plexus injury: burden in a publicly funded, universal healthcare system. *Journal of Neurosurgery-Pediatrics* 17: 222–229.

Coroneos CJ, Voineskos SH, Coroneos MK, et al. (2015b) Primary nerve repair for obstetrical brachial plexus injury: a meta-analysis. *Plastic and Reconstructive Surgery* 136: 765–779.

Dedini RD, Bagley AM, Molitor F, et al. (2008) Comparison of pediatric outcomes data collection instrument scores and range of motion before and after shoulder tendon transfers for children with brachial plexus birth palsy. *Journal of Pediatric Orthopaedics* 28: 259–264.

Duff SV and DeMatteo C (2015) Clinical assessment of the infant and child following perinatal brachial plexus injury. *Journal of Hand Therapy* 28: 126–133; quiz 134.

Evans-Jones G, Kay SP, Weindling AM, et al. (2003) Congenital brachial palsy: incidence, causes, and outcome in the United Kingdom and Republic of Ireland. *Archives of Disease in Childhood - Fetal and Neonatal Edition* 88: F185–F189.

Gunz AC, Canizares M, Mackay C, et al. (2012) Magnitude of impact and healthcare use for musculoskeletal disorders in the paediatric: a population-based study. *BMC Musculoskeletal Disorders* 13: 98.

Hale HB, Bae DS and Waters PM (2010) Current concepts in the management of brachial plexus birth palsy. *The Journal of Hand Surgery* 35: 322–331.

Holst M, Vlieland TP, van de Sande MA, et al. (2015) Translation and adaptation of the Pediatric Outcome Data Collecting Instrument (PODCI) into the Dutch language and preliminary validation in children with neonatal brachial plexus palsy. *Journal of Pediatric Rehabilitation Medicine* 8: 219–226.

Huffman GR, Bagley AM, James MA, et al. (2005) Assessment of children with brachial plexus birth palsy using the pediatric outcomes data collection instrument. *Journal of Pediatric Orthopaedics* 25: 400–404.

Lagerkvist AL, Johansson U, Johansson A, et al. (2010) Obstetric brachial plexus palsy: a prospective, population-based study of incidence, recovery, and residual impairment at 18 months of age. *Developmental Medicine & Child Neurology* 52: 529–534.

Lopez MA, Cruz AT, Kowalkowski MA, et al. (2014) Trends in hospitalizations and resource utilization for pediatric pertussis. *Hospital Pediatrics* 4: 269–275.

Louden EJ, Broering CA, Mehlman CT, et al. (2013) Meta-analysis of function after secondary shoulder surgery in neonatal brachial plexus palsy. *Journal of Pediatric Orthopaedics* 33: 656–663.

Malessy MJ, Pondaag W, Yang LJ, et al. (2011) Severe obstetric brachial plexus palsies can be identified at one month of age. *PLoS One* 6: e26193.

Pondaag W and Malessy MJ (2014) The evidence for nerve repair in obstetric brachial plexus palsy revisited. *BioMed Research International* 2014: 434619.

Pondaag W, Malessy MJ, van Dijk JG, et al. (2004) Natural history of obstetric brachial plexus palsy: a systematic review. *Developmental Medicine & Child Neurology* 46: 138–144.

Ralston SL, Harrison W, Wasserman J, et al. (2015) Hospital variation in health care utilization by children with medical complexity. *Pediatrics* 136: 860–867.

Ramos LE and Zell JP (2000) Rehabilitation program for children with brachial plexus and peripheral nerve injury. *Seminars in Pediatric Neurology* 7: 52–57.

Shah AK, Zurakowski D, Jessel RH, et al. (2006) Measuring surgeons' treatment preferences and satisfaction with nerve reconstruction techniques for children with unique brachial plexus birth palsies. *Plastic and Reconstructive Surgery* 118: 967–975.

Smania N, Berto G, La Marchina E, et al. (2012) Rehabilitation of brachial plexus injuries in adults and children. *European Journal of Physical and Rehabilitation Medicine* 48: 483–506.

Socolovsky M, Costales JR, Paez MD, et al. (2015) Obstetric brachial plexus palsy: reviewing the literature comparing the results of primary versus secondary surgery. *Childs Nervous System* 32(3): 415–425.

Strombeck C, Krumlinde-Sundholm L and Forssberg H (2000) Functional outcome at 5 years in children with obstetrical brachial plexus palsy with and without microsurgical reconstruction. *Developmental Medicine & Child Neurology* 42: 148–157.

Strombeck C, Krumlinde-Sundholm L, Remahl S, et al. (2007) Long-term follow-up of children with obstetric brachial plexus palsy I: functional aspects. *Developmental Medicine & Child Neurology* 49: 198–203.

Tse R, Kozin SH, Malessy MJ, et al. (2015) International federation of societies for surgery of the hand committee report: the role of nerve transfers in the treatment of neonatal brachial plexus palsy. *Journal of Hand Surgery* 40: 1246–1259.

van der Holst M, van der Wal CW, Wolterbeek R, et al. (2016) Outcome of secondary shoulder surgery in children with neonatal brachial plexus palsy with and without nerve surgery treatment history: a long-term follow-up study. *Journal of Rehabilitation Medicine* 48: 609–617.

van der Holst M, Vlieland TP, Meesters JJ, et al. (2015) Evaluation of shoulder function after secondary surgery in children with Neonatal Brachial Plexus Palsy. *Journal of Rehabilitation Medicine* 8: 187–196.

Waters PM (1999) Comparison of the natural history, the outcome of microsurgical repair, and the outcome of operative reconstruction in brachial plexus birth palsy. *The Journal of Bone and Joint Surgery* 81: 649–659.

Waters PM (2005) Update on management of pediatric brachial plexus palsy. *Journal of Pediatric Orthopaedics B* 14: 233–244.

Zhong W, Finnie DM, Shah ND, et al. (2015) Effect of multiple chronic diseases on health care expenditures in childhood. *Journal of Primary Care & Community Health* 6: 2–9.