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Original article

Health-related quality of life, anxiety, depression and distress of mothers and fathers of children on Home parenteral nutrition

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SUMMARY

Background & aims: Parents of children with intestinal failure, dependent on Home Parenteral Nutrition (HPN), may experience psychosocial problems due to the illness and intensive treatment of their child. Literature concerning psychosocial problems is scarce. Therefore, we aimed to investigate Health-Related Quality of Life (HRQOL), levels of anxiety, depression, distress and everyday problems of these mothers and fathers.

Methods: A multicenter study was conducted among 37 mothers and 25 fathers of 37 children on HPN (response-rate 37/49 = 76%, mean age children = 5.1 years, SD = 4.6). Parents completed three questionnaires to measure different outcomes on the KLIK website (www.hetklikt.nu): the TNO-AZL QOL Questionnaire (TAAQOL) to measure HRQOL, the Hospital Anxiety and Depression Scale (HADS) to measure anxiety and depression, and the Distress Thermometer for Parents (DT-P) to measure distress. Scores were compared to Dutch reference mothers and fathers using Mann–Whitney U-tests.

Results: No differences were found in HRQOL, measured by the TAAQOL, between HPN parents compared to the reference groups, except for the subscale 'depressive emotions' for mothers ($p = .01$) and 'daily activities' for fathers ($p = .04$). HPN mothers reported higher levels of depression compared to reference mothers ($p = .001$). In addition, HPN mothers and fathers reported higher levels of distress than reference mothers ($p = .001$) and fathers ($p = .03$). HPN mothers reported significantly more problems in the practical, emotional, cognitive and parenting domains, fathers in the social, emotional and parenting domains.

Conclusions: On HRQOL, anxiety and depression, HPN parents generally did not show much differences compared to reference parents. However, when asked about parental distress and everyday problems, HPN treatment of their child seems highly stressful for some parents and influences daily functioning. Therefore, structural screening for parental psychosocial problems in clinical practice, e.g. using the DT-P, is necessary in order to improve the well-being of both these parents and their children dependent on HPN.

Introduction

Intestinal failure, characterized by inadequate absorption of food or fluids for adequate growth, is a condition requiring the use of parenteral nutrition (PN) as long as the intestinal failure persists [1]. PN is an artificial nutritional technology whereby nutrients are administered intravenously in the hospital, and can be safely administered at home by well-trained caretakers (HPN) [2]. Because prolonged hospitalizations impair children's and families'

Abbreviations: PN, Parenteral Nutrition; HPN, Home Parenteral Nutrition; HRQOL, Health-Related Quality of Life; TAAQOL, TNO-AZL Quality Of Life Questionnaire; HADS, Hospital Anxiety and Depression Scale; DT-P, Distress Thermometer for Parents; PROMs, Patient Reported Outcome Measurements; SPSS, Statistical Package for Social Sciences; ORs, Odds Ratios.

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quality of life, an HPN program should be considered when a child needs PN for more than three months [3]. Reported prevalence of HPN varies across studies ranging from 9.6 children per million in the Netherlands to 13.7 children per million in the UK [4,5]. The parents must be educated in catheter care and the procedures necessary to connect and disconnect the parenteral nutrition to the central line. In addition, parents often are responsible for other nursing procedures, such as providing tube feeding and taking care of a stoma. This treatment is complex, of high risk for complications and will put much social and psychological pressure upon the child and parents [6].

Parents spend on average over 2 h a day on preparing and connecting the infusions. The burden on parents is high, as the majority cannot rely on someone else to connect or disconnect the parenteral nutrition [7]. In addition, hospitalizations are common due to their child's underlying disease but also complications such as the loss of vascular access or sepsis [8]. A French study showed that these hospitalizations occur on average at least twice a year [9].

In the last decade, survival rates for patients with intestinal failure have significantly improved, causing Health-Related Quality of Life (HRQOL) to be a new focus of interest [10]. Despite the large impact HPN has on the daily life of children and their families, little is known about parental HRQOL and the psychosocial consequences for these parents, and results are quite ambiguous (Table 1). Only two quantitative studies on this topic using validated questionnaires were carried out. Gottrand et al. [11] found that parental QOL, measured with the French Subjective QOL Profile questionnaire, was significantly impaired, especially for mothers. Wong et al. showed that seven of the 11 parents of children on HPN exceeded the threshold for psychiatric morbidity, and described a significant deterioration before and after the child's disease for social life, family life, sex life and work compared with controls [12]. In two qualitative studies, families with children on HPN seemed to cope well [13] and were resilient [14]. However in two other qualitative studies, mothers of children on HPN reported fear, frustration, anger and isolation [15] and nursing their child at home was a huge physical and psychological burden to parents [16].

In conclusion, only six studies were conducted [11–16]. Therefore, more quantitative, recent studies on functioning of parents, including both fathers and mothers of a child dependent on HPN are needed. It is essential to gain more insight into how these parents are functioning psychosocially in order to decide if, and which kind of help is required. Therefore, the aims of this study are to determine the 1) HRQOL, 2) degree of anxiety and depression, and 3) levels of parental distress and everyday problems in mothers and fathers of a child dependent on HPN (HPN mothers and fathers) compared to Dutch reference mothers and fathers.

