

Empowering Patients and Health Professionals to Address Sexual Health in the Context of Anorectal Malformations and Hirschsprung's Disease

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Abstract

The aim of this study is to develop a tool that is aligned with patients' and health professionals' needs to address sexual health in the context of anorectal malformations and Hirschsprung's disease. A multiphased participatory action–research was conducted. First, an inventory of needs was made through interviews (11 patients, 11 professionals), three online focus groups (4 patients, 20 professionals), and a questionnaire (38 patients). Subsequently, four cocreation sessions with in total four patients and nine professionals were organized to translate the needs into a tool (in the form of a website). The websites' functionality was assessed via a questionnaire ($n = 34$). The website, directed to patients, their parents, and professionals, stimulates awareness, fills knowledge gaps, and shows possibilities for support. The website is expected to change restrictive attitudes toward sexual health and improve the legitimization of the topic needed for the allocation of resources and sexologists' involvement in current care pathways.

Keywords

users' experiences; health care; holistic care; experiences; illness and disease; power; empowerment; qualitative; quality of life; sexuality; sexual health; the Netherlands

Introduction

Globally, the congenital diseases, anorectal malformations (ARM) and Hirschsprung's disease (HD), occur one in every 5,000 live births (Rintala, 2009). These diseases are, respectively, caused by the incomplete or insufficient development of the anus or rectum (Holschneider & Puri, 2008) or the absence of enteric nervous system along the intestine (Amiel et al., 2008). Although ARM and HD are often diagnosed and treated during the neonatal period, patients often experience long-term gastrointestinal morbidity such as chronic incontinence, constipation, or defective bowel control (Rigueros Springford et al., 2016; Rintala & Pakarinen, 2012; Stenström et al., 2014). In addition, psychological problems are frequently observed, such as shame due to fecal incontinence. Moreover, wearing a stoma (Grano et al., 2011; Leitner et al., 2017) or dilatations during childhood (Athanasakos et al., 2006; Witvliet et al., 2017) may contribute to a negative body image. These psychological problems may make patients hesitant to seek friendships and sexual relationships in adulthood (Witvliet et al., 2018) and cause sexual impairment such as erectile dysfunction, vaginism, or dyspareunia—genital

pain that interferes with sexual intercourse (Konuma et al., 2006; Schmidt et al., 2012; van den Hondel et al., 2015). Sexual impairment could also be the result of postoperative problems, such as scar tissue, or constrictions in the genital area due to urogenital anomalies (Athanasakos et al., 2006; Manderson, 2005; Witvliet et al., 2017).¹ These problems can negatively affect the sexual health of these patient groups (Kyrklund et al., 2016; Rigueros Springford et al., 2016; van den Hondel et al., 2015).

Good sexual health is essential for a person's physical and emotional well-being and thus quality of life (Grano et al., 2012). A study by van den Hondel et al. (2015) highlighted that patients with ARM and HD experience a need for support in managing their sexual problems. Nevertheless, there is a lack of attention to sexual health in clinical practice (Schmidt et al., 2012; Witvliet et al., 2017). van den Hondel et al. (2015) showed that 93.5% of the patients they questioned did not receive any support—in the form of counseling or patient education—regarding (future) sexual problems. Of these patients, 60% (ARM) and 59% (HD) felt a need for education (van den Hondel, 2015). In addition, Witvliet et al. (2017)

found that only one out of 168 patients with ARM and HD was referred to a specialist for sexual-related issues. Various authors suggested the need for better follow-up during adulthood to improve support (Stenström et al., 2014; Witvliet et al., 2018). Previous studies concerning other diseases showed that neither patients nor health professionals initiate the discussion on sexual health (McInnes, 2003; Traumer et al., 2019). Patients feel reluctant to ask for help regarding sexual issues (Hautamäki et al., 2007; Vermeer et al., 2015) due to a lack of trust (Fitch et al., 2013; Hughes & Lewinson, 2014) and feelings of embarrassment (Dyer & das Nair, 2013; Traumer et al., 2019). Health professionals also find it hard to bring up the topic due to personal discomfort (e.g., Dyer & das Nair, 2013; Traumer et al., 2019; Vermeer et al., 2015). Moreover, a lack of knowledge and (conversational) skills (Dyer & das Nair, 2013; Hautamäki et al., 2007; Ho & Fernández, 2006), limited time (van Ek et al., 2015; Vermeer et al., 2015) and resources, giving low priority to the topic, and not feeling responsible have been found to constrain health professionals from providing sexual health support (Magnan et al., 2005; Saunamäki et al., 2010).

To facilitate communication about sexual health during consultations, various tools have been developed, such as the “(extended) PLISSIT² model,” “Talking of Sex,”³ and “SeCZ TaLK”⁴ (Annon, 1976; Macdowall et al., 2010; Taylor, 2006; Taylor & Davis, 2007; van der Stege et al., 2010). The level of abstractness or practical implementation differs between these models. Although found to be useful for providing care in general, their

applicability in the context of ARM and HD is considered limited. Health professionals face difficulties in integrating either of these tools into practice, as also shown by van der Stege et al. (2014) in another care context. It is therefore relevant to align the tools to the specific needs of patients with ARM and HD and the health professionals involved. This also requires a better understanding of the mechanisms that underlie the barriers to discussing sexual health (Saunamäki et al., 2010). This study aims to acquire insights into patients’ and health professionals’ needs to address sexual health in the context of ARM and HD and—from this understanding—to develop a tool that is aligned with patients’ and health professionals’ needs through a participatory approach.

Theoretical Background

To obtain insights into patients’ and health professionals’ needs to address sexual health, insights into their empowerment could be helpful. To guide this study, several patient and professional empowerment models were integrated into a health professional–patient empowerment model (see Figure 1). The structure of the model is based on the model of Spence Laschinger et al. (2010). Following this model patients’ outcomes are the result of their empowerment, which depends among other factors, on support from health professionals. In turn, for health professionals to feel equipped to support patients, they also need to be empowered.

