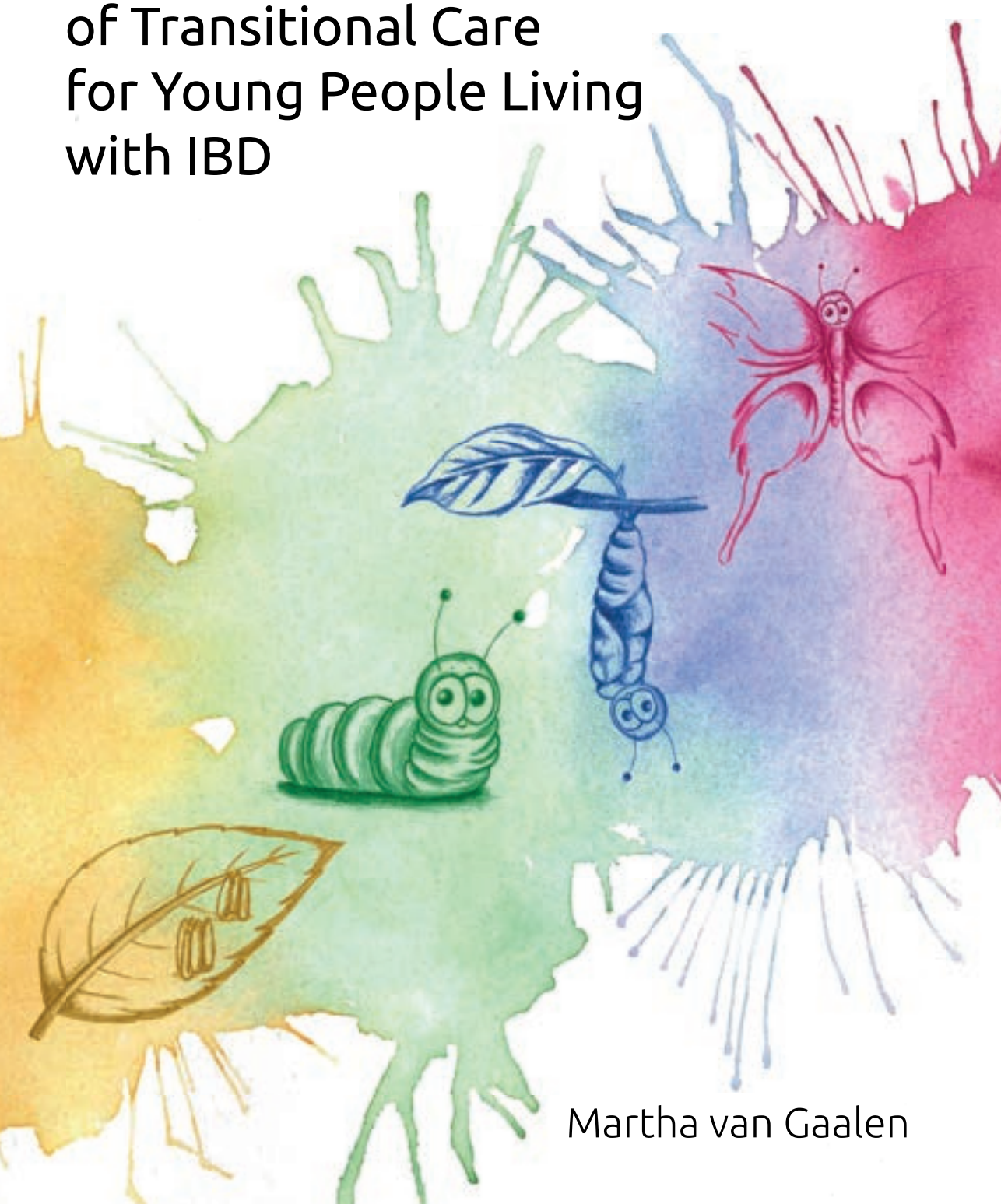
Searching for Success;

Improving the Quality of Transitional Care for Young People Living with IBD



Martha van Gaalen

Searching for Success; Improving the Quality of Transitional Care for Young People Living with IBD
The publication of this thesis was supported by Erasmus University, dr. Falk pharma B.V., Ferring
Author: Martha van Gaalen
Cover layout: Veerle en Niels Hoffmans
Printing: ProefschriftMaken
Layout and design: ProefschriftMaken

Promotiecommissie:

Promotor:	Prof. dr J.C. Escher
Overige leden:	Prof. dr E.H.H.M. Rings
	Prof. dr M. van Dijk
	Dr. P.F. Van Rheenen
Copromotor:	Dr. L. de Ridder

Searching for Success;

Improving the Quality of Transitional Care for Young People Living with IBD

Op zoek naar succes;
verbetering van de kwaliteit van transitiezorg voor jongeren die leven met IBD

Proefschrift
ter verkrijging van de graad van doctor aan de
Erasmus Universiteit Rotterdam
op gezag van de
rector magnificus

Prof.dr.ir. A.J. Schuit

en volgens besluit van het College voor Promoties

De openbare verdediging zal plaatsvinden op

woensdag 21 mei 2025 om 13.00 uur

door

Martha van Gaalen
geboren te Breukelen.

Erasmus University Rotterdam

Contents

Chapter 1

General Introduction, Aims and Outline

Inflammatory Bowel Disease

Crohn's disease (CD) and ulcerative colitis (UC) are the two main types of inflammatory bowel disease (IBD). IBD is a chronic relapsing inflammatory disease that affects the gastrointestinal tract.[1] The pathogenesis of IBD is not yet fully understood. It is multifactorial, with a combination of genetic susceptibility, dysregulation of the immune response, and environmental factors (including gut microbiome composition, diet, and lifestyle) playing a role.[2]

The incidence of IBD has increased worldwide over the last decade, both in adults and children.[3,4] Currently, studies vary in the exact number of people with IBD who develop the disease before the age of 18, but it is thought to be between 2 and 20%.[4-7] Onset of IBD before the age of 18 is associated with increased risk of a more severe phenotype, extensive disease location, higher rates of bowel complications (perianal disease, intestinal strictures, colonic surgery) and greater reliance on immunomodulatory therapy accompanied by elevated risk of malignancy later in life.[8-11]

The young IBD patient

Symptoms of abdominal pain, (bloody) diarrhoea, weight loss and fatigue are common in both CD and UC and may depend on the location and extent of the disease.

In CD, the disease can be localized anywhere in the gastrointestinal tract, from the mouth to the anus. In CD, endoscopy and histology often show transmural granulomatous inflammation with a discontinuous pattern. Perianal fistulae and abscesses may exist or develop during the course of disease. When the disease is confined to the ileum, non- specific symptoms such as decreased appetite, weight loss and delayed puberty may exist, that may lead to a delay in diagnosis.

UC is usually diagnosed earlier because it generally presents with more alarming symptoms such as bloody diarrhoea, urgency and cramping abdominal pain. In contrast to CD, endoscopy and histology show a continuous, superficial inflammation of the colon. The inflammation usually starts in the rectum, extending towards the proximal colon in more severe cases. Compared to adults, most pediatric patients have pancolitis, involving the whole colon.

IBD can also cause symptoms outside of the bowel; about 50% of people with IBD develop an extraintestinal manifestation during their lifetime. The most common are arthritis and skin abnormalities, which are more common in CD (10-20%). In UC, primary sclerosing cholangitis (PSC) is more common (5-7.5%).[12,13]

IBD is a disease with an unpredictable course, characterized by periods of remission alternating with flares. There is currently no definitive cure for IBD. The objective of lifelong treatment is to induce and maintain clinical remission, with the aim of achieving mucosal healing, and to prevent long-term complications of the disease. It is essential to balance these considerations against the potential complications of treatment with immunosuppressive and/or biological medications, dietary modifications, and in some cases, surgery.

While the disease may be in (clinical) remission, patients may still experience abdominal pain[14] and fatigue, affecting social interaction and quality of life.[15] An active lifestyle and a healthy diet can have a beneficial impact on fatigue, quality of life, and potentially even gastrointestinal symptoms.[16]

In addition to this, about 50% of adolescents with IBD experience psychological issues[17], compared to about one third of the total population of Dutch adolescents.[18] IBD patients are more prone to exhibit symptoms of anxiety and depression, with approximately 4% developing an anxiety or depressive disorder[19]. These symptoms contribute to suboptimal therapeutic outcomes, increased morbidity, and elevated mortality rates.[20] It is postulated that there is a bidirectional relationship between IBD and psychological symptoms, which is known as the "brain-gut axis". More specifically, the brain-gut axis posits that the presence of gut inflammation may have a negative effect on mental status, leading to anxiety and/or depression. Conversely, anxiety and/or depression may exacerbate intestinal inflammation and possibly accelerate a relapse of IBD.[21]

All these physical and psychological factors, combined with the unpredictability of the disease, negatively affect the quality of life of patients with IBD.[22] School performance can also be affected by the disease, due to lack of understanding from teachers, absenteeism because of symptoms, and time consuming hospital visits. Adolescent age, rather than disease-specific characteristics, appears to be a principal predictor of impaired school performance in patients with IBD. It is conceivable that the elevated risk of psychosocial issues in adolescents may contribute to this problem.[23,24]

Unexpected disease exacerbations, or flares, can lead to significant disruptions in the family life of the young IBD patient. During such episodes, caregivers may experience elevated levels of parental stress, indirectly contributing to a reduction in the quality of life of the pediatric patient with IBD.[25] Furthermore, having a child with IBD can lead to substantial limitations in work and daily activities for caregivers. The provision of family support is therefore of paramount importance.[26]

Adolescence

The term 'adolescence' is used to describe the natural transition between childhood and adulthood. The onset of adolescence is frequently accompanied by a series of hormonal fluctuations, collectively known as puberty. During the period of puberty, which typically starts between the ages of 9 and 12, with girls often entering puberty earlier than boys, numerous biological processes within the body undergo transformation under the influence of hormones. The typical physical features of adolescence include rapid growth in height, the development of gender characteristics and (in boys) changes in voice. However, there is also a wide range of social, behavioural and emotional changes that occur during this period. During this period, adolescents are compelled to establish their own identity and to navigate a multitude of social challenges. The final stage of becoming an independent and responsible adult, which can last until the early 20s, is often accompanied by changes in social roles and responsibilities. The formation of friendships, romantic relationships, religious beliefs and family structures, in addition to the development of personal achievements and career plans, are significant aspects of this period of life. The cognitive changes that occur during this time enable young people to regulate their emotions and behaviour in a manner that is conducive to the pursuit of long-term goals. It is important to note that the experience of adolescence varies considerably from one individual to another, and there is no definitive age at which this phase of life comes to an end.[27,28]

Adolescence and IBD

The process of growing up is challenging enough, but it can be even more so when living with a chronic disease (see Figure 1). When a patient is diagnosed at a young age (before the age of 12), caregivers often take on the responsibility of managing the disease. However, as they grow older, patients need to develop the ability to act as independent and responsible adolescents or young adults in managing their condition. Exploring this new role can be enlightening, but also overwhelming and not always without pitfalls.[28] For example, adherence to medications can be a challenge for adolescents and young adults with IBD, which can lead to unnecessary exacerbations of the disease.[29,30]

The search for self-identity and the confrontation with caregivers can complicate this process and present challenges for both caregivers and adolescents. Caregivers are often fearful of losing the knowledge they have gained over the years, find it challenging to help their child to develop more autonomy, find communication with adolescents difficult and are fearful of disrupting established patterns of care.[28]

Figure 1: Adolescent IBD patient with their challenges

a. Symptoms of IBD, b. Family, c. Smoking/ alcohol/ drugs, d. Hospital appointments, e. Anxiety and depression, f. Colon cancer, g. Lifestyle, h. Growing up, i. Transition in care, j. Social media, k. Relationships, l. Education

Transition from pediatric to adult healthcare in IBD patients.

Case study from daily practice as a pediatric IBD nurse specialist

Wednesday morning, quarter past 10, and I'm sitting in an empty consulting room. Once again, Mo hasn't shown up for his appointment at the hospital.

Mo is almost 18 years old and has had Crohn's disease since he was 15. When he was first diagnosed, he was really unwell. After a while, his illness calmed down and Mo passed his exams despite being absent from school a lot. He has a busy life with school, soccer, friends and a part-time job delivering pizzas.

Today, Mo is scheduled for his outpatient visits at the IBD Young Adult Clinic. The clinic sees adolescents with IBD from the age of 16 and gets them ready for the transfer to adult healthcare around the age of 18. The first time, when he was 16, Mo came with his parents, but he soon started coming alone because it was easier for him with his studies and his parents' work schedules. However, for the past nine months, Mo hasn't been attending at all. After a few phone calls, I was finally able to speak to him; Mo said he'll definitely come to the next appointment because he is doing well overall, regularly forgets his medication and is wondering why all this is necessary.

On this Wednesday morning at quarter past 10, sitting in my empty consulting room, I wonder what will happen to Mo after his transfer to adult healthcare. Does he know enough about his condition to make treatment decisions? In adult care, patients are expected to be more independent, so does he have enough disease-related self- management skills to cope in adult care? How can we make sure Mo's transition goes well? And what exactly is a successful transition?

The earliest articles about transition appeared in the 1980s. The quality of medical care was improving, and an increasing number of young people with chronic diseases was reaching adulthood. The question was posed as to the optimal means of providing support to this patient group, given the specific challenges.[31,32] A PubMed search reveals that the first article on transition care specifically for IBD patients was published in 2003.[33] In this article, the similarities and differences between pediatric IBD and IBD in adult patients were described. Furthermore, the article delineated the difficulties encountered by young adult patients, including

adherence issues.[33] Today, in 2024, hundreds of articles are published every year on healthcare transition, seeking to support patients with chronic illnesses during this period of change.

Terminology

Transition; A period (phase) of at least two years - both before and after the transfer of healthcare - during which a young person and their caregivers receive coordinated, structured care from medical and nursing specialists in pediatric -, and adult healthcare.

Transfer; The moment of transfer to adult healthcare, usually around the age of 18 in the Netherlands.

IBD is a chronic condition that requires life-long meticulous and regulated treatment in order to prevent complications and significant morbidity, including bowel obstruction, perforation, severe infections, and cancer. At some point in time, in the Netherlands around the age of 18, the adolescent or young adult patient will need to transfer to adult healthcare. This major change may lead to discontinuity of care, and increased risk that the young adult will not attend outpatient appointments. In the absence of disease monitoring, there is increased likelihood of non-adherence to medication, followed by a higher risk of disease exacerbation. This may then lead to progression to more severe disease or to disease complications, which may ultimately necessitate surgical intervention.[34-36]

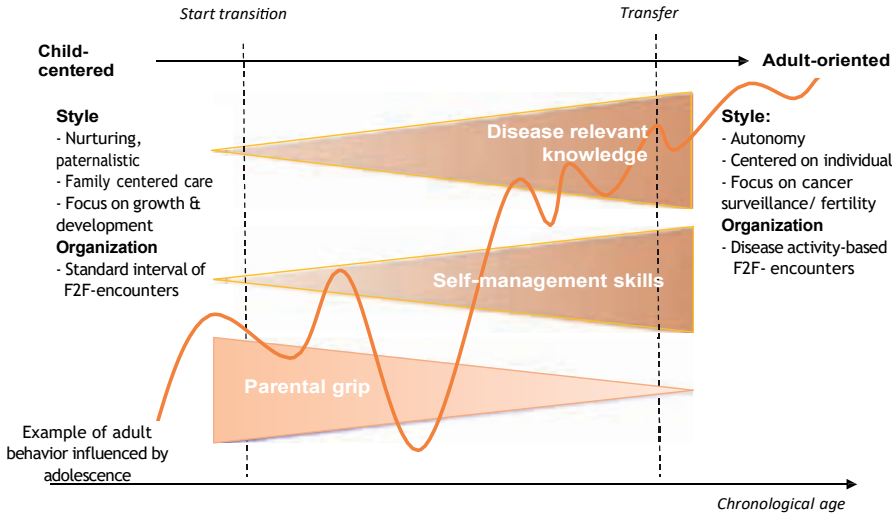
In order to successfully navigate in the adult healthcare system, the young adult patient should possess the skills to independently manage their illness and lifestyle and demonstrate the ability to respond effectively to challenges that may arise. In adult healthcare, patients are expected to engage in discourse during (live, online or telephone) consultations and to assume a participatory role in the care process. Figure 2, based on the Figure from the ECCO Topical Review[34], illustrates the development of skills that facilitate the transition to an independent and responsible adult patient.[34]

Knowledge of the disease and its treatment enable the young person to communicate effectively with healthcare professionals and to assume responsibility for managing their own condition. It is crucial to educate young adults about adult

topics such as the risks associated with alcohol, smoking, vaping, and drugs, but also about adult topics such as sexuality and pregnancy.[34,37-40]

The process of increasing independence and decreasing parental control is associated with the stages of adolescence. The course of adolescence is not uniform in all patients and can extend into the early 20s.[27] This is why Figure 2 shows that the transition continues in adult care, meaning that the patient does not need to know everything and be completely self- managing ('ready') at the time of transfer. In fact, the development towards independence as a patient continues in adult healthcare. This also requires commitment and skills from the receiving adult healthcare provider.

Figure 2: overview transition process based on Topical review of the ECCO[34]



F2F= face to face

Terminology

Self-management aims to empower patients with long-term conditions to take control of their treatment.

Self-efficacy: a person's belief in their capability to organize and execute the actions required to deal with prospective situations.

Knowledge: all the patient knows about his disease/treatment

The transition process is contingent upon optimal collaboration between healthcare providers from both sides of the care spectrum, necessitating a multidisciplinary approach. It is crucial that healthcare providers from both sides are aware of the differences between pediatric and adult care (see Table 1). These differences, combined with the pathway to independence and adulthood, represent the challenges faced by the young adult with IBD. The transition to adult care is frequently perceived as a natural progression towards independence and adulthood. However, this transition also gives rise to concerns.

Caregivers may express concerns regarding their child's preparedness for this transition and the quality of healthcare they will receive. In addition to the change in the healthcare team from pediatric to adult, young adults may encounter challenges in adapting to variations in healthcare provision (Table 1).[34,35,41]

Table 1: Differences between pediatric and adult healthcare in IBD.

