Including the voice of children

Towards patient-centered care

Lorynn Teela

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General introduction

Patient-centered care

Patient-centered care[1] (PCC), also referred to as person-centered care, is defined by the Institute of Medicine [1] as "providing care that is respectful of and responsive to individual patient preferences, needs and values, and ensuring that patient values guide all clinical decisions". This approach puts the patient (and not the disease) at the center of health care and encourages a collaboration between patient, family, and clinicians to deliver care tailored to the unique wishes and needs of patients [2,3]. Although different models of PCC exist [1-4], the New England Journal of Medicine (NEJM) Catalyst [2] describes seven common elements of PCC:

1) The health care system's mission, vision, and drivers for quality improvement are aligned with patient-centered goals.
2) Care is collaborative, coordinated, and accessible. The right care is provided at the right time and place.
3) Care is not only focused at physical comfort, but also on emotional well-being.
4) Care is respectful to patients' and families preferences and values.
5) Patients and families are part of the care team and play a role in the decision making at both patient and system level.
6) Involvement of family in the health care is encouraged and facilitated.
7) Information is shared with patients and families so that they can make informed decisions.

The most important reason for providing PCC is the improvement of individual health outcomes (i.e., medical outcomes, social well-being, and satisfaction with care) [2,5,6]. However, clinicians and health care organizations may also benefit from this approach as it results in enhanced patient satisfaction, greater job satisfaction, better productivity, and a reduction of health care costs (e.g., PCC is associated with a reduction of diagnostic tests and referrals) [2,5-8]. Despite the benefits of PCC, its implementation is challenging, as it calls for a different way of thinking about health care delivery and a changing role of patients and families, i.e., transitioning from a more passive role to an active member of the team [2,3].

This thesis focuses on the development of tools and scientific approaches on how to engage children and families in pediatric care, specifically (Figure 1):

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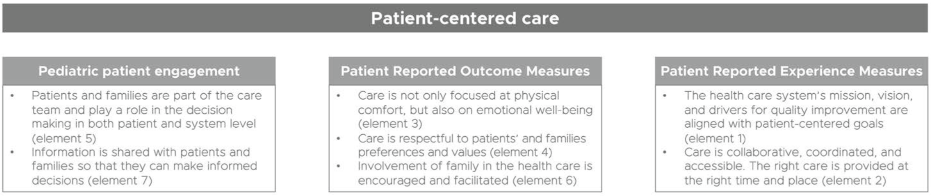
[1] In this thesis the term patient-centered care is used. However, we recognize that this term is broader in pediatrics and also means the engagement of parents and other family members.

- Part 1: Pediatric patient engagement
- Part 2: Patient Reported Outcome Measures
- Part 3: Patient Reported Experience Measures



Patient-centered care

Pediatric patient engagement

- Patients and families are part of the care team and play a role in the decision making in both patient and system level (element 5)
- Information is shared with patients and families so that they can make informed decisions (element 7)

Patient Reported Outcome Measures

- Care is not only focused at physical comfort, but also on emotional well-being (element 3)
- Care is respectful to patients' and families preferences and values (element 4)
- Involvement of family in the health care is encouraged and facilitated (element 6)

Patient Reported Experience Measures

- The health care system's mission, vision, and drivers for quality improvement are aligned with patient-centered goals (element 1)
- Care is collaborative, coordinated, and accessible. The right care is provided at the right time and place (element 2)

Figure 1. An overview of the different part of this thesis in relation to the patient-centered care model of NEJM Catalyst

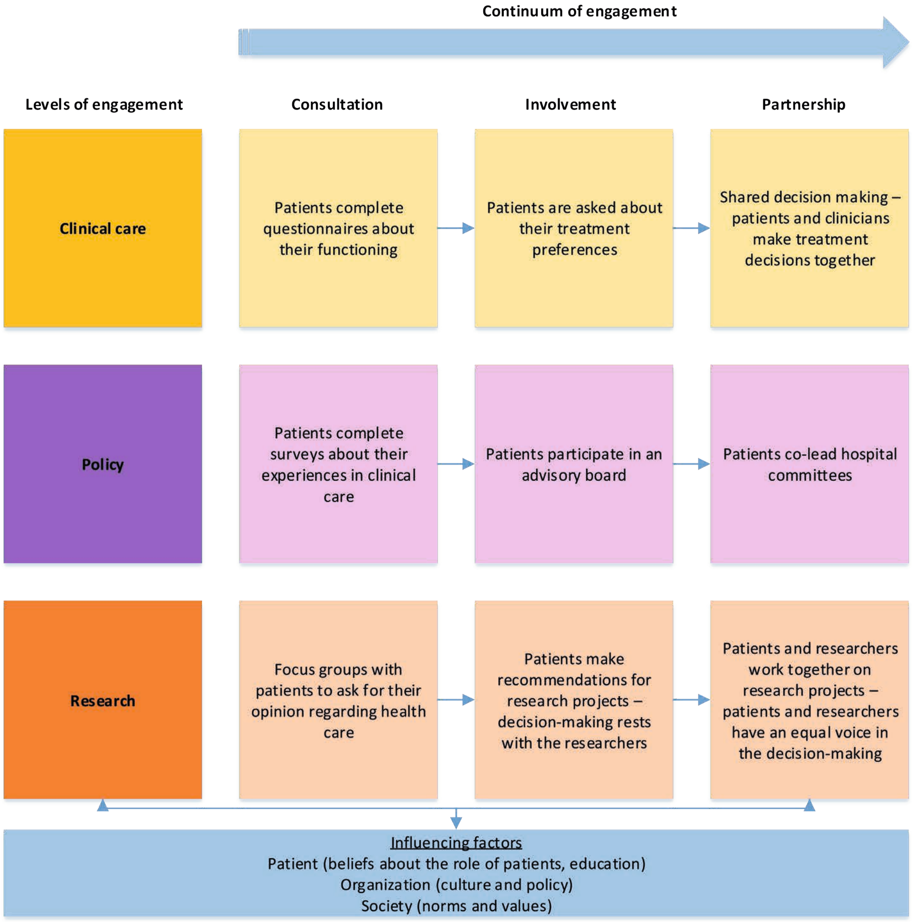
1. Pediatric patient engagement

An essential aspect of providing PCC, according to the NEJM model, is to involve patients in the decision-making in both clinical care and research (common elements 5 and 7) [2]. This involvement of patients, with the aim of improving health (care), is referred to as patient engagement [9,10]. Patient engagement is an umbrella concept and can be explained using the Multidimensional Framework for Patient Engagement in Health Care developed by Carman et. al. [11] (Figure 2). This framework shows that patients can be involved in all areas of health care, including clinical care, policy, and research. In addition, it states that patient engagement is a continuum and patients can influence decision-making to a different extend, ranging from consultation (e.g., patients are asked for their opinion, but have limited influence on decision-making) to active partnership (e.g., patients cooperate as equal partners). For which type of patient engagement is chosen depends on factors such as patients' knowledge, the culture of the organization, time and budget constraints, and societal norms and values regarding patient engagement [11,12].

Patient engagement is beneficial for both patients and organizations. Studies have shown that patient engagement leads to decisions better matching patients' wishes and needs, resulting in improved health outcomes, higher quality of care, and increased inclusion rates in research [13-15]. Furthermore, engaging patients enables them to be more responsible for their own health, which enhances self-confidence and sense of control [12,16].

While the added value of patient engagement is increasingly recognized, clinicians and researchers struggle with the engagement of patients [17,18]. Clinicians doubt whether patients are knowledgeable, involving patients is time consuming, and scheduling meetings with groups of patients is difficult [19-23]. Involving pediatric patients seems to be especially challenging for clinicians, as the competence of children is even more questioned than adults [20,21,24,25]. Also, pediatric patient engagement is complicated by the influence of parents and the lack of experiences by clinicians, policymakers, and researchers [20,25].

Since the United Nations Convention on the Rights of the Child (UNCRC) states that every child has the right to be heard and to have a say in matters that affect them [26], it is necessary to give clinicians more support to involve pediatric patients meaningfully and usefully [27]. However, it is unclear to what extent and in what way patients are engaged in clinical care and research. In addition, currently no tools are available to help clinicians with the engagement of pediatric patients.



Levels of engagement	Consultation	Involvement	Partnership
Clinical care	Patients complete questionnaires about their functioning	Patients are asked about their treatment preferences	Shared decision making – patients and clinicians make treatment decisions together
Policy	Patients complete surveys about their experiences in clinical care	Patients participate in an advisory board	Patients co-lead hospital committees
Research	Focus groups with patients to ask for their opinion regarding health care	Patients make recommendations for research projects – decision-making rests with the researchers	Patients and researchers work together on research projects – patients and researchers have an equal voice in the decision-making

Influencing factors
Patient (beliefs about the role of patients, education)
Organization (culture and policy)
Society (norms and values)

Figure 2. A multidimensional framework for patient engagement in health care, based on the model of Carman [11]

2. Patient Reported Outcome Measures

Common elements of providing PCC are to incorporate and respond to patient and families preferences, needs, and values (NEJM common element 4 and 6) and to also focus on emotional well-being (NEJM common element 3) [2,28]. Patient Reported Outcome Measures can be used to elicit information regarding the perceived impact of disease and/or treatment directly from the patients themselves and from their family members [28,29]. PROMs are thus useful to incorporate the patient perspective into decision-making in clinical care and are therefore seen as an effective way to provide PCC [28,29].

PROMs are standardized, validated questionnaires completed by patients regarding their health status, symptoms, or well-being [30-32]. PROMs can be disease-specific (i.e., applicable for patients with a specific disease) or generic (i.e., applicable for everyone, regardless of disease) and are available for different age-ranges [33]. PROMs can be used for several purposes. Originally, PROMs were developed for use in scientific research to measure the effect of healthcare interventions [34]. However, PROMs can also be used for quality registration of care, where aggregated PROM data is used to gain insight into the quality of care and opportunities for quality improvement [35]. Additionally, PROMs can be used on an individual level in clinical care enabling PCC [36]. The effects of using PROMs in clinical care have been widely studied. These studies showed that using PROMs increases awareness for patients' problems and concerns, enhances patient-clinician communication, improves patient satisfaction with health care and is associated with improved treatment outcomes [28,37-42].

Given the added value of PROMs, it is beneficial for PCC to implement PROMs in clinical practice. Currently, there is a wide variety in how PROMs are implemented, which impacts the intended effects [43]. For example, PROMs are used in clinical practice to monitor symptoms, make diagnoses, decide whether patients' needs an outpatient visit or to facilitate the communication between patients and clinicians [36,44]. The focus of this thesis is on the implementation of PROMs as part of the conversation with the clinician. There are several initiatives that provide guidelines about PROM implementation, such as the International Society for Quality of Life Research (ISOQOL) User's Guide to implementing patient-reported outcomes in clinical practice [45] and the PROM-toolbox, developed by the Dutch National Healthcare Institute [46].

KLIK PROM portal

To facilitate the use of PROMs in clinical practice, the Emma Children's hospital Amsterdam UMC developed the evidence-based KLIK PROM portal (www.hetklikt.nu) [47-50]. In the last 12 years, the KLIK PROM portal has been implemented in more than 40 hospitals in the Netherlands for pediatric and adult outpatient clinics [51]. In these hospitals, patients (children or adults) and/or caregivers complete online PROMs before an outpatient consultation to assess their physical, mental and/or social health (see Figure 3). Answers are converted into an electronic individual dashboard offering a broad range of feedback options customized to each specific PROM [52]. Clinicians discuss the KLIK dashboard with patients during the consultation with the aim to facilitate communication, monitor well-being over time, identify problems, and subsequently provide tailored advice and interventions.

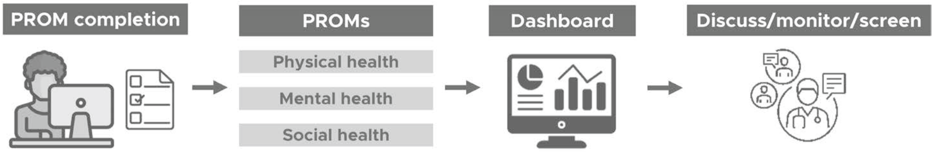


Figure 3. The KLIK workflow

Implementation of the KLIK PROM portal

The KLIK expertise center for PROMs and PREMs guides the implementation of PROMs in different multidisciplinary teams. Implementation strategies are crucial to reach the intended effects, but it remains challenging [53,54]. Implementation research can help to get insight into barriers and facilitators prior to implementation and can therefore ease the integration of PROMs in clinical care [55]. Implementation research is defined as the "scientific study of the use of strategies to adopt and integrate evidence-based health interventions into clinical settings in order to improve patient outcomes and benefit population health" [56]. Implementation research thus studies the mechanisms that influence implementation outcomes. Different implementation sciences models, theories and frameworks can be used to identify determinants (both barriers and facilitators) that influence the implementation outcome and provide implementation strategies as potential solution to barriers [56]. The Consolidated Framework for Implementation Research (CFIR) is a commonly used framework within PROM implementation [57]. This framework consists of five domains (intervention characteristics, outer setting, inner setting, characteristics of individuals, and implementation process), each with a number of constructs focusing on different aspects of implementation (Table 1) [30,57]. With the increasing interest in PROM implementation, several barriers have been identified in the literature within the different CFIR domains [30]. Table 1 provides an overview of these barriers.

Table 1. Barriers in the PROM implementation described using the CFIR framework

Domain	Description	Barriers identified in literature
Intervention characteristics	For PROM implementation, the design of PROMs, PROM administration, and feedback, e.g., complexity	- PROMs are perceived as burdensome - PROMs are not comparable due to different scoring methods
Outer setting	External factors that may impact the implementation, e.g., patients' needs	- Patients are not involved in the selection of PROMs - Patients do not have the ability to complete PROMs due to literacy issues
Inner setting	Internal factors that may impact the implementation, e.g., available resources	- Clinicians experience a lack of time - Insufficient support from ICT
Characteristics of individuals	The views of individuals working with the intervention on implementation, e.g., knowledge about the intervention	- Lack of knowledge on how to utilize and interpret PROMs
Implementation process	Factors related to the implementation process, e.g., planning and evaluation	- Difficulties with embedding PROMs in the existing workflow

Applying the CFIR model to the KLIK implementation process might yield valuable insights into barriers and facilitators, where after strategies can be identified to optimize the KLIK implementation. The involvement of patients/parents is of great importance in determining these factors.

PROMIS

To overcome the challenges associated with the burden of completing PROMs (i.e., long completion time, repetitive and irrelevant questions) [58,59], the National Institute of Health (NIH) developed the Patient-Reported Outcomes Measurement Information System (PROMIS®) [60,61]. PROMIS provides a set of person-centred, standardized instruments to measure a broad range of health domains (physical, mental, and social health) in both adults and children [60,62]. In contrast to legacy instruments, based on Classical Test Theory, PROMIS measures were developed according to Item Response Theory (IRT) [63,64]. An important advantage of IRT is the option of using Computerized Adaptive Testing (CAT) [62,63]. With CAT, questions are presented to patients based on their previous responses. In this way, patients answer a few questions per construct to get a reliable score. Consequently PROMIS measures are shorter, items are more tailored to the patients' situation, and the measurement is more reliable in comparison to legacy instruments resulting in a reduced burden for patients [62-64].

3. Patient Reported Experience Measures

According to NEJM Catalyst, delivering PCC is only possible if care is in line with patient-centered goals and provided at the right time and place (common element 1 and 2) [2]. Therefore, it is necessary to gain patients' feedback about the received care and the extent to which care is experienced as patient-centered. Patient Reported Experience Measures (PREMs) are questionnaires asking patients about their experience with the care received, and are commonly used to gain insight into the quality of care [55,65,66]. PREMs thus offer patients the opportunity to provide information, with the aim to improve care [67]. Contrary to PROMs, PREMs do not measure outcomes of care, but assess the impact of process of care such as communication between clinician and patient, information sharing, and involvement of patients in decision-making. PREMs and satisfaction questionnaires are often used interchangeably, although there are important differences between these instruments [65,66]. PREMs assess whether something that should happen in the health care setting actually happened or how often it happened (i.e., objective experiences), while satisfaction questionnaires assess whether the patient's expectation about the care received were met (i.e., subjective views).

PREMs are increasingly used to pursue PCC. However, guidelines regarding the use of PREMs are lacking, resulting in a wide variety of PREMs and PREM use in pediatrics.

Aim and outline of this thesis

The ultimate goal of this thesis is to enhance pediatric PCC in clinical care and research. Therefore, this thesis focuses on the development of tools and scientific approaches to provide PCC in pediatric care. This thesis is divided into three parts that discuss different parts of PCC.

Part 1: Pediatric patient engagement

Pediatric patient engagement is developing in recent years, and pediatric patients are more often involved in health care (projects). However, clinicians and researchers still struggle with the engagement of pediatric patients [17,18]. To gain insight into the extend of pediatric patients engagement and the methods used, **Chapter 2** provides a comprehensive overview of the literature about pediatric patient engagement in clinical care, policy, and research. To support clinicians with a tool to involve pediatric patients meaningfully and usefully, we developed a pediatric patient engagement tool with adolescents, as described in **Chapter 3**.

Part 2: Patient Reported Outcome Measures

As PROM implementation is a challenging process, insight into barriers and facilitating factors that influence implementation outcomes and stakeholder involvement is required. Understanding the wishes and needs of clinicians, patients, and parents (all stakeholders) is crucial. Therefore, the perspective of clinicians on the implementation of PROMs is investigated in **Chapter 4**. In **Chapter 5**, patients' and parents' perspective on the implementation of PROMs in clinical care is described. **Chapter 6** retrospectively describes the implementation of the KLIK PROM portal using the CFIR framework and identifies implementation strategies to address the identified barriers. Insights into determinants of successful PROM implementation can both improve the implementation of the KLIK PROM portal and help others working on PROM implementation.

PROMIS CATs can offer a promising solution to the burdensomeness of PROMs. Several steps are necessary to use these new instruments in clinical practice. As the PROMIS measures were initially developed in the United States [61], the first step was translation into Dutch [68] and validating the PROMIS instruments in the general population in the Netherlands [69,70]. A second step is to investigate the applicability and feasibility of the PROMIS CATs in a clinical population. Therefore, in **Chapter 7** the psychometric properties of the PROMIS item banks were assessed in boys with hemophilia.

Part 3: Patient Reported Experience Measures

PREMs gather patients' views of their experience with the care received and are commonly used to measure the quality of care, with the goal to make care more patient-centered [55,65,66]. With the growing adoption of PREMs in pediatric care, it is relevant to identify suitable PREMs. **Chapter 8** therefore provides a systematic review of the available PREMs that can be used in pediatric care.

This thesis ends with **Chapter 9;** a general discussion including a reflection on the main findings, clinical implications, methodological considerations, and future perspectives to promote PCC in clinical care and research.

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Part 1

Pediatric patient engagement

2

Pediatric patient engagement in clinical care, research and intervention development: a scoping review

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Abstract

Background

In the last decades, pediatric patient engagement has received growing attention and its importance is increasingly acknowledged. Pediatric patient engagement in health care can be defined as the involvement of children and adolescents in the decision-making of daily clinical care, research and intervention development. Although more attention is paid to pediatric patient engagement, a comprehensive overview of the activities that have been done regarding pediatric patient engagement and the changes over time is lacking. Therefore, the aim of this study is to provide an overview of the literature about pediatric patient engagement.

Methods

The methodological framework of Arksey & O'Malley was used to conduct this scoping review. The bibliographic databases Medline, Embase, and PsycINFO were searched for eligible articles. All retrieved articles were screened by at least two researchers in two steps. Articles were included if they focused on pediatric patient engagement, were carried out in the context of clinical care in pediatrics, and were published as full text original article in English or Dutch. Data (year of publication, country in which the study was conducted, disease group of the participants, setting of pediatric patient engagement, used methods, and age of participants) were extracted, synthesized, and tabulated.

Results

A total of 288 articles out of the 10,714 initial hits met the inclusion criteria. Over the years, there has been an increase in the number of studies that engage pediatric patients. Pediatric patients, especially patients with multiple conditions or oncology patients, were most involved in studies in the United States, United Kingdom, and Canada. Pediatric patients were most often asked to express their views on questions from daily clinical care and the individual interview was the most used method. In general, the extent to which pediatric patients are engaged in health care increases with age.

Discussion

This scoping review shows that there is an increasing interest in pediatric patient engagement. However, lack of uniformity about the definition of pediatric patient engagement and clear information for clinicians hinders engagement. This overview can inform clinicians and researchers about the different ways in which pediatric patient engagement can be shaped and can guide them to engage pediatric patients meaningfully in their projects.

Introduction

In 1989, over 190 countries, including the Netherlands, signed the United Nations Convention on the Rights of the Child (UNCRC) [1]. The UNCRC describes the human rights for every child, such as self-determination, freedom of thoughts and religion, and the right to have a say in matters that affect them. It is with this convention that the engagement of children in health care, research and intervention development became more important [2,3]. From that moment on, clinicians, researchers and policymakers more often tried to carry out their health care projects and decision-making together *with* pediatric patients rather than *about* or *for* pediatric patients [3].

Involving children in decision-making about daily clinical care, research and intervention development is referred to as 'pediatric patient engagement' [4,5]. The extent to which children influence the decision-making processes can vary from consultation (e.g., patients are asked for their opinion, but have limited influences on decision-making) to active partnership (e.g., patients cooperate as equal partners with other stakeholders and share responsibility) [6,7]. Notwithstanding the extent of involvement, pediatric patient engagement has important value for health care. Previous research shows that pediatric patient engagement increases children's self-confidence and sense of control, which results in better treatment outcomes [8]. Moreover, pediatric patient engagement leads to higher inclusion rates in research and improves the translation from research to clinical practice [9].

Although the importance of pediatric patient engagement is acknowledged, pediatric patients are not always involved in the decision-making process in health care [8,10]. Clinicians, researchers, and policymakers are, for example, reserved in involving pediatric patients in health care as they doubt the capacity of children required for participating, and they lack experience in engaging children [8,11,12]. In addition, pediatric patient engagement is complicated by the tendency of adults to protect children from making difficult decisions [8,11]. Professionals therefore need more support to involve pediatric patients meaningfully and usefully [13].

In the last years, a few systematic reviews on pediatric patient engagement in clinical care have been conducted [2,3,14]. These systematic reviews are relatively outdated (over 10 years old), given the fact that pediatric participation is a developing practice. The focus of the conducted systematic reviews were only on engagement in the decision-making process in the consultation room and the challenges involved [2,14]. Also, in one paper, the included articles are only summarized and interpreted by one author [2] , as opposed to systematically collating, summarizing, and reporting the results. A recent scoping review describes the involvement of adolescents and young adults (12-25 years) with a chronic condition in health and social care [3]. This review, only including 23 studies, provides a synopsis of the used definitions of patient engagement, goals, methods, and impact of the involvement of youth in research and implementation projects. However, a comprehensive overview of the activities that have been done in the past regarding pediatric patient engagement, also including primary school-aged children (4-18 years) in health care is lacking, as well as insights into how patient engagement takes place in clinical care, research, and intervention development. In addition, we want to know how pediatric patient engagement has developed in recent years to learn more about the different ways pediatric patients can be involved in health care. Therefore, the aim of this study is to provide an overview of the literature about pediatric patient engagement in clinical care, research, and intervention development.

Methods

Due to the broad nature of the study aim, a scoping review was conducted. Scoping reviews can be used to provide an overview and map the available evidence around a certain topic [15,16]. The methodological framework of Arksey & O'Malley [16] was used to guide this scoping review. This framework consisted of the following 5 stages:

Stage 1: Identifying the research question

The research question of this scoping review was: *What is known from the literature about pediatric patient engagement in clinical care, research and intervention development?* A comprehensive approach was chosen to examine the extent and nature of pediatric patient engagement in the broad field of pediatrics. Key parameters were **patient engagement** (defined as: actively involving children in the clinical care, medical research, and intervention development. This means that children were asked for their opinion on certain topics or that they played a role in the decision-making process), **children and adolescents** (defined as people aged 4-18 years), and **pediatrics** (defined as the medical care of children and adolescents in a hospital/clinical setting and the associated science).

Stage 2: Identifying relevant studies

A comprehensive search strategy was developed and carried out in collaboration with a medical research librarian (JGD). To obtain a clear description of the construct, both published and unpublished literature about engagement of children and adolescents was collected and reviewed by at least two research-psychologists (FW, MV, LH). Subsequently, a visualization of similarities (VOS) analysis [17] was carried out with the software tool VOSviewer® to remove irrelevant terms from the search strategy by NOTing [18]. Medline, Embase and PsycINFO were searched for eligible articles from inception (May 2017). The construct of the search strategy can be summarized as follows: ([hospitalized patient] AND [patient participation]) NOT [irrelevant terms identified by VOS analysis]. See Additional file 1 for full search details.

In February 2021, an update of the literature search was done. The same search strategy was applied. The bibliographic databases were searched for eligible articles in the period January 2017 until February 2021. For practical reasons, duplicate articles from the period January 2017 – May 2017 were removed in the last step of the study selection.

Stage 3: Study selection

Title and abstract of the articles retrieved were assessed by at least two members of the research team (LT, LEV, EEWK, FW, MV, LH) using the software tool Rayyan [19]. To reduce individual bias during the screening process and to refine inclusion and exclusion criteria, consultation took place between the members of the research team after screening the first 300 articles. The full text of potentially relevant articles was obtained and assessed by at least two members of the team (LT, LEV, EEWK). If necessary, a third member (LH) made the decision regarding inclusion of an article. An article was included if the study described all following inclusion criteria:

- Focused on engagement of children and adolescents (4-18 years).
  Studies that included pediatric patients in a broader age range or studies that included both pediatric patients and young adults were also included.
- Participants were asked for their opinion regarding clinical care, research, policy and/or intervention development.
- Carried out in the context of clinical care/pediatrics.
- Published as a full text original article (i.e. not an abstract, review, commentary, dissertation or study protocol).
- Published in English or Dutch.

Studies that reported only on the engagement of representatives of pediatric patients (i.e., caregivers, family members) or studies that did not clearly distinguish pediatric patients as a subgroup were excluded. In addition, studies that were conducted in the field of dentistry or psychiatry or studies that described the engagement of pediatric patients in a school or home setting were excluded. Also, studies that explored the experiences of children living with a medical condition in general (e.g., experiences of children living with HIV) were excluded, unless the studies reported on the life-experiences of these children with the aim to improve a medical treatment or to develop an intervention/tool. Furthermore, studies describing the involvement of pediatric patients in developing measurements using cognitive interviews for checking the understanding of questions or icons were excluded. The research team does not consider using cognitive interviews for this purpose to be part of pediatric patient engagement. The opinion of children and adolescents is thus not being asked in these cognitive interviews. Finally, studies that only described the importance of pediatric patient engagement, but did not discuss the application of pediatric patient engagement, were also excluded.

Stage 4: Charting the data

A data extraction form was developed by the team, and data were extracted from the included articles by one members of the team (LT, LEV, or EEWK). A second member of the team (LT, LEV, or EEWK) cross-checked a selection of the extracted data. The following data were extracted from the articles: year of publication, country in which the study was conducted, disease group of the participants, number of participants, setting of pediatric patient engagement (health care, research, or development of interventions or tools), method used for patient engagement, and age of participants.

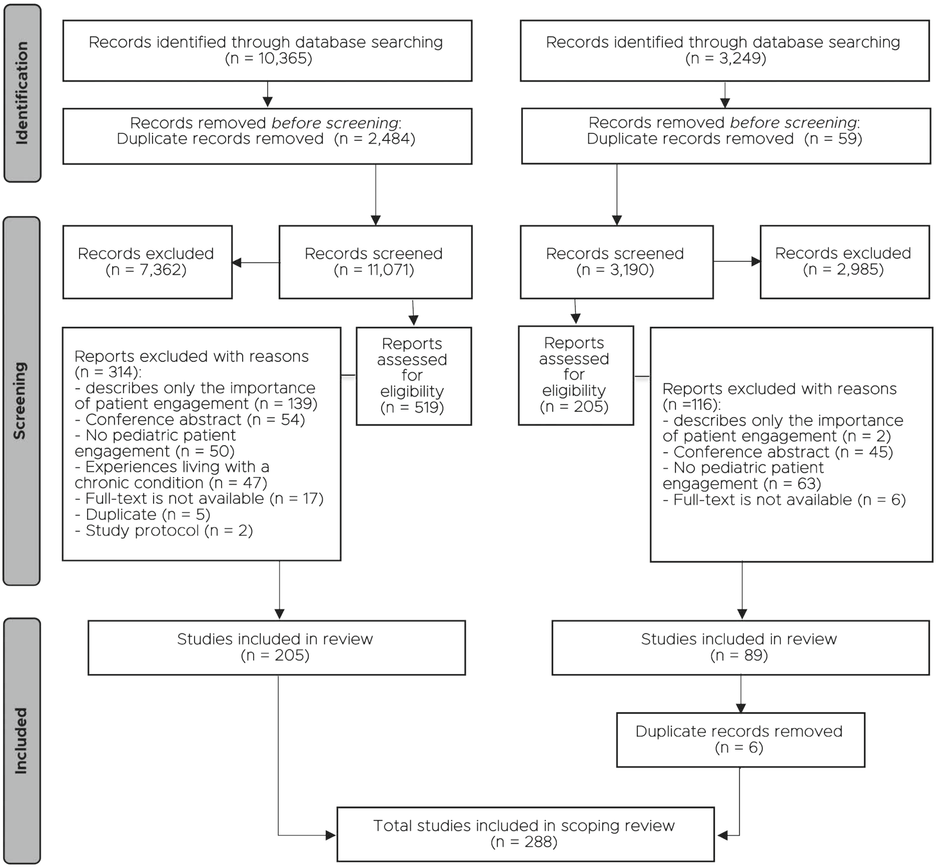
Stage 5: Collating, summarizing, and reporting the results

Extracted data were analyzed quantitatively with the use of the Statistical Package for Social Sciences (SPSS) version 28. This quantitative data provided an overview of the nature and extent of pediatric patient engagement. To learn more about the goals of pediatric patient engagement, the data were screened by the research team and examples were cited.

Results

Search and selection results

The study selection process is presented in the PRISMA flow diagram of Fig 1. The literature search yielded 10,365 (2017) and 3249 (2021) articles. After removing duplicates, title and abstracts of 11,071 (2017) and 3190 (2021) articles were assessed. Of these, 519 (2017) and 205 articles (2021) were eligible for full-text review. A total of 288 articles met the inclusion criteria and were included. An overview of the characteristics of included studies can be found in Additional file 2.

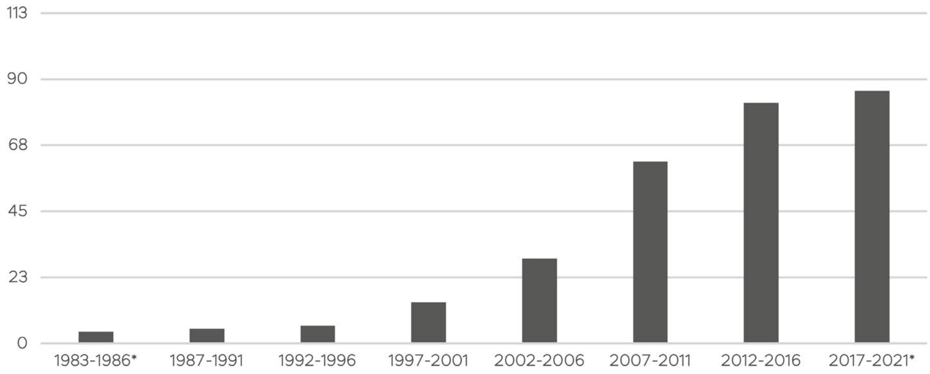


From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

Figure 1. Flow chart of the identification and selection process of studies [20]

Pediatric patient engagement through the years

The included articles are published between 1983 and February 2021, as shown in Fig 2. Over the years there has been an increase in the number of studies that include pediatric patient engagement.



* Range is less than 5 years

Figure 2. Overview of the included articles (number) per 5 years

Pediatric patient engagement per country

Figure 3 shows the number of studies in which pediatric patient engagement is included per country. Most studies involving pediatric patient engagement are performed in the United States of America, followed by the United Kingdom, and Canada.

Pediatric patient engagement per disease group

The largest group of studied patients encompasses pediatric patients from different disease groups (26%) in their clinical care, research or development of intervention, and 10% of the studies concerned children being admitted to the hospital for various reasons. When looking at individual disease groups, pediatric oncology patients (22%) are most often engaged about their opinion, followed by pediatric patients undergoing surgery (7%), diabetes patients (5%), asthma patients (4%), transplant patients (4%), patients with Juvenile Idiopathic Arthritis (2%), and pediatric patients in palliative care (2%).

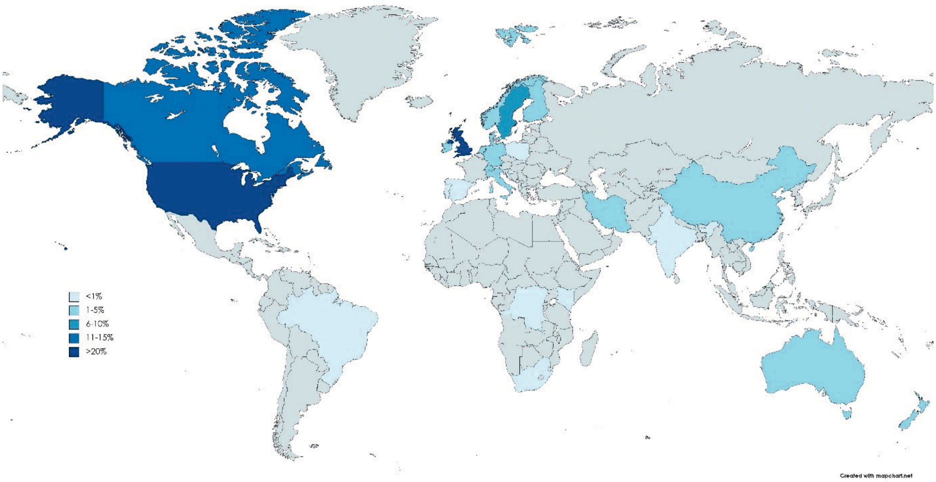


Figure 3. Overview of the articles (% of total) that include pediatric patient engagement per country

Setting of patient engagement

Pediatric patients were asked for their opinion or experiences in different settings: in clinical care (81%), research (10%), and intervention development (9%). One study on adolescents' beliefs about making treatment decisions and trial participation decisions following a cancer diagnosis was included in both the clinical care and research setting [21].

Clinical care: The majority of the included articles were about pediatric patient engagement in clinical care. The aims of these studies were diverse. For example, children's perspectives on the disclosure of medical errors were asked [22], children were asked about their experiences with postoperative pain and pain management [23], and adolescents' preferred level of involvement in the decision-making process in cancer care was investigated [24]. The ultimate goal of engaging pediatric patients in these kind of studies was improving daily clinical care.

Research: In the field of research, pediatric patients were mainly involved to gain more understanding into the reasons why pediatric patients do or do not participate in research, what factors influence their decision, and what adolescents' preferences were regarding the organization of research participation [25,21,26-28]. With this information, researchers aimed to improve recruitment strategies. In addition, a few studies evaluate the benefits and limitations of the use of a specific study design, for example a participatory research approach with chronically ill children as coresearchers [29], or asked pediatric patients with chronic conditions about their research priorities [30,31].

Intervention development: Pediatric patients were involved in the development and evaluation of various tools, such as a toolkit for advanced care planning [32], a therapeutic platform that provides health information to pediatric patients to prepare them for hospital procedures [33], a smartphone app developed to enhance medical adherence [34], and educational videos to motivate adolescents to become more actively involved during the outpatient visit [35].

Used methods for pediatric patient engagement

In the included articles different methods were used for pediatric patient engagement, as shown in Fig 4. The most commonly used method to engage pediatric patients in clinical care, research and intervention development was an individual interview (227 studies), followed by focus groups (40 studies), and draw & write/tell techniques (30 studies). Other used methods were an open-ended questionnaire (11 studies), photo and video techniques (9 studies), sentence completion (8 studies), and keeping a diary (4 studies). Multiple methods were sometimes used in one study. Below is an overview of the different techniques used in the studies and examples of studies that used these methods to include pediatric patients in their projects.

Individual Interview

In individual interviews, the interviewer questions the pediatric patient about the experienced facts and perception of the topic of the research question [36]. In the included studies, pediatric patients were for example interviewed about their expectations regarding the quality of the nursing care [37] or about their experiences and wishes with regard to their first conversation about epilepsy with their clinician [38]. The interviews were conducted in different ways. Almost all studies used a semi-structured interview [38-40], but a few studies conducted an unstructured interview [41]. Furthermore, the majority of interviews were held face-to-face in the clinical setting [37-39] or at the patients' home [40], and a few interviews were conducted by telephone [39].

Focus group

A focus group is a group interview with several participants (the number of participants varies per study from 2 to 8 participants) [25,36,42,43]. Focus groups were held about a wide variety of research questions, for example 'What do adolescents with a rheumatic condition think about research involvement and how should adolescents involvement in research be organized? [25]', 'What are the perceptions and wishes of children with cancer regarding information exchange during their illness? [42]' or 'What do pediatric patients with life-limiting conditions think of the Implementing Pediatric Advance Care Planning Toolkit?' [32]. Focus groups are often composed on shared characteristics, such as age or disease group, in order to obtain a homogeneous group [25,44]. In most studies, focus groups are held with children from 11 years and older [25,32,43,44]. An advantage of a focus group is that patients ask each other for explanations, resulting in more information in comparison to the sum of individual interviews. Disadvantages are that sometimes not every participant gets the chance to express their opinion due to the group composition and that experiences can be presented more polarized. An experienced discussion leader is necessary for a successful focus group [36].

Draw & write/tell techniques

With the use of the draw & write/tell technique, pediatric patients are asked to draw a picture around the theme of the research question. The researcher uses the drawing as starting point for the conversation. An advantage of this technique is that the drawing increases the ability of children to talk about their experiences [45-47]. Most of the times, the draw & write/tell technique is used to ask for the experiences (e.g., experiences of children with regard to the treatment of recurrent cancer or to identify characteristics of a good nurse from the perspective of hospitalized children) of younger children (4-12 years) [45-47]. Draw & write/tell techniques are often used in combination with other quantitative or qualitative techniques [33,47].

Photo/video techniques

With photo/video techniques, pediatric patients are asked to choose/make photos or videos that represent their thoughts of feelings. For example, the things they did or did not like in the hospital [48,49]. Subsequently, children are asked to provide an explanation to the pictures in an interview. An advantage of these techniques is that children are completely free to indicate what is important for them [48]. Examples of research questions for which photo/video techniques are used are 'What are the experiences of adolescents living with type 1 diabetes, and what are their support needs during the transition from child- to adulthood'? [50] and 'What are the experiences of children with the hospital care, and how could services be improved according to them?' [48]. Photo/video techniques are used for a wide age group (from about 6 years) [48-50].

Sentence completion

In this elicitation technique, patients are presented with half of a sentence and are asked to complete this. For example, the sentence started with *'In my view, the best things about the hospital have been …'*. An advantage of the sentence completion technique is that it offers pediatric patients the opportunity to express their opinion in their own words, without being influenced by others [36,51]. Sentence completion was used in studies that try to identify the experiences and wishes of pediatric patients with health care, with the ultimate goal to improve the quality of care [51,52].

Diary

Both unstructured and structured diaries can be used in study designs. With unstructured diaries, pediatric patients can write anything about a certain theme in their diary. While with the use of structured diaries, patients are asked to answer a number of questions on a daily basis. The included studies mainly used unstructured diaries in their research design [53]. Aims for which diaries are used are for example 'Exploring the extent to which adolescents are involved in care planning' and 'Identify factors that affect pediatric patients while receiving pediatric palliative care' [53,54].

Other

Other techniques that are used in the included studies to engage pediatric patients are, for example, participation in design meetings [55], advisory member of the research team, or other elicitation techniques like games, quizzes [56], and informal conversations [57].

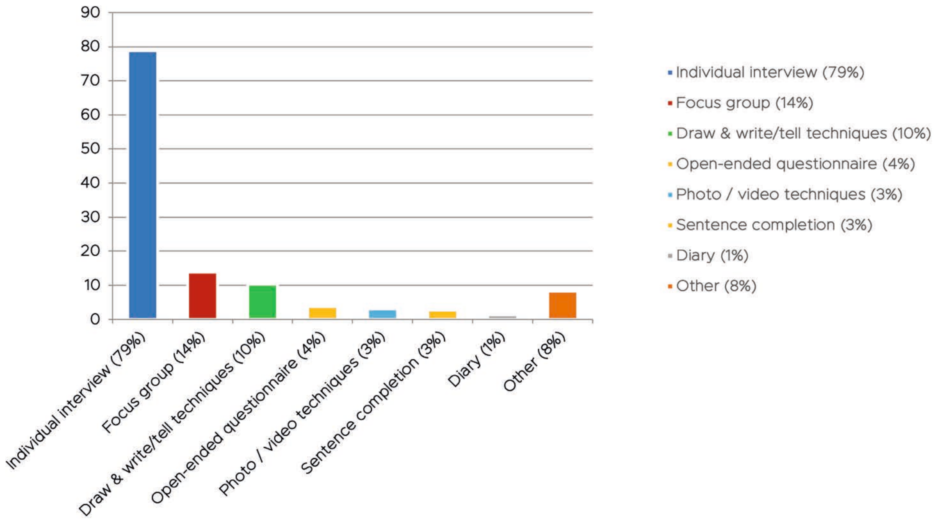


Figure 4. Overview of the methods used for pediatric patient engagement in the included articles

Patient engagement by age group

While some studies included young adults up to age 35 (some studies included both pediatric patients and young adults), analysis of engagement methods in this paper focuses on children up to age 18. In Fig 5, an overview is provided of the number of studies that included pediatric patients in a specific age range. Pediatric patients in the age range 13-17 years were most often engaged in studies, followed by the age groups ranging from 9 to 12 years, and from 4 to 8 years. For 14 studies the age of the included pediatric patients was not clearly specified. The reason for this is in some cases that pediatric patient engagement has been conducted in a subset of the study population.

2-3 years: The youngest age at which pediatric patients were involved in studies regarding clinical care, research or intervention development was 2 years. These young children were asked about their views of, for example, the hospital clown [58], their nurse or doctor [59], or their preferences for the used design/color in their hospital environment [60]. Except for one study (draw & write/tell technique) [59], interviewing was the used method for pediatric patient engagement in this age group. In most cases, parents were present to help their child or they were afterwards asked to reflect on the experiences of their child.

4-8 years & 9-12 years: Pediatric patients in the age range 4-12 are regularly asked for their opinion in the health care setting. All described methods were used in this age group. The draw & write/tell technique is used more often in this age group compared to other age groups.

13-17 years: Adolescent patients are most often included in pediatric patient engagement. Also in this age group, all described methods for patient engagement are used. However, focus groups were used more often in this age group compared to the younger age groups.

≥*18 years:* Most studies involved pediatric patients in their projects until the age of 18/19 years. Some studies involved a wider population and included both pediatric patients and young adults till the age of 35. This was the case, for example, in a study that aimed to establish a research agenda for patients with pediatric inflammatory bowel disease [30] or a study that investigated the views of adolescents and young adults (AYAs) with regard to their wishes and needs for a smart phone app that could be used to improve adherence to medication in the oncology setting [34].

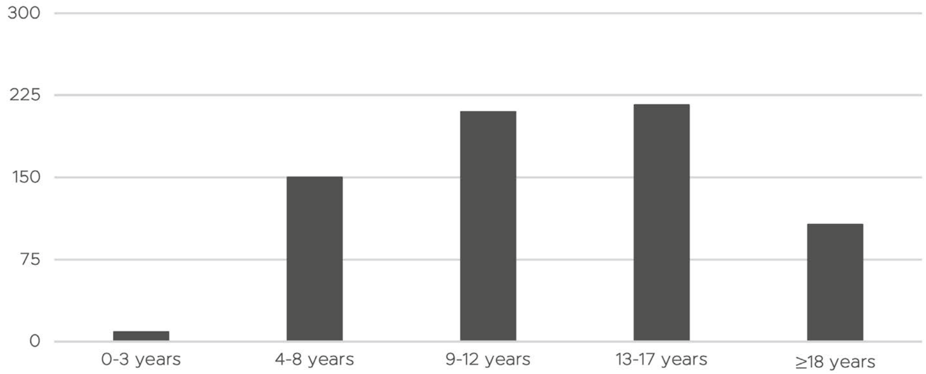


Figure 5. Overview of the number of articles that included pediatric patients in a specific age group

Discussion

This scoping review provided an overview of the existing literature about pediatric patient engagement in clinical care, research, and intervention development. The results showed an increase in the number of studies that report on pediatric patient engagement in the past decades, suggesting an increased interest in this topic. In the United States and Europe in particular, pediatric patients are more often involved in studies about clinical care, research, and intervention development compared to other countries and continents. A mix of patients from different disease groups were mostly asked for their opinion in the included studies, followed by oncology patients. Pediatric patients in the age range 9-17 years were most often engaged in a wide variety of projects compared to the other age groups. The individual interview is the most commonly used method to engage pediatric patients, followed by focus groups (for older children) and draw & write/tell techniques (for younger children). The majority of the included studies focused on the engagement of pediatric patients in clinical care with the aim to improve the quality of daily clinical care for patients.

The increased attention for pediatric patient engagement in the last decade is in line with the scoping review from Van Schelven et al. [3] about the involvement of adolescents (12-25 years) in research and implementation projects. Although our scoping review has a broader scope, included many studies, and focused on younger patients (4-18 years) in daily clinical care, the findings are comparable. Also in the study from Van Schelven et al. [3] the most important goal for patient engagement is improving the quality of care. In addition, the authors mentioned the lack of uniformity around the definition of patient engagement in the literature, which we underline. In the future, consensus needs to be reached about the definition of pediatric patient engagement and about the way clinicians and researchers should engage pediatric patients in their studies [3,5,9]. While conducting this study, it became evident that information on how pediatric patients were engaged was lacking. Therefore, we recommend, as a next step in the field, the development of a guideline to secure a uniform way to report on pediatric patient engagement in scientific papers. This guideline should include information on operationalization of patient engagement, goal, setting, age of patients, methods used, feasibility, and should be established in co-creation with all relevant stakeholders, definitely including patients and parents.

Regardless the external pressure/reinforcement (for example, pediatric patient engagement is increasingly mentioned as a requirement for grand applications by subsidy providers) for researchers to involve pediatric patients in their projects, only a few research projects include pediatric patients. This suggests that researchers need more tools and (financial) support to engage pediatric patients meaningfully. For example, we recently developed a patient engagement game for adolescents with a chronic condition, in cocreation with all stakeholders [61]. This game provides researchers and clinicians with a tool that can help them to engage pediatric patients meaningful in decision-making about clinical care, research and intervention development. In addition, we saw in some included studies that a small number of pediatric patients were involved in the project without having influences on the choices made, leading to tokenistic participation (a symbolic or perfunctory form of patient engagement, in which patients have no influence on decision-making [62]). Breaking through tokenism is difficult, as long as the added value and impact of pediatric patient engagement is not fully recognized, and challenges as funding, representativeness, changing power relations, and letting go of control over the project are not yet overcome [2,3]. In addition, there are reasons and situations in which it may be particularly challenging or even inappropriate to engage children, because they may not have the capacity to understand some aspects of their care, and ultimately their parents can legally override their decisions about their own care.

Different methods were used to involve patients, with the individual interview being the most common method [9]. The methods used in pediatric patient engagement correspond with previous literature about patient engagement with both children and adult patients [3,9]. Yet, there is no known best method to use for patient engagement. Which method is chosen depends on the project in which patients are involved, the age of the participants, and the availability of patients to participate [3,9]. Future research should focus on increasing knowledge about the used methods and their suitability and impact for different research questions and target groups.

This scoping review provides a descriptive overview of the existing literature about pediatric patient engagement (4-18 years) in clinical care, research, and intervention development. This overview can inform clinicians or researchers, who are insecure about how to engage pediatric patients, about the different ways in which patient engagement can be shaped, and guide them to engage pediatric patients in their project. A strength of this study is the broad approach, making it possible to map the existing literature about pediatric patient engagement in a wide range of health care. However, due to its descriptive nature, the study also has a number of limitations. First, this study did not pay attention to the impact of pediatric patient engagement in the included studies. This might be an interesting area for future research as it could give us insight into the added value of patient engagement. Second, scoping reviews do not assess the quality of the included articles [16]. However, assessing the quality of studies could help us to better understand and interpret the results found. Third, due to geographical differences, pediatric care can be interpreted differently. Therefore, we did not include populations as dentistry and psychiatry. In addition, only articles published in English were included. Last, lack of uniformity about the definition of pediatric patient engagement and the influence of tokenism made it difficult to determine what exactly is done in the studies and whether patients actually influence the decision-making process. Therefore, it is possible that we missed studies in this review or that we incorrectly included studies.

In conclusion, this scoping review shows that there is an increasing interest in pediatric patient engagement. Pediatric patients are more often asked to express their views on questions in daily clinical care with the aim of improving the quality of care and tailoring care to patients' needs. However, lack of uniformity about the definition of pediatric patient engagement and clear information and support for clinicians to engage patients in a meaningful way hinders engagement and can lead to tokenistic engagement. Guides, such as this overview, and sharing lessons learned can help clinicians to feel more confident about engaging pediatric patients in their daily practice.

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Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Availability of data and materials

All data generated or analyzed during this study are included in this published article and its Additional files.