Professionals’ empowerment is conceptualized at a structural level – the work settings - and a psychological

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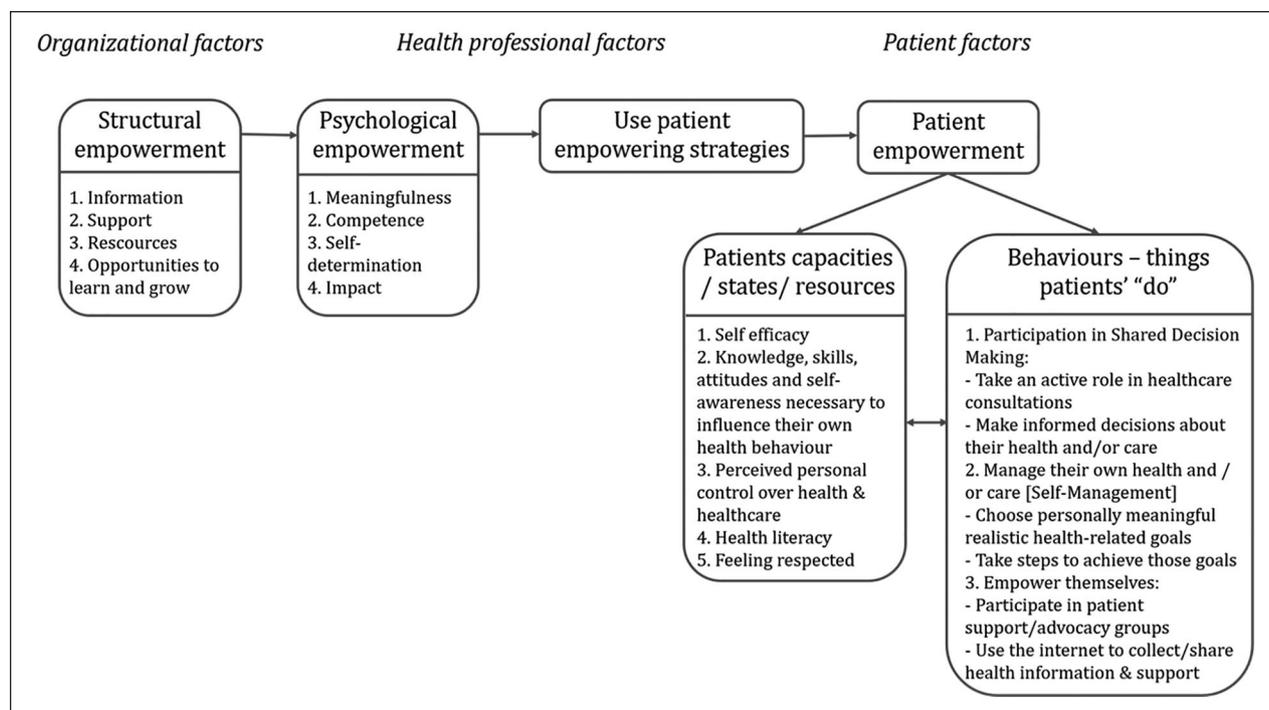


Figure 1. Health professional–patient empowerment model based on the extended nurse–patient empowerment model of Spence Laschinger et al. (2010), the professional empowerment models of Kanter (1987) and Thomas and Velthouse (1990), and the patient empowerment model of Bravo et al. (2015).

level – health professionals’ reaction to these work settings (Laschinger et al., 2001). To further specify these work settings, the model of Kanter (1987) offers insights. Following this model, four categories of work settings are distinguished: (a) *information*: the access to knowledge about, for instance, organizational policies, goals, and values; (b) *support*: the availability of feedback and instructions from managers and colleagues; (c) *resources*: access to requirements, such as materials, money, and time needed to fulfill organizational objectives; and (d) *opportunities to learn and grow*: opportunities to learn and grow refer to the availability of rewards and challenges in the work settings (Kanter, 1987). Using the model of Thomas and Velthouse (1990), psychological empowerment is described as professionals’ intrinsic motivation in response to the workplace environment, including the following four dimensions: (a) *meaningfulness*: the perceived value of certain tasks or goals; (b) *competence*: individuals’ belief in their capability to perform their work well; (c) *self-determination*: the degree to which people perceive that they have a choice in introducing and regulating actions; and (4) *impact*: the degree to which people can influence outcomes and issues at work (Thomas & Velthouse, 1990).

To conceptualize the empowerment of patients, the patient empowerment model of Bravo et al. (2015) is helpful. According to the model, patients’ empowerment level can be determined by two indicators: (a) patients’ “states”—what patients need to become empowered—and

(b) patients’ “behaviors”—the actions that increase the empowerment “states.” The “states” (patients’ capacities, states, and resources) consist of the elements (a) self-efficacy; (b) knowledge skills, attitudes, and self-awareness necessary to influence their own health behavior; (c) perceived personal control over health and health care; (d) sense of meaning and coherence about their condition; (e) health literacy; and (f) feeling respected. Three types of “behaviors” are distinguished: (a) shared decision-making, (b) self-management, and (c) patient activation. There is a reciprocal relationship between these indicators. For example, patients’ knowledge influences patients’ activation, which in turn leads to gaining more knowledge.

Method

This study was a multidisciplinary collaboration conducted in the period December 2017 to April 2019 in the Netherlands. To guide the study, a project group⁵ was set up. The group met before the start of each project phase to discuss the plans and to reflect on the actions and observations of the previous phase.

Participatory Action Research (PAR)

To develop a tool to empower both patients and health professionals to address sexual health, a PAR was conducted. PAR is a participatory approach that aims to realize

Table 1. Patient characteristics of interview, OFG and questionnaire respondents ($n = 53$).

Aspect	Category	Interviews	OFG	Questionnaire	Total
Gender	Female	5	3	23	31
	Male	6	1	15	22
Age (years)	16–20	0	3	2	5
	21–30	6	1	16	23
	31–45	2	0	9	11
	45–60	3	0	6	9
	60+	0	0	2	2
	Unknown	0	0	3	3
Diagnosis	ARM	10	2	32	44
	HD	1	2	6	9

Note. OFG = online focus group; ARM = anorectal malformations; HD = Hirschsprung's disease.

meaningful social change (Baum, 2016; Savin-Baden & Wimpenny, 2007; Shamrova & Cummings, 2017). Because the academic researchers who led the project work in close collaboration with the relevant stakeholders—in this case patients and health professionals—actions better reflect their knowledge and are better tailored to their wishes and needs (Baum, 2016; McIntyre, 2008). The study consisted of three phases: (a) *articulating barriers and (empowerment) needs*; (b) *translating needs into empowerment tools*; and (c) *assessing the empowerment tools*. In each phase, a cyclical process took place of *planning, taking action, observing, and reflecting* (Gray, 2013). The learnings from each phase were integrated in the subsequent one.

