

**EVALUATION OF A GROUP-BASED PATIENT EDUCATION PROGRAM
PROMOTING SELF-MANAGEMENT IN ADULTS
WITH HIRSCHSPRUNG DISEASE AND ANORECTAL MALFORMATIONS**

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ABSTRACT

Introduction

Adults with Hirschsprung disease (HD) and anorectal malformations (ARM) may experience persisting and new somatic and psychosocial problems. Patient education programs (PEPs) may improve self-management in patients with chronic illnesses. The aim of this study was to explore HD and ARM adults' experiences with and evaluation of a group-based PEP. We also looked at factors that might influence the attendance rate.

Method

Non-intellectually impaired HD and ARM adults were invited to attend a diagnosis specific PEP at a pelvic floor interdisciplinary center. Eight health care professionals lectured. Aspects of the PEP were graded anonymously in a patient reported experience measure (PREM). Ethical approval was obtained.

Results

17% (21/125) of invited adults (10HD, 11ARM) attended four PEPs. 19/21 (90%) PREMs were returned. Participants found meeting peers and sharing experiences especially valuable in addition to improved disease knowledge. Lectures by the pediatric and colorectal surgeons, stoma nurse, and sexologist were rated highest by the participants. The majority reported that the PEP would be helpful in managing everyday life. All participants recommended PEP in adolescence. Factors such as gender and travel distance did not affect attendance rate, but participants were older than non-participants, median 37 versus 24 years ($p=0.01$).

Conclusion

Attendance rates were low among HD and ARM adults invited to a PEP, but participants were overall highly satisfied. Peer support, mutual learning, and increased disease knowledge were seen as invaluable assets of the PEP. A web-based PEP was discouraged, while physical PEPs for both adults and adolescents were encouraged.

Keywords

Hirschsprung disease, anorectal malformations, patient education program, self-management, transitional care

Level of evidence

III

Abbreviations

ARM: Anorectal malformations

HD: Hirschsprung disease

PEP: Patient education program

PREM: Patient reported experience measure

QoL: Quality of life

1. Introduction

Hirschsprung disease (HD) and anorectal malformations (ARM) are rare congenital colorectal diseases, each affecting around 1 in 5000 births. HD is characterized by lack of ganglion cells in the distal bowel resulting in bowel obstruction, while ARM consists of a wide spectrum of malformations of the anus ranging from minimal to large changes of the normal anatomy of the anorectal region [1, 2]. Most patients are operated during the first year of life. Many children with HD and ARM have impaired bowel function and reduced quality of life (QoL), but improvements of both somatic and psychosocial function are common when they reach adolescence and adulthood [3-6]. In spite of this, there is a growing recognition that HD and ARM adults may have persisting problems related to bowel function and psychosocial health [7, 8]. In addition, new problems such as sexual dysfunction and infertility may occur [9-12].

Patient education programs (PEPs) aim at promoting self-management for people with a chronic illness. PEPs may vary greatly in content and form. Common goals are to increase skills related to problem solving and decision making regarding own health, knowing where to find help, and forming a patient-health care provider partnership [13]. A recent scoping review of papers including 7003 adults with various chronic illnesses found that participants in group-based PEPs experienced peer support, learning and hope, less symptom distress, greater awareness of own health, and improved self-management [14]. To our knowledge, no previous study has reported experiences with a group-based PEP designed for HD and ARM adults. The aim of this study was, therefore, to explore HD and ARM adults' experiences with and evaluation of a group-based PEP. We also looked at factors that might influence the attendance rate.

2. Methods

2.1 Study design, patients, clinical setting and data collection

This is a cross sectional study utilizing a patient reported experience measure (PREM) in HD and ARM adults' participating in a group-based PEP. Guidelines for Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) were applied [15].

Non-intellectually impaired adults (>18 years), fluent in Norwegian, having undergone surgery for HD or ARM as children at Oslo University Hospital were eligible for inclusion. Intellectual impairment was defined as mental retardation with or without a syndrome which would hinder participation in a group setting. Potential participants were identified from

surgical logbooks and electronic medical journals, and invited by letter to attend diagnosis specific and gender mixed one-day PEPs at a pelvic floor interdisciplinary center. The participants paid the same deductible (35€) as if they were to see a medical specialist. Travel, lunch and accommodation expenses as well as sick leave were covered by the national health care system.