Keywords

Patient participation, Patient involvement, Pediatrics, Chronic diseases

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Supplement 1 – Search strategy

Ovid MEDLINE(R) ALL <1946 to February 15, 2021>
Search date: 17 February 2021

#	Searches	Results
1	(adolescent, hospitalized/ or child, hospitalized/) and patient participation/	61
2	(adolescent, hospitalized/ or child, hospitalized/) and (participat* or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion or engagement).ab,kf,ti.	1016
3	(youngster or pubert* or pubescent or prepubescent or school or schools or schoolkid* or schoolchild* or highschool* or kid or kids or underage* or youth? or boy or boys or girl? or sibbling* or child or children or "children's" or schoolchild* or adolescents or adolescence or juvenile or minors or teen or teens or teenager* or p?ediatric? or adolescent? or adolescence). ab,kf,ti. and patient participation/	1957
4	((youngster or pubert* or pubescent or prepubescent or school or schools or schoolkid* or schoolchild* or highschool* or kid or kids or underage* or youth? or boy or boys or girl? or sibbling* or child or children or "children's" or schoolchild* or adolescents or adolescence or juvenile or minors or teen or teens or teenager* or p?ediatric? or adolescent? or adolescence) adj4 (participation or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion)).ab,kf,ti.	43793
5	exp hospitals/ or hospitalization/ or exp academic medical centers/ or exp hospital units/ or exp nursing research/	569687
6	(hospital* or multihospital or multicent* or multi cent* or clinical decision making or health care setting? or nursing research or (clinical adj2 practice) or (p?ediatric adj2 care)).ab,kf,ti.	1732120
7	(participat* or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion or engagement).ab,kf,ti. or patient participation/	3711271
8	4 and (5 or 6) and 7	6234
9	or/1-3,8	8844
10	(Lecture or Medical sudent or director or Dean or Educator or Adult service or Curriculum or Profession or Philosophy or mental health problem or Mental illness or Child psychiatry or Colleague or Psychiatry or psychiatrist or adolescent psychiatry or Art or Health promotion or Court or Labor or Breast cancer or child Birth or Mental disorder or Student or National institute or Abortion or Public Health or Gynecologist or pregnant wom* or first month).ab,ti. [VOS red irrelevant terms]	1226792
11	(New born or Autism or Psychosis).ab,ti. [VOS green irrelevant terms]	88418
12	(Psychiatric hospitalization or Smoking or Schizophrenia or Psychopathology).ab,ti. [VOS blue irrelevant terms]	368277
13	(Terrain vehicle or atv or Motor vehicle collision or alcohol or substance or substance abuse or suicide or Suicide attempt or Abuse or Abused Child or Child abuse or "Substance use" or Street or rural area or City or Mechanism or Cent or product or Juvenile justice system).ab,ti. [VOS yellow irrelevant terms]	2291914
14	Breastfeeding.ab,ti. [VOS pink irrelevant terms]	27484
15	or/10-14	3759560
16	9 not 15	7021

Supplement 1 – *(continued)*

#	Searches	Results
	Ovid Embase Classic+Embase <1947 to 2021 February 15> **Search date: 17 February 2021**	
#	**Searches**	**Results**
1	(*hospitalized adolescent/ or *hospitalized child/) and *patient participation/	8
2	(*hospitalized adolescent/ or *hospitalized child/) and (participat* or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion or engagement).ab,kw,ti.	317
3	(youngster or pubert* or pubescent or prepubescent or school or schools or schoolkid* or schoolchild* or highschool* or kid or kids or underage* or youth? or boy or boys or girl? or sibbling* or child or children or "children's" or schoolchild* or adolescents or adolescence or juvenile or minors or teen or teens or teenager* or p?ediatric? or adolescent? or adolescence).ab,kw,ti. and *patient participation/	833
4	or/1-3	1145
5	((youngster or pubert* or pubescent or prepubescent or school or schools or schoolkid* or schoolchild* or highschool* or kid or kids or underage* or youth? or boy or boys or girl? or sibbling* or child or children or "children's" or schoolchild* or adolescents or adolescence or juvenile or minors or teen or teens or teenager* or p?ediatric? or adolescent? or adolescence) adj4 (participation or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion)).ab,kw,ti.	58773
6	exp *hospital/ or *hospitalization/ or *university hospital/ or exp *"hospital subdivisions and components"/ or exp *nursing research/	375014
7	(hospital* or multihospital or multicent* or multi cent* or clinical decision making or health care setting? or nursing research or (clinical adj2 practice) or (p?ediatric adj2 care)).ab,kw,ti.	2711622
8	(participat* or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion or engagement).ab,kw,ti. or *patient participation/	4896091
9	5 and (6 or 7) and 8	9118
10	(Lecture or Medical sudent or director or Dean or Educator or Adult service or Curriculum or Profession or Philosophy or mental health problem or Mental illness or Child psychiatry or Colleague or Psychiatry or psychiatrist or adolescent psychiatry or Art or Health promotion or Court or Labor or Breast cancer or child Birth or Mental disorder or Student or National institute or Abortion or Public Health or Gynecologist or pregnant wom* or first month).ab,ti. [VOS red irrelevant terms]	1657004
11	(New born or Autism or Psychosis).ab,ti. [VOS green irrelevant terms]	129949
12	(Psychiatric hospitalization or Smoking or Schizophrenia or Psychopathology).ab,ti. [VOS blue irrelevant terms]	535521
13	(Terrain vehicle or atv or Motor vehicle collision or alcohol or substance or substance abuse or suicide or Suicide attempt or Abuse or Abused Child or Child abuse or "Substance use" or Street or rural area or City or Mechanism or Cent or product or Juvenile justice system).ab,ti. [VOS yellow irrelevant terms]	2988625
14	Breastfeeding.ab,ti. [VOS pink irrelevant terms]	34172
15	or/10-14	4983444
16	4 not 15	973
17	9 not 15	7041
18	16 or 17	7879
19	limit 18 to medline	1833
20	18 not 19	6046

Supplement 1 – *(continued)*

Ovid APA PsycInfo <1806 to February Week 2 2021>
Search date: 17 February 2021

#	Searches	Results
1	((youngster or pubert* or pubescent or prepubescent or school or schools or schoolkid* or schoolchild* or highschool* or kid or kids or underage* or youth? or boy or boys or girl? or sibbling* or child or children or "children's" or schoolchild* or adolescents or adolescence or juvenile or minors or teen or teens or teenager* or p?ediatric? or adolescent? or adolescence) adj4 (participation or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion)).ab,id,ti.	59961
2	hospitals/ or hospitalized patients/ or hospitalization/ or nursing.jx.	89947
3	(hospital* or multihospital or multicent* or multi cent* or clinical decision making or health care setting? or nursing research or (clinical adj2 practice) or (p?ediatric adj2 care)).ab,id,ti.	227858
4	(participat* or voice or feedback or feeling? or whish* or view? or perspective? or involv* or opinion or engagement).ab,id,ti. or client participation/	1306499
5	1 and (2 or 3) and 4	2601
6	(Lecture or Medical sudent or director or Dean or Educator or Adult service or Curriculum or Profession or Philosophy or mental health problem or Mental illness or Child psychiatry or Colleague or Psychiatry or psychiatrist or adolescent psychiatry or Art or Health promotion or Court or Labor or Breast cancer or child Birth or Mental disorder or Student or National institute or Abortion or Public Health or Gynecologist or pregnant wom* or first month).ab,ti. [VOS red irrelevant terms]	546377
7	(New born or Autism or Psychosis).ab,ti. [VOS green irrelevant terms]	87189
8	(Psychiatric hospitalization or Smoking or Schizophrenia or Psychopathology).ab,ti. [VOS blue irrelevant terms]	198247
9	(Terrain vehicle or atv or Motor vehicle collision or alcohol or substance or substance abuse or suicide or Suicide attempt or Abuse or Abused Child or Child abuse or "Substance use" or Street or rural area or City or Mechanism or Cent or product or Juvenile justice system).ab,ti. [VOS yellow irrelevant terms]	480436
10	Breastfeeding.ab,ti. [VOS pink irrelevant terms]	4144
11	or/6-10	1168655
12	5 not 11	1761
13	limit 12 to ("0100 journal" or "0110 peer-reviewed journal" or "0400 dissertation abstract")	1667

Supplement 2 - Characteristics of the included studies

First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Abrines Jaume [1]	2015	United Kingdom	36	Multiple disease groups	4	13	2	Intervention development
Adams [2]	2010	Canada	80	Multiple disease groups	5	18	1,4	Clinical care
Akard [3]	2013	United States	8	Oncology	7	12	1	Intervention development
Alderfer [4]	2017	United States	12	Oncology	12	25	1	Clinical care
Alderson [5]	2006	United Kingdom	24	Diabetes	3	12	1	Clinical care
Alvarez [6]	2020	United States	8	Hospital admission	7	15	1	Clinical care
Ammerlaan [7]	2015	The Netherlands	13	Juvenile Idiopathic Arthritis	16	25	1	Clinical care
Anderson [8]	2017	United Kingdom	6	Transplantation	15	18	1	Clinical care
Anderzén-Carlsson [9]	2012	Sweden	6	Oncology	14	16	1	Clinical care
Angstrom-Brannstrom [10]	2008	Sweden	7	Multiple disease groups	4	10	1,3	Clinical care
Applebaum [11]	2013	United States	20	Multiple disease groups	13	21	2	Clinical care
Arruda-Colli [12]	2015	Brazil	8	Oncology	5	12	3	Clinical care
Arvidsson [13]	2016	Sweden	5	Oncology	6	11	1,3,8	Intervention development
Bagley [14]	2007	United States	42	Multiple disease groups	5	15	1	Research
Barned [15]	2018	Canada	25	Gastrointestinal diseases	10	17	1	Research
Baron [16]	2011	Australia	26	Oncology	4;6	8;2	1	Clinical care
Beck [17]	2014	Canada	7	Hematological disorders	10	18	2	Clinical care
Belsky [18]	2020	United States	22	Oncology	15	30	1	Intervention development
Belsky [19]	2020	United States	20	Oncology	10	28	1	Clinical care
Bemmels [20]	2013	United States	10	Surgery	12	18	1	Clinical care
Berkwitt [21]	2015	United States	22	Multiple disease groups	7	18	1	Clinical care
Bice [22]	2019	United States	16	Hospital admission	4	8	1	Intervention development
Black [23]	1995	Australia	104	Hospital admission	12	18	1	Clinical care
Board [24]	2005	United States	21	Hospital admission	7	12	1	Clinical care
Boisen [25]	2015	Denmark	NR	Oncology	NR	NR	8	Clinical care

Supplement 2 - *(continued)*

First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Boyd [26]	1998	Canada	6	Multiple disease groups	10	13	1,3,6	Clinical care
Brady [27]	2009	United Kingdom	22	Hospital admission	7	12	1,3	Clinical care
Bray [28]	2007	United Kingdom	7	Surgery	13	16	1,6	Clinical care
Bray [29]	2019	United Kingdom	32	Multiple disease groups	8	12	1	Clinical care
Bray [30]	2019	United Kingdom	106	Multiple disease groups	8	12	1,3	Clinical care
Bray [31]	2020	United Kingdom	80	Multiple disease groups	8	12	3	Intervention development
Breitwieser [32]	2014	United States	18	Transplantation	4	25	1,2,4	Clinical care
Broome [33]	2003	United States	23	Multiple disease groups	8	22	1	Research
Brorsson [34]	2017	Sweden	13	Diabetes	11;5	18	1	Clinical care
Bsiri-Moghaddam [35]	2011	Iran	12	Surgery	7	11	1	Clinical care
Buckley [36]	2010	Ireland	9	Surgery	6	9	1,3	Clinical care
Burnfield [37]	2019	United States	15	Mobility problems	3	11	8	Clinical care
Byczkowski [38]	2010	United States	170	Multiple disease groups	11	17	1	Clinical care
Cahill [39]	2019	Australia	200	Multiple disease groups	12	20	8	Clinical care
Callery [40]	2013	United Kingdom	13	NR	NR	NR	1	Clinical care
Cameron [41]	1983	United States	1	Oncology	11	11	8	Intervention development
Carpenter [42]	2014	United States	296	Asthma	8	16	1	Clinical care
Castensoe-Seidenfaden [43]	2016	Denmark	9	Diabetes	15	19	1,4	Clinical care
Childerhose [44]	2018	United States	7	Obesity	16	21	1	Clinical care
Christofides [45]	2016	Canada	19	Cystic Fybrosis	8	18	1	Research
Chuong [46]	2019	Canada	28	Gastrointestinal diseases	9	17	1	Clinical care
Clark [47]	2019	United States	40	Hospital admission	6	17	1,3	Clinical care
Clarke [48]	2020	Ireland	5	Hospital admission	10	11	2	Intervention development
Clift [49]	2007	United Kingdom	6	Hospital admission	11	15	1	Clinical care
Coad [50]	2008	United Kingdom	30	Hospital admission	3	18	1	Clinical care

Supplement 2 - *(continued)*

First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Corbo [51]	1985	United States	14	Patients on a ventilator	12	16;9	1	Clinical care
Corsano [52]	2015	Italy	27	Multiple disease groups	6	15	1	Clinical care
Coyne [53]	2006	United Kingdom	11	Multiple disease groups	7	14	1	Clinical care
Coyne [54]	2006	United Kingdom	11	Multiple disease groups	NR	NR	1,5	Clinical care
Coyne [55]	2016	Ireland	20	Oncology	7	16	1	Clinical care
Coyne [56]	2014	Ireland	20	Oncology	7	16	1	Clinical care
Coyne [57]	2011	Ireland	55	Multiple disease groups	7	18	1,2	Clinical care
Coyne [58]	2012	Ireland	55	Multiple disease groups	7	18	1,5,8	Clinical care
Crnkovic [59]	2009	Croatia	190	Multiple disease groups	7	19	5	Clinical care
Dagg [60]	2020	Canada	7	Surgery	13	17	1	Clinical care
Davies [61]	2007	United States	4	Palliative care	6	17	1	Clinical care
Davies [62]	2005	United States	4	Palliative care	6	19	1	Clinical care
Deatrick [63]	1984	United States	24	Surgery	NR	NR	1	Clinical care
Dedding [64]	2015	The Netherlands	30	Diabetes	8	12	1,2	Clinical care
Dekking [65]	2015	The Netherlands	2	Oncology	13	18	1	Research
Dell'Api [66]	2007	Canada	5	Chronic pain	10	17	1	Clinical care
Dellenmark-Blom [67]	2016	Sweden	18	Surgery	8	17	2	Intervention development
Di Ciommo [68]	2012	Italy	20	Metabolic diseases	8	23	1	Clinical care
Dixon-Woods [69]	2002	United Kingdom	21	Asthma	8	16	1	Clinical care
Dowler [70]	2016	United Kingdom	25	Hospital admission	NR	NR	3	Clinical care
Earle [71]	2006	Canada	5	Multiple disease groups	4;5	17	1	Clinical care
Ekra [72]	2015	Norway	9	Diabetes	5	12	1,4	Clinical care
Elliott [73]	2019	Canada	2	Juvenile Idiopathic Arthritis	NR	NR	1,2	Research
Erdem Atak [74]	2019	Cyprus	31	Multiple disease groups	5	16	3	Clinical care
Fahner [75]	2020	The Netherlands	13	Palliative care	11	18	1,2	Intervention development

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Fallon [76]	2008	United Kingdom	7	Oncology	14	23	2	Intervention development
Farjou [77]	2014	Canada	200	Oncology	12	20	7	Clinical care
Ford [78]	2011	Australia	10	Surgery	6	12	1	Clinical care
Forsner [79]	2005	Sweden	7	Hospital admission	7	10	1	Clinical care
Foster [80]	2019	New Zealand	26	Hospital admission	5	15	3	Clinical care
Francis [81]	1988	United States	7	Multiple disease groups	12	16	8	Clinical care
Freeman [82]	2000	United States	7	Oncology	5	15	1,2	Clinical care
Gaab [83]	2013	New Zealand	7	Palliative care	9	18	6	Clinical care
Gagnon [84]	2008	Canada	15	Head / brain injury	12	16	1	Clinical care
Garth [85]	2009	Australia	10	Head / brain injury	8	12	1	Clinical care
Geehan [86]	2003	United Kingdom	1	Oncology	17	17	8	Clinical care
Gibson [87]	2009	New Zealand	23	Oncology	12	22	2	Clinical care
Gibson [88]	2010	United Kingdom	38	Oncology	4	19	1,2,3,8	Clinical care
Gillies [89]	2001	United Kingdom	351	Surgery	12	18	1	Clinical care
Gillis [90]	1990	Canada	30	Surgery	6	12	1	Clinical care
Gilljam [91]	2016	Sweden	20	Juvenile Idiopathic Arthritis	8	17	1,2	Clinical care
Gonzalez-Morkos [92]	2011	United States	6	Oncology	15	18	8	Clinical care
Grant [93]	2019	Canada	25	Gastrointestinal diseases	11	35	2,8	Research
Grinyer [94]	2009	United Kingdom	28	Oncology	16	24	1	Clinical care
Grover [95]	2013	India	12	Asthma	7	12	1	Clinical care
Haase [96]	2020	United States	14	Transplantation	13	22	1	Clinical care
Haase [97]	1994	United States	7	Oncology	5	18	1	Clinical care
Hall [98]	2001	United States	34	Multiple disease groups	8	22	1	Research
Hall [99]	2012	United Kingdom	17	Surgery	8	17	1,3,8	Clinical care
Han [100]	2011	China	29	Oncology	7	14	1	Clinical care

Supplement 2 - *(continued)*

First author	**Year**	**Country**	***N***	**Disease group**	**Age (min)**	**Age (max)**	**Used method***	**Setting**
Hanghoj [101]	2020	Denmark	12	Oncology	16	29	1	Intervention development
Happ [102]	2013	United States	11	Cystic Fibrosis	10	16	1	Clinical care
Hart [103]	2020	United Kingdom	18	Oncology	17	26	1	Clinical care / research
Hein [104]	2015	The Netherlands	35	Multiple disease groups	8	16	1	Research
Heiney [105]	1991	United States	NR	Oncology	13	22	2,8	Clinical care
Hellier [106]	1986	United States	20	Radiology	5	11	1	Clinical care
Hentinen [107]	1996	Finland	51	Diabetes	13	17	1	Clinical care
Hoffman [108]	2019	United States	21	Ear, nose and throat disorders	6	12	1	Intervention development
Horstman [109]	2002	United Kingdom	50	Multiple disease groups	6	10	1,3	Clinical care
Hsiao [110]	2007	United States	20	Palliative care	9	21	1	Clinical care
Huby [111]	2017	United Kingdom	26	Kidney diseases	5	17	1	Intervention development
Huijer [112]	2013	Lebanon	85	Oncology	7	18	1	Clinical care
Hutton [113]	2005	Australia	7	Multiple disease groups	13	18	1,3	Clinical care
Ingersgaard [114]	2017	Denmark	5	Oncology	14	19	1	Research
Jamalimoghadam [115]	2019	Iran	16	Multiple disease groups	12	18	1	Clinical care
Jamalimoghadam [116]	2019	Iran	13	Multiple disease groups	12	18	1	Clinical care
Jedeloo [117]	2010	The Netherlands	31	Multiple disease groups	12	19	1	Clinical care
Jelbert [118]	2010	United Kingdom	5	Chronic Fatigue Syndrome	13	18	1	Clinical care
Jeschke [119]	2020	Germany	101	Epilepsy	6	17	1	Clinical care
Jiang [120]	2020	Australia	14	Juvenile Idiopathic Arthritis	15	23	1	Clinical care
Jibb [121]	2017	Canada	16	Oncology	12	18	1,8	Intervention development
Johnson [122]	2008	United States	27	Kidney diseases	NR	NR	1	Clinical care
Jolley [123]	1992	United Kingdom	20	Surgery	NR	NR	7	Clinical care
Jones [124]	2001	United States	8	Multiple disease groups	13	17	2	Research
Jordan [125]	2019	United States	19	Multiple disease groups	13	19	1	Clinical care

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Jordan [126]	2020	Canada	1	Unknown	17	17	8	Clinical care
Kada [127]	2019	Norway	22	Radiology	8	16	1	Clinical care
Karande [128]	2005	India	50	Hospital admission	5	12	1	Clinical care
Karnieli-Miller [129]	2009	Israel	13	Gastrointestinal diseases	9;6	16;6	1	Clinical care
Kaziunas [130]	2016	United States	17	Transplantation	NR	NR	1	Clinical care
Kebbe [131]	2019	Canada	19	Obesity	13	17	1	Clinical care
Kebbe [132]	2019	Canada	19	Obesity	13	17	1	Clinical care
Kelly [133]	2017	United States	29	Oncology	9	17	1,8	Clinical care
Kelsey [134]	2007	United Kingdom	10	Multiple disease groups	13	16	1	Clinical care
Kiernan [135]	2005	Ireland	240	Multiple disease groups	7	16	7	Clinical care
King [136]	2020	Canada	10	Neuromuscular disorders	8	18	1	Clinical care
Klassen [137]	2015	Canada	38	Oncology	15	26	1	Intervention development
Kleye [138]	2020	Sweden	13	Multiple disease groups	4	12	1,8	Clinical care
Klosinski [139]	2015	Germany	22	Multiple disease groups	12	17	1,2	Intervention development
Klostermann [140]	2005	United States	54	Multiple disease groups	11	19	2	Clinical care
Koller [141]	2016	Canada	26	Multiple disease groups	5	18	1	Intervention development
Koller [142]	2018	Canada	14	Multiple disease groups	9	17	2	Clinical care
Koller [143]	2010	Canada	21	Multiple disease groups	5	19	1	Clinical care
Koller [144]	2006	Canada	5	Severe Acute Respiratory Syndrome	6	18	1	Clinical care
Kortesluoma [145]	2008	Finland	44	Hospital admission	4	11	1	Clinical care
Korus [146]	2015	Canada	21	Transplantation	12	18	1	Clinical care
Kulandaivelu [147]	2018	Canada	19	Sickle cell disease	12	19	1	Clinical care
Lam [148]	2017	China	25	Oncology	9	18	1	Clinical care
Lambert [149]	2008	Ireland	49	Hospital admission	6	16	3,8	Clinical care
Lambert [150]	2013	Ireland	49	Hospital admission	6	16	3,8	Clinical care

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First author	**Year**	**Country**	***N***	**Disease group**	**Age (min)**	**Age (max)**	**Used method***	**Setting**
Laster [151]	2009	United States	19	Asthma	8	17	2	Clinical care
Leite [152]	2019	Brazil	16	Multiple disease groups	7	12	1	Clinical care
Lewis [153]	2013	United Kingdom	30	Epilepsy	13	19	1,2	Clinical care
Lindeke [154]	2006	United States	120	Hospital admission	4	20	7	Clinical care
Linder [155]	2021	United States	19	Oncology	6	12	1	Intervention development
Lindstrom Nilsson [156]	2019	Sweden	50	Multiple disease groups	3	18	1	Clinical care
Linge [157]	2012	Sweden	9	Multiple disease groups	2	18	1	Clinical care
Lipstein [158]	2013	United States	15	Multiple disease groups	11	18	1	Clinical care
Livesley [159]	2013	United Kingdom	15	Kidney diseases	5	15	5	Clinical care
Lock [160]	2010	United Kingdom	12	Ear, nose and throat disorders	4	16	1	Clinical care
Lööf [161]	2019	Sweden	22	Surgery	4	15	1	Clinical care
Lopez-Vargas [162]	2018	Australia	3	Multiple disease groups	8	14	2	Research
Loureiro [163]	2020	Portugal	252	Hospital admission	7	11	5	Clinical care
Luchtenberg [164]	2015	United Kingdom	25	Multiple disease groups	10	23	1	Research
Luchtenberg [165]	2020	United Kingdom / The Netherlands	52	Multiple disease groups	9	18	1	Research
Luseno [166]	2017	Kenya	29	Human Immunodeficiency Virus	15	19	1	Clinical care
Macartney [167]	2014	Canada	12	Oncology	9	18	1	Clinical care
Mansson [168]	2013	Sweden	12	Multiple disease groups	5	10	1	Clinical care
Mant [169]	2019	United Kingdom	6	Oncology	8	12	1	Clinical care
Marcinowicz [170]	2016	Poland	22	Multiple disease groups	10	16	1	Clinical care
Mari Ruberg Ekra [171]	2012	Norway	9	Diabetes	7	12	1,4	Clinical care
Martin-Kerry [172]	2019	United Kingdom	21	Multiple disease groups	6	19	1,2	Research
McCann [173]	2014	United Kingdom	12	Oncology	17	25	1	Clinical care
McDonald [174]	2006	United Kingdom	63	Hospital admission	2	15	3	Clinical care

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Miller [175]	1995	United Kingdom	7	Multiple disease groups	12	19	1	Clinical care
Miller [176]	2013	United States	20	Oncology	14	21	1	Research
Mirlashari [177]	2021	Canada	5	Oncology	13	15	1	Clinical care
Mitchell [178]	2014	United States	27	Degenerative disorders	13	22	1	Clinical care
Morgan [179]	2018	United Kingdom	3	Oncology	13	18	2	Clinical care
Morgan [180]	2009	United Kingdom	31	Oncology	13	24	7	Clinical care
Morgan-Trimmer [181]	2016	United Kingdom	7	Diabetes	7	16	1	Clinical care
Morrow [182]	2017	Australia	21	Multiple disease groups	7	17	1	Intervention development
Moules [183]	2009	United Kingdom	9	Hospital admission	12	16	1	Clinical care
Nahata [184]	2019	United States	14	Oncology	12	25	1	Research
Needle [185]	2020	United States	10	Transplantation	14	27	5	Clinical care
Nguyen [186]	2020	United States / Canada	32	Transplantation	12	24	2	Clinical care
Nicholas [187]	2007	Canada	9	Multiple disease groups	4	17	1	Clinical care
Nicholas [188]	2011	Canada	5	Multiple disease groups	4	17	1	Clinical care
Nilsson [189]	2010	Sweden	39	Trauma	5	10	1	Clinical care
Noyes [190]	2000	United Kingdom	18	Patients on a ventilator	6	18	1	Clinical care
O'Callaghan [191]	2011	Australia	26	Oncology	4;6	8;2	1	Clinical care
Omondi [192]	2013	United States	10	Sickle cell disease	12	16	2	Research
Orr [193]	2020	Canada	25	Migraine	10	18	1	Clinical care
Owen [194]	2013	United Kingdom	11	Obesity	5	18	1	Clinical care
Padding [195]	2018	The Netherlands	41	Asthma	5	6	1	Clinical care
Paquette [196]	2018	United States	20	Hospital admission	12	29	1	Research
Parsons [197]	2018	United Kingdom	63	Juvenile Idiopathic Arthritis	11	24	2	Research
Patterson [198]	2015	United States	29	Sickle cell disease	10	29	1	Research
Pearce [199]	2016	United Kingdom	21	Oncology	15	24	1	Research

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Pelander [200]	2004	Finland	40	Multiple disease groups	4	11	1	Clinical care
Pelander [201]	2007	Finland	388	Hospital admission	7	11	3,7	Clinical care
Pelander [202]	2009	Finland	388	Hospital admission	7	11	1,3	Intervention development
Pelander [203]	2010	Finland	388	Hospital admission	7	11	5	Clinical care
Pena [204]	2014	Spain	30	Multiple disease groups	8	14	1	Clinical care
Penza-Clyve [205]	2004	United States	36	Asthma	9	15	2	Clinical care
Perlman [206]	1987	Canada	25	Multiple disease groups	4	7	1	Clinical care
Perrott [207]	2017	United Kingdom	249	Surgery	NR	NR	7	Clinical care
Petronio-Coia [208]	2020	United States	7	Oncology	8	12	1	Clinical care
Pflugeisen [209]	2019	United States	7	Oncology	16	34	8	Clinical care
Picchietti [210]	2010	United States	33	Restless Legs Syndrome	6	17	1	Intervention development
Pichini [211]	2016	Canada	11	Genetic disorders	12	18	1	Clinical care
Polkki [212]	1999	Finland	20	Multiple disease groups	7	11	1,8	Clinical care
Pope [213]	2018	Australia	15	Acute pain	4	8	1,3	Clinical care
Pradel [214]	2001	United States	32	Asthma	7	12	1,3	Clinical care
Preti [215]	2011	Italy	6	Multiple disease groups	NR	NR	1	Clinical care
Przybylska [216]	2019	United Kingdom / Australia / United States	9	Hospital admission	3	15	1	Clinical care
Radovic [217]	2018	United States	31	Multiple disease groups	13	18	1	Clinical care
Ramsdell [218]	2016	United States	10	Trauma	8	16	1	Clinical care
Randall [219]	2012	United Kingdom	21	Multiple disease groups	6	12	1,2,4	Clinical care
Rasmussen [220]	2017	New Zealand	6	Hospital admission	NR	NR	1	Clinical care
Ray [221]	2017	United States	2	Multiple disease groups	14	21	1	Clinical care
Reverend Alister [222]	2007	United Kingdom	5	Multiple disease groups	8	11	8	Clinical care
Reynolds [223]	2007	United States	32	Diabetes	12	17	1	Research

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Roper [224]	2018	United Kingdom	16	Asthma	7	15	1	Clinical care
Rosenberg [225]	2016	United States	18	Oncology	14	25	1	Clinical care
Ruhe [226]	2016	Switzerland	17	Oncology	9	17	1	Clinical care
Ruhe [227]	2016	Switzerland	17	Oncology	9	17	1	Clinical care
Ruland [228]	2007	Norway	7	Oncology	8	12	1,2	Intervention development
Runeson [229]	2000	Sweden	26	Multiple disease groups	6	17	1	Clinical care
Sartain [230]	2000	United Kingdom	7	Multiple disease groups	8	14	1	Clinical care
Sartain [231]	2001	United Kingdom	11	Hospital admission	5	12	1,3	Clinical care
Savedra [232]	1988	United States	6	Multiple disease groups	13	16	1,4	Clinical care
Schalkers [233]	2015	The Netherlands	63	Multiple disease groups	6	18	1,4,5,8	Clinical care
Schmidt [234]	2007	United States	65	Multiple disease groups	5	18	1,7	Clinical care
Schmidt [235]	2012	United States	20	Diabetes	8	15	1	Clinical care
Schwellnus [236]	2020	Canada	10	Head / brain injury	6	19	1	Clinical care
Sease [237]	2021	United States	2	Obesity	6	18	1	Clinical care
Sepponen [238]	2003	Finland	31	Asthma	7	13	1	Clinical care
Shaw [239]	2006	United Kingdom	28	Asthma	NR	NR	1	Clinical care
Sherratt [240]	2018	United Kingdom	8	Juvenile Idiopathic Arthritis	8	16	1	Research
Sjoberg [241]	2015	Sweden	10	Surgery	8	11	1	Clinical care
Skolin [242]	2006	Sweden	22	Oncology	2	17	1	Clinical care
Sleath [243]	2016	United States	18	Asthma	11	17	2	Intervention development
Smeland [244]	2019	Norway	20	Surgery	8	16	1	Clinical care
Smith [245]	2005	United Kingdom	9	Surgery	7	11	3	Clinical care
Smith [246]	2019	United States	7	Oncology	7	17	2	Clinical care
Squitieri [247]	2013	United States	18	Nerve damage	10	17	1	Clinical care
Stegenga [248]	2013	United States	12	Sickle cell disease	6	14	1	Clinical care

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Stegenga [249]	2018	United States	17	Transplantation	9	18	1	Clinical care
Stegenga [250]	2008	United States	3	Oncology	13	15	1	Clinical care
Stegenga [251]	2009	United States	10	Oncology	13	17	1	Clinical care
Stevens [252]	2006	Canada	14	Oncology	7	16	1	Clinical care
Stevens [253]	1998	United States	59	Surgery	12	17	1	Clinical care
Stewart [254]	2005	United States	6	Oncology	8	16	1	Intervention development
Sutters [255]	2007	United States	80	Surgery	6	15	1	Clinical care
Taylor [256]	2010	United Kingdom	21	Multiple disease groups	7	16	1	Clinical care
Taylor [257]	2016	Australia	26	Head / brain injury	6	15	7	Intervention development
Tenniglo [258]	2017	The Netherlands	11	Oncology	12	18	2	Clinical care
Tercyak [259]	1998	United States	12	Diabetes	NR	NR	1	Clinical care
Tong [260]	2013	Australia	13	Juvenile Idiopathic Arthritis	14	19	1,2	Clinical care
Trace [261]	2020	United Kingdom	5	Kidney disease	6	14	1	Clinical care
Travlos [262]	2016	Australia	11	Neuromuscular disorders	14	21	1	Clinical care
Ullan [263]	2012	Spain	126	Multiple disease groups	14	17	4,7	Clinical care
Unguru [264]	2010	United States	37	Oncology	7	19	7	Research
Van Niekerk [265]	2020	South Africa	15	Burns	9	15	2	Clinical care
Van Staa [266]	2010	The Netherlands	9	Multiple disease groups	15	17	1	Research
Van Staa [267]	2011	The Netherlands	31	Multiple disease groups	12	19	1	Clinical care
Vaz [268]	2010	Congo	8	Human Immunodeficiency Virus	8	17	1	Clinical care
Vejzovic [269]	2015	Sweden	17	Gastrointestinal diseases	10	17	1	Clinical care
Viklund [270]	2009	Sweden	31	Diabetes	12	17	1	Clinical care
Visentin [271]	2006	Australia	10	Diabetes	15	18	1	Clinical care
Wangmo [272]	2017	Switzerland	17	Oncology	9	17	1	Clinical care
Weaver [273]	2015	United States	40	Oncology	12	18	1	Clinical care

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First author	Year	Country	*N*	Disease group	Age (min)	Age (max)	Used method*	Setting
Weaver [274]	2020	United States	15	Palliative care	NR	NR	1	Clinical care
Wennstrom [275]	2008	Sweden	20	Surgery	6	9	1,3	Clinical care
Wiener [276]	2014	United States	24	Transplantation	5	17	1	Intervention development
Wilkinson [277]	2003	United Kingdom	14	Oncology	15	20	1,2	Clinical care
Wilson [278]	2010	United States	93	Hospital admission	5	9	3	Clinical care
Wise [279]	2002	United States	9	Transplantation	7	15	1	Clinical care
Wollenhaupt [280]	2012	United States	25	Spina Bifida	12	21	1	Clinical care
Woltermann [281]	2020	Germany	100	Epilepsy	6	18	1,3	Clinical care
Woodgate [282]	1995	Canada	11	Acute pain	2	6;5	1	Clinical care
Woodgate [283]	1998	Canada	23	Multiple disease groups	13	16	1	Clinical care
Woodgate [284]	2010	Canada	51	Multiple disease groups	6	19	1,2	Research
Woodgate [285]	2014	Canada	13	Oncology	8	17	1,3,6	Clinical care
Woynarowska-Soldan [286]	2015	Poland	24	Multiple disease groups	14	15	2	Clinical care
Xie [287]	2016	China	21	Oncology	7	12	1	Clinical care
Zitzelsberger [288]	2014	Canada	11	Kidney diseases	7	17	3,8	Clinical care

* 1 = individual interview, 2 = focus group, 3 = draw & write/tell techniques, 4 = photo/video techniques, 5 = complete sentences, 6 = diary, 7 = questionnaire with open questions, 8 = other

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3

Including the voice of paediatric patients: Cocreation of an engagement game

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Abstract

Background

Engaging patients in health care, research, and policy is essential to improving patient-important health outcomes and the quality of care. Although the importance of patient engagement is increasingly acknowledged, clinicians and researchers still find it difficult to engage patients, especially pediatric patients. To facilitate the engagement of children and adolescents in health care, the aim of this project is to develop an engagement game.

Methods

A user-centred design was used to develop a patient engagement game in three steps: (1) identification of important themes for adolescents regarding their illness, treatment and hospital care, (2) evaluation of the draft version of the game and (3) testing usability in clinical practice. Adolescents (12-18 years) were engaged in all steps of the development process through focus groups, interviews or a workshop. These were audio-recorded, transcribed verbatim, and analyzed in MAXQDA.

Results

(1) The important themes for adolescents (N=15) were included: visiting the hospital, participating, disease and treatment, social environment, feelings, dealing with staff, acceptation, autonomy, disclosure and chronically ill peers. (2) Then, based on these themes, the engagement game was developed and the draft version was evaluated by 13 adolescents. Based on their feedback, changes were made to the game (e.g., adjusting the images and changing the game rules). (3) Regarding usability, the pilot version was evaluated positively. The game helped adolescents to give their opinion. Based on the feedback of adolescents, some last adjustments (e.g., changing colours and adding a game board) were made, which led to the final version of the game, *All Voices Count*.

Conclusions

Working together with adolescents, *All Voices Count,* a patient engagement game was developed. This game provides clinicians with a tool that support shared decision-making to address adolescents' wishes and needs.

Patient or Public Contribution

Paediatric patients, clinicians, researchers, youth panel of Fonds NutsOhra, and patient associations (Patient Alliance for Rare and Genetic Diseases, Dutch Childhood Cancer Organization) were involved in all phases of the development of the patient engagement game – from writing the project plan to the final version of the game.

Introduction

Nowadays, engaging patients in health care is central to improving health outcomes that matter to them [1,2]. In health care, the concept of patient engagement applies to involving patients in decisions about their daily clinical care while addressing patients' wishes and needs [1-6]. In day-to-day care, this means that patients are informed about the choice in treatment options to make decisions that are aligned with patients' preferences [3,4]. In addition, efforts are increasingly being made to engage patients at a broader level of health care, including the level of the hospital organization, research and policy [4,6,7]. The extent to which patients influence the decision-making processes varies from consultation to active partnership – and everything in between [4,8]. For example, studies showed the involvement of adolescent patients in designing a youth-friendly ward and identifying their preferences regarding a study design or measurement of outcomes [9-12]. In whatever shape, patient engagement benefits both patients and organizations: It not only improves the quality of care but also improves patient experience and self-confidence, resulting in better health outcomes and higher inclusion rates in research [2,3,7,13].

Although the benefits of patient engagement are beyond dispute, clinicians and researchers still struggle with engaging patients in health care and research [14,15]. Mentioned reasons are that clinicians doubt whether patients are knowledgeable [16], involving patients is time consuming [16,17] and scheduling meetings with groups of patients is difficult [18]. Involving paediatric patients seems to be especially challenging [19,20], as the competence of children to participate is even more questioned [21-23]. Also, the involvement of parents makes the process of engaging complex because of the paternalist approach to care [13,21,22]. Finally, clinicians have little experience in *how* to involve children in matters pertaining health care [22].

Boenink et al. [24] developed a tool to engage adults in translational research, *The Voice of Patients*. With this card game, patients can reflect on various topics regarding biomedical research. The uptake of the tool was positive, exceeding expectations from both patients and researchers [24]. However, an engagement tool for children and adolescents is missing, but would be valuable to facilitate engaging paediatric patients. Thus, to fill in this gap, the aim of this study is to develop a patient engagement game for adolescents with a chronic condition that can be used by clinicians and researchers to incorporate what matters to paediatric patients in hospital care, research and policy. This game was developed in cocreation with adolescents through three different steps: (1) identification of the most important themes for adolescents in health care and finding out preferences for patient engagement, (2) development and evaluation of the game and (3) test the game usability in clinical practice.

Methods

An user-centred design, as described in the literature by Gulliksen et al., [25], was used. Key principles of an user-centred design include *user-focused* and *active user involvement* throughout the entire development process. These principles were guaranteed by actively involving all representative users, including adolescents with a chronic condition, clinicians, researchers, the Patient Alliance for Rare and Genetic Diseases (VSOP), the Dutch Childhood Cancer Organization (VKN) and a youth panel of Fonds NutsOhra (FNO). This youth panel consists of adolescents with a chronic condition, who contributed with ideas and suggestions to several projects aiming to improve social engagement in health care. All representative users were involved in all phases of the design process - from writing the project plan to the final version of the game. Other principles, such as *prototyping* and *evaluate use in context*, were applied by developing, testing, and continuously adapting the draft versions of the game. In addition, the draft versions were tested at every stage of the development process with the end-users in a real-life context. For the development and design of the game, we collaborated with design agency Studio Dam (*professional attitude – www.studiodam.nl)*.

The development of the patient engagement game was an iterative process consisting of three steps (Figure 1):

1. Identification of import themes for adolescents regarding their illness, treatment, hospital care, and influence on daily life and preferences for an engagement game. The identified themes will serve as a starting point for the development of the patient engagement game.
2. Evaluation of the draft version of the game
3. Testing usability in clinical practice

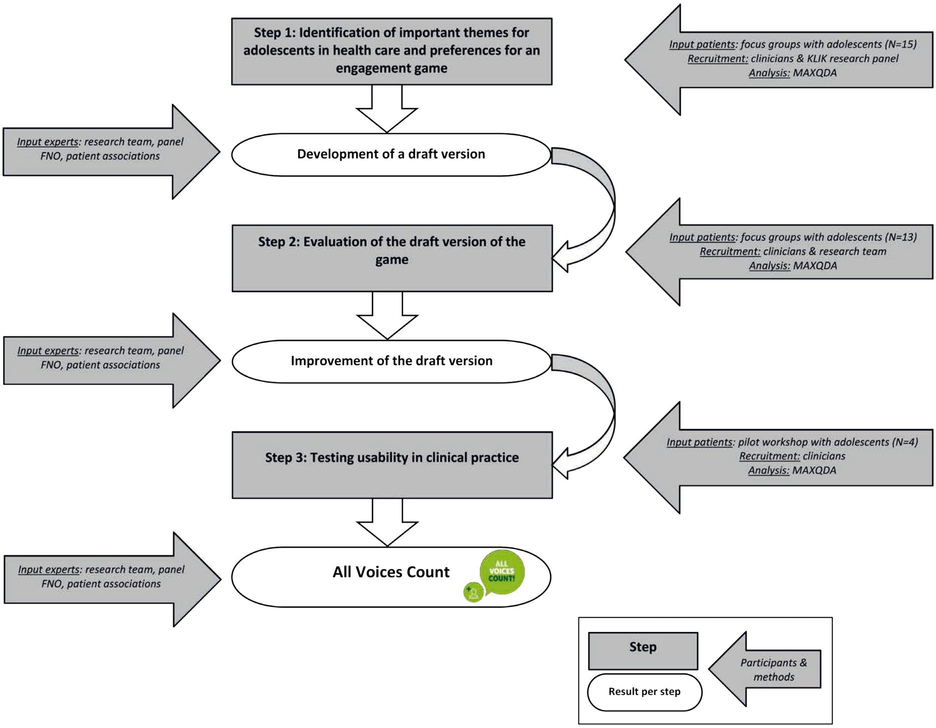

Figure 1. The development process of the game All Voices Count

Each step resulted in the development/improvement of the game and provided input for the next step.

Step 1: Identification of important themes for adolescents in health care and preferences for an engagement game

For the first step, adolescents (12-18 years) with a chronic condition, under treatment at the Emma Children's Hospital Amsterdam UMC, were invited to participate in 60-minute focus groups and individual interviews (30 – 60 min) to identify important themes for adolescents in health care. Adolescents were recruited for this study by their clinician in June and July 2017. Additionally, patients who were part of the research panel of the KLIK patient-reported outcome measures (PROM) portal (www.hetklikt.nu) were approached by the research team [26]. The KLIK research panel consists of patients who have indicated, during registration for the KLIK PROM portal, that they would like to be approached for research projects in the Emma Children's Hospital.

During the focus groups and interviews, the elicitation technique 'Complain and Cheer wall' [27] was used. Adolescents were invited to write down things they did not like about living with a chronic condition, their treatment and the hospital care on the 'Complain wall' and things they did like on the 'Cheer wall'. Thereafter, topics were discussed and grouped into themes by the adolescents and discussion leader following the Metaplan method (a workshop technique used to form a common understanding) [28]. In addition, adolescents were asked for their opinion regarding the development of a patient engagement card game. Data collection was continued until data saturation was reached. Data saturation was considered reached when no new themes emerged during the analyses of the focus groups. The result of these focus groups and interviews was a list of important themes for adolescents in health care and their preferences for an engagement game, which was used for the development of the first draft version of the game.

Step 2: Evaluation of the draft version of the game

In the second step, the first draft of the game was tested and evaluated with adolescents in a fictional context. Again, adolescents (12-18 years) with a chronic condition, under treatment at the Emma Children's Hospital Amsterdam UMC, were invited to participate in these focus groups and individual interviews. Adolescents were recruited from November 2017 till January 2018 in three ways: (1) participating adolescents in Step 1 were asked to participate again in this evaluation, (2) adolescents were approached by their clinician or (3) adolescents could sign up themselves after reading an information leaflet in the waiting room.

During 90-min focus groups and interviews (45-60 min), the game was played with the adolescents in a fictional context (opinion about the use of patient-reported outcome measures [PROMs]). Afterwards, adolescents were asked to evaluate the engagement game with the use of traffic light colours. Adolescents were invited to write down what they liked about the game (green), which parts of the game they were doubting about (yellow) and which parts of the game they did not like (red). Adolescents were asked to provide feedback on both the content and layout of the game. These topics were discussed, and adolescents were asked for suggestions for improvement and their opinion about specific aspects of the game (i.e., completeness of the included themes, desired game time and the use of photos or clip-arts). Data collection was continued until data saturation was reached. The result of these focus groups and interviews was a list of improvements for the game, which was used to develop a pilot version of the game.

Step 3: Testing usability in clinical practice

The third step involved usability testing of the pilot version of the game. In this field test, a pilot workshop was held with patients from the educational facility (educational support service for patients and their parents) of the Emma Children's Hospital Amsterdam UMC. Participating patients were recruited via clinicians of the educational facility in April 2018.

During the 90-min pilot workshop the game was played with the adolescents to answer a question from the educational facility: 'What can the educational facility do (even more) for you to ensure that things go even better at school?'. Afterwards, adolescents were asked to evaluate the engagement game using traffic light colours and were asked for suggestions for improvement. The result was that insight was gained into the usability of the engagement game and a list of improvements was obtained for the game, which was used to develop a final version of the game.

For all steps, participating adolescents and their parents (for adolescents <16 years) provided written informed consent and a sociodemographic questionnaire (i.e., age, gender, type of chronic disease) was completed by parents. Participants received a gift card (with an amount of 10 euro) and compensation for their travel expenses. Additionally, all focus groups, interviews and the pilot workshop were audio-recorded, transcribed verbatim, and analysed in MAXQDA [29] following the methodology for thematic analysis [30]. The focus groups, interviews, and pilot workshop were conducted by two members of the research team. These members have been trained in conducting qualitative research.

Results

The results are reported for every step of the development process. In total, 23 adolescents (range: 12-18 years, 57% female) participated in the cocreation of the patient engagement game, of whom nine adolescents participated in multiple steps (Table 1).

Table 1. Sociodemographic characteristics of participants in every step of the development process

	Step 1: identification of important themes (N=15)	**Step 2: evaluating draft version (N=13*)**	**Step 3: usability testing (N=4)**
	M (range)	M (range)	M (range)
Age (years)	15.0 (12-18)	15.5 (13-18)	14.5 (13-16)
	%	**%**	**%**
Gender (female)	60	61.5	50
Type of chronic disease	**%**	**%**	**%**
Cancer	20	23.0	100
Sickle cell disease	26.6	15.4	
Cystic fibrosis	13.3	15.4	
Juvenile idiopathic arthritis	13.3	15.4	
Kidney disease		15.4	
Chronic eczema	6.7	7.7	
Asthma		7.7	
Chronic pain	6.7		
Crohn's disease	6.7		
Muscular diseases	6.7		

* Nine adolescents also participated in Step 1

Step 1: Identification of important themes for adolescents in health care and preferences for an engagement game

In total, 15 adolescents (mean age: 15.0 years, range 12-18 years, 60% female) participated in four focus groups and four interviews (Table 1). Ten major themes for adolescents regarding their illness, treatment, and hospital care were identified: visiting the hospital, participating, disease and treatment, social environment, feelings, dealing with staff, acceptation, autonomy, disclosure and chronically ill peers (Table 2). Most of the adolescents liked the idea of a patient engagement card care. A few adolescents mentioned that they would prefer an online game. Based on the identified themes, a draft version of the game was developed by design agency Studio Dam, in consultation with the research team consisting of psychologists and medical doctors, representatives of the youth panel of FNO and the patient associations (VSOP, VKN).

Table 2. Overview of the identified themes and associated quotes (Step 1), and the adjusted names for the engagement game (Step 2)

Identified themes	Name in the game	Quotes
Visiting the hospital	My hospital	'I like the shops in the hospital' 'The things they organize for patients are very nice so I won't get bored'
Participating	I can (not) do this	'When I'm admitted to the hospital, it feels like I'm missing a few weeks of my life' 'I miss normal things, like hanging out with friends or going to school'
Disease & treatment	My disease & treatment	'I don't like that I am getting tired due to the antibiotics that I have to take' 'They made some mistakes in my treatment, for example once I got too much morphine'
Social environment	Me & others	'It is nice when people sympathize, because then I know that there are people who care about me' 'My friends always tell me that they can't imagine how it is to have juvenile arthritis'
Feelings	I feel this	'I was always really afraid that something was wrong when I got the results back' 'It feels very lonely when you think about your friends who are not sick'
Dealing with staff	The people in my hospital	'Sometimes doctors talk for hours and ask a lot of questions. I don't want that' 'I like doctors and nurses to be honest, don't tell me that it won't hurt if it will hurt'
Acceptation	I am okay	'I just want to be normal, I want to participate in class and not feel tired or sick' 'I feel a bit of an outsider'
Autonomy	I do (not do) it myself	'Because I am young, they don't take me seriously. That is annoying' 'I take care of myself'
Disclosure	Talk about it	'I'm not willing to tell my life story' 'I like to share my story'
Chronically ill peers	Just like me	'They understand me better than my normal friends' 'Kids who are sick too are more interested in my illness'

Note. All quotes were translated into English

Step 2: Evaluation of the draft version of the game

The opinion of 13 adolescents (mean age: 15.5 years, range: 13-18 years, 61.5% female, Table 1) was asked about the draft version (Figure 2) of the engagement game in three focus groups and five interviews. Overall, the adolescents were positive about the game as it gave them the opportunity to get involved and it helped them to express their views. They indicated that the use of themes and images made it easier for most adolescents to associate and think of other topics to express their opinion on. The game element was appreciated; it was fun, exciting and motivates competition. Furthermore, the design of the game was attractive and easy to play. Suggestions for improvement were about the explanation of the game and the images on the playing cards. Although adolescents mentioned a preference for images rather than clip-arts, the majority mentioned that the images on the cards were not clear and that the persons in the images were too old, preventing them to relate to the depicted situation. Therefore, we adapted the images and tried to match the age group of 12-18 years. A few adolescents suggested the

First version of the game

Pilot version of the game

Final version of the game All Voices Count

Figure 2. Overview of the different versions of the engagement game during the development process

addition of keywords about the situations to the playing cards, but we decided not to because it can reduce the possibility of free association. Furthermore, adolescents indicated that a map with an overview of the themes would be helpful. In addition, a few adolescents made some suggestions to improve the design of the game, for example changing the rules of the game or create an online version. These suggestions were discussed with the research team, the design agency and the representative of the youth panel. We decided to change the images, the lay-out of the playing cards, the rules of the game and the game explanation, and we added an personal overview card of the themes and associated subthemes. This card clarifies to adolescents what kind of subthemes are related to the specific themes (Table 3). Finally, we discussed the naming of the themes within the project group, as we noticed that the naming did not always match the perception of adolescents. We decided to rename the themes to make them more appealing and understandable for adolescents: my hospital, I can (not) do this, my disease & treatment, me & others, I feel this, the people in my hospital, I am okay, I do (not do) it myself, talk about it and just like me (Table 2).

Step 3: Testing usability in clinical practice

The pilot version (Figure 2) of the game was tested for usability by four patients (mean age: 14.5 years, range 13-16 years, 50% female, Table 1) in clinical practice. During this pilot workshop, the adolescents gave their opinion about a question of the educational facility, and a report on this workshop has been presented to the education facility to help them improve their daily clinical care. At the end of the workshop, the adolescents gave their opinion about the engagement game. All adolescents were enthusiastic about the game and enjoyed giving their opinion. Although it was difficult for some adolescents to give an opinion on all themes, the cards helped adolescents to come up with ideas about topics to talk about. The adolescents mentioned that not all themes were applicable for the educational facility. Therefore, we added some instructions to the game manual for professionals about the selection of themes and the minimum number of themes to be included in the game (Table 3). Furthermore, adolescents suggested the addition of keywords about the situations to the playing cards. In consultation with the research team, we decided to add the theme name to all playing cards. Adolescents evaluated the overview card of the themes (as developed and added after step 2) positively; however, we noticed that these personal maps were distracting and reduced the group feeling. By introducing the themes on a game board (Table 3), the focus of all players is on the game, and they are invited to help each other as not all words are visible from every corner.

Table 3. Overview of the feedback of adolescents and the adjustments that were made to the game (step 2 & 3)

Step 2: evaluating draft version		**Step 3: pilot workshop**	

*Points of improvement**
• Images on the playing cards
  · It is not always clear which situation is depicted in the images
  · The images are too much about the same situations
• Explanation
  · The explanation of the game is not clear enough
	Changes
	• We changed the images on the playing cards so that the depicted situations are easier to interpret and are more in line with the perceptions of adolescents
	• We have rewritten the explanation of the game
		Points of improvement
		• Images on the playing cards
		  · Not all images apply to every subject
			Changes
			• We agree with the adolescents that the images and themes are not relevant for every question. Therefore, we added instructions to the game manual about omitting irrelevant themes

*Tips for improvement**
• Images on the playing cards
  · Add keywords to the playing cards about the depicted situation or theme
	Changes
	• We did not add keywords to the playing cards as we believed that this would hinder adolescents from associating freely
		Tips for improvement
		• Images on the playing cards
		  · A keyword on the playing cards would be helpful
			Changes
			• We added the name of the theme to every playing card. We did not add keywords about the depicted situation, because we believed that this would hinder free association

• Game design
  · Increase the tension during the game by pressing the bell with 3 equal cards instead of 5
  · Create an online version of the game
	• We changed the rules of the game (i.e. the bell may be pressed when there are 3 cards of the same colour on the table). For financial reason, we did not develop an online version of the game		

• Lay-out
  · Develop an overview card of the themes
	• We developed an overview card of the themes and improved the lay-out of the pilot version	• Lay-out
		  · The colours of the cards can be brighter
			• We changed the colours of the cards and tried to use brighter colours

Table 3. *(continued)*

Step 2: evaluating draft version		**Step 3: pilot workshop**	
*Good points**		*Good points*	*Changes*
• Giving their opinion · The game helps to give an opinion · The game helps to think about other topics when expressing an opinion · It is fun to give your opinion using a game		• Giving their opinion · It is nice to be able to give your opinion	
• Images on the playing cards · The images on the playing cards are clear and helps to associate		• Images on the playing cards · The cards help to come up with ideas about topics you can say something about	
• Game element · The bell is a fun game element · The game motives to want to win			
• Difficulty level · The game is easy to play		• Difficulty level · The game is easy to play	
• Lay-out · The design of the game and the used colours are attractive		• Overview card of the themes · The additional words (subthemes) in the overview card were helpful	• We changed the overview card into a game board
		• Duration of the game · The duration of the game was good	

*Mentioned by adolescents in 2 or more focus groups / interviews

Finally, we changed the colours (brighter colours) of the themes and cards and after that a final version of the engagement game, called *All Voices Count,* was ready (Box 1). To support clinicians with the use of *All Voices count*, we developed a game manual, a website (www.allestemmentellen.nl), and a training.

