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Original article

Patients, family members and healthcare professionals' top ten research priorities for adults receiving home parenteral nutrition for malignant or benign disease



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SUMMARY

Background & aims: Home parenteral nutrition (HPN) is the primary treatment for chronic intestinal failure (CIF) due to non-malignant disease and is increasingly used in patients with a diagnosis of cancer. This project engaged with patients, family members and healthcare professionals to ascertain what questions they want researched.

Methods: This study followed the five-stage process of the James Lind Alliance that involved (1) setting up a steering group, (2) carrying out an initial survey to gather participants' questions, (3) data processing, (4) an interim priority setting survey and (5) final priority setting workshop. Surveys were translated and back translated into Italian, Danish and French.

Results: The project was delivered by an international steering committee with representation from Denmark, Italy, the United Kingdom and United States consisting of three patients, six healthcare professionals and facilitated by University researchers. For the first survey, 633 questions were submitted by 292 respondents from 12 countries. There were 79 questions removed as out of scope or already in the published literature. Responses were collated into two interim surveys of 41 questions for benign CIF and 13 questions for HPN and cancer. In the second survey, 216 respondents prioritised their top ten questions. The ordering from the cancer and HPN survey was taken as definitive; top priorities were quality of life, survival, when to commence HPN, using HPN with anti-cancer treatments, access barriers, measuring benefit and ethical implications. For CIF with benign disease, 18 questions were discussed in two workshops attended by 13 patients and 7 healthcare professionals. The questions were ranked using a modified nominal group technique; the top research priorities were prevention and treatment of liver disease, improving central infusion lines, oral absorption, avoiding long-term negative consequences, vascular access, side effects, line infections, decreasing stoma output, quality of life and sleep.

Conclusions: Priorities identified will assist researchers to focus on research questions important to patients, family members and healthcare professionals.

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1. Introduction

Chronic intestinal failure (CIF) is defined as the “reduction of gut function below the minimum necessary for the absorption of macronutrients and/or water and electrolytes, such that intravenous supplementation is required to maintain health and/or growth” (1). Home parenteral nutrition (HPN) is the primary treatment for those with CIF due to benign disease and is increasingly used in patients with a cancer diagnosis [1,2]. The life-saving role of HPN for patients with CIF and its supporting role for the treatment of cancer patients demands continuous investigation efforts.

The research agenda has been traditionally driven by researchers, industry and to a certain extent, healthcare professionals [3,4]. Internationally, there has been a move away from this approach to involving patients and the public with organisations such as Patient-Centered Outcomes Research Institute in the United States, INVOLVE in the UK and Strategy for Patients-Oriented Research in Canada supporting this process [5–7]. Patient and public involvement has been defined as “research being carried out ‘with’ or ‘by’ members of the public (including patients and carers) rather than ‘to’, ‘about’ or ‘for’ them” [8]. This involvement leads to better research with improved recruitment and retention rates and ultimately results in improved patient satisfaction, health outcomes and reduced costs [9–11]. Some research into HPN has included patients in the management of the research and interviewing patients for development of questionnaires measuring quality of life [12–15]. However, there has been limited engagement of patients in setting the research agenda [16].

The James Lind Alliance has facilitated the inclusion of patients, family members and healthcare professionals in setting research agendas by enabling partnerships to be formed between these groups and researchers [17]. They have a standard procedure for investigating what patients and healthcare professionals think should be researched for particular diseases or conditions [17]. This procedure involves having these groups represented on the steering committee so that patients and healthcare professionals manage the research process.

This project investigated the research agenda of patients receiving HPN, their family members and healthcare professionals. The James Lind Alliance methodology informed this process. The aim was to determine the questions that patients, family members and healthcare professionals want answered by research around HPN for CIF due to benign disease and also HPN used in people with cancer.

1.1. Methods

This project employed the five stages of the James Lind Alliance, which are setting up the steering group, gathering the questions participants want researched, data processing, interim priority setting, and final priority setting (see Fig. 1) [18].

