

Reaching adulthood with Hirschsprung's disease: Patient experiences and recommendations for transitional care

Anders T Hoel ^{a,*}, Louise Tofft ^b, Kristin Bjørnland ^a, Helene Gjone ^c, Catherine J Teig ^d, Tom Øresland ^e, Pernilla Stenström ^b, Marit H Andersen ^f

^a Department of Pediatric Surgery, Oslo University Hospital and Institute of Clinical Medicine, University of Oslo, Oslo, Norway

^b Department of Pediatric Surgery, Skåne University Hospital and Department of Clinical Sciences, Lund University, Malmö, Sweden

^c Department of Child and Adolescent Mental Health in Hospitals, Oslo University Hospital, Oslo, Norway

^d The Pelvic Floor Center, Division of Surgery, Akershus University Hospital, Oslo, Norway

^e The Pelvic Floor Center, Division of Surgery, Akershus University Hospital and Institute of Clinical Medicine, University of Oslo, Oslo, Norway

^f Department of Transplantation Medicine, Oslo University Hospital and Institute of Health and Society, University of Oslo, Oslo, Norway

ARTICLE INFO

Article history:

Received 23 February 2020

Received in revised form 28 April 2020

Accepted 5 May 2020

Keywords:

Experiences
Hirschsprung
Transitional care
In-depth interview
Long-term results
Health literacy

ABSTRACT

Background/Purpose: The need for transitional care has gained increased focus in the treatment of patients with congenital colorectal disorders. We aimed to acquire in-depth knowledge about the experiences of adult patients with Hirschsprung's disease (HD) and their suggestions for transitional care.

Methods: Binational study applying gender equal focus group interviews (FGI).

Results: Seventeen (9 men) of 52 invited patients with median age 29 (19–43) years participated. Three themes evolved from the FGI. "Scarred body and soul" describes the somatic and psychosocial challenges the patients experienced and "limited health literacy on HD" refers to the patients' lack of HD knowledge. "Absent transition" depicts missing transitional care and the patients' inability to find adult HD specialists. The adult HD patients strongly recommended transitional care from early teens with focus on information about HD and establishment of a peer-to-peer program. They also emphasized the possibility of being referred to a pelvic floor center.

Conclusions: HD negatively influences patients' somatic and psychosocial health in childhood, adolescence and adulthood. Adult HD patients strongly recommend transitional care from early teens and the possibility for referral to a center working with pelvic floor dysfunctions.

Level of evidence: IV

Type of research: Clinical

© 2020 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Quality of life can be affected by impaired bowel function in children and adolescents operated for Hirschsprung's disease (HD) [1–4]. Therefore, most pediatric surgical centers operating HD patients have regular multidisciplinary follow-up programs. Until recently, it was assumed that HD related problems disappeared as the patients got older [1,5]. However, some recent reports have shown that a surprisingly high number of adult HD patients struggle with the same problems as HD children and adolescents [6–8]. This has led to an increasing awareness of the need for transitional care for HD patients [9,10]. Health literacy is an important coping strategy for patients with chronic conditions like HD [11]. However, health literacy has not been studied in HD patients previously.

At present, few centers have initiated transitional care programs for HD patients. Furthermore, there are no established guidelines on how

transitional care should be implemented. In order to design appropriate transitional care for HD adolescents, more knowledge about HD related problems in teenagers and adults is needed. Secondly, it is important to know what the patients themselves think a transitional program should include. Thus, in this study the main aim was to acquire in-depth knowledge about how HD had influenced somatic and psychosocial health from childhood to adulthood. We also wanted to explore if HD patients want transitional care and what it should include.

1. Materials and methods

1.1. Study design

A qualitative research-design using focus group interviews (FGI) was chosen to generate a deep understanding of growing up with HD [12,13].

* Corresponding author at: Department of Pediatric Surgery, Oslo University Hospital, Postbox 4950 Nydalen, 0424 Oslo, Norway. Tel.: +47 984 83 047.

E-mail address: andho@ous-hf.no (A.T. Hoel).

Table 1

Interview guide for focus group interviews among adults with Hirschsprung's disease (HD) in Norway.