Materials and methods

Participants and procedures

This study was conducted during 2012–2016 in mothers and fathers of children dependent on HPN, and undergoing treatment in the Emma Children's Hospital/Academic Medical Center Amsterdam or Sophia Children's Hospital/Erasmus Medical Center Rotterdam, the Netherlands. Inclusion criteria were, parents: 1) of children aged 0–18 years, 2) of children receiving HPN for $\geq$3 months, 3) should be able to complete Dutch questionnaires. For this study, we used data from the KLIK database. Parents were invited by a letter of the HPN team to register themselves on the KLIK website (www.hetklikt.nu) as part of standard care. KLIK is an online portal, to systematically monitor different aspects of children with various chronic diseases and their parents over time. Parents and children are asked to complete Patient Reported Outcome Measurements (PROMs) about HRQOL and psychosocial functioning one week prior to the outpatient consultation with the pediatrician or other healthcare professional [17]. Answers on the PROMs are converted into a KLIK PROfile and discussed during the consultation. In this way, communication on psychosocial topics between the patient/parent and the pediatrician is increased, and problems can be detected at an early stage [18,19]. For this study, we used the questionnaires completed for the first time on the KLIK website by the HPN parents, and only of parents who gave online informed consent for use of their data for scientific purposes. The Medical Ethics Committee of both hospitals approved the study.

Measurements

Sociodemographics and medical characteristics

Mothers or fathers completed an online questionnaire concerning their socio-demographic characteristics: age, country of

Table 1
Summary of previous studies on parents of children on HPN.

First author	Year	Short title	Country	N of participants	Questionnaire	Type of study	Main findings
Gottrand	2005	Life satisfaction in children on HPN and their families	France	68 mothers and 62 fathers	Subjective QOL Profile questionnaire	Quantitative	Parental QOL was impaired, especially for mothers. Mothers lower level of satisfaction than fathers on work, inner life, and freedom.
Wong	2000	QOL of parents of children on HPN	UK	11 parents, gender unknown	General Health Questionnaire (GHQ-28)	Quantitative	7/11 parents exceeded the threshold for psychiatric morbidity Significant deterioration before and after child's disease for social life, family life, sex life and work compared with controls. Parents more physically tired, difficulties in taking holidays, going shopping and spending time with their partners. Many felt frustrated, annoyed, stressed and having problems sleeping.
Carlsson	1997	HPN in children in Sweden	Sweden	12 parents, gender unknown	n/a	Qualitative	Families seemed to cope well. Parents were successful and skilled caregivers, and many carried on with their professional careers. However, the interviews did not cover family stress or interpersonal relation problems.
Kawakami	2013	Experiences of parents with children on HPN	Japan	5 mothers and 1 father	n/a	Qualitative	Families were resilient. Information about child's health status and care plan were important. Relationship with healthcare providers minimizes stress responses.
Silver	2004	The lived experience of HPN	USA	3 mothers	n/a	Qualitative	Mothers reported fear, frustration, anger and isolation, and intense awareness of potential adverse events related to HPN use.
Sexton	2005	Homecare packages for paediatric HPN patients	UK	20, over half were mothers	n/a	Qualitative	All the families expressed immense physical, psychological issues and burden of care.

birth, educational level, employment status, marital status, number of children living at home, and age, gender and education of their child on HPN. Information regarding underlying disease of the child, duration of PN and HPN, and number of hospital admissions in the past six months and since the start of HPN was provided by the healthcare professionals.

Health-Related Quality of Life

Parental HRQOL was assessed with the TNO-AZL Questionnaire for Adult's HRQOL (TAAQOL) [20]. This validated questionnaire measures health status problems weighted by the impact of problems on well-being on 12 scales (see Table 3). Higher scores indicate a better HRQOL (range 0–100). The psychometric properties, validity and reliability of the TAAQOL were satisfactory [20]. Cronbach's alpha values, indicating internal consistency, in the present study were moderate to good, ranging from .53–.91 for mothers and .70–1.00 for fathers. We used reference data of Dutch mothers and fathers of healthy children [21].

Anxiety and depression

Parental anxiety and depression were assessed with the Hospital Anxiety and Depression Scale (HADS) [22]. This questionnaire is divided into two 7-item scales, with answers on a four-point scale (0–3). Higher scores indicate a higher level of anxiety or depression (range 0–21). A scale score of ≥8 (cut-off score) indicates clinically significant anxiety or depression. The Dutch version of the HADS showed satisfactory validity and reliability [23]. In the present study, Cronbach's alpha values were moderate to good; .74-.78 for mothers and .64-.84 for fathers. A reference group of Dutch mothers and fathers is available [24].