1.1.1. The steering group

An international steering group was formed comprised of patient representatives and healthcare professionals working with patients receiving HPN and was facilitated by university researchers (AMS, DJ and CF). The steering group managed the process of determining the research priorities of patients, family members and healthcare professionals.

1.1.2. Gathering research questions

Four initial surveys were written following the guidance of the James Lind Alliance and informed by other surveys published on their website. The surveys included two aimed at patients over 18 years receiving HPN and their family members; one for people with

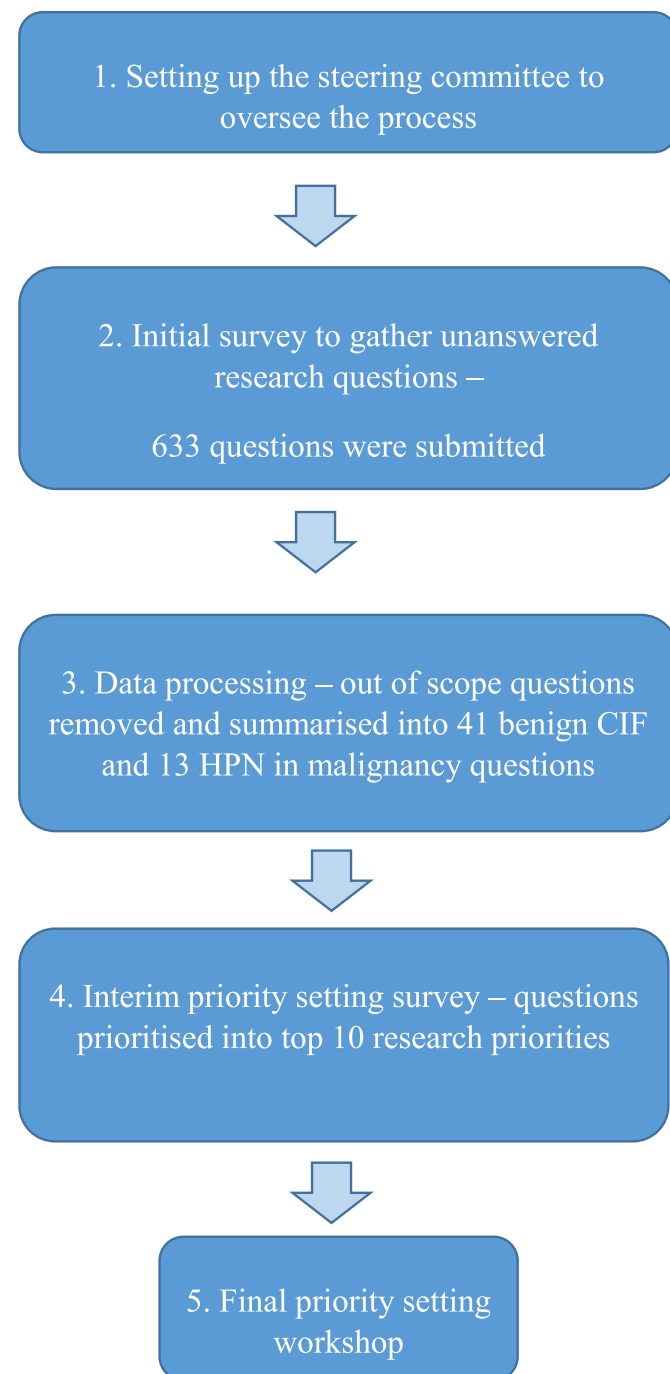


Fig. 1. The five stage process of the project.

CIF due to benign disease and the other for people with cancer. The other two surveys were aimed at healthcare professionals treating these patients. The surveys were translated from English into Italian, Danish, and French and back translated to ensure accuracy (English versions are shown in supplementary S1). The steering committee discussed and agreed the surveys before distribution. These surveys asked for ‘uncertainties’; that is questions or topics that need answering by research. The surveys were promoted through patient support groups; PINNT, Support and advocacy for people on home artificial nutrition support in the UK, Transplant Unwrapped in the USA and the parenteral nutrition patient’s association in Denmark, healthcare professional networks; British

Association for Parenteral and Enteral Nutrition (BAPEN), British Dietetic Association Oncology Specialist Group, sent to members of the Home Artificial Nutrition and Chronic Intestinal Failure Special Interest Group of the European Society for Clinical Nutrition and Metabolism and promoted on social media channels; LinkedIn, in appropriate Facebook groups and on Twitter. In addition, members of the steering committee sent the survey to healthcare professionals, and patient representatives contacted patients and family members known personally to them.