Phase 1: Articulating experienced barriers and (empowerment) needs. Both patients' and health professionals' experiences with sexual health support and their respective needs were identified through an iterative process using a mixed-method design consisting of semi-structured interviews, online focus groups (OFGs) and a questionnaire.

Participant recruitment. Letters were sent to individual patients treated at three collaborating medical centers. In addition, patients were recruited via meetings, emails, newsletters, and social media of the three patient organizations. Due to a limited response rate, it was decided to conduct mainly interviews ($n = 11$) instead of OFGs. An additional OFG was organized to specifically take the perspectives of adolescents ($n = 4$) into account. These adolescent patients (16–25 years old) were recruited during a weekend trip of two patient organizations and were familiar with each other. Furthermore, a questionnaire ($n = 38$) was disseminated via the patient organizations to validate the results of the interviews and the OFG. Patients were included who were above the age of 16, treated in the Netherlands and who speak either Dutch

or English. In total, 53 patients were involved who live in 11 of the 12 different provinces of the Netherlands. Table 1 gives an overview of the participating patients' characteristics.

Insights into health professionals' experiences and needs were obtained via interviews ($n = 11$) and two OFGs ($n = 20$). All health professionals who are working with the patient groups at the relevant medical centers were contacted via email and/or phone. Recruitment of relevant health professionals working outside the centers was done via snowball sampling. Health professionals of nine different specializations and working at six different medical centers were involved. An overview of the characteristics of all participating health professionals is shown in Table 2.

Data collection. The semi-structured interviews took 45 to 90 minutes. The interviews with patients were structured by discussing their disease and care process by making a timeline. Each step in time was related to their experiences and needs regarding their sexual health.

For the OFGs, a specific OFG tool was used, developed by Nivel (Tates et al., 2009). This tool consists of a secured online forum in which participants digitally respond on questions or statements or the comments of other participants. Because participants are able to join anonymously, this tool has been found useful for discussing taboo subjects, such as sexual health (Tates et al., 2009). In addition, the tool facilitates the participation of patients living in different regions and health professionals working at different medical centers. To generate an iterative learning process, the discussions were organized asynchronously. Each OFG lasted for ten days. This duration was found appropriate based on an informal talk with the developers of the tool and previous studies in which the tool was applied (Petit-Steeghs et al., 2019; Tates et al., 2009). During this period, participants could log in 24 hours a day. Participants were not online at the same

Table 2. Characteristics of participating health professionals ($n = 31$).

Aspect	Category	Interviews	OFG1	OFG2	Total
Profession	Pediatric surgeon	4	0	3	7
	(Pediatric) urologist	1	1	0	2
	Pediatrician	0	1	0	1
	Adult gastrointestinal specialist	1	0	0	1
	Nurse specialist	2	3	4	9
	Gynecologist	1	1	0	2
	Psychotherapist	1	2	1	4
	Sexologist	1	1	1	3
	Pelvic therapist	0	0	1	1
	Pediatric occupational therapist	0	0	1	1
	Deployment	Institutional	11	9	8
Outside the hospital		0	0	3	3
Gender	Female	9	7	10	26
	Male	2	2	1	5

Note. OFG = online focus group.

time but read each other's responses and commented on those within the time frame of ten days. Before the start of the OFGs, participants received written and—in the case of the patients also oral—instructions. Two researchers (first and third authors) moderated the discussions. The main questions for patients and health professionals—posted on days 1, 3, and 6—were structured in three parts. For the patients, these questions related to (a) experiences with sexual health, (b) experiences with discussing sexual health, and (c) needs to discuss sexual health. In the case of the health professionals, questions were directed to (a) current support of patients regarding their sexual health, (b) possibilities to improve the support for patients, and (c) needs to improve the current support. To obtain a more in-depth explanation and to stimulate interaction, the moderator posted supplementary questions or asked other participants about their view on a certain subject on a daily basis.

The questionnaire was developed by the online tool Qualtrics and comprised 14 open and closed questions. The questions were based on the outcomes of the previous consultation and were divided into four parts: (a) demographics, (b) experiences with sexual health problems, (c) needs regarding their sexual health, and (d) experiences with and needs regarding sexual health support.

Phase 2: Translating needs into empowerment tools. Based on the analysis of the findings of Phase 1, the project group decided to develop tangible empowering tools in the form of a website for both patients and health professionals. For each tool, a work group was set up consisting of two patient representatives and, respectively, five and four health professionals who also participated in Phase 1

of the research. Among the health professionals, five different disciplines were involved (pediatric surgeon, pediatrician, specialized nurse, sexologist, and a psychologist). Two cocreation sessions were organized per work group to generate a dialogue in which the articulated needs of Phase 1 were integrated into tools which aim to empower both patients and health professionals to address sexual health. The sessions lasted three hours and were moderated by two researchers (second and last authors). During the first sessions, the structure, format, and required content of the tangible empowering tools were determined. Subsequently, a draft tool was set up in collaboration with a text writer and a designer. In the second sessions, participants reflected upon the draft and attention was given to language, visualization, and implementation. Based on the feedback of the second session and the project group, the tools were improved and integrated into one website.

In addition, the process of the cocreation sessions was evaluated by an online questionnaire consisting of five open and four closed questions, of which one included 18 statements. The questions focused on both the process (stakeholders' representation, organization, facilitation, and interaction) and the (direct and indirect) outcomes of the cocreation process. The questionnaire was completed by 10 of the 13 participants.