Box 1. Game rules *All Voices Count*

All Voices Count

All Voices Count is a patient engagement game that helps clinicians to engage adolescents (12-18 years) with a chronic condition in their hospital care, research or policy. The game is played at the initiative of a clinician and before the meeting of All Voices Count the clinician prepares a 2-4 min video pitch, in which the question on which the clinician would like to hear the opinion of adolescents is presented to the participants. This question is the central topic of the meeting.

Examples of questions:

- *What do you think of this new treatment?*
- *What does it mean for you to have a coagulation disease and how can we help you?*

All Voices Count is accompanied by a game leader. The clinician (of the adolescent) is not present during the meeting to avoid that adolescents are being inhibited in expressing their opinion.

The course of the game

The game starts with a short introduction in which the participants get te know each other and watch the pitch of the clinician. Then, the game starts:

1. In turns, players turn over the top card of their deck and place it face upwards anywhere on the game board. Each subsequent card is placed anywhere on the game board, so that all card played remains visible for every player.
2. When three cards of the same colour (same theme) are visible on the game board, every player may press the bell. The player who presses the bell first wins all the cards of the same colour (same theme) that are visible on the game board.
3. The player who wins the cards may give his or her opinion first on the question posed by the clinician, regarding to the theme of the cards won. To get ideas, the player may look at the pictures on the cards won or the words belonging to the theme on the game board. All other players are allowed to react and give their own opinion.
4. When the players are done talking about the cards won, a new round starts.

The game ends when all cards have been played or if the fixed play time has elapsed. The player with the most cards won is the winner of the game.

Additional remarks

- All Voices Count can be played with 3-6 adolescents.
- The game leader plays an important role in steering the meeting (e.g., to make sure that the question of the clinicians remains the central topic and that all participants get the opportunity to express their opinion) and to help the participants with expressing their opinion whenever they are struggling with this. The game leader does not express his or her own opinion.
- Not all themes are relevant for every question. The game leader may decide to remove irrelevant themes. All Voices Count can be played with a minimum of five themes.

Discussion

Working together with adolescents, we co-developed and tested the usability of a paediatric patient engagement game, *All Voices Count*. This resulted in a valued tool that makes it easier for clinicians to include the input from paediatric patients in the decision-making process of hospital care, research and policy. Overall, adolescents were pleased with *All Voices Count* as it enables them to express their opinion and experiences regarding different topics in health care more easily.

The first step in developing *All Voices Count* was to identify important themes for adolescents regarding their chronic condition, treatment and hospital care. Further development of the game was based on these themes to connect to the perception of adolescents with a chronic condition. The identified themes were aligned with previous studies [31-33], in which participation in daily life, being normal, treatment, social environment and communication about their disease were also seen as important themes by adolescents with other conditions. This corroboration showed that adolescents, regardless of their chronic condition, face similar difficulties and supportive factors.

Since the development of *All Voices Count*, the game has been used to include the opinion of adolescents in several projects in our hospital. For example, *All Voices Count* was used during the development of an International Core Outcome Set for acute simple appendicitis in children [34]. With the use of *All Voices Count*, important outcomes for adolescents in determining the effectiveness of treatment were identified (*What do you think is important to know to make an informed choice between two treatments for appendicitis?)* and subsequently prioritized. In addition, we are planning to use *All Voices Count* for questions from physiotherapist of the department of oncology (*How can we make exercising more fun for you during treatment?*) to improve daily hospital care, for questions from researchers and clinicians from the haematology department (*What does it mean for you, as a girl, to have a coagulation disease and how can we improve the care?*), and for questions from clinicians and policy makers from the paediatric surgery department *(How should the follow-up programme look like and which themes should be discussed by the clinician?)* for setting up a new follow-up programme. Other purposes for which the game could be used are within the Kids Advisory Board of the children's hospitals and to discuss new research ideas with adolescents while writing a grant proposal. Engaging patients in the development of new research projects is increasingly mentioned as a requirement for research funding [35,36].

The next step is a further distribution and implementation of *All Voices Count* in other children's hospitals and rehabilitation centres in the Netherlands. Our goal is to bring *All Voices Count* to the attention of professionals working in different areas within the health care sector. We will therefore present *All Voices Count* at international conferences and to policymakers and division boards of hospitals in the Netherlands. Furthermore, we train clinicians in how to use the game and in the way in which they can use the results obtained in their daily clinical care, research or policy. To be able to deploy the game widely, we recently translated the Dutch version of *All Voices Count* into English.

The strengths of the user-centred design used in this study were that it provided insight into the perspective of the users and that it facilitated new ideas, so that it meet the needs of the users [25,37]. Especially, the input from adolescents was very valuable to us during the development process. Adolescents thought critically about the game and came up with valuable suggestions to improve the game. *All Voices Count* has been tested in a real-life context, making it usable and appropriate to the cultural context, and it increases the chances of a successful implementation [25,38].

Challenges or limitations in our user-centred design were the degree of influence and control of the participants and the representativeness [17]. While adolescents were involved throughout all phases of the development process, the research team included researchers, clinicians, representatives of a youth panel and patient associations, reviewed the final version. Regarding the representativeness, we invited adolescents with different chronic conditions to participate in this study. Now, during the evaluation of the pilot version of the game, only adolescents with cancer participated, which may have limited the representativeness of our study. However, this study showed that adolescents, regardless of their chronic condition, showed the same problems and supportive factors; therefore, we do not believe that this has influenced the results. Furthermore, earlier research showed that paediatric patients willing to participate in codesign studies tend to be more self-confident, critical and assertive adolescents, which can further hinder representativeness [17,39]. Finally, we tried to include the same adolescents in several steps of the development process to give them the opportunity to be a part of the project and to hear their views on the changes that were made based on their feedback. The engagement of adolescents multiple times can have advantages such as adolescents can express their views on the changes made to the game and are well-informed, and disadvantages like adolescents can express views that are a bit more one-sided, and fewer new ideas. During the different steps of the development process, we therefore included adolescents that did not participate in earlier steps as adolescents that participated in earlier steps.

Some barriers to engagement in this study included logistic difficulties related to travelling to the hospital, time constraints and difficulties in scheduling a meeting with a group of adolescents. These barriers have been mentioned by both adolescents and adults in other fields [7,18,40]. Developing an online version of the game could potentially reduce these barriers, according to the adolescents in our study. Adolescent indicated that the advantages of an online game are that they do not have to visit the hospital, that it takes less time and that they can fit it more flexibly into their time schedule. For this reason, we would like to develop an online version of *All Voices Count* in the future.

Conclusion

In conclusion, we developed a patient engagement game called *All Voices Count*, working together with all stakeholders. This game lowers the barrier to include the voice of adolescents in decision-making about hospital care, research, and policy.

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Ethics statement

All procedures performed in this study were in accordance with the ethical standards of the international and/or national research committee (Medical Ethics Committee of the Amsterdam UMC – W17_068#17.086) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in this study.

Conflict of interest

The authors declare that they have no competing interests.

Data availability statement

The data generated and analyzed during this study are available from the corresponding author on reasonable request.

Keywords
Adolescent, codevelopment, patient engagement, patient participation, paediatrics, shared decision-making, user-centred design

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Part 2

Patient Reported Outcome Measures

4

Clinicians' perspective on the implemented KLIK PROM portal in clinical practice

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Abstract

Purpose

Since 2011, the evidence-based KLIK Patient Reported Outcome Measure (PROM) portal has been implemented in clinical practice in > 20 Dutch hospitals. Patients and/or parents complete PROMs on Health Related Quality of Life, symptoms and psychosocial functioning before their outpatient consultation. Answers are converted into an ePROfile and discussed by clinicians during consultation to monitor well-being over time and detect problems early. This study aims to get insight into the KLIK implementation from the clinician's perspective.

Methods

As part of the KLIK implementation process, annual meetings were held with multidisciplinary teams to evaluate the use of KLIK. An online questionnaire was sent regarding (1) overall satisfaction, (2) feeling competent to discuss PROMs, (3) use of KLIK during the consultation, (4) influence of KLIK on the consultation, (5) usability of the KLIK PROM portal, (6) satisfaction with PROMs and feedback, and (7) support of the KLIK expert team. Open questions about (dis)advantages were included. Descriptive analyses were used.

Results

One hundred and forty-eight clinicians (response-rate 61%) from 14 hospitals in the Netherlands participated. Results show that: (1) clinicians report an overall satisfaction of median = 69/100 (visual analogue scale), (2) 85.8% feel competent discussing the ePROfile, (3) 70.3% (almost) always discuss the ePROfile, (4) 70.3% think that KLIK improves consultation, (5) 71.6% think KLIK is easy to use, (6) 80.4% are satisfied with the feedback of the overall KLIK ePROfile, 7) 71.6% experience sufficient support of the KLIK team.

Conclusion

Participating clinicians are generally satisfied with KLIK. Improvements to the KLIK PROM portal are now realized based on the mentioned disadvantages (e.g., shorten PROM completion by use of PROMIS and integrating KLIK with Electronic Health Records).

Introduction

In the past decades, there has been increased attention for the use of Patient Reported Outcome Measures (PROMs) in daily clinical practice enabling patient-centered care [1]. Discussing PROMs in the consultation room empowers patients, enhances patient-clinician communication and promotes shared decision making [2-5]. Monitoring patients by using PROMs increases awareness for patients' concerns, facilitates recognition of physical or psychological problems, improves patient satisfaction with health care and is associated with improved treatment outcomes, including survival [3, 4, 6-8].

After two efficacy studies [9, 10], the KLIK PROM portal (www.hetklikt.nu) is being implemented in daily clinical practice since 2011. These studies showed that the feedback and discussion of PROMs in the consultation room resulted in more attention for, and improved identification of, psychosocial and emotional problems and increased satisfaction of pediatricians with the provided care [9, 10]. Within the KLIK PROM portal, pediatric patients (≥ 8 years) and/or their parents and adult patients are asked to complete PROMs regarding Health Related Quality of Life (HRQOL), symptoms and/or psychosocial functioning online at home prior to the outpatient consultation. The answers are converted into an electronic PROfile (KLIK ePROfile, Figure 1) that contains a broad range of feedback options tailored to each specific PROM [11]. The clinician discusses the KLIK ePROfile during the outpatient consultation with patients and/or parents in order to monitor well-being over time, detect problems at an early stage and provide tailored advice and interventions. Currently, more than 17,000 patients from 70 different patient groups (e.g., rheumatology, diabetes, oncology) have registered themselves on the KLIK website and around 1,000 clinicians (e.g., physicians, nurses, psychologists, social workers, physiotherapists, dieticians, and speech therapists) have been trained (around 800 active users) in the use of KLIK in daily clinical practice in > 20 different hospitals in the Netherlands [12] and 3 hospitals in the United Kingdom (www.klik-uk.org).

Nevertheless, implementing a PROM portal in clinical practice is a challenging process in which the interests of different stakeholders are involved [12, 13]. For a successful implementation, different determinants can be distinguished on the level of intervention characteristics, the clinician, the patient (and parent), and the socio-political context. In the past years, the intervention characteristics of the KLIK PROM portal have been evaluated repeatedly and adapted so that identified barriers for implementation for this determinant have been addressed [12, 13].

1a

Child

○ 03-11-2016 ☑ 03-06-2017 ○ 23-12-2017 ☑ 26-04-2018

Physical	**03-06-2017**	**26-04-2018**
It is hard for me to walk more than one block	Sometimes ●	Never ●
It is hard for me to run	Often ●	Almost always ●
It is hard for me to do sports activity or exercise	Often ●	Often ●
It is hard for me to lift something heavy	Sometimes ●	Almost always ●
It is hard for me to take a bath or shower by myself	Never ●	Never ●
It is hard for me to do chores around the house	Almost always ●	Often ●
I hurt or ache	Almost never ●	Sometimes ●
I have low energy	Often ●	Sometimes ●
Emotional	**03-06-2017**	**26-04-2018**
I feel afraid or scared	Never ●	Never ●
I feel sad or blue	Never ●	Almost never ●
I feel angry	Almost never ●	Sometimes ●
I have trouble sleeping	Never ●	Sometimes ●
I worry about what will happen to me	Never ●	Never ●
Social	**03-06-2017**	**26-04-2018**
I have trouble getting along with other kids	Never ●	Never ●
Other kids do not want to be my friend	Never ●	Never ●
Other kids tease me	Never ●	Almost never ●
I cannot do things that other kids my age can do	Sometimes ●	Often ●
It is hard to keep up when I play with other kids	Never ●	Never ●
School	**03-06-2017**	**26-04-2018**
It is hard to pay attention in class	Never ●	Never ●
I forget things	Almost never ●	Sometimes ●
I have trouble keeping up with my schoolwork	Sometimes ●	Never ●
I miss school because of not feeling well	Sometimes ●	Sometimes ●
I miss school to go to the doctor or hospital	Sometimes ●	Sometimes ●

1b

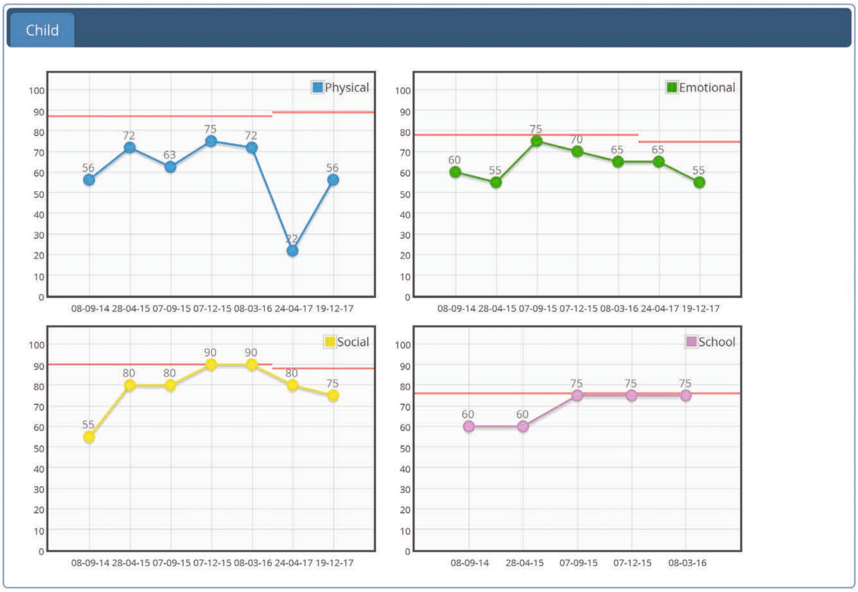


Figure 1. a KLIK ePROfile – literal feedback of the individual items on the Pediatric Quality of Life Inventory (PedsQL) **b** KLIK ePROfile – graphical feedback of the PedsQL, including norm lines

For example, PROMs are now available in multiple languages and KLIK has become an adaptable system to meet many individual wishes of the multidisciplinary teams [11]. However, little is known about barriers at the level of both clinicians and patients/parents. More insight is needed to fully understand the experienced barriers and to be able to optimize the KLIK PROM portal with regard to the wishes and needs of the user. Therefore, the aim of this study is to get more systematic insight into the experiences with KLIK from a clinician's perspective.

Methods

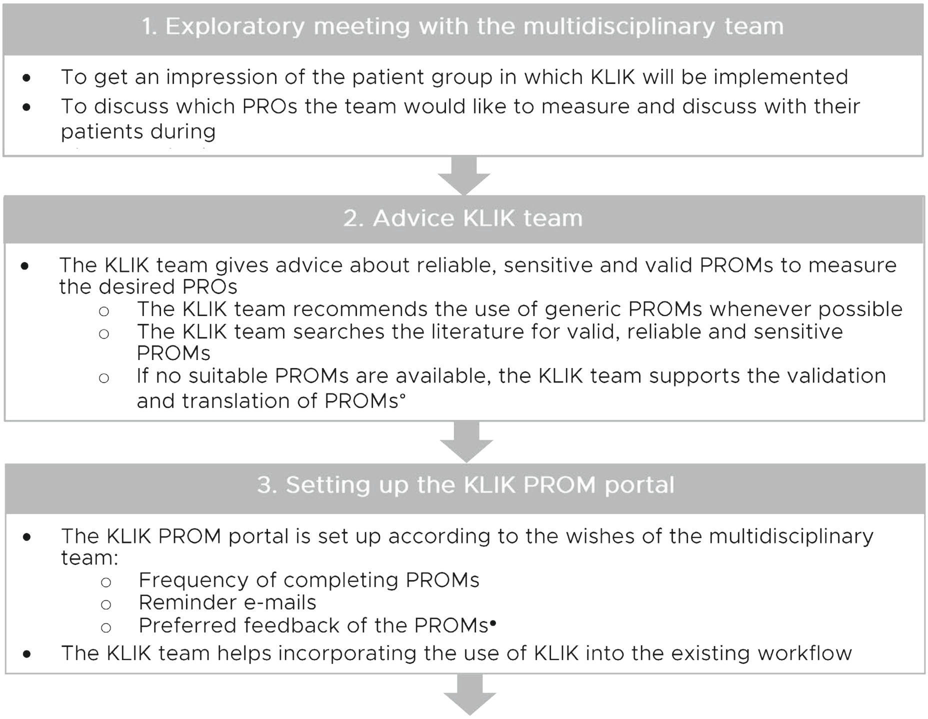
KLIK implementation process

KLIK can be implemented for every multidisciplinary team (e.g., diabetes, dermatology) in health care [12]. The implementation process starts at request of a multidisciplinary team and is guided by the KLIK expert team (consisting of researchers with expertise in the field of PROMs and HRQOL research) of the Emma Children's Hospital Amsterdam UMC through the following phases (Figure 2):

1. The KLIK expert team has an exploratory meeting with the clinicians of the multidisciplinary team to get an impression of the patient group and the Patient Reported Outcomes (PROs) they would like to discuss in the consultation room.
2. The KLIK expert team gives advice about reliable, sensitive and valid PROMs to measure the desired PROs. Whenever possible, PROMs with high reliability for specific populations and settings are selected to be able to use them on an individual level. However, sometimes the psychometric properties are not sufficient or unknown for the specific population, but no alternatives are available (e.g., in pediatrics, or in rare diseases).
3. The KLIK website is set up according to the wishes and workflow of the multidisciplinary team (e.g., frequency of completing PROMs, which reminder e-mails should be sent etc.). At this moment, over 300 PROMs have been built into the KLIK PROM portal. PROMs are offered to patients depending on age and patient group. Each member of the multidisciplinary team sees feedback of their preferred outcome measure set in a personal KLIK ePROfile.
4. Prior to the start of the implementation, all clinicians are trained in the use of KLIK in the consultation room. The 1.5 h training consists of a theoretical and a practical part. In the theoretical part, attention is paid to the definition of PROs and PROMs, the importance of discussing PROMs in the consultation room, and the use of the

KLIK PROM portal including the different feedback options. In the practical part clinicians are trained in discussing the KLIK ePROfile with patients [14].

5. Throughout the implementation process, the KLIK expert team acts as a helpdesk for both clinicians and patients. For example, the KLIK expert team supports the integration of KLIK into the existing workflow of a multidisciplinary team and helps patients and/or parents to log into the KLIK website and complete PROMs.
6. As standard part of the KLIK implementation process, the KLIK expert team offers annual one-hour evaluation meetings to multidisciplinary teams to evaluate the use of KLIK in daily clinical practice and to identify and overcome barriers in the implementation process.



1. Exploratory meeting with the multidisciplinary team

- To get an impression of the patient group in which KLIK will be implemented
- To discuss which PROs the team would like to measure and discuss with their patients during

2. Advice KLIK team

- The KLIK team gives advice about reliable, sensitive and valid PROMs to measure the desired PROs
    - The KLIK team recommends the use of generic PROMs whenever possible
    - The KLIK team searches the literature for valid, reliable and sensitive PROMs
    - If no suitable PROMs are available, the KLIK team supports the validation and translation of PROMs°

3. Setting up the KLIK PROM portal

- The KLIK PROM portal is set up according to the wishes of the multidisciplinary team:
    - Frequency of completing PROMs
    - Reminder e-mails
    - Preferred feedback of the PROMs•
- The KLIK team helps incorporating the use of KLIK into the existing workflow

4. Training clinicians

- All clinicians are trained in the use of KLIK in the consultation room, prior to the start□
- The training (1.5 hour) consists of two parts:
    - Theoretical part
        - Background of using PROMs in clinical practice
        - Use of the KLIK PROM portal for both clinicians and patients
        - Feedback options in the KLIK ePROfile
    - Practical part
        - Video material is used to show examples of clinicians discussing the KLIK ePROfile with patients
        - Support tools (decision tree & summary of the KLIK ePROfile) are provided to clinicians

5. The implementation process

- Invitation letters are sent to (new) patients by the secretariat of the multidisciplinary team or KLIK team*
- Patients themselves create a KLIK account on the website and complete PROMs online at home. If patients do not have internet access at home, it is possible to complete PROMs at the outpatient clinic (98% of the Dutch households have access to internet~)
- Clinicians discuss the KLIK ePROfile with patients during the consultation
- For a follow-up visit, the consultation date is added to the KLIK account by the patient, secretariat or KLIK team*
- Automatic e-mails are sent to patients when the PROMs are available
- If necessary, automatic e-mail reminders are sent to patients one day prior to the consultation

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- The KLIK team acts as a helpdesk during the implementation process
    - The KLIK team answers questions from both clinicians and patients
    - The KLIK team trains new clinicians in the use of KLIK
- The KLIK team helps incorporating the use of KLIK in the existing workflow
- The KLIK team facilitates the KLIK implementation process in the Amsterdam UMC

6. Evaluation

- One-hour annual evaluation meetings are held with the multidisciplinary team
    - Experiences with the use of the KLIK PROM portal
    - Workflow regarding the use of the KLIK PROM portal
- If possible, adjustments are made to the specific settings of the KLIK PROM portal

Note. °[15, 16], •[11], □ [14], *The KLIK implementation process is different for every multidisciplinary team depending on their wishes and workflow, ~[17]

Figure 2. Overview of the KLIK implementation process for one multidisciplinary team

Design and procedure

From February 2018 until August 2019, online evaluation questionnaires were sent out one week prior to each evaluation meeting. Reminder e-mails were sent to clinicians who had not completed the questionnaire one day before the meeting. The answers of the clinicians on the questionnaire on a team level provided a starting point for the evaluation meeting. Clinicians who had not completed the questionnaire prior to this meeting were asked to do so afterwards. This study has been approved by the Medical Ethics Committee of the Amsterdam University Medical Centers (Amsterdam UMC).

Participants

Two hundred and forty-three team members (independent of their presence during the evaluation meeting) of 36 multidisciplinary teams in 14 hospitals that use KLIK were approached to participate in this study prior to a KLIK evaluation meeting. Multidisciplinary teams who use the KLIK PROM portal only for scientific purposes (6 multidisciplinary teams), where the implementation process started less than a year ago (N=14) or teams that did not respond (N=14) were not eligible. Supplement 1 provides an overview of the inclusion process.

Measure

An evaluation questionnaire (Supplement 2) was developed to obtain the opinion of clinicians about the use of KLIK in daily clinical practice. The evaluation questionnaire was composed by four researchers of the KLIK expert team and reviewed by three nurses and one pediatrician. The questionnaire consisted of 20 closed questions (response options: three- and five-point Likert Scales, Visual Analogue Scales (VAS) and check boxes) and four mandatory open questions ((a) advantages and (b) disadvantages of KLIK, (c) incentives for patients and (d) frequently heard reactions of patients about KLIK) regarding (1) overall satisfaction, (2) feeling competent to discuss PROMs, (3) use of KLIK during the consultation, (4) influence of KLIK on the consultation, (5) usability of the KLIK PROM portal, (6) satisfaction with PROMs and feedback, and (7) support of the KLIK expert team. There was room to add a comment or explanation with each question. Since every multidisciplinary team uses a different subset of PROMs and feedback options, not all questions in the domain 'satisfaction with PROMs and feedback' could by answered be all clinicians.

Analysis

The Statistical Package for Social Sciences (SPSS) version 25.0 was used for descriptive statistics (percentages) to provide insight into the opinion of clinicians regarding KLIK and to study barriers and facilitators for the implementation process. Open questions of the evaluation questionnaire were analyzed qualitatively, by clustering the answers of all clinicians into main themes. This was done by two researchers (LT & HAvO) following the method for thematic analysis in Psychology [18]. Themes are ranked based on the number of times they have been mentioned by the clinicians (most often to fewest times).

Results

Participants

The online evaluation questionnaire was completed by 148 clinicians (61%), who were part of 36 different multidisciplinary teams from the following 14 different hospitals (Supplement 1): Emma Children's Hospital (N = 57 participating clinicians), Amsterdam UMC locations VU Medical Center (N = 24) and Academic Medical Center (N = 4), Kidz & Ko – diabetes collaboration centers (N = 18), Reade (N = 8), University Medical Center Groningen (N = 7), Spaarne Hospital (N = 6), VieCuri Medical Center (N = 6), Zuyderland Medical Center (N = 5), Maasstad Hospital (N = 5), Kempenhaeghe epilepsy center (N = 3), Sophia Children's Hospital (N = 2), Radboud University Medical Center (N = 2), and Wilhelmina Children's Hospital (N = 1). Discipline and disease group of participating clinicians are shown in Table 1. On average, participating clinicians used KLIK for 3.3 years (range 0.2-8.8 years). Most participating clinicians were employed as medical doctor (N = 57), psychologist (N = 39) or nurse (N = 36), and multidisciplinary teams were divided into pediatrics (32 teams) and adult health care (4 teams).

1. Overall satisfaction

Clinicians (N = 147) reported an overall satisfaction with the KLIK PROM portal of *median* = 69, range 13-100, on a VAS ranging from 0 (not satisfied) to 100 (very satisfied). One clinician could not fill in the VAS due to technical problems.

2. Feeling competent to discuss PROMs

Almost all clinicians (89.9%) indicated that the KLIK training had prepared them sufficiently to use KLIK in daily clinical practice (8.1% neutral, 2% disagree). In addition, 85.8% of the clinicians felt competent to discuss the KLIK ePROfile with patients and/or parents in the consultation room (7.4% neutral, 6.8% disagree).

Table 1. Characteristics of participants

	Participants (N = 148)
	N (% response-rate within discipline or group)
Discipline	
Medical doctor	57 (63.3)
Psychologist	39 (52.0)
Nurse	36 (66.7)
Dietitian	5 (71.4)
Physiotherapist	4 (100.0)
Social worker	3 (50.0)
Occupational therapist	2 (66.7)
Speech therapist	2 (100.0)
Disease group	
Diabetes (6 hospitals)	42 (63.6)
Juvenile Idiopathic Arthritis (2 hospitals)	12 (80.0)
Medical psychology (2 hospitals)	10 (52.6)
Sickle cell disease	9 (100.0)
Gender dysphoria	8 (27.6)
Coagulation diseases (4 hospitals)	7 (77.8)
Diagnostic Center Nutritional problems	6 (100.0)
Gastrointestinal diseases	6 (75.0)
Marfan syndrome	5 (100.0)
Neonatology follow-up	5 (71.4)
Spina Bifida	5 (55.6)
Cystic Fibrosis	4 (100.0)
Nephrology (2 hospitals)	4 (50.0)
Epidermolysis Bullosa	4 (44.4)
Surgery follow-up	4 (36.4)
Epilepsy	3 (75.0)
Human Immunodeficiency Virus	3 (50.0)
Congenital hand and arm disorders	2 (100.0)
Home Parenteral Nutrition	2 (66.7)
Metabolic diseases (2 hospitals)	2 (66.7)
Dermatology	2 (40.0)
Neurofibromatosis type 1	1 (100.0)
Muscle diseases	1 (50.0)
Endocrinology	1 (33.3)

3. Use of KLIK during the consultation

Table 2 gives an overview of the use of KLIK reported by the clinicians. Most clinicians (70.3%) indicated they discuss the KLIK ePROfile (almost) always with patients and/or parents, 18.2% reported to discuss the KLIK ePROfile sometimes and 11.5% indicated to (almost) never discuss the KLIK ePROfile. Reasons for not discussing the KLIK ePROfile with patients and/or parents, as indicated by clinicians in the comments section, were lack of time, PROMs not completed, forgot to discuss, technical problems, no priority, no problems reported in the KLIK ePROfile, the KLIK ePROfile was discussed by another team member or KLIK was no longer part of standard care. Clinicians indicated they discuss the KLIK ePROfile at the start (42.6%), middle (37.8%) or end (19.6%) of the consultation. Clinicians estimated that they spend on average 15% of the consultation (broad range of consultation time; 10–50 min) on discussing the KLIK ePROfile and 85.8% of the clinicians were satisfied with this percentage.

The majority of the clinicians (70.3%) invited all patients to participate in the KLIK PROM portal. Patients were not invited for the following reasons: absence of a chronic health condition, presence of a language barrier, a mental disability, illiteracy or not falling into a specific age range. In addition, clinicians mentioned they sometimes forgot to invite patients or they did not see it as their responsibility. 43.2% of the clinicians estimated that 75-100% of their patients and/or parents completed the PROMs. According to clinicians, reasons for not completing PROMs by patients were no Internet access, language barrier, forgetting and loss of motivation.

Table 2. Scores on the domain 'use of KLIK during the consultation' (N = 148)

			(Almost) always (%)	**Sometimes (%)**	**(Almost) never (%)**
Clinicians					
I discuss the KLIK ePROfile with patients/ parents			104 (70.3)	27 (18.2)	17 (11.5)
			Start (%)	**Middle (%)**	**End (%)**
I discuss the KLIK ePROfile at the … of the consultation			63 (42.6)	56 (37.8)	29 (19.6)
			Median (range)		
On average, I spend % of the consultation on discussion of the KLIK ePROfile (N=147)			15 (0-100)		
			Yes (%)	**No, I need more time (%)**	**No, I need less time (%)**
I am satisfied with the time I spent discussing the KLIK ePROfile			127 (85.8)	20 (13.5)	1 (0.7)
About patients			**Agree (%)**	**Neutral (%)**	**Disagree (%)**
All patients are invited to participate in the KLIK PROM portal			104 (70.3)	13 (8.8)	31 (20.9)
	100 (%)	**75 (%)**	**50 (%)**	**25 (%)**	**0 (%)**
I estimate that …% of patients/parents complete the PROMs	2 (1.4)	62 (41.8)	50 (33.8)	33 (22.3)	1 (0.7)

4. Influence of KLIK on the consultation

According to 70.3% of the clinicians, their consultation improved by the use of the KLIK PROM portal (24.3% neutral, 5.4% disagree) and 60.1% of the clinicians detected problems in functioning of patients and/or parents sooner (33.8% neutral, 6.1% disagree). Reasons for not detecting problems sooner with the use of KLIK were that another team member discussed the KLIK ePROfile with patients and/or parents or that the clinician was already aware of the functioning of the patients. Half of the clinicians (48.6%) indicated that they thought patients and/or parents were satisfied with the use of KLIK, 45.3% of the clinicians indicated that they did not know and 6.1% of the clinicians indicated that they thought patients and/or parents were not satisfied. Reasons why patients were not satisfied according to clinicians were: *many questions* (time intensive, having to complete PROMs too often, repetition in questions), *practical problems* (no Internet, login problems) and/or *no motivation* (annoying, no added value).

Regarding the open questions (Table 3), main advantages of KLIK for clinicians were: *insight in patient's functioning*, *improved communication*, *detecting problems*, *insightful feedback, patients being better prepared, easy to use, time saving,* and *clinician was better prepared.* Main disadvantages of KLIK for clinicians were: *low response-rate*, *takes time for clinician*, *irrelevant content of PROMs*, *complex procedure*, *technical aspects*, *no integration with Electronic Health Record (EHR)*, and *takes time for patients*. Table 3 shows the most important advantages and disadvantages of KLIK, expressed by clinicians.

According to clinicians, incentives for patients to use the KLIK PROM portal were: *insight in functioning* (reflection, awareness), *preparation for consultation* (time to think, conversation topics), *improved communication* (starting point for conversation, structure, comprehensive), *feeling heard* (being taken seriously, acknowledgement), *to be offered interventions in time* (signaling, intervene), and *empowerment* (involvement, request for help). Ten clinicians (6.8%) indicated that they do not know what the benefits for patients are.

Table 3. Advantages and disadvantages of KLIK and the use of PROMs, according to clinicians (N = 148)

Advantages of KLIK/PROM use	Examples
1. Insight in patient's functioning	'You quickly can get an impression of the things that are (not) going well' 'Monitoring the patient over time' 'Quick overview of how the patient is doing'
2. Improved communication	'The KLIK ePROfile structures the consultation' 'It provides a starting point for the conversation on difficult topics' 'Makes it possible to go in depth more quickly'
3. Detecting problems	'Problems are recognized earlier' 'It provides information about the disease/person that I would not have discovered otherwise' 'Standardized screening'
4. Insightful feedback	'Graphs provide insight' 'Convenient that scores are calculated directly and automatically' 'Better overview of the results through traffic light colors and graphs'
5. Patients being better prepared	'Provides patients the opportunity to think in advance about questions and concerns. They are not confronted with these during the consultation' 'Patients and parents talk to each other about items that matter' 'Patients think in advance about their own functioning and request for help'
6. Easy to use	'User-friendly' 'Accessible' 'Completing PROMs at home is easier for patients/parents'
7. Time saving	'The consultation is quicker' 'Saves time' 'As a clinician, it takes me less time than PROMs on paper'
8. Clinician was better prepared	'Better and more targeted preparation of the consultation' 'Prior to the consultation, I have important information from patient and parents' 'Before the consultation, I already have an impression of the complaints'
Disadvantages of KLIK/PROM use	**Examples**
1. Low response-rate	'Patients often do not complete PROMs' 'Patients with problems, for whom KLIK adds value, rarely complete the questionnaires' 'Reminders are necessary for patients to complete PROMs'
2. Takes time for clinician	'Extra time is needed to prepare the consultation' 'It takes time to discuss, since KLIK is not integrated into the EHR' 'Motivating patients to complete PROMs takes time'
3. Irrelevant content of PROMs	'Not all questions are relevant for every patient' 'Patients misunderstand questions' 'Many questions'
4. Complex procedure	'Patients lose username and password' 'PROMs are not easy to complete for parents with a cognitive disability or foreigners' 'Not all patients have access to Internet'
5. Technical aspects	'It takes effort to log in' 'I do not receive an automatic message when patients have completed PROMs' 'I have to print the KLIK ePROfile, because we do not have computers in the consultation room'
6. No integration with EHR	'The data from KLIK does not end up directly in the EHR' 'No integration with Epic©' 'Need to open a separate window, besides EHR'
7. Takes time for patients	'Requires time investment of patients' 'Patients indicate that they sometimes spend a long time completing PROMs' 'Extra burden for busy parents'

5. Usability of the KLIK PROM portal

According to 71.6% of the clinicians, the KLIK portal is easy to use (19.6% neutral, 8.8% disagree) and 83.8% of the clinicians indicated that the KLIK portal has an attractive lay-out (15.5% neutral, 0.7% disagree).

6. Satisfaction with PROMs and feedback

In general, 64.9% of the clinicians were satisfied with the selected PROMs (Table 4). Reasons why clinicians were not satisfied with the PROMs were too many PROMs, PROMs are not suitable for every patient and not all PROMs are available in multiple languages. Regarding the feedback of answers of the PROMs, 80.4% of the clinicians were satisfied with the feedback in the overall KLIK ePROfile. In the KLIK ePROfile the individual items in traffic light colors (Figure 1a) were viewed most frequently by the clinicians (84.7%). Of these traffic light colors, clinicians discussed the red answers most often with patients/parents (84.7%), followed by orange (58.4%) and green answers (34.3%). The graphs (scores over time resp. comparison with peers) are discussed by 47.4% resp. 33.6% of the clinicians. Clinicians thought that the traffic light colors of the KLIK ePROfile are most important (*median* = 72), followed by literal answers (*median* = 71) and graphs (*median* = 70) (Figure 1b), reported on a VAS, ranging from 0 (not important) to 100 (very important).

7. Support KLIK expert team

82.5% of the clinicians indicated to know where to ask their questions regarding the use of the KLIK PROM portal (10.1% neutral, 7.4% disagree) and 71.6% indicated that there is enough support from the KLIK expert team (25.7% neutral, 2.7% disagree).

Table 4. Scores on the domain 'satisfaction with PROMs and feedback'

	N	**Agree (%)**	**Neutral (%)**	**Disagree (%)**			
I am satisfied with the PROMs offered	134	87 (64.9)	36 (26.9)	11 (8.2)			
I am satisfied with the feedback of:							
Overall KLIK ePROfile	148	119 (80.4)	26 (17.6)	3 (2.0)			
Literal answers	148	112 (75.7)	33 (22.3)	3 (2.0)			
Traffic light colors	137	115 (83.9)	19 (13.9)	3 (2.2)			
Graphs (scores over time and comparison with peers)	137	105 (76.6)	25 (18.3)	7 (5.1)			
		Literal answers (%) (N=148)	**Traffic light colors (%) (N=137)**	**Graphs (%) (N=137)**			
I look at the following parts of the feedback in the KLIK ePROfile (multiple answers possible)		124 (83.8)	116 (84.7)	97 (65.5)			
			Literal answers			**Graphs**	
		Green answers (%)	**Orange answers (%)**	**Red answers (%)**	**Comparison with peers (graph) (%)**	**Scores over time (graph) (%)**	**Other (%)**
I discuss the following parts of the KLIK ePROfile (multiple answers possible)	137	47 (34.3)	80 (58.4)	116 (84.7)	46 (33.6)	65 (47.4)	24 (16.2)*
		Median (range)					
I think the following parts of the feedback of the KLIK ePROfile are important:							
Literal answers	147	71 (35-100)					
Traffic light colors	136	72 (11-100)					
Graphs	136	70 (12-100)					

* Other parts of the KLIK ePROfile that clinicians discuss with patients: open questions and changes in literal answers over time. A part of the clinicians does not discuss the KLIK ePROfile with patients.

Discussion

This study provided insight into the experiences of clinicians with the use of the KLIK PROM portal in daily clinical care, at a group level. Overall, clinicians were satisfied with discussing PROMs in the consultation room via the KLK PROM portal. Clinicians indicated that discussing PROMs helps them to gain more insight into patient functioning, to improve the communication with patients, to detect psychosocial or physical problems, and to empower patients. These benefits are in line with previous effectiveness studies [3, 4, 6]. In addition, clinicians valued specific characteristics of the KLIK ePROfile, such as ease of use and the well-developed and insightful feedback. Regarding this feedback, clinicians mentioned they appreciated and looked at the individual item feedback in traffic light colors most often. This preference was also found in previous research on the feedback of the QLIC-ON Profile [19].

Although clinicians indicated that the KLIK training sufficiently prepared them to use KLIK in clinical practice, they also indicated that the training did not fully meet their needs. More explanation about the interpretation of PROM results and the use of cut-off scores would increase their sense of competence. In addition, a refresher course every few years would be desirable. For this reason, the KLIK expert team is now revising the KLIK training. More information and tips and tricks about the interpretation and communication of PROM results will be included.

Clinicians indicated that they do not always discuss the PROMs with patients and/or parents due to lack of time, technical problems or lack of clarity regarding the workflow. For some clinicians it is unclear which team member of the multidisciplinary team discusses the PROMs with patients and/or parents or who sends invitations. This indicates that continuous support with the implementation process and annual evaluation meetings with all team members of a multidisciplinary team remains necessary. Also, patients do not always complete PROMs prior to the outpatient consultation. Forgetting, loss of motivation or no Internet access were reasons from the clinicians' perspective. In supporting the implementation process, a commonly heard argument from patients for not completing the PROMs is that the clinician does not discuss the PROMs during the consultation. This indicates how important it is for clinicians to discuss the PROMs with patients and/or parents. In addition, it was mentioned that for patients (or parents) with low health literacy skills and for non-native Dutch speakers it is sometimes difficult to complete the PROMs. Although the most frequently used generic PROMs in KLIK are available in multiple languages, this is not the case for all PROMs. When compiling the PROMs outcome sets with the multidisciplinary teams, more attention should also be paid to the needs of non-native Dutch speakers and patients with low health literacy skills.

Clinicians reported several main barriers for using PROMs via the KLIK portal. The first one is a lack of integration between KLIK and the EHRs. Opening a separate website to view the KLIK ePROfile is an added operation for clinicians, with the consequence that the KLIK ePROfile is sometimes not discussed with patients and/or parents. Therefore, in September 2019 a front-end integration with the two most often used EHRs in the Netherlands, Epic© and HiX© was realized in four hospitals. Clinicians can now view the KLIK ePROfile via the EHR, which increases the user-friendliness and makes it a better fit into the clinical workflow.

Second, clinicians indicated that they are not always satisfied with the content of PROMs. Reasons were mostly focused on the burden of completing PROMs for patients, such as a long completion time, many repetitions in questions and irrelevant questions. These challenges with PROMs correspond with previous research [20]. To address these problems, the National Institute of Health (NIH) developed the Patient-Reported Outcomes Measurement Information System (PROMIS) [21, 22]. PROMIS consists of various dynamic item banks (each measuring a separate construct) that can be administered through computerized adaptive testing (CAT) [20, 23]. By using a CAT, questions are offered based on the person's previous answer. In this way, patients and/or parents only have to answer a few questions per PROMIS construct to get a reliable score. As a result, the burden for patients and/or parents can be reduced [24]. Since November 2019, it is possible to administer the PROMIS item banks via KLIK, by linking KLIK with the Dutch Assessment Center. To realize this, the PROMIS item banks were translated and validated in the Netherlands [11, 16].

Third, clinicians mentioned that the use of PROMs is time intensive. Clinicians indicated that it takes more time to prepare themselves for the consultation and to discuss the PROMs in the consultation room. This is a remarkable finding, since previous research has shown that the use of the QLIC-ON Profile did not lengthen the consultation [9]. In addition, clinicians who are responsible for inviting patients for the KLIK PROM portal indicated that it takes a lot of effort to motivate patients to complete PROMs. A case manager that supports the KLIK implementation would be helpful.

There were a few limitations to this study. First, not all clinicians that use KLIK in the consultation room have been included in this study, because not all multidisciplinary teams were open to an evaluation meeting despite the importance for the implementation process. However, the experiences of clinicians from different disciplines, working with various disease groups in multiple hospitals and different outcome measure sets were included. Second, completing the VAS of the domains ‘overall satisfaction’ and ‘satisfaction with PROMs and feedback’ was not always possible when using a tablet. For these clinicians, it was not possible to move the bar to the desired position, causing a score around 50. Unfortunately, it could not be traced who had had this problem and therefore the results of these questions should be interpreted carefully. Third, the question ‘I am satisfied with the PROMs offered’ was not always understood by the clinicians. Prior to this question, there was a question about specific PROMs. The explanations showed that some clinicians referred to the specific PROMs when answering this question. That is why the answers to this question of 14 clinicians were not included. Fourth, due to the used method, this study provides no insight into the actions clinicians take with regard to the completed PROMs. In addition, no questions were asked about how clinicians use the information from the completed PROMs in daily clinical care. Therefore, recommendations for future PROM implementation research are to gain more insight into the actions of clinicians with regard to the discussed PROMs and how this can lead to more patient-centered care. The use of video observations in the consultation room may provide this information.

To conclude, the KLIK PROM portal is a valuable tool for clinicians to systematically monitor the functioning of their patients in clinical practice, so that extra support can be offered when needed. Overall, clinicians were enthusiastic about the feedback and user-friendliness of the KLIK PROM portal and the added value of using PROMs in clinical practice. However, some challenges and barriers were also identified. Therefore, a next step is to address the mentioned feedback points in the KLIK portal to improve the user-friendliness. Also, the perspective of the other user group, the patients and parents, is needed to further adapt the KLIK PROM portal to their wishes. Therefore, a similar study will be performed in the near future evaluating the KLIK PROM portal from the patients’ perspective, with the ultimate goal to further optimize the KLIK PROM portal and to improve the quality of health care.

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Ethical approval

All procedures performed in this study were in accordance with the ethical standards of the international and/or national research committee (Medical Ethics Committee of the Amsterdam UMC – W18_091 # 18.117) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in this study.

Conflict of interest

The authors declare that they have no conflict of interest.

Keywords

Patient Reported Outcomes, Patient Reported Outcome Measures, healthcare professionals, providers, implementation

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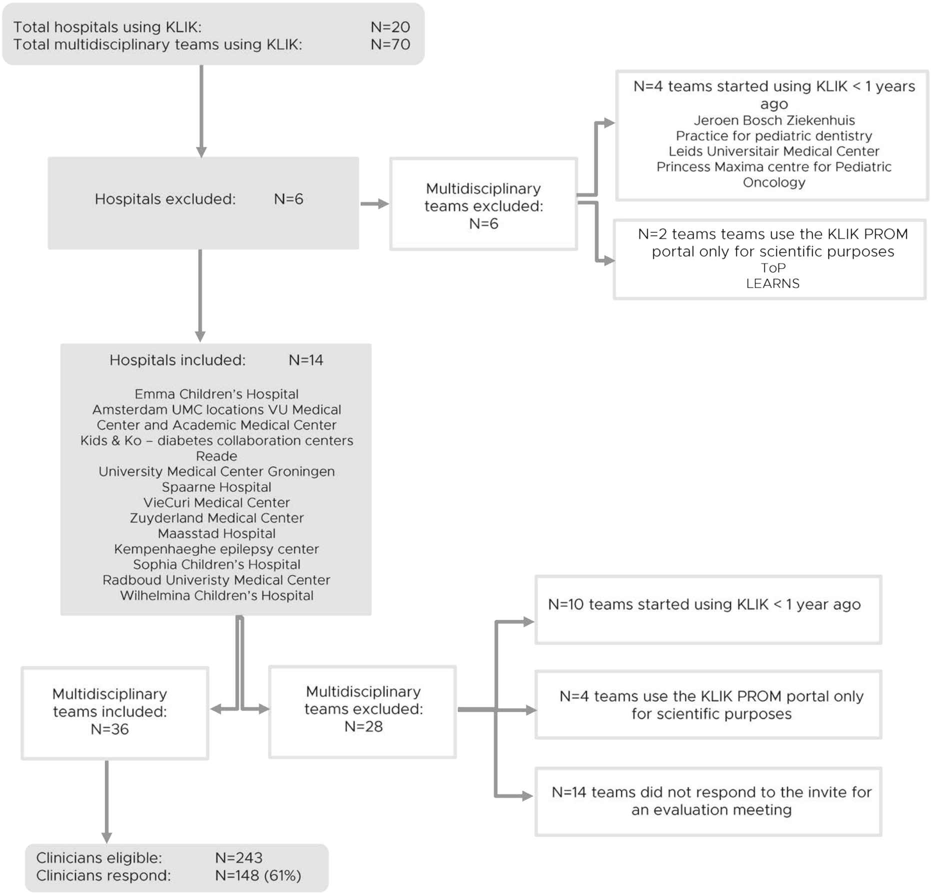
Supplement 1 - Flow diagram of participating hospitals, multidisciplinary teams and clinicians



Supplement 2 - Evaluation questionnaire KLIK

Evaluation questionnaire KLIK

Overall satisfaction

1. In general, how satisfied are you with the use of KLIK?

Not satisfied at all ………………………………. very satisfied *(VAS 0-100)*

Feeling competent to discuss PROMs

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
2. The KLIK training has prepared me sufficiently to start using KLIK Explanation:	☐	☐	☐	☐	☐
3. I feel competent to discuss the KLIK ePROfile with patients/parents Explanation:	☐	☐	☐	☐	☐

Use of KLIK during the consultation

4. I discuss the KLIK ePROfile with patients/parents ☐ Always ☐ Usually ☐ Sometimes ☐ Almost never ☐ Never
Explanation:

When: usually, sometimes, almost never and never, then:

4a. The reason why I do not always discuss the KLIK ePROfile is:
Open answer

5. I discuss the KLIK ePROfile at the … of the consultation ☐ Start ☐ Middle ☐ End
Explanation:

6. On average, I spend …. % of the consultation on discussing the KLIK PROfile
☐ I am satisfied with this
☐ I would like to spend more time on discussing the KLIK PROfile, because …
☐ I would like to spend less time on discussing the KLIK PROfile, because …

Patient	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
7. All patients are invited to participate in the KLIK PROM portal Explanation:	☐	☐	☐	☐	☐
When: strongly disagree, disagree, neither disagree, nor agree:					
7a. That not everyone is asked to participate is based on: *Open answer*					
8. I estimate that … % of the patients/parents complete the PROMs Explanation:	☐ 100%	☐ 75%	☐ 50%	☐ 25%	☐ 0%

Influence of KLIK on the consultation

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
9. KLIK improves my consultation Explanation:	☐	☐	☐	☐	☐
10. With the use of KLIK, I detect problems sooner Explanation:	☐	☐	☐	☐	☐

11. Advantages of KLIK are:
Open answer

12. Disadvantages of KLIK are:
Open answer

Patient	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
13. I think that patients/parents are satisfied with the use of KLIK Explanation:	☐	☐	☐	☐	☐

14. I think that incentives for patients/parents to use KLIK are:
Open answer

15. The most frequently heard reactions of patients/parents about KLIK are:
Open answer

Usability of the KLIK PROM portal

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
16. The KLIK PROM portal is easy to use Explanation:	☐	☐	☐	☐	☐
17. The KLIK PROM portal has an attractive lay-out Explanation:	☐	☐	☐	☐	☐

Satisfaction with PROMs and feedback

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
18. I am satisfied with the PROMs offered Explanation:	☐	☐	☐	☐	☐

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree

19. I am satisfied with the feedback of:

a. Overall KLIK ePROfile	☐	☐	☐	☐	☐
b. Literal answers	☐	☐	☐	☐	☐
c. Traffic light colors	☐	☐	☐	☐	☐
d. Graphs (scores over time and comparison with peers) Explanation:	☐	☐	☐	☐	☐

20. I look at the following parts of the feedback in the KLIK ePROfile (multiple answers possible) ☐ Literal answers ☐ Traffic light colors ☐ Graph ☐ Otherwise
Explanation:

21. I discuss the following parts of the KLIK ePROfile (multiple answers possible)
- ☐ Green answers
- ☐ Orange answers
- ☐ Red answers
- ☐ Comparison with peers (graph)
- ☐ Scores over time (graph)
- ☐ Other.....