1.1.3. Data processing

Responses from the Italian, Danish and French surveys were translated into English. The responses from all the surveys were collated, categorised into themes and formed into questions by the university researchers. Any out of scope responses were removed. The three researchers met weekly to discuss any discrepancies and agree questions. A long list of questions was compiled and duplicate questions removed. The steering committee checked potential questions, and highlighted any that had already been answered by research. These questions were checked against the published literature and removed.

The submitted questions within the same theme were collected and written as summary questions where possible. The steering committee members subsequently checked and agreed the reformulated summary questions.

1.1.4. Interim priority setting

The summary questions were separated into those relating to benign CIF and those pertaining to cancer and HPN and two surveys were created. The surveys were translated into Danish and Italian. Patients, family members and healthcare professionals were asked to choose their top ten questions and prioritise them in order of importance. To prioritise their top 10, participants were asked to rank the questions from 1 to 10, with 1 being the most important. Respondents were invited to take part in the second survey regardless of whether they had participated in the initial survey. This survey was publicised using the same channels as the first survey.

1.1.5. Final priority setting

The questions that were ranked the highest by participants from the interim survey for CIF due to benign disease were taken forward to a workshop of patients and healthcare professionals. The workshop was advertised in the second survey and participants could provide their contact details if they were interested in taking part. The top ten priorities were decided at the workshop using a modified nominal group technique [19]. Questions were sent out in advance to participants, who were asked to rank them in order of importance. The workshop was held on-line due to the COVID-19 pandemic and so that participants from around the world could be included.

The workshop consisted of an initial introduction followed by small break-out groups. In the break-out groups all participants had the opportunity to give their opinion and discuss the ordering of questions. The opinions of patients and healthcare professionals were treated with equal weight. There were two rounds of ranking with the makeup of the groups being mixed up between the first and second round of ranking. The order of the top ten priorities was created from collating the ordering of the different break-out groups.

1.1.6. Ethical approval

This project was patient and public engagement as determined by NHS Health Research Authority Decision Tool and, as such, did not need formal ethical approval. However, participants' data were handled according to the General Data Protection Regulations and University of Manchester data handling policy. Participants filled in

the surveys anonymously, unless they wanted to have the results emailed to them or if they wanted details about taking part in the workshop. Participants' personal data were only used for the purpose of this project.

2. Results

2.1. Setting up the steering committee

The project steering committee was convened in March 2021 and was a mix of patients and healthcare professionals with representation from different countries. It consisted of three patients representing patient organisations in the UK, Denmark and the USA, two gastroenterologists – one from the UK and one from Italy, two dietitians, one nurse and one oncologist. The members of the committee were chosen to represent perspectives relevant to the project: three consultants specialising in benign CIF and HPN in cancer, three other healthcare professionals (a nurse and dietitians), and three patients. The project was facilitated by three University researchers from the UK.

2.2. The priority setting process

2.2.1. Gathering research questions

The first survey ran from April to June 2021. This resulted in 292 questionnaires containing 633 questions being submitted from participants in 12 countries with 69% coming from those with lived

Table 1
Demographic details of population.