Phase 3: Assessing the empowerment tools. The empowerment tools—in the form of a website—were assessed regarding its user-friendliness and expected (empowerment) outcome. Two online questionnaires were developed via the online tool Qualtrics: one for patients and parents/partners of patients, and one for health professionals. The questionnaires consisted of, respectively, 27 and 18 closed and open questions. The questions were

Table 3. Characteristics of patient and patients' parent respondents of the evaluation questionnaire.

Aspect	Category	Patients	Parents' Child	Total
Gender	Female	10	7	17
	Male	3	2	5
Age (years)	0–4	0	0	0
	5–8	0	5	5
	9–12	0	4	4
	12–16	0	0	0
	17–20	1	0	1
	21–30	4	0	4
	31–45	5	0	5
	45–60	0	0	0
	60+	3	0	3
Diagnosis	ARM	11	3	14
	HD	2	6	8

Note. ARM = anorectal malformations; HD = Hirschsprung's disease.

divided into four parts: (a) demographics; (b) feedback on the website; (c) impact of the website on their empowerment based on the models of Bravo et al. (2015), Kanter (1987), and Thomas and Velthouse (1990) for, respectively, patients and health professionals; and (d) preferences regarding the implementation of the website. The questionnaires were completed by 13 patients, nine parents, and 13 health professionals from eight different health care organizations. See Tables 3 and 4 for respondents' characteristics.

Data analysis

The interviews were transcribed verbatim. Subsequently, the transcripts of the interviews and OFGs were analyzed via the qualitative data-analysis program MAXqda 2007. A process of inductive and thematic analysis (Green & Thorogood, 2018) was performed by the first three authors. Deductively, codes were derived from the empowerment models of Bravo et al. (2015), Kanter (1987), and Thomas and Velthouse (1990). Inductive

coding was used for additional findings. Next, the categorized segments were clustered into subthemes. Field notes of the cocreation sessions were coded manually based on the coding scheme of the interviews and the online discussions. The data of the questionnaires were analyzed via descriptive analysis using Microsoft Excel based on the health professional–patient empowerment models. The categories and themes were structured following the model nurse–patient empowerment model of Spence Laschinger et al. (2010) and discussed among all authors.

Validation and Ethics

In the assessment of the formal medical ethical committee of Erasmus University Medical Center, it was confirmed that the study does not fall within the remit of the Medical Research Involving Human Subjects Act (IRB approval MEC-2017-1133). The study is non-invasive, does not include medical research, and involves patients who are above the age of 16. The

Table 4. Characteristics of health professional respondents of the evaluation questionnaire.

Aspect	Category	Total
Profession	Pediatric surgeon	2
	Pediatric urologist	1
	Gynecologist	2
	Sexologist	2
	Nurse specialist	1
	Incontinence and stomas consultant ^a	2
	General practitioner	1
Deployment	Institutional	10
	Outside the hospital	3

^aIn Dutch: WIS (Wond-, Incontinentie- en Stomazorg) consulent.

Table 5. Patients' Barriers and (Empowerment) Needs.

Barriers	Need	Needs Empowerment 'States'
Avoidance due to distress	Feeling 'normal'	Sense of meaning about their condition
Unaware of possibilities of professional support	Knowledge of possibilities professional support	Knowledge
Difficult to broach the topic	Empathetic response	Feeling respected
Not willing to discuss	Normalizing the (sexual) problems	Sense of meaning about their condition
Unaware of relation between disease and sexual health problems	Knowledge on (possible) sexual health problems	Knowledge
Unaware of possibilities of managing sexual health problems	Knowledge about self-management	Knowledge and skills

medical ethical committees of two other collaborating centers have followed this advice. The research complied with the national Code of Ethics for Research in the Social and Behavioural Sciences involving Human Participants (VCWE, 2018). All respondents received written and/or verbal information on the voluntariness and incentives of participation, the nature and purpose of the study and the right to withdraw at any time without giving reasons. Verbal informed consent was obtained for recording the interviews and cocreation sessions. All data were anonymized and stored securely. Summaries of the interviews and online discussions were sent to the participants for a member check.

Results

The results are described based on the three phases of this PAR study: (a) *articulation of patients' and health professionals' experienced barriers and needs*; (b) *translation of these needs into empowerment tools*; and (c) *assessment of the empowerment tools*. Examples are illustrated by quotes.

Phase I: Articulating Experienced Barriers and (Empowerment) Needs

Patients and health professionals experienced various barriers to and needs regarding patients' sexual health. The barriers and needs are discussed according to the empowerment concept of Bravo et al. (2015) for patients, and of Kanter (1987) and Thomas and Velthouse (1990) for health professionals.

Patients' barriers and needs. Patients' barriers and need are presented according to the empowerment "states" of Bravo et al. (2015): *sense of meaning about their condition*, *feeling respected*, and *knowledge and skills* (see Table 5).

Sense of meaning about their condition. More than half of the questionnaire respondents regarded their sexual health as meaningful by indicating its important contribution to

their physical and psychological well-being. Nevertheless, various interviewed patients addressed that they qualified their sexual health as less important than other needs, such as feeling "normal" and "safe," which were considered more apparent in daily life. The (sexual) problems were seen by patients as not "normal". Patients were often not aware that other people (not having the disease) could also experience similar sexual health problems due to other causes. To feel "normal" and "safe", these patients ignored their (sexual) problems by, for instance, having no sexual contact or not discussing their problems with others. By focusing on other things, patients did not have to deal with their problems. Avoidance was often used as a strategy to cope with their (sexual) problems and reduce stress:

It was a kind of mental denial. If I deny it, it will turn out better than expected. If I ignore it, it will prove better than anticipated. That was kind of the line of thought. (Interview patient)

In line with this, patient representatives hypothesized that many members of the patient organizations cancel their membership during puberty due to their need to feel "normal" and not being considered as a "patient". In addition, some patients indicated that they saw sexual health as a very intimate or even taboo topic which you do not discuss. Of the questionnaire respondents, less than a half of them (44%) discussed sexual health with their friends, a quarter (26%) with their parents, and only a few (17%) with health professionals. A few patients who indicated they had a more open family culture described their sexual problems as part of their life, sharing experiences with family and friends. Normalizing the topic helped these patients to address them:

And I think that if a health professional starts talking about it, it's more likely that you start thinking "Oh it's normal for me to talk" And if that person starts talking about it, then you think I can also talk about it. (Interview patient)

Feeling respected. Another reason for patients to avoid the topic were feelings of shame and anxiety:

Yeah, shame of course. Shame also in general. When discussing the topic with a medical specialist, who is also a person. (Interview patient)

Various patients described situations in which they clammed up and therefore were not able to ask questions or join the shared decision-making process. These feelings were reinforced by distrust as a result of bad experiences in the past caused by being “different”. A few patients who did address their problems during a consultation with a general practitioner or gynecologist felt that their concerns were not taken seriously. The health professionals’ limited response resulted in patients clamming up, precluding a constructive conversation. Patients addressed their need for a relationship of trust with a health professional with whom they are familiar to feel respected and able to discuss the topic.