22. I think the following parts of the feedback of the KLIK ePROfile are important:

a. Literal answers Not important at all ....Very important *(VAS 0-100)*

b. Traffic light colors Not important at all ....Very important *(VAS 0-100)*

c. Graphs Not important at all ....Very important *(VAS 0-100)*

Support KLIK expert team

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
23. I know where I can ask my questions regarding the KLIK PROM portal Explanation:	☐	☐	☐	☐	☐
24. There is sufficient support from the KLIK team when I need it Explanation:	☐	☐	☐	☐	☐

5

Patients' and parents' perspective on the implementation of Patient Reported Outcome Measures in pediatric clinical practice using the KLIK PROM portal

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Abstract

Introduction

The KLIK Patient Reported Outcome Measures (PROM) portal (www.hetklikt.nu) has been implemented since 2011 in clinical practice in over 20 Dutch hospitals. Patients and/or parents complete PROMs before the outpatient consultation and answers are subsequently discussed by clinicians during consultation. This study aims to provide insight into patients' and parents' perspective on the use of the KLIK PROM portal in order to optimize its implementation in pediatric clinical practice.

Methods

Patients (12-19 years) and parents (of children 0-19 years) from the Emma Children's Hospital were invited to participate. A mixed-method design was used; (1) Focus groups were held and analyzed using thematic analysis in psychology, (2) a questionnaire was sent out and analyzed using descriptive statistics.

Results

(1) Eight patients and 17 parents participated. Patients mentioned that KLIK has an attractive layout. However, PROMs were sometimes considered irrelevant and repetitive. Parents valued that KLIK provides insight into their child's functioning, but they were not satisfied with the extent to which PROMs were discussed by clinicians. (2) 31 patients and 130 parents completed the questionnaire. Overall, patients and parents reported a satisfaction score of 7.9/10 and 7.3/10, respectively. 81% of patients and 74% of parents indicated that KLIK is easy to use.

Conclusion

Patients and parents are generally satisfied with KLIK, however, points of improvement were mentioned. These are currently being addressed by e.g., upgrading the KLIK website, implementing PROMIS item banks in KLIK to reduce irrelevancy and repetitiveness of PROMs, and implementation strategies to improve the discussion-rate. In this way, implementation of the KLIK PROM portal can be further optimized, with the ultimate goal to improve quality of care.

Introduction

Patient Reported Outcome Measures (PROMs) are increasingly used to monitor and discuss symptoms, Health-Related Quality of Life (HRQOL) and psychosocial functioning of patients in the consultation room with the ultimate goal to enable shared-decision making and patient-centered care [1-3]. Using PROMs in clinical practice has been shown valuable, as it results in more awareness for and increased discussion of patient concerns, higher patient satisfaction, better communication between patient and clinician, and improved treatment outcomes [4-9].

A system that facilitates the use of PROMs in clinical practice is the evidence-based KLIK PROM portal (www.hetklikt.nu) [10-13], which has been implemented in over 20 hospitals in the Netherlands since 2011 [14]. With KLIK, pediatric patients and/or their parents, and adult patients complete PROMs before the outpatient consultation. Answers are converted into an electronic KLIK PROfile (KLIK ePROfile) which the clinician discusses with patients and parents during the consultation [14]. The most important stakeholders in the development and implementation process of the KLIK PROM portal are the users; clinicians as well as patients/parents. From the onset of KLIK, clinicians' opinions were asked during these processes. For example, clinicians' preferences for PROM feedback options in the KLIK ePROfile were studied [10], clinicians were involved in the selection of PROs and PROMs for their disease group, and they were consulted in annual evaluation meetings to identify and overcome barriers in the implementation process [14]. Two studies were performed to gain more insight into the experiences of clinicians with KLIK and to identify barriers in the implementation process, with the goal to improve the KLIK PROM portal according to their needs [15, 16]. However, the opinion of the other stakeholder, patient/parents, is also important [17], as engaging patients in KLIK could result in higher patient satisfaction and higher enrollment rates [18-21].

Worldwide, patients are increasingly engaged in PROM development (e.g., item development, comprehensibility) [22] and PROM visualization to patients and clinicians [23]. However, the experiences of patients regarding the use of PROMs in daily clinical practice has received less consideration [24-31]. Available studies explored the experiences of adult patients regarding the use of PROMs in daily clinical practice. Both positive (e.g., improved communication, insight into patient's functioning, and increased awareness of psychosocial problems) [25, 26, 28-31] and negative experiences (e.g., negative and irrelevant questions in PROMs, unclear purpose of using PROMs) [25-27] were identified. To our knowledge, no studies have been performed focusing on the experiences of pediatric patients and their parents with using PROMs in daily clinical practice. To be able to optimize and further implement the KLIK PROM portal, it is also necessary to gain understanding of their wishes and needs. Therefore, the aim of this study is to provide more insight into the perspective of patients and parents on the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal.

Methods

KLIK workflow

The KLIK workflow for pediatric patients and parents consists of several steps; (1) creation of a KLIK account by patients/parents, (2) completion of PROMs by patients/parents before the outpatient consultation, (3) conversion of answers into a KLIK ePROfile, and (4) discussion of the KLIK ePROfile by the clinician during consultation (Fig 1).

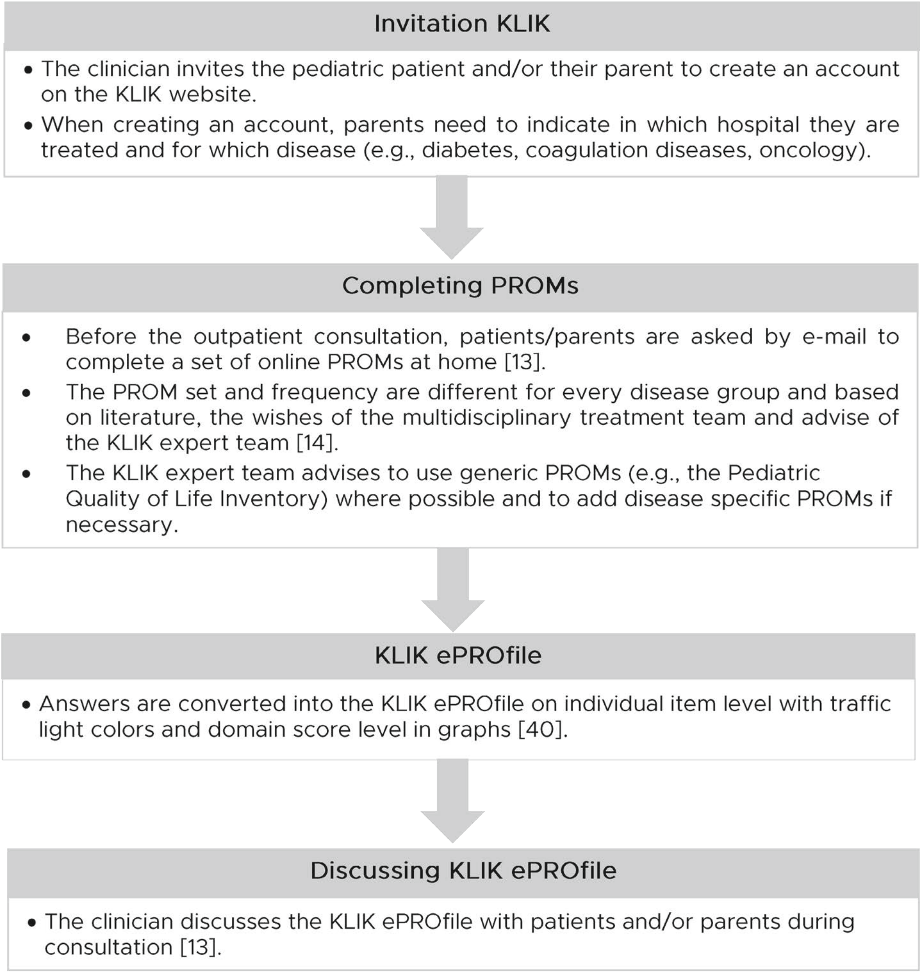
Design

This study is part of a larger participation study where KLIK users' (patients/parents) opinion was asked about several aspects of health care and the use of the KLIK PROM portal. This sub-study reports on the evaluation of the KLIK PROM portal. A mixed-method design was used where qualitative and quantitative methodologies were combined: (1) focus groups were held with patients and parents and (2) an evaluation questionnaire was sent out to pediatric patients and parents. The Medical Ethics Committee of the Amsterdam University Medical Centers (Amsterdam UMC-AMC) approved this study. All participants provided informed consent.

Participants

Patients (12-19 years) and parents (of children 0-19 years) who consult a pediatric department of the Emma Children's Hospital Amsterdam UMC that uses KLIK as standard part of care, completed KLIK PROMs at least once (questionnaire) or twice (focus groups), and were part of the 'KLIK panel' could participate in this mixed-method study. Patients with any chronic health condition could participate in this study as the workflow of the KLIK PROM portal is similar for all patient groups. The 'KLIK panel' consists of patients and parents that indicated, during registration on the KLIK PROM portal, that they give permission to be invited for research projects. Eligible patients/parents were invited by e-mail to take part in the focus groups

(March 2018) and/or to complete the evaluation questionnaire (June-December 2019). Socio-demographics (age and gender child), information on chronic health condition of the child and years of using KLIK were obtained from the KLIK PROM portal. All participants received a gift card of 5 euros (focus groups) or 10 euros (questionnaire) after participation.



Invitation KLIK

- The clinician invites the pediatric patient and/or their parent to create an account on the KLIK website.
- When creating an account, parents need to indicate in which hospital they are treated and for which disease (e.g., diabetes, coagulation diseases, oncology).

Completing PROMs

- Before the outpatient consultation, patients/parents are asked by e-mail to complete a set of online PROMs at home [13].
- The PROM set and frequency are different for every disease group and based on literature, the wishes of the multidisciplinary treatment team and advise of the KLIK expert team [14].
- The KLIK expert team advises to use generic PROMs (e.g., the Pediatric Quality of Life Inventory) where possible and to add disease specific PROMs if necessary.

KLIK ePROfile

- Answers are converted into the KLIK ePROfile on individual item level with traffic light colors and domain score level in graphs [40].

Discussing KLIK ePROfile

- The clinician discusses the KLIK ePROfile with patients and/or parents during consultation [13].

Figure 1. Patient journey of patients and parents using the KLIK PROM portal

Procedure

Focus groups

Focus groups with patients and parents were held separately and for each focus group inclusion of three to six participants was pursued [32]. Focus groups consisted of a group discussion guided by two moderators (MvM, LT, HvO, or LH). At the start of the focus group, the aim of the study was explained and a short recapitulation of KLIK was provided. Then, to obtain patients' and parents' opinion about KLIK, positive and negative experiences with KLIK were discussed using the evaluation technique 'Complain and Cheer wall' [33]. Participants were asked to write down their positive experiences on a flip over at one side of the room, what we called the 'Cheer wall', and points of improvement on another flip over at the other side of the room, the 'Complain wall'. Thereafter a group discussion took place and topics on the walls were grouped together into main themes. Duration of each focus group was 60 minutes. All focus groups were audio recorded.

Questionnaire

The questionnaire (separate version for patients and parents, with minor differences regarding language use–Supplement 1) was developed by five researchers of the KLIK expert team and reviewed by five other researchers and one psychologist. Both versions of the questionnaire consisted of 17 closed questions (response options: three- and five-point Likert Scales and Visual Analogue Scales (VAS)) and two mandatory open questions (advantages and disadvantages of KLIK), regarding (1) overall satisfaction with the KLIK PROM portal, (2) completion of PROMs in the KLIK PROM portal, (3) discussing PROMs with the clinician, (4) influence of KLIK on the (preparation of) the consultation, (5) usability of the KLIK PROM portal, and (6) content of PROMs. For three closed questions, an additional mandatory open question was provided, asking about the reason for their answer.

Analyses

Descriptive analyses were performed using the Statistical Package for Social Sciences (SPSS) version 25.0 to characterize the participants.

Regarding the focus groups, all audio recordings were transcribed verbatim and the transcripts were analyzed independently by MvM and LT in MAXQDA (2018) following the thematic analysis in psychology [34]: (1) highlighting relevant parts of the manuscript, (2) organizing data into meaningful groups by generating initial codes, (3) collating initial codes into themes, (4) refining themes into main- and subthemes, (5) defining the final themes. Analyses were discussed until consensus was reached on the themes. Data saturation was considered attained when no new themes emerged during the analyses of the focus groups.

Regarding the questionnaire, SPSS was used for descriptive statistics (percentages) to provide insight into the experiences of patients and parents with the use of the KLIK PROM portal. Open questions of the evaluation questionnaires were analyzed qualitatively by MvM and LT. This was done by clustering the answers of both patients and parents into main themes following the thematic analysis in psychology [34].

Results

Participants

Figure 2 shows the study and participant flowchart of this study. In total, 8 patients (three focus groups) and 17 parents (three focus groups) participated in six focus groups. Regarding the questionnaire, 31 patients (response rate: 21.8%) and 130 parents (response rate: 19.6%) participated. One patient and 5 parents participated in the focus groups and completed the questionnaire. Table 1 shows the sociodemographic characteristics of all participants.

Focus groups

Data saturation was attained as no new themes emerged after analyzing the focus groups. Table 2 (patients) and 3 (parents) depict the most important positive experiences with KLIK and points of improvement for KLIK and corresponding examples of statements. Themes are ranked based on the number of times mentioned (most often to fewest times) by patients and parents during the focus groups.

Patients

In all focus groups, patients came up with a broad range of experiences with KLIK, both positive, negative and mixed. Themes that were unanimously rated as positive were that the KLIK website has an attractive *layout* (due to the use of colors and pictures), that KLIK *provides insight* into their daily functioning and that KLIK improves the *conversation content* during the consultation, where a broader range of topics is discussed. Furthermore, patients indicated that the *consultation* is more *efficient* when using KLIK and that they are happy about how *secure* the KLIK website is and how their data remains *anonymous*. There were five themes on which patients disagreed. Some patients rated the *content of PROMs* positively, as they cover all important topics and are clear, while other patients indicated that the questions in the PROMs are difficult to understand, repetitive and not relevant for every patient.

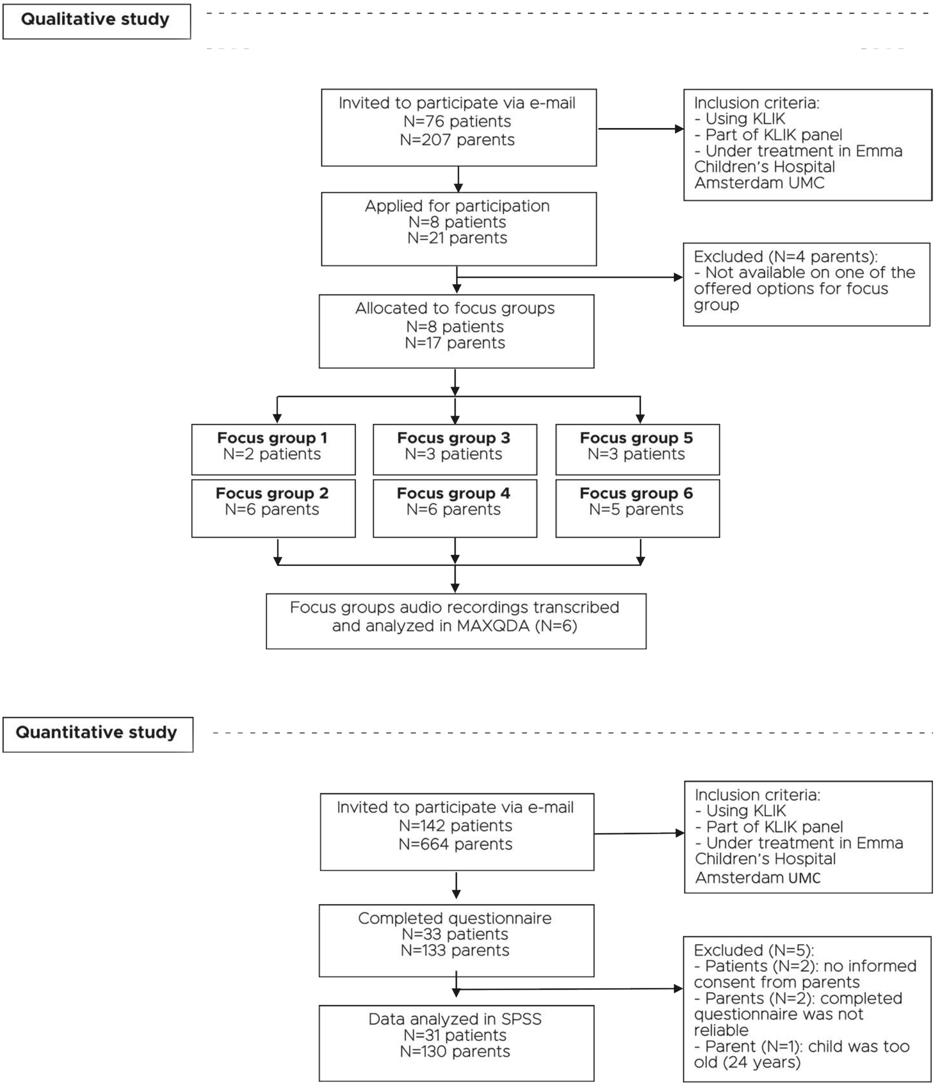


Figure 2. Study and participant flowchart of the qualitative (focus groups) and quantitative study (questionnaire)

Table 1. Sociodemographic characteristics of focus group and questionnaire participants

	Focus groups			**Questionnaire**		
Patients	***N***	***M***	**Range**	***N***	***M***	**Range**
KLIK user since (years)	8	3.2	1.1-6.1	31	5.2	1.0-8.2
Age	8	15.3	13.1-18.8	31	15.7	12.4 -19.2
		%			**%**	
Gender (female)	6	75.0		15	48.4	
Chronic health condition						
Juvenile idiopathic arthritis	2	25.0		7	22.6	
Cystic Fibrosis	2	25.0		1	3.2	
Cancer	2	25.0		0	0	
Gastrointestinal diseases	1	12.5		4	12.9	
Home parenteral nutrition	1	12.5		0	0	
Sickle cell disease	0	0		4	12.9	
Other*	0	0		15	48.4	
Parents	***N***	***M***	**Range**	***N***	***M***	**Range**
KLIK user since (years)	17	2.8	0.8-6.1	130	3.2	0.3 - 8.1
Age (of child in KLIK)	17	10.4	2.1-16.9	130	9.3	0.9 – 19.1
		%			**%**	
Chronic health condition (child)						
Cancer	6	35.3		0	0	
Juvenile idiopathic arthritis	2	11.8		13	10.0	
Hemophilia	2	11.7		4	3.1	
Home parenteral nutrition	2	11.7		3	2.3	
Gastrointestinal diseases	1	5.9		20	15.4	
Neonatology follow up	0	0		28	21.5	
Other*	4	23.5		62	47.7	

*Only most common conditions groups (>10% in one of the study groups) are reported, other: cleft lip, endocrinology, nephrology, HIV, dermatology, craniofacial abnormalities, spherocytosis, cystic fibrosis, lysosomal storage disorders, intensive care follow-up, Marfan syndrome, feeding disorders, phenylketonuria, and muscular disorders.

In addition, *completion time* was rated by some as good and by others as time-consuming, and the KLIK ePROfile is always *discussed* by the *clinician* according to some patients, but not enough by others. Finally, KLIK helps only some patients in *preparing for the consultation*, and patients were ambiguous about *ease of use* of KLIK. The lack of motivation for completing the KLIK PROMs was only mentioned as a negative experience by some patients.

Table 2. Positive experiences and points of improvement mentioned by patients (N=8) in the focus groups (ranked from most often to fewest times mentioned)

Themes	Positive experiences	Points of improvement
Content of PROMs	'The questions are clear, recognizable and easy to answer'	'There is a lot of repetition in questions'
	'All topics are covered in the questionnaires, not only topics about your disease'	'The questions are not relevant for every patient and sometimes questions are difficult to understand'
		'It would be good if questions were administered based on previous answers'
Completion time PROMs	'Completing the questionnaires does not take too much time'	'Completing the questionnaires takes a lot of time'
Layout	'The KLIK website looks nice with the colors that are used'	
	'Nice that you can see a picture of your doctor'	
Discussion by clinician	'The answers in the KLIK ePROfile are discussed by the clinician'	'The clinician often does not discuss the KLIK ePROfile'
		'Sometimes the clinician does not ask more questions based on my answers'
Insight patients' functioning	'By completing the questionnaires you see how you are doing'	
	'It is good that parents know what is going on'	
	'With KLIK, clinicians know how you are doing'	
Conversation content	'With KLIK, not only physical health, but also mental health is discussed' 'It helps in discussing topics that you would otherwise not think about'	
Preparation of consultation	'Completing the questions before the appointment helps you to come up with topics you want to discuss during the consultation'	'Completing KLIK questionnaires does not help you in preparing for the consultation, it is just something you need to do'
Motivation child		'I think it is not always necessary to complete the KLIK questionnaires'
		'I sometimes just do not want to talk about the KLIK topics'
Consultation efficiency	'The consultation is more efficient when KLIK is used, as the doctor immediately has an overview of how you are doing'	
Anonimity and security	'It is good that KLIK is well secured'	
	'As KLIK PROMs are completed on the computer, it feels more anonymous, which results in completing the PROMs more honestly'	
Ease of use	'It is nice that the KLIK questionnaires can be completed on the computer at home'	'You cannot go back to the questionnaire if you completed all questions'

All quotes were translated into English.

Parents

Parents mentioned many similar experiences with KLIK as patients (Table 3). Themes that were unanimously rated as positive were that KLIK helps in *preparing for the consultation* and *provides insight into the patients' functioning*, although for some parents this insight was also confronting when many problems were reported. In addition, parents were satisfied that by using KLIK *problems are detected* at an early stage and that support can be provided timely. All other themes were evaluated both positively and negatively. Some parents indicated that they are satisfied with the *content of PROMs*, as all topics are covered and questions are easy to understand, while other parents disagreed and indicated that questions are hard to understand for their child, are confronting and repetitive. Parents also had mixed opinions regarding *ease of use* of KLIK, where some thought completing PROMs online is working great, and others thought this could be improved by developing a KLIK app and linking KLIK to the Electronic Health Records (EHR). Furthermore, *discussion* of the KLIK ePROfile *by clinicians* always happens according to some parents, but not often enough by even more parents. Most parents mentioned that the *conversation content* improves as more and different topics are discussed, while some did not recognize this. *Completion time* is manageable for some, but too long for others and the *layout* of the KLIK website is attractive and child-friendly according to most parents, but could be made more attractive by using visuals according to some parents. Finally, some parents indicated that they do not see the added *value and goal* of KLIK, while others disagreed and indicated that KLIK is of great value to the consultation.

Questionnaire

Overall satisfaction with the KLIK PROM portal

Patients and parents reported an overall satisfaction with the KLIK PROM portal of mean = 7.9 and mean = 7.3, respectively, on a VAS ranging from 0 (not satisfied) to 10 (very satisfied).

Table 3. Positive experiences and points of improvement mentioned by parents (N=17) in the focus groups (ranked from most often to fewest times mentioned)

Themes	Positive experiences	Points of improvement
Content of PROMs	'The questions are easy to understand for children'	'The questions are sometimes not relevant and confronting for children'
	'All important topics are covered in the questionnaires'	'It is annoying that every time the same questions are asked'
		'There is no attention for brothers, sisters and the family situation'
		'The questions are difficult to understand for young children. I would suggest to make the questions more visual'
Ease of use	'KLIK is easy to use and it is nice that you can complete questionnaires online'	'KLIK should be connected with the EHRs, so appointments are automatically linked'
	'I like the reminder e-mails that are sent by KLIK'	'I would like KLIK to be available as an app'
Insight patients' functioning	'It is nice that parents have insight into the functioning of their child over time'	
	'With KLIK the clinician knows what is going on and can follow the child over time'	
Discussion by clinician	'The clinician takes KLIK seriously and always discusses the answers'	'The KLIK questionnaires are often not discussed by the clinician'
		'Especially questionnaires about the functioning of parents are not discussed'
Conversation content	'KLIK is a conversation tool and provides structure and more depth to the conversation'	'Our consultation has already a fixed structure, so KLIK does not help with that'
	'It is nice that with KLIK psychosocial functioning is also taken into account'	
Preparation of consultation	'KLIK helps to start a conversation with your child or partner about the situation before the consultation'	
	'KLIK helps to think about how it is going and to prepare questions before the consultation'	
Layout	'The KLIK website is attractive and looks nice for children'	'It would be good if smileys were used to make KLIK more attractive'
	'The layout of KLIK is clear and understandable'	
Completion time PROMs	'The completion time is manageable and not too long'	'Too many questions have to be completed'
		'Before I start completing the questionnaires I would like to see how much time it will take'
Detecting problems	'With KLIK problems are detected early and your child can be referred for help'	

Table 3. *(continued)*

Themes	Positive experiences	Points of improvement
Value and goal	'I like that with KLIK there is the possibility to report difficulties'	'Completing KLIK questionnaires feels not useful when it is going well'
		'It is not totally clear what is done with your answers and if they can be used against you by the government'

All quotes were translated into English.

Completion of PROMs in the KLIK PROM portal

As shown in Table 4, 78% of the patients and 84% of the parents agreed that they know why there are asked to complete PROMs via the KLIK PROM portal. Patients and parents reported that the frequency in which they are asked to complete these PROMs varies from once every three years to more than four times a year. Most patients and parents were satisfied with this frequency. When patients and parents are asked to complete PROMs, the majority indicated that they almost always do this. Reasons for not completing the PROMs were: lack of time, forgot to complete, little change in functioning since the last PROM completion, and no motivation. Patients and parents spent on average 13.8 and 15.2 min on completing the PROMs, respectively. More than 80% of both patients and parents were satisfied with this completion time.

Discussing PROMs with the clinician

About half of the patients and parents indicated that their clinician (almost) always discusses the KLIK ePROfile with them during the consultation (Fig 3). If the clinician does not discuss the completed PROMs, 52% of the patients and 72% of the parents indicated they dare to start the discussion about PROMs themselves.

Influence of KLIK on the (preparation of the) consultation

KLIK is of added value for the conversation with their clinician, according to 58% of the patients and 59% of the parents (Fig 3). Less than half of the patients and parents indicated that more topics are discussed by using the KLIK PROM portal in comparison with not using the KLIK PROM portal and that the use of KLIK provides more structure to the conversation. Clinicians' failure to discuss the KLIK ePROfile was a frequently mentioned reason why KLIK has no value during the consultation. More than half of the parents reported that the use of KLIK provides them more insight into the functioning of their child and helps in preparing for the consultation (62% and 54% respectively), in contrast to only 39% and 42% of the patients. Patients indicated that they know very well how they are doing, even without completing a PROM.

Table 4. Scores on the domain 'completion of PROMs in the KLIK PROM portal' (patients: $N = 31$, parents: $N = 130$)

			Agree–*N* (%)	**Neutral–*N* (%)**	**Disagree - *N* (%)**
I know why I am being asked to complete KLIK PROMs	Patients		24 (78)	1 (3)	6 (19)
	Parents		109 (84)	13 (10)	8 (6)
		4 times a year–*N* (%)	**2 times a year–*N* (%)**	**Yearly–*N* (%)**	**Other–*N* (%)**
How often are you asked to complete the PROMs in KLIK?	Patients	7 (22)	12 (39)	8 (26)	4 (13)
	Parents	21 (16)	29 (22)	38 (30)	42 (32)
			Yes–*N* (%)	**No, too often–*N* (%)**	**No, too infrequent–*N* (%)**
Are you satisfied with this frequency?	Patients		29 (94)	1 (3)	1 (3)
	Parents		111 (85)	11 (9)	8 (6)
			(Almost) always–*N* (%)	**Sometimes–*N* (%)**	**(Almost) never–*N* (%)**
When you are asked to complete the PROMs in KLIK, how often do you do this?	Patients		28 (90)	3 (10)	-
	Parents		123 (95)	2 (1)	5 (4)
			***M* (range)**		
I spend on average .. minutes on completing the KLIK PROMs	Patients		13.8 (5-30)		
	Parents		15.2 (0-60)		
			Yes–*N* (%)	**No, too long–*N* (%)**	**No, too short–*N* (%)**
Are you satisfied with the completion time?	Patients		25 (81)	6 (19)	-
	Parents		109 (84)	20 (15)	1 (1)

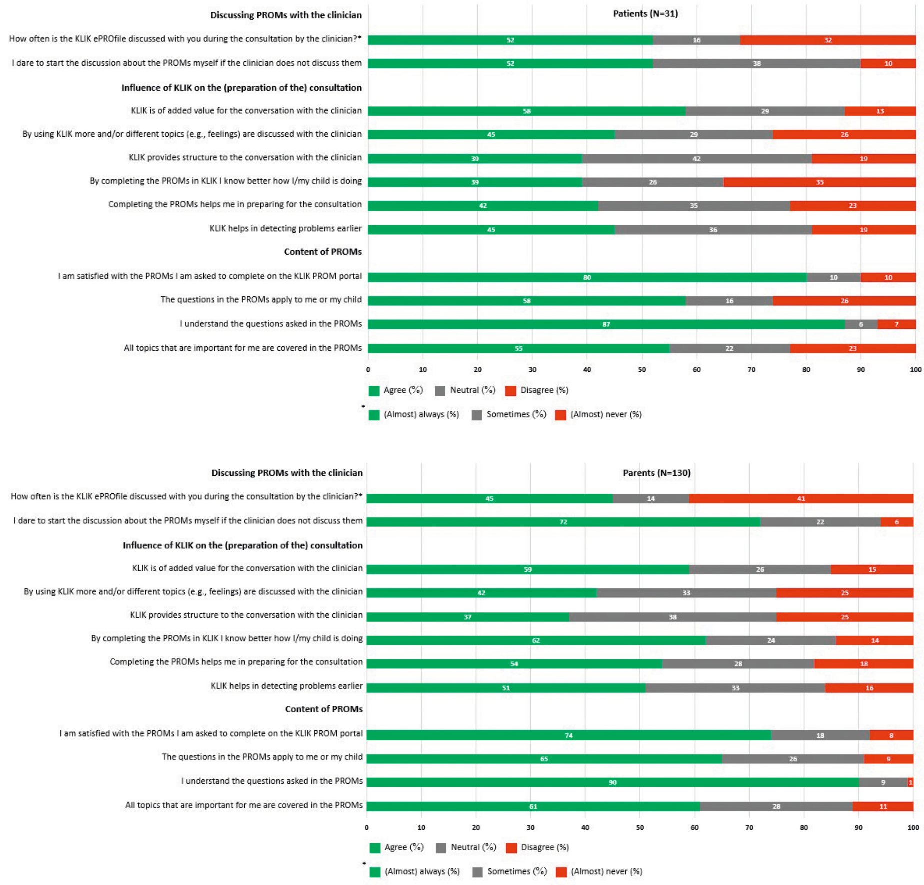


Figure 3. Scores on the domains 'discussing PROMs with the clinician', 'Influence of KLIK on the (preparation of the) consultation', and 'content of PROMs' (patients: N=31, parents: N=130)

Table 5 shows the most important advantages and disadvantages of KLIK, as reported in the open questions. The themes are ranked based on the number of times mentioned by patients and parents in the open-ended questions. Main advantages of KLIK for patients and parents were: *easy to use, clinician is better prepared, patients and parents are better prepared*, and *insight into functioning (of my child)*. Main disadvantages of KLIK for patients and parents were: *not easy to use, irrelevant content of PROMs*, and *takes time*. Eleven patients (35%) and 48 parents (37%) did not experience any disadvantages with using the KLIK PROM portal.

Table 5. Advantages and disadvantages of the KLIK PROM portal, mentioned by patients (*N* = 31) and parents (*N* = 130) in the open questions of the evaluation questionnaire

Advantages KLIK PROM portal	**Examples**
Easy to use	'Simple and clear' 'It is easy that you can complete questionnaires online at home'
Clinician is better prepared	'The clinician can see my questions before the appointment at the outpatient clinic' 'The clinician is already aware of my child's health situation and can immediately respond to it'
Patient and parents are better prepared	'It is valuable that you can ask the clinician questions in advance so that you do not forget them' 'Subjects are discussed which you normally do not bring up yourself'
Insight into functioning (of my child)	'KLIK provides insight into how I am doing' 'Provides the opportunity to compare the health situation of my child now with the situation just after diagnosis'
Disadvantages KLIK PROM portal	**Examples**
Not easy to use	'I keep forgetting my password' 'Annoying that I get multiple reminders'
Irrelevant content of PROMs	'Not all questions apply to our situation''It is boring to complete the same questionnaires every time'
Takes time	'Completing the questionnaires takes sometimes more time than I hope''It is a lot of work to complete the questionnaires'

All quotes were translated into English.

Usability of the KLIK PROM portal

The KLIK PROM portal is easy to use, according to 81% of the patients (13% neutral and 6% disagree) and 74% of the parents (18% neutral, 8% disagree). In addition, 48% of the patients (39% neutral, 12% disagree) and 55% of the parents (36% neutral, 9% disagree) indicated that KLIK has an attractive layout.

Content of PROMs

Most patients and parents are satisfied with the PROMs they are asked to complete (Fig 3). Almost all participants indicated that they understand the questions asked in the PROMs. Reasons why patients and parents are not satisfied with the offered PROMs were that the questions in the PROMs do not apply to them or their child, PROMs are too generic, the different questions are very similar, and the PROMs are too long. Some of the patients and parents felt that the offered PROMs do not cover all topics that are important for them. For example they miss topics like growth, parenting support, and side jobs.

Discussion

This study provided insight into the experiences of patients and parents with the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal. Overall, patients and parents were satisfied with the use of KLIK. They indicated that KLIK provides insight into the patient's functioning, helps parents and clinicians in preparing for the consultation, is easy to use, and results in discussion of a broad range of topics (e.g., from disease-specific to psychosocial functioning) during the consultation. However, points of improvement were indicated regarding the content of PROMs, the layout of the KLIK PROM portal, and the discussion of PROMs by the clinician. The results described in this study are in line with previous studies [15, 25, 26].

Although patients and parents responded to the closed question of the evaluation questionnaire that they are generally satisfied with the offered PROMs in KLIK, they mentioned in the focus groups and open-ended questions that the content of PROMs is the most important point of improvement. For example, they indicated that there is repetition in questions, that irrelevant questions are administered, and that the completion time is long, resulting in a burden of completing PROMs. These challenges with PROMs have been mentioned in previous research [16, 35, 36]. To address these challenges, the self-report and proxy-versions of the Patient-Reported Outcomes Measurement Information System (PROMIS®) item banks [37-39] were implemented in the KLIK PROM portal in the past year and are currently used in several clinics [16, 40, 41]. The PROMIS item banks each measure a separate construct that can be administered using Computerized Adaptive Testing (CAT). With CAT, questions are presented to patients based on their previous responses. Hence, patients only have to answer a small number of questions per item bank to obtain a reliable score [42] and have to answer less irrelevant questions. Consequently, the burden of completing PROMs can be reduced.

Another difference between the focus groups and the questionnaire was the rating of the ease of use of the KLIK PROM portal. While in the questionnaire the majority of participants indicated that KLIK is easy to use, in the focus groups especially parents had quite some remarks on how the ease of use could be improved. Parents mentioned that an app would be a valuable addition to the KLIK website in order to complete PROMs on your mobile phone. Additionally, they would like an integration of KLIK with the EHR so that appointments are automatically linked to KLIK by which PROMs are directly available. To address these suggestions, we made the KLIK

PROM portal adaptable for mobile phone use, and realized a front-end (hybrid) integration with the EHR in 2019. With this integration, clinicians can now view the KLIK ePROfile in the EHR and discuss the PROMs more easily. However, to be able to automatically link the appointments to KLIK, a full integration is necessary, which can hopefully be realized in the future.

A final difference between the focus group and questionnaire outcomes was the satisfaction with the layout of the KLIK PROM portal, which was mainly mentioned as a point of improvement in the questionnaire. Patients and parents indicated that the website looks a bit old-fashioned and could be made more attractive by using visuals. For this reason, the homepage of the KLIK website was upgraded recently. The design of the website was changed (e.g., by using visuals and creating a more professional look). In addition, specific information pages are now available for all KLIK users (pediatric patients, parents, adult patients, and clinicians).

Patients and parents mentioned in both the focus groups as the questionnaires that clinicians often do not discuss PROMs during the consultation. This is worrisome, as patients and parents indicated that this is an important reason why KLIK sometimes has no added value for the consultation which consequently may lead to loss of motivation to complete KLIK PROMs. To improve this discussion rate, several implementation strategies were used. For example, the KLIK expert team revised the KLIK training in which more attention is now paid to the importance of discussing PROMs [43] and this topic is discussed more thoroughly during annual evaluation meetings with clinicians [16], with the goal to increase their knowledge, awareness and confidence in discussing PROMs. Additionally, finding champions for each multidisciplinary team to motivate clinicians to use and discuss KLIK PROMs would be beneficial as this was identified as the most important implementation strategy in two KLIK studies [15, 17]. When clinicians do not discuss the completed PROMs, patients and some parents indicated that they do not dare to bring up for them important themes themselves. To empower patients/parents and increase their self-efficacy, educational videos were developed and made available on the KLIK homepage (article in preparation). In these videos tips and tricks are provided how patients and parents can prepare themselves for the consultation and bring up topics they want to discuss with the clinician.

When comparing this study with the KLIK evaluation study with clinicians [16], similar experiences regarding the KLIK PROM portal were mentioned. For example, insight into patients' functioning, improved communication, and better preparation of the consultation were positive points they agreed on, and content of PROMs was the most important point of improvement mentioned by both user groups. However, patients/parents and clinicians mentioned a different PROM completion rate. Patients and parents indicated a very high completion rate, whereas clinicians estimated that this completion rate is much lower and that it takes a lot of effort to motivate patients to complete PROMs [16]. A possible reason for this difference might be a bias in the current sample, as only patients and parents that were part of the KLIK panel were invited for participation. These patients/parents might be more assertive in comparison to the other KLIK users, which might have resulted in an overestimation of the PROM completion rate. Therefore, continuous support and explanation about the goal of the use of KLIK remains very important to both user groups.

There are some limitations to this study that should be mentioned. First, there was a low response rate in the evaluation questionnaire (around 20%) which was unexpected as this questionnaire was sent to participants of the KLIK panel (who indicated that they were willing to be invited for research projects). Possible reasons for the low response rate might be that (1) the willingness of patients and parents has changed as participation in the KLIK panel was only asked during registration, (2) patients and parents do not actively use the KLIK PROM portal anymore, or (3) patients and parents might be tired of completing surveys. Second, it was also difficult to motivate patients to participate in the focus groups. This resulted in a small number of participants per patient focus group (2 to 3 participants) with two moderators, which may have influenced the dynamics. Additionally, we noticed that pediatric patients found it very difficult to formulate and express their opinion and needed a lot of guidance which could have led to a bias in the results. Third, we used a self-developed questionnaire which makes comparisons with other evaluation studies difficult. However, other studies also made use of self-conducted questionnaires [44] or adapted questionnaires from prior studies [29-31], as the questions needed to be specific about features of the tool used.

In conclusion, pediatric patients and parents were satisfied with the usability and effect of the KLIK PROM portal in clinical care. KLIK provides them insight into their functioning and helps them to communicate with the clinician. However, some points of improvement were also identified, which are currently being addressed. We now have insight into the experiences of the most important stakeholders (patients/parents and clinicians) of KLIK. In the future it is important to continuously evaluate the use of the KLIK PROM portal with all stakeholders (including adult patients) to match their needs. In this way, we can further optimize and implement the KLIK PROM portal in clinical care with the ultimate goal to improve the quality of care.

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Conflict of interest

The authors declare that they have no conflict of interest.

Data availability

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

Code availability

Not applicable

Ethics approval

All procedures performed in this study were in accordance with the ethical standards of the international and/or national research committee (Medical Ethics Committee of the Amsterdam UMC – W18_023 # 18.034 and Medical Ethics Committee of the Amsterdam UMC – W19_272 # 19.324) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Consent to participate

Informed consent was obtained from all individual participants included in the study.

Keywords

Patient Reported Outcomes, Questionnaires, Patient engagement, Pediatrics

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Supplement 1 - Evaluation questionnaire KLIK: patient version

Overall satisfaction

1. In general, how satisfied are you with the use of KLIK?
Not satisfied at all 0 1 2 3 4 5 6 7 8 9 10 Very satisfied

Completion of PROMs in the KLIK PROM portal

2. I know why I am being asked to complete KLIK questionnaires
Explanation:
☐ Strongly disagree ☐ Disagree ☐ Neither disagree nor agree ☐ Agree ☐ Strongly agree

3. How often are you asked to complete the questionnaires in KLIK?
Explanation:
☐ 4 times a year ☐ 2 times a year ☐ Once a year ☐ Other

3a. Are you satisfied with this frequency?
☐ Yes, I am satisfied ☐ No, it is too often ☐ No, it is too infrequent

4. When you are asked to complete the questionnaires in KLIK, how often do you do this?
☐ Always ☐ Usually ☐ Sometimes ☐ Almost never ☐ Never

When: usually, sometimes, almost never and never, then:

4a. What is the reason that you do not always complete the questionnaires in KLIK?
Open answer

5. I spend on average … minutes on completing the KLIK questionnaires

5a. Are you satisfied with the completion time?
Explanation:
☐ Yes, I am satisfied ☐ No, it is too long ☐ No, it is too short

Discussing PROMs with the clinician

6. How often is the KLIK ePROfile (completed questionnaires on the KLIK website) discussed with you during the consultation by the clinician?
Explanation:
☐ Always ☐ Usually ☐ Sometimes ☐ Almost never ☐ Never

When usually, sometimes, almost never and never, then:

6a. I dare to start the discussion about the questionnaires myself if the clinician does not discuss them
Explanation:
☐ Strongly disagree ☐ Disagree ☐ Neither disagree nor agree ☐ Agree ☐ Strongly agree

Influence of KLIK on the (preparation of the) consultation

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
7. KLIK is of added value for the conversation with the clinician Explanation:	☐	☐	☐	☐	☐
8. By using KLIK more and/or different topics (e.g., feelings) are discussed with the clinician Explanation:	☐	☐	☐	☐	☐
9. KLIK provides structure to the conversation with the clinician Explanation:	☐	☐	☐	☐	☐
10. By completing the questionnaires in KLIK I know better how I am doing Explanation:	☐	☐	☐	☐	☐
11. Completing the questionnaires in KLIK helps me in preparing for the consultation Explanation:	☐	☐	☐	☐	☐
12. KLIK helps in detecting problems earlier Explanation:	☐	☐	☐	☐	☐

13. Advantages of KLIK are:
Open answer

14. Disadvantages of KLIK are:
Open answer

Usability of the KLIK PROM portal

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
15. The KLIK PROM portal is easy to use Explanation:	☐	☐	☐	☐	☐
16. KLIK has an attractive lay-out Explanation:	☐	☐	☐	☐	☐

Content of PROMs

	Strongly disagree	Disagree	Neither disagree nor agree	Agree	Strongly agree
17. I am satisfied with the questionnaires I am asked to complete on the KLIK PROM portal Explanation:	☐	☐	☐	☐	☐
When: strongly disagree, disagree, neither disagree, nor agree:					
17a. Why are you not satisfied? *Open answer*					
18. The questions in the questionnaires apply to me Explanation:	☐	☐	☐	☐	☐
19. I understand the questions asked in the questionnaires Explanation:	☐	☐	☐	☐	☐
20. All topics that are important for me are covered in the questionnaires	☐	☐	☐	☐	☐
When: strongly disagree, disagree, neither disagree, nor agree:					
20a. Which topics do you miss? *Open answer*					

6

A retrospective assessment of the KLIK PROM portal implementation using the Consolidated Framework for Implementation Research (CFIR)

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Abstract

Purpose

The KLIK Patient Reported Outcome Measure (PROM) portal is an evidence-based intervention implemented in clinical practice in >25 Dutch hospitals for patients (children and adults) who regularly visit the outpatient clinic. Implementation science frameworks can be used to understand why implementation succeeded or failed, to structure barriers and enablers, and to develop implementation strategies to overcome barriers. This paper aimed to A) retrospectively describe determinants of successful KLIK PROM implementation using the Consolidated Framework for Implementation Research (CIFR), and B) identify current barriers and match implementation strategies.

Methods

A) The KLIK implementation process was described retrospectively based on literature and experience, using the 39 CFIR constructs organized in five general domains: intervention characteristics, outer setting, inner setting, characteristics of individuals and implementation process. B) The CFIR-Expert Recommendations for Implementing Change (ERIC) Implementation Strategy Matching tool identified current barriers in the KLIK implementation and matched implementation strategies that addressed the identified barriers.

Results

A) The most prominent determinants of successful KLIK PROM implementation lie in the following CFIR domains: intervention characteristics (e.g. easy to use), characteristics of individuals (e.g. motivation) and process of implementation (e.g. support). B) 13 CFIR constructs were identified as current barriers for implementing the KLIK PROM portal. The highest overall advised ERIC strategy for the specific KLIK barriers was to identify and prepare champions.

Conclusion

Using an implementation science framework, e.g. CFIR, is recommended for groups starting to use PROMs in clinical care as it offers a structured approach and provides insight into possible enablers and barriers.

Introduction

Patient-Reported Outcome Measures (PROMs) are standardized, validated questionnaires that are completed by patients, such as a person's perspective on their health, well-being or symptoms [1,2]. PROMs can be used for several purposes: at group level to study differences between disease populations, to describe the effects of treatment in clinical trials, and to assess quality of care or on an individual level to promote patient-centered care, guide clinical decision-making and to facilitate communication [3]. There is widespread evidence for the effects of PROM applications on an individual level regarding an increase in Health-related Quality of Life (HRQOL) scores, satisfaction with care and communication about PROs in research settings, both in adult [4-6] and pediatric [7-12] samples. Yet the implementation of these evidence-based (EB) PROMs interventions is challenging.

The KLIK PROM portal (www.hetklikt.nu and www.klik-uk.org) is an example of an EB PROM intervention for patients (children or adults) who regularly visit the outpatient clinic [13]. Patients complete PROMs online, prior to their visit. Answers are transformed into an electronic PROfile (ePROfile; Fig 1). Clinicians discuss this ePROfile with patients, to monitor well-being over time, identify problems, and provide tailored advice and interventions. The effects of using the KLIK PROM portal have been demonstrated in pediatric oncology [7] and in pediatric rheumatology [12], by showing an increased and more detailed discussion of HRQOL and psychosocial functioning during the consultation, less undetected problems, and a higher clinician-reported satisfaction with provided care, without lengthening the consultation duration.

Despite the availability of several EB PROM interventions across the world, the actual implementation of PROM interventions in clinical practice remains limited [14-17]. There is a critical gap in behavioral medicine between what we know can optimize patient health and care outcomes and what gets implemented in everyday practice [1]. If EB PROM interventions are not successfully implemented in clinical practice, then intended effects are not reached, which limits the impact on patients' health outcomes [18,19]. Traditional randomized controlled trials study the effectiveness of PROM interventions under ideal circumstances. Yet for the implementation of PROMs in clinical practice, a different, more flexible approach is needed. Often, a "voltage drop" (a dramatic decrease in effectiveness) is seen once interventions get implemented in clinical practice [20]. Implementation research is defined by the National Institute of Health as the "scientific study of the use of strategies to adopt and integrate evidence-based health interventions into clinical and community settings in order to improve patient outcomes and benefit population health" [21]. Therefore, a scientific approach to the change process is crucial. In order to know what drives successful implementation of PROMs in clinical practice, we need to study the mechanisms that influence implementation outcomes [17,22,23]. Implementation science models, theories, or frameworks support in identifying factors that influence an implementation process or outcome.

In general, three overarching aims of theoretical approaches and five *categories* of theories, models and frameworks used in implementation science can be distinguished [24]: (1) guiding the process of translating research into practice (*process models*), (2) understanding and/or explaining what influences implementation outcomes (*determinant frameworks, classic theories*, and *implementation theories*), and (3) evaluating implementation (*evaluation frameworks*). Specifically, determinant frameworks are useful in understanding or explaining what influences implementation outcomes and to support the design of implementation strategies or maximizing the use of enablers to implementation [24].

A widely cited and comprehensive determinant framework in the implementation science literature in health is the Consolidated Framework for Implementation Research (CFIR). Damschroder et al. [22] aimed to develop a framework that comprises common constructs from published implementation theories and includes, therefore, missing key constructs in other theories. It contains 39 constructs which are organized in five general domains: (1) intervention characteristics (e.g., evidence, complexity, adaptability, costs), (2) outer setting (e.g., peer pressure and external policies), (3) inner setting (e.g., structural characteristics, implementation climate, and culture), (4) characteristics of individuals (e.g., knowledge about the intervention and self-efficacy), and (5) implementation process (e.g., planning, engaging stakeholders, champions, and execution), see Fig 3. Determinant frameworks, such as CFIR, are specifically useful in understanding or explaining what influences implementation outcomes and to support the design of implementation strategies or maximizing the use of enablers to implementation [24]. This paper aimed to (A) retrospectively describe the most prominent determinants and reasons of successful KLIK PROM implementation using CFIR and (B) use the CFIR-ERIC Implementation Strategy Matching tool to identify *current* barriers of the KLIK PROM portal implementation and match implementation strategies that address the identified barriers. In our specific study context, the CFIR framework seemed particularly useful as it covers a wide range of implementation constructs and domains and it allowed

1a

Child

○ 03-11-2016 ☑ 03-06-2017 ○ 23-12-2017 ☑ 26-04-2018

Physical	03-06-2017	26-04-2018
It is hard for me to walk more than one block	Sometimes ●	Never ●
It is hard for me to run	Often ●	Almost always ●
It is hard for me to do sports activity or exercise	Often ●	Often ●
It is hard for me to lift something heavy	Sometimes ●	Almost always ●
It is hard for me to take a bath or shower by myself	Never ●	Never ●
It is hard for me to do chores around the house	Almost always ●	Often ●
I hurt or ache	Almost never ●	Sometimes ●
I have low energy	Often ●	Sometimes ●

Emotional	03-06-2017	26-04-2018
I feel afraid or scared	Never ●	Never ●
I feel sad or blue	Never ●	Almost never ●
I feel angry	Almost never ●	Sometimes ●
I have trouble sleeping	Never ●	Sometimes ●
I worry about what will happen to me	Never ●	Never ●

Social	03-06-2017	26-04-2018
I have trouble getting along with other kids	Never ●	Never ●
Other kids do not want to be my friend	Never ●	Never ●
Other kids tease me	Never ●	Almost never ●
I cannot do things that other kids my age can do	Sometimes ●	Often ●
It is hard to keep up when I play with other kids	Never ●	Never ●

School	03-06-2017	26-04-2018
It is hard to pay attention in class	Never ●	Never ●
I forget things	Almost never ●	Sometimes ●
I have trouble keeping up with my schoolwork	Sometimes ●	Never ●
I miss school because of not feeling well	Sometimes ●	Sometimes ●
I miss school to go to the doctor or hospital	Sometimes ●	Sometimes ●

1b

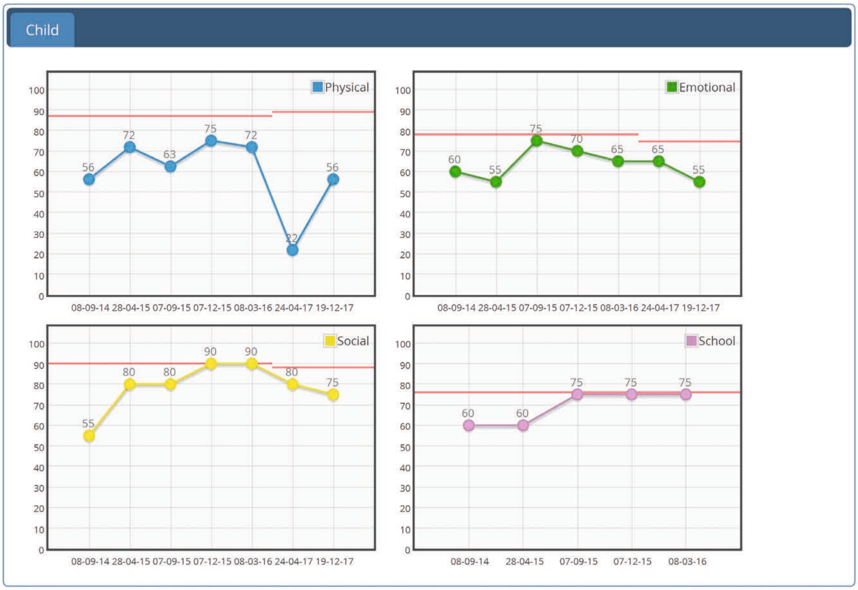


Figure 1. A. KLIK ePROfile - literal feedback of the individual items on the Pediatric Quality of Life Inventory (PedsQL) B. KLIK ePROfile - graphical feedback of the PedsQL, including norm lines

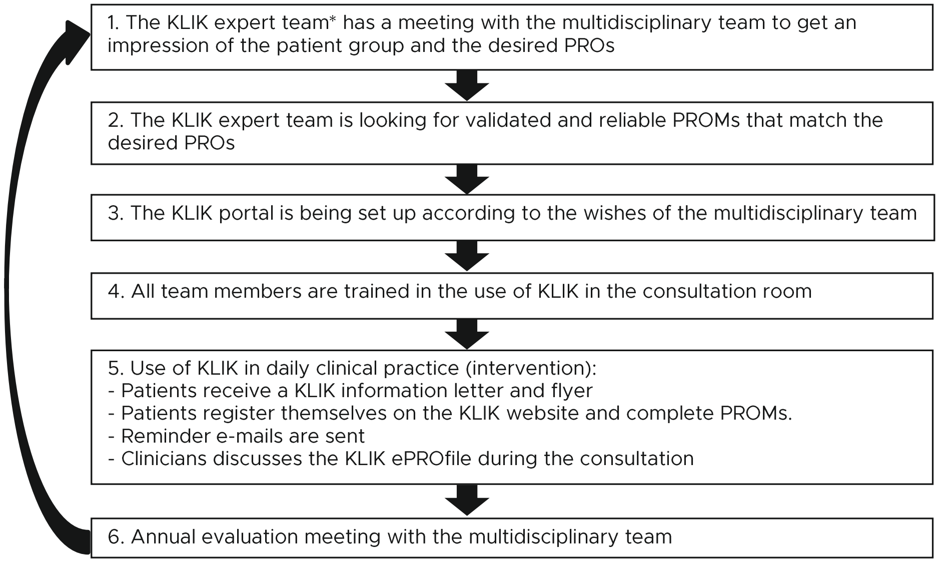
us to use a standardized framework to explain the influence of each domain on the implementation outcomes of an evidence-based PROM portal. With years of experience in the development and implementation, the KLIK PROM portal is now in a phase of understanding what barriers and facilitators have already been resolved and determining what major determinants are currently of influence to move to the next area of implementation: sustainability.