Parental distress

Parental distress and everyday problems were assessed with the Distress Thermometer for Parents (DT-P) [25], a validated screening instrument frequently used in clinical practice to quickly identify distress and everyday problems in parents of children with a chronic condition. The DT-P consists of 1) a 'thermometer' ranging from 0 (no distress) to 10 (extreme distress) on which parents rate their overall distress in the past week, where a score ≥4 indicates clinically elevated distress, 2) a problem list which inquires the occurrence ('yes' or 'no') of 36 or 34 everyday problems, (depending on the age of the child; <2 years or ≥2 years, respectively) across six problem domains where problem domain scores are the sum of item scores (yes = 1, no = 0) within that problem domain, and 3) additional questions (see Table 5a) and an open question. In developing the DT-P, we distinguished the parenting problem domain into two age categories (<2 years or ≥2 years) because, based on reactions of social workers and psychologists, the problems in parenting are different for these age categories [25]. Cronbach's alpha values in the present study were moderate to good, ranging from .54–.88 for mothers and .59–.91 for fathers. A reference group of Dutch mothers and fathers of healthy children is available [26].

Statistical analyses

The Statistical Package for Social Sciences (SPSS) version 24 was used for all statistical analyses. First, socio-demographic characteristics of mothers, fathers and children were analyzed using descriptive analyses. Participants and non-participants were compared on age, gender of the child and underlying diagnosis, using an independent samples t-test and Chi-square tests, respectively.

Second, HRQOL differences between HPN mothers and fathers and mothers and fathers in the reference groups were non-parametrically analyzed using Mann–Whitney U-tests. By calculating the effect size (r), the extent of the differences between both groups was measured. An effect size of .10 was considered as small, .30 was considered as medium and .50 was considered as large [27].

Third, the proportion of HPN mothers and fathers scoring in the clinical range (score ≥8) on anxiety or depression was compared to the proportion of mothers and fathers in the reference groups scoring in the clinical range, using Chi-square tests. In addition, median levels of anxiety and depression were compared using Mann–Whitney U-tests. Odds ratios (OR) and effect sizes (r) were calculated.

Fourth, median DT-P thermometer and domain scores of HPN mothers and fathers were compared with scores of reference mothers and fathers using Mann–Whitney U-tests. Chi-square tests were used to test differences in clinical thermometer score (≥4), everyday problem scores and additional questions. Odds ratios (OR) and effect sizes (r) were calculated. Answers on the open question of the DT-P were described explorative.

Results

Socio-demographics and medical characteristics

In total, parents of 49 children dependent on HPN were eligible and 62 parents (37 mothers, 25 fathers) of 37/49 (76%) children completed one or more questionnaires (response rate AMC: 81%, Erasmus MC: 65%). Mean age of the child did not differ (p = .81) between participants (N = 37, M = 5.1 years, SD = 4.6) and non-participants (N = 12, M = 4.7 years, SD = 6.2), neither did gender (46% vs 33% female, p = .44) or underlying diagnosis. Characteristics of the participating parents and their children are shown in Table 2. Mean duration of HPN was 3.1 years (SD 2.9). Most children suffered from chronic intestinal pseudo-obstruction syndrome (32%), short bowel syndrome (22%) or intestinal obstruction due to congenital malformations (19%).

Health-Related Quality of Life

HRQOL of HPN parents did not differ significantly from HRQOL of parents of healthy children on 11 of 12 TAAQOL scales (Table 3). Only on the 'depressive emotions' scale, mothers showed a significantly lower median score compared to reference mothers (75 vs 83, p = .01, r = −.13). On the 'daily activities' scale, fathers showed a significantly lower score compared to reference fathers (88 vs 100, p = .04, r = −.21).

Anxiety and depression

The proportion of HPN mothers and fathers scoring in the clinical range (score ≥8) on anxiety or depression did not differ significantly from mothers and fathers in the reference groups (Table 4a). However, when medians were compared, HPN mothers showed significantly higher levels of depression than reference mothers (p = .001, r = −.16) (Table 4b).

Parental distress

Thermometer score

When comparing DT-P thermometer median scores, both HPN mothers (5 vs 3, p = .001) and fathers (3 vs 2, p = .03) reported significantly higher overall distress than reference mothers and fathers. Only HPN mothers reported significantly more often elevated distress (score ≥4) than mothers of healthy children (64.9% vs 42.3%, p = .007), see Table 5a.

Table 2
Socio-demographic characteristics of mothers and fathers of a child on Home Parenteral Nutrition.