Demographics and length of the aganglionic segment or type of anorectal malformation were recorded from medical records of the participants. Travel distance, between the registered address of all potential participants and the pelvic floor interdisciplinary center, was calculated in a direct line using a digital map. In addition to travel distance, age and gender were recorded for all potential participants to look at factors that might influence the attendance rate between participants and non-participants. Due to Covid-19 restrictions, one PEP had to be web-based on short notice and travel distance was therefore not excluded for these participants. The web-based PEP was organized as similar as possible as the physical PEPs with the same structure and lectures. Those who had accepted the web-based PEP were sent a Whereby-link to log on to the encrypted web-based platform.

Oslo University Hospital has both local-, regional-, and national functions serving approximately 3 million people. Akershus University Hospital, situated 20 minutes by car from Oslo University Hospital, serves a population of 0.6 million people and has a pelvic floor interdisciplinary center. The pelvic floor interdisciplinary center has a multidisciplinary team which coordinates investigations and treatment of adults with complex diseases of the pelvic floor and/or pelvic organs. In addition, the pelvic floor interdisciplinary center offers PEPs to patients with pelvic floor disorders.

Eight health care professionals lectured during the PEPs held in 2019 and 2021 at the pelvic floor interdisciplinary center; pediatric surgeon, adult colorectal surgeon, stoma nurse, psychiatrist, sexologist, physiotherapist, nutritionist, social worker (Table 1). The participants were encouraged to ask questions and comment during the PEP, and it was emphasized that everything said was confidential.

At the end of the PEPs the participants were handed a self-designed PREM with 11 questions regarding their experiences with and evaluation of the PEP (Appendix 1). The PREM was answered anonymously. The different questions were answered with text, dichotomously, or

graded on a 4-point Likert scale. For those who attended a web-based PEP, the PREM contained 12 questions and was mailed to them and returned in a stamped envelope.

2.2 Statistics

Continuous variables are presented as median (min-max) and were analyzed with Mann Whitney U test. Statistical significance was set at $p < 0.05$. IBM SPSS software for Windows version 25 (Armonk, NY: IBM Corp.) was used.

2.3 Ethics

Written informed consent was obtained from all participants. The study was approved by the regional committee for medical and health research ethics (2017/1895) and Akershus University Hospital's Data Protection Officer (2018-008-1).

3. Results

3.1 Participants

125 adults (64 HD, 61 ARM) were invited, and 28 (22%) (12 HD, 16 ARM) signed up for the PEPs. 21/28 (75%) of those who had accepted the PEP invitation participated (10 HD, 11 ARM) (Table 2). Thus, 17% (21/125) of the invited adults participated at a PEP. Four PEPs, two for HD and two for ARM participants were held. One PEP for ARM participants in 2021 was web-based due to Covid-19 restrictions, whereas the other three PEPs were arranged at the pelvis floor interdisciplinary center at Akershus University Hospital. Median 5 (4-7) participants attended each PEP. The HD participants had aganglionosis confined to rectosigmoid colon (3), proximal to rectosigmoid colon (2), and unknown (5). The ARM participants had been treated for perineal (4), recto urethral (3), recto vestibular (1) and unknown (3) malformations.

3.2 Experiences with and evaluation of the patient education program

19/21 (90%) questionnaires were returned. All participants highlighted that the most valuable aspect of the PEPs was to meet peers and to share experiences with them (Table 3). Most participants met peers for the first time, and realized that they were not alone having HD or ARM. Furthermore, the participants appreciated the information they received about their diagnosis. Several participants reported that what they had learnt during the PEPs had made

them understand their own body better and that symptoms they had, could be related to HD or ARM.

The HD and ARM participants evaluated the lectures relatively similar. Overall, they were most satisfied with the lectures by the pediatric and colorectal surgeons, the stoma nurse, and the sexologist (Table 3). The HD participants rated information about their diagnosis and nutrition higher than the ARM participants. The ARM participants, in contrast, found information from the social worker more important than the HD participants.