Areas of interest	Examples of questions
Disease literacy and subjective symptoms	Do you have any questions about HD? Please elaborate on your present bowel function
Disease impact on somatic and psychological aspects of life	Do you have to adjust your daily routines because of your bowel? What emotional concerns have you felt growing up with HD?
Use of health care in adult life	What kind of health providers would you contacted if you had HD related problems? If you have had bowel problems, who helped you?
Relationships and sexuality	Has your bowel influenced relations to friends and partners? Do you think HD has influenced your sexual life?
Follow up of HD in adulthood	How is your present follow-up organized? If we were to design a transition plan, what should it include?

1.2. Clinical context

The study was collaboration between four tertiary referral centers; two centers for pediatric surgery and two pelvic floor centers in Norway and Sweden.

1.3. Study sample

Patients were identified from hospital records. Inclusion criteria were over 18 years, having been treated for HD in childhood at one of the pediatric surgical centers and fluent in either Norwegian or Swedish. In Norway, patients living within a 2-hour drive from the hospital were invited, whereas patients from the whole southern part of Sweden were invited. Patients with intellectual retardation and/or major comorbidities were not invited.

1.4. Data collection

The facilitator in Norway moderated discussions from a list of topics (Table 1), whereas in Sweden fictive case stories were used (Table 2). The FGI were held between January and September 2018 in four gender divided groups of 3–5 patients in each group. The FGI were facilitated by same gender psychiatrists in Norway and by a female psychotherapist in Sweden, all familiar with the methodology.

1.5. Data analysis

The FGI were verbatim transcribed and then analyzed according to Kvale and Brinckmann's principles for qualitative content analysis [14]. First, two of the authors (ATH, KB) read the FGI transcripts to acquire a general impression of their content. Second, the meaning units, representing the most significant in the transcript, were

identified. Third, meaning units discussing similar topics were placed into subcategories prior to reduction into categories with a broader content. Lastly, the categories were reduced to three main themes. The three main themes are abstracted phrases representing the main results of the FGI. Consensus was reached through binational research meetings. Consolidated criteria for reporting qualitative research (COREQ) were applied [15].

1.6. Data quality

A set of qualitative research criteria (i.e. credibility, transferability, dependability, and confirmability) was applied for reaching trustworthiness. Credibility was ensured by review of recent literature to create the research questions, the interview guide and the fictive patient stories, a binational study, researchers with both distance and close relation to the investigated topic, and researchers with extensive experience with qualitative research. The rich, saturated and varied data strengthened the external validity and thereby the transferability. Dependability was ensured by constant comparison back and forth between the data material and the analysis, always based on the patients' unique experiences. Confirmability was reached through the use of triangulation, continuous discussion between the researchers during analysis and consensus meetings to determine results in a methodically translucent audit trail.

1.7. Ethical considerations

The study was approved by the Regional Committee for Medical and Health Research Ethics in both countries (Norway: 2017/1895, Sweden: 2017/867). An invitation letter including information about the study was sent to eligible patients, and written consent was obtained. Oral information was given at the day of the FGI. Anonymity was maintained

Table 2

Condensed fictive patient stories presented to adults with Hirschsprung's disease in Sweden as a base for the focus group interviews.

Girl, 17 years, with bowel emptying problems is invited to go abroad on holiday with friends.
Man, 20 years, gets constipated if sitting too much at work, reluctant to tell his boss.
Man, 20 years, has experienced one episode with impotence, now met a girl, afraid of experiencing impotence again.
Girl, 15 years, needing regular enemas. She is offered to meet an adult colorectal surgeon together with a pediatric surgeon. She does not want this, but her parents do.
Woman, 19 years, needs to empty her bowels 5–6 times daily to avoid fecal incontinence. She is worried about going to bed with a partner.
Man, 18 years, with a scar after previous stoma. He is embarrassed when people ask why he has a scar.

Table 3

Demographics of adults with Hirschsprung's disease taking part in focus group interviews (FGI) in Norway and Sweden.