	Mothers (N = 37)	Fathers (N = 25)
Age in years, M (SD), median, range	36.0 (8.2), 36.4, 23.4–55.1	40.0 (8.6), 38.9, 29.2–60.4
Born in the Netherlands, N (%)	31 (83.8%)	21 (84.0%)
Educational level, N (%)[a,b]		
Low	7 (18.9%)	0 (.0%)
Intermediate	19 (51.4%)	14 (58.3%)
High	11 (29.7%)	10 (41.7%)
Paid employment, N (%)	17 (45.9%)	23 (92.0%)
Marital status, N (%) [b]		
Married/living together	35 (94.6%)	24 (100.0%)
Single/separated	2 (5.4%)	0 (.0%)
Children living at home, N (%)		
1	12 (32.4%)	7 (28.0%)
2	14 (37.8%)	11 (44.0%)
≥ 3	11 (29.8%)	7 (28.0%)
	Children on HPN (N=37)	
Age in years, M (SD), median, range	5.1 (4.6), 3.6, .3–17.4	
Female gender, N (%)	17 (45.9%)	
Undergoing treatment in, N (%)		
Amsterdam (AMC)	26 (70.3%)	
Rotterdam (Erasmus MC)	11 (29.7%)	
Duration of PN in years, M (SD), median, range	3.5 (3.0), 3.1, .2–10.2	
Duration of HPN in years, M (SD), median, range	3.1 (2.9), 2.6, .1–10.0	
Underlying disease, N (%)		
Chronic intestinal pseudo-obstruction syndrome	12 (32%)	
Short bowel syndrome	8 (22%)	
Intestinal obstruction due to congenital malformations	7 (19%)	
Microvillous inclusion disease	4 (11%)	
Chronic intractable diarrhea	4 (11%)	
Hirschsprung's disease	1 (3%)	
Severe failure to thrive	1 (3%)	
Number of admissions in past 6 months, M (SD), median, range [c]	1.1 (.9), 1, 0-3	
Number of admissions since start HPN, M (SD), median, range [d]	8.3 (10.2), 2, 0-36	
Education, N (%)		
None (yet)	15 (40.5%)	
Daycare	6 (16.2%)	
Regular primary school	8 (21.6%)	
Special primary school	5 (13.5%)	
Regular secondary school	2 (5.4%)	
Special secondary school	1 (2.7%)	

[a] Highest educational level completed. Low: primary education, lower vocational education, lower or middle general secondary education; Intermediate: middle vocational education, higher secondary education, pre-university education; High: higher vocational education, university.
[b] Score of 1 father missing.
[c] Scores of 4 children missing.
[d] Scores of 3 children missing.

Table 3
HRQOL (according to the TAAQOL) of mothers and fathers of children on HPN compared with reference groups of mothers and fathers of healthy children.

	HPN mothers			Reference mothers					HPN fathers			Reference fathers				
HRQOL	N	Med	IQR	N	Med	IQR	*P*	r	N	Med	IQR	N	Med	IQR	*p*	r
Gross motoric functioning	35	100	88–100	359	100	81–100	.18	−.07	24	100	100–100	73	100	100–100	.96	−.01
Fine motoric functioning	35	100	100–100	358	100	100–100	.08	−.09	24	100	100–100	73	100	100–100	.96	.00
Cognitive functioning	35	88	63–100	358	88	63–100	.49	−.03	24	94	66–100	72	97	75–100	.52	−.07
Sleep	35	63	44–94	360	75	50–94	.25	−.06	24	84	56–100	72	81	56–100	.86	−.02
Pain	35	81	63–94	361	75	56–88	.12	−.08	24	88	64–100	72	81	64–100	.67	−.04
Social functioning	35	88	69–100	356	88	75–100	.47	−.04	24	84	52–100	73	94	75–100	.13	−.16
Daily activities	35	100	63–100	355	94	75–100	.79	−.01	24	88	58–100	73	100	88–100	**.04**	−.21
Sexuality	35	100	88–100	341	100	75–100	.20	−.07	24	100	50–100	73	100	69–100	.67	−.04
Vitality	35	58	33–75	360	67	46–75	.09	−.08	24	67	42–75	73	75	58–83	.08	−.18
Positive emotions	35	67	42–75	359	67	58–75	.53	−.03	24	67	60–73	73	67	58–75	.82	−.02
Depressive emotions	35	75	67–83	360	83	67–92	**.010**	−.13	24	83	75–92	73	83	75–92	.21	−.13
Aggressive emotions	35	89	78–100	358	89	78–100	.39	−.04	24	89	89–100	73	100	89–100	.40	−.08

Higher scores indicate a higher HRQOL.
P-values according to Mann Whitney U-tests. Significant differences at p < .05 are presented in bold.
HRQOL=Health-Related Quality of Life; HPN=Home Parenteral Nutrition; TAAQOL = TNO-AZL Questionnaire for Adult's HRQOL, Med = Median, IQR = interquartile range.

Problem domain scores

HPN mothers reported a significantly higher total problem score than mothers of healthy children (6 vs 4, p = .006). HPN fathers did not differ significantly on total problem score (6 vs 2, p = .13). Both HPN mothers and fathers reported more problems on 4 out of 7 problem domains, compared to mothers and fathers of healthy children; mothers reported more problems in the practical, emotional, cognitive and parenting (≥2 years) domain, fathers

Table 4a
Clinical scores of anxiety and depression: score of ≥8 in HPN mothers and fathers compared with reference groups of mothers and fathers: percentages and Odds Ratio (OR).