	Pediatric healthcare	Adult healthcare
Specific focus on	• growth, development, school, nutrition	• cancer surveillance, fertility, pregnancy, new drug treatment options (biologicals, small molecules)
Healthcare provision general	• holistic • combination/coordination of appointments with other specialists involved in the field. • often small population of IBD patients (up to 250 patients) • often standard psychological support	• focused on IBD, not multidisciplinary. • often large population of IBD patients (up to thousands of patients)
Outpatient appointments	• regular, every three months with blood sampling in hospital • missed appointments will be contacted and rescheduled. • telephone contact/appointments via caregivers	• trend to do appointments once or at most twice a year or only on demand (in case of increased symptoms) or using telemonitoring. • a missed appointment is the patient's responsibility. • telephone contact via thepatient
Procedures	• endoscopy standard under anesthesia or deep sedation with propofol • if necessary, bowel preparation in hospital • if necessary, lidocaine patch for injections or blood collection • support by pedagogical care in case of fear of needles, for example.	• endoscopy under midazolam, (deep sedation only if explicitly requested). • bowel preparation always at home • no support during interventions for injection or blood collection
Hospitalization	• one caregiver is allowed in the room 24 hours a day. • all consultations are conducted in the presence of caregiver(s) • all-day visiting possible • all kinds of activities are often organized	• consultations are often done after the rounds, i.e., outside visiting hours. • visitors (including caregivers) only during visiting hours
Infusion therapy at day clinic	• often a small room • separate room for venous canula insertion • Distraction with games/computer games and wide choice of food and drink • Caregivers must be present	• often large rooms • for venous canula insertion on the chair in infusion room • staffing is tightly planned • Caregivers are usually not allowed

To optimize the transition from pediatric to adult healthcare and minimize the risk of complications, a structured transition program is recommended to prepare patients and their caregivers for the actual transfer. During this program, which should be started in a timely manner, attention should be paid to the above-mentioned skills of increasing knowledge and independence regarding the illness, as well as to the differences in care. Caregivers should be taught how to support their child in the care process in a different way; more alongside the young adults rather than holding control over the care.[42,43]

The Dutch Quality Standard 'Young people in Transition from pediatric to adult healthcare', based on the NICE guidelines[44], describes a number of core interventions during structured transition:[45]

- Appointing a transition coordinator,
- Making an Individual Transition Plan together with the Adolescent and Young Adult (AYA)
- Organizing a warm handover of healthcare (with both teams, patient and caregivers present) around the age of 18,
- Comprehensive consultation for extended introductions the 1st year of adult healthcare.

The implementation of a structured transition program in IBD has been shown to improve self-management skills, disease knowledge, satisfaction and quality of life in adolescents[34,35,46-48] At the moment, costs of transitional care are not reimbursed, and the provision of transitional care depends on the affinity of healthcare providers in both pediatric and adult healthcare. As a result, care still varies greatly between departments and hospitals. If it can be shown that a particular program leads to a more successful outcome of transition, this could eventually lead to reimbursement by health insurers and more consistency in transition care.[35,49]

However, there is no single preferred transition model. This is partly because there is no clear definition of transition success or scoring system to measure transition success in IBD patients. The absence of a scoring system hinders accurate assessment of existing tools and transition programs. Studies evaluating the success of transition intervention programs often rely on unvalidated or qualitative instruments based on patient experience or satisfaction. Therefore, there is a need for a validated scoring system for transition success.[50,51]

Aim and Outline of this thesis.

The focus of this thesis is on how healthcare providers can best guide adolescent and young adult IBD patients during the transition period to adult healthcare. The objective was to optimize the tools used for monitoring of skills building during transition (Part A), and to establish a valid method to measure success (or effectiveness) of the transition process (Part B).

Part A: Developing tools to support the transition process.

Chapter 2 describes the development and validation of an open-ended questionnaire to measure and monitor knowledge about IBD during the transition process, the Rotterdam Transition Test.
Chapter 3 provides the translation and validation with reference scores for IBD patients of the Dutch version of the self-management tool Transition Readiness Assessment Questionnaire; TRAQ-NL.
In **Chapter 4** we describe the implementation of Patient Reported Outcome Measures (PROMs) in routine healthcare at the outpatient IBD clinic, as well as assess its feasibility.

Part B: The quest of making success of transition measurable.

In **Chapter 5**, we explore the relationship between self-efficacy and transition outcomes with a composite score to measure transition success.
In **Chapter 6**, we have taken the first step in the development of a tool to measure transition success. Through a multinational Delphi study, a large panel of experts and patients were asked to identify outcomes that reflect successful transition.
Chapter 7 describes the development and validation of the Transition Success Score (TSS) and shows the results of a prospective, national multicentre study on the outcome of transition.
Chapter 8 discusses the main findings of this thesis and provides recommendations for clinical practice and future research.
Chapter 9 summarizes our findings as described in this thesis in English and Dutch.

References

1. Rosen MJ, Dhawan A, Saeed SA. Inflammatory bowel disease in children and adolescents. *JAMA Pediatr* 2015;**169**:1053-60.
2. Peloquin JM, Goel G, Villablanca EJ, Xavier RJ. Mechanisms of pediatric inflammatory bowel disease. *Annu Rev Immunol* 2016;**34**:31-64.
3. Ng SC, Shi HY, Hamidi N, *et al.* Worldwide incidence and prevalence of inflammatory bowel disease in the 21st century: A systematic review of population-based studies. *Lancet* 2017;**390**:2769-78.
4. Wang Y, Pan CW, Huang Y, *et al.* Global epidemiology and geographic variations of pediatric-onset inflammatory bowel disease: A comprehensive analysis of the global burden of disease study 1990 to 2019. *Inflamm Bowel Dis* 2024.
5. Ghione S, Sarter H, Fumery M, *et al.* Dramatic increase in incidence of ulcerative colitis and crohn's disease (1988-2011): A population-based study of french adolescents. *Am J Gastroenterol* 2018;**113**:265-72.
6. Kern I, Schoffer O, Kiess W, *et al.* Incidence trends of pediatric onset inflammatory bowel disease in the years 2000-2009 in saxony, germany-first results of the saxon pediatric ibd registry. *PLoS One* 2021;**16**:e0243774.
7. Burgess CJ, Henderson P, Jones GR, *et al.* Paediatric patients (less than age of 17 years) account for less than 1.5% of all prevalent inflammatory bowel disease cases. *J Pediatr Gastroenterol Nutr* 2020;**71**:521-3.
8. Van Limbergen J, Russell RK, Drummond HE, *et al.* Definition of phenotypic characteristics of childhood-onset inflammatory bowel disease. *Gastroenterology* 2008;**135**:1114-22.
9. Pigneur B, Seksik P, Viola S, *et al.* Natural history of crohn's disease: Comparison between childhood- and adult-onset disease. *Inflamm Bowel Dis* 2010;**16**:953-61.
10. Nasiri S, Kuenzig ME, Benchimol EI. Long-term outcomes of pediatric inflammatory bowel disease. *Semin Pediatr Surg* 2017;**26**:398-404.
11. Granot M, Kopylov U, Loberman-Nachum N, *et al.* Differences in disease characteristics and treatment exposures between paediatric and adult-onset inflammatory bowel disease using a registry-based cohort. *Aliment Pharmacol Ther* 2024.
12. Greuter T, Bertoldo F, Rechner R, *et al.* Extraintestinal manifestations of pediatric inflammatory bowel disease: Prevalence, presentation, and anti-tnf treatment. *J Pediatr Gastroenterol Nutr* 2017;**65**:200-6.
13. Vavricka SR, Schoepfer A, Scharl M, *et al.* Extraintestinal manifestations of inflammatory bowel disease. *Inflamm Bowel Dis* 2015;**21**:1982-92.
14. Limbri LF, Wilson TG, Oliver MR. Prevalence of irritable bowel syndrome and functional abdominal pain disorders in children with inflammatory bowel disease in remission. *JGH Open* 2022;**6**:818-23.
15. Bevers N, Van de Vijver E, Hanssen A, *et al.* Fatigue and physical activity patterns in children with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2023;**77**:628- 33.
16. Scheffers LE, Vos IK, Utens E, *et al.* Physical training and healthy diet improved bowel symptoms, quality of life, and fatigue in children with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2023;**77**:214-21.
17. van den Brink G, Stapersma L, Vlug LE, *et al.* Clinical disease activity is associated with anxiety and depressive symptoms in adolescents and young adults with inflammatory bowel disease. *Aliment Pharmacol Ther* 2018;**48**:358-69.
18. van der Velden PG, van Bakel HJA, Das M. Mental health problems among dutch adolescents of the general population before and 9 months after the covid-19 outbreak: A longitudinal cohort study. *Psychiatry Res* 2022;**311**:114528.
19. Stapersma L, van den Brink G, Szigethy EM, Escher JC, Utens E. Systematic review with meta-analysis: Anxiety and depression in children and adolescents with inflammatory bowel disease. *Aliment Pharmacol Ther* 2018;**48**:496-506.
20. Katon WJ. Epidemiology and treatment of depression in patients with chronic medical illness. *Dialogues Clin Neurosci* 2011;**13**:7-23.
21. Fairbrass KM, Lovatt J, Barberio B, *et al.* Bidirectional brain-gut axis effects influence mood and prognosis in ibd: A systematic review and meta-analysis. *Gut* 2022;**71**:1773-80.

22. Knowles SR, Graff LA, Wilding H, *et al.* Quality of life in inflammatory bowel disease: A systematic review and meta-analyses-part i. *Inflamm Bowel Dis* 2018;**24**:742-51.
23. Barnes C, Ashton JJ, Borca F, *et al.* Children and young people with inflammatory bowel disease attend less school than their healthy peers. *Arch Dis Child* 2020;**105**:671-6.
24. Carreon SA, Bugno LT, Wojtowicz AA, Greenley RN. School functioning in adolescents with inflammatory bowel diseases: An examination of disease and demographic correlates. *Inflamm Bowel Dis* 2018;**24**:1624-31.
25. Sălcudean A, Nan AG, Bodo CR, *et al.* Association between childhood onset inflammatory bowel disease and psychiatric comorbidities in adulthood. *Diagnostics (Basel)* 2023;**13**.
26. Klomberg RCW, Aardoom MA, Kemos P, *et al.* High impact of pediatric inflammatory bowel disease on caregivers' work productivity and daily activities:
An international prospective study. *J Pediatr* 2022;**246**:95-102 e4.
27. Crone EA, Dahl RE. Understanding adolescence as a period of social-affective engagement and goal flexibility. *Nat Rev Neurosci* 2012;**13**:636-50.
28. Lerch MF, Thrane SE. Adolescents with chronic illness and the transition to self- management: A systematic review. *J Adolesc* 2019;**72**:152-61.
29. Spekhorst LM, Hummel TZ, Benninga MA, van Rheenen PF, Kindermann A. Adherence to oral maintenance treatment in adolescents with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2016;**62**:264-70.
30. Alsous MM, Hawwa AF, Imrie C, *et al.* Adherence to azathioprine/6-mercaptopurine in children and adolescents with inflammatory bowel diseases: A multimethod study. *Can J Gastroenterol Hepatol* 2020;**2020**:9562192.
31. Hirst MA. Young people with disabilities: What happens after 16? *Child Care Health Dev* 1983;**9**:273-84.
32. Will MC. Youth with disability. The transition years. Opening remarks. *J Adolesc Health Care* 1985;**6**:79-83.
33. Désir B, Seidman EG. Transitioning the paediatric ibd patient to adult care. *Best Pract Res Clin Gastroenterol* 2003;**17**:197-212.
34. van Rheenen PF, Aloi M, Biron IA, *et al.* European crohn's and colitis organisation topical review on transitional care in inflammatory bowel disease. *J Crohns Colitis* 2017;**11**:1032-8.
35. Brooks AJ, Smith PJ, Cohen R, *et al.* Uk guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. *Gut* 2017;**66**:988-1000.
36. Blum RW, Garell D, Hodgman CH, *et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the society for adolescent medicine. *J Adolesc Health* 1993;**14**:570-6.
37. Zijlstra M, De Bie C, Breij L, *et al.* Self-efficacy in adolescents with inflammatory bowel disease: A pilot study of the "ibd-yourself", a disease-specific questionnaire. *J Crohns Colitis* 2013;**7**:e375-85.
38. Hait EJ, Barendse RM, Arnold JH, *et al.* Transition of adolescents with inflammatory bowel disease from pediatric to adult care: A survey of adult gastroenterologists. *J Pediatr Gastroenterol Nutr* 2009;**48**:61-5.
39. Benchimol EI, Walters TD, Kaufman M, *et al.* Assessment of knowledge in adolescents with inflammatory bowel disease using a novel transition tool. *Inflamm Bowel Dis* 2011;**17**:1131-7.
40. Cole R, Ashok D, Razack A, Azaz A, Sebastian S. Evaluation of outcomes in adolescent inflammatory bowel disease patients following transfer from pediatric to adult health care services: Case for transition. *J Adolesc Health* 2015;**57**:212-7.
41. Bihari A, Hamidi N, Seow CH, *et al.* Defining transition success for young adults with inflammatory bowel disease according to patients, parents and health care providers. *J Can Assoc Gastroenterol* 2022;**5**:192-8.
42. Sattoe JNT, Peeters MAC, Haitsma J, *et al.* Value of an outpatient transition clinic for young people with inflammatory bowel disease: A mixed-methods evaluation. *BMJ Open* 2020;**10**:e033535.
43. Goodhand J, Hedin CR, Croft NM, Lindsay JO. Adolescents with ibd: The importance of structured transition care. *J Crohns Colitis* 2011;**5**:509-19.
44. Willis ER, McDonagh JE. Transition from children's to adults' services for young people using health or social care services (nice guideline ng43). *Arch Dis Child Educ Pract Ed* 2018;**103**:253-6.

45. Specialisten FM. Kwaliteitsstandaard - jongeren in transitie van kinderzorg naar volwassenenzorg. https://richtlijnendatabase.nl/richtlijn/kwaliteitsstandaard_-_jongeren_in_transitie_van_kinderzorg_naar_volwassenenzorg/startpagina_-_jongeren_in_transitie_van_kinderzorg_naar_volwassenenzorg.html Accessed 10-10-2024, 2022.
46. Fishman LN, Ding J. Optimizing the transition and transfer of care in pediatric inflammatory bowel disease. *Gastroenterol Clin North Am* 2023;**52**:629-44.
47. Kim J, Ye BD. Successful transition from pediatric to adult care in inflammatory bowel disease: What is the key? *Pediatr Gastroenterol Hepatol Nutr* 2019;**22**:28-40.
48. Crowley R, Wolfe I, Lock K, McKee M. Improving the transition between paediatric and adult healthcare: A systematic review. *Arch Dis Child* 2011;**96**:548-53.
49. Chan P, McNamara J, Vernon-Roberts A*, et al.* Systematic review: Practices and programs in inflammatory bowel disease transition care. *Inflamm Bowel Dis* 2024.
50. Bihari A, Olayinka L, Kroeker KI. Outcomes in patients with inflammatory bowel disease transitioning from pediatric to adult care: A scoping review. *J Pediatr Gastroenterol Nutr* 2022;**75**:423-30.
51. Sattoe JNT, Hilberink SR, van Staa A. How to define successful transition? An exploration of consensus indicators and outcomes in young adults with chronic conditions. *Child Care Health Dev* 2017;**43**:768-73.

A

Part A

Developing Tools to Support the Transition Process

2

Chapter 2

Rotterdam Transition Test: A Valid Tool for Monitoring Disease Knowledge in Adolescents With Inflammatory Bowel Disease

MAC van Gaalen, M van Pieterson, G van den Brink, L de Ridder, D Rizopoulos, JC Escher.

J Pediatr Gastroenterol Nutr 2022;74:60–7

ABSTRACT

Objectives

Disease knowledge is important in adolescents with inflammatory bowel disease (IBD) transitioning to adult care. We developed an IBD-specific knowledge questionnaire, the Rotterdam Transition Test (RTT), and aimed to validate this tool.

Methods

This is a prospective longitudinal validation study. The RTT has 25 open questions on IBD, medication, lifestyle and transition to adult care. A scoring model was developed, and inter-rater agreement was assessed. Using a Rasch model, we determined the difficulty and performance of the questions. Cronbach's alpha was used to demonstrate reliability. Patient factors (age, disease, education, medication use, illness acceptance and independence) were correlated to RTT score.

Results

A total of 207 RTTs were evaluated in 111 adolescent IBD patients. The scoring model showed a kappa score of > 0.61 for all questions. Reliability with Cronbach's alpha was good (0.81). Mean total result of the RTT was 58% (girls) and 55% (boys) of maximal score.

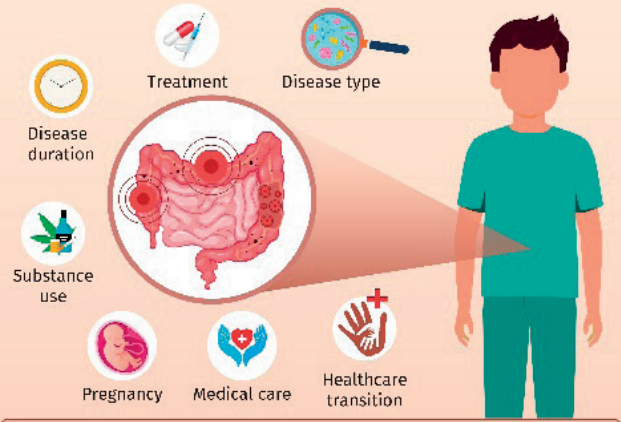
The RTT discriminated well between the different levels of knowledge. Knowledge scores increased in patients who did repeated RTTs during the transition period. Male sex, low educational level, disease acceptance issues and dependence on parents associated with a significantly lower total RTT score. Prednisone use within three months and treatment without biologics associated with significantly higher RTT scores. Disease activity was not a significant factor.

Conclusion

The Rotterdam Transition Test is a reliable and valid tool to assess IBD knowledge. The RTT can be used to detect and discuss knowledge gaps in adolescents with IBD transitioning to adult healthcare.

The Rotterdam Transition Test for Measuring Knowledge Gaps in Inflammatory Bowel Disorder: Is it Valid?