	Total	Norway	Sweden
Included/invited patients	17/52 (33%)	10/23 (43%)	7/29 (24%)
Age, median (min-max), years	29 (19 – 43)	39 (19–43)	27 (24–30)
Gender (male/female)	9/8	5/5	4/3
Duration of FGI median, (min-max) minutes	85 (56 – 105)	88 (72 – 105)	77 (56 – 98)
Patients with appendicostomy	1	1	0

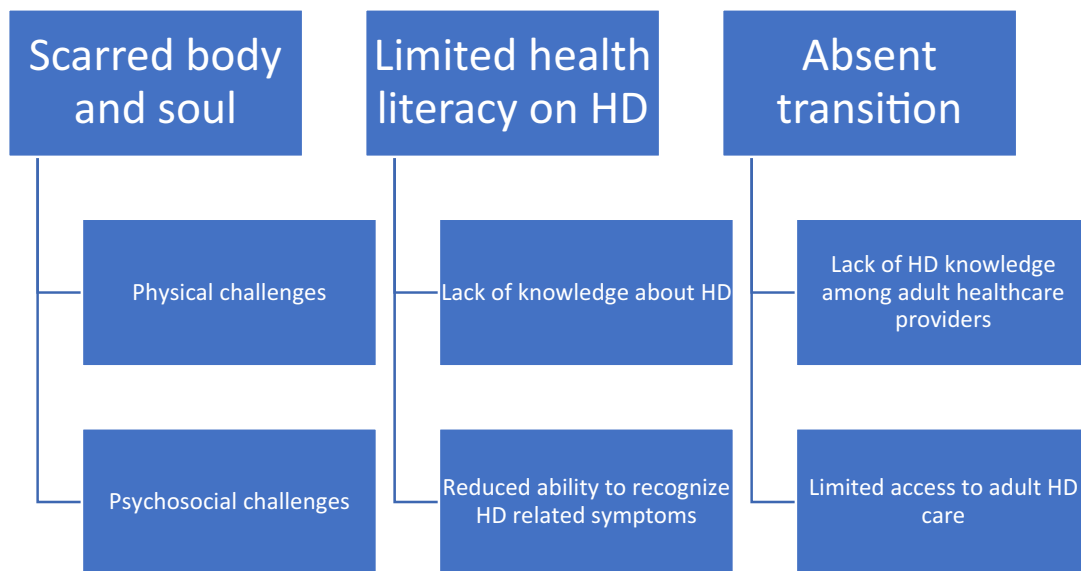


Fig. 1. Themes and categories emerging from focus group interviews with adult Hirschsprung's disease (HD) patients.

throughout the FGI by a personalized identification number in Norway. In Sweden, the principal investigator linked the individual patient to the verbatim transcript. The patients were offered individual medical or psychological support outside the study if needed.

2. Results

2.1. Patient characteristics

Seventeen (nine men) of 52 invited patients were included (Table 3). All, but one patient, were operated with the Duhamel technique. One patient had total colonic aganglionosis, whereas the others had aganglionosis in varying lengths of the colon.

2.2. Themes

Three major themes evolved from the FGI (Fig. 1). The first theme “Scarred body and soul” describes how HD affects somatic and psychosocial health. The second theme “Limited health literacy on HD” refers to the patients’ lack of knowledge about HD and how persisting gastrointestinal symptoms may be related to HD. The last theme “Absent transition” refers to the patients’ inability to find health providers with knowledge and understanding of possible long-term effects of HD.

2.3. Theme 1: Scarred body and soul

All patients had experienced a negative influence of HD on physical and psychosocial well-being during from adolescence and adulthood.

Table 4 describes how the major theme “Scarred body and soul” evolved through the audit trail.

2.3.1. Physical challenges

2.3.1.1. Impaired bowel function. All patients reported varying degrees of bowel emptying problems and fecal incontinence, and the majority had experienced more severe problems when they were younger. They had learned through trial and error to manage their bowel problems. Present bowel dysfunctions varied from minor interference with everyday life to daily time-consuming bowel emptying regimens. Moreover, all patients reported both previous and present episodes of painful bowel movements. It seemed to be easier for the men to tell about their bowel problems. The men used humor when talking about impaired bowel function, whereas the women were more reluctant to discuss this topic and had prolonged pauses during the FGI.

“It may hurt to poop; it can be diarrhea with painful contractions.”

2.3.1.2. Food intolerance and abdominal discomfort. All patients struggled with various degrees of abdominal discomfort. They struggled constantly to find food that would reduce flatulence, constipation or diarrhea or simply “calm down the stomach”. They were unsure if this was related to HD or if they had any food intolerances. In line with this, the patients pointed out that all HD patients should see a dietician (Table 5).

Table 4

Example of content analysis of the major theme “Scarred body and soul” through an audit trail from focus group interviews with adult Hirschsprung's disease (HD) patients.