	Mothers							Fathers						
	HPN (N = 34)		Reference (N = 368)					HPN (N = 24)		Reference (N = 368)				
	N	%	N	%	*p*	OR	95% CI of OR	N	%	N	%	*p*	OR	95% CI of OR
Anxiety	9	26.5	76	20.7	.43	1.39	.62–3.13	2	8.3	64	17.4	.40*	.43	.10–1.89
Depression	5	14.7	44	12.0	.64	1.27	.47–3.45	4	16.7	56	15.2	.77*	1.11	.37–3.33

Differences according to Chi-square tests.
CI = Confidence Interval.
* = Fishers Exact (N < 5 in one cell).

Table 4b
Median scores of anxiety and depression in HPN mothers and fathers compared with reference groups of mothers and fathers.

	Mothers						Fathers					
	HPN (N = 34)		Reference (N = 368)				HPN (N = 24)		Reference (N = 368)			
	Median	IQR	Median	IQR	*p*	Effect size r	Median	IQR	Median	IQR	*p*	Effect size r
Anxiety	4	3–8	4	2–7	.39	-.04	4	2–5.75	3	1–6	.64	−.02
Depression	4	2–6.25	2	1–5	**.001**	-.16	4	1–6	2	1–6	.56	−.03

Higher scores represent higher levels of anxiety and depression.
Differences calculated with Mann–Whitney U test.
Significant differences at p < .05 are presented in bold.
IQR = interquartile range.

Table 5a
DT-P thermometer score, problem domain scores and additional questions in HPN mothers and fathers compared with reference groups of mothers and fathers of healthy children.

	Mothers					Fathers				
	HPN (N = 37)	Reference (N = 671)	*p*	r/OR	95% CI	HPN (N = 24)	Reference (N = 463)	*p*	r/OR	95% CI
Thermometer score										
Median (IQR)	5 (3–7)	3 (1–6)	**.001**	-.13		3 (2–7)	2 (1–5)	**.03**	-.10	
Clinical, %	64.9	42.3	**.007**	2.50	1.27–5.00	45.8	32.2	.17	1.79	.78–4.00
Total problem score, Med (IQR)[a]	6 (3–12.5)	4 (1–8)	**.006**	−.10		6 (1–9)	2 (1–5)	.13	-.07	
Practical problems, Med (IQR)	1 (0–3)	1 (0–2)	**.04**	−.08		1 (0–2)	0 (0–1)	.12	-.07	
Social problems, Med (IQR)	0 (0–1.5)	0 (0–1)	.09	−.06		0 (0–1)	0 (0–0)	**.002**	-.14	
Emotional problems, Med (IQR)	2 (.5–5)	1 (0–3)	**.005**	−.10		2 (0–4)	0 (0–2)	**.02**	-.11	
Physical problems, Med (IQR)	2 (1–3)	2 (0–3)	.42	−.04		1 (0–2)	1 (0–2)	.78	-.01	
Cognitive problems, Med (IQR)	0 (0–2)	0 (0–1)	**.02**	−.10		0 (0–1)	0 (0–0)	.14	-.07	
Parenting problems ≥2, Med (IQR) [b]	1 (0–2.5)	0 (0–0)	**.001**	−.21		1 (0–2)	0 (0–0)	**.000**	-.22	
Parenting problems <2, Med (IQR) [c]	0 (0–2)	0 (0–1)	.44	−.05		2 (0–2)	0 (0–1)	**.02**	-.24	
Additional questions										
Enough support from surroundings, %	91.1	92.1	1.00*	.97	.29–3.23	87.5	93.3	.23	.50	.14–1.79
People react with a lack of understanding, %	18.9	11.3	.16	1.82	.78–4.35	20.8	10.2	.10	2.33	.83–6.67
Parental chronic illness	10.8	20.3	.20	.48	.17–1.37	25.0	14.0	.14	2.04	.78–5.26
Would like to talk to a professional about situation - Yes/Maybe, %	40.5	17.1	**.000**	3.33	1.67–6.67	33.3	12.5	**.004**	3.45	1.43–8.33

Significant differences at p < .05 are presented in bold.
Domain scores: Mann–Whitney U-tests with effect sizes r, item scores: Chi^2 tests with OR and 95% CI.
* = Fishers Exact (<N = 5 in one cell).
[a] Total problem score = the sum of item scores (yes = 1, no = 0) within 5 problem domains (practical, social, emotional, physical and cognitive).
[b] N = 25 HPN mothers, N = 560 reference mothers, N = 17 HPN fathers, N = 370 reference fathers.
[c] N = 12 HPN mothers, N = 111 reference mothers, N = 7 HPN fathers, N = 93 reference fathers.

reported more problems in the social, emotional, parenting (≥2 years) and parenting (<2 years) domain. Effect sizes (*r*) ranged from −.08 to −.24.

Everyday problem scores

In total, mothers reported significantly more problems on 15/34 everyday problems when their child was ≥2 years, or 12/36 everyday problems when their child was <2 years (Table 5b). HPN mothers reported significantly less problems about work/study than mothers of a healthy child (8.1% vs 25.3%, p = .02). Fathers reported significantly more problems on 9/34 everyday problems when their child was ≥2 years, or 8/36 everyday problems when their child was <2 years. Everyday problems on which both HPN mothers and fathers reported significantly more problems were: child care, leisure time, dealing with friends, interacting with child (ren), loneliness, concentration, independence of child, and following advice about treatment/giving medication.