Increasing disease knowledge in adolescents with inflammatory bowel disease (IBD) during their transition from adolescent IBD care to adult care is crucial for future disease management



This study aimed to validate the open questionnaire-based Rotterdam Transition Test (RTT) which measures IBD knowledge

Prospective longitudinal validation study

111 (Adolescents with IBD)

207 RTT

Validation parameters

- ✓ Difficulty and performance of RTT (Rasch model)
- ✓ Reliability (Cronbach's alpha)
- ✓ Correlation between patient factors and RTT score

Scoring of RTT

Reliability of RTT
(Cronbach's alpha = 0.81)

GOOD

Mean total RTT score
(% of maximal score)

Female 58%

Male 55%

RTT discriminated between knowledge levels

Low knowledge level

Higher knowledge level

Factors associated with low total RTT score

- Male sex
- Low education level
- Disease acceptance issues
- Dependence on parents

Low total RTT score

The Rotterdam Transition Test is a reliable and valid tool to assess IBD knowledge in adolescents

Rotterdam Transition Test: a Valid Tool for Monitoring Disease Knowledge in Adolescents with Inflammatory Bowel Disease
van Gaalen et al. (2021) | DOI:

JPGN

INTRODUCTION

In 15 to 25% of patients, Inflammatory Bowel Disease (IBD) has its onset during late childhood or adolescence. (1, 2) Compared to adult-onset, paediatric-onset IBD patients often have more extensive disease with a more severe disease course. (2) The care provided in the paediatric setting is different from that in adult IBD healthcare. (2, 3) In general, paediatric healthcare is family-centred, multidisciplinary and more holistic, with an important role for the parents and focus on growth and development. (3, 4) Adult care tends to be patient-centred, focuses more on cancer surveillance, sexual function and pregnancy, while patients are expected to be autonomous and independent. (2, 3, 5)

Despite these differences, all adolescent patients need to undergo transfer to adult care, usually around 18 years. "Transfer" is defined as a moment in time, when handover of care to the adult health-care team is done, and should be considered part of [and not necessarily the end of] transition.(6) Absence of or insufficient quality of transitional care may have serious negative consequences, such as non-adherence, non-attendance to clinic visits, more hospitalization and higher surgery rates. (3, 7, 8) "Transition" is defined as a period of time when the purposeful, planned movement of adolescents with chronic medical conditions into adult-orientated health-care systems is organised.(9) And is advised to prepare patients and parents for the actual transfer. (3, 10, 11) Our adolescent IBD patients aged 16-18 years are routinely managed by a multidisciplinary team in the transition clinic as described by Sattoe et al (12). Besides disease management, the focus is mainly on preparing the adolescent for adult care by increasing self-management skills and disease knowledge. (13) An international Delphi study involving IBD nurses and doctors in paediatric and adult care and as well as patients was carried out to identify outcome factors that represent a successful transition, with decision-making regarding IBD as the most important factor. (14) In order to be able to make decisions, a certain degree of knowledge is required. Adolescents however have been reported to have little knowledge of their disease. (3, 13, 15, 16) In order to measure and monitor disease knowledge, we developed a questionnaire: the Rotterdam Transition Test (RTT). In contrast to other questionnaires (13, 15, 17, 18) , the RTT has no multiple choice, but only open questions. The purpose of this study was to develop and validate the RTT in adolescent IBD patients.

MATERIALS AND METHODS

The study was a single-centre, longitudinal, prospective validation study that was divided into three phases: questionnaire development, questionnaire testing and performance assessment. The Research Ethics Review Board of the Erasmus MC approved this study. Patients were asked for written informed consent before recruitment into the study.

For this cohort study we followed the STROBE statement methodology.

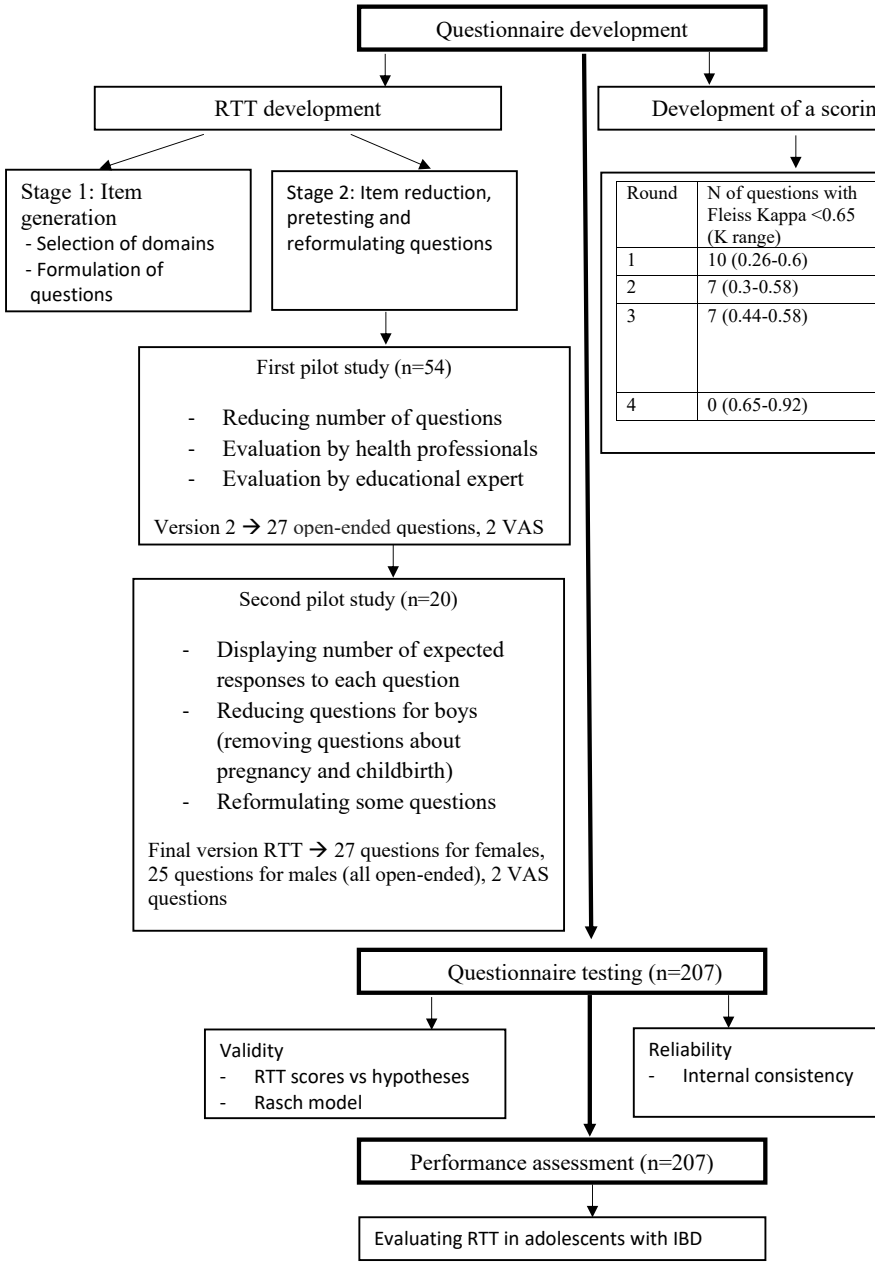
Development of the Rotterdam Transition Test
This phase of the study had several stages: item generation, item reduction, pre-testing, reformulation of questions and development of a scoring model (Figure 1).

Stage 1: item generation
The first steps in the RTT development were taken in 2011. The initial RTT was developed and used as an instrument to test IBD knowledge and self-management in our transition clinic, in order to determine in what areas the patients' knowledge and self-management skills were deficient and needed improvement. The questions were generated from the available literature (4, 16, 19-22) and the opinion of an expert in transitional care and a panel of IBD specialists (two paediatric gastroenterologists, one adult gastroenterologist, a PhD student and two IBD nurses). The first version of the test consisted of 27 open-ended questions on knowledge, 11 multiple choice and two visual analogue scale (VAS) questions on self-management.

Stage 2: Item reduction, pretesting and reformulating questions
A pilot study was conducted in 54 adolescents visiting the IBD transition clinic. With their feedback on the time investment, we shortened the questionnaire by removing multiple choice questions about self-management, thus focusing only on disease knowledge. Review by the expert panel resulted in minor textual changes.

After linguistic review and corrections (made by an educational scientist), the questionnaire was again piloted in 20 adolescents during their transition clinic visits. Patients were interviewed after completing the questionnaire to identify areas of difficulty. To rule out misinterpretation, incorrectly answered questions were reviewed together with the patients. Patient feedback resulted in elimination of two questions (about pregnancy and childbirth) for the male patients. Another suggestion of the patients was to show (per question) how many answer

Figure 1: Study overview



K range: Kappa range, RTT: Rotterdam Transition Test, VAS: visual analogue scale

items should be provided in order to get a maximum score. This testing-retesting process resulted in a final version of the RTT, consisting of 27 open-ended questions for girls and 25 open-ended questions for boys. For a listing of question subjects, see Table 1.

Table 1: Overview of the Rotterdam Transition Test, total patients n= 207

Question	Subject of question	Number (%) of patients with correct responses
1a	VAS independence (n=200)	Median 80 IQR: 20 (70-90)
1b	Name of disease	202 (97%)
2	Location of IBD	156 (75%)
3	Symptoms of disease	203 (98%)
4	Abbreviation IBD	71 (34%)
5a	Exacerbation of disease	149 (72%)
5b	Symptoms of disease exacerbation	168 (81%)
6	Long-term consequences of disease	46 (22%)
7	Alcohol and IBD	15 (7%)
8	Smoking and IBD	41 (20%)
9	Drugs and IBD	13 (6%)
10	Fertility and IBD	29 (14%)
11	Pregnancy and IBD (only for girls; n= 77)	14 (7%)
12	Childbirth and IBD (only for girls; n= 77)	4 (2%)
13	Heredity	61 (30%)
14	Diagnostic procedures for diagnosing IBD	172 (83%)
15	Diagnostic procedures for assessing disease exacerbation IBD	42 (20%)
16	Reasons for performing endoscopy in paediatric care	31 (15%)
17	Reasons for performing endoscopy in adult care	19 (9%)
18	Transition	70 (34%)
19	Adult team	74 (36%)
20a	Differences between paediatric and adult care	7 (3%)
20b	VAS transfer readiness (n= 195)	Median 75 IQR: 40 (55-95)
21	Name of medication	174 (84%)
22	Dosage of medication	140 (67%)
23	Maintenance therapy	130 (63%)
24	Medication to induce remission (at exacerbation)	83 (40%)
25	Side effects of medication	56 (27%)
26	Other treatments (diet, surgery, etc.)	24 (12%)

IBD: inflammatory bowel disease, IQR: interquartile range, RTT: Rotterdam Transition Test, VAS: visual analogue scale (range 0–100).

Development of a Scoring Model
As the RTT has only open questions, a scoring model had to be made to quantitatively score responses to questions. The types of responses in the scoring model were formulated by the expert panel, with support of the available literature.

A four-stage point system was used for each response: one point for false response / I don't know; two points for a not entirely wrong answer; three points for an almost right but not completely correct answer; and four points for a completely correct answer. In general, when in doubt how to score one of the answers, it was decided to round up to a more positive score. If there was no response, it was assumed that the adolescent did not know the answer. For girls, the RTT (27 questions, score ranging from 1-4 points) score ranged from 27-108 points, and for boys (25 questions), RTT score ranged from 25-100 points.

Kappa (K) was used to assess the inter-rater reliability of the scoring model. We aimed for questions that showed substantial agreement (K > 0.61) to almost perfect agreement (K > 0.81). In each round of testing, twenty RTTs were independently assessed by three different assessors. After four rounds, substantial or almost perfect agreement was reached on all 27 questions. (Figure 1).

Testing of the Rotterdam Transition Test

Validity testing
In 2016 to 2018, all patients visiting the IBD transition clinic were asked to fill in the RTT yearly during their transition process (maximal four time points: age 16, 17, at the last transition clinic visit (around 18) and one year after transfer to adult care). The RTT was sent by a secure online survey program (Lime Survey version 3.1.1, LimeSurvey GmbH, Hamburg, Germany). Questionnaires obtained during the second round of pretesting (Figure 1) were also included in this analysis as the questionnaire then used was essentially identical to the final version.

A Rasch analysis was used to analyse characteristics of the questions and to ascertain whether any questions needed further adjustment, as well as to assess the capacity of the questions to discriminate between different levels of knowledge. (23) Rasch analysis resulted in a standardized knowledge score per patient, ranging from 0 to 1. The minimum value 0 corresponded to no knowledge level, 0.5 to average knowledge level, 1 to excellent knowledge level.

As an external criterion for IBD-related knowledge was not available, the validity of the RTT was assessed using a number of constructs (Table 2), which were hypotheses regarding relations between the RTT knowledge scores from the Rasch analysis and presumed surrogate markers of IBD-related knowledge. (7, 8, 10, 17) The information required for the hypotheses was obtained by additional questions about disease acceptance, dependence on parents and VAS (0-100) about transfer readiness and self-management.

Table 2: Hypothetical constructs and assessment of validity and discriminatory ability of the RTT

Construct	Statistic
Association of total RTT scores of patients with IBD with specific variables:	
Scores should correlate positively with patient age	Pearson correlation coefficient
Scores should correlate positively with disease duration	Pearson correlation coefficient
Scores should not differ between CD and UC patients	ANOVA
Females often score better in questionnaires than males	*t* test
Patients with lower educational levels score lower than those with higher educational levels	Post-hoc Tukey test
Scores should correlate positively with repeated RTT administration	Rasch model
Scores should correlate positively with higher VAS self-management	Spearman correlation coefficient
Scores should correlate positively with higher VAS transfer readiness	Spearman correlation coefficient
Patients who were without parents for more than two nights should score higher (independency score)	*t* test
Patients who accepted having IBD should score higher	*t* test

ANOVA; analysis of variance, CD; Crohn disease, IBD; inflammatory bowel disease, RTT; Rotterdam Transition Test, UC; ulcerative colitis, VAS; visual analogue scale

Reliability testing

The internal consistency of the questionnaire was assessed using Cronbach's alpha, which is an indicator of the extent to which different items on a scale assess a single entity.

Rotterdam Transition Test responses of the adolescents with IBD

We further dichotomised answers as "incorrect" when the answer scored 1-2 points, and "correct" in case of 3-4 points. To analyse the adolescents' RTT responses we calculated percentage of correct answers versus incorrect answers. Clinical disease activity at time of RTT was determined by the Pediatric Ulcerative Colitis Activity Index (PUCAI) and the Pediatric Crohn's Disease Activity Index (PCDAI), with values >10 defined as active disease. Exacerbation in the last year

was noted if there were medication changes based on increased symptoms or presence of endoscopic disease activity.

Statistical Analysis

All statistical analyses were performed using SPSS for Windows, version 25 (IBM SPSS Statistics for Windows, Armonk, NY, USA). Results were considered significant if $p < 0.05$. Associations between RTT knowledge scores as extracted from the Rasch model and a number of independent variables were evaluated in bivariate or multivariate analyses. Most multivariate answers were categorized. Independent variables were the same as the hypothetical constructs are listed in Table 2. In addition, *t*-tests, Pearson's and Spearman's correlation coefficients were used for comparisons using dichotomous, continuous and discontinuous independent variables, respectively. To investigate whether the education level was associated with the RTT score we performed an analysis of variance. RTT was considered the dependent variable, and the education level the categorical predictor, with categories, low, medium and high. The Rasch model was also used to study the characteristics of the questions. Cronbach's alpha test was used to analyse the reliability of overall knowledge as assessed by the RTT. As questions about pregnancy and childbirth were intended for girls only, we did not include these in this validation.

If an item is missed, it will not be included in the analyses and will be reflected for the specific analysis in the reduction of participants (n).

RESULTS

Testing of the Rotterdam Transition Test

Rotterdam Transition Test Validity

For validity assessment, we collected 207 RTTs from 111 adolescent patients (response rate 89%) with median age at diagnoses of 14.08 yrs (IQR 2.65 (range 2.8-17.6)), 63% being male and 59% having CD. Table 3 lists demographics at the time of RTT. 110 RTTs (53.1%) were filled in for the first time and almost all RTTs (201; 97.1%) were taken before transfer .

We found significant positive correlations of RTT knowledge scores from the Rash model (ranging from 0 and 1) with the following hypothetical constructs (as presented in Table 2): age (Pearson's correlation coefficient 0.42, $p < 0.001$), higher VAS of self-management (Spearman's correlation coefficient 0.22, $p=0.002$), transfer readiness (Spearman's correlation coefficient 0.20, $p=0.006$), being without

Table 3: Demographics of IBD patients at the moment of completing the Rotterdam Transition Test (n=207)

		N. (%) or Median
Age (years)	Range Median; IQR	15.34-19.97 17.14; 1.37 (16.5-17.8)
Disease duration (years)	Range Median; IQR	0.18-13.97 3.07; 3 (1.6-4.6)
RTT completed before/after transfer	Before transfer After transfer	201 (97.1%) 6 (2.9%)
Number of times that RTT was completed by study subject	Once 2 times 3 times 4 times	110 (53.1%) 70 (33.8%) 26 (12.6%) 1 (0.5%)
Educational level	Low (lower secondary) Medium (upper secondary) High (pre-university)	110 (53.1%) 62 (30%) 36 (16.9%)
Disease activity	Remission	148 (71,5%)
Current medication use	Aminosalicylates Immunomodulators Biologics Corticosteroids Topical treatment No medication	64 (30.9%) 114 (55.1%) 75 (36.2%) 20 (9.7%) 10 (4.8%) 18 (8.7%)
Steroids use 3 months before RTT	Yes	30 (14.5%)
Surgical during disease (peri-anal or bowel resection)	Yes	35 (16.9%)
Relapses in preceding year	None 1 relapse More than 1 relapse	107 (51.7%) 87 (42%) 13 (6.3%)

IQR; interquartile range, RTT; Rotterdam Transition Test

parents for more than two days (0.59 ±0.16 vs 0.44 ±0.18, p <0.001) and accepting the disease (0.58 ±0.17 vs 0.52 ±0.16, p=0.05). Female patients scored significantly higher although there was a relatively large spread in the two groups (0.62 ±0.14 vs 0.53 ±0.17, p <0.001). Adolescents with a low educational level scored significantly lower than adolescents with medium (p = 0.029) and higher (p= 0.002) educational levels.

The Rasch model showed that RTT discriminated well between different levels of knowledge (results ranging from 0.14 to 0.96), with best discrimination between low (0.25) and high (0.75) levels of knowledge. This implies that total RTT score (the total score of the answers given by correction model) showed what adolescents knew. Discrimination was poorer at lower knowledge levels, from low (0.25

SDS) to very low scores (0 SDS). In other words, if adolescents knew just a little bit, total RTT score did not distinguish them from adolescents who knew nothing at all (see Supplemental Figure 1).

Certain questions discriminated better between the different levels of knowledge. The response to questions about disease exacerbation were associated with low levels of knowledge (0.25 and 0.5); i.e., if a patient answered these incorrectly, total RTT score was low. On the other hand, response to questions about reasons for endoscopy in adult care and the differences between the two care systems gave more information about high levels of knowledge; if a patients answered these correctly, total RTT score was high (data not shown).

Questions that provided little information on differences between levels of knowledge were questions about smoking, alcohol, drugs and heredity.