17/19 (89%) reported that the PEP would help them to a large or very large extent to manage everyday life. The majority, 16/19 (84%), found the size of the group appropriate to share experiences and ask questions. 14/19 (74%) participants were either satisfied or very satisfied with talking about sex with both genders present. In line with that, 17/19 (89%) participants were either positive or neutral about gender mixed groups in a future PEP. 17/19 (89%) participants reported that they did not miss any topic in the PEP. All participants thought they would have benefited from an age-adapted PEP in their teens.

3.3 Factors that might influence attendance rate

Gender and travel distance did not seem to influence the attendance rate. Participants were older than those who chose not to attend the PEP (Table 4).

3.4 Evaluation of a web-based patient education program

Four of the eight adults who had signed up to attend the web-based PEP for ARM adults, did not log on. Furthermore, the only two participants who did not return the questionnaire, were attending the web-based PEP. The two ARM participants who returned the questionnaire, were unsatisfied with the PEP. As for those who attended the PEP at the pelvic floor interdisciplinary center, they highly valued meeting peers (median 4), but rated sharing experiences lower (median 3) than those participating physical PEPs (median 4).

3.5 Referrals to the pelvic floor interdisciplinary center after the patient education program

3/21 (14%) participants were referred to the pelvic floor interdisciplinary center after the PEPs. All three had ARM, leaving 3/11 (27%) ARM participants in need of a referral. These three participants required treatment for fecal incontinence, constipation, and anal prolapse.

4. Discussion

The main finding of this study is that few of invited HD and ARM adults chose to attend a group-based PEP. However, those who participated, were overall very satisfied with the PEP. Meeting peers, sharing experiences, and getting a better understanding of their congenital disease were rated as the most valuable assets of the PEP.

This is the first study where HD and ARM adults' evaluation of a group-based PEP is presented. As in a recent review of 7003 adults, we found that many of the participants met others with the same disease and facing similar problems for the first time [14]. The feeling of “togetherness” and not being the only one with a rare disease were reported as an immediate beneficial experience of PEPs [16-18]. The participants reported that sharing experiences with peers were one of the most important parts of the PEP. Mutual learning among peers makes participants feel better and strengthens their confidence and identity [16, 19]. In addition, the majority of participants reported that new knowledge they had received through the lectures had made them aware that symptoms they had, might be related to HD or ARM. That new knowledge obtained during a PEP may lead to improved self-management skills, has also been described previously [17, 20-23].

Only 17% of the invited HD and ARM adults participated at the PEP, and the rate of no-show was 25%. How many of invited patients that attend a PEP, is rarely reported [14, 24, 25]. For patients with chronic obstructive pulmonary disease and diabetes, attendance rates at PEPs vary a lot, from 7-92% [26, 27]. Since there is limited information on how patients are recruited to PEPs, it is difficult to compare our results with those of others. Nevertheless, it seems that low attendance rates are common [26-28]. Whether this means that those who did not attend, would not have benefitted from attending a PEP or that other reasons kept them from attending, is impossible to know. In our study, travel distance and gender did not affect attendance rate. In other studies, travel distance is a common barrier for attending a PEP [28]. A systematic review studying why patients with diabetes chose not to attend PEPs, found that non-participants were predominantly male [26]. This same systematic review found that non-participants were younger than participants, which is in line with our study. This could imply that patients with a chronic illness get increasingly interested in topics discussed at PEPs as they get older. On the other hand, it could also mean that older patients overall have got less information and poorer follow-up than younger patients. Since the evaluations of the PEP in

this study were overall very positive, we are concerned that many more than those who came, would have profited from taking part. An important result from this study is that recruiting patients to a PEP is challenging. A recent review assessing effects of PEPs on self-management and empowerment, found that all PEPs resulted in some form of improvement for the participants [14]. Thus, it seems that making efforts to make patients attend PEPs is important to promote health and self-management in patients with chronic illnesses. Measures to increase attendance rates of HD and ARM adults participating in PEPs may include early information about PEPs to HD and ARM adolescents in transitional care clinics, information to HD and ARM patients attending adult clinics (eg. pelvic floor interdisciplinary center), and advertisement through national HD and ARM patient organizations. We have noted that an invitation letter including that previous participants have given positive ratings, a detailed program, “first come, first served”-principle for registration through a QR-code to a web-based form, and contact information (phone and e-mail) to the PEP organizer, have improved the attendance rates.