Additional questions

When asked about support, people reacting with a lack of understanding, and parental chronic illness, HPN parents did not differ significantly from reference parents (Table 5a). HPN parents

Table 5b
DT-P everyday problem scores of HPN mothers and fathers compared with reference groups of mothers and fathers of healthy children.

	Mothers					Fathers				
	HPN (N = 37)	Reference (N = 671)	*p*	OR	95% CI	HPN (N = 24)	Reference (N = 463)	*p*	OR	95% CI
Practical problems										
Housing, %	10.8	5.5	2.63*	2.08	.70–6.25	4.2	3.7	.60*	1.14	.15–9.09
Work/study, %	8.1	25.3	**.02***	.26	.08–.85	25.0	25.9	.92	.95	.37–2.44
Finances/insurance, %	10.8	16.7	.49*	.61	.21–1.75	4.2	14.5	.23*	.26	.03–1.92
Housekeeping, %	35.1	21.6	.05	1.96	.98–4.00	20.8	12.1	.21	1.92	.68–5.26
Transport, %	8.1	4.6	.41*	1.82	.53–6.25	.0	3.9	1.00*	[c]	
Child care/child supervision, %	32.4	10.1	**.000**	4.17	2.04–9.09	16.7	5.4	**.047***	3.45	1.11–11.11
Leisure activities/relaxing, %	45.9	22.4	**.001**	2.94	1.52–5.88	54.2	14.9	**.000**	6.67	2.94–16.67
Social problems										
Dealing with (ex)partner, %	16.2	12.4	.49	1.37	.56–3.33	16.7	11.7	.51*	1.52	.50–4.55
Dealing with family, %	16.2	10.9	.32	1.59	.64–3.85	4.2	6.7	1.00*	.61	.08–4.55
Dealing with friends, %	13.5	3.7	**.004**	4.00	1.45–11.11	29.2	1.5	**.000**	25.00	8.33–100.00
Interacting with your child (ren), %	24.3	11.8	**.02**	2.38	1.10–5.26	25.0	7.8	**.003**	4.00	1.47–11.11
Emotional problems										
Controlling emotions, %	43.2	27.4	**.04**	2.00	1.03–4.00	25.0	11.9	.06	2.50	.94–6.67
Self-confidence, %	24.3	22.7	.81	1.10	.51–2.38	8.3	12.7	.76*	.62	.14–2.70
Fears, %	18.9	10.7	.12	1.92	.82–4.55	16.7	6.5	.08*	2.86	.93–9.09
Depression, %	43.2	31.9	.15	1.61	.83–3.23	45.8	22.2	**.008**	2.94	1.28–6.67
Feeling tense or nervous, %	54.1	36.1	**.03**	2.08	1.08–4.00	41.7	26.3	.10	2.00	.86–4.55
Loneliness, %	21.6	7.7	**.003**	3.23	1.43–7.69	20.8	3.7	**.000**	6.67	2.33–20.00
Feelings of guilt, %	24.3	17.4	.29	1.52	.70–3.33	16.7	7.3	.11*	2.50	.81–7.69
Use of substances (e.g. alcohol, drugs and/or medication), %	5.4	2.7	.28*	2.08	.46–9.09	8.3	3.0	.18*	2.94	.63–14.29
Intrusive/recurrent thoughts about a specific event, %	43.2	20.4	**.001**	2.94	1.52–5.88	25.0	13.8	.13	2.08	.79–5.56
Physical problems										
Eating, %	5.4	12.4	.30*	.40	.10–1.72	.0	4.8	.62*	[c]	
Weight, %	27.0	26.2	.92	1.04	.50–2.17	4.2	16.6	.15*	.22	.03–1.64
Sleep, %	37.8	29.7	.29	1.45	.73–2.86	25.0	21.4	.67	1.22	.47–3.13
Fatigue, %	73.0	55.7	**.04**	2.13	1.02–4.55	50.0	44.1	.57	1.27	.56–2.86
Out of shape/condition, %	27.0	20.9	.37	1.41	.66–2.94	25.0	19.0	.47	1.43	.55–3.70
Pain, %	24.3	24.3	.99	1.00	.46–2.17	4.2	18.1	.10*	.20	.03–1.47
Sexuality, %	8.1	10.6	.79*	.75	.22–2.50	8.3	8.9	1.00*	.93	.21–4.17
Cognitive problems										
Concentration, %	35.1	17.9	**.009**	2.50	1.23–5.00	29.2	11.2	**.009**	3.23	1.28–8.33
Memory, %	40.5	22.4	**.01**	2.38	1.20–4.76	20.8	13.6	.32	1.67	.60–4.55
Parenting problems ≥2 [a]										
Dealing with your child, %	8.0	10.9	1.00*	.02	.16–3.13	11.8	9.7	.68*	1.23	.27–5.56
Dealing with the feelings of your child, %	44.0	9.3	**.000**	7.69	3.33–16.67	17.6	8.6	.19*	2.27	.62–8.33
Talking about the disease/consequences with your child, %	20.0	3.0	**.000**	7.69	2.70–25.00	11.8	2.7	.09*	4.76	.96–25.00
Independence of your child, %	48.0	7.5	**.000**	11.11	4.76–25.00	23.5	7.6	**.04***	3.70	1.15–12.50
Following advice about treatment/giving medication, %	20.0	3.4	**.000**	7.14	2.44–20.00	41.2	3.0	**.000**	25.00	7.14–100.00
Parenting problems <2 [b]										
Feeling connected with your child	.0	1.8	1.00*	[c]		14.3	1.1	.14*	14.29	.85–250
Caring for your child	16.7	1.8	**.047***	11.11	1.39–100.00	.0	3.2	1.00*	[c]	
Feeding your child	25.0	17.1	.45	1.61	.40–6.67	42.9	9.7	**.04***	7.14	1.35–33.33
Development of your child	8.3	7.2	1.00*	1.18	.13–10.00	28.6	4.3	.06*	9.09	1.30–50.00
Following advice about treatment/giving medication, %	8.3	2.7	.34*	3.23	.31–33.33	14.3	1.1	.14*	14.29	.85–250
Your child's sleeping	33.3	23.4	.48*	1.64	.45–5.88	28.6	20.4	.64*	1.56	.28–8.33
Behavior/crying of your child	16.7	16.2	1.00*	1.03	.21–5.00	14.3	17.2	1.00*	.08	.09–7.14