	First survey n (%)	Second Survey n (%)
Gender		
Female	190 (65)	109 (50)
Male	94 (32)	46 (21)
Other	2 (1)	0
Prefer not to say	5 (2)	1 (1)
Missing	1 (0.3)	60 (28)
Age		
<20	3 (1)	4 (2)
20–29	28 (10)	17 (8)
30–49	107 (37)	88 (40)
50–69	115 (39)	60 (28)
70–79	31 (11)	18 (8)
80+	2 (1)	2 (1)
Prefer not to say	3 (1)	2 (1)
Missing	3 (1)	25 (12)
Participants		
Patient benign	156 (53)	76 (35)
Past patient benign	4 (1)	7 (3)
Patient cancer	12 (4)	2 (0.9)
Family member	31 (11)	37 (17)
HCP	87 (30)	69 (32)
Missing	2 (1)	25 (13)
Country		
Australia	10 (3)	0
Canada	1 (0.3)	3 (1)
Denmark	33 (11)	11 (5)
France	2 (1)	1 (0.5)
Germany	2 (1)	1 (0.5)
Holland	2 (1)	0
Israel	1 (0.3)	0
Italy	41 (14)	42 (19)
Ireland	0	1 (0.5)
Portugal	1 (0.3)	0
Switzerland	1 (0.3)	0
United Kingdom	150 (51)	96 (44)
United States of America	47 (16)	33 (15)
Missing	1 (0.3)	28 (13)
Total	292	216

n – number HCP – healthcare professional.

Table 2
Themes and number of questions submitted by respondents.

Themes	Number of questions submitted by respondents
Absorption of nutrients	11
Adverse events	38
Blood sugar	5
Caloric variation between days	3
Cancer	75
Causes of IF	35
Contents of PN bags	22
Disrupted sleep	9
Doctor/HCP awareness	11
Drug issues in SBS	8
Environment	5
Exercise	2
Finances	3
Further conditions	25
Gut adaption	5
Infections	41
Infusion rates	41
Lines	22
Liver	26
Managing daily life	16
Multi-chamber bags	13
Mental Health	11
Monitoring	16
More freedom	4
Muscle mass	3
Patient education	40
Pregnancy	2
Pumps	15
Quality of life	18
Support	21
Medications	2
Vascular	6

HCP – healthcare professional, IF – intestinal failure, PN – parenteral nutrition, SBS – short bowel syndrome.

experience and 30% from healthcare professionals. Further demographic details of the respondents are reported in Table 1.

2.3. Organising research questions

Of the 633 questions submitted, 71 questions were deemed out of scope and 8 questions were already addressed in the published

Table 3
The top 13 cancer and home parenteral nutrition priorities and number of participants who selected each question.

Number of participants selected question				
Rank	Pt	HCP	Unknown	
1	2	24	2	Does PN improve quality of life for patients with cancer?
2	1	22	1	Does early intervention with PN in cancer patients at risk of inoperable malignant bowel obstruction improve quality of life and extend survival?
3	2	21	2	How do we define the most appropriate time to commence PN in patients with advanced cancer?
4	0	16	1	When should PN be used concurrently with active anti-cancer treatments (eg. Radiotherapy/ chemotherapy) and does this improve treatment outcomes?
5	2	23	2	What are the barriers to accessing PN for patients with advanced cancer and how can these be overcome?
6	2	21	3	Does PN improve survival for patients with advanced cancer?
7	2	21	2	Can fast-track HPN protocols be used safely to reduce length of hospital stay in patients with advanced cancer?
8	1	23	1	Can simple prognostic tools be developed to predict which patients with advanced cancer will benefit, or not, from PN in routine clinical practice?
9	2	15	1	What are the best tools to measure quality of life in cancer patients receiving PN?
10	1	15	1	What are the ethical implications of PN for patients with advanced cancer?
11	2	20	2	When is it appropriate to stop/withdraw PN in patients with advanced cancer?
12	2	23	2	Do patients with advanced cancer need PN of a different nutritional composition to those with benign IF?
13	1	16	0	Can different PN regimens/approaches (individualised vs generic vs intravenous hydration) be used in patients with advanced cancer dependent on the clinical setting?

PN – parenteral nutrition IF – intestinal failure.

literature; these included patients commenting that they had no problems, dealing with line occlusions and conditions for storing HPN (See Supplementary S2). Similar questions were collated and categorised into 32 themes (see Table 2). This resulted in 54 questions. Each member of the steering committee was given between 6 and 11 summary questions to check against the submitted questions to validate the summary questions. The 54 summary questions were discussed and agreed by the committee.