The web-based PEP had lower attendance rate than the physical PEPs, and fewer participants returned the questionnaire after the web-based PEP. Most importantly, ratings of the web-based PEP were much poorer than of the physical PEPs. It has been suggested that web-based PEPs may improve accessibility for patients living in rural areas, while others find it difficult to know when web-based PEPs are preferable to physical meetings [29, 30]. The feedback from the participants in this study was that a web-based PEP was unsuitable for a group-based PEP like this. A plausible explanation is that topics such as fecal and urinary incontinence and problems with intimacy, sexuality, and fertility are too sensitive to be discussed on a web-based platform with strangers.

All HD and ARM adults attending the PEP thought that they would have benefitted from a PEP in their teens. This corresponds with findings from our previous study where HD and ARM adults discussed transitional care during focus group interviews [31, 32]. Even though there is limited evidence of the effectiveness of PEPs for adolescents with chronic illnesses, our findings encourage arrangement of PEPs for HD and ARM adolescents [33-35].

The main strengths of this study are that it was conducted by a multidisciplinary team experienced in arranging PEPs, transparency of the recruitment process, and a high response rate for those participating. Participation only costed a small fee, and travel and

accommodation expenses were covered. The participants also got a sick leave to avoid loss of income. All these measures were taken to avoid that the invited adults did not want to attend the PEPs. It is a limitation that the study has few participants and an overrepresentation of participants with poor functional outcome (Table 2). That we used a self-designed questionnaire to assess participant satisfaction did not strengthen the study.

In conclusion, attendance rates were low among HD and ARM adults invited to a group-based PEP aimed to promote self-management. Importantly, the participants who did attend, were highly satisfied with the PEP. Support from peers, mutual learning and increased disease knowledge were seen as invaluable assets of the PEP. A web-based PEP was discouraged, while PEP for adolescents was encouraged.

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Conflict of interest

The authors report no conflict of interest.

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Figure legends

Appendix 1. Questionnaire evaluating a patient education program for adults with Hirschsprung disease or anorectal malformations.

Table 1. Overview of the patient education program (PEP) for adults with Hirschsprung disease (HD) and anorectal malformations (ARM) with allocated time for each lecturing health care professional and which themes that were covered by them.

Table 2. Demographics of adults with Hirschsprung disease (HD) and anorectal malformations (ARM) participating in a patient education program.

Table 3. The evaluation of a patient education program (PEP) for adults with Hirschsprung disease (HD) and anorectal malformations (ARM) graded on a 4-point Likert scale where one was “unsatisfied” and four “very satisfied”.

Table 4. Factors that might influence attendance rate at a patient education program for adults with Hirschsprung disease or anorectal malformations.

Appendix 1. Questionnaire evaluating a patient education program for adults with Hirschsprung disease or anorectal malformations.

1. How satisfied were you with the patient education program to improve your knowledge on living with Hirschsprung disease/anorectal malformation?

Unsatisfied			Very satisfied
1	2	3	4

2. How satisfied were you with these aspects of the patient education program?

	Unsatisfied			Very satisfied
a) Meeting peers	1	2	3	4
b) Sharing experiences	1	2	3	4

3. How satisfied were you with the lectures by the:

	Unsatisfied			Very satisfied
Psychiatrist	1	2	3	4
Pediatric/colorectal surgeon	1	2	3	4
Stoma nurse	1	2	3	4
Physiotherapist	1	2	3	4
Sexologist	1	2	3	4
Nutritionist	1	2	3	4
Social worker	1	2	3	4

4. To what degree did the patient education program help you in managing your situation?

Very little			Very much
1	2	3	4

5. How may the patient education program help you with self-management in everyday life?

6. What was the most important in the patient education program?

7. Did you miss any topics during the patient education program?

Yes, if so what?

No

8. How satisfied were you with the size of the group when it came to sharing experiences and asking questions during the lectures?

- Unsatisfied, too small or too large group
- Satisfied, right size of the group

9. How satisfied were you to talk about sex with both genders present?

- Unsatisfied
- Somewhat satisfied
- Neutral
- Satisfied
- Very satisfied

10. In the future, the group should consist of:

- Both men and women
- Neutral
- Only men and only women

11. Would you have benefited from a patient education program as a teenager?

- Yes
- No
- Do not know

12. How satisfied were you with a web-based patient education program? (Only for the web-based patient education program)

Unsatisfied				Very satisfied
1	2	3	4	

Table 1. Overview of the patient education program (PEP) for adults with Hirschsprungs disease (HD) and anorectal malformations (ARM) with allocated time for each lecturing health care professional and which themes that were covered by them.