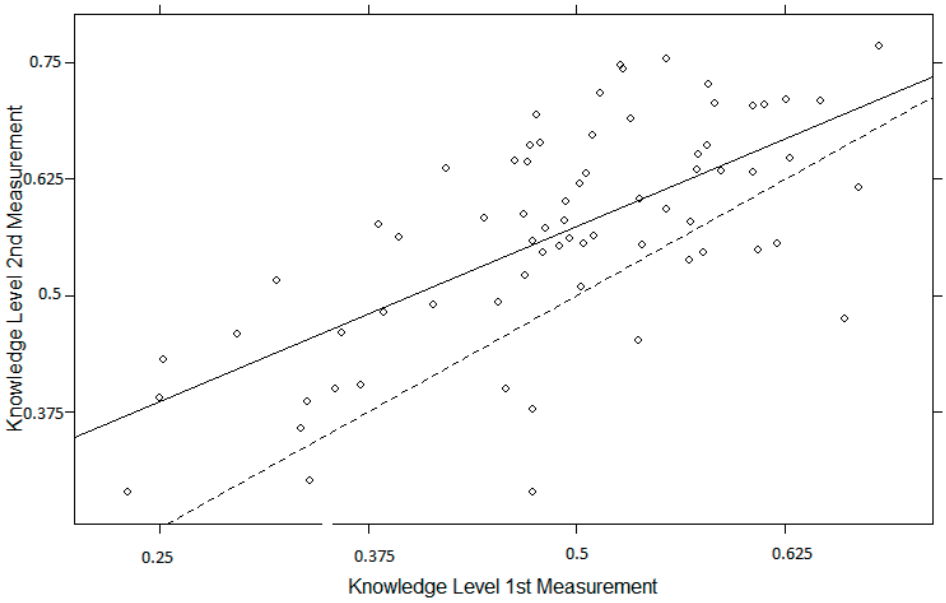
We also evaluated changes over time in the knowledge levels of patients who took the questionnaire at two points in time, during their transition process. Figure 2 showed an improvement in knowledge levels, that became smaller for patients who did well on the RTT the first time.

Reliability of the total Rotterdam Transition Test score
The internal consistency was measured using Cronbach's alpha. The reliability of the total RTT was good with a Cronbach's alpha of 0.81.

Responses of the Adolescents With IBD on the Rotterdam Transition Test
Scoring the RTT took on average 5 to 10 minutes per patient. Mean total RTT result for girls was 63 (range 39-95 ±11.1; 58% of maximal score (108)) and 55 (range 30-78 ± 10.4; 55% of maximal score (100)) for boys (p =<0.001).

The bivariate analysis, based on the knowledge score of the Rasch model, shows that adolescents who used corticosteroids in the past three months scored significantly better on the total RTT (results ranging from 0-1) (steroids (n=30); 0.64 ± 0.12 SDS vs no steroids (n=177); 0.55 ±0.17 SDS , p=0.001). Similarly, but not significant, adolescents who had a disease exacerbation in the prior year seemed to have more knowledge (exacerbation (n=100) 0.58 ±0.17SDS vs no exacerbation (n=107) 0.55 ±0.17SDS, p=0.25). This was also true for disease activity at the time of the RTT (remission (n= 148) 0.56 ± 0.16SDS vs disease activity (n=59) 0.57 ±0.18SDS, p=0.77). Adolescents who took biologics (anti-TNF medication, vedolizumab) at the time of RTT scored lower (biologics use (n=75) 0.53 ±

Figure 2: Relationship between knowledge level over time.



The x-axis denotes the knowledge level in the first assessment, and the y-axis the same level after the second assessment. The dashed lines denotes no improvement over time. The solid line is the regression line between the 2 assessments.

0.19SDS) compared to patients not using biologics ((n= 132) 0.58 ± 0.15SDS, p= 0.035), reflecting less knowledge.

Table 1 shows correct responses of the adolescents on each question of the RTT.

DISCUSSION

In this prospective, longitudinal study we demonstrated that the Rotterdam Transition Test, the first completely open IBD-knowledge questionnaire, is valid and reliable to detect gaps in knowledge in adolescents with IBD, transferring to adult care. The RTT has only open questions, drawing on various levels of knowledge, discriminating better in the higher knowledge levels. As it assesses the knowledge in adolescents, it enables health care providers to see clearly on what issues adolescents need to improve their knowledge. The RTT is a reliable questionnaire given its high Cronbach's alpha (0.81). The scoring model proved to be usable and reliable, with a Fleiss Kappa of substantial or almost

perfect agreement on all 27 questions. Major strengths of our study are its rigid methodology, as well as its prospective and longitudinal design that enabled both complete validation and preliminary testing in a large group of adolescent IBD patients.

Female sex, older age, higher educational level, better disease acceptance and level of independence from parents are significant factors contributing to higher RTT scores. These factors are logically explainable, but not all comparable to other IBD knowledge questionnaires. We found no association between levels of knowledge and time since diagnosis, which was also shown using other IBD knowledge tools (10, 15, 17, 18, 24-28), while in adult asthma and diabetes disease knowledge was reported to improve with disease duration. (29, 30) One explanation may be that core information is obtained shortly after diagnosis, while little new information is obtained subsequently or forgotten over time. Moreover, if there is little burden of disease, patients may have less interest in the disease, with knowledge decreasing over time. As repeating the RTT in one year's time shows significantly higher scores, particularly if there was a prior low knowledge level, it may be important to keep repeating important disease information to maintain patients' level of knowledge. (27) Adolescents taking prednisone in the last 3 months before the RTT had significantly higher knowledge scores, a finding that we think is clinically significant as knowledge of disease understandably increases during or shortly after a disease exacerbation. Our finding that adolescents scored better without a biological treatment is less evident and with the small difference in RTT score (0.53 vs 0.58), we are unsure about the clinical significance here. Further research in a larger cohort should provide more clarity on this.

As the RTT is the first completely open questionnaire for assessing knowledge in adolescents with IBD, there is no comparison questionnaire. If we compare the RTT questions with the open questions of MY IBD Passport (15), we see a smaller difference in percentage of adolescents correctly answering questions about diagnosis (97.2% vs 78.2%) and medication (84.1% vs 76.9%). Compared with multiple-choice questionnaires. Compared to multiple-choice questionnaires (IBD-yourself (13) and IBD-KID (17)), the open questions in the RTT seem more difficult to answer correctly: a set of questions about transition, such as ''I know who my doctor will be in the adult team'', ''I can easily identify the differences between pediatric and adult IBD care'' and ''I know the reason why there is a transition clinic'' were answered correctly by 73% of patients in the IBD-yourself, while similar, open questions in the RTT revealed that only 36% actually knew the names of the adult team, 3.4% were able to list the differences and 33.8% could explain

the meaning of transition. As both questionnaires test knowledge, this may suggest that, with multiple-choice questions, adolescents have the opportunity to hazard a guess, but that knowledge gaps are more easily detected with an open questionnaire such as the RTT. Moreover, open questions and answers given (or left open) enable healthcare providers to provide personalized education during transitional care.

Our study does have a number of limitations. Compared to a multiple-choice questionnaire, completing an open questionnaire such as the RTT takes more time: around 30 minutes. This is quite a challenge for busy adolescents, especially for those who do not wish to be aware of their disease. Our first analyses (Cronbach alpha and the Rasch analysis) did not yield any questions that did not contribute to a distinction in high and low knowledge levels. In the future, when the RTT is evaluated in a greater cohort, we will again test whether the questionnaire can be shortened without affecting completeness. Also, using the scoring model to check and score the open answers of the RTT takes more time than just counting correct multiple choice answers. In our hands, it took 5 to 10minutes to rank the responses for a test. When using the RTT is in daily practice, however, the open answers gives immediate insight into what the adolescent does and does not know about his/her disease. Another limitation about the open questionnaire is the well-known fact that school-aged adolescents may give (partially) incorrect answers while indeed knowing the right answer (31). For this reason we chose to "round up" scores when unsure, and to give our patients the "benefit of the doubt" whenever we could. The intent of the RTT also to target education to what is not known will not be affected by this, as partly incorrect answers to the questions are clearly visible to the healthcare provider. For targeting education in daily practice, knowing the total score is not important. And when we do use the total score, as in outcome research, we feel that after rounding up, patients still scoring low are the real targets for extra help. Furthermore, when asked for feedback on this open questionnaire related to knowledge, some of our adolescent patients felt like being tested or evaluated, as associated with school exams. Another limitation is that this was a single-centre study and that, hence, disease-related knowledge may have been related to the individual care received by patients. This may limit the generalizability of our findings. In our clinic, however, adolescents with IBD receive transitional care from a multidisciplinary team of IBD physicians and nurses, and patients did not have a fixed healthcare provider. In other words, there may have been variations in educational strategies applied by the IBD transition team members.

In conclusion, the RTT is a reliable and valid questionnaire with a good scoring model, to be used in the adolescent clinic to detect knowledge gaps in adolescents with IBD. This tool can identify low-scoring patients, who may be at risk for dropping out of the healthcare system after transfer to adult care. RTT results will guide transition teams as to what issues they should discuss with their adolescent IBD patients. The RTT can also be used to assess changes in knowledge over time. The overall results of testing showed a RTT score of 58% and 55% (of maximal score) in girls and boys, respectively. We plan to further validate the RTT in a multicentre cohort of adolescent IBD patients. As a next step, we aim also to investigate whether the RTT score has predictive value for transition readiness and outcome of transition. Meanwhile, by focusing on low scoring patients, who need the most help to become self-managing adults, we can improve the quality of care delivered to this vulnerable patient population.

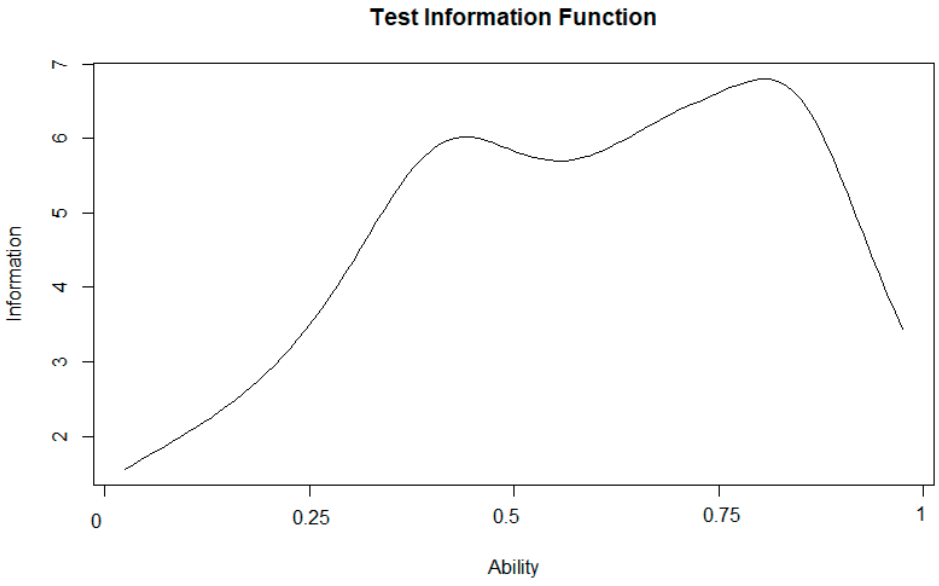
REFERENCES

1 Afzali A, Katz S Inflammatory Bowel Disease in the Baby to Baby Boomer: Pediatric and Elderly Onset of IBD. Curr Treat Options Gastroenterol 2018;16(3):289-305.
2 Kim J, Ye BD Successful Transition from Pediatric to Adult Care in Inflammatory Bowel Disease: What is the Key? Pediatr Gastroenterol Hepatol Nutr 2019;22(1):28-40.
3 van Rheenen PF, Aloi M, Biron IA, et al. European Crohn's and Colitis Organisation Topical Review on Transitional Care in Inflammatory Bowel Disease. J Crohns Colitis 2017;11(9):1032-38.
4 Hait E, Arnold JH, Fishman LN Educate, communicate, anticipate-practical recommendations for transitioning adolescents with IBD to adult health care. Inflamm Bowel Dis 2006;12(1):70-3.
5 de Silva PS, Fishman LN Transition of the patient with IBD from pediatric to adult care-an assessment of current evidence. Inflamm Bowel Dis 2014;20(8):1458-64.
6 Goodhand J, Hedin CR, Croft NM, et al. Adolescents with IBD: the importance of structured transition care. J Crohns Colitis 2011;5(6):509-19.
7 Bollegala N, Brill H, Marshall JK Resource utilization during pediatric to adult transfer of care in IBD. J Crohns Colitis 2013;7(2):e55-60.
8 Cole R, Ashok D, Razack A, et al. Evaluation of Outcomes in Adolescent Inflammatory Bowel Disease Patients Following Transfer From Pediatric to Adult Health Care Services: Case for Transition. J Adolesc Health 2015;57(2):212-7.
9 Blum RW, Garell D, Hodgman CH, et al. Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. J Adolesc Health 1993;14(7):570-6.
10 Brooks AJ, Smith PJ, Cohen R, et al. UK guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. Gut 2017;66(6):988-1000.
11 Pearlstein H, Bricker J, Michel HK, et al. Predicting Sub-Optimal Transitions in Adolescents with Inflammatory Bowel Disease. J Pediatr Gastroenterol Nutr 2020.
12 Sattoe JNT, Peeters MAC, Haitsma J, et al. Value of an outpatient transition clinic for young people with inflammatory bowel disease: a mixed-methods evaluation. BMJ Open 2020;10(1):e033535.
13 Zijlstra M, De Bie C, Breij L, et al. Self-efficacy in adolescents with inflammatory bowel disease: a pilot study of the "IBD-yourself", a disease-specific questionnaire. J Crohns Colitis 2013;7(9):e375-85.
14 Brink GVD, Gaalen M, de Ridder L, et al. Health Care Transition Outcomes in Inflammatory Bowel Disease: a Multinational Delphi Study. J Crohns Colitis 2019.
15 Benchimol EI, Walters TD, Kaufman M, et al. Assessment of knowledge in adolescents with inflammatory bowel disease using a novel transition tool. Inflamm Bowel Dis 2011;17(5):1131-7.
16 Hait EJ, Barendse RM, Arnold JH, et al. Transition of adolescents with inflammatory bowel disease from pediatric to adult care: a survey of adult gastroenterologists. J Pediatr Gastroenterol Nutr 2009;48(1):61-5.
17 Haaland D, Day AS, Otley A Development and validation of a pediatric IBD knowledge inventory device: the IBD-KID. J Pediatr Gastroenterol Nutr 2014;58(3):313-9.
18 Tung J, Grunow JE, Jacobs N Pilot Development of an Electronic Pediatric Inflammatory Bowel Disease Quiz Game. J Pediatr Gastroenterol Nutr 2015;61(3):292-6.
19 Leung Y, Heyman MB, Mahadevan U Transitioning the adolescent inflammatory bowel disease patient: guidelines for the adult and pediatric gastroenterologist. Inflamm Bowel Dis 2011;17(10):2169-73.
20 Pinzon JL, Jacobson K, Reiss J Say goodbye and say hello: the transition from pediatric to adult gastroenterology. Can J Gastroenterol 2004;18(12):735-42.
21 Fishman LN, Barendse RM, Hait E, et al. Self-management of older adolescents with inflammatory bowel disease: a pilot study of behavior and knowledge as prelude to transition. Clin Pediatr (Phila) 2010;49(12):1129-33.
22 Zorg LTi. Op eigen benen. https://www.opeigenbenen.nu/. Accessed April, 2011.
23 Baker F The Basics of Item Response Therapy. University of Maryland, College Park: ERIC Clearinghouse on Assessment and Evaluation; 2001.

24 Gumidyala AP, Plevinsky JM, Poulopoulos N, et al. What Teens Do Not Know Can Hurt Them: An Assessment of Disease Knowledge in Adolescents and Young Adults with IBD. Inflamm Bowel Dis 2017;23(1):89-96.
25 Subasinghe D, Wijekoon NS, Nawarathne NM, et al. Disease-related knowledge in inflammatory bowel disease: experience of a tertiary care centre in a developing country in South Asia. Singapore Med J 2010;51(6):484-9.
26 Kim JY, Yoon H, Hwang JS, et al. Comparison of Disease-related Knowledge of Patients With Inflammatory Bowel Disease Between the West and the East Using an Updated Questionnaire (IBD-KNOW). J Clin Gastroenterol 2019.
27 Vernon-Roberts A, Otley A, Frampton C, et al. Validation of a Revised Knowledge Assessment Tool for Children with Inflammatory Bowel Disease (IBD-KID2). Inflammatory Intestinal Diseases 2020;5(2):70-77.
28 Eaden JA, Abrams K, Mayberry JF The Crohn's and Colitis Knowledge Score: a test for measuring patient knowledge in inflammatory bowel disease. Am J Gastroenterol 1999;94(12):3560-6.
29 Firestone DN, Jimenez-Briceno L, Reimann JO, et al. Predictors of diabetes-specific knowledge and treatment satisfaction among Costa Ricans. Diabetes Educ 2004;30(2):281-92.
30 Emtner M, Hedin A, Andersson M, et al. Impact of patient characteristics, education and knowledge on emergency room visits in patients with asthma and COPD: a descriptive and correlative study. BMC Pulm Med 2009;9(43.
31 Reja U, Manfreda K, Hlebec V, et al. Open-ended vs. close-ended questions in Web questionnaires. 2003.

Supplementary Figure 1

Rasch analysis showing relation between level of knowledge



(X-axis, ranging from 0 (no knowledge) till 1 (perfect knowledge)) and information value (Y-axis)

3

Chapter 3

Validation and Reference Scores of the Transition Readiness Assessment Questionnaire in Adolescent and Young Adult IBD patients

MAC van Gaalen, E van Gijn, M van Pieterson, L de Ridder, D Rizopoulos, CJ van der Woude, JC Escher.

J Pediatr Gastroenterol Nutr 2023;77:381–8.

ABSTRACT

Objectives

Transition readiness can predict a successful transition from paediatric to adult care. This study aimed to validate and develop age-dependent reference scores for the (Dutch version of) Transition Readiness Assessment Questionnaire (TRAQ), in adolescents and young adults (AYA) with inflammatory bowel disease (IBD).

Methods

TRAQ has 20 items (score 1-5) distributed over five domains (total sum score 100) and is completed by AYA. Following the COSMIN methodology, we conducted the translation, back-to back translation, pretesting, and validation of the final TRAQ-NL questionnaire. We used a RASCH model for structural validation, hypothesis testing for construct validity, and Cronbach's alpha to demonstrate reliability. Reference scores were calculated using percentiles.

Results

250 TRAQ questionnaires were evaluated in 136 AYA with IBD (56% Crohn's disease, 58% male, median age 17.5 years (range 15.7-20.4)).