Methods

The evidence-based KLIK PROM portal

The development and implementation of the KLIK PROM portal is based on multiple studies (Supplemental Table 1). The predecessor of the KLIK ePROfile was the QLIC-ON PROfile [25]. During the QLIC-ON study, two generic HRQOL questionnaires widely used in pediatrics (TAPQOL [26] and PedsQL [27]) were converted into digital questionnaires. Patients were asked to complete a HRQOL questionnaire on a laptop in the waiting room of the outpatient clinic, prior to the visit. The literal answers and graphs were printed out, fed back to the pediatrician in a QLIC-ON PROfile on paper, and discussed with patients and parents during the consultation [25]. However, completing PROMs at the outpatient clinic and providing hard copy PROfiles was logistically complicated, and therefore, they are hard to implement in a real-world situation. As a result, the KLIK website (www.hetklikt.nu) was developed during the KLIK study in pediatric rheumatology [28]. From that moment, children and parents completed the questionnaires online at home. The implementation of KLIK, as part of standard care, started in 2011 [7,12]. To gain more insight into the implementation process and outcomes, a study was conducted to identify barriers and enablers in this process in pediatric oncology [29].

Currently, KLIK is part of standard care in >70 different patient groups (e.g., diabetes, nephrology) in >20 centers in the Netherlands and 3 centers in the United Kingdom. Over 17,000 patients are registered on the KLIK website and around 1,000 clinicians (e.g., physicians, nurses, psychologists) have been trained in the use of KLIK. KLIK is implemented in various settings, including hospital outpatient clinics, rehabilitation centers, and recently in dentistry. KLIK was initially developed for use in pediatrics, but since 2017 KLIK has also been implemented in adult care (e.g., coagulation diseases and medical psychology). The KLIK expert team of the Emma Children's Hospital Amsterdam UMC coordinates the implementation of the KLIK PROM portal in pediatrics and adult healthcare in 20 hospitals in the Netherlands. The KLIK expert team in the Princess Máxima Center for pediatric oncology coordinates the implementation of KLIK in this center. KLIK can be implemented for any patient group, on request of a multidisciplinary team. The implementation procedure of the KLIK PROM portal has previously been described according to the guidelines of the International Society for Quality of Life Research (ISOQOL) [13]. A core element of the KLIK implementation process is to train all team members in the use of KLIK and discussing PROMs in the consultation room. A summary of the implementation process is shown in Fig 2.



1. The KLIK expert team* has a meeting with the multidisciplinary team to get an impression of the patient group and the desired PROs

2. The KLIK expert team is looking for validated and reliable PROMs that match the desired PROs

3. The KLIK portal is being set up according to the wishes of the multidisciplinary team

4. All team members are trained in the use of KLIK in the consultation room

5. Use of KLIK in daily clinical practice (intervention):
- Patients receive a KLIK information letter and flyer
- Patients register themselves on the KLIK website and complete PROMs.
- Reminder e-mails are sent
- Clinicians discusses the KLIK ePROfile during the consultation

6. Annual evaluation meeting with the multidisciplinary team

* The KLIK expert team consists of researchers with expertise in the field of (implementing) PROMs and HRQOL research

Figure 2. Overview of the KLIK implementation process for one multidisciplinary team

Design

To retrospectively assess the KLIK PROM implementation using the CFIR framework, a mixed methods design was used. Part A consisted of a qualitative description regarding the most prominent determinants of successful KLIK PROM implementation. Part B consisted of an evaluation of current barriers in the KLIK implementation process and matching potential future strategies to reduce these barriers using the CFIR-ERIC Implementation Strategy Matching tool v1.0 [30,31] and a qualitative description of the identified barriers and strategies that have been used already by the KLIK expert team.

A. Retrospectively describing the most prominent determinants of successful KLIK PROM implementation using CFIR

The CFIR framework was used to retrospectively describe the implementation process of the KLIK PROM portal in different patient groups and hospitals throughout the Netherlands and to identify determinants in this process. Only the determinants relevant for the KLIK implementation process were described. To define which determinants were relevant for successful KLIK PROM implementation the following steps were taken. First, the KLIK PROM implementation process was described and discussed by the KLIK expert team, using all 39 CFIR constructs. However, for the reason of clarity, only the most prominent CFIR determinants relevant for the KLIK PROM implementation were extracted here (see Table 1). Second, the authors discussed which facilitators they found most prominent to describe. If the majority of authors considered a CFIR construct as valuable, it was included in the qualitative description. The KLIK implementation process was described based on published literature regarding the development, effectiveness, and implementation of KLIK in various settings and options for visual feedback of the PROMs (Supplemental Table 1) and unpublished literature (e.g., the KLIK user manual and training) about the KLIK portal and on experiences of the KLIK expert team.

B. CFIR-ERIC Implementation Strategy Matching Tool to identify *current* barriers of the KLIK PROM portal implementation

The CFIR-ERIC Implementation Strategy Matching tool v1.0 [31] was used to identify current barriers in the ongoing KLIK implementation and to match implementation strategies that address the identified barriers. The CFIR-ERIC tool is based on the CFIR framework and the 73 Expert Recommendations for Implementing Change (ERIC) implementation strategies [32]. During the development of this tool [30], implementation researchers and clinicians (panelists) were presented with brief descriptions of barriers based on CFIR construct definitions. They were asked to rank implementation strategies that would best address each barrier.

Within the provided Excel tool, one can indicate which CFIR constructs are barriers to implementation. Five KLIK expert team members based in the Emma Children's hospital Amsterdam UMC and three in the Princess Máxima Center for pediatric oncology involved in the implementation of the KLIK PROM portal independently indicated which of the 39 CFIR constructs were perceived as *current* barriers in the overall KLIK implementation. These eight expert team members include all authors. When the majority (5 or more members) of the KLIK expert team identified a CFIR construct as barrier, this was entered in the matching tool. Specific agreement (both positive and negative, including 95% confidence intervals) was calculated according to De Vet et al. [33] using R.

Consequently, the tool provided output with percentages showing which ERIC implementation strategies can best be used to reduce these specific CFIR barriers. Percentages reflect the proportion of panelists endorsing a strategy appropriate for that barrier. Strategies are sorted by the cumulative percentage value. According to the tool, the strategies with the highest cumulative percentages are most effective in reducing the combined identified barriers [30]. In the results, the ten highest cumulative percentages, and, therefore, the overall advised strategies for the specific KLIK barriers will be shown. In addition, for every identified barrier using the CFIR-ERIC tool, the authors discussed what was already done in the past to reduce the impact of this barrier on the KLIK implementation process and the reasons why it still remains a barrier.

Results

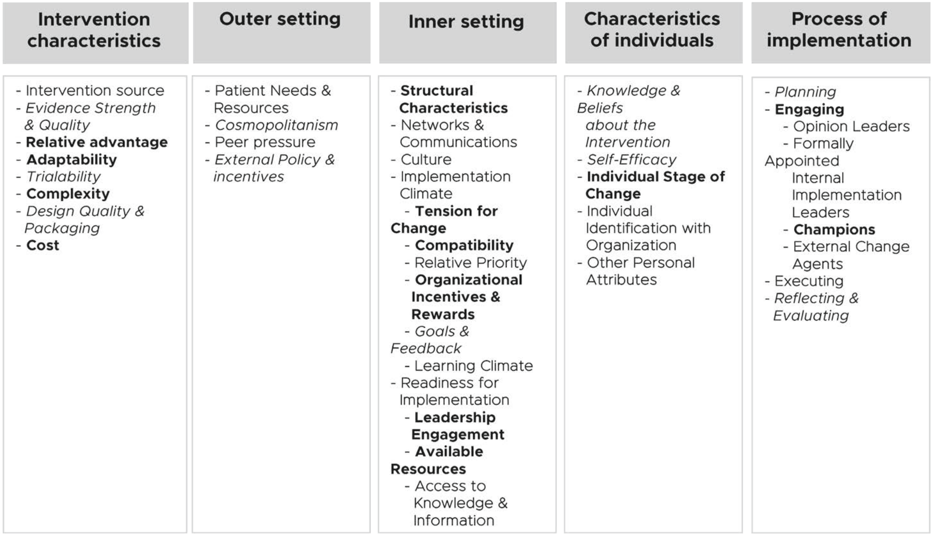
A. Retrospectively describing the most prominent determinants of successful KLIK PROM implementation using CFIR

Based on previous research and on multiple years of experience implementing the KLIK PROM portal in clinical practice, the most prominent determinants were identified by the KLIK expert team (Fig 3) and reasons for successful KLIK implementation are depicted in Table 1.

Several of the CFIR constructs were not applicable to the implementation of the KLIK PROM portal, unknown or differ too much between the different multidisciplinary teams and hospitals. These constructs include patient needs, networks & communications, culture, relative priority, learning climate, individual identification with organization, other personal attributes, and executing.

B. CFIR-ERIC Implementation Strategy Matching Tool to identify *current* barriers of the KLIK PROM portal implementation

Of the 39 CFIR constructs, 13 were identified by the KLIK expert team as current barriers for implementing the PROM portal using the CFIR-ERIC matching tool. The total specific agreement was 68.1% (95% CI 59.6%-77.6%), positive agreement (CFIR barrier) was 75.9% (95% CI 68.1%-84.6%), and negative agreement (no CFIR barrier) was 53.1% (95% CI 44.0%-63.2%). In Table 2 and Fig 3, the 13 barriers are shown. Per barrier is described what is already done as well as the challenges that remain.



Intervention characteristics	Outer setting	Inner setting	Characteristics of individuals	Process of implementation

- Intervention source
- *Evidence Strength & Quality*
- **Relative advantage**
- **Adaptability**
- *Trialability*
- **Complexity**
- *Design Quality & Packaging*
- **Cost**
	- Patient Needs & Resources
	- *Cosmopolitanism*
	- Peer pressure
	- *External Policy & incentives*
		- **Structural Characteristics**
		- Networks & Communications
		- Culture
		- Implementation Climate
		  - **Tension for Change**
		  - **Compatibility**
		  - Relative Priority
		  - **Organizational Incentives & Rewards**
		  - *Goals & Feedback*
		  - Learning Climate
		- Readiness for Implementation
		  - **Leadership Engagement**
		  - **Available Resources**
		  - Access to Knowledge & Information
			- *Knowledge & Beliefs about the Intervention*
			- *Self-Efficacy*
			- **Individual Stage of Change**
			- Individual Identification with Organization
			- Other Personal Attributes
				- *Planning*
				- **Engaging**
				  - Opinion Leaders
				  - Formally Appointed Internal Implementation Leaders
				  - **Champions**
				  - External Change Agents
				- Executing
				- *Reflecting & Evaluating*

Most prominent determinants are indicated in *italics*.
The 13 identified current barriers using the CFIR-ERIC matching tool are indicated in **bold**.

Figure 3. Overview of the five domains of CFIR, indicating determinants and barriers for the implementation of the KLIK PROM portal

Matching ERIC strategies to CFIR barriers

The identified barriers were matched to the 73 ERIC strategies using the CFIR-ERIC matching tool. Of these ERIC implementation strategies, the top 10 strategies matching the 13 identified CFIR barriers are shown in Table 3, sorted by the cumulative percentage value. Percentages reflect the proportion of panelists endorsing a strategy for that specific CFIR barrier. The tool shows that the strategy 'identify and prepare champions' is most effective in addressing the combination of identified barriers, followed by 'promote adaptability' and 'assess for readiness and identify barriers and facilitators'.

Table 1. Description of the most prominent determinants of successful KLIK implementation using CFIR

CFIR domain	CFIR determinants	Reasons for successful implementation
Intervention characteristics	Evidence Strength & Quality	• Effectiveness studies showed that KLIK is acceptable, valuable, and feasible [7,12] • The evidence of KLIK is emphasized in the training for clinicians [34]
Intervention characteristics	Trialability	• KLIK started small and has found its way, step by step, in many hospitals and has scaled up to adult healthcare and other countries • A license agreement is signed at the start, which can be ended and therefore undo the implementation if needed • The implementation process and workflow are adapted according to the wishes of every multidisciplinary team, as the KLIK team experienced that a 'one size fits all' approach was not feasible
Intervention characteristics	Design Quality and Packaging	• Clear and direct available feedback of PROMs on a well-designed dashboard • The design of the KLIK PROM portal is evaluated positively, both by clinicians and patients [35] • A strength of KLIK is the design of the PROM feedback and the variety of options [36] • Optimization of the PROM feedback in KLIK is an ongoing process, based on scientific knowledge [37] and user experience
Outer setting	Cosmopolitanism	• Worldwide, there is increased motivation for the use of PROMs in clinical practice, e.g., Value Based Healthcare supports the use of PROMs, which facilitates the implementation climate • The KLIK expert team shares common experiences with other hospitals through collaborations and networks (e.g., ISOQOL, PROMIS, research projects, implementation in many Dutch hospitals and the UK). Therefore, the KLIK PROM portal is increasingly well known and more visible for interested stakeholders
Outer setting	External Policy & incentives	• Former research showed lack of formal agreements, such as policy and work plans on using KLIK at a hospital level [29]. However, this is changing, because from a governmental perspective, collecting PROMs or using Routine Outcome Monitoring for benchmarking purposes is increasingly encouraged or even obligated
Inner setting	Goals and feedback	• During the KLIK training goals on implementing PROMs are clearly communicated, as previously different expectations were noticed (e.g., discussing PROMs in the consultation room versus collecting PROMs for research purposes), which may hinder the implementation • Clinicians receive feedback regarding the implementation process during the annual evaluation meetings

Table 1. *(continued)*

CFIR domain	**CFIR determinants**	**Reasons for successful implementation**
Characteristics of individuals	Knowledge & Beliefs about the intervention	• Multidisciplinary teams initiate implementation themselves and are, therefore, motivated to use KLIK. However, some clinicians of a team may have a negative attitude and show resistance, because they do not know the added value of using PROMs in clinical practice. The KLIK training provides knowledge of underlying principles and helps to generate enthusiasm • Research shows that clinicians are more satisfied about their provided care when using PROMs [35] and that the majority of clinicians experience personal benefit from using KLIK, e.g., by helping them in communicating with patients/parents [36]
Characteristics of individuals	Self-efficacy	• The KLIK training provides clinicians with knowledge, tools, and skills to feel competent to implement KLIK in their practice. However, there could even be more emphasis on training communication skills, as some clinicians report low confidence in discussing psychosocial topics with their patients • Research shows that most clinicians have sufficient knowledge to use KLIK as intended [29] • Current focus is on empowering patients to discuss PROMs with their clinician, for example by developing educational videos
Process of implementation	Planning	• The steps necessary to implement KLIK are clearly defined (see Figure 2) and adapted to the specific situation of each multidisciplinary team. • The KLIK expert team supports this process.
Process of implementation	Reflecting & Evaluating	• During the annual evaluation meetings, the team members are provided with feedback regarding response rates and progress, in order to improve implementation outcomes [35]. • Patient panels are consulted about their needs/preferences with regard to the implementation process.

Table 2. Identified ***current*** barriers for the KLIK PROM portal implementation using the CFIR-ERIC matching tool

CFIR domain	CFIR barrier	What is already done?	Why still a barrier?
Intervention characteristics	1. Relative advantage	• Overall, clinicians working with the KLIK PROM portal think it is a valuable tool to monitor PROMs in their patients [12,29,35] • The advantages about the KLIK PROM portal and discussing PROMs in the consultation room are spread during conferences and in scientific papers	Some stakeholders are reluctant to change and do not see the advantages of using PROMs, or suggest that alternative solutions (e.g., administering PROMs using EHRs or on paper) can be used
Intervention characteristics	2. Adaptability	• KLIK is a very flexible system where many individual wishes of the multidisciplinary teams can be met (e.g., different PROMs for different patient groups at different timeslots, for different ages, various forms of feedback different for specific clinicians) • To make KLIK as user-friendly as possible. For example, KLIK is available in different languages and proxy reported PROMs are offered for patients with disabilities [13]	Clinicians prefer the intervention as tailored as possible. A standard set of PROMs is currently being offered to patients automatically based on age and patient group, not yet on an individual patient level (selecting specific PROMs per individual patient per visit)
Intervention characteristics	3. Complexity	• The KLIK PROM portal is easy to use as a result of its origin in pediatrics. A recent evaluation study shows that the majority of clinicians (72%) think KLIK is easy to use [35] • Together with the use of the KLIK PROM portal, hospitals receive support and advise from the KLIK expert team during all steps of the implementation (see Fig 3)	For some clinicians KLIK remains complex to use, for example if they are not familiar with ICT. In addition, it requires additional actions, because clinicians need to actively motivate patients and sometimes send out the PROMs to their patients
Intervention characteristics	4. Cost	• KLIK is being offered at low costs, as we are a non-profit organization, and alternative portals are often more expensive	Within healthcare there are often insufficient financial resources. Therefore, some teams still decide to refrain from using PROMs because of the additional costs
Inner setting	5. Structural characteristics	• In general, KLIK is being implemented bottom-up, where small multidisciplinary teams show their interest in using KLIK	Hospitals are large organizations, and obtaining permission to change existing workflows can be a long process The board of the hospital might not be aware of bottom-up processes and can, therefore, be perceived as a barrier in larger scale implementation
Inner setting	6. Tension for change	• Champions (clinicians who are enthusiastic about using KLIK) can explain the added value of using PROMs in clinical practice and persuade colleagues in trying out KLIK as well.	Some clinicians do not see the current situation (not using PROMs in clinical practice) in a need of change.

Table 2. *(continued)*

CFIR domain	CFIR barrier	What is already done?	Why still a barrier?
Inner setting	7. Compatibility	• At the start of the KLIK implementation, the KLIK expert team advices on how to fit KLIK best into the existing workflow • Recently, in four hospitals, a front-end integration with KLIK and the EHR (Epic© and HiX©) is realized	A study showed that a perceived barrier for stakeholders was compatibility (24% of clinicians indicated that the KLIK method did not fit well with current routines) [29]. To make it better fit with existing workflows, KLIK should be integrated into the EHR in all hospitals
Inner setting	8. Organizational incentives & rewards	• Clearly communicate incentives (e.g. communication tool, improvement of quality of care, data can also be used for scientific purposes) of using PROMs in clinical practice for both patients and clinicians	Sometimes there are no incentives in the opinion of multidisciplinary teams and they, therefore, do not promote the use of PROMs in clinical practice
Inner setting	9. Leadership engagement	• License agreements are signed by an authorized signatory and it therefore approves the implementation	Key organizational leaders or managers could show more commitment and involvement in KLIK by promoting it actively. In addition, in the current situation, they are not held accountable for implementation of the innovation
Inner setting	10. Available resources	• KLIK has received several grants for the implementation and developed a business model to provide financial resources for the KLIK expert team in addition to the external resources	There is no structural funding yet for the KLIK expert team. To continue implementing KLIK, we are currently working on a new business model where we are not dependent on external funds, but can provide the use of KLIK at low costs For clinicians, money or time to discuss PROMs can be a barrier in implementing PROMs
Characteristics of individuals	11. Individual stage of change	• When clinicians experience benefits from implementing PROMs, they become more enthusiastic • By training clinicians, the skills necessary to implement and discuss PROMs are provided • During the annual evaluation meetings we identify clinicians that do not perceive enough benefits or forget using KLIK. These meetings keep the clinicians focused on the goal of discussing PROMs • From the perspective of patients, information letters, flyers and educational videos are provided to give them the skills to complete and discuss PROMs. In addition, focus groups are held to explore their experiences regarding KLIK, in order to further optimize KLIK	Clinicians that do not feel skilled or enthusiastic about using the innovation in a sustained way are resistant to use the intervention. Feedback from patients includes that PROMs are not discussed by the clinician, they sometimes do not see the added value, and PROMs can be long and repetitive

Table 2. *(continued)*

CFIR domain	**CFIR barrier**	**What is already done?**	**Why still a barrier?**
Process of implementation	12. Champions	• Most teams have a champion (an individual who support the KLIK implementation in a way that helps to overcome indifference of resistance by key stakeholders) who is motivated to start implementing KLIK for their patients	Some champions seem to have insufficient influence to convince their colleagues
Process of implementation	13. Engaging (Key stakeholders)	• Clinicians are involved in the entire implementation process • Also patients are more and more involved in the KLIK PROM portal, e.g., by asking their opinion in both qualitative and quantitative studies, developing educational videos to prepare them for the outpatient consultation, and by collaborating with patient associations	Patient engagement can be increased, for example, at the start of the implementation to explore the relevant PROs for patients

Table 3. Output of the CFIR-ERIC matching tool: top 10 ERIC strategies matched to the 13 identified CFIR barriers for current KLIK implementation

CFIR Barriers **ERIC Strategies**	Cumulative Percent	1. Relative advantage	2. Adaptability	3. Complexity	4. Cost	5. Structural Charac-teristics	6. Tension for Change	7. Compatibility	8. Organizational Incentives & Rewards	9. Leadership Engagement	10. Available Resources	11. Individual Stage of Change	12. Champions	13. Key Stakeholders
Identify and prepare champions	449%	45%	23%	30%	12%	27%	48%	21%	25%	41%	4%	44%	67%	63%
Promote adaptability	312%	24%	73%	40%	16%	23%	17%	45%	4%	9%	4%	28%	11%	17%
Assess for readiness and identify barriers and facilitators	310%	24%	31%	30%	16%	36%	35%	34%	13%	14%	13%	12%	15%	38%
Alter incentive/ allowance structures	305%	28%	0%	7%	44%	18%	22%	10%	71%	32%	17%	32%	7%	17%
Conduct local consensus discussions	287%	24%	31%	7%	4%	14%	43%	41%	8%	27%	0%	20%	26%	42%
Inform local opinion leaders	261%	28%	15%	13%	12%	14%	39%	3%	17%	18%	0%	28%	44%	29%
Access new funding	226%	10%	0%	3%	72%	5%	0%	3%	38%	9%	78%	0%	4%	4%
Tailor strategies	218%	17%	35%	27%	12%	18%	13%	38%	17%	5%	9%	8%	4%	17%
Create a learning collaborative	218%	7%	23%	33%	8%	18%	9%	14%	13%	5%	9%	28%	19%	33%
Identify early adopters	217%	17%	27%	20%	8%	23%	13%	10%	13%	9%	0%	24%	41%	13%

Percentages shown reflect the proportion of panelists [30] endorsing a strategy appropriate for that barrier.

Discussion

This paper aimed to retrospectively describe the most prominent determinants of successful KLIK PROM portal implementation using the Consolidated Framework for Implementation Research (CFIR) and to identify current barriers and matching implementation strategies for the KLIK implementation using the CFIR-ERIC Implementation Strategy Matching Tool.

This retrospective evaluation shows that the strength of the KLIK PROM portal implementation lies particularly in the following CFIR domains: intervention characteristics (e.g., easy to use, direct feedback), characteristics of individuals (e.g., motivated clinicians), and process of implementation (e.g., support of the KLIK expert team). In addition, the climate of the outer setting is changing and patient-reported outcomes are more valued, which facilitates the implementation of the KLIK PROM portal. On the other hand, barriers in the implementation lie mainly in the domain of the inner setting and the intervention characteristics. Regarding the inner setting, involving and motivating all stakeholders at various levels of the multidisciplinary teams and hospitals is challenging. Regarding the intervention characteristics, mainly the tension field of providing optimal support of the KLIK expert team and the use of the KLIK PROM portal on the one hand and keeping low costs on the other hand is difficult. These findings are in line with another study discussing PROM implementation [38], where the authors describe the same relevant CFIR domains. This implies that the CFIR domain ‘outer setting’ might be less relevant than the other four domains when describing PROM implementation. However, a recent study on PREM implementation did find relevant outcomes regarding the outer setting, or macro level [39], and other literature on PROMs in palliative care also conclude that all CFIR domains need consideration for effective implementation [40].

Most CFIR domains were applicable to implementation of the KLIK PROM portal, showing that CFIR can be used in the context of implementing PROMs. However, the framework is not specifically developed for this context, resulting in insufficient attention for specific parts of the PROM implementation. For example, the content, length, and psychometric properties of PROMs are important factors for successful implementation of PROMs in clinical practice and are not addressed by the CFIR framework.

The CFIR is a comprehensive framework based on various published implementation theories [22], resulting in a very extensive framework consisting of many constructs, which can make it complicated to use. The five domains of the framework are intertwined and interacting, making it hard to determine where points of attention can be placed without iteration. In particular, the domain inner setting consists of many overlapping subdomains with intangible concepts. In addition, a recent systematic review on implementing e-health interventions shows blind spots in current literature about contextual factors (such as the organization), which makes it difficult for clinicians and researchers to understand these concepts and to translate it to clinical practice [41]. In previous literature, other weaknesses of CFIR are mentioned. In their systematic review on PROM implementation, Foster et al. identified the importance of different stages of the implementation process, which is not captured by CFIR [1].

The CFIR can be described as a determinant framework [24]. Determinant frameworks specify which factors (determinants) have a facilitating or inhibiting effect on the implementation. These frameworks thus describe the influence of processes on the implementation outcomes, but do not address these implementation outcomes, in contrary to evaluation frameworks. Therefore, it would be useful to use the CFIR in combination with another type of model. For instance, a widely used model on implementation outcomes is the "conceptual model of implementation research", as described by Proctor and colleagues [18]. In order to improve outcomes for patients, it is important to be able to determine which determinants relate to which specific implementation outcomes. Only then can be reliably concluded which specific strategies work for which implementation outcomes.

The CFIR-ERIC Implementation Strategy Matching tool provided implementation strategies for the identified CFIR barriers [30]. Some of the suggested implementation strategies can be explored and used in the KLIK PROM portal implementation in the upcoming years. For example, assess key stakeholders for readiness is an ongoing process and still a challenge. By conducting individual interviews with the more reluctant clinicians underlying resistance can be better understand and addressed. In addition, identifying expected barriers and facilitators in the implementation process by actively discussing these topics in multidisciplinary team meetings in a more structured way is necessary. Also, incentives for patients in using the KLIK PROM portal could be explored further by increasing patient engagement.

However, not all suggested strategies by the matching tool provided new insights as they were directly linked to the perceived barrier (e.g., identify and prepare champions for the barrier 'champions' and access new funding for the barrier 'cost') and therefore were already known by the KLIK expert team. In addition, some strategies are currently being worked on (e.g., tailoring strategies, inform local opinion leaders, and identify barriers in the implementation process). Though, these strategies are difficult to implement and the tool underlines the need to pay more attention to these important strategies.

To further improve the KLIK implementation process in daily clinical practice, both the identified current barriers as well as the strategies extracted from the CFIR-ERIC tool can be used, to provide some examples:

- Recently, more and more evidence has become available for the *relative advantage* of implementing PROMs [42,43]. We incorporate this information in the training to clinicians (step 4 in Fig. 2) and in the information we send to interested stakeholders to overcome this barrier. This might also affect the barrier *tension for change*.
- To overcome the barrier of *structural characteristics*, creating awareness within the board of hospitals to facilitate larger scale implementation can be an opportunity. This might also affect the barrier *leadership engagement*.
- Regarding *engaging key stakeholders*, patients and patient associations should be more involved in e.g., selecting PROs and PROMs and choices regarding frequency (step 1 in Fig. 2).

On the other hand, some current barriers will likely remain or even become more prominent in the future. For example *complexity*, due to increased privacy legislation, the KLIK PROM portal requires now the use of two-factor authentication, which does not benefit the usability of KLIK for some users.

At the time the implementation of the KLIK PROM portal in clinical practice started, a variety of implementation frameworks (including CFIR) and instruments to monitor and evaluate the implementation process from the start were not yet available. Just as we have evolved as a group, implementation science has evolved over the past decade as well. Implementation of the KLIK PROM portal was therefore essentially a process of "learning by doing". Each time a specific multidisciplinary team showed interest in using KLIK, novel challenges appeared. As a result, a wide range of implementation strategies were used to tackle these particular issues. Notably, without realizing it at the time, many of the principles and strategies that are outlined in the CFIR tool were applied.

We recommend groups starting to implement PROMs in their setting to use an implementation science framework, like the CFIR, as knowing which factors need to be taken into account can lead to a more successful implementation in a specific context. The CFIR authors have developed an Interview Guide Tool (https://cfirguide.org/tools/) that can help researchers to question constructs of the CFIR that apply for the specific context. As every individual implementation process is different, also the constructs that are applicable differ.

Strengths of this study include the broad view of the retrospective description; multiple populations and multicenter experiences have been taken into account. In addition, the description is based on long-term experience and on published literature. However, this paper has several limitations. First, although a deliberate choice, no standardized qualitative research methods were used in this paper as the aim of this paper was to give a retrospective description of the KLIK PROM implementation process using the CFIR framework with the overarching purpose to create more awareness for the use of implementation science in PROM research. Second, the determinants and barriers for successful KLIK PROM implementation were described based on the experiences of the KLIK expert team (existing of members from two different centers) and this could have led to a selective view from the KLIK expert team. However, the KLIK expert team works closely with a variety of stakeholders on a day-to-day basis, including clinicians, patients and parents. They furthermore provide opportunities for stakeholders to provide feedback during regular evaluation meetings. In addition, recently two evaluation studies were carried out to gain more insight into the perspectives of clinicians [35] and pediatric patients and parents [44]. Thus, even though other stakeholders were not literally represented as co-authors, it can be assumed that their opinions are represented throughout this study.

In conclusion, this retrospective approach showed that the CFIR provides clinicians and scientists guidance during a healthcare implementation process and can be used in all phases of implementation, although it is a quite extensive and complex framework with some overlapping constructs. For example, the CFIR can be used retrospectively, reflected in this article, to describe the implementation process and its determinants and to identify remaining barriers. An advantage of using this theoretical framework prior to start of implementation is that clinicians become aware of the possible facilitating determinants and barriers for implementation. Using an implementation science framework, like the CFIR, is recommended for groups starting to use PROMs in clinical care as knowing which factors need to be taken into account can lead to a more successful implementation in a specific context.

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Keywords

Implementation science, PROMs, Clinical practice, Framework

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Supplement 1 - Overview of the KLIK studies

Authors	Year	Short title	Setting	Journal	Main outcomes
Engelen et al [25]	2010	Development of QLIC-ON (predecessor of KLIK)	Pediatric oncology	Patient Educ Couns	A description of the development and implementation of the QLIC-ON Profile in pediatric oncology.
Haverman et al [28]	2011	Development of KLIK	Pediatric rheumatology	Pediatrics	The use of the KLIK ePROfile makes a positive contribution to systematic monitoring and discussing HRQOL issues in the consultation room.
Engelen et al [7]	2012	Effectiveness of KLIK	Pediatric oncology	Pediatr Blood Cancer	Using KLIK, emotional and psychosocial problems are more often discussed and identified during the consultation.
Engelen et al [45]	2012	Influences of PROs on discussion of psychosocial issues	Pediatric oncology	Pediatr Blood Cancer	The use of PROs increases the amount of psychosocial topics that are discussed during the consultation.
Haverman et al [12]	2013	Effectiveness of KLIK	Pediatric rheumatology	Pediatrics	The use of the KLIK ePROfile increased discussion about psychosocial topics and satisfaction of the clinician with the provided care.
Haverman et al [13]	2014	Implementation of KLIK	Pediatrics	Clin Pract Pediatr Psychol	The implementation of KLIK is feasible and workable.
Schepers et al [46]	2014	PROs in pediatric oncology: suggestions for future use	Pediatric oncology	Pediatr Blood Cancer	Parents and clinicians consider the use of PROs as an important part of standard care.
Santana et al [34]	2015	Training clinicians	Adult oncology, lung transplant and pediatrics	Qual Life Res	An important step in the implementation of PROs is training clinicians in how to use and act on PROMs in clinical practice.
Schepers et al [29]	2017	Real-world implementation of PROs	Pediatric oncology	Psycho-oncology	Implementing the KLIK PROM portal in pediatric oncology and a description of the barriers and enablers for this implementation process.
Schepers et al [47]	2017	Electronic feedback of the psychosocial assessment tool	Pediatric oncology	Support Care Cancer	Implementation of the electronic version of the psychosocial assessment tool seems feasible.
Schepers et al [48]	2018	The use of the electronic psychosocial assessment tool	Pediatric oncology	Cancer	Scores of the electronic psychosocial assessment tool at diagnosis are good predictors of parental stress in the future.
Haverman et al [36]	2019	Feedback options of PROMs in KLIK	Clinical practice	Med Care	Customization of the KLIK PROM portal is needed per patient group and per PROM.
Teela et al [35]		Experiences of clinicians with the use of KLIK	Clinical practice	Submitted	Clinicians are generally satisfied with the usability of the KLIK PROM portal and the feedback of the KLIK ePROfile.
Van Muilekom et al [44]		Experiences of patients and parents with the use of KLIK		In progress	Patients and parents are satisfied with the use of the KLIK PROM portal. It helps them during the conversation with the clinician.

7

Psychometrics of the patient-reported outcomes measurement information system measures in hemophilia; the applicability of the pediatric item banks

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Abstract

Background

The use of patient-reported outcomes measures (PROMs) is important in hemophilia care, as it facilitates communication between patients and clinicians and promotes patient-centered care. Currently, a variety of PROMs with insufficient psychometric properties are used. Patient-reported outcomes measurement information system (PROMIS) measures, including Computer Adaptive Tests, were designed to measure generically and more efficiently and, therefore, are an alternative for the existing PROMs.

Objectives

To assess the feasibility, measurement properties, and outcomes of 8 PROMIS pediatric measures for boys with hemophilia.

Methods

In this multicenter study, boys with hemophilia completed 8 PROMIS measures and 2 legacy instruments. Feasibility was determined by the number of completed items and floor or ceiling effects (percentage of participants that achieved the lowest or highest possible score). Reliability was assessed as the percentage of scores with a standard error ≤ 4.5. Construct validity was evaluated by comparing the PROMIS measures with the legacy instruments. Mean PROMIS T-scores were calculated and compared with the Dutch general population.

Results

In total, 77 boys with hemophilia participated. Reliability was good for almost all PROMIS measures and legacy instruments. The total number of completed items varied from 49 to 90 for the PROMIS pediatric measures, while the legacy instruments contained 117 to 130 items. Floor and ceiling effects were observed in both the PROMIS measures (0-39.5%) and legacy instruments (0-66.7%), but were higher for the legacy instruments.

Conclusions

The PROMIS pediatric measures are feasible to use for boys with hemophilia. With the use of the PROMIS measures in clinical care and research, a step toward worldwide standardization of PROM administration can be taken.

1. Introduction

Hemophilia A or B are x-linked bleeding disorders that are caused by a deficiency of the coagulation proteins factor (F) VIII (haemophilia A) or FIX (haemophilia B), resulting in excessive bleeding typically in joints and muscles, spontaneously or after minor trauma. The risk of bleeding is related to the severity of the factor deficiency, and repeated bleeds can cause pain, functional impairment, and acute and long-term disabilities, especially when treated inadequately [1-3]. In recent decades, the treatment of hemophilia has greatly improved. In children with a severe phenotype of hemophilia the treatment is now mainly focused on the prevention of bleeding by prophylactic therapy with factor concentrates (eg, prophylaxis) or non-factor alternatives (eg, emicizumab) [1,4-6].

With these treatment advancements, health outcomes and the health-related quality of life (HRQoL) of children with hemophilia have significantly improved. Children now have a near-normal life expectancy and HRQoL, experience a lower treatment burden, and are less limited in activities of daily living [5,7-10]. However, hemophilia treatment still has an impact on the lives of these children. Therefore, comprehensive care focusing on both physical and psychosocial outcomes is standard in high-income countries [4,11,12]. The use of patient-reported outcomes measures (PROMs) are of important value in comprehensive care to gain insight into the consequences of hemophilia treatment [13].

PROMs are self-reported questionnaires that measure patients' perspectives on their health, well-being, and the impact of disease and treatment on their life [14,15]. PROMs can be used both at a group level to study differences between populations or to measure the effect of treatment modalities in clinical research, or at an individual level to increase awareness for patients' problems and concerns, facilitate communication, and to guide clinical decision-making [16-18]. PROMs can be disease-specific (ie, applicable to patients with a specific disease) or generic (ie, applicable to everyone, regardless of disease) and are preferably standardized and validated [19]. For measuring outcomes in children, it is important to have PROMs available for different age ranges and parent proxy questionnaires.

In hemophilia research, a wide variety of PROMs are used which makes comparisons difficult due to differences in content, age ranges, and scoring methods [13,15,20-22]. Specifically for pediatric hemophilia care, a wide variety of disease-specific PROMs (eg, CHO-KLAT, Haemo-QoL) are used without established psychometric properties to justify the use of these disease-specific PROMs in daily clinical practice [20]. For these reasons, standardization of outcomes and PROM administration in hemophilia care and research is essential, as described by Van Hoorn et al. [11,15,20,23]. Several initiatives have recently worked on core outcome sets for patients with hemophilia [24-26], resulting in the patient-reported outcomes measurement information system (PROMIS) being selected as one of the included measurement tools [26].

PROMIS provides a set of person-centered, standardized instruments to measure a broad range of health domains (physical, mental, and social health) in children [27,28]. In contrast to legacy instruments that are based on the *Classical Test Theory* (CTT), PROMIS measures were developed according to the *Item Response Theory* (IRT) [29,30]. An important advantage of the use of IRT is the option of Computerized Adaptive Testing (CAT) [28,29]. With CAT, items are offered to patients based on their previous answers. Consequently, PROMIS measures are shorter, items are more tailored to the patients' situation, and the measurement is more reliable in comparison to existing PROMs [28,30]. Recent studies showed that, in (young) adult patients with hemophilia, PROMIS measures are effective, reliable, and valid with low floor- and ceiling effects [15,31-33]. However, it is unclear if PROMIS instruments are also suitable for children with hemophilia. Therefore, the aim of this study is to evaluate the feasibility, measurement properties, and outcomes of 8 relevant PROMIS pediatric measures for boys with hemophilia.

2. Methods

2.1. Study population and procedure

All boys (8-17 years) treated for mild to severe hemophilia A or B in one of the hemophilia treatment centers in the Netherlands (Amsterdam University Medical Centers, Van Creveldkliniek, Erasmus University Medical Center, Radboud University Medical Center, or the University Medical Center Groningen) were eligible to participate in this multicenter study. Between June 2021 and December 2021, patients were invited to participate by email and received a personal link to the study website (https://promis.hetklikt.nu/hemofilie/) of the KLIK PROM portal [34]. Caregivers were asked to complete a sociodemographic questionnaire, and children were asked to complete 8 PROMIS instruments and 2 legacy instruments (Haemophilia Quality of Life Questionnaire for Children (Haemo-QoL) and Pediatric Hemophilia Activities List (PedHAL)). Children with insufficient knowledge of the

Dutch language or children who were unable to complete the PROMs were excluded, as determined by the treating clinician.

The Medical Ethics Committees of the participating centers approved this study. All participants signed online informed consent.

2.2. Measurements

2.2.1. Patient characteristics

The sociodemographic questionnaire included questions about the caregivers (eg, country of birth, educational level, marital status), the child (eg, position in family, school, sports), and clinical characteristics/variables (eg, type and severity of hemophilia, treatment, bleeding episodes, comorbidities).

2.2.2. PROMIS pediatric measures

Six PROMIS pediatric measures were assessed as CAT: V2.0 Pain Interference [35], V2.0 Fatigue [36], V2.0 Anxiety [37], V2.0 Depressive Symptoms [37], V2.0 Mobility [38], and V2.0 Peer Relationships [39]. For two domains, no CAT was available; therefore, we used the fixed scales; V2.0 Anger 9a scale [40] and V1.0 Global Health scale (7+2) [41]. All PROMIS pediatric measures use a 7-day recall period. Items are scored on a 5-point Likert scale ranging from 1 (“never”) to 5 (“almost always”), except for the domains Mobility (ranging from “not able to” to “with no trouble”) and Global Health (response categories differ for each item, eg, ranging from “excellent” to “poor”). The CAT automatically stopped when the standard error of the estimate (SE) was ≤ 3.2 (90% reliability) and/or a maximum of 12 items was administered. PROMIS total scores were calculated by transforming the item scores into a T-score ranging from 0 to 100. For all PROMIS pediatric measures, higher scores represent more of the construct (eg, more pain interference or better peer relationships). The scores of the total scales were calculated with use of the *PROMIS Assessment Centre Scoring Service* (https://www.assessmentcenter.net/ac_scoringservice).

2.2.3. Legacy instruments

The Haemo-QoL is a widely-used disease-specific instrument developed for the assessment of HRQoL of children with hemophilia [42]. The Haemo-QoL consists of different age versions. For this study, we used the Dutch versions for children 8 to 12 years (64 items) and adolescents 13 to 16 years (including children aged 17; 77 items). The Haemo-QoL measures 10 domains (*Physical Health, Feeling, Attitude, Family, Friends, Coping, Other People, Sport and School, Dealing, and treatment*), and two additional domains for the adolescent version (*Future and Relationship*).

Items are disease-specific and ask about complaints due to hemophilia (eg, the past 4 weeks I was sad due to my hemophilia). The Haemo-QoL uses a 4-week recall period and items are scored on a 5-point Likert Scale ranging from "never" to "always". Positively formulated items were inversely recoded and sum scores were calculated for each domain. Sum scores were transferred to transformed domain and total scores ranging from 0-100. Lower scores indicating better HRQoL.

The PedHAL is a validated disease-specific instrument that assesses the self-reported limitations in activities and participation for children (4-18 years) with hemophilia [43]. The PedHAL consists of 53 items, distributed over 7 domains (sitting/kneeling/standing, functions of the legs, functions of the arms, use of transportation, self-care, household tasks, and leisure activities and sports). The PedHAL uses a recall period of a month (eg, in the previous month, did you have any difficulty, due to hemophilia with walking short distances). Items are scored on a 6-point Likert Scale ranging from "impossible" to "never a problem", and a response option "not applicable". Domain scores and a summary score were calculated and converted to normalized scores ranging from 0 to 100, were higher scores represent better functioning. No scores were calculated if >50% of the items on a domain were scored as "not applicable".

2.3. Statistical analyses

The Statistical Package for Social Sciences version 26.0 was used for all statistical analyses. Descriptive analyses (means and percentages) were performed to characterize the patients.

2.3.1. Reliability & feasibility

Reliability was assessed for the PROMIS instruments under IRT and for the legacy instruments under CTT. In IRT modeling, each response pattern results in a T-score and an associated reliability (SE of measurement). An SE of ≤ 4.5 corresponds to a reliability of 80%, which has been considered the minimum acceptable level of reliability for group comparisons with the PROMIS pediatric measures [40]. To assess the reliability of the PROMIS pediatric measures, the percentage of T-scores with an SE ≤ 4.5 was calculated. Internal consistency estimates (Cronbach α) were calculated to assess the reliability of the legacy instruments through CTT.

To assess the feasibility of the instruments for use in clinical practice the number of items (for CAT: mean, minimum, maximum) that patients completed were described. In addition, floor and ceiling effects for all instruments were calculated. Floor and ceiling effects were presented as the percentage of the participants who achieved the lowest or highest possible score, respectively. A floor or ceiling effect was considered present if the commonly accepted threshold of 15% was exceeded [44,45]. Both the number of completed items as well as the floor and ceiling effects were compared between the PROMIS pediatric measures and the legacy instruments.

2.3.2. Construct validity

To evaluate the convergent validity of the PROMIS pediatric measures, hypotheses regarding the correlations between the PROMIS pediatric measures and the legacy instrument were formulated by researchers of the project group (Table 1) and tested. Moderate correlations (Spearman's rho, 0.40-0.69 [46]) were expected between PROMIS Pain Interference and Haemo-QoL Physical Health, PROMIS Depressive Symptoms and Haemo-QoL Feeling, PROMIS Mobility and PedHAL, and PROMIS Global Health and Haemo-QoL total score. Weak correlations (Spearman's rho, 0.10-0.39 [46]) were expected between PROMIS Anxiety, PROMIS Anger and Haemo-QoL Feeling, and between PROMIS Peer Relationships and Haemo-QoL Other Persons. Although the constructs of these measures were closely related, the content differs due to the disease-specific vs. generic approach.

Although no differences were expected between subgroups of boys with hemophilia based on previous literature [47,48], secondary analysis were performed comparing the mean PROMIS T-scores of the subgroups severe vs. non-severe (mild and moderate) hemophilia.

2.3.3. Outcomes

To determine which PROMIS pediatric measures were relevant for patients with hemophilia, mean T-scores were calculated and compared with Dutch reference data [49-53] from the general male population (8-18 years) using independent t-tests. In addition, transformed/normalized total and scale scores of the legacy instruments were calculated.

2.4. Synthesis of the results

Comparisons between the PROMIS pediatric measures and the legacy instruments are described regarding the number of completed items, floor and ceiling effects, and reliability.

Table 1. (Predefined) correlations between the PROMIS pediatric measures and the legacy instruments

PROMIS pediatric measures	Legacy instruments	Version	Predefined hypothesized correlations	Spearman's correlation	Confirmed
Pain Interference	Haemo-QoL Physical Health				
		8-12 years	≥0.40	0.49	Yes
		13-17 years	≥0.40	0.42	Yes
Anxiety	Haemo-QoL Feeling				
		8-12 years	≥0.10	0.60	Yes
		13-17 years	≥0.10	0.35	Yes
Depressive Symptoms	Haemo-QoL Feeling				
		8-12 years	≥0.40	0.63	Yes
		13-17 years	≥0.40	0.29	No
Mobility	PedHAL				
		8-17 years	≥0.40	0.41	Yes
Peer Relationships	Haemo-QoL Other People				
		8-12 years	≥-0.10	-0.33	Yes
		13-17 years	≥-0.10	-0.04	No
Anger	Haemo-QoL Feeling				
		8-17 years	≥0.10	0.44	Yes
		13-17 years	≥0.10	0.48	Yes
Global Health	Haemo-QoL total score				
		8-12 years	≥-0.40	-0.51	Yes
		13-17 years	≥-0.40	-0.20	No

Note: Predefined correlations were either weak (> 0.10) or moderate (>0.40) based on the content of the items and the domains assessed. Haemo-QoL, Haemophilia Quality of Life Questionnaire for Children; PedHAL, Pediatric Hemophilia Activities List; PROMIS, patient-reported outcomes measurement information system.

3. Results

3.1. Patient characteristics

A total of 77 boys with hemophilia participated (response rate: 47.5%). Of these, 70 participants (90.9%) completed all PROMs. The data of one participant was excluded, because this participant ticked the first answer for almost all questions in the PROMs (*N*=76).

Patient characteristics are shown in Table 2. The mean age was 13.5 years (range, 8-17 years). The majority of the participants (86.8%) had hemophilia A. In addition, 40.8% of the participants had a severe form of hemophilia. These participants were treated with prophylaxis with factor concentrates (19 participants) or with emicizumab

(12 participants). Participants with a moderate form of hemophilia (18.4%) received prophylaxis with factor concentrates (5 participants) or on-demand treatment (in case of a bleed; 9 participants). On-demand treatment was used for all participants with a mild form of hemophilia (35.5%).

Table 2. Patient characteristics (N=76)

	N	Mean (SD)
Sociodemographic characteristics		
Age (yrs)	76	13.5 (2.8)
	N	**%**
Age groups		
8-12 years	31	40.8
13-17 years	45	59.2
Country of birth parents		
Both parents born in the Netherlands	62	81.6
At least one parent born in foreign country	10	13.1
Unknown	4	5.3
Hemophilia characteristics	**N**	**%**
Type of hemophilia		
Hemophilia A	66	86.8
Hemophilia B	6	7.9
Unknown	4	5.3
Severity of hemophilia		
Mild (5-50%)	27	35.5
Moderate (2-5%)	14	18.4
Severe (<1%)	31	40.8
Unknown	4	5.3
Type of treatment hemophilia		
Prophylaxis with factor concentrates	24	31.5
Prophylaxis with emicizumab	12	15.8
On demand – in case of a bleed	36	47.4
Unknown	4	5.3
Inhibitor		
Current	0	0
Historically	9	11.8
No inhibitor / unknown	67	88.2

3.2. Reliability & feasibility

Table 3 and Table 4 show data on the number of completed items, reliability, and floor and ceiling effects of the PROMIS pediatric measures and legacy instruments, respectively. The reliability of the PROMIS pediatric measures was excellent (>90%

of the scores were reliable) or good (>70% of the scores were reliable) for almost all measures, except for the CAT Mobility (56.2% of the scores was reliable). The reliability of the legacy instruments was excellent with Cronbach α ranging 0.92 to 0.99.

The mean number of completed items per PROMIS pediatric measure varied from 8.8 items (range, 5-12) for the item bank *Peer Relationships* to 11.6 items (range, 8-12) for the item bank *Anxiety*. For the Haemo-QoL, the number of completed items varied from 6 items for the subscale *Haemo-QoL Other People* to 8 items for the subscale *Haemo-QoL Feeling*. The total number of items for the Haemo-QoL were 64 (8-12 years) and 77 items (13-17 years). The PedHAL consisted of 53 items. The selected set of PROMIS pediatric measures contained an average of 80.4 items (range, 49-90), while the selected legacy instruments contained 117 items for children aged 8-12 years and 130 items for children aged 13-17 years. This means a reduction of items by 31% for patients aged 8-12 years and a reduction of 38% for patients aged 13-17 years.