Meaning unit	Subcategory	Category	Theme
“I don't think I have had a single day with normal stool. It's either diarrhea or constipation. So, my diet always includes potato flour, lactulose and prunes.”	Impaired bowel function	Physical challenges	Scarred body and soul
“I don't know if it is related to my condition or not, but typically greasy food make my stomach bloat and hurt.”	Food intolerances and abdominal discomfort		
“I have some scars. I have always preferred to cover them, never liked to wear a bikini.”	Scars		
“I have friends who still, today, do not know about my condition and that I have a stoma. I have even had boyfriends who didn't know.”	Emotional effects of impaired bowel function	Psychosocial challenges	
“HD has always held me back sexually. I never tell partners straight away, it takes time.”	Sex and reproductive health		
“I feel I am walking around in the world alone, looking for answers.”	Loneliness		

Table 5

Adult Hirschsprung's disease (HD) patient's recommendations for transitional care.

Domains	Challenges	Recommendations
Physical	Impaired bowel function Food intolerances Embarrassing scars	Support for optimal bowel function Information from a dietician Address scars
Psychosocial	Emotional effects of impaired bowel function Sexuality and fertility Feelings of loneliness	Psychological support to learn coping strategies Address sex and fertility Peer-to-peer program
Educational	Limited health literacy on HD	Strengthen the patient's ability to get access to, understand and use health information about own medical history and possible long-term effects of HD from early teens
Transitional	Absence of transition	Consultations partly without parents Transition should start in early teens Establish contact with adult multidisciplinary team with HD expertise

"As a child I had a lot of stomachache. I grew up not eating meat, fruit, vegetables, berries, and fish. What was left was literally just grains, oat, cheese, and dairy products."

2.3.1.3. Scars. During childhood and adolescence, all patients had been embarrassed of their scars, and females expressed more concerns than the males. The scars were a constant reminder of HD. Some had consulted plastic surgeons, and a few had undergone scar revisions. Patients advised pediatric surgeons to discuss scars during follow-up (Table 5).

"I've had my scar corrected. Before the scar was fixed, I always wore a bathing suit, never a bikini. When I look at photos of myself from childhood, I always cover my stomach with my hands."

2.3.2. Psychosocial challenges

2.3.2.1. Emotional effects of impaired bowel function. Feelings of being different and a desire to keep HD secret were common. Fecal incontinence, excessive noise during toilet visits and need for extra time at the toilet caused both stress and shame. Some reported bullying from classmates because of their ostomy or abnormal bowel habits.

"I have sent partners out of the house when I had to use the bathroom."

"There were comments at school when I was younger. One boy said I should taste my own poop, since I had it in a bag outside my body."

The patients emphasized the invaluable emotional support from their mothers, stoma nurses and psychosocial health providers (Table 5). Those with family members with HD had less feelings of being the only one with bowel problems. All patients thought that psychosocial support during follow up should be mandatory (Table 5).

2.3.2.2. Sex and reproductive health. Sex issues were discussed differently by the men and the women. Generally, the men expressed good sexual self-esteem and few problems with intimate relationships and fertility. The women were more reluctant to discuss intimacy, had more problems with sexual relationships and were concerned about telling partners about HD. The females who had experienced problems to conceive, wondered if the infertility was related to HD. As a consequence, especially female patients would like pediatric surgeons to discuss sex and fertility during follow-up (Table 5).

"I needed IVF to conceive. In my medical records, the reason for my infertility is regarded unknown. But I think there must be a correlation with my previous operations. My tubes are clogged."

2.3.2.3. Loneliness. The patients valued highly the opportunity to meet other HD adults during the FGI. Most did not know anyone with HD and discussed HD related experiences with other patients for the first time during the FGI. They would have liked to have known others with HD during childhood and adolescence for two reasons; feel less abnormal and to get helpful advice. Therefore, the patients suggested establishment of a peer-to-peer program from adolescence (Table 5).

"You feel a bit alienated since it's a rare disease. So, it's good to meet others who know what you have gone through (bowel problems)."

2.4. Theme 2: Limited health literacy on HD

Our findings indicate that the patients had limited health literacy concerning HD. Consequently, it was difficult for them to interpret symptoms and to seek appropriate medical assistance.