Significant differences at $p < .05$ are presented in bold.
Item scores: Chi-square tests with OR and 95% CI.
* = Fishers Exact (<N = 5 in one cell).
[a] N = 25 HPN mothers, N = 560 reference mothers, N = 17 HPN fathers, N = 370 reference fathers.
[b] N = 12 HPN mothers, N = 111 reference mothers, N = 7 HPN fathers, N = 93 reference fathers.
[c] When N = 0 in HPN parents, OR cannot be calculated.

did indicate more often than reference parents (mothers: 40.5% vs 17.1%, $p < .001$, fathers: 33.3% vs 12.5%, $p = .004$) a possible wish to talk to a professional about their situation. About one third of HPN parents answered the open question (results not shown). The topics they brought up were very diverse; about the next appointment, financial issues regarding the care of their child on HPN, work, medical staff, emotional issues, and the wish to talk to a psychologist.

Discussion

This study compared mothers and fathers of children dependent on HPN with reference parents. On most subscales of the HRQOL questionnaire, no significant differences were found between both groups. Fathers only reported more problems on the subscale daily activities, and mothers on the subscale depressive emotions. On the anxiety and depression questionnaire, HPN mothers also showed

higher mean levels of depression. However, when comparing proportions of mothers and fathers scoring in the clinical range, no significant differences were found between HPN parents and reference parents. Regarding overall parental distress, both HPN mothers and fathers did report significantly higher levels than reference parents.

An unexpected result of our study is that only small differences were found on HRQOL, with quite small effect sizes. A large study in parents of children with various chronic conditions, such as asthma or metabolic disease, using the same questionnaire and reference group, did show lower scores on HRQOL [21]. An explanation might be that, for the HPN parents, practical everyday problems are more prominent, as shown in the DT-P results, rather than general HRQOL. Parents might also adjust their expectations after taking care of a child with a chronic illness for a long time. Qualitative research might give more insight into these processes. Similar results were found in a recent study in parents of children with Down's syndrome, implying that asking about distress in general is not sufficient and healthcare professionals should address or ask about specific problems [28].

Specific problems that were reported by mothers as well as by fathers, e.g. dealing with friends, may represent relevant everyday problems in these families. This example is consistent with earlier findings [7], where HPN parents did not differ from the normal population on overall social interaction, but showed a different pattern in the subscales. Superficial and simple social contacts were high, whereas deeper, emotional relations were affected. In addition, in the current study, HPN parents reported problems with the independence of their child, loneliness and leisure time. This corresponds with results of another qualitative study in HPN patients and their parents, where isolation, e.g. finding a source of support, was a major theme [15], and also with the results of another Dutch study, which showed that parents of chronically ill children spent less time doing leisure activities [29]. Although HPN parents in this study did not report more problems with their (ex)partner than reference parents, and the vast majority of the parents is married/living together, it is important to pay attention to the quality of the partner relation in clinical practice. This is shown in research within parents of children with Down's syndrome, where fathers also did not report more problems with their (ex)partner, measured with the same DT-P questionnaire [28], but the same fathers did indicate a worse partner relation (marital satisfaction, conflict-management, support and trust) on another questionnaire [30]. Furthermore, our findings reflect the results of a small survey [31], which indicated that almost all HPN families experienced great difficulty trying to find a person to look after their child and going out. Sleep disturbance was common for parents, as they had to attend to the equipment and their child at night. Many families experienced deterioration in their family life, i.e. social activities, and overall quality of life after their child started HPN. Although families coped well with HPN, they frequently suffered from social isolation [31].