Floor and ceiling effects were present in both the PROMIS pediatric measures and the legacy instruments. For the PROMIS pediatric measures, floor effects were observed in 4 CATs: *Pain Interference*, *Fatigue*, *Anxiety*, and *Depressive Symptoms*. A ceiling effect was observed in the CAT Mobility. In case of floor or ceiling effects, participants had to complete the maximum of 12 items. For the legacy instruments, floor effects were observed for the *Haemo-QoL Physical Health*, *Feeling*, and *Other People*. Ceiling effects were observed for the *Haemo-QoL Physical Health*, *Feeling*, *Other People*, and *PedHAL (total score)*.

3.3. Construct validity

The correlations between the PROMIS pediatric measures and the legacy instruments are shown in Table 1. Of the 13 hypothesized correlations for convergent validity, 10 correlations were confirmed. The correlations between *PROMIS Depressive Symptoms* and *Haemo-QoL Feeling 13 to 17 years* (weak correlation), *PROMIS Peer Relations* and *Haemo-QoL Other People 13-17 years* (negligible correlation), and *PROMIS Global Health* and *Haemo-QoL total scores 13-17 years* (weak correlation) did not meet the predefined correlations.

Secondary analysis showed that boys with severe hemophilia reported more fatigue (41.2 vs. 38.0, $p = 0.04$, $d = 0.42$) compared to boys with non-severe hemophilia.

Table 3. Number of completed items, reliability, floor and ceiling effects, and mean scores of the PROMIS pediatric measures

	Number of items			Reliability	Floor	Ceiling		
PROMIS pediatric measures	**Mean**	**Minimum**	**Maximum**	**%**	**%**	**%**	***Mean T-score (σ)***	***N***
Computerized Adaptive Tests								
Pain Interference	9.2	3	12	100	39.5	0	42.1 (6.5)	76
Fatigue	11.4	8	12	100	16.2	0	39.9 (7.9)	74
Anxiety	11.6	8	12	100	28.0	0	42.3 (5.9)	75
Depressive Symptoms	10.3	4	12	100	22.7	0	43.9 (7.4)	75
Mobility	11.1	3	12	55.4	0	32.4	52.4 (6.2)	74
Peer Relationships	8.8	5	12	100	0	13.7	49.6 (7.5)	73
Scale								
Anger	9	9	9	100	9.5	0	43.6 (7.3)	74
Global Health	9	9	9	87.5	0	1.4	50.1 (7.9)	72

Note. Reliability: scores were considered reliable as SE ≤ 4.5. PROMIS, patient-reported outcomes measurement information system.

Table 4. Number of completed items, reliability, floor and ceiling effects, and mean scores of the legacy instruments

			Reliability	Floor	Ceiling		
Legacy instruments	**Version**	**Number of items**	**α**	**%**	**%**	***M (σ)***	***N***
Haemo-QoL Physical Health							
	8-12 years	7	0.92	30.0	0	16.7 (23.2)	30
	13-17 years	7	0.95	43.2	5.4	15.5 (25.9)	37
Haemo-QoL Feeling							
	8-12 years	7	0.99	60.0	3.3	10.4 (25.7)	30
	13-17 years	8	0.97	51.4	5.4	12.9 (26.5)	37
Haemo-QoL Other People							
	8-12 years	6	0.97	66.7	3.3	9.3 (24.6)	30
	13-17 years	6	0.97	56.8	5.4	13.2 (26.7)	37
Haemo-QoL total score							
	8-12 years	64	0.95	0	0	20.2 (15.4)	30
	13-17 years	77	0.98	0	0	24.5 (20.2)	37
PedHAL (total score)							
	8-17 years	53	0.98	0	44.8	96.5 (9.4)	67

Note. Data of 4 patients (Haemo-QoL 13-17 years) was excluded as these patients experienced technical difficulties during completion. Haemo-QoL, Haemophilia Quality of Life Questionnaire for Children; PedHAL, Paediatric Haemophilia Activities List.

3.4. Outcomes

Figure 1 shows the mean PROMIS T-scores for boys with hemophilia and the Dutch reference group. In comparison with the Dutch reference data, boys with hemophilia reported more pain interference ($P < .001$, mean difference = 3.85, $d = 0.60$) and they scored worse on the domain *Mobility* ($P < .001$, mean difference = -6.33, $d = -1.02$). In contrast, boys with hemophilia scored better on the domain *Peer Relationships*. On the other domains, no differences were found between boys with hemophilia and the Dutch reference group.

On the legacy instruments, boys with hemophilia scored a mean transformed total score of 20.2/24.5 (range, 0.4 – 91.6) on the Haemo-QoL (8-12 years and 13-17 years, respectively). On the PedHAL, boys with hemophilia scored a mean normalized score of 96.5 (range, 40-100) (Table 4).

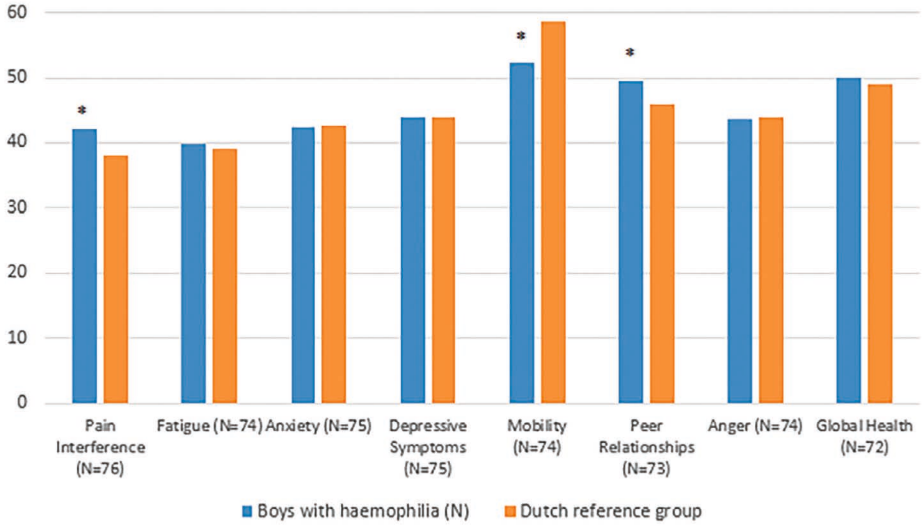

Figure 1. Mean PROMIS T-scores for boys with hemophilia and the Dutch reference group

3.5. Synthesis of the results

Table 5 presents a synthesis of the results for the PROMIS pediatric measures and the legacy instruments.

Table 5. Comparison between the measurement properties of the PROMIS pediatric measures and legacy instruments

PROMIS pediatric measures	Feasibility			Measurement properties	
	N_items	**Floor**[a]	**Ceiling**[a]	**Reliability**[b]	**Convergent validity**[c]
Pain Interference	9.2	-	+	+	+
Fatigue	11.4	+/-	+	+	n/a
Anxiety	11.6	+/-	+	+	+
Depressive Symptoms	10.3	+/-	+	+	+/-
Mobility	11.1	+	-	-	+
Peer Relationships	8.8	+	+	+	+/-
Anger	9	+	+	+	+
Global Health	9	+	+	+	+/-
Legacy instruments					
Haemo-QoL Physical Health	7	-	+	+	
Haemo-QoL Feeling	7/8[d]	-	+	+	
Haemo-QoL Other People	6	-	+	+	
Haemo-QoL total score	64/77[d]	+	+	+	
PedHal (total score)	53	+	-	+	

Haemo-QoL, Haemophilia Quality of Life Questionnaire for Children; PedHAL, Paediatric Haemophilia Activities List; PROMIS, patient-reported outcomes measurement information system.

Table footnotes
[a] floor/ceiling effect: + = <15% , +/- = 15-30%, - = ≥30%
[b] reliability: + = SE ≤ 4.5, - = SE >4.5
[c] convergent validity: + = predefined correlations are met, +/- = predefined correlations are partially met
[d] Different number of items for the age versions 8 to 12 and 13 to 17 years

4. Discussion

This study evaluated the feasibility, measurement properties and outcomes of 8 PROMIS pediatric measures in boys with hemophilia. Almost all PROMIS pediatric measures were considered feasible and reliable for use in clinical hemophilia care. The number of completed items in the selected set of PROMIS measures was lower than that of the legacy instruments, resulting in a lower burden of completing PROMs. However, at domain level, the number of completed items was higher for the PROMIS pediatric measures, except for the measures Mobility and Global Health. Floor and ceiling effects of the PROMIS pediatric measures were substantially less than that of the legacy instruments. This implies that PROMIS measures adequately cover the range of functioning of boys with hemophilia. The reliability of the PROMIS pediatric measures and the legacy instruments was good, with exception of the PROMIS CAT Mobility.

4.1 Validity

For testing convergent validity, we choose the widely-used disease specific PROMs within hemophilia pediatric care and research (Haemo-QoL and PedHAL) [20]. These PROMs aim to measure the effect of hemophilia on daily life, and specifically ask if children experience symptoms like pain, sadness, or problems with friends due to their hemophilia (eg, I was angry because of my hemophilia). This is different from the PROMIS pediatric instruments that measure a generic domain of health and assume that symptoms can occur due to multifaceted reasons (eg, I was angry) [19]. Due to these different approaches, strong correlations were not expected, and it was hard to accurately assess convergent validity. For example, the *PROMIS Peer Relationships* item bank was expected to correlate minimally with the *Haemo-QoL Other People* scale as they assess different domains of social health. The *Other People* scale of Haemo-QoL relates more to the ability to participate in social roles due to hemophilia, whereas the *Peer Relationships* item banks relates to the overall quantity and quality of relationships with peers. Similarly, the *Haemo-QoL Feeling* scale does not cover the same unidimensional domains as measured by the PROMIS item banks. Nonetheless, most convergent validity hypotheses were met in both age groups, except for *PROMIS Peer Relationships* and *PROMIS Depressive Symptoms* item banks and the *Global Health* scale for 13-17 year olds. Previous studies have shown that the subjective questioning of the *Global Health* scale ("How would you rate your own health?") may be influenced by social norms, which could be a possible explanation for a low correlation with the more objective questioning of the Haemo-QoL (total score of all subdomains), which relates much more to reported symptoms [53].

In addition, the correlation between the PROMIS pediatric measures and the legacy instruments could be negatively affected by the high floor and ceiling effects and the differences in recall period [33]. The PROMIS instruments use a recall period of 7 days, while the legacy instruments apply recall periods of 4 weeks/month [54].

A limitation of this study is that we were unable to directly compare the reliability of the PROMIS instruments and legacy instruments, due to the use of different measurement theories (IRT vs CTT). Results showed that both the PROMIS pediatric measures as well as the legacy instruments measure reliably. However, higher floor and ceiling effects were found for the legacy instruments than for the PROMIS item banks (except for the *PROMIS Mobility* item bank) negatively affecting content validity and reliability. This in accordance with previous studies on the PedHAL and Haemo-QoL instruments, where floor and ceiling effects were also found [42,43].

High floor and ceiling effects implicate that distinctive items are missing at the ends of the scale, making it difficult to distinguish patients with few or no complaints from each other [44], which results in an unreliable measurement for these patients. This also may explain the low reliability for the *PROMIS Mobility* Item bank.

4.2 Health Related Quality of Life

The results of this study showed that the HRQoL of boys with hemophilia is comparable to the Dutch general population, except for the domains *Pain Interference* and *Mobility*. The high HRQoL found in this study is comparable to other studies assessing the HRQoL of boys with hemophilia with the legacy instruments [9,10,42]. Boys with a severe phenotype of hemophilia in the Netherlands experience few joint bleeds because the annual bleeding rate is low due to adequate prophylactic therapy. It is therefore recommended to repeat this study in a group of boys with hemophilia in low-income countries with less access to effective treatment.

A limitation of this study is that as a measure of sociocultural determinants of the population, we did not have information on the race or ethnicity of participants, but did present information on place of birth of parents as a proxy for this.

4.3 Future research

The number of the PROMIS CAT items administered was still relatively high. The reason for this is that available items on the high or low end of the scale are limited and more difficult to measure reliably. Consequently, patients with no problems or complaints have to answer the maximum amount of items to reach the CAT stopping rule (SE ≤ 3.2 and/or a maximum of 12 items). To reduce the burden of administration of PROMs for patients, initiatives are currently exploring the possibility to optimize these CAT stopping rules [55]. There also have been initiatives to shorten the legacy instruments [56,57].

Conclusion

The PROMIS pediatric measures are reliable and feasible to use in hemophilia clinical care and research. Although, more research is needed to further reduce the burden of completing PROMs and to get more insight into the minimal important changes in patients with hemophilia. Innovative therapies are currently implemented of researched in clinical trials [4,5]. The need for reliable and valid instruments is crucial to measure the impact and cross-benefit of these innovative treatments. We conclude that the PROMIS measures are valid alternatives to the well-known legacy instruments, and importantly demonstrate lower floor and ceiling effects. With the use of generic PROMIS pediatric measures as used in our study, a leap can be made towards worldwide standardization of PROM administration, realizing comparisons between patient populations, the general population, patients from other disease groups and other health care settings [23].

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Conflicts of Interest

MAJL and LH are members of the Dutch-Flemish PROMIS group. SCG received an unrestricted research grant from Sobi. The other authors have no conflict of interest regarding this work/project.

Ethics Approval

The study was approved by the Medical Ethic Research board from all participating centers. All procedures performed in this study were in accordance with the ethical standards of the international and/or national research committee (Medical Ethics Committee of the Amsterdam UMC – W19_349 # 21.111) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Patient Consent

Informed consent was provided by all patients for the use of their data for this study.

Keywords

feasibility studies, hemophilia, patient-reported outcome measures, psychometrics

Essentials

- This study investigates the Patient Reported Outcomes Measurement Information System (PROMIS).
- This is a multicenter study in 76 Dutch children with hemophilia.
- The PROMIS pediatric measures are feasible to use for boys with hemophilia.
- The PROMIS pediatric measures are valid alternatives to the well-known legacy instruments.

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Part 3

Patient Reported Experience Measures

8

Use of Patient-Reported Experience Measures in Pediatric Care: A Systematic Review

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Abstract

Introduction

Patient-reported Experience Measures (PREMs) are validated questionnaires, that gather patients' and families' views of their experience receiving care and are commonly used to measure the quality of care, with the goal to make care more patient and family-centered. PREMs are increasingly being adopted in pediatric population, however knowledge gaps exist around understanding the use of PREMs in pediatrics.

Objective

To identify and synthesize evidence on the use of PREMs in pediatric healthcare settings and their characteristics.

Evidence review

Preferred Reporting Items for Systematic Reviews and Meta-Analysis guidelines governed the conduct and reporting of this review. An exhaustive search strategy was applied to MEDLINE, EMBASE, PsycINFO, Cochrane Library, and CINAHL databases to identify relevant peer-reviewed articles from high-income countries. Additionally, gray literature was searched to capture real-world implementation of PREMs. All the articles were screened independently by two reviewers in two steps. Data was extracted independently, synthesized, and tabulated. Findings from gray literature was synthesized and reported separately. Risk of bias for the studies identified through scientific databases was assessed independently by two reviewers using the National Institutes of Health Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies.

Results

The initial search identified 15,457 articles. After removing duplicates, the title and abstracts of 11,543 articles were screened. Seven hundred ten articles were eligible for full-text review. Finally, 83 articles met the criteria and were included in the analyses. Of the 83 includes studies conducted in 14 countries, 48 were conducted in USA, 25 in European countries and 10 in other countries. These 83 studies reported on the use of 39 different PREMs in pediatric healthcare settings. The gray literature retrieved 10 additional PREMs. The number of items in these PREMs ranged from 7 to 89. Twenty-three PREMs were designed to be completed by proxy, 10 by either pediatric patients or family caregivers, and 6 by pediatric patients themselves.

Conclusion and Relevance

This comprehensive review is the first to systematically search evidence around the use of PREMs in pediatrics. The findings of this review can guide health administrators and researchers to use appropriate PREMs to implement patient and family-centered care in pediatrics.

Introduction

Pediatric healthcare systems around the world continue to evolve and are increasingly acknowledging the importance of delivering patient and family-centered care (PFCC) to improve all dimensions of quality, including patients' and families' experience with care received [1]. Encouraged by the American Academy of Pediatrics, PFCC is key in the planning, delivery, and evaluation of healthcare that is grounded in mutually beneficial partnerships among healthcare providers, patients, and families. To improve and sustain the practice of PFCC, measuring patient and families' experience with the care received is necessary [2].

Patient-reported Experience Measures (PREMs) are validated questionnaires, that gather patients' and families' views of their experience receiving care. PREMs assess the impact of the process of care including communication between patient, their families and healthcare providers, information sharing, involvement of patients and their families in decision-making and are commonly used as indicators to evaluate the quality of care [2,3]. In the context of the Institute for Healthcare Improvement (IHI) Triple Aim Framework, the implementation of PREMs in healthcare leads to improved outcomes while lowering healthcare costs [4]. In addition, it allows the voice of patients and their family to inform care improvement, an important concept included in the learning health system paradigm [5].

The growing adoption of PREMs in pediatric care requires the identification of appropriate PREMs and their subsequent use in healthcare settings. PREMs are centered around the experience while receiving care (e.g., hospital environment, ease of parking, call buttons near bed etc.) rather than clinical outcomes. Moreover, most of the validated PREMs are developed in high income countries which have comparable healthcare systems and services. Thus, the objective of this systematic review is to identify and synthesize evidence on the types of PREMs used in pediatric care, and their subsequent use in healthcare systems in high income countries to inform care improvement and support pediatric learning health systems paradigm.

Methods

Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines governed the conduct and reporting of this review [6]. The protocol has been registered with OSF (DOI 10.17605/OSF.IO/3RMNC).

MeSH (Medical Subject Headings) terms, keywords and their variations were used to develop a search strategy, which was initially applied to MEDLINE database to randomly screen 100 abstracts to refine this strategy. The final search strateg was applied to MEDLINE, EMBASE, PsycINFO, Cochrane Library, and CINAHL databases. Gray literature was searched through the websites of health institutes, pediatric hospitals, conferences, professional agencies, and search engines manually, which provided an overview of real-world implementation of PREMs.

Box 1. Inclusion and Exclusion Criteria

Inclusion Criteria

1. Population: Studies that focused only on pediatric populations (≤ 18 years)
2. Measure: Studies that implemented previously validated pediatric PREMs with explicit information regarding how the PREM was validated by mentioning either the validation process or referencing a previous article that described the validation and development process.
3. Geography: Included studies also needed to have been conducted in high-income countries, loosely defined by World Bank [7].
4. Articles published from January 2000 to April 2021.

Exclusion Criteria:

1. Population: Studies that focused on adult or general populations alongside pediatric populations
2. Measure: Studies that utilized a non-validated PREM or a satisfaction survey. Editing a validated PREM threatens its validity, therefore we excluded studies where PREMs were either edited or researcher created their own questionnaires without conducting any validity testing.
3. PREM validation studies
4. Study design: opinion pieces and reviews
5. Language: Studies in languages other than English, French, Spanish, or Dutch

Covidence was used for article screening and selection against pre-defined inclusion and exclusion criteria (Box 1) [8]. In the first step, two independent reviewers screened titles and abstracts. Then, two reviewers independently screened selected articles by going through their full text. In both the steps, conflicts were resolved by discussion and consensus or by involving a third reviewer.

Following screening, two reviewers independently extracted the data. Due to heterogeneity among the studies in both statistical and methodological domains, conducting a meta-analysis was neither warranted nor plausible. We instead synthesized the results inductively by tabulating identified PREMs in various geographic locations, their type, use and characteristics. Similarly, findings from gray literature are synthesized and reported separately.

Risk of bias for all the studies identified through scientific databases was assessed independently by two reviewers using the National Institutes of Health Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies [9].

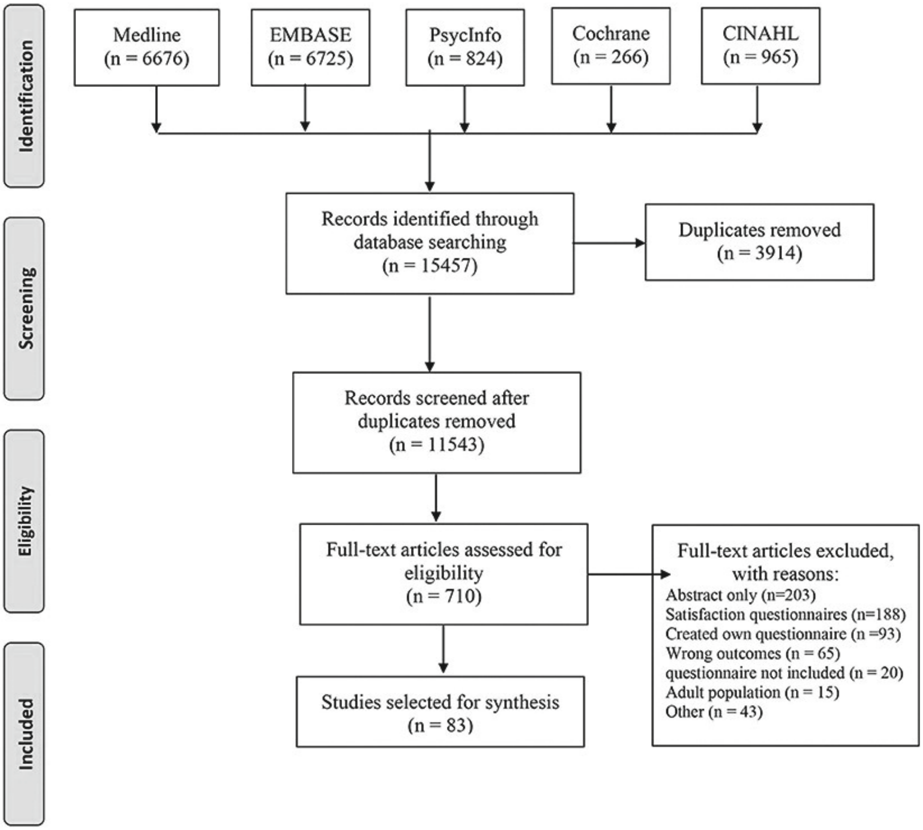

Figure 1. Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) flow diagram of identification and selection process of studies

Table 1. Summary of the pediatric patient-reported experience measures

No.	PREM Name	Year and country of origin	Type (generic/disease specific)	Patient or proxy completion	Number of items	Number of domains	Domain descriptors
1	Child HCAHPS	2015, United States [10]	Generic	Proxy	62	5	Communication with parent, Communication with child, Attention to safety and comfort, Hospital environment, Global ratings.
2	Children and Young People's Inpatient and Day Case Survey 2014	2014, United Kingdom [11]	Generic	Both	74	10	Presence of pain, Pain relief, Overall experience, Involvement in decisions, Communication on arrival, Communication about care and treatment, Communication before operation/procedure, Communication after operation/procedure, Discharge communication, Advice on post-discharge care.
3	Children's Revised Humane Care Scale (CRHCS)	2019, Finland [12]	Generic	Both	41	6	Professional practice, Information and participation in own care, Cognition of physical needs, Human resources, Pain and apprehension management, Interdisciplinary collaboration.
4	Clinician and Group Consumer Assessment of Healthcare Providers and Systems (CG-CAHPS)	2007, United States [13]	Generic	Proxy	33	N/A	N/A
5	Consultation and Relational Empathy measure (CARE)	2004, United Kingdom (Scotland) [14]	Generic	Both	10	4	Not specified.
6	Consumer Assessment of Health Plan Study (CAHPS) 2.0 Child Core Questionnaire	2014, United States [15]	Generic	Proxy	7	5	Courtesy and respect of office staff, Helpfulness of office staff, Providers' communication skills with parents, Respect shown to parents by providers, Providers' communication skills with children.
7	Consumer Assessment of Health Plans Survey (CAHPS)	2002, United States [16]	Generic	Proxy	18	9	Getting care quickly, Doctor's communication; Health plan customer service, Getting prescription medicines, Getting specialized services, Family centred care-shared decision making, Family centred care-getting needed information, Family centred care-personal doctor.
8	Consumer Assessment of the Healthcare Providers and Systems (CAHPS)	2012, United States [17]	Generic	Proxy	29	9	Care from nurses, Care from doctors, The hospital environment, Experiences in this hospital, Post-discharge overall rating, Understanding care post-discharge, About patient.

Table 1. *(continued)*

No.	PREM Name	Year and country of origin	Type (generic/disease specific)	Patient or proxy completion	Number of items	Number of domains	Domain descriptors
9	Disease-specific patient satisfaction questionnaire	2014, Germany [18]	Disease specific (IBS)	Patient	32	N/A	N/A
10	Epilepsy 12	2002, United Kingdom [19]	Disease specific (epilepsy)	Both	18	N/A	N/A
11	EMPATHIC-30	2011, Netherlands [20]	Generic (ICU)	Proxy	30	5	Information, Care and treatment, Organization and coordination of care, Parents and family engagement, Team care (pediatrician and other clinicians involved in the care of the children), Overall score.
12	Evaluation of the Quality of Diabetes Care' (PEQ-D)	2002, Netherlands [21]	Disease specific (diabetes)	Patient	14	N/A	N/A
13	Experience of Service Questionnaire (ESQ)	2002, United Kingdom [22]	Disease specific (mental health)	Both	12	N/A	N/A
14	Family-Provider Relationships Instrument-NICU (FAMPRO-NICU)	2001, United States [23]	Generic (NICU)	Proxy	65	3	Belief-desire, Feelings, Intentions.
15	FCCS (Family Centered Care Survey)	2006, Canada [24]	Generic	Proxy?	20	N/A	N/A
16	GYV (Give Youth a Voice)	2008, Canada [25]	Generic	Patient	56	4	Supportive and respectful relationships, Information sharing and communication, Support of independence, Teen centered services. Note: adapted from MPOC.
17	Inpatient Survey (IS)	2013, United Kingdom [26]	Generic	Patient	86	N/A	N/A
18	McLean Hospital's Perception of Care survey	2002, United States [27]	Generic (inpatient psychiatric care)	Both	20	4	Interpersonal aspects of care, Continuity/coordination of care, Communication/information received from treatment providers, Global evaluation of care.

Table 1. *(continued)*

No.	PREM Name	Year and country of origin	Type (generic/disease specific)	Patient or proxy completion	Number of items	Number of domains	Domain descriptors
19	MPOC-20, MPOC-32, MPOC-56	1996, Canada [28]	Generic	Proxy	20, 32, 56	5	Enabling and partnership, providing general information, Providing specific information about the child, Coordinated and comprehensive care, Respectful and supportive care.
20	Mind the Gap	2007, United Kingdom [29]	Generic	Both	22	3	The environment, Care processes, Healthcare provider characteristics.
21	Neonatal Instrument of Parent Satisfaction (NIPS)	1996, Canada [30]	Generic (NICU)	Proxy	27	N/A	N/A
22	NRC Health Patient Survey	2020, United States [31]	Generic	Proxy	20	N/A	N/A
23	P-MISS (Medical Interview Satisfaction Scale)	1986, United States [32]	Generic	Proxy	23	3	Parent communication and child communication, Parent communication and adherence intent, Distress relief and adherence
24	Parent's Perceptions of Primary Care (P3C)	2001, United States [33]	Generic	Proxy	23	6	Continuity of care, Accessibility of care, Contextual knowledge of physicians, Communication skills of physicians, Comprehensiveness of care, Coordination of care.
25	Pediatric Family Satisfaction Questionnaire (PFSQ)	2002, United States [34]	Generic	Proxy	35	3	Hospital services and accommodation, Nursing care, Medical care
26	Pediatric Family Satisfaction-ICU (pFS-ICU)	2001, United States [35]	Generic (ICU)	Proxy	24	5	How did we treat your family member (the patient), Symptom management: how well the ICU staff assessed and treated your child's symptoms, How did we treat you?, Information needs, Process of making decisions.
27	PedsQL - Healthcare Satisfaction Generic Module	2005, United States [36]	Generic	Both	26	6	Information, Family inclusion, Communication, Technical skills, Emotional needs, Overall satisfaction.
28	Picker Inpatient Survey	1990's, United States [37]	Generic	Proxy	35	7	Partnership, Overall care, Physical comfort, Information to parents, Confidence and trust, Continuity and transition, Coordination of care.

Table 1. *(continued)*

No.	PREM Name	Year and country of origin	Type (generic/disease specific)	Patient or proxy completion	Number of items	Number of domains	Domain descriptors
29	Press Ganey Inpatient Pediatric Survey	1998, United States [38]	Generic	Proxy	38	8	Admission, Nursing care, Tests and treatments; Family and visitors, Child's physician, Discharge, Personal issues, Overall assessment.
30	Press Ganey Medical Practice Survey	United States [39]	Generic	Proxy	29	6	Access to care, Visit processes, Nursing, Care provider, Personal issues, Overall assessment.
31	Press Ganey Patient Satisfaction Survey	United States [40]	Generic	Patient	Not provided	4	Inpatient overall, ED overall, Inpatient speed of admission, ED wait times to treatment
32	Press Ganey Physician Specialties Survey	United States [41]	Generic	Proxy	39	N/A	N/A
33	Press Ganey Satisfaction Survey (unique to each study)	United States [42]	Generic	Proxy	Varies	Varies	Varies
34	Swedish Pyramid Questionnaire (Quality of Patient Care Questionnaire - Parents Version)/ Swedish parent satisfaction questionnaire	2001, Sweden [43]	Generic	Proxy	63	8	Information on illness, Information on routines, Accessibility, Medical treatment, Care processes, Staff attitudes, Parent participation, Staff work environment.
35	The Picker Institute's Neonatal Intensive Care Unit Family Satisfaction survey	2014, United States [44]	Generic (NICU)	Proxy	80	8	Information and education to parents, Environment and visitation policies, Family and infant support by the NICU, Confidence and trust in the NICU, Continuity and transition, Family participation in care, Overall impressions, Coordination of care.
36	The Children's Hospital Boston Inpatient Experience Survey	2013, United States [45]	Generic	Proxy	62	8	Care from nurses, Care from doctors, Doctors/nurses/parents working together, Hospital experiences (procedures, pain management, comfort), Hospital environment, Child's medication, Arrival at and discharge from the hospital, Overall ratings.

Table 1. *(continued)*

No.	PREM Name	Year and country of origin	Type (generic/disease specific)	Patient or proxy completion	Number of items	Number of domains	Domain descriptors
37	The national cancer patient experience survey	2010-2014, United Kingdom [46]	Disease Specific (cancer)	Patient	79 (varied each year)	N/A	N/A
38	The patient-reported experience measure (PREM) for children in urgent and emergency care.	2012, United Kingdom [47]	Generic (emergency care)	Both	29	N/A	N/A
39	Young Patient Survey	2004, United Kingdom [48]	Generic	Both	89	9	Respect for patient preferences, Coordination of care, Information and education, Physical comfort, Emotional support, Involvement of family and friends, Continuity and transition, Overall quality of care, Confidentiality and privacy.

Results

Search results

The PRISMA flow diagram (Figure 1) summarizes the study selection process. The initial search identified 15,457 articles. After removing duplicates, the title and abstracts of 11,543 articles were screened. Of these, 710 were eligible for full-text review. In total, 83 articles met the inclusion criteria and were included in the analyses. These studies reported on the use of 39 different PREMs in pediatric healthcare settings (Table 1). The gray literature retrieved 10 additional PREMs that are used in clinical practice. Since many PREMs are usually copy-righted by the developers, so we did not contact authors or developer of the surveys for more information.

Characteristics of included studies

The included studies were conducted in 14 countries, including 48 studies in the United States of America, 25 studies in European countries (Austria, Finland, Germany, Greece, Iceland, the Netherlands, Norway, Slovenia, Spain, and the United Kingdom), 8 studies in Canada, and 1 study each in Australia and Singapore. Figure 2 provides an overview of the number of different PREMs that are used per country. Regarding study design, 41 of the included studies used a cross-sectional study design. The remaining study designs include 13 cohort studies, 6 mixed-methods, 6 observational, 3 quasi-experimental, 2 randomized-control trials, 2 quality improvement studies, 2 secondary data analyses, 2 retrospective data studies, and 1 of each of tlowing study designs: program evaluation, descriptive, longitudinal, case study, and pilot/feasibility studies.

The duration of studies ranged from 2 weeks to 5 years and study populations ranged from 0 to 25 years. Disease of interest varied across studies, although most studies addressed a general disease group. Disease-specific populations included epilepsy, diabetes, asthma, neurological conditions, and juvenile arthritis, among others. The identified studies also presented a diverse range of healthcare settings, including emergency rooms, NICU and PICUs, inpatient wards, and outpatient ambulatory clinic, among others. Overall, paper-pencil was the most common mode of administering PREMs (60.2%), followed by electronic (26.5%), telephone (12.0%), and/or via interview (1.2%) modes. The PREMs were completed by proxy only in 60 studies (71.4%), by patient and/or the proxy in 14 studies (16.7%), by only the patient in 7 studies (8.3%), while 3 studies did not specify (3.6%) (one study used two different PREMs that had different methods of completion) [49].

Among the studies, the most commonly used PREMs were the various Press Ganey surveys [50] (*n* = 18), the Measure of Processes of Care (MPOC)[28] (*n* = 15), Child HCAHPS [10] (*n* = 10) and HCAHPS surveys [51] (*n* = 5). Among all the included studies, there was a high amount of variation in the purpose of using PREMs. PREMs were most commonly used to evaluate factors that affected overall patient experience and to assess the experiences after an intervention, most often an improvement in quality of care. A complete overview of the characteristics of the included studies can be found in the Supplement 1.

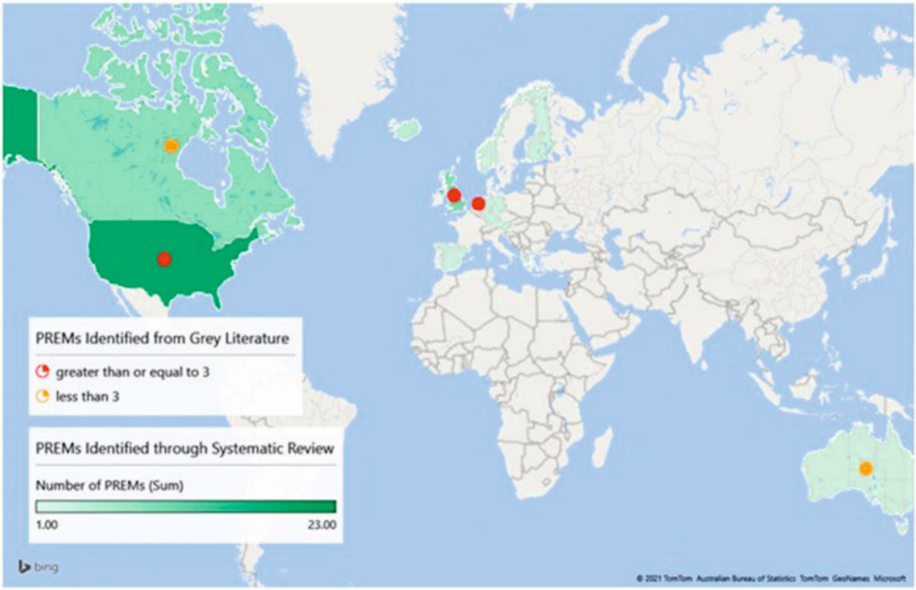

Figure 2. Geographic locations of pediatric PREMs identified through systematic review and grey literature

PREMs

This extensive review identified 39 previously validated pediatric PREMs (Table 1). These PREMs were developed in 7 countries. The greatest number of PREMs were developed in the United States of America (*n* = 21), followed by the United Kingdom (*n* = 9), Canada (*n* = 4), the Netherlands (*n* = 2), Finland (*n* = 1), Germany (*n* = 1), and Sweden (*n* = 1). Most of the included PREMs were generic (*n* = 34), aiming to measure general experiences of healthcare regardless of the disease or care sector. The included disease specific PREMs (*n* = 5) have been developed to capture the experience of healthcare from patients with inflammatory bowel disease, epilepsy, diabetes, cancer, and mental health conditions.

Of the identified PREMs, 23 were designed to be completed by proxy (59.0%), 10 PREMs (25.6%) could be completed by either or both of the pediatric patient and their caregiver(s), and 6 PREMs (13.4%) had been explicitly developed for completion by the pediatric patients themselves. The PREMs designed for completion by the pediatric patients were Disease-Specific Patient Satisfaction Questionnaire [18], Evaluation of the Quality of Diabetes Care [21], the Give Youth a Voice [25], the Inpatient Survey [26], the Press Ganey Patient Satisfaction Survey [40], and the National Cancer Patient Experience Survey [46].

Among the studies reporting questionnaire characteristics, the number of items ranged from 7 to 89, and the number of domains ranged from 3 to 10. The number of items was not provided for 5 PREMs, and the number of domains was not provided for 12 PREMs. Domains related to communication were most common, such as "communication with parent", "communication with child", "communication about care and treatment", and "provider's communication skills". Other examples of reported domains include "information", "respect", "coordination of care", "patient and family engagement", "respectful and supportive care", and "overall experiences".

Quality Assessment

The quality assessment of the included studies is presented in the Supplement 1. The quality of all studies was rated as either fair (40%) or good (60%). Overall, the risk of bias of the included studies was moderate.

Grey literature

Ten additional PREMs were identified through the grey literature search (Supplement 2). These PREMs were used to evaluate the experiences of pediatric patients and/or caregivers with daily clinical healthcare in the United States of America, the United Kingdom, the Netherlands, Canada, and Australia. The majority of hospitals in these countries asked patients to share their experiences with the use of PREMs. The gray literature search showed that a variation of PREMs, often self-developed, were used in the hospitals. Some hospitals administered PREMs to all their patients/caregivers, though most hospitals randomly invited recently discharged patients/caregivers to complete PREMs. The modes of administering PREMs identified through gray literature were similar to the ones identified through scientific databases, as listed previously.

Discussion

In this review, we document the geographic distribution of pediatric PREMs used and quantify the different PREMs administered in clinical care. PREMs are often falsely synonymized with patient reported outcome measures (PROMs) and satisfaction questionnaires, but these three types of questionnaires have distinct purposes and target different elements of patient care. In contrast to PROMs, which assess the patient's health status and measures quality of life, PREMs focus on care processes and their perceived impact on overall patient experience [52]. While dissimilar in outlook, PREMs and PROMs are often used in tandem to gather information related to the patient's care experience and outcome contentment. The terms patient satisfaction and patient experience despite being often used interchangeably, are different. Patient experience assesses whether something that should happen in a healthcare setting (such as clear communication with a provider) actually happened or how often it happened. On the other hand, satisfaction is about whether a patient's expectations about a health encounter were met [52-54] .PREMs also differ from patient satisfaction surveys, which relate to patient expectations and often involve a degree of subjectivity that is not seen in PREMs [52,55-57].

The results of our review demonstrate an international uptake of pediatric PREMs in clinical care, totaling 49 different PREMs, 39 from peer-reviewed articles and 10 from gray literature that were used in 14 developed countries spanning four continents. While administered in 14 different countries, the development of these PREMs only occurred in seven, with the greatest heterogeneity in both pediatric PREM development and implementation occurring in the United States (21), followed by the United Kingdom (9). While primarily utilized for quality improvement purposes, various research groups implemented pediatric PREMs to gauge how the responses varied between patient populations or between the patients and their family caregivers.

Measuring patient and family experience has a critical role in informing PFCC. Previous studies have explored the development and psychometric evaluation of PREMs, assessed their validity and reliability, and compared different PREM instruments for their respective utilization [58-60]. Studies have also noted differences between proxy ratings, usually coming from a family caregiver, and the ratings of a patient themselves, where the patient tends to provide lower rating regarding their own experiences of care [49,61]. Additionally, there exists a paucity of information regarding the use of pediatric PREMs, and their type (i.e., generic, disease-specific, health-setting-specific), as well as their purpose and impact on quality of care in clinical practice. These findings can be used to inform PFCC initiatives at a system-level, helping to achieve the Triple Aim and supporting the learning health system paradigm [62,63].

Additionally, research has acknowledged the correlation between PREM-implementation, the establishment of the PFCC, and the promotion of quality improvement initiatives [64]. While this information is accepted in the context of adult PREMs, much less research exists regarding the implementation and assessment of pediatric PREMs [65]. This study will inform future work in the area of PREM implementation in pediatric care.

The identified PREMs feature important domains addressing PFCC concepts such as shared-decision making and respecting patient values. A main gap identified in our review suggests that the use of disease-specific PREMs warrants more attention, with only five of the validated PREMs being disease-specific. Even among studies conducted in disease-specific settings, generic PREMs were more often chosen over an appropriate disease-specific tool. This may be related to the versatility and applicability of generic PREMs in more healthcare settings compared to disease-specific PREMs. However, disease-specific PREMs issues more specific to the corresponding disease. For example, MPOC [28] is a validated PREM commonly used for children with variety of neurodevelopmental disabilities or maxillofacial disorders. MPOC assesses family caregiver's perception of the care that their children receive at rehabilitation treatment centres, and thus can provide a better contextual understanding of patient experience specifically related to those clinical conditions. Therefore, future research examining why disease-specific PREM use and development is lacking should be explored. Additionally, while all included studies discussed the utility of using these pediatric PREMs, few examined the practicality of implementing them [66-68]. Future research examining the capacity of hospitals and physicians to incorporate these measures into clinical care is needed to pragmatically assess the likelihood of pediatric PREM administration.

A significant strength of this systematic review is the inclusion of gray literature. As this review aimed to explore the range of pediatric PREMs currently in use, gray literature sources provided an exploration of real-world PREM implementation in pediatric healthcare settings around the world. We also incorporated the perspectives of international researchers with expertise in the topics of PREMs and PROMs. This bolstered the knowledge and experience of the research team and allowed for the inclusion of different perspectives on PREM implementation from different countries.

Despite being successful in identifying the number of pediatric PREMs currently in use, this review was not without limitations. Regarding gray literature, the information about the PREMs and their implementation were often not explicitly described on hospital websites, meaning we could only provide a global description of these PREMs. PREMs created in or translated to different languages or cultural contexts may have not been available in a language that the reviewers could understand, and therefore those studies were excluded. Furthermore, the inclusion criteria of "high-income countries" potentially limited the scope of this study by geographically restricting the results. Lastly, because of the interchangeable use of the terms "experience" and "satisfaction", it is possible that due to the phrasing of study surveys, some PREMs were inadvertently excluded. However, the likelihood of this occurring was minimized due to the continual implementation of dual reviewers and the inclusion of the terms "satisfaction" and "satisfaction survey" in our initial search strategy.

The objective of this systematic review was to identify pediatric PREMs and their use in care settings. Although there are tools like the COSMIN Checklist to critically appraise the validity and reliability of PROs (PROMs and PREMs), there are no such standard tools to evaluate the strengths and weaknesses of PREMs. Moreover, evaluating these measures for their strengths and weaknesses would be subjective and context specific. Therefore, this systematic review did not evaluate the strengths and weaknesses of the PREMs, but further studies focused on assessing the strengths and weaknesses of individual PREMs may be warranted in the future.

Conclusion

This systematic review details the international use of pediatric PREMs in different pediatric clinical settings and provides an overview of the current validated pediatric PREMs available for use. The findings of this review can guide health administrators and researchers to use appropriate PREMs to implement PFCC in pediatric settings. In most of the studies included in this review, the usefulness of pediatric PREMs was highlighted. However, future additional research into the views of implementing PREMs held by clinical practitioners and patients and their families is warranted to best gauge the practicality of widespread pediatric PREM implementation.

Abbreviations

Child HCAHPS: The Child Hospital Consumer Assessment of Healthcare Providers and Systems
IHI: Institute for Healthcare Improvement
MeSH: Medical Subject Headings
MPOC: Measure of Process of Care
PFCC: Patient and Family-Centered Care
PREM: Patient-Reported Experience Measure
PRISMA: Preferred Reporting Items for Systematic Review and Meta-Analysis
PROM: Patient-Reported Outcome Measure

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Conflict of Interest

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

Data availability statement

The original contributions presented in the study are in the article/supplementary material, further inquires can be directed to the corresponding author/s.

Keywords

patient-centered care, paediatrics, systematic review, routine clinical care, patient-reported experience measures (PREMs)

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Supplement 1 - Overview of the characteristics of included studies

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
1	Krugman, Scott D; Suggs, Adrienne; Photowala, Hasnain Y; Beck, Adam, 2007, USA[1]	To measure the effects of combining an emergency department and pediatric inpatient unit.	Interventional study	18 months	General	Emergency department and inpatient care units	NR	NR	Press Ganey patient satisfaction survey	Patient	NR	Fair
2	Matziou, Vasiliki; Boutopoulou, Barbara; Chrysostomou, Anthi; Vlachioti, Efrosini; Mantziou, Theodora; Petsios, Konstantinos, 2011, Greece[2]	To record parental satisfaction with care during hospitalization and assess its determinants.	Descriptive, non-experimental	4 months	General	Surgical and inpatient care units	< 14	206	Swedish Pyramid Questionnaire (Quality of Patient Care Questionnaire - Parents Version)	Proxy	In-person interview	Good
3	Williams, Fiona; McCafferty, Aileen; Dunkley, Colin; Kirkpatrick, Martin, 2018, UK[3]	To survey patients experiences with epilepsy in the UK.	Cross-sectional	12 months	Epilepsy	Audit units	NR (many were 5-15)	2335	"Epilepsy 12", patient satisfaction experience questionnaire	Either the patient and proxy	NR	Fair
4	Voos, Kristin C; Ross, Gail; Ward, Mary J; Yohay, Anne-Lise; Osorio, Snezana Nena; Perlman, Jeffrey M, 2011, USA[4]	To evaluate the satisfaction of care providers and parents following practice changes.	Quasi-experimental	10 months	General	NICU	Neonates	28	Neonatal Instrument of Parent Satisfaction (NIPS[26])	Proxy	Paper and electronic	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
5	Nichol J.R.; Fu R.; French K.; Momberger J.; Handel D.A., 2016, USA[5]	To determine the key variables found in 3 separate PREMs.	Retrospective cohort	3 years 6 months	General	Emergency department	<20	810	Press Ganey patient satisfaction surveys	Either the patient or proxy	Paper and electronic	Fair
6	Sigurdardottir, Anna Olafia; Garwick, Ann W.; Svavarsdottir, Erla Kolbrun, 2017, Iceland [6]	To assess parental predictors of healthcare satisfaction throughout an Icelandic hospital.	Cross-sectional	2 years	General	Children's hospital (neonatal, emergency, day, and outpatient units)	≤18	219	PedsQL - Healthcare Satisfaction Generic Module	Proxy	Paper and electronic	Good
7	Timmer, Antje; Peplies, Jenny; Westphal, Max; Kaltz, Birgit; Ballauff, Antje; Clasen, Martin; Laass, Martin W; Koletzko, Sibylle, 2017, Germany and Austria[7]	To assess patients' experiences with transitioning between pediatric and adult care settings.	Cross-sectional	NR	Irritable Bowel Diseases	Varied	15-25	583	"Disease-specific patient satisfaction questionnaire"	Patient	Paper	Good
8	Sleath, Betsy; Ayala, Guadalupe X; Washington, Deidre; Davis, Stephanie; Williams, Dennis; Tudor, Gail; Yeatts, Karin; Gillette, Chris, 2010, USA[8]	To examine the correlation between parental ratings of participatory decision making and satisfaction with their pediatric asthma visits.	Cross-sectional	NR	Asthma	Pediatric Practices	8-16	320	P-MISS (Medical Interview Satisfaction Scale)	Patient	Paper	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
9	De Wit M.; Delemarre-van De Waal H.A.; Bokma J.A.; Haasnoot K.; Houdijk M.C.; Gemke R.J.; Snoek F.J., 2008, Netherlands [9]	To analyze the effects of discussing health-related quality of life (HRQoL) in pediatric patients with type 1 diabetes.	Randomized control trial	1 year	Type 1 Diabetes	Outpatient clinics	13-17	81	Patients' Evaluation of the Quality of Diabetes Care (PEQ-D)	Patient	NR	Good
10	Cohen, Eyal; Austin, Janice; Weinstein, Michael; Matlow, Anne; Redelmeier, Donald A, 2008, Canada[10]	To assess quantitatively and qualitatively the care provided to isolated versus non-isolated patients.	Prospective observational	3 months	General	Inpatient unit	NR	65	Pediatric Family Satisfaction Questionnaire (PFSQ)	Proxy	Paper	Good
11	Galbraith, Alison A; Semura, Jeanne; McAninch-Dake, Becky; Anderson, Nancy; Christakis, Dimitri A, 2004, USA[11]	To determine whether a correlation exists between emergency department use and perceived lags in accessing acute care.	Cross-sectional	5 months	General	Emergency department inpatient unit	10-14	5142	CAHPS	Proxy	Paper and telephone	Good
12	Adams, E Kathleen; Ketsche, Patricia; Zhou, Mei; Minyard, Karen, 2008, USA[12]	To track patient experience and satisfaction with a new insurance program (PeachCare).	Cross-sectional	3 years	General	Varied	≤18	2089	CAHPS	Proxy	Paper and telephone	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
13	Agra Tuñas, Maria del Carmen; Pérez Várela, Fátima; Bello Rama, Eva; Mato Guerra, Paula; Calviño Vieito, Nuria; Garrudo Díaz, Rubén; Pérez Rivas, Manuela; Rodríguez Núñez, Antonio, 2018, Spain[13]	To evaluate perceived quality of care in a PICU from the viewpoint of a patient's relative.	Cross-sectional	1 year 1 month	General	Intensive care unit	1month -16	181/308	EMPATHIC- 30 questionnaire	Proxy	Paper and electronic	Fair
14	Mah, Jean K; Tough, Suzanne; Fung, Thomas; Douglas-England, Kathleen; Verhoef, Marja, 2006, Canada[14]	To assess patient responsiveness to PREMs, the association between patient care experience and HRQoL, and whether patient and parent responses differ.	Cross-sectional	4 months	Neurologic diseases	Outpatient care	12-18	104	Family Centered Care Survey (FCCS), MPOC-20, Give Youth a Voice (GYV)	Both the patient and participant	NR	Good
15	Mason, Sheila L; Chike-Harris, Katherine E; Gyr, Bettina M; Johnson, Emily, 2019, USA[15]	To evaluate whether implementing a discharge facilitator will improve the discharge process.	Retrospective cohort	Pre-intervention: 3 months, Post-intervention: 3 months	Orthopedic diseases	Orthopedic surgery ambulatory clinic (inpatient and outpatient)	8months -18	219	Press Ganey Satisfaction Survey	Proxy	Electronic	Fair