2.4.1. Lack of knowledge about HD

The patients knew little about HD pathophysiology, including heredity and possible long-term effects. They said that during follow up, the doctors had mostly talked to their parents. Most patients did not know whom to consult about possible HD related problems and would contact the pediatric surgical department even as adults. Few patients knew that pelvic floor centers existed. The general impression was that the patients thought that there was no treatment for their problems, and consequently few had been referred to any specialist. The patients recommended age-adapted information about HD (Table 5). Furthermore, they thought HD teenagers should talk with the pediatric surgeon alone so that issues like sexuality could be brought up (Table 5).

"It's today, in the FGI, that I understand what HD is."

"I wondered if I would function normally during sex. I did not dare ask anyone, but luckily I found out I was perfectly normal"

2.4.2. Reduced ability to recognize HD related symptoms

Most of the patients did not think of themselves as "patients". Since the diseased part of the large bowel was removed, they considered themselves cured of HD. Nevertheless, the patients reported several HD related problems, such as abdominal pain, food intolerances and infertility. Some wondered if this could be possible long term effects of HD (Table 5).

"I've always thought you get rid of the disease after the operation."

2.5. Theme 3: Absent transition

Overall, the patients found adult healthcare providers ignorant of HD and possible long-term effects. When HD related problems occurred, they had limited access to healthcare providers with experience of HD.

2.5.1. Lack of HD knowledge among adult healthcare providers

All patients reported lack of knowledge about HD outside the pediatric surgical departments. It seemed that the patients' feeling of being the only one with a rare disease was reinforced by ignorant adult healthcare providers. Consequently, the patients did not find it worthwhile to contact local doctors for their HD related problems. The patients emphasized the need for adult healthcare providers with sufficient knowledge on HD long term effects (Table 5).

"You come to a general practitioner, and he is fascinated when hearing about HD, but doesn't know much. But he finds it exciting."

2.5.2. Limited access to adult HD care

At the time of the FGI only one patient had planned follow-up for HD. When asked who could be of help, they listed nutritionist, gynecologist, obstetrician, colorectal surgeon, stoma nurse, psychologist, plastic surgeon, social worker, and sexologist (Table 5). The FGI revealed a uniform agreement that a pelvic floor center was the ideal referral center for adults with HD related problems (Table 5). Regular follow-up as adults were not regarded necessary since most by now had learnt to manage their problems.

"I wanted to get my stomach checked because I had so much stomach pain. I went to the general practitioner, and then he referred me to one specialist. Then I got referred to another specialist, and I went back and forth. In the end I gave up, could not stand being sent everywhere".

3. Discussion

This study shows that HD continues to negatively influence both physical and psychosocial health in adulthood and is the first to have applied qualitative research methods to obtain in-depth knowledge of patient experiences. Furthermore, the HD patients exhibited limited health literacy on HD. The patients suggested transitional care from early teens to increase health literacy and knowledge of multidisciplinary expert centers they could contact if HD related problems occurred.

3.1. Physical challenges

The term "scarred body" was chosen to summarize the persisting bodily challenges reported by adult HD patients. They described a variety of symptoms such as bowel emptying problems, food intolerances and embarrassing scars.

Previous studies have reported fecal incontinence and obstructive symptoms in HD adults, but these studies have mainly applied questionnaires [1,5,16,17]. The FGI revealed new findings such as abdominal pain, excessive flatulence and troublesome toilet visits. Furthermore, the patients reported various food intolerances, and they expressed a frustrating search for a suitable diet. The few studies on food intolerances and HD have similar findings as our study [1,18].

Not surprisingly, the female patients reported more concerns about abdominal scars than the men. Problems related to scars have not been reported earlier in HD literature. Even though one-stage minimally invasive HD pull-through operations are the most common techniques now, a significant number still get an ostomy prior to the pull-through, and some are still operated with a laparotomy [19,20].

Pediatric surgeons should be aware of this and address the possibility of scar revisions if that is indicated.

3.2. Psychosocial challenges

"Scarred soul" embraces psychosocial challenges HD patients experience in adolescence and adult life from impaired bowel function and intimate relationships. Emotional effects of impaired bowel function include feelings of being different, loneliness and difficult intimate relationships. This important finding has not been addressed earlier. Supporting our findings is a Swedish national population-based study showing an increased risk of depression in HD patients compared with controls [21].

Another important finding from the FGI is that the females reported more psychosocial challenges than the men when it came to sexual function. This observation is in line with studies in patients with inflammatory bowel disease and diabetes showing that female sex is a mental health risk factor [22,23]. Furthermore, a study on 329 healthy women found a significant association between sexual dysfunction and surgical scars [24]. Fertility has to a low degree been investigated in the adult HD population. Thus, our observations on fertility suggest that health providers should have extra focus on female HD patients during transition and in adult care.