The overall mean item score was 3.87 (range 1.45-5). With good reliability (Cronbach's alpha 0.87), TRAQ-NL discriminated well between knowledge levels, especially in the lower levels. Transition readiness was defined as low, moderate, adequate, or excellent in patients with TRAQ percentile scores <25th (< 3.375 mean item score), 25th-50th (3.375 - 3.9), 50th-90th (3.91- 4.7) or >90th (>4.7). Younger patients, concomitant illness, fewer visits to the transition clinic, and parental dependence were associated with significantly lower TRAQ scores.

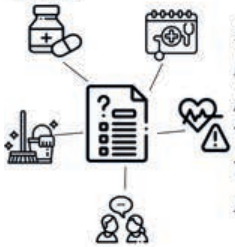
Conclusion

TRAQ(-NL) is reliable and valid, with age-dependent percentile scores to identify (in)adequate transfer readiness. TRAQ can now be more easily used as a patient-reported outcome measure to monitor transition readiness longitudinally in routine care for adolescent and young adult IBD patients.

How to score transition readiness (TRAQ)* in IBD patients

Background

The Transition Readiness Assessment Questionnaire (TRAQ) is a commonly used patient-reported outcome measure (PROM) during transition to adult care.

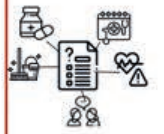


5 Domains:
Managing medication
Appointments Keeping
Tracking Health issues
Talking to providers
Managing daily activities

But without good validation and reference scores for IBD patients; how to use in daily practice?

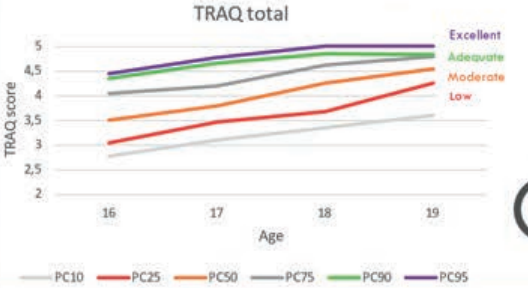
Results

136 adolescents with IBD



250 TRAQ's

Reference scores (total and domains)



Association with lower TRAQ scores

Younger patients

Concomitant illness

Parental dependence

Fewer visits transition clinic

Validation & translation in Dutch by COSMIN methodology

*Wood et al; 2014

Validation and reference scores of the Transition Readiness Assessment Questionnaire in adolescent and young adult IBD patients.
M. van Gaalen et al. 2023

JPGN Journal of Pediatric Gastroenterology and Nutrition

INTRODUCTION

Most pediatric IBD patients present during adolescence(1). During this period, in most European countries, the transition to adult health care is organized around the age of 18. Pediatric healthcare tends to be more family-centered, holistic, supportive, and focused on growth and development, with an important role for parents, while adult healthcare is patient-centred, focused on new biological treatments, cancer surveillance and reproductive health. Most importantly, patients transitioning to adult health care are suddenly expected to be self-reliant and self-managed (2-5). To ensure that these changes are not so "sudden", it is essential that adolescents are offered a structured transition program (2, 5). Transition is defined as a period during which the purposeful, planned movement of adolescents with chronic medical conditions into adult-oriented healthcare systems is organised (6). Adequate preparation on disease relevant knowledge, help in development of self-management skills and coaching of parents to gradually loosen their grip are issues that make a patient more transfer ready, so they are at less risk of adverse health outcomes (2, 7-10).

A commonly used patient-reported outcome measure (PROM) during transition is the Transition Readiness Assessment Questionnaire (TRAQ-20), a generic questionnaire (3, 11-18). TRAQ has good internal reliability and has been validated in AYA with various chronic diseases (19), but not in IBD. Although TRAQ scores are likely to increase with age, it would be helpful to have age-dependent reference values and cut-off scores that can guide the transition process in individual patients. TRAQ has been translated into several languages (20-23), but not into Dutch. With all this in mind, our research goal was to translate and validate TRAQ (TRAQ-NL) and facilitate its use in clinical care by providing age-dependent reference scores specifically for IBD.

MATERIALS AND METHODS

The study was a single-centre, longitudinal, prospective validation study that followed the COSMIN methodology(24): translation with back-to-back translation, pre-testing, performance assessment and development of baseline scores.

The Research Ethics Review Board of Erasmus MC approved this study (MEC-2017-459). Patients were asked for written informed consent for recruitment into the study.

Transition Readiness Assessment Questionnaire (TRAQ)

TRAQ is divided into five domains: Managing medication (four questions about prescriptions and how to take the medication); Appointments Keeping (seven questions about making an appointment, calling the hospital with questions, checking the results and insurances); Tracking health issues (four questions about medical history form, making a list of questions and financial help); Talking to providers (two questions about conversation with health care providers); and Managing daily activities (three questions about doing household chores). Each question is rated on a 5-point Likert scale, ranging from score 1 ("No, I don't know how") to score 5 ("Yes, I always do this when necessary"), with higher scores indicating better readiness for transition.

Translation of the TRAQ

The TRAQ was translated into Dutch after a cross-cultural adaptation process (25). Figure 1 shows the overview of the stages of development and the changes that were made. Based on cultural differences, question 9 from the original TRAQ (19); "Will you apply for health insurance if you lose your current coverage?" was changed to "When you turn 18, there will be changes regarding your health insurance; Do you know how to arrange health insurance as an adult?".

The final version of the TRAQ-NL has 20 questions with a 5-point Likert scale (identical to the original TRAQ version) (19).

IBD transition clinic

In the Netherlands, youngsters with a chronic disease are transferred from pediatric to adult care around the age of 18. Transitional care is organised depending on the local situation of the healthcare providers of both departments (pediatric and adult). In our academic hospital, where the pediatric and adult hospital are under one roof, we see all IBD patients aged 16-18 years for their regular appointments at the IBD outpatient transition clinic, located in the adult

gastroenterology department. In addition to providing regular medical care by the pediatric team, the transition clinic aims to guide adolescents in developing self-management skills, while parents receive assistance in gradually releasing their grip. The adult IBD team gets to know their future patients through a multidisciplinary meeting prior to each (biweekly) clinic, and an introductory meeting around age 17. Around age 18, both the paediatric and adult IBD teams meet with the patient and parents for a warm transfer of care. About 75% of our IBD patients are transferred within our academic hospital. The remaining 25% are referred to a hospital closer to home or near the school or university the patients chooses to go to. For these patients, a detailed transfer letter is made (with a copy sent to the patient). As of today (but not yet during the time of this study), a warm handover of care is organised as a web consultation where the pediatric IBD nurse specialist (in our clinic) and IBD nurse in adult care (in the receiving hospital) join virtually to meet with the patient and parents.

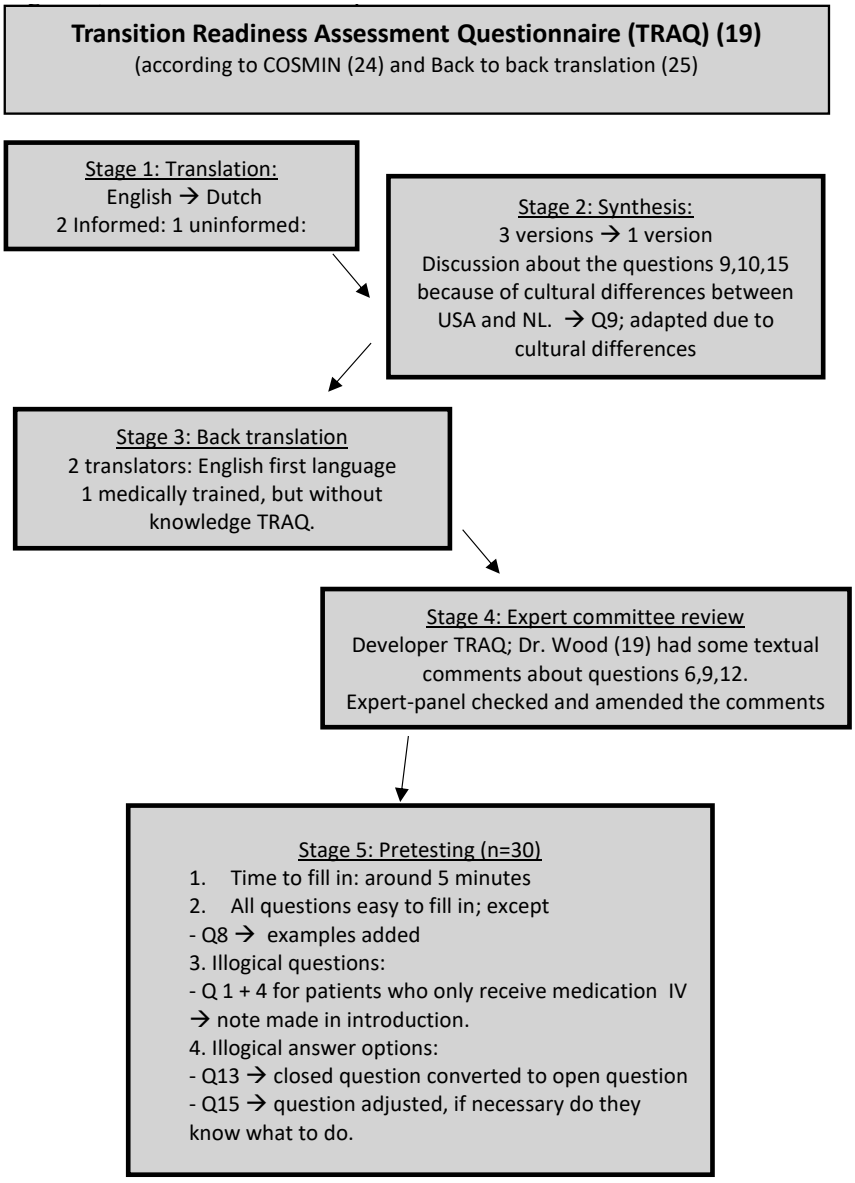
Testing of the TRAQ

Pretesting TRAQ-NL
The final stage in translation process (25) was pre-testing of TRAQ-NL.

30 IBD patients, who visited the IBD transition clinic, completed TRAQ-NL. The time to complete the TRAQ was about 5 minutes. The AYAs were then interviewed whether the questions and answer options were clear. Some questions and answer options required textual adjustments (see Figure 1).

Validity testing
In 2016-2020, all patients attending the IBD transition clinic were asked to complete the TRAQ annually during the years of their transition process (up to four time points: age 16, 17, at the last visit to the transition clinic (around 18). Post-transfer TRAQ data were only collected in patients who were transferred within our hospital and who reached the one-year post transfer moment within the timeframe of this study. So patients who went to other facilities were not asked to fill in a TRAQ questionnaire after their transfer. A Rasch analysis was used for structural validation. Rasch analysis assesses the characteristics of the questions, identifies whether questions need further modification, and assesses the ability of the questions to discriminate between different levels of transfer readiness (26). The construct validity of TRAQ-NL was tested using several hypotheses concerning relationships between TRAQ sum score and hypothesised surrogate markers of transition readiness (Table 1), (2, 3, 5, 13, 14, 27). The information needed for the hypotheses was obtained by additional questions on disease acceptance (4-point

Figure 1: Overview translation process



Likert scale to the question, "I can accept that I will have this disease for the rest of my life"), dependence on parents asking, "Have you ever been away from your parents for at least one weekend (e.g. sleepover or holiday)?", a 10-point Visual Analogue Score to self-assess readiness for transition and self-management, and a chart review (on IBD type and education level).

TRAQ (combined with the supplementary questions) was sent via a secure online survey program (Lime Survey version 3.1.1, LimeSurvey GmbH, Hamburg, Germany). Questionnaires obtained during pretesting were included in the final validation analysis, as the TRAQ version used during pretesting was essentially identical to the final version (Figure 1).

Table 1: Hypothetical constructs and assessment of validity and discriminatory ability of TRAQ

Construct	Statistic
Association of total mean TRAQ scores of patients with IBD with specific variables:	
Scores should correlate positively with patient age	Pearson correlation coefficient
Scores should correlate positively with disease duration	Pearson correlation coefficient
Scores should not differ between CD and UC patients	ANOVA
Females often score better in questionnaires than males	*t* test
Patients with lower educational levels score lower than those with higher educational levels	Post-hoc Tukey test
Scores should correlate positively with repeated TRAQ administration	Rasch model
Scores should correlate positively with higher VAS self-management	Spearman correlation coefficient
Scores should correlate positively with higher VAS transfer readiness	Spearman correlation coefficient
Patients who were without parents for more than two nights should score higher (independency score)	*t* test
Patients who accepted having IBD should score higher	*t* test

CD = Crohn disease; IBD = inflammatory bowel disease; TRAQ = Transition Readiness Assessment Questionnaire; UC = ulcerative colitis; VAS = visual analogue scale.

Reliability tests

Cronbach's alpha was used to assess the internal consistency of TRAQ-NL, which indicates the extent to which different items of a scale assess one whole.

Measuring instruments for TRAQ responses of adolescents with IBD

Paediatric Ulcerative Colitis Activity Index (PUCAI) (28) and Paediatric Crohn's Disease Activity Index (PCDAI) (29), with values >10 defined as active disease, assessed clinical disease activity at the time of completing TRAQ. Exacerbation of IBD was noted when increased symptoms or presence of endoscopic disease activity led to escalation of treatment in the year prior to assessment of TRAQ. Extra-intestinal disease (such as dermatological disorders or PSC) and other diseases (such as asthma) were recorded as concomitant disease when it led to an outpatient appointment with a specialist or new medical treatments.

Reference scores

To make TRAQ more practical in routine clinical care, age-related reference scores (between 16 and 18 years) were generated. Reference scores for both total TRAQ score and individual domains were calculated using percentiles based on TRAQ mean scores. Transition readiness was defined as moderate in patients with scores between the 25th-50th percentile, adequate in scores between the 50th-90th percentile and excellent in scores above the 90th percentile. Transition readiness was low for scores below the 25th percentile.

Statistical analysis

All statistical analyses were performed using SPSS for Windows, version 28.0.1.0 (IBM SPSS Statistics for Windows, Armonk, NY, USA). Results were considered significant at a p-value <0.05. The total mean score of TRAQ was used for the analyses. Bivariate or multivariate analyses evaluated associations between a number of independent variables and mean TRAQ scores. For some question items the response categories were merged to two options. The independent variables were the same as the hypothetical constructs in Table 1. For comparisons of the independent variables, t-test (dichotomous), Pearson's (continuous) and Spearman's (non-normally distributed) correlation coefficients were used. To examine whether education level (categorised as low, medium or high) was related to TRAQ score, we conducted an analysis of variance. TRAQ was considered as the dependent variable, and education level as the categorical predictor. The reliability of the overall TRAQ was assessed using Cronbach's alpha. If an item was missed, it was not included in the analysis, which was reflected in a reduction in the number of participants (n) for that particular analysis.

RESULTS

Testing the TRAQ

We prospectively collected 250 TRAQs from 136 adolescent patients who consecutively visited the IBD outpatient clinic (87% response rate). The median age at diagnosis was 14.5 years (IQR 3.07 (range 2.7-17.5)), 58.1% were male and 55.9% had CD. Table 2 shows the demographic data at the time the TRAQ was completed. 136 TRAQs (54.4%) were completed for the first time and almost all TRAQs (212; 84.8%) were completed before transfer to adult care. Data from one outlier (AYA scored very low, total mean score 1.45) were not used to obtain evenly distributed data (mean score range 2.25-5).

Table 2: Demographics at the time of completing TRAQ (250 questionnaires in 136 IBD patients)

		N. % or Median
Age (years)	Range	15.67-20.38
	Median; IQR	17.53; 1.36
Disease duration (years)	Range	0.12-14.71
	Median; IQR	3.14; 3.48
Timepoint of filling in TRAQ	Before transfer	212 (84.8%)
	After transfer	38 (15.2%)
Timepoint of filling in TRAQ	First time	136 (54.4%)
	Second time	77 (30.8%)
	Third time	31 (12.4%)
	Fourth time	6 (2.4%)
Duration of transitional care: Time since first visit to the IBD Transition clinic (y)	Range	0 – 4.02
	Median; IQR	0.91; 1.63
Educational level	Low (lower secondary)	142 (56.8%)
	Medium (upper secondary)	56 (22.4%)
	High (pre-university)	52 (20.8%)
Clinical Disease activity	Remission	162 (64.8%)
	Mild	68 (27.2%)
	Moderate	20 (8.0%)
	Severe	0
Current medication use	Aminosalicylates	63 (25.2%)
	Immunomodulators	124 (49.6%)
	Biologics (infusions/ injection)	112 (44.8%)
	Subcutaneous injections at home	27 (10.8%)
	Corticosteroids	25 (10%)
	Topical treatment	8 (3.2%)
	No medication	23 (9.2%)
Induction treatment with EEN/steroids used within three months before completing TRAQ	EEN	3 (1.2%)
	Steroids	35 (14%)
Prior surgery (peri-anal or bowel resection)	Yes	57 (22.8%)
Concomitant diagnosis	Yes	64 (25.6%)
	Joint disease	12 (18.8 %)
	Liver disease (PSC/AIH)	11 (17.2%)
	Joints + Liver	1 (1.6%)
	Skin disease	16 (25%)
	Other (such as asthma, epilepsy)	24 (37.5%)
Being without parents for more than two days	Yes, without problems	198 (79.2%)
	Yes, but with problems/ inconveniences	21 (8.4%)
	Never	31 (12.4%)
Acceptance of disease (n=247)	Yes, always or often	198 (80.2%)
VAS independence (n=246)	Range	0-100
	Median; IQR	80; 15
VAS transfer readiness (n=246)	Range	10-100
	Median; IQR	80; 35

AIH = autoimmune hepatitis; EEN = exclusive enteral nutrition; IQR = interquartile range; PSC = primary sclerosing cholangitis; TRAQ = Transition Readiness Assessment Questionnaire; VAS = visual analogue scale.

Structural validation of TRAQ-NL

The Rasch model (ranging from 0 to 1) showed that the TRAQ discriminated well between different levels of transfer readiness, with the best discrimination between low/absence (0) and high (0.75) levels of transfer readiness. This implies that the TRAQ is best used to identify patients with low transfer readiness. Questions that provided little information on differences between knowledge levels were questions on transport to hospital (Q7), financial overview (Q11) and preparing dinner (Q18).