Knowledge and skills. Patients repeatedly mentioned the need for knowledge and skills. Patients lacked knowledge about the potential sexual problems and the possibilities for self-management and professional support to manage these problems. Fewer than 25% of the questionnaire respondents received information on sexual health, which was mostly regarded as limited and ill-timed. About half of the questionnaire respondents indicated that—due to their lack of knowledge—they were limited able to manage their sexual problems. Yet, in time—through their experiences—patients often developed *skills* and coping strategies to be able to live with their problems. Various interviewed patients indicated, for instance, that they obtained control by restricting their sexual contact to times when the problems were limited. They controlled flatulence and fecal incontinence during sex by regulating their diet and managing their toilet visits and bowel enemas. Sexual foreplay, the use of lubrication, and relaxation exercises helped to relax and prevent vaginism. Most patients avoided one-night stands. Sexual contact was usually limited to the context of a long-term relationship, in which trust had been established. Talking with others—mainly their partner—helped some of the patients to try to offload their emotional baggage and find a place to put their emotions.

Various patients said that they had (now and/or in the past) limited knowledge of the possibilities of professional support. After the age of 18 years, patients often lacked a contact person at the hospital because the treatment had usually ended. As a result, they did not know whom to contact. The general practitioner was thought to have insufficient expertise. The patients who were still treated at the outpatient clinic were often uncertain whether the first responsible practitioner—due to her or his specialization—was the right person to contact. Pediatric surgeons or urologists were not thought to have knowledge on these topics and patients did not dare to

bother them about their concerns. This idea was reinforced by the fact that these health professionals did not mention this topic spontaneously. Patients were often not familiar with the specific expertise of a pelvic physiotherapist or sexologist or found out only later:

On my sisters’ advice I went to a pelvic physiotherapist. I was 18 years old then. If she would not have told me, I would not have known that such a person existed. (Interview patient)

Health professionals’ barriers and needs. Only half of the health professionals indicated having discussed sexual health with patients in the past and most of them do not discuss this topic on a regular basis. “Many topics are tackled, but this [sexual health] is rarely discussed” (Interview pediatric surgeon) and “Structurally discussing [sexual health] is still not common practice” (OFG specialized nurse). Health professionals mentioned various barriers to providing sexual health support to patients. To overcome these barriers, health professionals mentioned various (empowerment) needs at a structural and psychological level (see Table 6).

Structural empowerment. In relation to their structural empowerment, health professionals discussed the need for *information* and *resources*. Regarding *information*, they indicated that clarity is needed about who should be responsible for providing sexual health support. Currently there are different opinions on who should be responsible. Some health professionals thought the pediatric surgeon—as principal provider of health care—should be in charge. Due to the heavy workload of pediatric surgeons and their medical focus, others—including some of the pediatric surgeons—disagreed. Instead, they felt that this was the role of a specialized nurse, who is more accessible and easier for patients to approach. Different opinions were also expressed on whether the responsible health professional should have a long-term relationship of trust with the patient. Some regarded such relationship as beneficial, others as an obstacle for an open conversation on sexual health. A few health professionals mentioned that the whole multidisciplinary team should be responsible. If only one health professional is responsible, it is difficult to guarantee continuity in providing sexual health support. A need for information was also addressed regarding whether, when, and to whom patients should be transferred if sexual problems are diagnosed. Health professionals were seldom aware of how a sexologist and pelvic physiotherapist might be deployed. As a result, only a few patients were referred to those specialists:

That there is someone like a sexologist is already an eye-opener for some [health professionals]. And what a sexologist does. It would be good to know why you refer

Table 6. Health Professionals' Barriers and (Empowerment) Needs.

Barriers	Need	Empowerment Need-Level
Responsibilities regarding sexual health unclear	Clear role and task division	Information—structural
Uncertainties transfers	Information possibilities sexologist/pelvic physiotherapist	Information—structural
No time to discuss the topic	Time during consults	Resources—structural
No time to obtain knowledge and skills	Time for professional development	Resources—structural
Lack of priority given to the topic	Awareness of meaning and importance sexual health	Meaningfulness—psychological
Difficult to broach the topic	Conversational skills	Competences—psychological
Difficult to cope with defensive reactions of (parents) patients	Conversational skills	Competences—psychological
Difficult to diagnose problems	Knowledge about (possible) sexual problems	Competences—psychological
Difficult to provide support	Knowledge on (possible) solutions for problems	Competences—psychological
Sexologist involved to limited degree	More central role within patients' care path	Impact—psychological

someone . . . Well, we are not very visible on the digital platform of the hospital. (Interview sexologist)

A majority of the health professionals described a lack of *resources* (time and money) to provide sexual health support. According to many health professionals, there is a need for extra time during the consultations to discuss sexual health in addition to other topics, such as incontinence, which are considered to have a higher priority. Time is also needed to obtain knowledge on sexual problems and develop conversational skills to discuss sexual health:

From the institution, we need an infrastructure to provide education on sexual health to different health professionals of the colorectal team. In addition, at any event one team member should be available for discussing sexuality as common practice during consults, supporting the other team members and to arrange the information provision to patients. (OFG pediatrician)