2.4. Second surveys

The 54 summary questions were split into 2 questionnaires consisting of 41 questions related to CIF due to benign disease and 13 questions pertaining to HPN and cancer and 2 questionnaires were created. The second survey ran from August to October 2021, and 216 people took part in the surveys; further demographic details are given in Table 1.

The top 18 questions for benign CIF were put forward for a workshop where participants discussed the ordering. As there were only 13 questions for the second survey for HPN and cancer, the steering committee decided that ordering from the survey would be published as definitive. The final ranking can be seen in Table 3 and the top ten in Fig. 2.

2.5. Final workshop

The workshops took place in December 2021 with one running in the morning, GMT, and one in the evening on two consecutive weeks. There were thirteen patients and seven healthcare professionals of which there were two nurses, two dietitians, two gastroenterologists and one pharmacist. There were two participants from Italy, one from Denmark one from the USA and the rest (sixteen) from the UK. Some family members were sent information about the workshop, but decided not to participate. For the final ranking see Table 4 and the top ten can be seen in Fig. 3.

3. Discussion

This was the first time to our knowledge that those directly affected by HPN, as patients, family members, or healthcare professionals treating them have been consulted for their opinion on

- Does PN improve quality of life for patients with cancer?
- Does early intervention with PN in cancer patients at risk of inoperable malignant bowel obstruction improve quality of life and extend survival?
- How do we define the most appropriate time to commence PN in patients with advanced cancer?
- When should PN be used concurrently with active anti-cancer treatments (eg. Radiotherapy/ chemotherapy) and does this improve treatment outcomes?
- What are the barriers to accessing PN for patients with advanced cancer and how can these be overcome?
- Does PN improve survival for patients with advanced cancer?
- Can fast-track PN protocols be used safely to reduce length of hospital stay in patients with advanced cancer?
- Can simple prognostic tools be developed to predict which patients with advanced cancer will benefit, or not, from PN in routine clinical practice?
- What are the best tools to measure quality of life in cancer patients receiving PN?
- What are the ethical implications of PN for patients with advanced cancer?

Fig. 2. The top ten priorities in home parenteral nutrition and cancer.

research priorities. Following the methodology of the James Lind Alliance, participants from across the globe were involved in creating top ten research priorities for HPN used with CIF due to

benign disease and used with malignancy. For CIF due to benign disease these included avoiding complications, improving physiological function and lived experience. In contrast for HPN in cancer,

Table 4

The top 18 benign intestinal failure and home parenteral nutrition priorities, including the ranking from the second survey and the number of participants who selected each question in the second survey.

Workshop Final Rank	Second Survey		Number of participants selected question				
	Interim Rank		Pt	Family	HCP	Unknown	
1	1		46	16	22	10	How can liver damage from PN be better identified, prevented and treated?
2	2		48	28	14	13	Can central lines be improved; more discrete, have a longer life span, be less easily dislodged, less prone to infection and complications, and easier to repair?
3	6		27	18	15	7	Can absorption be improved for oral nutrition and fluids?
4	3		51	2	9	12	What are the long term negative consequences of PN and how can they be prevented?
5	5		33	21	14	6	How can sustainable vascular access be achieved and preserved?
6	8		37	10	16	8	Can the side effects of PN be reduced e.g. cramp, nausea, night sweats, fatigue, skin reactions?
7	4		43	14	19	13	What are the causes and best ways to prevent and treat line infections?
8	7		38	11	15	13	Does PN effect patients' quality of life and what would improve it?
9	9		26	11	17	6	What factors help the body more effectively adapt to a shortened gut to increase passage time and decrease stoma output?
10	15		22	8	10	5	What are the issues related to sleep whilst having PN over night?
11	14		23	8	11	6	What is the impact of PN on relationships and family life?
12	13		26	15	8	9	Can the number of days on PN be reduced?
13	17		23	8	9	6	What drugs are effective with SBS, can oral drugs be absorbed effectively or should IV drugs be used?
14	11		28	11	10	4	Does gut bacteria change or influence gut response to nutrition during PN?
15	10		27	14	6	8	What do we know about the underlying diseases that lead to intestinal failure and are there any alternative treatments to PN?
16	12		25	18	6	7	What is the safest protocol for swimming and showering with central lines?
17	16		24	13	4	3	What is the optimum infusion rate that can be achieved considering time, safety and absorption?
18	18		27	8	7	10	Can pumps be improved so that they are quieter, lighter and more efficient?