Construct validity with hypothesis testing

Significant positive correlations of TRAQ total mean scores (score 1-5) were found with the following hypotheses (Table 1): older age (Pearson correlation coefficient 0.47, p <0.001), more frequent visits to the transition clinic (Pearson correlation coefficient 0.42, p <0. 001), higher VAS of self-management (Spearman correlation coefficient 0.34, p <0.001), higher VAS of transfer readiness (Spearman correlation coefficient 0.37, p <0.001), having experienced transfer of care (4.38 ±0.43 vs 3.78 ±0.63 p <0.001), being without parents for more than two days (3.91 ±0.64 vs 3.61 ±0.64, p = 0.007). Using the Rasch model, we also evaluated changes over time in the level of transfer readiness of patients who completed the TRAQ at two time points, during their transition process. This showed an increase in the level of transfer readiness; this increase was smaller in patients who already scored well on the TRAQ at the first completion time. Some patients scored lower during the second time the TRAQ was completed. Female patients scored almost significantly higher than male patients (3.95 ±0.64 vs. 3.82 ±0.63, p = 0.06). Acceptance of living with IBD seemed to correlate with transfer readiness, with (almost significantly) higher scores in those who accept compared with patients who experience acceptance problems (3.91 ±0.64 vs. 3.75 ±0.6, p = 0.06). Education level showed no correlation with TRAQ results (data not shown).

Reliability of total TRAQ score

Internal consistency was measured using Cronbach's alpha of 0.87 (good reliability of the total TRAQ).

Responses of adolescents with IBD on TRAQ

The mean total TRAQ score for all age groups was 3.88 (range 2.25-5.0; SD 0.64). Question 9 (about impending changes in health insurance) was scored lowest (mean score 2.82 (SD 1.49)). Bivariate analysis showed that adolescents with concomitant disease scored significantly lower on the overall mean score TRAQ (score 1-5) (concomitant disease (n=63); 3.7 ± 0.71 SDS versus no concomitant disease (n=186); 3.93 ±0.60 SDS, p=0.02). Other IBD-specific characteristics such as (type of) medication, disease activity, surgery, and disease type showed no correlation with TRAQ score (data not shown).

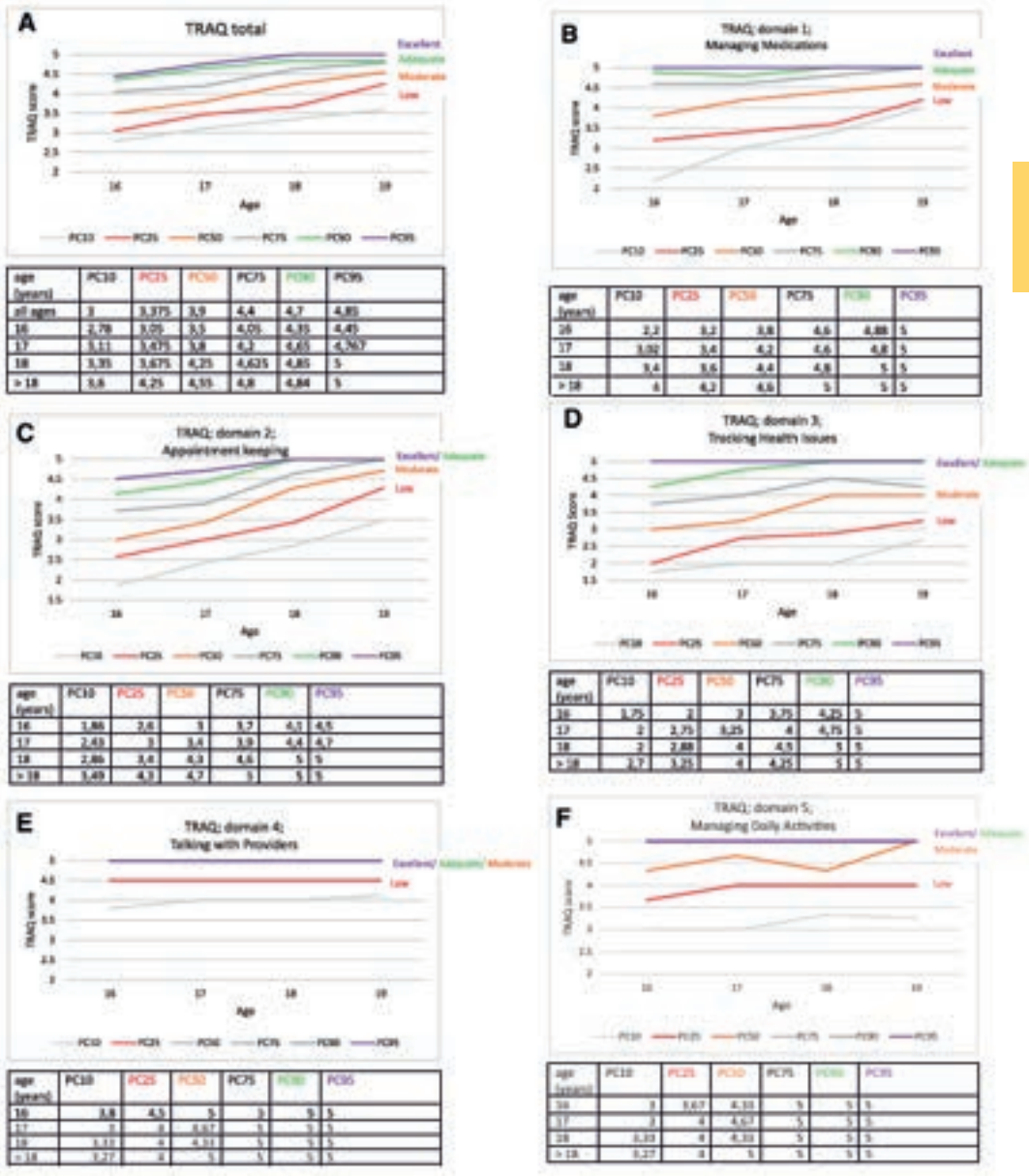
Reference scores

The graphs in Figure 2 show the age-specific percentile (PC) scores on the total TRAQ and its domains. Both total TRAQ and domain scores increased with age, and the gap between pc10 and 95 decreased with age. AYA of all ages had the most difficulty with domain 3 (keeping track of health issues) and 2 (keeping track of appointments), while domain 5 (talking to healthcare providers) seemed the easiest. Domain 4, managing daily activities, distinguished the least between the different levels of transfer readiness. Transition readiness was moderate in patients with scores between the 25th-50th percentile (total mean score 3.375 - 3.9), adequate scores in the 50th-90th percentile, (total mean score 3.91- 4.7) and excellent scores above the 90th percentile (total mean score >4.7). Transition readiness was low with a score below the 25th percentile (total mean score <3.375).

DISCUSSION

This longitudinal, prospective study aimed to facilitate the use of TRAQ in daily practice in the AYA IBD population, with a scoring model and reference values. Following the stages of back-to-back translation (25), we showed that TRAQ-NL is a valid and reliable tool to detect (in)adequate transfer readiness in adolescents with IBD transitioning to adult care. TRAQ-NL is now available for routine clinical care and research purposes (Supplement). Age-dependent percentile scores facilitate the interpretation of TRAQ and put into perspective the transfer readiness and specific needs of individual patients. Longitudinal monitoring of TRAQ as a patient-reported outcome measure will contribute to patients' understanding of their transition readiness and self-management skills. As previous studies have shown (12-15, 19-21, 30), age is the most important factor influencing patients' transition readiness. Although others have shown that girls score higher than boys (12-14, 20, 21) this difference is not significant in our cohort (p=0.06), and thus we did not show gender-specific reference data. As described in the study by Hart et al. (14), the gender difference becomes smaller as adolescents get older. This may explain why we did not find the age difference in our 16-plus cohort. We found that having concomitant diseases resulted in a significantly lower score on TRAQ. With more complicated illness, parents may be more concerned and protective, leaving less room for the development of self-management skills (13, 31). Along this line, AYA with IBD and concomitant disease were less independent of their parents (never 2 nights without parents; 15.6%) than AYA without concomitant disease (11.3%). The VAS score of transfer readiness and independence was significantly related to the transfer readiness score, as shown previously (13, 21).

Figure 2: Age-related percentile scores on the total transition readiness (TRAQ) and the domains.



age (years)	PC10	PC25	PC50	PC75	PC90	PC95
all ages	3	3,375	3,9	4,4	4,7	4,85
16	2,78	3,05	3,5	4,05	4,35	4,45
17	3,11	3,475	3,8	4,2	4,65	4,767
18	3,35	3,675	4,25	4,625	4,85	5
> 18	3,6	4,25	4,55	4,8	4,84	5

age (years)	PC10	PC25	PC50	PC75	PC90	PC95
16	2,2	3,2	3,8	4,6	4,88	5
17	3,02	3,4	4,2	4,6	4,8	5
18	3,4	3,6	4,4	4,8	5	5
> 18	4	4,2	4,6	5	5	5

age (years)	PC10	PC25	PC50	PC75	PC90	PC95
16	1,86	2,4	3	3,7	4,1	4,5
17	2,43	3	3,4	3,9	4,4	4,7
18	2,86	3,4	4,3	4,6	5	5
> 18	3,49	4,3	4,7	5	5	5

age (years)	PC10	PC25	PC50	PC75	PC90	PC95
16	1,75	2	3	3,75	4,25	5
17	2	2,75	3,25	4	4,75	5
18	2	2,88	4	4,5	5	5
> 18	2,7	3,25	4	4,25	5	5

age (years)	PC10	PC25	PC50	PC75	PC90	PC95
16	3,8	4,5	5	5	5	5
17	3	4	4,67	5	5	5
18	3,33	4	4,33	5	5	5
> 18	3,27	4	5	5	5	5

age (years)	PC10	PC25	PC50	PC75	PC90	PC95
16	3	3,67	4,33	5	5	5
17	3	4	4,67	5	5	5
18	3,33	4	4,33	5	5	5
> 18	3,27	4	5	5	5	5

(A) TRAQ total. (B) TRAQ domain 1: Managing medications. (C) TRAQ domain 2: Appointment keeping. (D) TRAQ domain 3: Tracking health issues. (E) TRAQ domain 4: Talking with providers. (F) TRAQ domain 5: Managing daily activities. TRAQ = Transition Readiness Assessment Questionnaire.

As described by others (12, 14, 20, 21, 30) domain 2; "Keeping an appointment" and 3; "Detecting health problems" have the lowest scores, implying that more attention should be paid to these domains during a structured transition program.

TRAQ is an extensively validated questionnaire used in different diseases and in different languages (21-23). Until now, we did not know which cut-off scores identify patients who are not yet ready for transfer. With age-specific reference percentile scores, results from the TRAQ domains will guide the type of support to be provided to individual patients transitioning to adult care. Comparing our results with US data (14),the only other study that has produced TRAQ reference data for IBD patients, our cohort appears to score higher than US patients in all domains. One explanation is the different patient selection in the US data, that was extracted from a large database of the patient organization, with no information on availability or type of transitional care that was offered. The largest "transcontinental" difference in mean TRAQ score occurred at age 17. This may be due to the success of the structured transition program we offer at the IBD transition outpatient clinic, starting at age 16. The patients in the US may not have had similar high quality transitional care, and in our opinion this emphasizes the need to organize transitional care in a structured way. The program aims to increase disease-related knowledge and independence to prepare AYA for adult care. What underlines this line of thought is that having more appointments at the transition clinic correlated positively with TRAQ score (Pearson's correlation coefficient 0.42, $p < 0.001$). The difference between the US cohort and ours disappeared on almost all domains around age 18. This may again indicate that age is the most important determinant of transition readiness. Looking at the increase in TRAQ score during the adolescent years, the upward curve in TRAQ score in our cohort is more gradual than in the US cohort. Our overall mean TRAQ score varies by age from 0.2-0.4 per year, while in the US it ranges between 0.1-0.6, with the largest increase between 17 and 18 years (0.6). This may be an indication that a transition outpatient clinic (starting at age 16) leads to a smoother increase in self-management skills.

TRAQ states that it measures transition readiness, but the question remains: when is a patient truly "transfer-ready"? And one can also question whether our adolescent IBD patients can and should be completely ready at age 18, at the time of transfer. In our opinion, transition and transfer should be organized together with a co-responsible adult care team that is well informed about the readiness of their new patient. That way, the adult care team does not simply accept a "perfect patient," but (in the ideal situation) continues to provide transitional care as usual. In current practice, however, not all hospitals have a transition clinic with

a structured program, and pediatricians need to know when their patient can be safely and properly transferred to adult care with minimal risk of dropping out of medical care. To truly know the implications of a patient's readiness for transition, we need to prospectively study the relationship between transfer readiness and the outcome of transition (32), which we intend to do in the near future.

Our study has some limitations, the first being age as a confounding factor in TRAQ monitoring. This also makes it difficult to say what is the exact effect of transition interventions over time, such as frequency of appointments on the IBD transition clinic, repeated completion of TRAQ or assessment of TRAQ after transfer (in patients older than 18 years). Another limitation is the single-centre design. All our patients were offered the same high quality structured transitional care which may have affected the level of TRAQ scores. Thus, the reference values we generated may be higher than in other cohorts and should be validated in a larger multicentre study. In addition, to make TRAQ useful in daily practice, we developed a digital dashboard in our electronic patient record. Each time the AYA completes the TRAQ digitally (once or twice a year, always prior to a visit to the transition clinic), the progression of total and domain scores is displayed against a background of age-related reference values. The TRAQ dashboard is part of a value-based healthcare dashboard that facilitates monitoring of self-management skills, quality of life and mental health issues.

In conclusion, TRAQ-NL is a valid and reliable patient-reported outcome measure of transfer readiness. It can support both pediatric and adult caregivers in guiding individual AYA with IBD toward independence and self-management. Finally, engaging patients by repeatedly asking them to self-report transition readiness will have a positive effect on skill development.

References

1 Ghione S, Sarter H, Fumery M, et al. Dramatic Increase in Incidence of Ulcerative Colitis and Crohn's Disease (1988-2011): A Population-Based Study of French Adolescents. Am J Gastroenterol 2018;113(2):265-72.
2 van Rheenen PF, Aloi M, Biron IA, et al. European Crohn's and Colitis Organisation Topical Review on Transitional Care in Inflammatory Bowel Disease. J Crohns Colitis 2017;11(9):1032-38.
3 van Gaalen MAC, van Pieterson M, van den Brink G, et al. Rotterdam Transition Test: A Valid Tool for Monitoring Disease Knowledge in Adolescents With Inflammatory Bowel Disease. J Pediatr Gastroenterol Nutr 2022;74(1):60-67.
4 de Silva PS, Fishman LN Transition of the patient with IBD from pediatric to adult care-an assessment of current evidence. Inflamm Bowel Dis 2014;20(8):1458-64.
5 Brooks AJ, Smith PJ, Cohen R, et al. UK guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. Gut 2017;66(6):988-1000.
6 Blum RW, Garell D, Hodgman CH, et al. Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. J Adolesc Health 1993;14(7):570-6.
7 van den Brink G, van Gaalen MAC, Zijlstra M, et al. Self-efficacy did not predict the outcome of the transition to adult care in adolescents with inflammatory bowel disease. Acta Paediatr 2019;108(2):333-38.
8 Kim J, Ye BD Successful Transition from Pediatric to Adult Care in Inflammatory Bowel Disease: What is the Key? Pediatr Gastroenterol Hepatol Nutr 2019;22(1):28-40.
9 Cole R, Ashok D, Razack A, et al. Evaluation of Outcomes in Adolescent Inflammatory Bowel Disease Patients Following Transfer From Pediatric to Adult Health Care Services: Case for Transition. J Adolesc Health 2015;57(2):212-7.
10 Crume B, Mitchell PD, Fishman LN Infusion Appointment Self-Management Among Adolescents With Inflammatory Bowel Disease. J Pediatr Gastroenterol Nutr 2023;76(3):319-24.
11 Carlsen K, Haddad N, Gordon J, et al. Self-efficacy and Resilience Are Useful Predictors of Transition Readiness Scores in Adolescents with Inflammatory Bowel Diseases. Inflamm Bowel Dis 2017;23(3):341-46.
12 Arvanitis M, Hart LC, DeWalt DA, et al. Transition Readiness Not Associated With Measures of Health in Youth With IBD. Inflamm Bowel Dis 2021;27(1):49-57.
13 Gray WN, Holbrook E, Morgan PJ, et al. Transition readiness skills acquisition in adolescents and young adults with inflammatory bowel disease: findings from integrating assessment into clinical practice. Inflamm Bowel Dis 2015;21(5):1125-31.
14 Hart LC, Arvanitis M, Sawicki GS, et al. Trajectories of the Transition Readiness Assessment Questionnaire Among Youth With Inflammatory Bowel Disease. J Clin Gastroenterol 2022;56(9):805-09.
15 Rosen D, Annunziato R, Colombel JF, et al. Transition of Inflammatory Bowel Disease Care: Assessment of Transition Readiness Factors and Disease Outcomes in a Young Adult Population. Inflamm Bowel Dis 2016;22(3):702-8.
16 Sönmez HE, Koç R, Karadağ Ş G, et al. The readiness of pediatric rheumatology patients and their parents to transition to adult-oriented treatment. Int J Rheum Dis 2021;24(3):397-401.
17 Sawicki GS, Lukens-Bull K, Yin X, et al. Measuring the transition readiness of youth with special healthcare needs: validation of the TRAQ--Transition Readiness Assessment Questionnaire. J Pediatr Psychol 2011;36(2):160-71.
18 Teh KL, Hoh SF, Chan SB, et al. Transition readiness assessment in adolescents and young adults with rheumatic diseases: The Singapore experience. Int J Rheum Dis 2022;25(3):344-52.
19 Wood DL, Sawicki GS, Miller MD, et al. The Transition Readiness Assessment Questionnaire (TRAQ): its factor structure, reliability, and validity. Acad Pediatr 2014;14(4):415-22.
20 Chapados P, Aramideh J, Lamore K, et al. Getting ready for transition to adult care: Tool validation and multi-informant strategy using the Transition Readiness Assessment Questionnaire in pediatrics. Child Care Health Dev 2021;47(5):645-53.
21 González F, Roizen M, Rodríguez Celin ML, et al. Validation of the Argentine Spanish version of Transition Readiness Assessment Questionnaire for adolescents with chronic conditions.