Psychological empowerment. Regarding their psychological empowerment, the elements *meaningfulness*, *competences*, and *impact* were discussed by health professionals. Although most health professionals described sexual health as a *meaningful* topic in (general) patient care, many—apart from nurses—did not regard sexual health as a meaningful topic in relation to their daily practice. Various health professionals mentioned that the topic was not relevant for them because they treated patients before the age of 12 years. Moreover, sexual health was in general seen by health professionals as having a lower priority than other medical issues such as incontinence. A lack of prioritization was especially apparent for pediatric surgeons and an adult gastroenterologist, who did not regard sexual health as part of their professional responsibilities:

I am not a sexologist that deals with the psychological issues. The question is whether I can help them [patients] with their incontinence. When their incontinence improves, they are doing better as well . . . We do not treat patients on sexual issues. (Interview adult gastrointestinal specialist)

Several health professionals mentioned that sexual health was not a priority for their patients, because they do not bring it up during the consultations. Awareness of the sexual health problems and the need to address them were mentioned as necessary for prioritizing the topic. Adopting sexual health as a standard topic in patients' medical history and protocols or using a top three topic list (in which patients can indicate the three main issues they would like to discuss) were suggested as ways to improve prioritization. Yet, some participants indicated that patients will probably feel hesitant to indicate sexual health on the topic list due to feelings of shame.

Various health professionals indicated that they do not feel *competent* to provide sexual health support. When bringing up the topic, health professionals are confronted with their own interests and values and those of the patient. This is especially the case for male health professionals who have to discuss sexuality with a young girl (especially in relation to the current “#me too discourse”):

In daily practice it [sexual health] is not discussed, from both sides. I never had a child that told me that he or she would like to discuss this [their sexual health] with you . . . that's from both sides. Maybe it is due to my conversational skills. Or the fact that I am dealing with young girls that won't like to discuss such topic with a guy of 40 years old. (Interview pediatric surgeon)

Gaining more experience in discussing sexual health—and especially with children and adolescents—would help in dealing with feelings of shame and overcoming discomfort.

Health professionals also cope with defensive reactions of patients' parents. Preparing parents for a future consultation which will be only with their child due to reasons of privacy was suggested as a possible solution for coping with parents' resistance. However, not all health professionals agreed on leaving parents outside the consulting room. According to sexologists, parents should be empowered to discuss sexual health with their children by mentioning it from early age. Parents could be informed—via consultations or a group session—about the problems that can arise at the different sexual stages and how they can address these potential problems:

I think the subject [sexual health] will be more easily discussed if there is already attention for the topic from early age on. A good moment is when the health professionals informs about primary school. Children with an atypical phenotype or scars get questions from other children: "what do you have there?" It is good that parents are aware that these questions can arise and that it is important for their child to be able to answer those questions. (OFG psychologist)

In addition, most of the health professionals indicated that they did not know what would be a "good age" to address sexual health. Some thought just before puberty, others at the age of 12, after achieving continence—because of the priority given to continence before that time—or at the age of three, because of the start of children's sexual development. A few health professionals mentioned that, in order to be empowering, the moment of introducing the topic should depend on the child's development. According to sexologists, knowledge about the stages of sexual development would be helpful in improving the timing of the support provided by health professionals. This knowledge was also found to be worthwhile for identifying problems (at an earlier stage). The development of a negative body image during childhood can, for instance, hinder starting intimate relations during adulthood. Underdiagnoses of sexual health problems by health professionals was also thought to be caused by framing sexuality only in relation to sexual intercourse:

Many health professionals stop after questioning whether you are sexually active. That [if you are not sexually active] does not mean that you do not have any sexual feelings or thoughts. It does not cross their mind that you can be sexually active in your mind and that you can be a sexual person. Everyone experiences sexuality differently. Irrespective whether you are sexually active. (Interview pediatric urologist)

Moreover, health professionals described a need to have greater knowledge about sexual health problems in patients with ARM and HD and possible solutions through, for instance, training or information to be able to diagnose sexual health problems and provide support.

The need to create more attention for sexual health support in general was mentioned by sexologists. Sexologists indicated that they are limited involved in current care pathways and have difficulties in getting more attention for the topic within the health organization/institution (and experienced thus limited *impact*):

For years, I am addressing the importance of developing an organization-wide policy on the topic [sexual health]. I think it will help to integrate the topic into the organization and that you will be less dependent on a few health professionals who have affinity with the topic There are still few health professionals who consider sexual health and would like to give attention to it. It is often seen as "luxurious" medicine. (OFG sexologist)

Phase 2: Translating Needs Into Empowerment Tools

The results of the first phase show patients' need for normalizing the topic, feeling respected and having knowledge and skills. Moreover, the first phase showed health professionals' need for having knowledge, skills, resources (time), and clear arrangements for providing sexual health support and impact (in the case of sexologists) on creating awareness for sexual health and how sexual health could be addressed. Based on these empowerment needs, we decided to develop tangible empowerment tools for patients, their parents, and health professionals. The empowerment tool for patients and patients' parents aimed to educate patients and their parents and stimulate individualized verbal support by increasing patients' help-seeking behavior. Because of patients' preference for digital information—for privacy and easy availability—an online tool was created. In addition, online information was developed for health professionals to obtain more knowledge and competences to discuss the topic, identify problems, and to empower patients by providing support. This information should also provide tools for health organizations to facilitate the integration of sexual health issues within current protocols.

Cocreation sessions were organized to translate the needs from Phase 1 into these empowerment tools by discussing their format and content. The tools "Talking of Sex" and "SeCZ talk" were, respectively, not available for the Dutch context and not applicable for individual patient consultations. In addition, patients indicated that due to feelings of shame group meetings were not their preference. The PLISSIT model was suggested as a useful guideline for discussing sexual health, but the participants found it very generic and lacked specific tips for patients with ARM and HD.

Based on patients' needs, the website for patients should aim to (a) increase knowledge on sexual health and opportunities for self-management; (b) share

experiences; (c) normalize the problems; and (d) provide support consisting of tips and suggestions for contacts. The normal psychosexual development stages were found to be an appropriate basis for structuring the website. A neutral though positive tone was chosen for the texts to prevent being perceived as over-dramatic. Based on health professionals' needs, the digital information for health professionals aimed to (a) increase awareness about the importance of patients' sexual health among health professionals; (b) normalize the topic; and (c) provide tools to facilitate discussion of the topic, to identify problems and to support patients and parents. The information was structured based on the organizational and professional barriers to integrating sexual health as a standard of care. To make the information more accessible outside the hospital setting, we decided—based on the input of the first phase and discussions with the project team—to integrate both tools into a digital information source.