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
16	Stefansdottir, Sara; Thora Egilson, Snaefridur, 2016, Iceland[16]	To assess parental and therapist perceptions of FCS, the factors that influence parental satisfaction with care, and how parents understood the service.	Mixed methods	1 year	Physical rehabilitation	Outpatient care	0-18	236	MPOC-32	Proxy	Electronic	Fair
17	Siebes, R C; Wijnroks, L; Ketelaar, M; van Schie, P E M; Gorter, J W; Vermeer, A, 2007, Netherlands[17]	To describe parental involvement in their child's rehabilitation, their satisfaction with treatment quality, and ideas that they have pertaining to increasing parental involvement in rehabilitation.	Longitudinal mixed methods	Phase 1:1 year 7 months, Phase 3: 6 months	Physical rehabilitation	Outpatient care	1-20	679	MPOC-56	Proxy	NR	Fair
18	Van Riper M., 2001, USA[18]	To assess maternal perceptions of their patient-provider relationship and how this relates to patient well being.	Cross-sectional	NR	Prematurity	NICU	NR	55	Family-Provider Relationships Instrument- NICU (FAMPRO- NICU)	Proxy	Paper	Fair
19	Nieman, Carrie L; Benke, James R; Ishman, Stacey L; Smith, David F; Boss, Emily F, 2014, USA[19]	To evaluate race and insurance-based disparities in PREM participation.	Cross-sectional	7 months	Otolaryngo-logic diseases	Tertiary otolaryngology clinic	0-17	130	Press Ganey Medical Practice Survey	Proxy	Paper and electronic	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
20	Wells, R D; Dahl, B; Wilson, S D, 2001, USA[20]	To assess differences in care satisfaction between patients and their primary care provider.	Prospective cohort	9 months	Asthma, bronchitis, gastro-enteritis, pneumonia	Inpatient care unit	1month -18	181	Press-Ganey Physician Satisfaction Scale	Proxy	Paper and telephone	Good
21	Toomey, Sara L; Elliott, Marc N; Zaslavsky, Alan M; Klein, David J; Ndon, Sifon; Hardy, Shannon; Wu, Melody; Schuster, Mark A, 2017, USA[21]	To measure the performance of different hospitals in a national field test.	Observational study	15 months	General	Inpatient care units	0-17	17727	Child HCAHPS	Proxy	Paper and telephone	Fair
22	Terwiel, M; Alsem, M W; Siebes, R C; Bieleman, K; Verhoef, M; Ketelaar, M, 2017, Netherlands [22]	To assess the importance of each PREM item related to FCS.	Observational study	NR	Cerebral palsy	Rehabilitation centers (outpatient care)	3-9	175	MPOC-56	Proxy	NR	Fair
23	Williams, Geraint; Pattison, Giles; Mariathas, Chrishan; Lazar, Joanna; Rashied, Muhammad, 2011, UK[23]	To pinpoint aspects of patient care that could be improved to increase patient satisfaction.	Prospective observational	10 months	Orthopedic diseases	Inpatient care units	2-6	104	Swedish parent satisfaction questionnaire	Proxy	NR	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
24	Nieman, Carrie L; Benke, James R; Boss, Emily F, 2015, USA[24]	To record patient satisfaction with clinical care and stratify results according to patient race, ethnicity, or SES.	Cross-sectional	1 year	General	Outpatient surgical care	0-17	527	Press Ganey Medical Practice Survey	Proxy	Paper and electronic	Fair
25	Tothy, Alison S; Limper, Heather M; Driscoll, James; Bittick, Nicholas; Howell, Michael D, 2016, USA[25]	To assess the effects of implementing a package of improvement activities on patient care satisfaction.	Cross-sectional	11 months	General	Emergency department inpatient care	NR	352	Press Ganey Satisfaction Survey	Both the patient and proxy	Paper	Fair
26	Tilly-Gratton A.; Nadon M.A.; Houle A.; Pelaez S.; Ducharme F.M., 2018, Canada[26]	To investigate patient satisfaction with clinical care and identify modifiable factors that may increase treatment adherence.	Mixed methods	NR	Asthma	Outpatient clinic	1-17	24	P-MISS (Medical Interview Satisfaction Scale)	Proxy	NR	Good
27	Sonneveld, H. M.; Strating, M. M. H.; van Staa, A. L.; Nieboer, A. P., 2013, Netherlands[27]	To examine the perceptions of parents, providers, and patients on transitional care experiences and to determine the extent to which perspective differences were disease specific.	Cross-sectional	NR	Type 1 diabetes, juvenile rheumatoid arthritis, neuromuscular disorder	Outpatient clinic	12-25	319	Mind the Gap	Both the patient and proxy	Paper and electronic	Fair

Supplement 1 - *(continued)*

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28	Siebes, R C; Wijnroks, L; Ketelaar, M; van Schie, P E M; Vermeer, A; Gorter, J W, 2007, Netherlands[28]	To assess the stability of the MPOC survey 1 year post implementation.	Longitudinal survey	1 year 1 month	Chronic diseases	Rehabilitation centers (outpatient care)	1-20	205	MPOC-56	Proxy	Paper	Good
29	Locke, Robert; Stefano, Mariane; Koster, Alex; Taylor, Beth; Greenspan, Jay, 2011, USA[29]	To assess parental satisfaction with the care provided in the emergency department.	Retrospective cohort	6 months	General	Emergency department outpatient care	IQ range 1-10	456	Press Ganey Satisfaction Survey	Proxy	NR	Fair
30	Camilon, P Ryan; Levi, Jessica R; Carrion, Rose A; Josephson, Gary D, 2019, USA[30]	To identify PREM items that are most closely correlated with the "likelihood of recommending practice".	Retrospective cohort	1 year	Otolaryngo-logic diseases	Outpatient care	NR	3401	Press Ganey Outpatient Medical Practice Survey	NR	NR	Fair
31	Shaw, K L; Southwood, T R; McDonagh, J E; British Society of Paediatric and Adolescent Rheumatology, 2007, UK[31]	To document the expectations and satisfaction of parents and patients with their transitional care experiences prior to and following the implementation of a related programme.	Cohort	NR	Juvenile idiopathic arthritis	Outpatient care	11-18	308	Mind the Gap Scale	Both the patient and proxy	NR	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
32	Shaw, K L; Watanabe, A; Rankin, E; McDonagh, J E, 2014, UK[32]	To evaluate transition care quality through examination of patient adherence to guidelines and whether adherence was associated with improved experience.	Cross-sectional	2 weeks	Chronic diseases	Outpatient tertiary care	11-21	787	Mind the Gap Scale	Both the patient and proxy	Paper	Fair
33	Shevell, Michael; Oskoui, Maryam; Wood, Ellen; Kirton, Adam; Van Rensburg, Esias; Buckley, David; Ng, Pamela; Majnemer, Annette, 2019, Canada[33]	To describe characteristics of children with cerebral palsy and factors that impact parent perceptions of FCS.	Cross-sectional	4 years	Neuro-developmental disabilities (cerebral palsy)	Rehabilitation centers (outpatient care)	Infants	312	MPOC-56	Proxy	Paper	Good
34	Singh, Suprit C; Sheth, Raj D; Burrows, James F; Rosen, Paul, 2016, USA[34]	To identify key factors in facilitating overall satisfaction with ambulatory neurological care.	Cross-sectional	3 years	Neurologic disease	Ambulatory care units	NR	2890	Press Ganey Satisfaction Survey	Proxy	Paper and electronic	Good
35	Singleton I.M.; Garfinkel R.; Temkit H.; Belthur M.V., 2020, USA[35]	To determine which factors impact parental satisfaction with their care provider in an outpatient clinic.	Cross-sectional	1 year 8 months	Orthopedic diseases	Orthopedic outpatient clinic	Mean age of 6	122	Clinician and Group Consumer Assessment of Healthcare Providers and Systems (CG-CAHPS)	Proxy	Paper	Fair

Supplement 1 - *(continued)*

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36	Sng, Qian Wen; Kirk, Angela H P; Buang, Siti Nur Hanim; Lee, Jan Hau, 2017, Singapore[36]	To assess whether different parental satisfaction scores in a PICU are related to differences in ethnicity and to isolate areas of the PICU that need improvement.	Prospective observational	1 year	General	PICU	Median age of 3	206	EMPATHIC- 30	Proxy	Paper	Good
37	Schreiber, Joseph; Benger, Jennifer; Salls, Joyce; Marchetti, Gregory; Reed, Lindsey, 2011, USA[37]	To evaluate parental perceptions regarding the amount of FCC exhibited at the rehabilitation facility.	Mixed methods	NR	Neuro-developmental diseases	Rehabilitation centers (outpatient care)	1-26	246	MPOC-20	Proxy	Paper	Fair
38	Rahi J.S.; Manaras I.; Tuomainen H.; Lewando Hundt G., 2005, UK [38]	To consolidate information on the needs and experiences of parents whose children have recently been diagnosed with an ophthalmic disorder.	Cross-sectional + qualitative	2 years	Ophthalmic disorders	Tertiary clinical practice	Mean age of 2	147	MPOC-56	Proxy	Paper	Good

Supplement 1 - *(continued)*

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39	Rahi, Jugnoo S; Manaras, Irene; Tuomainen, Helena; Hundt, Gillian Lewando, 2004, UK [39]	To monitor changes in the health service experiences of parents and care providers following the introduction of a key worker service.	Cross-sectional + qualitative	1 year	Ophthalmic disorders	Tertiary referral center of pediatric ophthalmology	Mean age of 2	147	MPOC-56	Proxy	Paper	Good
40	Hargreaves, Dougal S; McDonagh, Janet E; Viner, Russell M, 2013, UK[40]	To assess the association between the PREMs criteria and reported patient satisfaction in national inpatient surveys.	Cross-sectional	NR	General	Inpatient care unit	12-17 (YPS); 16-19 (IS)	7657 (YPS); 988 (IS)	Young Patient Survey (YPS); Inpatient Survey (IS)	Patient	Paper	Fair
41	Hargreaves, Dougal S; Sizmur, Steve; Pitchforth, Jacqueline; Tallett, Amy; Toomey, Sara L; Hopwood, Bridget; Schuster, Mark A; Viner, Russell M, 2018, UK[41]	To quantify the number of patients that completed the CYP PREM portion on their own and how their responses resemble their parents' experience. Secondly, CYP responses were separated and compared according to how they were completed (by the child, parent, or together).	Cross-sectional	2 months	General	Inpatient care unit	8-15	6204	Children and Young People's Inpatient and Day Case Survey 2014	Both the patient and the proxy	Paper	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
42	Myrhaug, Hilde Tinderholt; Jahnsen, Reidun; Ostensjo, Sigrid, 2016, Norway [42]	To assess the perceptions of parents and patients on the family-centeredness of their care. This was then compared to the extent of parental involvement, child well being, and parental satisfaction.	Cross-sectional	2 months	Cerebral palsy	Primary clinical practice	<6	121	MPOC-20	Proxy	Paper	Fair
43	Nagarajan, Neeraja; Rahman, Sydur; Boss, Emily F, 2017, USA[43]	To characterize the connection between patient race and their satisfaction with their care.	Cross-sectional	1 year	Inpatients	Tertiary inpatient care unit	<18	904	Press Ganey Inpatient Pediatric Survey	Proxy	Paper and electronic	Fair
44	Peng, Frederick B; Burrows, James F; Shirley, Eric D; Rosen, Paul, 2018, USA[44]	To identify the primary factors involved in orthopedic patient satisfaction.	Cross-sectional	3 years	Orthopedic diseases	Orthopedic outpatient care	NR	6195	Press Ganey Satisfaction Survey	Proxy	Paper and electronic	Fair
45	Petitgout, Janine M, 2015, USA[45]	To monitor changes in patient satisfaction following the implementation of a quality improvement initiative.	Case study	NR	General	Tertiary inpatient care unit	<21	NR	Press Ganey Satisfaction Survey	Proxy	NR	Fair

Supplement 1 - *(continued)*

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46	Kemp, Kyle A; Ahmed, Sadia; Quan, Hude; Johnson, David; Santana, Maria J,, 2018, Canada[46]	To illustrate the provincial uptake of the Child HCAHPS survey in Alberta.	Cross-sectional	1 year 6 months	General	Inpatient care unit	<17	3389	Child HCAHPS	Proxy	Telephone	Good
47	Kleinsorge, Christy A; Roberts, Michael C; Roy, Kimberlee M; Rapoff, Michael A, 2010, USA[47]	To determine how well a primary care training facility met primary care standards and how satisfied the patients were with their care.	Program evaluation	NR	General	Primary clinical care	2-18	102 (P3C); 105 (CSQ-8)	Parent's Perceptions of Primary Care (P3C); CAHPS 2.0 Child Core Questionnaire	Proxy	Paper	Fair
48	Knapp, Caprice; Madden, Vanessa; Sloyer, Phyllis; Shenkman, Elizabeth, 2012, USA[48]	To examine the effects of introducing an integrated care system on the patients' perception of care quality and their satisfaction with their child's treatment.	Quasi-experimental	2 years	General	Varied	1-21	1727	Consumer Assessment of Health Plans Survey (CAHPS)	Proxy	Telephone	Good
49	Knox V.; Menzies S., 2005, UK[49]	To record the experiences of families receiving therapy, and their perceptions of their quality of care.	Cross-sectional	1 year	Cerebral palsy	Tertiary clinical care	<18	72	MPOC-56	Proxy	Paper	Good

Supplement 1 - *(continued)*

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50	Kuo, Dennis Z.; Sisterhen, Laura L.; Sigrest, Ted E.; Biazo, James M.; Aitken, Mary E.; Smith, Christopher E., 2012, USA[50]	To assess the effects of family centered rounds on improved family experiences and health care utilization.	Prospective cohort	1 year 3 months	General	Inpatient care unit	<12	97	Consumer Assessment of Healthcare Providers and Systems measures	Proxy	Telephone	Good
51	Koves I.H.; Boucher A.; Ismail D.; Donath S.; Cameron F.J., 2008, Australia[51]	To correlate parental or patient satisfaction with better patient outcomes, improved health literacy, and elevated HRQoL.	Cross-sectional	2 months	Diabetes mellitus	Outpatient care	5-18	217	Evaluation of the Quality of Diabetes Care' (PEQD)	Both the patient and the proxy	Paper	Good
52	Miceli PJ; Clark PA, 2005, USA[52]	To document the experiences of hospitalized pediatric patients.	Retrospective database study	1 year	General	Inpatient care unit	<21	50446	Press Ganey Pediatric Inpatient Survey	Proxy	Paper	Fair
53	Janhunen, Katja; Kankkunen, Päivi; Kvist, Tarja, 2019, Finland [53]	To compare the experiences and satisfaction of patients and parents in the emergency department, and to identify key factors that predict satisfaction.	Cross-sectional	6 months	General	Emergency department	7-16	196	Children's Revised Humane Care Scale (CRHCS)	Both the patient and the proxy	Paper	Good

Supplement 1 - *(continued)*

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54	Lee, Brian; Hollenbeck-Pringle, Danielle; Goldman, Victoria; Biondi, Eric; Alverson, Brian, 2019, USA[54]	To assess the accuracy of the child HCAHPS survey in reflecting patient experiences and to determine whether external factors were influencing survey responses.	Retrospective cohort study	1 year	General	Inpatient care unit	<18	363	Child HCAHPS (C-HCAHPS)	Proxy	Paper	Fair
55	Hurtubise, Karen; Shanks, Robin; Benard, Laura, 2017, Canada[55]	To evaluate patient satisfaction with a new orthopedic physiotherapy-led clinic.	Pilot/ feasibility study	1 year 10 months	Cerebral palsy	Outpatient clinic	NR	50	MPOC-20	Proxy	Paper	Fair
56	Harder, Valerie S; Krulewitz, Julianne; Jones, Craig; Wasserman, Richard C; Shaw, Judith S, 2016, USA[56]	To assess whether 2 different aspects of care are associated with improved patient experience, and whether this association changes depending on the type of practice.	Cross-sectional	2 years	General	Primary clinical care	<18	2599	Consumer Assessment of Health care Providers and Systems (CAHPS).	Proxy	Paper	Fair
57	Groleger Srsen, Katja; Vidmar, Gaj; Socan, Gregor; Zupan, Anton, 2014, Slovenia [57]	To evaluate clinical processes of care and identify provider characteristics that are correlated with increased patient satisfaction.	Cross-sectional	NR	Chronic diseases	Inpatient and outpatient care	<18	235	MPOC-20	Proxy	NR	Good

Supplement 1 - *(continued)*

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58	Hall, Allyson G; Landry, Amy Yarbrough; Lemak, Christy Harris; Boyle, Erin L; Duncan, R Paul, 2014, USA[58]	To assess how patient and parent satisfaction fluctuates with the child's health status and their prescribed treatment plan.	Cross-sectional	3 years	General	Varied	<21	11067	Consumer Assessment of Health Providers and Systems (CAHPS) surveys	Proxy	Telephone	Good
59	Fustino, Nicholas J; Moore, Paige; Viers, Sandy; Cheyne, Ken, 2019, USA[59]	To increase patient experience care provider scores and assess how improvement efforts impact practice expansion, patient complaint rates, and staff engagement.	Prospective interventional study	5 years	General	Multiclinic children's hospital (tertiary care, ambulatory practices)	NR	11705	Press-Ganey Satisfaction Survey	Proxy	Paper and electronic	Fair
60	Fustino, Nicholas J; Kochanski, Justin J, 2015, USA[60]	To improve patient satisfaction with care in an outpatient clinic.	Quality improvement initiative	2 years	Hematology-oncology disorders	Outpatient clinic	<18	207	Press-Ganey Satisfaction Survey	Proxy	Paper	Fair
61	Davis-Dao, Carol A; Ehwerhemuepha, Louis; Chamberlin, Joshua D; Feaster, William; Khoury, Antoine E; Fortier, Michelle A; Kain, Zeev N, 2020, USA[61]	To compile a list of factors that predict patient satisfaction in a pediatric urology setting.	Cross-sectional	2 years	Urologic disorders	Urology outpatient clinic	NR	3232	NRC Health Patient Survey	Proxy	Telephone and electronic	Good

Supplement 1 - *(continued)*

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62	Fustino, Nicholas J; Wohlfeil, Margo; Smith, Hayden L, 2018, USA[62]	To determine key drivers of high patient satisfaction and factors which increase the probability of the patient recommending the practice to others.	Cross-sectional descriptive survey	2 years 5 months	Hematology-oncology disorder	Hematology-oncology unit	<21	281	Consumer Assessment of Healthcare Providers and Systems Clinician & Group Survey (CG-CAHPS)	NR	Electronic	Fair
63	Furness, Caroline L; Smith, Lesley; Morris, Eva; Brocklehurst, Caroline; Daly, Sasha; Hough, Rachael E, 2017, UK[63]	To identify areas of improvement in teen and young adult cancer care and to see if there has been a stepwise increase in patient satisfaction over time. Research also assessed whether care in a principle treatment centre affects patient experience.	Retrospective cohort	4 years	Cancer	Varied	13-24	1367	The national cancer patient experience survey	Patient	Paper	Good
64	Gray, J E; Safran, C; Davis, R B; Pompilio-Weitzner, G; Stewart, J E; Zaccagnini, L; Pursley, D, 2000, USA[64]	To evaluate the efficacy of an Internet-based telemedicine program in the NICU through assessing parental quality assessments of their care.	Randomized control trial	Nov 1997-April 1999	General (NICU - low birth weight)	NICU	Neonates	31	The Picker Institute's Neonatal Intensive Care Unit FamilySatisfaction survey	Proxy	NR	Good

Supplement 1 - *(continued)*

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65	Norman, Shelley Marie; Ford, Tamsin; Henley, William; Goodman, Robert, 2016, UK[65]	To examine the associations between patient-reported outcome measures, practitioner-reported outcome measures, and PREMs.	Cohort study	1 year	Mental health	CAMHS facilities	<16	189	Experiences of Services Questionnaire	Proxy	NR	Good
66	Madan, Alok; Sharp, Carla; Newlin, Elizabeth; Vanwoerden, Salome; Fowler, J Christopher, 2016, USA[66]	To assess patient and parental satisfaction with inpatient psychiatric care and to examine the association between satisfaction of patient outcomes.	Cohort study	1 year 9 months	Psychiatric disorders	Inpatient care unit	Adolescents	129	McLean Hospital's Perception of Care survey	Both the patient and proxy	NR	Good
67	Mah, Jean K; Tough, Suzanne; Fung, Thomas; Douglas-England, Kathleen; Verhoef, Marja, 2006, Canada[67]	To collect patient responses to a PREM covering patient satisfaction and FCC, and to see how the responses related to the patients' HRQoL. Additionally, patient responses were compared to their parents to identify possible discrepancies.	Cross-sectional	4 months	Neurologic diseases	Ambulatory clinic	12-18	104	MPOC-20 ; Give Youth a Voice survey (GYV)	MPOC-20 - proxy; GYV - patient	NR	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
68	Hummel, Kevin; Presson, Angela P; Millar, Morgan M; Larsen, Gitte; Kadish, Howard; Olson, Lenora M, 2020, USA[68]	To determine key factors for family satisfaction in the PICU and to formulate the best approach for survey dissemination.	Mixed methods	7 months	General	PICU	<18	206	Pediatric Family Satisfaction-ICU (pFS-ICU)	Proxy	Electronic	Good
69	Toomey, Sara L; Elliott, Marc N; Zaslavsky, Alan M; Quinn, Jessica; Klein, David J; Wagner, Stephanie; Thomson, Cassandra; Wu, Melody; Onorato, Sarah; Schuster, Mark A, 2019, USA[69]	To assess whether the administration of discharge surveys with tablets increases patient responsiveness and representativeness.	Quasi-experimental	4 months	General	Tertiary inpatient care unit	<18	1184	Child HCAHPS	Proxy	Paper and electronic	Good
70	Uhl, Tammy; Fisher, Kimberley; Docherty, Sharron L; Brandon, Debra H, 2013, USA[70]	To improve PCC and FCC through the analysis of parent-completed PREMs.	Mixed methods	5 months	Chronic diseases	Tertiary inpatient care unit	<18	134	The Children's Hospital Boston Inpatient Experience Survey	Proxy	Paper and electronic	Fair

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
71	Viner RM, 2007, UK[71]	To evaluate the effects of creating an adolescent-specific ward on perceived care quality, relative to treating these patients in child or adult settings.	Secondary data analysis	NR	General	Varied	<17	59815	National English Young Patient Survey 2004 (YPS)	Both the patient and proxy	Paper	Good
72	Christensen, Anna L; Brown, Jonathan D; Wissow, Lawrence S; Cook, Benjamin, 2016, USA[72]	To assess parental satisfaction with the PCC and FCC demonstrated by medical assistants, and to compare these survey results with other elements of their care.	Secondary data analysis	NR	General	Primary clinical care	18months -16	360	Consultation and Relational Empathy measure (CARE)	Proxy	NR	Good
73	Barsoom R.R.; Maugans T.A.; Burrows J.F.; Rosen P., 2017, USA[73]	To perform clinical quality improvements through the dissemination of PREMs after medical visits.	Cross-sectional	3 years	Neurologic diseases	Outpatient care	NR	458	Press Ganey Satisfaction Survey	Proxy	NR	Good
74	Barber AJ; Tischler VA; Healy E, 2006, USA[74]	To evaluate the likeness of CAMHS PREMs that were completed by either themselves or their carer.	Cross-sectional	NR	Mental health	Outpatient care	≥4	118	Experience of Service Questionnaire (ESQ)	Both the patient and the proxy	Paper	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
75	Ahmed, Sarah; Miller, Jonathan; Burrows, James F; Bertha, Ben Khallouq; Rosen, Paul, 2017, USA[75]	To determine the primary factors that are predictive of positive patient experience.	Retrospective	3 years	Dermatological diseases	Tertiary outpatient care	NR	516	Press Ganey Satisfaction Survey	Proxy	Paper and electronic	Good
76	Chen, Alex Y; Elliott, Marc N; Spritzer, Karen L; Brown, Julie A; Skootsky, Samuel A; Rowley, Cliff; Hays, Ron D, 2012, USA[76]	To assess and contrast the experiences of pediatric and adult patients.	Observational study	2 years	General	Primary clinical care	NR	668 parents/ 7823 adults	Consumer Assessment of Healthcare Providers and Systems Clinician & Group 1.0 (CG-CAHPS 1.0)	Proxy	Paper and electronic	Good
77	Burnet, Deborah; Gunter, Kathryn E; Nocon, Robert S; Gao, Yue; Jin, Janel; Fairchild, Paige; Chin, Marshall H, 2014, USA[77]	To evaluate the effects of increasing PCC characteristics on patient perceptions of their quality of care.	Cross-sectional	1 year 6 months	General	Primary clinical care	1-17	440	Consumer Assessment of Healthcare Providers and Systems Clinician & Group (CAHPS-CG)	Proxy	Paper	Good
78	Brousseau, David C; Mukonje, Terence; Brandow, Amanda M; Nimmer, Mark; Panepinto, Julie A, 2009, USA[78]	To compare parental dissatisfaction with sickle cell care to the dissatisfaction of parents receiving asthma or general pediatric care.	Cross-sectional	3 years	Sickle cell diseases, asthma, general pediatrics	Inpatient care unit	2-18	639	Picker Inpatient Survey	Proxy	NR	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Admini-stration method	Quality score
79	Bumpers, Bernadette; Dearmon, Valorie; Dycus, Paula, 2019, USA[79]	To examine whether the implementation of a "communication bundle" improves parental experiences with patient-nurse communication.	Quality improvement	15 weeks	Orthopedic and hematologic diseases	Tertiary inpatient care unit	NR	NR	Child HCAHPS	Proxy	Telephone	Fair
80	Boss, Emily F; Thompson, Richard E, 2012, USA[80]	To assess the satisfaction ratings of otolaryngic outpatients and whether these ratings differ between age groups.	Cross-sectional	1 year	Otolaryngo-logic diseases	Outpatient care	<17	5996	Press Ganey Medical Practice surveys	NR	NR	Good
81	Boss, Emily F; Thompson, Richard E, 2013, USA[81]	To examine how a teaching versus non-teaching setting influences parental satisfaction with otolaryngic care.	Cross-sectional	1 year	Otolaryngo-logic diseases	Outpatient care	<17	4704	Press Ganey Medical Practice surveys	Proxy	NR	Good

Supplement 1 - *(continued)*

No.	Author(s), year, country of study	Study objective	Study design	Study duration	Disease group	Study setting	Patients' age group (yrs)	Number of participants completing PREMs	PREM used in the study	Person completing the survey	Administration method	Quality score
82	Bal, Chandan; AlNajjar, Mohammad; Thull-Freedman, Jennifer; Pols, Erin; McFetridge, Ashley; Stang, Antonia S, 2020, Canada[82]	To collect information on patient experiences in an emergency department and to assess how care perceptions may differ between a child and their parent(s). Research also investigated whether extrinsic factors may be impacting survey responses.	Cross-sectional	2 months	Emergency department	Tertiary clinical care	<17	346	The patient-reported experience measure (PREM) for children in urgent and emergency care.	Both the patient and proxy	Paper and electronic	Good
83	Allam, Shalini D; Mehta, Mary; Ben Khallouq, Bertha; Burrows, James F; Rosen, Paul, 2017, USA[83]	To identify components of patient care that are indicative of high patient satisfaction in cardiology settings.	Retrospective cohort	2 years 2 months	Cardio-vascular diseases	Outpatient care	NR	2468	Press Ganey Physician Specialties Survey	Proxy	Paper and electronic	Good

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Supplement 2 - Pediatric Patient-reported Experience Measures identified through grey literature

Country	Sources	Used PREMs
Australia	*Pediatric hospitals* • Monash Children's Hospital • Perth Children's Hospital • Queensland Children's Hospital • Royal Children's Hospital Melbourne • Sydney Children's Hospital • Women's and Children's Hospital	• MySay Healthcare Survey A statewide online survey for parents of public hospital patients (inpatient care or day-procedure).
	Health institute • Australian Government Department of Health	
Austria	*Health institute* Federal Ministry: Social Affairs, Health, Care and Consumer Practice	No information
Canada	*Pediatric hospitals* • Alberta Children's Hospital • British Colombia Children's Hospital • Children's Hospital of Eastern Ontario • Children's Hospital of London Health Sciences Centre • CHU Sainte-Justine • Holland Bloorview Kids Rehabilitation Hospital • IWK Health Centre • Jim Pattison Children's Hospital • McMaster Children's Hospital • Montreal Children's Hospital • Sickkids	• The Canadian Patient Experiences Survey on Inpatient Care This bilingual PREM is based on the Hospital Consumer Assessment of Healthcare Providers and Systems (HCAHPS) survey.
	Health institutes • Alberta Health Services • Canadian Institute for Health Information • Provincial Health Services Authority	
Israel	*Pediatric hospitals* • Dana-Dwek Children's Hospital Tel Aviv Sourasky Medical Center	No information
Japan	*Pediatric Hospitals* • National Center for Child Health and Development	No information
	Health institute • Ministry of Health, Labour, Welfare	
New Zealand	*Pediatric hospitals* • Starship Hospital	No information
	Health institute • Ministry of Health	

Supplement 2 - *(continued)*

Country	Sources	Used PREMs
Sweden	*Pediatric hospitals* • Astrid Lindgren Children's Hospital • Huddinge Children's Hospital • Queen Silvia Children's Hospital • Sachsska Children's Hospital • Skane University Hospital • Uppsala University Children's Hospital	No information
	Health institute • Patient Centered Care Institute	
The Netherlands	*Pediatric hospitals* • Beatrix Children's Hospital • Emma Children's Hospital • Juliana's Children's Hospital • Princess Maxima Center for oncology • Sophia Children's Hospital • VU Medical Center • University Hospital Maastricht • Wilhelmina Children's Hospital • Willem-Alexander Children's Hospital	• Patient Ervaringsmeting (PEM) This PREM is based on the Picker Institute surveys and is available for inpatient and outpatient visits. • Consumer Quality index (CQ-index) Different versions of the CQ-index are available for different disease groups. • BeterMeter This survey is available for inpatient and outpatient visits.
	Health institute • Ministry of Health, Welfare and Sport	
United Kingdom	*Pediatric hospitals* • Alder Hey Children's Hospital • Birmingham Children's Hospital • Bristol Royal Hospital for Children • Derbyshire Children's Hospital • Great Ormond Street Hospital • Evelina London Children's Hospital • Noah's Ark Children's Hospital for Wales • Royal Aberdeen Children's Hospital • Royal Alexandra Children's Hospital • Royal Hospital for Sick Children • Royal Manchester Children's Hospital • Saint Mary's Hospital • Sheffield Children's Hospital • Southampton Children's Hospital • Tayside Children's Hospital • West Glasgow Ambulatory Care Hospital	• National Patient Surveys o Care Quality Commission Assessment All social care and healthcare have to be registered with the Care Quality Commission which assess hospitals on standards for patient care. • Healthcare Environment Inspectorate This questionnaire is distributed to patients, visitors and carers during inspection of the cleanliness of the hospital. • The under 16 Cancer Patient Experience Survey This survey is developed in partnership with the Picker Institute Europe and address children and young people's cancer experiences across England.
	Health institute • United Kingdom National Health Service (NHS)	

Supplement 2 - *(continued)*

Country	Sources	Used PREMs
United States of America	*Pediatric hospitals* • Akron Children's Hospital • Ann and Robert H. Lurie Children's Hospital • Arkansas Children's Hospital • Boston Children's Hospital • Bristol Myers Squibb Children's Hospital • Children's Hospital Colorado • Children's Hospital Los Angeles • Children's Hospital of Alabama • Children's Hospital of Illionois • Children's Hospital of Philadelphia • Children's Hospital of Savannah at Memorial Health Medical Center • Children's Mercy • Children's National Medical Center • Cincinnati Children's Hospital Medical Center • Cleveland Clinic Children's Hospital Rehabilitation Center • Cohen Children's Medical Center • Connecticut Children's Medical Center • Cook Children's Medical Center • Inova Children's Hospital • Intermountain Primary Children's Hospital • John Hopkins Children's Center • Joseph M. Sanzari Children's Hospital • Lucile Packard Children's Hospital • Mattel Children's Hospital UCLA • Miller Children's & Women's Hospital Long Beach • Monroe Carell Jr. Children's Hospital • Morgan Stanley Children's Hospital of New York-Presbyterian • Nationwide Children's Hospital • Nicklaus Children's Hospital • Peyton Manning Children's Hospital • Phoenix Children's Hospital • Rady Children's Hospital • Riley Children's Health • Seattle Children's Hospital • St. Jude Children's Research Hospital • St. Louis Children's Hospital • Texas Children's Hospital • The university of Chicago Comer Children's Hospital • University of South Alabama Children's & Women's Hospital • UCSF Benioff Children's Hospital • UPMC Children's Hospital of Pitssburg • Valley Children's Healthcare	• Child HCAHPS survey Survey for parents/guardians about their experiences with pediatric inpatient care. • Clinician and Group Consumer Assessment of Healthcare Providers and Systems (CG-CAHPS) survey This survey is developed by the Agency of Healthcare Research and Quality and measures patient experiences with a range of health services. • Press Ganey Satisfaction Survey Hospitals are working together with Press Ganey Associates, Inc., to measure patient experiences. • National Research Corporation (NRC) Survey Hospitals partnered with NRC Health, a third party patient experience company, to measure all aspects of patient experience. • Picker surveys Hospitals are working together with the Picker Institute to measure patient experiences. • Pediatric Integrated Care Survey A survey to measure family experiences of care integration. This survey is used to improve the quality of care. • Adolescent Assessment of Preparation for Transition survey This survey measures the quality of pre-transition counseling from adolescent-focused to adult-focused care for adolescents (16 and 17 years) with chronic conditions.
	Health Institutes • United Stated Department of Health and Human Services • National Institute of Health • Agency for Health Research and Quality	

9

Discussion

The ultimate goal of this thesis was to study and enhance pediatric patient-centered care (PCC). Therefore, this thesis focused on scientific approaches and development of tools to provide PCC. Specifically, three aspects of PCC were studied; pediatric patient engagement (part 1), Patient Reported Outcome Measures (part 2), and Patient Reported Experience Measures (part 3). This chapter provides an overview of the main findings and includes a reflection on the main findings, the clinical implications, methodological considerations, and directions for future research.

Main findings

Table 1 summarizes the main findings presented in this thesis.

Part 1: Pediatric patient engagement

To help clinicians with the engagement of pediatric patients in clinical care, policy, and research, the first part of this thesis provides information and tools to engage children meaningfully. **Chapter 2** gave an overview of the available literature about pediatric patient engagement in the past decades. This review showed that there is a growing attention for pediatric patient engagement in recent decades. Pediatric patients, especially in the age range 9- 17 years, are increasingly being asked for their opinion in health care (projects). The individual interview is the most commonly used method to engage pediatric patients, followed by focus groups (for older children) and draw & write/tell techniques (for younger children). Although pediatric patient engagement is increasing, the scoping review also showed that children are still rarely engaged in research projects. This suggests that researchers need more tools and support to engage pediatric patients. Therefore, **Chapter 3** described the development of a patient engagement tool, called All Voices Count. Working together with adolescents, we co-developed this tool in several steps. During the first step, ten important themes for adolescents with a chronic condition regarding their illness, treatment and hospital care were identified: visiting the hospital, participating, disease & treatment, social environment, feelings, dealing with staff, acceptation, autonomy, disclosure, and chronically ill peers. Based on these themes, the tool was developed. Subsequently, the tool was evaluated multiple times with adolescents and further developed based on the received feedback. Overall, adolescents were pleased with All Voices Count as it enables them to express their opinion regarding different topics in health care more easily.

Part 2: Patient Reported Outcome Measures

The second part of this thesis focuses on the use of Patient Reported Outcome Measures (PROMs) in clinical practice. PROM implementation is a challenging process and successful implementation requires involvement of all stakeholders, definitely including patients and parents. **Chapter 4** provided insight into the experiences of clinicians on the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal. Clinicians were generally satisfied with the use of the KLIK PROM portal. It gives them a valuable tool to systematically monitor and discuss patients' functioning. However, several barriers were mentioned such as irrelevant and long PROMs, low response rate, and no integration with the electronic health record (EHR). **Chapter 5** focused on the experiences of patients and parents. Both patients and parents were satisfied with the usability and effect of the KLIK PROM portal on the conversation with the clinician. However, some points of improvement were identified; again irrelevant and long PROMs, PROMs were not discussed by the clinician, and a suboptimal lay-out of the portal.

Applying implementation frameworks, such as the Consolidated Framework for Implementation Research (CFIR), can yield valuable insights into facilitating determinants and barriers in the implementation process. Subsequently, targeted implementation strategies can be identified. **Chapter 6** described the most prominent determinants for successful PROM implementation of the KLIK PROM portal; intervention characteristics, characteristics of individuals, and process of implementation. In addition, barriers were identified; inner setting and intervention characteristics. The strategy 'identify and prepare champions' was acknowledged as the most effective strategy in addressing the combination of identified barriers.

To overcome the barrier of burdensomeness of completing PROMs (i.e., long completion time and irrelevant questions), Patient-Reported Outcomes Measurement Information System (PROMIS®) Computerized Adaptive Testing (CAT) can be used. The feasibility, psychometric properties and outcomes of eight PROMIS pediatric measures for boys with hemophilia were assessed in **Chapter 7**. Results showed that almost all PROMIS pediatric measures were feasible and reliable for use in clinical hemophilia care. The PROMIS pediatric measures are therefore a valid alternative to the well-known legacy instruments, with lower floor- and ceiling effects. Although the number of completed items was higher for the PROMIS pediatric measures on domain level, the number of completed items in the selected set of PROMIS pediatric measures was lower compared to the legacy instruments, resulting in a lower burden of completing PROMs.

Part 3: Patient Reported Experience Measures

The third part of this thesis focuses on Patient Reported Experience Measures (PREMs). **Chapter 8** provided an overview of PREMs currently available for use in pediatrics. A total of 39 different PREMs were identified in the literature, developed in seven countries. Most identified PREMs were generic (87%), which aimed to measure experiences of health care, regardless of disease or department of care. More than half of the PREMs (59%) were designed to be completed by proxy. The number of items included in the different PREMs ranged from 7 to 89, and the number of domains ranged from 3 to 10. Communication (e.g., communication with parent, communication with child, communication about treatment, provider's communication skills) was the most prominent domain across the different PREMs. The various PREMs developed by Press Ganey (1) were most commonly used, followed by the Measure of Processes of Care (MPOC) (2) and Child HCAHPS (3).

Table 1. Overview of the main findings of this thesis

Part 1. Pediatric patient engagement

Chapter	Short title	Aim	Participants	Methods	Main findings & conclusion
2	Scoping review pediatric patient engagement	To provide an overview of the literature about pediatric patient engagement in clinical care, research and intervention development.	Not applicable	The methodological framework of Arksey & O'Malley was used: 1. Identifying the research question 2. Identifying relevant studies 3. Study selection 4. Charting the data 5. Collating, summarizing, and reporting the results	• There is an increase in the number of studies that include pediatric patient engagement in the past decades. • Most studies involving pediatric patient engagement were performed in the United States of America. • When looking at patient populations, a mix of patients from different disease groups were mostly asked for their opinion, followed by oncology patients. • The majority of studies focused on the engagement of pediatric patients in clinical care. • The individual interview is the most commonly used method to engage pediatric patients. • Pediatric patients in the age range 9-17 years were most often engaged. There is an increasing interest in pediatric patient engagement. However, lack of uniformity about the definition of pediatric patient engagement and clear information/support for clinicians hinders engagement.
3	Development of a patient engagement tool; All Voices Count	To develop a patient engagement game for adolescents (12-18y) with a chronic condition that can be used by clinicians/researchers to incorporate what matters to pediatric patients in clinical care, research and policy.	Adolescents with a chronic condition under treatment at the Emma Children's Hospital Amsterdam UMC	An uses-centered design was used: 1. Identification of important themes for adolescents regarding their illness and treatment *Focus groups with adolescents (N=15)* 2. Evaluation of the draft version of the game *Focus groups with adolescents (N=13)* 3. Testing usability in clinical practice *Pilot workshop with adolescents (N=4)*	• Ten major themes for adolescents regarding their illness, treatment and hospital care were identified: visiting the hospital, participating, disease & treatment, social environment, feelings, dealing with staff, acceptation, autonomy, disclosure, and chronically ill peers. • Development of a pediatric patient engagement game called All Voices Count. This game provides clinicians with a tool to include pediatric patients meaningfully in hospital care, research and policy.

Table 1. *(continued)*

Part 2. Patient Reported Outcome Measures

Chapter	Short title	Aim	Participants	Methods	Main findings & conclusion
4	Clinicians' perspective on PROM implementation	To gain insight into clinicians' perspective on the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal.	Clinicians (i.e., doctors, nurses, and psychologists) using the KLIK PROM portal in 14 hospitals in the Netherlands	Clinicians (N=148) completed an online evaluation questionnaire, regarding (1) overall satisfaction, (2) feeling competent to discuss PROMs, (3), use of KLIK during consultation, (4) influence of KLIK on the consultation, (5) usability of the KLIK PROM portal, (6) satisfaction with PROMs and feedback, and (7) support of the KLIK expert team.	1. Clinicians are generally satisfied with the KLIK PROM portal. 2. The majority of clinicians (85.8%) feels competent to discuss the KLIK dashboard. 3. 70.3% of the clinicians (almost) always discusses the KLIK dashboard. 4. KLIK improves the consultation according to 70.3% of the clinicians. 5. KLIK is easy to use according to 71.6% of the clinicians. 6. The majority of clinicians (80.4%) is satisfied with the feedback in the KLIK dashboard. 7. Clinicians (71.6%) experience enough support from the KLIK expert team. The KLIK PROM portal is a valuable tool for clinicians to systematically monitor patients' functioning. However, some barriers (i.e., irrelevant and long PROMs, low response rate, no integration with EHR) were also identified.
5	Patient's and parents' perspective on PROM implementation	To gain insight into patient's and parents' perspective on the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal.	Patients (12-19y) and parents (of children 0-19y) using the KLIK PROM portal in the Emma Children's Hospital Amsterdam UMC	A mixed-method design was used: 1. Focus groups with patients (N=8) and parents (N=17) 2. Online evaluation questionnaire regarding (1) overall satisfaction, (2) completion of PROMs in the KLIK PROM portal, (3) discussing PROMs with the clinicians, (4) influence of KLIK on the (preparation of) the consultation, (5) usability of the KLIK PROM portal, and (6) content of PROMs was completed by patients (N=31) and parents (N=130)	*Focus groups:* Patients and parents are generally satisfied with the use of the KLIK PROM portal. Patients mentioned that KLIK has an attractive lay-out and parents valued that KLIK provides insight into their child's functioning. *Evaluation questionnaire:* 1. Patients and parents reported an overall satisfaction of respectively 7.9/10 and 7.3/10. 2. Almost all patients (90%) and parents (95%) (almost) always complete PROMs. 3. The KLIK dashboard is (almost) always discussed by the clinician according to 52% of the patients and 45% of the parents.

Table 1. *(continued)*

					4. KLIK is of added value for the conversation with the clinician according to 58% of the patients and 59% of the parents. 5. KLIK is easy to use according to 81% of the patients and 74% of the parents 6. The majority of the patients (80%) and parents (74%) are satisfied with the PROMs they complete. Pediatric patients and parents were satisfied with the usability and effect of the KLIK PROM portal in clinical care. However, some points of improvements (i.e., irrelevant and long PROMs, no discussion of PROMs by clinician, suboptimal lay-out website) were also identified.
6	Assessment of the KLIK PROM portal implementation	(a) To retrospectively describe the most prominent determinants of successful PROM implementation. (b)To identify current barriers of the KLIK PROM portal implementation.	Not applicable	A mixed-method design was used: (a) The CFIR framework was used to retrospectively describe the implementation process of the KLIK PROM portal and to identify determinants in this process. (b)The CFIR-ERIC Implementation Strategy Matching Tool was used to identify current barriers in the KLIK implementation and to match implementation strategies that address these barriers.	(a) The strength of the KLIK PROM portal lies particularly in the following CFIR domains: intervention characteristics (e.g., easy to use, direct feedback), characteristics of individuals (e.g., motivated clinicians), and process of implementation (e.g., support KLIK expert team). (b)Barriers in the implementation lie mainly in the domain inner setting (e.g., involving/motivating all stakeholders) and intervention characteristics (e.g., complex to use for some clinicians, costs). The strategy 'identify and prepare champions' was identified as the most effective strategy in addressing the combination of identified barriers. CFIR provides clinicians and scientists guidance during a health care implementation process and can be used in all phases of implementation, although it is a quite extensive and complex framework with some overlapping constructs.

Table 1. *(continued)*

7	Psychometrics of the pediatric PROMIS measures in hemophilia	To assess the feasibility, measurement properties, and outcomes of eight PROMIS pediatric measures for boys with hemophilia.	Boys (8-17y) treated for mild to severe hemophilia A or B in one of the Hemophilia Treatment Centers in the Netherlands	Participants (N=77) were asked to complete the following measures online: • Socio-demographic questionnaire • PROMIS pediatric measures: v2.0 Pain Interference, v2.0 Fatigue, v2.0 Anxiety, v2.0 Depressive Symptoms, v2.0 Mobility, v2.0 Peer Relationships, v2.0 Anger 9a scale, and V1.0 Global Health scale (7+2) • Legacy instruments: Haemo-QOL and PedHAL	• Floor and ceiling effects of the PROMIS pediatric measures were substantially less compared to the legacy instruments. • The number of completed items in the selected set of PROMIS pediatric measures was lower compared to the legacy instruments. • The reliability of the PROMIS pediatric measures was good, except for the PROMIS CAT Mobility. • HRQoL of boys with hemophilia is comparable to the Dutch general population, except for the domains Pain Interference and Mobility (more complaints). The PROMIS pediatric measures are valid alternatives to the well-known legacy instruments.

Part 3. Patient Reported Experience Measures

Chapter	Short title	Aim	Participants	Methods	Main findings & conclusion
8	Systematic review PREMs	To identify and synthesize evidence on the use of PREMs in pediatric health care settings and their characteristics.	Not applicable	PRISMA guidelines were followed during the conduct and reporting of this review: • A search strategy was applied to MEDLINE, EMBASE, PsychINFO, Cochrane Library, and CINAHL to identify relevant articles. • Gray literature was searched to capture real-word implementation of PREMs. • Articles were screened by two reviewers • Data was extracted, synthesized, and tabulated. • Risk of bias was assessed for all identified studies.	• 83 studies reported on the use of PREMs in pediatrics. • Most included studies were conducted in the United States of America (48 studies). • 39 different PREMs were identified in the included studies and were used in different healthcare settings. • 10 additional PREMs were identified in gray literature. • Majority of the PREMs were generic (34/39 PREMs) • Most PREMs were designed to be completed by proxy (23/39 PREMs). • The number of items/domains in the identified PREMs ranged from 7 to 89 resp. 3 to 10. • Communication was the most prominent domain. This review provides an overview of the current PREMs available for use in pediatrics. These findings can guide clinicians and researchers to use appropriate PREMS to implement patient-centered care in pediatrics.

Reflection on the findings & clinical implications

Part 1: Pediatric patient engagement

Despite the increase in pediatric patient engagement in recent decades, there are still challenges that hinder the involvement of children in clinical care, policy, and research. Literature suggests that lack of uniformity about the definition of pediatric patient engagement, limited information about ways in which children can be engaged, insufficient resources for pediatric patient engagement, difficulties adapting to the shifting dynamics of control of decision-making, and little support for clinicians to engage pediatric patients meaningfully hinders engagement (4-7). These challenges, especially the struggles with the changing degree of influence and control, can even lead to tokenistic engagement (i.e., symbolic or perfunctory form of patient engagement, in which pediatric patients have no influence on the decision-making process (8, 9)). The reduction of these challenges is not easy and requires more uniformity in pediatric patient engagement, as well as more support for clinicians in the form of guidelines, shared lessons, and support tools. In this thesis, we have taken a first step towards this goal:

1. Shared lessons

   The literature overview in **Chapter 2** can inform clinicians, policy makers and researchers about the different ways in which patient engagement can be shaped. It gives them information regarding the methods used in earlier studies and goals for patient engagement. This knowledge can help clinicians to feel more confident and can guide them to engage pediatric patients meaningfully and usefully in their practice.
2. Support tools

   In **Chapter 3** the development of the engagement tool All Voices Count was described. Since the development, All Voices Count was used (or will be used in the near future) to include the opinion of adolescents in several projects in our hospital. Table 2 provides an overview of these projects and related questions for pediatric patients. In addition, the game has been translated to English to extend the scope.

Table 2. Overview of projects in which All Voices Count has been used

Area of engagement	Purpose	Question
Clinical care	Bringing physiotherapeutic treatment more in line with the wishes of pediatric oncology patients.	How can we make exercising more fun for you during treatment?
Clinical care / policy	Development of a follow-up program for children who have had surgery.	How should the follow-up program look like and which themes should be discussed by the clinician?
Clinical care / policy	Various questions that are submitted to the children's advisory board	-
Clinical care / research	Investigate what problems girls with a coagulation disease and carriers encounter and how can we best help them.	What does it mean for you, as a girl, to have a coagulation disease and how can we improve clinical care?
Clinical care / research	Studying the burden of disease in children with kidney disease	How does a chronic kidney disease affect your daily live?
Research	Development of an International Core Outcome Set for acute simple appendicitis in children (10).	What do you think is important to know to make an informed choice between two treatments for appendicitis?
Research	Investigate important Patient Reported Outcomes (PROs) for children treated by a pediatric physiotherapist.	How does your complaint (for which you are being treated by the physiotherapist) affect your daily life?

Part 2: Patient Reported Outcome Measures

KLIK PROM portal

The KLIK PROM portal was evaluated with all stakeholders (patients, parents, and clinicians) in this thesis (**Chapter 4** and **Chapter 5**). In addition, we retrospectively assessed the KLIK PROM portal with the KLIK expertise team (**Chapter 6**). Table 3 summarizes the identified facilitators and barriers in the implementation of the KLIK PROM portal in these studies. We observed a significant overlap in the facilitating factors mentioned by the various stakeholders. The same facilitators were also reported in previous studies focusing on the use of PROMs in the adult population and a recent review in pediatric care and can ultimately contribute to PCC (11-17).

Overlap is also observed in the mentioned barriers by the stakeholders. However, contradictions are visible as well. For example, clinicians reported that a low response rate of completing PROMs by patients and parents was a major barrier, while patients and parents reported that a low discussion rate by the clinicians was an important barrier. These findings may be interrelated, as patients and parents indicated that they see no added value in using PROMs if the answers are not discussed by the clinician, which potentially can result in a lower response rate. The mentioned barriers were consistent with other international studies focusing on the identification of barriers for using PROMs mainly performed in the adult care setting (18-25). In a paper series of case studies regarding the use of implementation science for PROM/PREM implementation, which includes **Chapter 6**, Stover et al. (21) indicated that barriers for PROM implementation were consistent across populations and settings, while facilitators were more context-specific. Implementation strategies can help to overcome the identified barriers. The paper series showed that identified implementation strategies in the different studies almost all match the 'Expert Recommendations for Implementing Change' (ERIC) (26), suggesting the value of these strategies independent of setting (21).

During the retrospective assessment of the KLIK PROM portal (**Chapter 6**), we used the CFIR-ERIC matching tool to identify implementation strategies (27). The strategy 'identify and prepare champions' was identified as the most effective strategy to address the mentioned barriers in the KLIK PROM portal. This means that we should identify an individual of the health care team that supports the implementation and can help to overcome indifference or resistance that implementation of the KLIK PROM portal can provoke at an organizational level (26). This strategy was not innovative for the KLIK PROM implementation, as we already were aware of the helping role of the champion. The KLIK implementation always starts at request of a health care team and therefore a large part of the team is motivated to implement PROMs. We noticed that it is particularly helpful if the opinion of one of the motivated team members is highly valued by their colleagues, as this team member can function as a champion and can motivate their colleagues to change (28). In our opinion, a bottom-up implementation as opposed to a top-down approach, which is regularly seen in PROM implementation initiatives, is valuable. However, (financial) support from management and the board of the hospital is necessary to enhance the sustainability of change.

We are currently working on the identified ERIC strategies to improve the KLIK PROM implementation. For example, we assess key stakeholders for readiness (i.e., we conduct evaluation meetings with the multidisciplinary teams and pay attention to the more reluctant team members to better understand their underlying resistance), inform local opinion leaders (i.e., we talk about the added value of using PROMs at conferences and symposia. This information is picked up by opinion leaders who talk about it with their colleagues), access new funding, and tailoring strategies (i.e., we tailor implementation strategies to address identified barriers). In addition, we added information about the evidence of implementing PROMs in the KLIK training for clinicians, create awareness within the board of hospitals to facilitate the funding and practical support of PROM implementation, and try to engage all stakeholders in the implementation process. For example, we should involve patients and patient associations more in selecting PRO's and PROM's.