Program	Allocated time (minutes)	Health care professional(s)	Themes
Welcome and introduction of participants	30	Pediatric surgeon and stoma care nurse	Outline of the program and presentation of health care professionals. Participants present themselves and their expectations of the PEP.
Information about HD and ARM	30	Pediatric surgeon	Pathogenesis, surgery, associated anomalies, functional outcomes, and current research in HD/ARM.
What the pelvic floor interdisciplinary center can offer adults with HD and ARM	30	Adult colorectal surgeon	Presentation of different health care professionals working at the pelvic floor interdisciplinary center. Investigations for incontinence and constipation. Surgical treatments.
Lunch	60		Lunch free of charge.
Mental health challenges	30	Psychiatrist	Emotional challenges and coping strategies related to living with a chronic disease.
Pelvic floor dysfunctions	30	Physiotherapist	Pelvic anatomy, investigations and non-surgical treatment of pelvic floor dysfunctions. Practical pelvic floor training aimed at strengthening and relaxing the pelvic floor.
Foods and bowel function	30	Nutritionist	Anatomy and function of the gastrointestinal tract. How different foods affect digestion and bowel function.
Social support	30	Social worker	Economic and labor rights. Public services offered to persons with functional impairments.
Break	15		
Impaired bowel and urinary function and self-management strategies	75	Stoma care nurse	Non-surgical treatment of fecal and urinary incontinence, constipation, and stoma care.
Sexuality	30	Sexologist	Treatment and aids for impaired sexual function and strategies to increase sexual confidence.
Closing remarks	15	Pediatric surgeon, adult surgeon, and stoma care nurse	Comments, questions, remarks, and answering and handing in the questionnaire.

Table 2. Demographics of adults with Hirschsprung disease (HD) and anorectal malformations (ARM) participating in a patient education program.

	All participants	HD	ARM
Age, median (range), years	37 (18-57)	37 (18-43)	37 (20-57)
Males	9/21 (43%)	5/10 (50%)	4/11 (36%)
Antegrade colonic enema	4/21 (19%)	2/10 (20%)	2/11 (18%)
Colostomy	2/21 (10%)	1/10 (10%)	1/11 (9%)
Urinary diversion	2/21 (10%)	0	2/11 (18%)

Table 3. The evaluation of a patient education program (PEP) for adults with Hirschsprung disease (HD) and anorectal malformations (ARM) graded on a 4-point Likert scale where one was “unsatisfied” and four “very satisfied”.

	Overall	HD	ARM
Meeting peers	4 (1-4)	4 (3-4)	4 (1-4)
Sharing experiences with peers	4 (1-4)	4 (3-4)	4 (1-4)
Increased knowledge	4 (2-4)	4 (2-4)	3 (3-4)
Improved self-management	3 (2-4)	3 (2-4)	3 (3-4)
<i>Lectures by</i>			
Pediatric and colorectal surgeons	4 (2-4)	4 (2-4)	4 (2-4)
Stoma nurse	4 (2-4)	4 (3-4)	4 (2-4)
Sexologist	4 (3-4)	4 (3-4)	4 (3-4)
Social worker	3 (3-4)	3 (3-4)	4 (3-4)
Nutritionist	3 (2-4)	3.5 (3-4)	3 (2-4)
Psychiatrist	3 (3-4)	3 (3-4)	3 (3-4)
Physiotherapist	3 (2-4)	3 (2-4)	3 (3-4)

Table 4. Factors that might influence attendance rate at a patient education program for adults with Hirschsprung disease or anorectal malformations.

		p-value
Gender		
Men (participants/invited)	9/72 (13%)	p=0.13
Women (participants/invited)	12/53 (23%)	
Age, median (range), years		
Non-participants	23 (18-50)	p=0.01
Participants	37 (18-57)	
Travel distance, median (range), km		
Non-participants	103.5 (4-1725)	p=0.88
Participants	124 (11-1393)	