Participants provided different types of input, which complemented each other. Patients mainly addressed the type of sexual problems experienced and the importance of personalization and involvement of specialists outside the hospital such as a pelvic therapist. Health professionals' input was mostly related to the feasibility and practicalities regarding implementation. In developing the materials, a balance between these different types of input was found. For instance, the use of the term “vulva” or “vagina” was discussed. Vulva was the correct term, but vagina was commonly used by most health professionals and familiar to patients. It was decided to use the term “vagina” in line with other related materials. Another example was the solution to advise health professionals to start the conversation at an early age so that patients and their parents become familiar with the topic, for the discussion whether to advise health professionals to exclude parents when the topic is introduced.

Participants found the sessions valuable. All participants felt involved and taken seriously $n = 10$ (100%), and almost all (80%) thought that the sessions led to more insights into the different perspectives and the integration of these perspectives into a better result:

Very meaningful. The interaction of health professionals from different disciplines and experience experts led to an efficient exchange of ideas and perspectives. As a result, decisions on the use of materials for the website could be easily made. (Questionnaire, pediatrician)

Based on the input of Phases 1 and 2, a draft website was developed by a designer and a text writer in collaboration with the researchers. The website addressed the multifactorial issues by giving empowering information for both patients, their parents, and health professionals directed at different development stages, already starting at an early

age. The website consisted of six sub-parts: (a) general information and information for (b) parents, (c) children, (d) adolescents, (e) adults, and (f) health care providers. It was decided to not to add ages to categories (c), (d), and (e), taking into consideration that someone's sexual development can develop differently and giving patients the possibility to decide for themselves which phase is most appropriate to their situation. The general information consisted of an overview of possible sexual problems, relevant health providers, other relevant websites, and a glossary. Gender-specific information was provided for both patients and their parents. Parents' information was based on the stages of normal sexual development and divided into three parts: (a) “normal” sexual development, (b) possible (future) problems related to the disease, and (c) tools to empower their child in coping with these problems. The information for adolescents and adults was structured based on the questions they addressed. Specific information was provided for health professionals to increase awareness and improve their knowledge and conversational skills (based on their needs for psychological empowerment). In addition, information was provided for health providers focused on the aspects “information” and “resources” of structural empowerment. The final Dutch version of the website has been published at www.seksualiteit-arm-zvh.nl. The website will also be translated into English to make it accessible for non-Dutch-speaking patients.

Phase 3: Assessing the Empowerment Tools

The draft website was tested for its expected empowering effect on both patients and health professionals.

Patients' empowerment. In general, patients and patients' parents positively evaluated the website (intelligibility $n = 21/22$ [96%], accuracy $n = 20/22$ [91%], and completeness $n = 18/22$ [82%]). Regarding their empowerment, the patients anticipated that the website will have a positive effect on their knowledge (67%),⁶ on how they will regard sexual health (62%) and whether they feel respected by health professionals (56%). A slight positive effect was expected regarding health literacy (44%) and perceived positive control (44%):

I am very happy that there is—at last—attention for it [sexual health] and I got more clarity on the relation to Hirschsprung disease. Now I understand why I had difficulties with being touched during relationships. It is rather emotional to find out the underlying reason of this problem. (Questionnaire patient)

Patients' parents expected that the website would positively affect their empowerment through greater knowledge (89%), self-efficacy (78%), meaningfulness (78%), health

literacy (67%), feeling respected (63%), and perceived personal control (59%).

Health professionals' empowerment. Although they were in general positive about the website—intelligibility $n = 12/13$ (92%), accuracy $11/13$ (85%), and professional $11/13$ (85%)—health professionals addressed various points of improvement regarding, for instance, reducing the amount of text. With regard to their empowerment, health professionals often indicated “neutral” or “not applicable,” especially with regard to their resources (67%; see Note 5), support (62%), and competences (62%). The number of neutral answers was higher among health professionals working outside the hospital because the work settings (structural empowerment) are probably less applicable to them. In addition, the need for competences is not relevant for sexologists, given that they are specialized in the topic. Diversity in responses were found regarding information (39% agree and 18% disagree) and self-determination (26% agree and 23% disagree). This diversity is probably due to differences in personal and professional characteristics, such as job function. An expected result of the website on health professionals' empowerment was found regarding their meaningfulness (67%), opportunities to learn and grow (62%), and possibly their expected impact on providing sexual health support (46%). Yet, as one of the respondents mentioned, “A website cannot solve all the problems.”

Discussion

This study consisted of a three step process of (a) identifying patients' and health professionals' needs to address sexual health in the context of ARM and HD, (b) the development of an empowerment tool in line with these needs, and (c) an evaluation of the expected outcomes of the tool. In line with the need for more education on sexual health highlighted in previous studies (Carlsson Wincrantz & Wahlberg, 2011; Mellor et al., 2013; Saunamäki et al., 2010), the tool—in the form of a website—provides information for patients, their parents, and health professionals. In contrast to the PLISSIT model—an existing tool which guides the development of interventions by nurses to address patients' sexual problems through one-way communication (Mercer, 2008)—the tool facilitates a collaborative dialogue. The tool can be used both for individual consultation and in patient–health professional interaction.

The use of the empowerment models is thought to assist in bringing about a change by including patients', parents', and health professionals' empowerment needs into the tool and thereby enabled the assessment of the tool's expected impact on their empowerment levels. In addition, this tool adds—in comparison with other tools—to the integration of knowledge on patients', parents', and health professionals'

empowerment needs in a cocreation process. By cocreating the tool, a widely supported website could be developed which contributes to the successful implementation of the tool. The results of this study were also included in the Dutch clinical guidelines of ARM and HD. Addressing the topic in the clinical guidelines will help in anchoring the tool in practice, which has been identified as a barrier for existing tools (van der Stege et al., 2014). In addition, the website was disseminated via patient and professional organizations, relevant websites, protocols of health care organizations, and professionals who acted as an ambassador within health care organizations.