Validación español-argentina del cuestionario de transición a la atención médica del adulto en adolescentes con enfermedades crónicas. Arch Argent Pediatr 2017;115(1):18-27.
22 Sato Y, Ochiai R, Ishizaki Y, et al. Validation of the Japanese Transition Readiness Assessment Questionnaire. Pediatr Int 2020;62(2):221-28.
23 Anelli CG, Len CA, Terreri M, et al. Translation and validation of the Transition Readiness Assessment Questionnaire (TRAQ). J Pediatr (Rio J) 2019;95(2):180-87.
24 Mokkink LB, de Vet HCW, Prinsen CAC, et al. COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. Qual Life Res 2018;27(5):1171-79.
25 Beaton DE, Bombardier C, Guillemin F, et al. Guidelines for the process of cross-cultural adaptation of self-report measures. Spine (Phila Pa 1976) 2000;25(24):3186-91.
26 Baker F The Basics of Item Response Therapy. University of Maryland, College Park: ERIC Clearinghouse on Assessment and Evaluation; 2001.
27 Pearlstein H, Bricker J, Michel HK, et al. Predicting Sub-Optimal Transitions in Adolescents with Inflammatory Bowel Disease. J Pediatr Gastroenterol Nutr 2020.
28 Turner D, Hyams J, Markowitz J, et al. Appraisal of the pediatric ulcerative colitis activity index (PUCAI). Inflamm Bowel Dis 2009;15(8):1218-23.
29 Hyams JS, Ferry GD, Mandel FS, et al. Development and validation of a pediatric Crohn's disease activity index. J Pediatr Gastroenterol Nutr 1991;12(4):439-47.
30 Cleverley K, Davies J, Allemang B, et al. Validation of the Transition Readiness Assessment Questionnaire (TRAQ) 5.0 for use among youth in mental health services. Child Care Health Dev 2023;49(2):248-57.
31 Paine CW, Stollon NB, Lucas MS, et al. Barriers and facilitators to successful transition from pediatric to adult inflammatory bowel disease care from the perspectives of providers. Inflamm Bowel Dis 2014;20(11):2083-91.
32 Brink GVD, Gaalen M, de Ridder L, et al. Health Care Transition Outcomes in Inflammatory Bowel Disease: a Multinational Delphi Study. J Crohns Colitis 2019.

4

Chapter 4

Implementing Routine Medical and Mental Health Screening in Children and Adolescents with Inflammatory Bowel Disease

M van Dalen, MAC van Gaalen, MM Favejee, MS den Hollander – Ardon, K Dulfer, L de Ridder, JC. Escher.

J Pediatr Gastroenterol Nutr 2025;Accepted

ABSTRACT

Objectives

Living with Inflammatory Bowel Disease (IBD) can have significant impact on children. Many children with IBD experience symptoms of anxiety or depression. Routine screening for mental health has been recommended. This preregistered study aimed to describe the implementation of Patient Reported Outcome Measures (PROMs) in routine healthcare at an outpatient IBD clinic, as well as assess its feasibility.

Methods

Prior to every outpatient clinic appointment, PROMs were distributed to all patients aged eight and older, and their parents. PROMs related to anxiety, depression, fatigue, pain and IBD-related quality of life and were selected by a paediatric gastroenterologist and psychologist, and integrated into electronic health records. Patients who completed PROMs on two occasions were invited to complete a user experience survey, focussing on their experiences with the PROMs.

Results

A total of 2910 questionnaires were distributed. Adherence was 44.7%, with 175 patients or parents completing at least one questionnaire. User experience results of 24 patients showed that they were satisfied with both the patient portal and discussion with the healthcare provider. Five patients perceived the length of the questionnaires as too long, or as having to complete the questionnaires too frequently. Outcomes of 114 patients with 187 sets of questionnaires, described in the supplement, showed that up to 82% reported pain and/or fatigue. About 20% of patients reported symptoms of anxiety and/or depression.

Conclusions

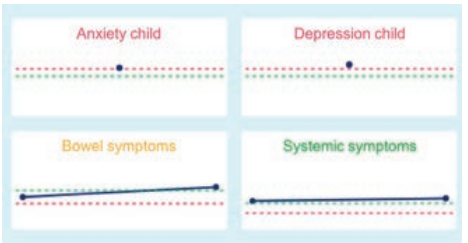
Implementation of PROMs is feasible, but the length and frequency of PROMs can be improved. Healthcare professionals wanting to work with PROMs should carefully decide which PROMs to select.

How should medical and mental health screening be implemented into the outpatient clinic?

Background

Adolescents with Inflammatory Bowel Disease (IBD) are at risk for anxiety and depression

Routine screening for medical and mental health outcomes is recommended



Dashboard in electronic health record

Implementation in outpatient clinic

Patients aged 8 and older. Self-report

One caregiver reports on child. Proxy-report

Patient-Reported Outcome Measures (PROMs) on anxiety, depression, fatigue, pain and IBD-related quality of life

Every 3 months

Discussed by healthcare provider during outpatient appointment

Compliance & results

2910 PROMs to 221 patients & parents
44.7% completed

20% symptoms of anxiety or depression

User experience

24 patients (median age 15.2)

Satisfied with PROMs, patient portal & healthcare provider

Less frequent PROMs

Implementing routine medical and mental health screening in children and adolescents with inflammatory bowel disease. van Dalen et al. (2025)

JPGN Journal of Pediatric Gastroenterology and Nutrition

4

INTRODUCTION

Paediatric and adolescent patients with Inflammatory Bowel Disease (IBD) can experience adverse psychosocial outcomes. For instance, they frequently encounter stigmatisation, regularly leading to social withdrawal.[1] Paediatric-onset IBD has been associated with mood disorders in childhood and adulthood[2]. Specifically, meta-analyses showed that 4.2-6% of adolescents with IBD are diagnosed with an anxiety disorder and 3.4-4% with a depressive disorder.[3,4] Symptoms of anxiety and depression occur in 16.4% and 15.0% of adolescents respectively.[3] These psychological problems are likely to have a bidirectional relationship to IBD-related physical problems,[5,6] although large, prospective, high-quality studies are currently lacking. Adolescents with IBD are also at increased risk of suicidal ideations and suicide attempts.[7]

As symptoms of anxiety and depression may lead to poor treatment adherence, higher morbidity and higher mortality,[8] screening for these symptoms at least once a year has been recommended both in research[9] and in clinical guidelines[10] to ensure early detection of psychosocial problems.

In recent years, screening for mental and medical health outcomes has been implemented in the context of value-based healthcare.[11,12] In outpatient clinics with adults, implementation of Patient Reported Outcome Measures (PROMs) has led to significantly fewer endoscopies, fewer surgeries, hospitalisations, emergency department visits and imaging studies.[13] Overall, IBD-related healthcare costs were 16% lower than expected.[14] In paediatric IBD patients, the Patient-Reported Outcomes Measurement Information System (PROMIS) Paediatric domain questionnaires have been shown to be responsive to changes in disease activity and quality of life.[15,16] These questionnaires assess symptoms of anxiety and depression, as well as health-related quality of life.

There is currently no gold standard for PROM implementation in paediatric IBD healthcare. We provide an example and use-case of how PROMs could be implemented in clinical care. We aimed to 1) describe the implementation of PROMs in routine outpatient healthcare for children and adolescents with IBD in an academic children's hospital, and 2) assess the feasibility of implementing PROMs on mental health and quality of life and its associated outcomes. We defined feasibility as asking *"whether something can be done, should we proceed with it, and if so, how"*.[17] Exploratory results on the PROM outcomes of 114 patients and referrals to psychological care can be found in the supplemental materials.

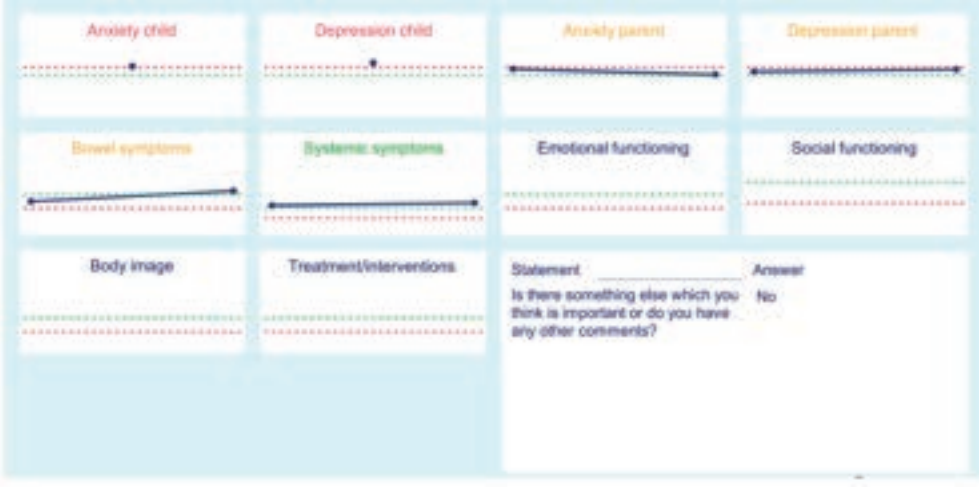
METHODS

This study was registered prospectively in the Open Science Framework (OSF; https://osf.io/ef8wm). The Medical Research Ethics Committee (MREC) of the Erasmus MC rated this study as exempt research (MEC-2022-0222). This study was conducted in accordance with the Declaration of Helsinki.[18]

IMPLEMENTATION

From May to November 2022, the PROMs were implemented into standard healthcare procedures. PROMs on mental and medical health outcomes were selected by a paediatric gastroenterologist and a psychologist. Patients reported on their own functioning, while parents also reported on their child's functioning. The distribution of PROMs was automated and linked to electronic health records. This allowed patients and parents to complete the PROMs through the patient portal and allowed healthcare providers to view the patient's results directly in the electronic health record. A dashboard was built, with the colours green, orange and red aiding the interpretation of the scores as normal, slightly elevated (+1 SD) and highly elevated (+2 SD) respectively. An example of the dashboard is shown in

Figure 1: Example of a dashboard integrated into the electronic health record



Note. Each blue dot in the dashboard represents a point of measurement. A single dot indicates one point of measurement, two dots indicate that a patient has completed two measurements. An absence of dots indicates that the patient has not provided any data related to this construct.

Figure 1. This dashboard was also designed to be used to discuss the results with patients and parents and decide together if referral to psychological healthcare is necessary. As part of routine care, patients and their parents received PROMs seven days prior to their scheduled appointment at the outpatient clinic, with a time interval of three months. This was done through an automated process. Patients or parents received an email notification that there were questionnaires for them to complete. They could then log into the electronic patient portal to complete the questionnaires in a secured environment.
Patients aged 8 and older completed all PROMIS measures, but not the IMPACT-III. Patients aged 9 and older completed all the PROMIS measures and the IMPACT-III.

To aid healthcare providers in the discussion of possible symptoms of anxiety and depression, a flow-chart and pocket cards were created. The flow-chart showed possible actions to be taken in the case of a green, orange or red score. The pocket cards detailed information on symptoms of anxiety and depression in children and adolescents, and possible questions to ask the patient and parents. This way, an informed decision could be made on whether to refer to psychological healthcare or not.

PARTICIPANTS

Recruitment for the feasibility study took place from November 2022 to the end of August 2023. Patients were eligible for participation if they 1) had an IBD diagnosis (Crohn's disease, ulcerative colitis or IBD-U), 2) were 8-18 years old, 3) were able to read and answer Dutch questionnaires and 4) completed PROMs on two occasions. Patients with an intellectual disability were excluded.

INSTRUMENTS/MATERIALS

Patient demographics

Patient characteristics were extracted from their electronic health records. These characteristics were: sex, age, medical diagnosis (Crohn's disease, ulcerative colitis or IBD-U), age at diagnosis, current medication use and disease activity. Medication use was categorised into the following categories: Oral 5-aminosalicylates, immunomodulators, biologics, corticosteroids, nutritional treatment and no current treatment. Disease activity was divided into remission, mild, moderate and

severe, based on a Physician Global Assessment (PGA).[19] Disease duration was calculated based on the age at diagnosis and the age of completing the first PROM.

Quality of life

Quality of life was assessed using both a generic and a disease-specific questionnaire. Generic quality of life was measured using the PROMIS Pediatric Scale v1.0 – Global Health 7+2 and the PROMIS Parent Proxy Profile v1.0 – Global Health 7+2. Both forms consist of nine questions and are completed by patients and their parent/caregiver respectively. Questions are rated on a 5-point Likert scale covering two domains (physical and mental health) and one question on general quality of life. Higher scores indicate better functioning. The forms have internal consistencies of $\alpha = .88$ for the paediatric form and $\alpha = .84$ for the parent proxy.[20]

Disease-specific quality of life was measured using the IMPACT-III questionnaire[21] completed by patients. The IMPACT-III contains 35 questions that are rated on a 5-point Likert scale. Higher scores indicate better quality of life. Answers were summed to create scores in several domains: bowel symptoms, systemic symptoms, emotional functioning, social functioning, body image and treatment/interventions. Internal consistency in these domains ranges from $\alpha = .57$ to $\alpha = .85$.[22]

Anxiety

Anxiety was assessed using the PROMIS Pediatric Short Form v2.0 – Anxiety 8a and the PROMIS Parent Proxy Short Form v2.0 – Anxiety 8a.[23] Both forms consist of eight questions and were completed by the patient and parent/caregiver respectively. Questions were rated on a 5-point Likert scale and higher scores indicate more symptoms of anxiety. Internal consistencies are $\omega = .89$ for the pediatric form and $\omega = .87$ for the proxy form.[24]

Depression

Depression was assessed using the PROMIS Pediatric short Form v2.0 – Depressive symptoms 8a and the PROMIS Parent Proxy Short Form v2.0 – Depressive symptoms 6a.[23] The forms were completed by the patient and the parent/caregiver respectively. The patient form consists of eight questions and the proxy form of six questions. Questions were rated on a 5-point Likert scale and higher scores indicate more depressive symptoms.

Cognitive functioning

Cognitive functioning was assessed using the PROMIS Pediatric Short Form v1.0 – Cognitive Function 7a and the PROMIS Parent Proxy Profile v1.0 – Cognitive Function 7a.[25] Both forms contain seven questions and are completed by the patient

and the parent/caregiver respectively. Questions are rated on a 5-point Likert scale and higher scores indicate better cognitive functioning.

Peer relationships

The perceived quality of peer relationships was measured using the PROMIS Pediatric short Form v2.0 – Peer Relationships 8a and the PROMIS Parent Proxy Short Form v2.0 – Peer relationships 7a.[26] The forms were completed by the patient and the parent/caregiver respectively. The patient form consists of eight questions and the parent form of seven questions. Questions were rated on a 5-point Likert scale and higher scores indicate better perceived quality of peer relationships.

Feasibility and user experience

A survey was created by a psychologist (MvD) and paediatric gastroenterologist (JE). The survey measures user experience related to the PROMs, the way the PROMs were discussed with patients during their hospital appointment and their experiences with the patient dashboard. The survey consisted of a mix of open-ended questions (e.g. "What was it like for you to complete the questionnaires?") and statements with a 5-point Likert scale ranging from completely disagree to completely agree (e.g. "It was easy to complete the questionnaires"). The survey consisted of 26 questions. The full survey is available in the supplemental materials .

PROCEDURE

Patients and their caregivers were informed of the feasibility study by their treating clinician. They received information letters through e-mail and gave electronic informed consent. If the patient was younger than 16 years old, informed consent was obtained from both legal parents/caregivers and assent was obtained from the patients. For patients aged 16 or older informed consent was obtained from the adolescent. Parents also signed informed consent for their data (i.e., answers to PROMs) to be used in the current study. Participants did not receive incentive upon participation.

PREREGISTERED STATISTICAL ANALYSIS

In this study we defined adequate feasibility as favourable feedback ratings (>3 on a 5-point Likert scale) by patients and a completion rate of ≥50% of scheduled measurements.

Quantitative data were analysed using descriptive statistics. Medians and interquartile ranges were calculated for the closed questions of the user experience survey.

Qualitative data were analysed using inductive content analysis[27] at a manifest level. Two researchers (MvD, MvG) read and reread the responses to familiarise themselves with the data. They simultaneously developed codes on a manifest level. These codes were discussed during a consensus meeting and code sheets were developed. A total of 41.7% of the data was coded independently. Intercoder-reliability was 81.1%. These codes sheets were used by one of the researchers (MvD) to code the remainder of the data.

All analysis scripts and corresponding output are publicly available on the OSF repository (https://osf.io/p2gjk/).

RESULTS

Implementation

Between the 7th of November 2022 and the 25th of July 2023, a total of 2910 questionnaires were sent to 221 patients and their parents. Of these, 1609 (44.7%) were completed. A total of 175 patients or parents completed at least one questionnaire. For children, depending on the age and PROM category, 25% to 50.4% of questionnaires was completed. For the proxy measures, the number of completed questionnaires ranged from 40% to 44.6%.

Feasibility

A total of 72 patients were approached for completing the user experience questionnaire. One person was not approached due to time constraints. Of these 72 patients, 16 declined participation, 26 received the user experience questionnaire and 30 did not respond to the request. Due to online recruitment process, reasons for non-participation are unknown. One person received the questionnaire, but did not complete it, resulting in a completion rate of 96.2%. One additional questionnaire was excluded from analysis, as this had been completed by a parent. Of the remaining 24 completers the median age was 15.2 years (IQR = 13.9-16.8) and 37.5% was male. Fourteen patients were diagnosed with Crohn's disease, nine with ulcerative colitis and one with IBD-U.

User experience relating to PROMs

All closed questions were rated from 0 (completely disagree) to 5 (completely agree). Patients were fairly neutral when rating whether they had to complete questionnaires too often (Median = 3, IQR = 2-3) or whether the questionnaires were too long (Median = 3, IQR = 2-4).

Results of content analysis are shown in Figure 2, panel A. Patients reported both negative and positive experiences related to the PROMs. The majority of negative responses related to perceiving the questionnaires as too long or having to complete them too often (n = 5). The most often reported positive experience related to the clarity of the questions (n = 8) or were positive in general (n = 12; i.e., participants reporting the questionnaires as "ok to complete"). When asked for changes, three patients reported wanting more room for own comments, or an option "other" with questions that were not applicable to them. Explanation of the codes is available in Table 1.

User experience relating to the patient portal

The patients thought the invitation e-mail they received was clear (Median = 4, IQR = 4-5) and everyone agreed or totally agreed that it was easy to log in (median = 4, IQR = 4-5). No one thought it was unclear which questionnaire was intended for the parent and which questionnaire was intended for the patient (Median = 4, IQR = 4-5).