Optimization KLIK PROM portal
The received feedback of all stakeholders (clinicians, patients and parents) was used to optimize the KLIK PROM portal. After the evaluation studies (**Chapter 4, Chapter 5**, and **Chapter 6**), the following changes were made to KLIK PROM portal:

- Front-end integration between the KLIK PROM portal and the EHR
  In 2019, a front-end integration between the EHRs EPIC© and HiX© was realized in the Amsterdam UMC, University Medical Center Utrecht, and Princess Maxima Center. With this integration, clinicians can view the KLIK dashboard in the EHR, which facilitates the ease of use of the KLIK PROM portal in the consultation room. However, to be able to automatically link hospital appointments to KLIK, data integration is necessary. This automation process would be valuable for the implementation of PROMs, because it reduces the administrative tasks for health care teams, which can boost the sustainable use of PROMs in clinical care. We hope to realize a full data integration with EHRs in the future.
- Mobile version of the KLIK PROM portal
  Patients and parents expressed that an app would be a valuable addition to the KLIK PROM portal. Therefore, a mobile version of the KLIK PROM portal was developed. This makes it easier for patients and parents to complete PROMs on their smartphone or tablet.
- Modernize the lay-out of the KLIK portal
  Children indicated that the lay-out of the KLIK portal was a bit old-fashioned. Therefore, we upgraded the lay-out of the KLIK portal by using visuals and creating a more professional look. In addition, we added specific information pages for all users (pediatric patients, adult patients, parents, and clinicians).
- Adding educational video's
  During the evaluation studies, it was recognized that a large part of the patients and parents were reluctant to initiate the communication about PROMs. To empower patients and parents, educational videos and a topic list were developed and made available on the KLIK website (29).These tools give patients and parents information on how to start the conversation with the clinician about topics that are important to them.

Table 3. A summary of facilitators and barriers in KLIK PROM implementation identified in this thesis using the CFIR framework

Domain	**Facilitators**			**Barriers**		
	Chapter 4	**Chapter 5**	**Chapter 6**	**Chapter 4**	**Chapter 5**	**Chapter 6**
Intervention characteristics	- Easy to use	- Easy to use	- Acceptable, valuable and feasible		- Not easy to use	
	- Improves communication	- Improves communication				
	- Gives insight into patient's functioning / earlier detection of problems	- Gives insight into functioning (of child)				
	- More efficient consultation (time-saving)	- More efficient consultation (time-saving)				
	- Helps in preparing for consultation	- Helps in preparing for consultation				
	- KLIK dashboard		- KLIK dashboard			
	- Attractive lay-out	- Attractive lay-out	- Attractive lay-out		- Out-dated lay-out	
	- KLIK training (knowledge, tools, skills)		- KLIK training (knowledge, tools, skills)			
				- No integration EHR		
		- Security KLIK PROM portal				
					- No app	
			- Flexibility (settings, languages)			- No possibility of selecting PROMs per individual

Table 3. *(continued)*

Domain	**Facilitators**			**Barriers**		
	Chapter 4	**Chapter 5**	**Chapter 6**	**Chapter 4**	**Chapter 5**	**Chapter 6**
Outer setting				- Content of PROMs (irrelevant and repetitive questions)	- Content of PROMs (irrelevant and repetitive questions)	
			- Increased motivation for PROM use in society			
			- Collaborations and networks			
			- Support hospital board			- No support hospital board
Inner setting				- Time-consuming	- Time-consuming	- Time-consuming
				- Low response-rate	- Low discussion-rate	
			- Bottom-up implementation			
						- Financial resources
Characteristics of individuals				- Insecure about skills to interpret and communicate about PROMs		
				- Patients with low health literacy skills / non-native speakers cannot complete PROMs		
			- Motivated clinicians		- Not seeing the added value and goal	- Not seeing the added value and goal

Table 3. *(continued)*

Domain	**Facilitators**			**Barriers**		
	Chapter 4	**Chapter 5**	**Chapter 6**	**Chapter 4**	**Chapter 5**	**Chapter 6**
Implementation process	- Support KLIK expert team			- Support KLIK expert team		
			- Adapted to the workflow	- Lack of clarity regarding the workflow		
			- License agreements			

Implementation strategies (Chapter 6)

1. Identify and prepare champions
2. Assess for readiness and identify barriers and facilitators
3. Inform local opinion leaders
4. Access new funding
5. Tailor strategies

PROMIS

The PROMIS pediatric measures, especially the Computerized Adaptive Tests (CAT), are promising in reducing the burden of completing PROMs. In **Chapter 7**, we assessed the psychometric properties of the PROMIS pediatric measures, were it was shown that these measures were reliable and feasible for use in clinical practice. The validity and reliability of the PROMIS pediatric measures were comparable to the validation studies of the Dutch-Flemish PROMIS group (30, 31). However, a known problem of the PROMIS pediatric measures is that the number of PROMIS items administered is still relatively high. Especially, patients with no problems or complaints have to answer the maximum amount of items to reach the CAT stopping rule (SE ≤ 3.2 and/or a maximum of 12 items). This problem arises due to limited availability of items on the high or low end of the scale and patients with no complaints are therefore more difficult to measure reliably (32). Future research is necessary to optimize the CAT stopping rule and to further reduce the burden of completing PROMs in clinical care (32, 33).

Despite this limitation, the PROMIS pediatric measures have been implemented in the KLIK PROM portal and are used by some health care teams in clinical care and/or research (34-36). The experiences of the first groups using PROMIS in the KLIK PROM portal are mixed. The groups are positive about the idea of PROMIS; shorter PROMs that are more tailored to the individual. However, problems with the CAT stopping rules hinder successful implementation in clinical practice. In addition, studies showed that individual item feedback is essential for clinicians to start the conversation with patients, to understand the PROMIS domain scores, and to easily detect problems (37). In a CAT, not all items of an item bank are administered to patients which can lead to confusion with interpreting the feedback. In addition, domain scores are not always understood. Therefore, clinicians need more training in interpreting the results and the use of this new method of administering PROMs.

The use of the PROMIS measures is in line with a shift towards the use of generic PROMs (38). Recently, Kennisplatform Uitkomstgerichte Zorg (www.platformuitkomstgerichtezorg.nl) published an advice report for a generic PROM core set for children and adults, in which the use of the PROMIS measures is advised (39). Worldwide standardization of PROMs will facilitate the uptake as it realizes the comparisons between different patient populations and the general population (40). Therefore, Kennisplatform Uitkomstgerichte Zorg advises to use the generic

PROM core set for children (consisting of ten patient-reported outcomes relevant for all conditions), if necessary supplemented with disease-specific PROMs. This message is supported by the KLIK expert team and health care teams are being informed about the importance of generic measurement. The PROMIS measures are available to use in the KLIK PROM portal and research is being conducted to reduce the above mentioned barriers.

Part 3: Patient Reported Experience Measures

Chapter 8 provided an overview of the available pediatric PREMs and their use in clinical care. This systematic review demonstrated the adoption of PREMs in developed countries worldwide, aligning with the growing recognition of the importance of delivering PCC (41). Just like PROMs, PREMs were used in different health care settings targeting different purposes. In accordance with the literature, PREMs were most often used to evaluate factors that affected overall patient experience or assess experiences after an intervention with the ultimate goal of improving the quality of care (42, 43). In contrast to PROMs , where often digital administration tools are used (44), the majority of PREMs was administered via paper-pencil. Possible explanations for this administering mode can be found in the purpose and manner in which PREMs are processed. Where PROMs are completed before the outpatient consultation and discussed by the clinician at an individual level (44-46), PREMs are completed afterwards and are usually processed anonymously at group level for quality of care purposes.

Noteworthy, PREMs are often confused with PROMs (assess patient's health status, symptoms or well-being (25, 47, 48)) and satisfaction questionnaires (assess whether patient's expectations about the received care were met (49)), but these measurement tools have different purposes that focus on different aspects of PCC (50, 51). Especially, the distinction between PREMs and satisfaction questionnaires is often overlooked, while satisfaction questionnaires involve a degree of subjectivity that is not seen in PREMs (52, 53). The interchangeability of concepts, around both pediatric engagement and PREMs, highlights the novelty of the research field of PCC and emphasizes the need for clarification of concepts (42).

Currently, there is limited use of PREMs in the KLIK PROM portal, despite the availabilities. Within the KLIK PROM portal it is possible to administer PREMs after the outpatient consultation and to keep the answers confidential on an individual level. At this moment, PREMs are only used in the KLIK PROM portal for quality improvement within programs for preterm born children (e.g., for a parenting intervention program (ToP program), Change study) (54). Other health care teams have expressed a desire to administer PREMs in order to make care more patient-centered, but lack of time, (financial) resources, and knowledge prevents them to actually use PREMs. In addition, it should be clear to patients which answers (on PROMs and PREMs) are being fed back to the clinician, as responses on PREMs may differ when results are being discussed or not.

Methodological considerations

Some overall limitations should be taken into account when looking at the findings of this thesis.

Representativeness of the participants

Patients, parents and clinicians were included in different studies in this thesis to gain insight into their perspective regarding All Voices Count and the KLIK PROM portal. Although we tried to include representative samples, some remarks need to be made. While we invited adolescents with different chronic conditions to participate in the various phases of the development of All Voices Count, only adolescents with cancer participated during the evaluation of the pilot study. However, we showed in **Chapter 3** that adolescents, regardless of their chronic condition, showed the same problems and supportive factors. Therefore, we believe that this did not influence the results. In addition, we only invited patients and parents that were part of the KLIK panel (consisting of patients/parents in the Emma Children's Hospital that indicated that they give permission to be invited for research projects) to participate in the KLIK evaluation studies. These patients/parents might be more assertive than other KLIK users. Earlier research showed that patient who participate in (co-design) studies tend to be more self-confident, critical, and assertive in comparison to patients who do not participate, which may hinder representativeness (4, 55).

Challenging to include sufficient participants

We have encountered difficulties in including an adequate number of participants, especially in studies focused on the engagement of children and adolescents. It has been challenging to motivate them to participate in research (both for quantitative as qualitative research). We noticed that adolescents preferred to spend their time differently, which is healthy from a developmentally perspective. Other barriers for engagement included logistic difficulties related to traveling to the hospital, time constraints, and problems with scheduling meetings with a group of adolescents. These barriers were also observed in other studies in different research domains (7, 56, 57).

Directions for future research

Part 1: Pediatric patient engagement

Challenges regarding pediatric patient engagement

Lack of uniformity in defining pediatric patient engagement and tokenism are major issues regarding the engagement of children and adolescents in clinical care, policy, and research. This is further exacerbated by external pressure from funders to include pediatric patients in research. As long as the added value and impact of pediatric patient engagement are not fully recognized, and challenges as funding, representativeness, and letting go of control are not yet overcome, it remains challenging to break through these issues (4, 58). Uniformity in pediatric patient engagement, as well as more support for clinicians in the form of guidelines, shared lessons, and support tools is necessary. In this thesis, we have taken a first step towards this goal (sharing information around pediatric patient engagement and development of an engagement tool). However, to work towards a more uniform way of (reporting on) pediatric patient engagement, we recommend the development of a guideline in co-creation with all stakeholders. This guideline should include information regarding the operationalization and goal of pediatric patient engagement, setting, participants, used methods, feasibility, and shifting dynamics of control. Changing power dynamics requires a paradigmatic shift and is often seen as the main reason for resistance among clinicians (59). Information regarding the shifting dynamics of control can facilitate patient engagement.

The importance of uniformity regarding (pediatric) patient engagement is recognized by other researchers in the field. For example, a scoping review will be conducted by Oravec et al. (60) to gain a better understanding of how patients and caregivers been engaged in scoping reviews that followed the methodology of Arksey and O'Malley (61) and Levac (62), and to work towards a guideline regarding the engagement of patients in scoping reviews. Such a guideline will help researchers to involve patients as partners in conducting scoping reviews. This can help to better understand patients' health conditions and experiences.

In addition, there is limited information available regarding the used methods to engage pediatric patients. Yet, methods are chosen based on availability of patients to participate, age of the participants, and target group (4, 7). Future research should focus on increasing knowledge about the used methods in pediatric patient engagement and their suitability and impact for different research questions and age groups.

Engagement of adolescents

In our studies it was difficult to include adolescents due to motivational and logistic barriers. The development of online tools to engage pediatric patients could reduce these barriers. During the development of All Voices Count, adolescents indicated important benefits of an online tool such as not paying an extra visit to the hospital, takes less time, and an online approach is more flexible to fit in their time schedule. Therefore, we would like to develop an online version of All Voices Count in the future. In this regard, we can learn from initiatives that look at new methodologies for online focus groups for hard-to-include populations in both pediatric and adult care (63, 64). These research groups investigated the benefits (e.g., flexibility, anonymity, feeling comfortable to express their opinion, cost-effective, inclusion of different populations) of online / chat-based focus groups, which can be conducted in real time (synchronously) or not in real time (asynchronously).

Impact pediatric patient engagement

This thesis provided descriptive information about pediatric patient engagement, as this is a first step towards uniformity about pediatric patient engagement. However, both in the scoping review as during the development of All Voices Count, we did not assess the impact of pediatric patient engagement. This might be an interesting area for future research as it could give us insight into the added value of patient engagement. Studies regarding the impact of patient engagement have mainly focused on adult patient engagement or pediatric patient engagement in the medical consultation room (65, 66). A recent review showed that the impact of pediatric patient engagement in medical consultations is still minor and innovations to enhance engagement are scarce (66). To enhance pediatric patient engagement more respect for the autonomy of children and parents is necessary. Research regarding skills needed to think along at different levels during treatment is desired (i.e., everyone can participate at their own level), as is research into intervention/tools/training to increase patient engagement. Tools, like All Voices Count, can contribute.

Children's Advisory Board

Pediatric patient engagement is considered important in the Emma Children's Hospital. Therefore, the Children's Advisory Board was re-established in June 2023. Children aged ten to eighteen years treated at the Emma Children's Hospital can participate. During four to six sessions per year, the Children's Advisory Board can talk about subjects on all levels of health care (i.e., clinical care, policy, and research). Examples of discussed questions are related to nutrition, development of a follow-up program, recruitment methods in research, and reviewing new information material. Regular evaluations are needed to keep these children motivated and to ensure that engagement contributes to decision-making.

Part 2: Patient Reported Outcome Measures

Patient Reported Outcome Measures and the impact on patient-centered care

This thesis showed that PROMs are effective to use in daily clinical practice to monitor patient's functioning and to improve patient-clinician communication. However, we did not study how PROM information is used in the consultation room by clinicians, patients, and parents. For future research, it would be interesting to gain insight into the actions taken with regard to the completed PROMs and to expand knowledge about how this contributes to the enhancement of PCC. The use of video observations in the consultation room may provide this information and can be used for both quantitative as qualitative analyses with regard to the content as well as non-verbal communication. In addition, future research should focus on patient preferences for visualization of PROM feedback (i.e., graphs) and testing the accuracy of interpretation of these visualizations. Only when patients can interpret PROM feedback, it gives them insight into their own functioning and PROMs can optimally be discussed in the consultation room.

Patient Reported Outcome Measures and shared-decision making

A part of PCC that has not been addressed in this thesis is shared-decision making (fits with common element 7 of the New England Journal of Medicine (NEJM) Catalyst model of PCC (67)). Shared-decision making is a conversation model involving patients in health care decisions (68, 69). In shared-decision making patients are engaged to think about the available treatment options and work together with the clinician to select the best tests and treatments based on the available evidence and the preferences and values of the patient (69, 70). PROM data, at individual level and aggregated level, can be helpful in shared-decision making, as it provides information about patient's preferences and values and benefits and harm of options (69, 71, 72). However, more information is needed about how PROMs can be linked to shared-decision making and how clinicians can be trained to use this information (69). Recent initiatives from our research group are focused on the use of PROMs in the shared-decision making process. This research will provide information and training on how to use PROMs in the process of shared-decision making (73).

PROMIS common metric

Another advantage of the use of PROMIS (item response theory) is the standardization of the scoring system, also referred to as the common metric (74). The common metric makes it possible to compare scores of different PROMIS measures and allows for the addition of new items to the existing measures (74-76). This makes PROMIS flexible and sustainable for the future (74). In addition, the common metric can contribute to PCC as it makes it possible to adapt items to different patient populations (for example for patients with low health literacy or reduced language proficiency). More research is needed in this field with regard to the content (i.e., to examine to what extent it is necessary to modify items for different patient populations) and statistical aspects (i.e., the replacement of items).

Part 3: Patient Reported Experience Measures

An important incentive for the use of PREMs is that it provides data that can be used for quality improvement (43). However, our experience is that the use of PREMS is difficult in clinical practice. There is a lot of uncertainty about how PREM data can be used meaningfully for quality improvement. Studies showed that lack of guidance hinders the application of PREMs to improve the quality of health care services (43). Other challenges in using PREMs include lack of time and resources for both the administration of PREMs as analyzing the data, variation in the use of PREMs, and lack of skills among clinicians, policymakers, and researchers to use and interpret PREM data (43, 77). Future research should focus on how PREM data can contribute to quality improvement to provide PCC. Lessons learned can be shared and merged to a guideline for the use of PREMs. Initiatives, such as the PREM working group from Linnean (www.linnean.nl), can contribute to this purpose (78). However, more in-depth conversations and profound collaboration on this important topic is necessary to make the use of PREMs in health care a success.

Conclusion

The importance of patient-centered care is recognized, but its application is still in its infancy. The shift towards patient-centered care is difficult as it requires a shift in thinking and a redefinition of roles and decision-making for patients, parents, clinicians, policymakers and researchers. This thesis contributes towards this goal as it provides scientific information and shared lessons for clinicians and tools to empower both patients and clinicians in their new roles.

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Addendum

Summary

Patient-centered care (PCC) is characterized by the provision of care that respects and responds to individual patient preferences, needs and values, and ensures that patient's wishes guide all clinical decisions. PCC places the patient, not the disease, at the center of health care and promotes a collaboration between patient, family, and clinicians to provide care that is tailored to the patient's wishes and needs. Providing PCC has many benefits for both patient and organization, including improved individual health outcomes, increased patient satisfaction, greater job satisfaction, and a reduction of health care costs. Although the importance of PCC is recognized, implementation is difficult because it requires a different mindset and changing roles of patients, parents, and clinicians.

This thesis focused on scientific approaches and the development of tools to enhance PCC in children. Three aspects of PCC were investigated. The first part focused on pediatric patient engagement, the second part focused on Patient Reported Outcome Measures, and the third part focused on Patient Reported Experience Measures. In **Chapter 1**, the general introduction, the different parts of PCC are described and the context of the research in this thesis is illustrated.

Part 1: Pediatric patient engagement

The involvement of children in the decision-making in clinical care and research with the aim to improve health (care) is defined as pediatric patient engagement. Pediatric patient engagement is an essential aspect of providing PCC and its importance is established in the United Nations Convention on the Rights of the Child. However, clinicians and researchers struggle with the engagement of pediatric patients.

To help clinicians in engaging pediatric patients, the first part of this thesis provided information and tools to meaningfully engage children. **Chapter 2** gave an overview of the existing literature on pediatric patient engagement. This review showed that interest in pediatric patient engagement has increased over the past decades. Pediatric patients, especially those between the ages 9 to 17 years, have been involved in a wide variety of health care projects more often. Although pediatric patient engagement is increasing, this review also showed that children are still rarely engaged in research projects. This suggests that researchers need more tools and support to engage pediatric patients. Therefore, **Chapter 3** described the development of a patient engagement tool, called All Voices Count. In co-creation with adolescents (N=23), the game All Voices Count was developed in several steps. First, ten themes were identified that were important to adolescents regarding their illness, treatment, and hospital care: visiting the hospital, participating, disease & treatment, social environment, feelings, dealing with staff, acceptation, autonomy, disclosure, and chronically ill peers. Based on these themes, a first version of All Voices Count was developed. Subsequently, All Voices Count was evaluated several times with adolescents and further developed based on feedback received. Adolescents were satisfied with All Voice Count as it helps them to give their opinion regarding different topics in health care more easily. Therefore, All Voices Count has the potential to lower the barrier of including the voice of adolescents in decision-making in health care, research and policy.

Part 2: Patient Reported Outcome Measures

Another aspect of PCC are Patient Reported Outcome Measures (PROMs). PROMs are standardized, validated questionnaires that can be used to include patients' perspective into decision-making in clinical care. The use of PROMs has shown to be effective in enabling PCC as it increases awareness for patients' problems and concerns, enhances patient satisfaction, and is associated with improved treatment outcomes.

To facilitate the use of PROMs in clinical practice, the Emma Children's Hospital Amsterdam UMC developed the KLIK PROM portal (www.hetklikt.nu). Over the years, the KLIK PROM portal has been implemented in more than 40 hospitals in the Netherlands. The KLIK expertise center for PROMs and PREMs guides the implementation process. However, PROM implementation is a challenging process and successful implementation requires insight into barriers and facilitators that influence implementation outcomes. In addition, the involvement of all stakeholders (i.e., pediatric patients, parents, clinicians) is crucial. Therefore, **Chapter 4** focused on clinicians' experiences with the implementation of PROMs in pediatric clinical practice using the KLIK PROM portal. In general, clinicians (N=148) were satisfied with the use of the KLIK PROM portal. It gives them a valuable tool for systematically monitoring and discussing patients' functioning. However, several barriers were identified, such as irrelevant and long PROMs, low response rate, and no integration with the electronic health record. Subsequently, the patients' and parents perspective on the use of the KLIK PROM portal was described in **Chapter 5**, which showed that patients (N=8 focus groups, N=31 questionnaire) and parents (N=17 focus groups, N=130 questionnaire) were satisfied with the use of PROMs through the KLIK PROM portal. However, some points of improvement were identified; irrelevant and long PROMs, PROMs were not discussed by the clinician, and a suboptimal lay-out of the KLIK PROM portal. In **Chapter 6**, implementation science was used by the KLIK expert team to get insight into facilitating determinants and barriers in the implementation process, where after implementation strategies were identified. The Consolidated Framework for Implementation Research (CFIR) was used. Facilitating determinants were found mainly in the CFIR domains ‘intervention characteristics’, ‘characteristics of individuals’, and ‘process of implementation’, while barriers were identified in the domains ‘inner setting’ and ‘intervention characteristics’. The strategy ‘identify and prepare champions’ was acknowledged as the most effective strategy in addressing the combination of identified barriers. Based on the identified barriers and facilitators by all stakeholders the KLIK PROM portal was optimized. For example, a front-end integration between the KLIK PROM portal and the electronic health record (EHR) was realized, a mobile version of the KLIK PROM portal was developed and the lay-out was modernized.

PROMs are often experienced as burdensome due to a long completion time and irrelevant and repetitive questions. To overcome this barrier, Computerized Adaptive Tests (CAT) of the Patient-Reported Outcomes Measurement Information System (PROMIS®) can be used. **Chapter 7** showed that the PROMIS pediatric measures are feasible and reliable to use in clinical hemophilia care (N=76). Compared with the well-known legacy instruments, the PROMIS pediatric measures had lower floor- and ceiling effects and the total number of items completed was lower, resulting in a reduced burden of completing PROMs. However, the number of PROMIS items administered was still relatively high. To be able to optimally implement PROMIS in clinical care, further research into the CAT stopping rule is necessary.

Part 3: Patient Reported Experience Measures

PCC requires feedback from patients about the received care and the extent to which care is perceived as patient-centered. Patient Reported Experience Measures (PREMs) can be used for this purpose. Although PREMs are increasingly used, there is a lack of guidelines, resulting in a wide variety of PREM use. **Chapter 8** provided an overview of the available PREMs and their use in pediatric clinical care. A total of 39 different PREMs were identified that were used in 14 countries worldwide, demonstrating the growing adoption of PREMs. Nearly all of the PREMS identified were generic, designed to measure the experience of health care, regardless of disease or care setting. In addition, more than half of the PREMs were completed by proxy. The PREMs included a wide variety of domains. Communication (e.g., communication with parent, communication with child, communication about treatment, provider’s communication skills) was the most prominent domain across the various PREMs.

Remarkably, PREMs are often confused with PROMs and satisfaction questionnaires, although these three types of questionnaires all have different purposes and focus on different aspects of PCC. The interchangeability of concepts, which is seen around the use of PREMs, but also visible in the field of pediatric patient engagement (part 1), demonstrates the novelty of this research field and emphasizes the need for conceptual clarification.

This thesis ends with **Chapter 9,** the general discussion. This chapter includes a reflection on the main findings, clinical implications, methodological considerations, and future perspectives to promote PCC in clinical care and research.

Conclusion

The importance of patient-centered care is recognized, but its application is still in its infancy. The shift towards patient-centered care is difficult as it requires a shift in thinking and a redefinition of roles and decision-making for patients, parents, clinicians, policy makers and researchers. This thesis contributes to this goal as it provides scientific information and lessons learned for clinicians and tools to empower both patients and clinicians in their new roles.

Key messages

Part 1:

- Pediatric patients are increasingly engaged in health care projects with the aim of improving the quality of care and tailoring care to the patient's needs.
- Lack of uniformity about the definition of pediatric patient engagement and absence of clear information and support hinders engagement.
- All Voices Count is a patient engagement game that can help clinicians engaging pediatric patients in various health care projects.

Part 2:

- Implementing PROMs in pediatric clinical practice is a challenging process and barriers and facilitators can be identified on multiple domains.
- The engagement of all stakeholders (i.e., patients, parents and clinicians) is necessary for successful implementation.
- The PROMIS pediatric measures are valid alternatives to the well-known legacy instruments and have the potential to reduce the burden of completing PROMs.

Part 3:

- PREMs are increasingly used in pediatric health care, but more research is needed to use the PREM results in a meaningful way for quality improvement.

Summary in Dutch – Nederlandse samenvatting

Patiëntgerichte zorg wordt gekenmerkt door het leveren van zorg die de individuele wensen, behoeften en waarden van de patiënt respecteert en hierop inspeelt, en ervoor zorgt dat de wensen van de patiënt de leidraad vormen voor alle klinische beslissingen. Patiëntgerichte zorg plaatst de patiënt, en niet de ziekte, in het middelpunt van de gezondheidszorg. Het vraagt om een samenwerking tussen patiënt, familie en zorgverleners om zorg te verlenen die is afgestemd op de wensen en behoeften van de patiënt. Het bieden van patiëntgerichte zorg heeft voordelen voor zowel de patiënt als de organisatie, waaronder betere gezondheidsuitkomsten, meer patiënttevredenheid, grotere tevredenheid met werk en lagere gezondheidszorgkosten. Hoewel het belang van patiëntgerichte zorg wordt erkend is implementatie lastig, omdat het een andere manier van denken vraagt en een verandering van rollen van patiënten, ouders en zorgverleners vereist.

Dit proefschrift richt zich op wetenschappelijke benaderingen en de ontwikkeling van hulpmiddelen om patiëntgerichte zorg bij kinderen te bevorderen. Drie aspecten van patiëntgerichte zorg worden onderzocht. Het eerste deel van het proefschrift richt zich op het betrekken van patiënten in de kindergeneeskunde, het tweede deel richt zich op Patient Reported Outcome Measures (patient-gerapporteerde uitkomsten vragenlijsten) en het derde deel richt zich op Patient Reported Experience Measures (patiënt-gerapporteerde ervaringen vragenlijsten). In **Hoofdstuk 1**, de algemene inleiding, worden de verschillende onderdelen van patiëntgerichte zorg beschreven en wordt de context van het onderzoek in dit proefschrift geschetst.

Deel 1: Betrekken van patiënten in de kindergeneeskunde

Het betrekken van kinderen bij de besluitvorming in de klinische zorg en onderzoek met het doel de gezondheidszorg te verbeteren wordt gedefinieerd als '*pediatric patient engagement*'. Het betrekken van patiënten (in dit proefschrift richten we ons specifiek op kinderen en jongeren) in zorg en onderzoek is een essentieel aspect van patiëntgerichte zorg en het belang hiervan is vastgelegd in het Internationaal Verdrag inzake de Rechten van het Kind van de Verenigde Naties. Echter zien we dat zorgverleners en onderzoekers worstelen met het betrekken van patiënten binnen de kindergeneeskunde.

Om zorgverleners hierbij te helpen, biedt het eerste deel van dit proefschrift informatie en hulpmiddelen om kinderen op betekenisvolle wijze te betrekken. **Hoofdstuk 2** geeft een overzicht van de bestaande literatuur op het gebied van *pediatric patient engagement*. Dit literatuuroverzicht laat zien dat de belangstelling voor het betrekken van patiënten binnen de kindergeneeskunde de afgelopen decennia is toegenomen. Patiënten, met name in de leeftijd van 9 tot 17 jaar, worden steeds vaker betrokken bij een breed scala aan gezondheidszorgprojecten. Daarentegen laat dit literatuuroverzicht ook zien dat kinderen nog steeds nauwelijks betrokken worden bij onderzoeksprojecten. Dit suggereert dat onderzoekers meer hulpmiddelen en ondersteuning nodig hebben. Daarom werd in **Hoofdstuk 3** de ontwikkeling van een hulpmiddel (Alle Stemmen Tellen) voor het betrekken van patiënten beschreven. In samenwerking met adolescenten (N=25) is het spel Alle Stemmen Tellen ontwikkeld. Eerst werden 10 thema's die belangrijk waren voor adolescenten met een chronische aandoening ten aanzien van hun ziekte, behandeling en ziekenhuiszorg geïdentificeerd: ziekenhuisbezoek, participatie, ziekte en behandeling, sociale omgeving, gevoelens, omgang met personeel, acceptatie, autonomie, praten over de ziekte en chronische zieke leeftijdsgenoten. Op basis van deze thema's werd een eerste versie van Alle Stemmen Tellen ontwikkeld. Vervolgens werd Alle Stemmen Tellen meerdere keren geëvalueerd met jongeren en verder ontwikkeld op basis van de ontvangen feedback. Adolescenten waren tevreden met Alle Stemmen Tellen, omdat het hen helpt gemakkelijker hun mening te geven over verschillende onderwerpen in de gezondheidszorg. Alle Stemmen Tellen kan daarom de drempel om adolescenten te betrekken bij de besluitvorming in de gezondheidszorg, onderzoek en beleid verlagen.

Deel 2: Patient Reported Outcome Measures

Een ander aspect van patiëntgerichte zorg zijn Patient Reported Outcome Measures (PROMs). PROMs zijn gestandaardiseerde, gevalideerde vragenlijsten die gebruikt worden om het perspectief van patiënten te betrekken bij de besluitvorming in de klinische zorg. Onderzoek laat zien dat het gebruik van PROMs effectief is in het realiseren van patiëntgerichte zorg, omdat dit het bewustzijn voor de problemen en zorgen van patiënten vergroot, de patiënttevredenheid verhoogt en geassocieerd wordt met verbeterde behandelresultaten.

Om het gebruik van PROMs in de klinische praktijk te faciliteren, heeft het Emma Kinderziekenhuis Amsterdam UMC het KLIK PROM portaal ontwikkeld (www.hetklikt.nu). KLIK is inmiddels geïmplementeerd in meer dan 40 ziekenhuizen in Nederland. Het KLIK expertisecentrum voor PROMs en PREMs begeleidt het implementatieproces. Het implementeren van PROMs is een uitdagend proces en voor een succesvolle implementatie is het nodig om inzicht te hebben in de barrières en faciliterende factoren die de implementatie beïnvloeden. Daarnaast is het cruciaal om alle belanghebbenden (d.w.z. patiënten, ouders en zorgverleners) bij de implementatie te betrekken. **Hoofdstuk 4** richtte zich daarom op de ervaringen van zorgverleners met de implementatie van PROMs in de kindergeneeskunde middels het KLIK PROM portaal. Zorgverleners (N=148) gaven aan tevreden te zijn met het gebruik van het KLIK PROM portaal. Het geeft hen een waardevol hulpmiddel om het functioneren van patiënten systematisch te monitoren en te bespreken. Zorgverleners noemden daarnaast ook een aantal barrières, zoals irrelevante en lange PROMs, een lage respons van patiënten en ouders en geen integratie met het elektronisch patiëntendossier. Het perspectief van patiënten en ouders ten aanzien van het gebruik van het KLIK PROM portaal werd vervolgens beschreven in **Hoofdstuk 5**. Hieruit bleek dat patiënten (N=8 focusgroepen, N=31 vragenlijst) en ouders (N=17 focusgroepen, N=130 vragenlijst) tevreden zijn met het gebruik van PROMs via het KLIK PROM portaal. De volgende barrières werden genoemd; irrelevante en lange PROMs, PROMs worden niet besproken door de zorgverlener en een suboptimale lay-out van het KLIK PROM portaal. In **Hoofdstuk 6** werd implementatiewetenschap door het KLIK team gebruikt om inzicht te krijgen in faciliterende factoren en barrières in het KLIK implementatieproces. Vervolgens werden implementatie strategieën geïdentificeerd. Het Consolidated Framework for Implementation Research (CFIR) werd hiervoor gebruikt. Faciliterende factoren werden met name gevonden in de CFIR domeinen ‘kenmerken van de interventie’, ‘kenmerken van het individu’ en ‘implementatieproces’, terwijl barrières werden gevonden in de domeinen ‘interne omgeving’ en ‘kenmerken van de interventie’. De implementatiestrategie ‘identificeren en voorbereiden van voorlopers’ werd gezien als de meest effectieve strategie om de combinatie van geïdentificeerde barrières aan te pakken. Naar aanleiding van de geïdentificeerde barrières en faciliterende factoren van alle belanghebbenden in **Hoofdstuk 4 t/m 6** werd het KLIK PROM portaal geoptimaliseerd. Zo werd er een front-end integratie gerealiseerd tussen KLIK en het elektronisch patiëntendossier, werd er een mobiele versie van het KLIK PROM portaal ontwikkeld en werd de lay-out gemoderniseerd.

PROMs worden vaak als belastend ervaren door patiënten en ouders vanwege de lange invultijd en irrelevante en herhalende vragen. Om deze barrière te verminderen, kunnen de Computer Adaptieve Tests (CATs) van het Patient-Reported Outcomes Measurement Information System (PROMIS®) worden gebruikt. **Hoofdstuk 7** liet zien dat de PROMIS vragenlijsten voor kinderen betrouwbaar zijn voor gebruik in de hemofiliezorg (N=76). In vergelijking met bekende, veelgebruikte vragenlijsten binnen de hemofilie hebben de PROMIS vragenlijsten minder vloer- en plafondeffecten en is het totaal aantal in te vullen items lager. Dit leidt er toe dat de belasting van het invullen van PROMs lager is. Echter was het aantal in te vullen PROMIS items nog steeds relatief hoog. Om PROMIS optimaal te kunnen implementeren in de klinische zorg is verder onderzoek nodig naar de stopregels van de CATs.

Deel 3: Patient Reported Experience Measures

Patiëntgerichte zorg vraagt om feedback van patiënten over de zorg die zij hebben ontvangen en de mate waarin de zorg als patiëntgericht wordt ervaren. Patient Reported Experience Measures (PREMs) kunnen voor dit doel gebruikt worden. Hoewel PREMs steeds vaker gebruikt worden, ontbreekt een richtlijn. Hierdoor wordt een grote verscheidenheid aan PREMs op verschillende manieren gebruikt. **Hoofdstuk 8** geeft een overzicht van de beschikbare PREMs en de manier waarop ze gebruikt worden binnen de kindergeneeskunde. In totaal werden er 39 PREMs geïdentificeerd die gebruikt worden in 14 landen wereldwijd. Bijna alle geïdentificeerde PREMs waren generiek. Dit wil zeggen dat deze PREMs ontworpen zijn om de ervaringen van de gezondheidszorg te meten, ongeacht aandoening of zorgsetting. Bovendien werd meer dan de helft van de PREMs ingevuld door de ouders. De PREMs gaan in op een breed scala aan domeinen. Communicatie (bijvoorbeeld communicatie met ouders, communicatie met het kind, communicatie over de behandeling, communicatievaardigheden van zorgverleners) is het meest voorkomende domein in de verschillende PREMs.

Het is opvallend dat PREMs vaak verward worden met PROMs en tevredenheidsvragenlijsten, terwijl deze vragenlijsten allemaal een ander doel hebben en zich richten op verschillende aspecten van patiëntgerichte zorg. De verwarring ten aanzien van concepten rondom het gebruik van PREMs en op het gebied van *pediatric patient engagement* (deel 1) toont aan dat dit onderzoeksveld nog erg nieuw is en benadrukt de behoefte aan verduidelijking van concepten.

Dit proefschrift eindigt met **Hoofdstuk 9**, de algemene discussie. Dit hoofdstuk bevat een reflectie op de belangrijkste bevindingen, klinische implicaties, methodologische overwegingen en toekomstperspectieven om patiëntgerichte zorg te bevorderen.

Conclusie

Hoewel het belang van patiëntgerichte zorg erkend wordt, staat de toepassing ervan nog in de kinderschoenen. De verschuiving naar patiëntgerichte zorg is lastig, omdat het vraagt om een andere manier van denken en een herdefiniëring van de rollen van patiënten, ouders, zorgverleners en beleidsmakers. Dit proefschrift draagt bij aan de bevordering van patiëntgerichte zorg. Het biedt wetenschappelijke informatie, een praktisch toepasbaar hulpmiddel en geleerde lessen die zorgverleners kunnen ondersteunen in hun nieuwe rol.

Kernboodschappen

Deel 1:

- Kinderen worden steeds vaker betrokken bij projecten binnen de gezondheidszorg met het doel de kwaliteit van zorg te verbeteren en de zorg beter af te stemmen op de behoeften van de patiënt.
- Gebrek aan een uniforme definitie van *pediatric patient engagement* en het ontbreken van duidelijke informatie en hulpmiddelen belemmeren het betrekken van patiënten binnen de kindergeneeskunde.
- Alle Stemmen Tellen is een spel dat zorgverleners kan helpen om patiënten te betrekken bij verschillende projecten in de gezondheidszorg.

Deel 2:

- Het implementeren van PROMS in de kindergeneeskunde is een uitdaging met barrières en faciliterende factoren op meerdere domeinen.
- Het betrekken van alle belanghebbenden (d.w.z. patiënten, ouders en zorgverleners) is noodzakelijk voor een succesvolle implementatie.
- De PROMIS vragenlijsten voor kinderen zijn goede alternatieven voor de huidige vragenlijsten. De PROMIS lijsten hebben de potentie om de belasting die het invullen van vragenlijsten met zich mee brengt te verminderen.

Deel 3:

- PREMs worden steeds vaker gebruikt in de kindergeneeskunde, maar er is meer onderzoek nodig om de uitkomsten op een zinvolle manier te gebruiken voor kwaliteitsverbetering.

List of publications

This thesis

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Chapter 2: Pediatric patient engagement in clinical care, research and intervention development: a scoping review

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LT collected data, assisted in data extraction (search 2017 & 2021), and drafted the first version of the manuscript. LEV assisted in data collection and extraction (search 2017). HAvO and MJS supervised the writing of the first draft and critically revised the manuscript. EEWK assisted in data collection and extraction (search 2021). JGD developed the search strategy and collected data (search 2017 & 2021). MPG and MAG critically revised the manuscript for intellectual content. LH conceptualized and designed the study, collected data, supervised the data collection and data analysis (search 2017 & 2021), and revised the manuscript. All authors read and approved the final manuscript and agreed to be accountable for all aspects of the work.

Chapter 3: Including the voice of paediatric patients:
Cocreation of an engagement game

Authors: Lorynn Teela, Lieke E. Verhagen, Mariken P. Gruppen, Maria J. Santana, Martha A. Grootenhuis, Lotte Haverman

LH conceived the study. LT and LV carried out the focus groups and interviews and performed the qualitative analyses. The first draft of the manuscript was written by LT. MpG, MS, MaG, and LH handled the supervision. All authors critically revised the manuscript for intellectual content and approved the final version of the manuscript.

Chapter 4: Clinicians' perspective on the implemented KLIK PROM portal in clinical practice

Authors: Lorynn Teela, Maud M. van Muilekom, Lieke H. Kooij, Anouk W. Gathier, Johannes B. van Goudoever, Martha A. Grootenhuis, Lotte Haverman, Hedy A. van Oers

LT, MMvM, LH and HAvO developed the evaluation questionnaire. LT, AG and HAvO carried out the data collection. LT, LK and HAvO performed the statistical analysis. The first draft of the manuscript was written by LT. JBvG, MAG, LH and HAvO handled the supervision. All authors critically revised the manuscript for intellectual content and approved the final version of the manuscript.

Chapter 5: Patients' and parents' perspective on the implementation of Patient Reported Outcome Measures in pediatric clinical practice using the KLIK PROM portal

Authors: Lorynn Teela*, Maud M. van Muilekom*, Hedy A. van Oers, Johannes B. van Goudoever, Martha A. Grootenhuis, Lotte Haverman

LH conceived the study. LT, MMvM, HAvO, and LH conceptualized and designed the study, performed the focus groups, and developed the evaluation questionnaire. LT and MMvM performed the qualitative and statistical analyses and wrote the first draft of the manuscript. JBvG, MAG, LH and HAvO handled the supervision. All authors critically revised the manuscript for intellectual content and approved the final version of the manuscript.

Chapter 6: Implementation of the KLIK PROM portal using the Consolidated Framework for Implementation Research (CFIR) retrospectively
Authors: Lorynn Teela*, Hedy A. van Oers*, Sasja A. Schepers, Martha A. Grootenhuis, Lotte Haverman

LH conceived the study. LT, HAvO, SAS, LH conceptualized and designed the study. LT, HAvO, SAS performed the analyses and wrote the first draft of the manuscript. MAG and LH handled the supervision. All authors critically revised the manuscript for intellectual content and approved the final version of the manuscript.

Chapter 7: Psychometrics of the patient-reported outcomes measurement information system measures in hemophilia; the applicability of the pediatric item banks
Authors: Lorynn Teela, Michiel A.J. Luijten, Isolde A.R. Kuijlaars, Tessa C.M. van Gastel, E. Shannon van Hoorn, Samantha C. Gouw, Karin C.J. Fijnvandraat, Kathelijn Fischer, Marjon H. Cnossen, Sasja Andeweg, Carolien van der Velden – van ‘t Hoff, Corinne Liem, Margreet E. Jansen-Zijlstra, Marjolein Peters, Lotte Haverman

MP and LH conceived the study. LT, IARK, ESvH, SCG, KCJF, KF, and MHC contributed to study design. Data collection was led by LT and supported by MHC, SA, CvdVvtH, CL, MEJZ. LT performed the statistical analyses and MAJL, TCMvG, MP, LH contributed to the interpretation of the data and modification of statistical analyses. The first draft of the paper was written by LT. MP and LH were responsible for the supervision. All authors critically revised the manuscript for intellectual content and approved the final version of the manuscript.

Chapter 8: Use of Patient-Reported Experience Measures in pediatric care: a systematic review
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MS, LH, SB, SA, EG, LT, and HvO contributed to the overall rationale and design of the review. ND provided expert input on developing search strategy. SB, MZ, SR, SA, and LT assessed studies for eligibility against inclusion and exclusion criteria including gray literature and assessed risk of bias and also extracted the data and synthesized results. SB, MZ, SR, and SA led the drafting of the manuscript. MS, LH, SB, SA, EG, LT, HvO, MZ, and SR contributed to the subsequent drafts of the manuscript. All authors contributed to the article and approved the submitted version.

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PhD Portfolio

Name: Lorynn Teela
PhD period: November 2018 – July 2023
Promotor: Prof. dr. J.B. van Goudoever
Co-promotores: dr. L. Haverman & dr. H.A. van Oers

I. PhD training	Year	Workload (ECTS)
Courses		
Didactical Skills	2019	0.4
Peer to Peer Group Coaching	2020	0.5
eBROK	2020	1.5
Scientific Writing	2020	1.5
Clinical Epidemiology: Systematic Reviews	2021	0.7
Practical Biostatistics	2021	1.4
Re-registration eBROK	2023	0.2
Seminars		
Two-weekly research meetings – department of Child and Adolescent Psychiatry & Psychosocial Care	2018-2023	4
Selected for the TULIPS PhD curriculum	2020-2022	4
One-monthly research seminar VKC Psyche	2021-2023	1
One-monthly PROMIS researchers network meeting, online	2021-2023	1
Oral presentations		
Patient participation in pediatric care: A systematic review. Amsterdam Pediatrics Symposium	2018	0.5
All Voices Count: Development of a game to facilitate pediatric patient participation. Amsterdam Pediatrics Symposium	2019	0.5
Alle Stemmen Tellen: Hét spel om patiëntenparticipatie te bevorderen. Symposium ‘Zorg voor tieners: een vak apart’, Prinses Máxima centrum voor Oncologie, Utrecht	2019	0.5
Evaluating the KLIK PROM portal in clinical practice: patients’ and parents’ point of view. Amsterdam Pediatrics Symposium	2020	0.5
How the COVID-19 pandemic impacts the psychosocial well-being of children and adolescents in the Netherlands. ISOQOL 27th Annual Conference, online	2020	0.5
Het KLIK PROM portaal. Refereeravond Medische Psychologie	2021	0.2
A retrospective assessment of the KLIK PROM portal implementation using the Consolidated Framework for Implementation Research. European Implementation Event, online	2021	0.5
Transition in haemophilia care: from paediatric to adult care. The European Association for Haemophilia and Allied Disorders (EAHAD), 15th Annual Conference, online	2022	0.5
Including the voice of pediatric patients: Cocreation of an engagement game. 2023 Person-Centred Care Forum, Calgary Canada, Hybrid	2023	0.5

Poster presentations		
The development of a game to facilitate pediatric patient participation in hospital care, research and intervention development. European Pediatric Psychology Conference, Ghent, Belgium	2018	0.5
Participation of pediatric patients in hospital care, research and intervention development: a systematic review. European Pediatric Psychology Conference, Ghent, Belgium	2018	0.5
Health-Related Quality of Life in children with severe skin diseases before and after medical treatment. Pediatric Psychology Conference, Ghent, Belgium	2018	0.5
The development of a game to facilitate pediatric patient participation in hospital care, research and intervention development. ISOQOL 25th Annual Conference, Dublin, Ireland	2018	0.5
Participation of pediatric patients in hospital care, research and intervention development: a systematic review. ISOQOL 25th Annual Conference, Dublin, Ireland	2018	0.5
The development of a game to facilitate pediatric patient participation in hospital care, research and intervention development. Amsterdam Public Health (APH) conference, Amsterdam	2018	0.5
The development of a game to facilitate pediatric patient participation in hospital care, research and intervention development. Science Exchange Day, Amsterdam	2019	0.5
Evaluating the KLIK PROM portal in clinical practice: patients' and parents' point of view. ISOQOL 26th Annual Conference, San Diego, USA	2019	0.5
Implementing the KLIK PROM portal in transitional care. ISOQOL 26th Annual Conference, San Diego, USA	2019	0.5
The use of generic and disease-specific patient reported outcome measures in daily clinical practice of a pediatric nephrology unit. ISOQOL 27th Annual Conference, online	2020	0.5
Patients' and parents' perspective on the implementation of Patient Reported Outcome Measures in pediatric clinical practice. European Pediatric Psychology Conference, online	2021	0.5
The use of pediatric PROMIS item banks in Dutch boys with haemophilia. ISOQOL 28th Annual Conference, online	2021	0.5
The use of pediatric PROMIS item banks in Dutch boys with haemophilia. ISOQOL 28th PROMIS Health Organization, 7th Annual Conference, online	2021	0.5
The use of pediatric PROMIS item banks in Dutch boys with haemophilia. The European Association for Haemophilia and Allied Disorders (EAHAD), 15th Annual Conference, online	2022	0.5

(Inter)national conferences		
Medical Psychology Research Meeting, Amsterdam	2017, 2019, 2021	0.75
TULIPS Young Researchers Day	2017-2021	1.2
Amsterdam Pediatrics Symposium	2018-2023	1.5
Conferentie Transitiezorg: niet zo, maar zo!, Nieuwegein	2018	0.2
European Pediatric Psychology Conference, Ghent, Belgium	2018	0.5
International Society for Quality of Life Research (ISOQOL), 25th Annual Conference, Dublin, Ireland	2018	0.5
International Society for Quality of Life Research in the Netherlands (ISOQOL-NL) conferences	2018, 2019	0.5
Amsterdam Public Health Annual Meeting, Amsterdam	2018	0.2
Implementation Science Conference, Utrecht	2019	0.2
Symposium ‘Zorg voor tieners: een vak apart’, Prinses Máxima centrum voor Oncologie, Utrecht	2019	0.1
Amsterdam Center for Health Communication (ACHC) Symposium – Gezondheid in Beeld: De rol van visuele communicatie, Amsterdam	2019	0.1
Annual CaRe Days, Eindhoven	2019, 2022	1
Science Exchange Day, Amsterdam	2019	0.2
Dutch Pediatric Psychology Network conference, Utrecht	2019	0.2
International Society for Quality of Life Research (ISOQOL), 27th Annual Conference, online	2020	0.5
Symposium Jeugd in Onderzoek, online	2021	0.2
Amsterdam Public Health, Spring Meeting, online	2021	0.1
AmsCis Network: How to engage key users for successful implementation?, online	2021	0.1
European Implementation Event, online	2021	0.5
Amsterdam Public Health, Junified, Amsterdam	2021	0.2
European Pediatric Psychology Conference, online	2021	0.2
AmsCis Network: Yes, we can, online	2021	0.1
International Society for Quality of Life Research (ISOQOL), 28th Annual Conference, online	2021	0.5
PROMIS Health Organization, 7th Annual Conference, online	2021	0.5
Amsterdam Reproduction & Development, Retreat, Amsterdam	2021	0.2
The European Association for Haemophilia and Allied Disorders (EAHAD), 15th Annual Conference, online	2022	0.5
Conferentie Uitkomstgerichte Zorg: Generieke PROMS, online	2022	0.1
Symposium Measuring What Matters, International Society for Quality of Life Research (ISOQOL), online	2022	0.2

Workshops		
Inspirational pitching	2020	0.1
PURE workshop	2021	0.1
Pitching workshop, Speech Republic	2022	0.1
Getting published, Nature Research Academies	2022	0.5
Coach Uitkomstgerichte Zorg	2022	0.3

II. Teaching	**Year**	**Workload (ECTS)**
Lecturing		
KLIK training for professionals	2017-2023	4
Supervising		
Interns KLIK team	2018-2020	2
Co-supervisor Master Thesis Medicine – Esmée Kramer Patient-centered care: placing the pediatric patient in the center, not in the middle.	2021	1

III. Parameters of esteem	**Year**	**Workload (ECTS)**
Awards and Prizes		
Poster award (first prize) at the Pediatric Psychology Conference, Ghent, Belgium	2018	
Poster award (second prize) at the Science Exchange Day, Amsterdam	2019	
Other		
Member of organising committee TULIPS Young Researchers Day	2019-2020	2
Member of organising committee AR&D Retreat	2021	1
Member of organising committee Amsterdam Pediatrics Symposium	2022-2023	2
Member of the Children's Advisory Board Emma Children's Hospital	2023	0.1

Curriculum Vitae

Lorynn Teela was born on 30 November 1989 in Hengelo (O), the Netherlands. After graduating from Twickel College Hengelo, she moved to Groningen and started with the bachelor Educational Sciences. After one year, she combined this study with the bachelor Psychology. After finishing her master in Educational Sciences cum laude, Lorynn was admitted to the 2-years master program in Medical Psychology and moved to Brabant. In the first year of this master, she took several courses at the intersection of psychology and medicine. In the second year, she did an internship at the Catharina Hospital Eindhoven and wrote her master thesis. She graduated with distinction in 2015.

After her study, Lorynn worked as a psychologist at Opdidakt. In November 2017, she started as a junior researcher / implementation assistant on the KLIK project at the department of Child and Adolescent Psychiatry & Psychosocial Care of the Emma Children's Hospital, Amsterdam UMC. After one year, she started working on her PhD focusing on pediatric patient engagement, implementation of Patient Reported Outcome Measures (PROMs) in daily clinical practice and the use of Patient Reported Experience Measures (PREMs) under the supervision of prof dr. Hans van Goudoever, dr. Lotte Haverman, and dr. Hedy van Oers.

In the past years, Lorynn combined her PhD project with the coordination of the national implementation of the KLIK PROM portal in clinical practice. In addition, she is involved in various research projects in the field of hemophilia.