3.3. Limited health literacy on HD

We found overall limited health literacy on HD among the patients. Health literacy has not been studied in HD patients previously [25]. Our finding is not surprising, since adolescents with a chronic disease are prone to hold reduced health literacy [11,26]. The patients' recommendation of planned transition with focus on age adapted information about HD, is supported in literature to increase health literacy [9,27].

3.4. Patients' recommendations for transitional care

This study provides novel knowledge on how transitional care for HD patients may be designed through firsthand experiences and recommendations from adult HD patients. The patients had firm opinions of what they had missed as adolescents and now as adults. Although several papers suggest transitional care for HD patients, none has suggested guidelines for transitional care [5,8,10,28]. Patients with anorectal malformations struggle with many of the same problems as HD patients, and the ARM-net consortium has proposed guidelines for transitional care [29]. Our patients' recommendations for transitional care are almost identical to that of the ARM-net consortium.

3.5. Strengths and limitations

Strengths of this study were inclusion from two countries and the use of consolidated criteria for reporting qualitative research (COREQ). Since patients' symptoms and problems often are underestimated by clinicians, the use of FGI strengthen our study due to the invaluable self-reported data [30]. We have limited clinical data, and some may regard this as a limitation. Like other FGI studies, less than half of invited patients participated [27,31]. This may represent a selection bias.

4. Conclusion

This study finds that HD negatively influences both physical and psychosocial health in adolescence and adulthood. To reduce negative long term effects of HD, adult HD patients strongly recommend transitional care with focus on age adapted information on HD and contact with pelvic floor centers.

Acknowledgements

We would like to thank each patient who so openly shared his or her experiences growing up with this rare congenital colorectal disease. We also acknowledge Svein Staff's facilitation during the FGI.

Conflicts of interest statement

The authors declare no conflict of interest.

Funding

This study was funded by the Dam Foundation.