PN – parenteral nutrition, SBS short bowel syndrome, IV – intravenous.

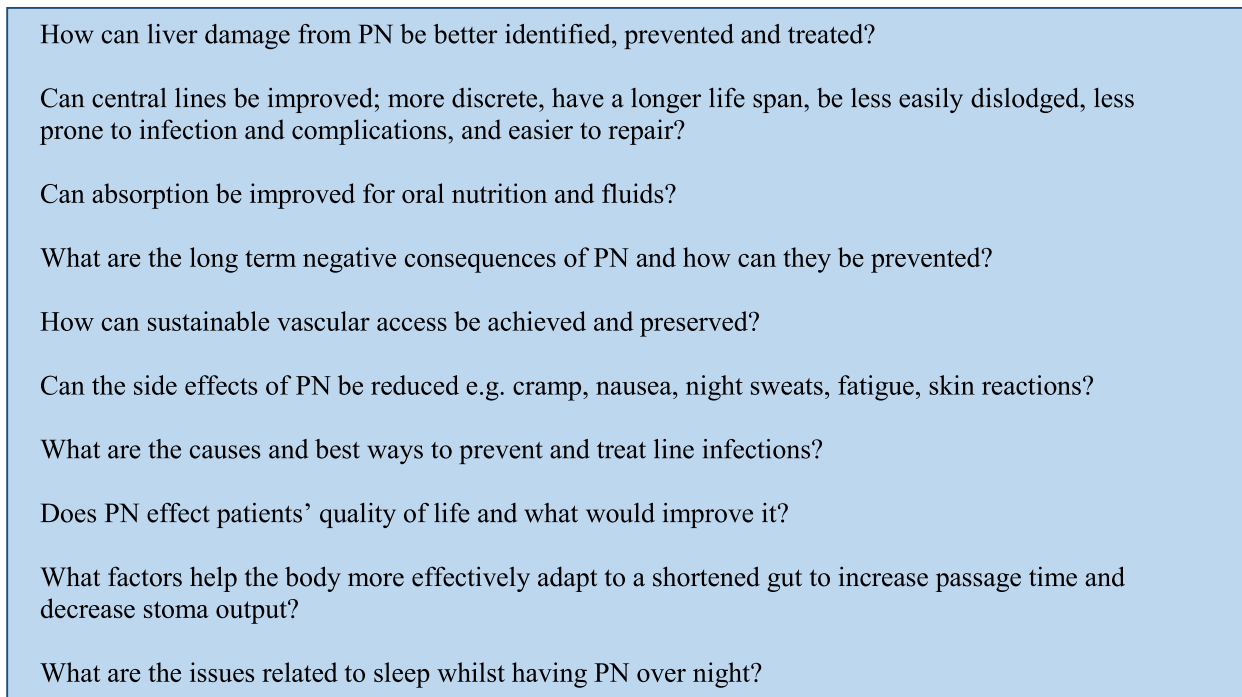


Fig. 3. The top ten priorities in home parenteral nutrition and benign intestinal failure.

they included establishing who would benefit, how to use HPN most effectively and access barriers. Quality of life was amongst the top ten priorities for both groups.

The top priority for benign CIF with patients and healthcare professionals was identifying, preventing and treating liver disease. Intestinal failure-associated liver disease (IFALD) can progress to end stage liver disease, which is a life-threatening complication of HPN causing 1–4% of deaths [20–22]. Although IFALD is serious, catheter-related blood stream infection is also a serious and more common complication [23]. That IFALD is a key area of concern is an important finding and warrants further investigation, in addition to research into preventing and treating the condition.