A striking finding is that HPN mothers report less problems regarding work/study than reference mothers. An explanation may be that the majority of the HPN mothers in our cohort do not have paid employment (Table 2), consistent with the earlier finding that mothers of chronically ill children work fewer hours a week [29]. However, it may also reflect a tendency to choose for a less demanding career, to combine work with the care for their child, as found in a previous study on parents of children with Down's syndrome [32].

Previous research showed that fear, frustration, and anger were frequently expressed negative feelings of HPN mothers [15]. The initial experience of being discharged with a child receiving PN was terrifying. However, HPN was seen as lifesaving and miraculous. Mothers expressed being overwhelmed with so much to learn at initiation of HPN. This does not reflect our findings, since both on the anxiety scale of the HADS as the item 'fears' on the DT-P, parents did not show significantly elevated scores compared to reference parents. This could be explained by the fact that children of the parents in this study were receiving HPN on average for over 3 years already, and the initial stress after the start of HPN might be reduced. This also reflects previous findings, where longer time intervals since hospitalization were associated with lower parental stress levels [10].

The current study is the first that systematically inquired about HRQOL, anxiety, depression, distress and everyday problems in both HPN mothers and fathers, and used reference groups. Although the number of participants might seem small, almost all parents of children on HPN in the Netherlands were approached to participate, as the centers in Amsterdam and Rotterdam treat the majority of Dutch pediatric HPN patients. Although participants and non-participants could not be compared on all characteristics, with the high response rate, our results can be seen as representative for the Dutch population of HPN parents.

Our study does, however, have some limitations. In this study, parents completed the questionnaires on the KLIK website in order to discuss the answers with the healthcare professional, who could see the answers, during the outpatient consultation. This is part of standard care to identify problems at an early stage and to see how parents and patients are doing longitudinally. Parents in all three reference groups completed the questionnaires anonymously. The effect of the difference between anonymous and non-anonymised responses is unknown. One might hypothesize that parents who do not complete questionnaires anonymously, report less problems than they in reality experience, because some parents do not want their answers to be discussed, or social acceptability is at stake. In that case, the difference in this study between HPN parents and reference groups might be bigger. Another limitation is that we do not know whether parents in our study already received psychosocial help. Therefore, the question about wanting to talk to a professional needs to be interpreted with caution.

Future research could be directed at potential risk- and protective factors of psychosocial functioning, e.g. socio-economic, medical factors (such as number of nights the child is on HPN), coping and social support, in order to detect and support the parents who are at risk for impaired psychosocial functioning at an early stage. Also, longitudinal studies to follow these parents would be interesting, for example to see if most parents are still married/living together and how this may affect their psychosocial wellbeing. Additionally, to be able to compare outcomes on how these parents are doing, the use of the same outcomes and questionnaires is needed in international studies. In addition, qualitative research, using semi-structured interviews, can give more insight into the (HPN) specific problems parents may experience, not captured by the three questionnaires in this study.

In conclusion, although the HRQOL of HPN parents is only affected on one subscale of the TAAQOL for mothers, and on one for fathers, some parents experience high levels of parental distress and everyday problems. The results stress the importance of implementing structured parental psychosocial screening in daily clinical practice. In the Netherlands, HPN mothers and fathers complete the DT-P on the KLIK website once a year. Therefore, annual screening of Dutch HPN parents is warranted. The aim of using a screening questionnaire such as the DT-P is an early identification of psychosocial problems. Parents can also use this questionnaire to ask the multidisciplinary team for additional practical or psychosocial support. After identifying which parents want and need support, referrals to a social worker or psychologist should be made and tailor-made interventions could be provided.

Practically, it is possible to receive homecare, including overnight support, for Dutch children dependent on HPN and their families. Parents receive a personal budget from the health insurance company, to hire nurses for homecare or other assistance, for example support at the school of their child. Psychosocially, interventions aimed at reducing depressive symptoms could be offered. A previous review also recommends that individual healthcare plans should include a strong psychosocial component to assess ongoing family needs [31]. Families must be encouraged to identify what kind of help they require [31]. With structural screening of parental psychosocial problems, the well-being of both these parents and their children dependent on HPN can be improved.

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Conflict of interest

The authors have indicated they have no potential conflicts of interest to disclose.

Statement of authorship

Drs. van Oers, Dr. Haverman, Prof. Grootenhuis and Dr. Tabbers conceptualized and designed the study. Drs. van Oers, Dr. Olieman, Drs. Neelis, Mrs. Jonkers-Schuitema, and dr. Tabbers performed the data collection. Drs. van Oers and dr. Haverman performed the statistical analysis. The first draft of the manuscript was written by drs. van Oers. Dr. Haverman, Prof. Grootenhuis and Dr. Tabbers handled the supervision.

All authors made critical revisions to the manuscript. Each author listed takes full responsibility for the manuscript and approved submission of the manuscript in its present form.

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