References

- [1] Bjornland K, Pakarinen MP, Stenstrom P, et al. A Nordic multicenter survey of long-term bowel function after transanal endorectal pull-through in 200 patients with rectosigmoid Hirschsprung disease. *J Pediatr Surg*. 2017;52(9):1458–64.
- [2] Collins L, Collis B, Trajanovska M, et al. Quality of life outcomes in children with Hirschsprung disease. *J Pediatr Surg*. 2017;52(12):2006–10.
- [3] Lane VA, Nacion KM, Cooper JN, et al. Determinants of quality of life in children with colorectal diseases. *J Pediatr Surg*. 2016;51(11):1843–50.
- [4] Neuvonen MI, Kyrklund K, Rintala RJ, et al. Bowel Function and Quality of Life After Transanal Endorectal Pull-through for Hirschsprung Disease: Controlled Outcomes up to Adulthood. *Ann Surg*. 2017;265(3):622–9.
- [5] Meinds RJ, van der Steeg AFW, Sloots CEJ, et al. Long-term functional outcomes and quality of life in patients with Hirschsprung's disease. *Br J Surg*. 2019;106(4):499–507.
- [6] Granstrom AL, Danielson J, Husberg B, et al. Adult outcomes after surgery for Hirschsprung's disease: Evaluation of bowel function and quality of life. *J Pediatr Surg*. 2015;50(11):1865–9.
- [7] Ieiri S, Nakatsuji T, Akiyoshi J, et al. Long-term outcomes and the quality of life of Hirschsprung disease in adolescents who have reached 18 years or older—a 47-year single-institute experience. *J Pediatr Surg*. 2010;45(12):2398–402.
- [8] Rintala RJ, Pakarinen MP. Long-term outcomes of Hirschsprung's disease. *Semin Pediatr Surg*. 2012;21(4):336–43.
- [9] Harrington AW, Gasior AC, Einarisdottir H, et al. Hirschsprung Disease: The Rise of Structured Transition and Long-term Care. *J Pediatr Gastroenterol Nutr*. 2019;69(3):306–9.
- [10] Muise ED, Cowles RA. Transition of care in pediatric surgical patients with complex gastrointestinal disease. *Semin Pediatr Surg*. 2015;24(2):65–8.
- [11] Mackey LM, Doody C, Werner EL, et al. Self-Management Skills in Chronic Disease Management: What Role Does Health Literacy Have? *Med Decis Making*. 2016;36(6):741–59.
- [12] Krueger RA, Casey MA. Focus groups : a practical guide for applied research. 5th ed. Los Angeles: Sage; 2015.
- [13] Rabiee F. Focus-group interview and data analysis. *Proc Nutr Soc*. 2004;63(4):655–60.
- [14] Brinkmann S, Kvale S. *InterViews : learning the craft of qualitative research interviewing*. 3rd ed. ed. Thousand Oaks, Calif: Sage; 2015.
- [15] Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care*. 2007;19(6):349–57.
- [16] Kyrklund K, Neuvonen MI, Pakarinen MP, et al. Social Morbidity in Relation to Bowel Functional Outcomes and Quality of Life in Anorectal Malformations and Hirschsprung's Disease. *Eur J Pediatr Surg*. 2018;28(6):522–8.
- [17] Tran VQ, Mahler T, Dassonville M, et al. Long-Term Outcomes and Quality of Life in Patients after Soave Pull-Through Operation for Hirschsprung's Disease: An Observational Retrospective Study. *Eur J Pediatr Surg*. 2018;28(5):445–54.
- [18] Catto-Smith AG, Trajanovska M, Taylor RG. Long-term continence after surgery for Hirschsprung's disease. *J Gastroenterol Hepatol*. 2007;22(12):2273–82.
- [19] Bradnock TJ, Knight M, Kenny S, et al. The use of stomas in the early management of Hirschsprung disease: Findings of a national, prospective cohort study. *Journal of pediatric surgery*. 2017;52(9):1451–7.
- [20] Menezes M, Corbally M, Puri P. Long-term results of bowel function after treatment for Hirschsprung's disease: a 29-year review. *Pediatr Surg Int*. 2006;22(12):987–90.
- [21] Amin L, Skoglund C, Wester T, et al. Swedish national population-based study shows an increased risk of depression among patients with Hirschsprung disease. *Acta paediatr*. 2019;108(10):1867–70. <https://doi.org/10.1111/apa.14801>.
- [22] Byrne G, Rosenfeld G, Leung Y, et al. Prevalence of Anxiety and Depression in Patients with Inflammatory Bowel Disease. *Can J Gastroenterol Hepatol*. 2017;2017:6496727.
- [23] Robinson DJ, Coons M, Haensel H, et al. Diabetes and Mental Health. *Can J Diabetes*. 2018;42(Suppl. 1):S130–s41.
- [24] Cihan A, Cihan E. Interrelation Between Appearance Anxiety and Sexual Functions in Women: The Role of Surgical Scars, Morphologic Features, and Accompanying Depression. *J Sex Med*. 2019;16(11):1769–78.
- [25] Institute of Medicine Committee on Health L. In: Nielsen-Bohlman L, Panzer AM, Kindig DA, editors. *Health Literacy: A Prescription to End Confusion*. Washington (DC): National Academies Press (US) Copyright 2004 by the National Academy of Sciences. All rights reserved; 2004.
- [26] Heijmans M, Waverijn G, Rademakers J, et al. Functional, communicative and critical health literacy of chronic disease patients and their importance for self-management. *Patient Educ Couns*. 2015;98(1):41–8.
- [27] Nah SA, Ong CCP, Lie D, et al. Understanding Experiences of Youth Growing Up with Anorectal Malformation or Hirschsprung's Disease to Inform Transition Care: A Qualitative In-Depth Interview Study. *Eur J Pediatr Surg*. 2018;28(1):67–74.
- [28] Cairo SB, Chiu PPL, Dasgupta R, et al. Transitions in care from pediatric to adult general surgery: Evaluating an unmet need for patients with anorectal malformation and Hirschsprung disease. *J Pediatr Surg*. 2018;53(8):1566–72.
- [29] Giuliani S, Grano C, Aminoff D, et al. Transition of care in patients with anorectal malformations: Consensus by the ARM-net consortium. *J Pediatr Surg*. 2017;52(11):1866–72.
- [30] Basch E. The missing voice of patients in drug-safety reporting. *N Engl J Med*. 2010;362(10):865–9.
- [31] Aasbo G, Solbraekke KN, Waller J, et al. Perspectives of non-attenders for cervical cancer screening in Norway: a qualitative focus group study. *BMJ Open*. 2019;9(8):e029505.