In contrast quality of life was the top priority for those having HPN due to cancer. This group of patients have a limited life span so quality of life is of utmost importance. The poignant question is often: do the benefits outweigh the inconveniences and risks of the burdens HPN places on patients and their families [24]? This question is crucial for patients and family members and is not easy to answer as people with terminal illness have different views of what constitutes quality of life for them [25–28]. Moreover, some patients will have HPN in addition to anti-cancer treatment and some will not [29]. Given the complexity of the question and its importance to this group of patients and families, it requires sufficient resources to answer definitively due to the sample size required.

This study has several strengths including employing the robust methodology of the James Lind Alliance, which has been used to set research priorities in other conditions such as malnutrition, inflammatory bowel disease and diabetes [30–32]. There has been a variation in the engagement of patients and family members in studies using this methodology; some such as advanced heart disease, have a low engagement, whereas conditions with a highly engaged patient population, such as diabetes, have a high response from those with lived experience [32,33]. CIF due to benign disease is something that some patients and families have to live with for many years and there are many active patient support groups

around the world [34–36]. The involvement of some of these support groups overseeing and promoting the project led to a high response rate from those with lived experience [37,38].

Another particular strength of this project was that it was an international study with patients and healthcare professionals from 13 countries taking part. The rates of HPN vary across the world, with one study finding prevalence ranging from 3.25 to 66 per million and management differs between countries [39,40]. However, despite these differences, we found concerns from different parts of the world were similar. Thus, the priorities generated in this project will be useful to researchers internationally.

A limitation of the study is that ethnic data were not collected. It proved difficult to translate ethnicities in a way that was sensitive to the different national situations and to ensure that compatible data were being gathered. Therefore, it was not possible to know the ethnic mix of people taking part in the surveys. A further limitation is that the surveys were distributed online via patient associations and on social media. Therefore, patients without access to the internet or active on social media would not have had an opportunity to take part.

4. Conclusion

All the priorities presented have been set following methodology of the James Lind Alliance. The resulting priorities are the main concerns of people living in various countries that are directly implicated in the use of HPN either due to their lived experience or as healthcare professionals. As such, they can provide guidance for researchers within the field, policy makers and research funders internationally.

Statement of authorship

Anne Marie Sowerbutts: Investigation, Formal analysis, Project administration, Writing - original draft; Simon Lal: Conceptualization, Supervision, Writing - review & editing; Loris Pironi:

Conceptualization, Supervision, Writing - review & editing; Debra Jones: Investigation, Formal analysis, Writing - review & editing; Chloe French: Investigation, Formal analysis, Writing - review & editing; Marianne Riis: Investigation, Supervision, Writing - review & editing; Andrew Clamp: Supervision, Writing - review & editing; Jennifer McCracken: Supervision, Writing - review & editing; Leanne Williamson: Supervision, Writing - review & editing; Carolyn Wheatley: Supervision, Writing - review & editing; Bethany Johnson: Supervision, Writing - review & editing; Sorrel Burden: Funding acquisition, Conceptualization, Investigation, Supervision, Writing - review & editing.

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Declaration of competing interest

AMS, SB, DJ, CF, LW and SL report an investigator initiated grant from Takeda into their departments during the conduct of the study. JM reports a Health Education England (HEE)/National Institute for Health Research (NIHR) ICA Predoctoral Clinical Academic Fellowship and a Royal Marsden Partners Research Fellowship. Unrelated to this study: SL reports grants into his department from Takeda and Baxter and consulting fees from Takeda, Baxter, Zealand, and VectivBio, LP reports consulting fees from Takeda, Baxter and Northsea Therapeutics, CW reports consulting fees from VectivBio, payments for presentations from Fresenius Kabi and Inspiration Healthcare and conference registration fees from Fresenius Kabi, payment for being on a clinical governance board from B Braun, and BJ reports an honoraria for speaking from Snow Companies.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.clnesp.2022.12.010>.

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