The information need of ARM and HD patients regarding sexual health shown by van den Hondel et al. (2015) was specifically addressed in this study with regard to potential sexual problems and possibilities for self-management and professional support. Besides a lack of knowledge, this study showed that patients were often not comfortable with and able to discuss their sexual health. Patients' reactive attitude was also caused by a strong wish to be “normal,” resonating with the hypothesis of Hartman et al. (2006). Health professionals could empower parents to support their children in having an open conversation on sexual health from an early age. Yet, health professionals still put little attention to patients' sexuality. Sexual health is often seen as a secondary need in which more emphasis is laid to more direct physical outcomes of the disease. This vision on sexual health was reinforced by patients' preference of discussing physical above psychological aspects of their disease, also brought forward by Hughes and Lewinson (2014) in the context of sexual problems among elderly women. In addition, the restricted view patients hold regarding health professionals' responsibility to discuss sexual health, further reinforced this vision. These visions underline that the medical model of care is still prevalent in current health care services. Besides, sexuality was often interpreted by health professionals in a narrow way as “having intercourse.” A more broad view on sexual health, in line with the definition “A state of physical, emotional, mental and social well-being in relation to sexuality; it is not merely the absence of disease, dysfunction or infirmity” of the World Health Organization (2006, p. 5), could be helpful. In addition, health professionals not always felt responsible for patients' sexual health or experienced a lack of competences and time and resources also highlighted in previous studies as barriers to the provision of sexual health support (Dyer & das Nair, 2013; Hautamäki et al., 2007; Ho & Fernández, 2006; Stead et al., 2003; Vermeer et al., 2015). This study specifically addresses the “invisibility” of sexologists within the hospital. Health professionals have been found to have limited knowledge about how to deploy sexologists. In addition, sexologists were seen to have limited impact on putting sexual health on the agenda. This resonates with the

study of Alarcão et al. (2016) who showed that sexology is not seen as a “full-fledged profession but rather as a specialization or a secondary field of action” (p. 1190) due to the historical development of the specialization from different backgrounds—psychologists, physicians, or nurses (Fugl-Meyer & Giami, 2006).

The website developed within this study is expected to increase awareness, fill current knowledge gaps among patients, patients’ parents, and health professionals and to improve their skills. It is expected that this knowledge will broaden stakeholders’ views on sexuality (being more than sexual intercourse; Hordern & Street, 2007), the importance of sexual health for patients’ well-being and normalize the inclusion of topic in patient–health professional interactions (Mellor et al., 2013), which is necessary to overcome restrictive attitudes to sexual health in the context of chronically ill patients (McInnes, 2003). Resources (time and money) are also needed so that health professionals can receive sufficient training (Jaarsma et al., 2011) to provide sexual health support or to integrate such support in current protocols. Previous studies indicate that structural empowerment underlies health professionals’ psychological empowerment (Knol & Van Linge, 2009; Wagner et al., 2010).

Strengths and Limitations

By using various methods, a broad range of patients and health professionals were involved within the study and the findings of each method could be validated. Another strength of this study is the nationwide collaboration of different academic medical centers and patient and professional organizations. A large proportion of the health professionals working with the patient groups in the Netherlands were involved (response rate of 60%).

The difficulty in recruiting patients for the OFG and interviews might be explained by the taboo nature of the

subject. Parents are important stakeholders in the empowerment of patients during childhood. Efforts were undertaken to recruit parents at the data collection in Phase 1, yet no-one applied. Patients’ parents’ perspectives were, however, considered in Phases 2 and 3 and throughout the project via the project group. Furthermore, the experiences and needs of non-Dutch or English-speaking and/or digital-illiterate patients were not taken into account. The tool assumes patients have Information and Communication Technology (ICT) resources and skills, thereby excluding patients with a lower income, education, or literacy and further exacerbating their vulnerability.

Conclusion

Patients and health professionals were found to have limited knowledge and skills to address the sexual health of ARM and HD patients. In addition, patients’ and health professionals’ impeding attitudes toward sexual health, health professionals’ lack of time and sexologists’ limited involvement in the care pathway, hinder their empowerment. By simultaneously educating patients, patients’ parents and health professionals, the empowerment tool—in the form of a website—is expected to change current attitudes to addressing sexual health. Subsequently, the tool is expected to help in legitimizing the topic, which is necessary for the allocation of resources and increasing the power of sexologists within current care pathways. This tool could be used as a format for other disease types.

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Notes

1. The studies of van den Hondel et al. (2015) and Witvliet et al. (2018) showed that among female patients, respectively, 50%/27.3% (anorectal malformations [ARM]) and 53%/50% (Hirschsprung's disease [HD]) experienced sexual dysfunction and 38%/50% (ARM) and 20%/37.5% (HD) sexual distress. Among the male patients, respectively, 13.6%/16% (ARM) and 4.4%/11% (HD) experienced erectile dysfunction.
2. The PLISSIT model has been developed by Jack Annon in 1976 to guide the development of intervention plans used by nurses to address patients' sexual problems (Annon, 1976; Taylor, 2006). The abbreviation stands for Permission, Limited Information, Specific Suggestions and Intensive Therapy. The "(extended) PLISSIT model" has originally been applied.
3. Talking about "sex," an electronic consultation aid developed to address sexual health during consults in primary care (Macdowall et al., 2010).
4. "SeCZ TaLK" has been developed to stimulate communication about sex among young patients with chronic diseases (van der Stege et al., 2010, 2016).
5. The project group consisted of five patient representatives of three patient organizations; 12 health professionals of six different specialisms of the medical centers Erasmus University Medical Center, Radboud University Medical Center, and University Medical Center Utrecht; three researchers of Athena Institute (Vrije Universiteit Amsterdam) who are specialized in transdisciplinary research within the health care context; and two MSc students of Vrije Universiteit Amsterdam. The project group was led by the first author, who is an academic researcher in the field of patient involvement, in collaboration with the last author and a colleague.
6. The percentages of the website, its expected effect on the empowerment of patients, patients' parents, and health professionals, are each based on three questions. In total, 13 patients, nine parents, and 13 health professionals filled in the questions.

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