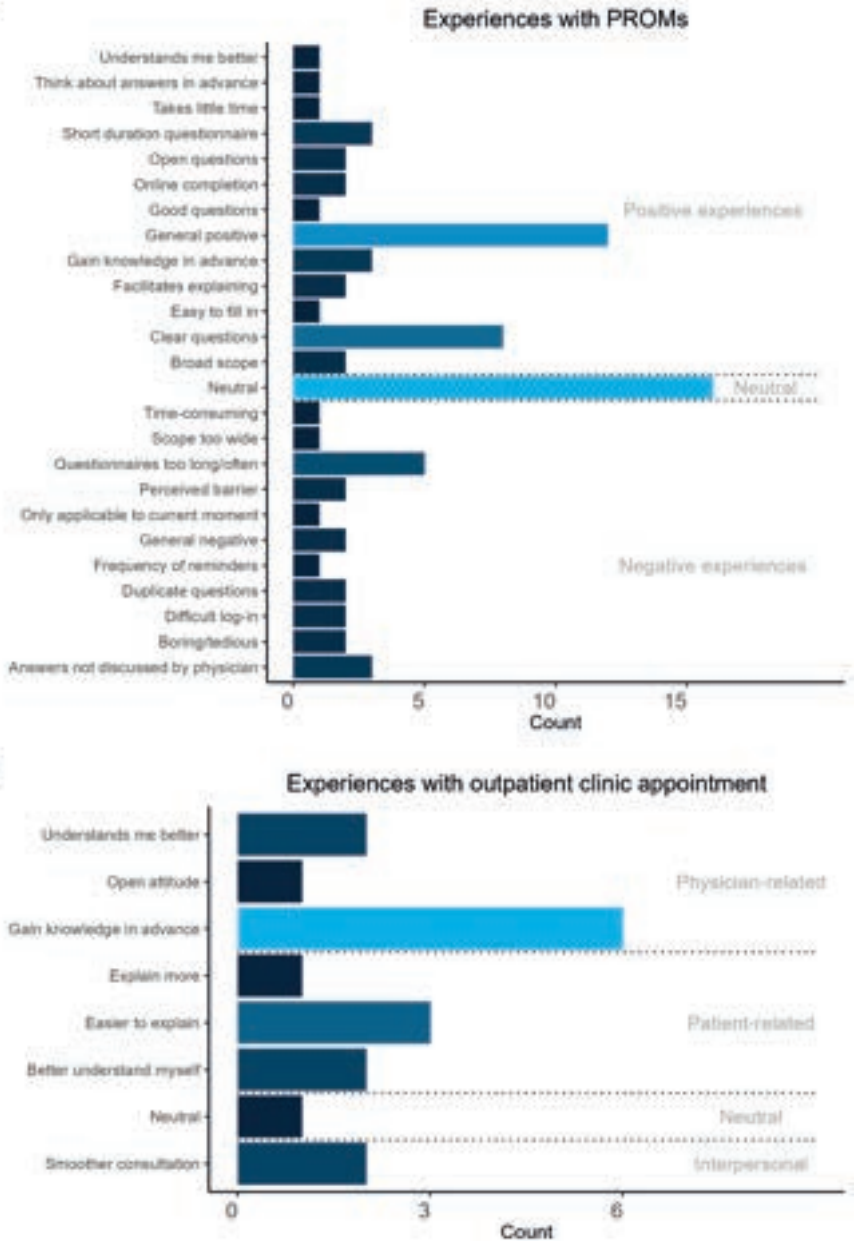
Open questions were asked to those rating the statements with disagree or totally disagree. One person did not like the fact that their parents received the invitation e-mail. Another person thought it was hard to login with their digital identification (*DigiD)*. No other feedback was provided by participants.

User experience relating to the healthcare provider

On average, patients were neutral when asked if the questionnaires allowed their healthcare professional to better help them (Median = 3, IQR = 3-4) or to better understand them (Median = 3, IQR 3-4).

A total of 14 participants (58.3%) indicated that the questionnaires were discussed with them by their healthcare professional. Overall, they were neutral to satisfied with the way they were discussed (Median = 3.5, IQR = 3-4).

Figure 2: Bar charts showing the results of the content analysis on user experience.



The length of the bars indicates the frequency of the code. Barchart A shows results relating to questions on positive and negative experiences with the PROMs. Barchart B shows results relating to questions on positive and negative experience with the discussion with the healthcare provider during the outpatient clinic appointment.

Table 1: Description of codes for user experience related to PROMs

Category name	Category description
Negative experiences	
Answers not discussed by physician	The answers to the PROMs were not discussed by the healthcare providers
Boring/tedious	The completion of PROMs was perceived as boring or burdensome
Difficult log-in	Patient reported difficulties logging in to the patient portal
Duplicate questions	Questions in the PROMs were perceived as occurring more than once, or as slightly the same
Frequency of reminders	The frequency of reminders was not correct
General negative	Short negative comments without elaboration (i.e., patients reporting they appreciated 'nothing' about the PROMs)
Only applicable to current moment	PROMs were perceived to be a snapshot and not applicable to other moments in time
Perceived barrier	Patient experienced motivational barrier
Questionnaires too long/ often	PROMs were being send out too often or were too long
Scope too wide	The questions in the PROMs were perceived as to broad
Time-consuming	It was time-consuming to complete the PROMs
Neutral experiences	
Neutral	General neutral comments to questions
Positive experiences	
Broad scope	The wide scope of the PROMs was appreciated
Clear questions	The questions in the PROMs were worded clearly
Easy to fill in	It was easy to complete the PROMs
Facilitates explaining	Completing the PROMs facilitates explaining patients' symptoms to their healthcare provider
Gain knowledge in advance	Appreciation by the patient that the healthcare provider knows about their symptoms before the consultation in the outpatient clinic
General positive	Short positive comments without elaboration (i.e., 'fine' or 'ok')
Good questions	The questions in the PROMs were perceived as good (without further elaboration)
Online completion	Patients appreciated the option to complete the PROMs online
Open questions	Appreciation of the open questions included in the PROMs
Short duration questionnaire	The duration of the PROMs was perceived as short
Takes little time	The completion of the PROMs took little time
Think about answers in advance	Completion of the PROMs forced the patient to think about their answers before the consultation at the outpatient clinic
Understands me better	The healthcare professional understood the patient better by having access to the PROMs

Results of the content analysis are shown in Figure 2, panel B. The themes related to interpersonal factors, patient-related factors, physician-related factors and neutral responses. Overall, patients appreciated that their physician had knowledge in advance to their outpatient clinic appointment (n = 6). Some patients also thought it was easier to explain using the PROMs (n = 2) or that the PROMs helped them understand themselves better (n = 2). Explanation of the codes is available in Table 2.

Table 2: Description of codes for user experience related to healthcare providers

Category name	Category description
Interpersonal	
Smoother consultation	The PROMs enabled smoother consultation and easier conversation with the healthcare provider
Neutral	
Neutral	General neutral responses
Patient-related	
Better understand myself	The PROMs helped the patient to better understand themselves
Easier to explain	It is easier to explain in-person
Explain more	The PROMs helped the patient to explain things more elaborately
Physician-related	
Gain knowledge in advance	The PROMs helped the healthcare provider to gain knowledge in advance, thereby being able to better help the patient
Open attitude	The healthcare provider had an open attitude and the patient could ask questions if they did not understand something
Understands me better	The PROMs helped the healthcare provider to better understand the patient

DISCUSSION

In this preregistered mixed-methods study, we aimed to describe the implementation of PROMs on mental and medical health outcomes in routine healthcare for children and adolescents with IBD in a large academic children's hospital, as well as assess the feasibility of implementing PROMs.

Although our compliance rate of 44.7% did not meet our predefined cut-off of ≥50%, we conclude that the implementation of PROMs was feasible, but that improvements should be made. Several explanations exist for the lower compliance rate. First, it can in part be explained by the fact that the PROMs were distributed to all patients, including those who were unable to login to the patient portal because they did not understand Dutch or did not have access to the Dutch digital

identifier (*DigiD;* f.e., expats). Second, results are comparable to other applications of PROMs in children and adolescents showing compliance rates around 50%,[28,29] although rates of 85% have also been reported.[30]

Regarding user experience, a score of 3 of higher, corresponding to at least a neutral rating, was set as cut-off for determining feasibility. Results showed that user experience regarding the patient portal and the discussion of PROMs by the healthcare provider was above the cut-off and can be seen as feasible. Regarding the PROMs themselves, the ratings were slightly below the predetermined cut-off. Negative feedback on the PROMs primarily related to the questionnaires being too long or being sent out too often. As a result, we have adjusted the time interval between PROMs from every three months to every six months.

A topic that was not studied directly, but is of importance to our results is mental health-related stigma. Many adolescents with mental healthcare needs do not access mental health care.[31] The implementation of PROMs can help facilitate discussions around mental health.[32,33] Specifically for adolescents with IBD, it is important that they trust their healthcare provider and that the healthcare provider inquires about their mental health.[33] As our results show that up to 40% of questionnaires were not discussed by the healthcare provider, it is likely that signals of mental health problems may be missed. On the other hand, if mental health scores were indicated as orange or red, the healthcare providers made an effort to always discuss these with their patients.

The current study has some strengths and limitations. Strengths of this study include the broad sample selection, with a range of ages and diagnoses. Further, this study incorporated the voice of the patient group. This can increase the adherence to the PROMs.[14] The experiences and lessons learned from our work can be used by other clinicians who may want to implement PROMs into routine care.

Limitations of this study relate primarily to the sample. Only patients that completed the PROMs on two occasions were invited for the user experience questionnaire. This could have resulted in omitting patients that are not compliant and have different views on the PROMs. However, as we gathered both positive and negative feedback on the implementation of the PROMs, we believe the data are an accurate representation of the preferences of the patients. Further, a sensitivity analysis, included in the supplemental material, showed that patients who completed the user experience survey were comparable to those that did not in terms of demographics (with the exception of disease duration) and PROMIS scores. A second limitation is that we were unable to extract data on ethnicity or

socioeconomic status, as these are not stored within the electronic health records. We could therefore not use this data in the current paper.

Future studies should examine the acceptability and feasibility of using PROMs in the children's hospital in general. As the use of PROMs will likely increase in the near future,[34] it is important to study whether all patient groups have the same needs, or whether patients with different diagnoses report different needs and preferences. It is also important to incorporate the patient's voice into the PROMs. Many studies on PROMs have included experts,[35,36] but incorporating patients' needs and preferences could have a direct effect on the compliance.

Healthcare professionals who intend to use PROMs in clinical care should carefully consider which PROMs they select, as current results show that the frequency and length of the PROMs, and the content of the questions, can be perceived as burdensome. For our outpatient clinic this meant reducing the frequency of the questionnaires from every three months to every six months. This way we capture variability in patients' physical and mental health status, but reduce the burden placed on patients and parents. We recommend including patients and parents as early as possible and evaluating the PROMs not only by clinical outcomes, but also by patient satisfaction. After implementing, an effort should be made to provide feedback based on the PROMs during each hospital appointment, as there are indications that this may motivate patients' compliance[37] and self-management.[38]

REFERENCES

1. Lenti MV, Cococcia S, Ghorayeb J, Di Sabatino A, Selinger CP. Stigmatisation and Resilience in Inflammatory Bowel Disease. *Internal and Emergency Medicine*. Mar 2020;15(2):211-223. doi:10.1007/s11739-019-02268-0
2. Jansson S, Malham M, Carlsen K, et al. Psychiatric disorders in paediatric-onset immune-mediated inflammatory diseases: a nationwide Danish study. *Archives of Disease in Childhood*. 2023;108(12):999-1007.
3. Stapersma L, van den Brink G, Szigethy EM, Escher JC, Utens EMWJ. Systematic Review with Meta Analysis: Anxiety and Depression in Children and Adolescents with Inflammatory Bowel Disease. *Alimentary Pharmacology & Therapeutics*. 2018;48(5):496-506. doi:10.1111/apt.14865
4. Jansson S, Malham M, Wewer V, Rask CU. Psychiatric comorbidity in childhood onset immune-mediated diseases—A systematic review and meta-analysis. *Acta Paediatrica*. 2022;111(3):490-499. doi:https://doi.org/10.1111/apa.16246
5. Fairbrass KM, Lovatt J, Barberio B, Yuan Y, Gracie DJ, Ford AC. Bidirectional Brain–Gut Axis Effects Influence Mood and Prognosis in IBD: a Systematic Review and Meta-Analysis. *Gut*. 2021:1-8. doi:10.1136/gutjnl-2021-325985
6. Bisgaard TH, Allin KH, Keefer L, Ananthakrishnan AN, Jess T. Depression and anxiety in inflammatory bowel disease: epidemiology, mechanisms and treatment. *Nature reviews Gastroenterology & hepatology*. 2022;19(11):717-726. doi:10.1038/s41575-022-00634-6
7. Xiong Q, Tang F, Li Y, et al. Association of inflammatory bowel disease with suicidal ideation, suicide attempts, and suicide: A systematic review and meta-analysis. *Journal of psychosomatic research*. 2022;160:110983.
8. Katon WJ. Epidemiology and Treatment of Depression in Patients with Chronic Medical Illness. *Dialogues in Clinical Neuroscience* 2011;13(1):7. doi:10.31887/DCNS.2011.13.1/wkaton
9. Mackner LM, Whitaker BN, Maddux MH, et al. Depression Screening in Pediatric Inflammatory Bowel Disease Clinics: Recommendations and a Toolkit for Implementation. *Journal of Pediatric Gastroenterology and Nutrition*. Jan 2020;70(1):42-47. doi:10.1097/MPG.0000000000002499
10. NVK. Inflammatoire darmziekten (IBD) bij kinderen. Federatie Medisch Specialisten. Accessed 01 February, 2023. https://richtlijnendatabase.nl/richtlijn/inflammatoire_darmziekten_ibd_bij_kinderen_en_adolescenten/startpagina_-_inflammatoire_darmziekten_ibd.html
11. de Jong MJ, Huibregtse R, Masclee AAM, Jonkers D, Pierik MJ. Patient-Reported Outcome Measures for Use in Clinical Trials and Clinical Practice in Inflammatory Bowel Diseases: A Systematic Review. *Clin Gastroenterol Hepatol*. May 2018;16(5):648-663 e3.
12. Field J, Holmes MM, Newell D. PROMs data: can it be used to make decisions for individual patients? A narrative review. *Patient Related Outcome Measures*. 2019/07/29 2019;10(null):233-241. doi:10.2147/prom.S156291
13. van Deen WK, Spiro A, Burak Ozbay A, et al. The Impact of Value-Based Healthcare for Inflammatory Bowel Diseases on Healthcare Utilization: a Pilot Study. *European Journal of Gastroenterology & Hepatology*. Mar 2017;29(3):331-337. doi:10.1097/MEG.0000000000000782
14. van Deen WK, Nguyen D, Duran NE, Kane E, van Oijen MG, Hommes DW. Value Redefined for Inflammatory Bowel Disease Patients: a Choice-Based Conjoint Analysis of Patients' Preferences. *Quality of Life Research*. Feb 2017;26(2):455-465. doi:10.1007/s11136-016-1398-z
15. Brenner EJ, Long MD, Mann CM, et al. Responsiveness of the Patient-reported Outcomes Measurement Information System (PROMIS) Pediatric Measures to Changes in Disease Status and Quality of Life Among Children and Adolescents With Crohn's Disease. *Inflammatory Bowel Diseases*. Feb 16 2021;27(3):344-351. doi:10.1093/ibd/izaa083
16. Brenner EJ, Long MD, Mann CM, et al. Validity and Responsiveness of the Patient-reported Outcomes Measurement Information System in Children With Ulcerative Colitis. *Journal of Pediatric Gastroenterology and Nutrition*. Jul 1 2021;73(1):67-72. doi:10.1097/MPG.0000000000003101
17. Eldridge SM, Lancaster GA, Campbell MJ, et al. Defining Feasibility and Pilot Studies in Preparation for Randomised Controlled Trials: Development of a Conceptual Framework. *PLOS ONE*. 2016;11(3):e0150205.

18. World Medical Association. World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects. *Jama*. 2013;310(20):2191-2194. doi:10.1001/jama.2013.281053
19. Brahmania M, Bernstein CN. Physician global assessments or blood tests do not predict mucosal healing in ulcerative colitis. *Canadian Journal of Gastroenterology & Hepatology*. Jun 2014;28(6):325-9. doi:10.1155/2014/737349
20. Forrest CB, Bevans KB, Pratiwadi R, et al. Development of the PROMIS ® pediatric global health (PGH-7) measure. *Quality of Life Research*. May 2014;23(4):1221-31. doi:10.1007/s11136-013-0581-8
21. Otley AR, Xu S, Yan S, et al. IMPACT-III Is a Valid, Reliable and Responsive Measure of Health-related Quality of Life in Pediatric Crohn's Disease. *Journal of Pediatric Gastroenterology and Nutrition*. 2006;43:S49. doi:10.1097/01.mpg.0000256260.38116.4a
22. Loonen HJ, Grootenhuis MA, Last BF, de Haan RJ, Bouquet J, Derkx BH. Measuring quality of life in children with inflammatory bowel disease: the impact-II (NL). *Quality of Life Research*. Feb 2002;11(1):47-56. doi:10.1023/a:1014455702807
23. Klaufus L, Luijten M, Verlinden E, et al. Validation of the PROMIS (R) pediatric item banks Anxiety and Depressive Symptoms in a general Dutch population. *Quality of Life Research*. 2020;29(SUPPL 1):S70-S70.
24. Freitag GF, Salem H, Conroy K, et al. The Patient□Reported Outcomes Measurement Information System (PROMIS) pediatric and parent-proxy short forms for anxiety: Psychometric properties in the Kids FACE FEARS sample. *Journal of Anxiety Disorders*. 2023;94:102677. doi:10.1016/j.janxdis.2023.102677
25. Lai J-S, Butt Z, Zelko F, et al. Development of a parent-report cognitive function item bank using item response theory and exploration of its clinical utility in computerized adaptive testing. *Journal of Pediatric Psychology*. 2011;36(7):766-779.
26. DeWalt DA, Thissen D, Stucky BD, et al. PROMIS Pediatric Peer Relationships Scale: development of a peer relationships item bank as part of social health measurement. *Health Psychology*. 2013;32(10):1093.
27. Elo S, Kyngäs H. The Qualitative Content Analysis Process. *Journal of Advanced Nursing*. 2008;62(1):107-115. doi:10.1111/j.1365-2648.2007.04569.x
28. El Miedany Y, El Gaafary M, Lotfy H, et al. Facilitating patient-centered care: the development of illustrated multidimensional patient-reported outcome measures for children/adolescents with juvenile idiopathic arthritis. *Clinical Rheumatology*. 2019/08/01 2019;38(8):2219-2226. doi:10.1007/s10067-019-04490-9
29. Mellor X, Buczek MJ, Adams AJ, Lawrence JTR, Ganley TJ, Shah AS. Collection of Common Knee Patient-reported Outcome Instruments by Automated Mobile Phone Text Messaging in Pediatric Sports Medicine. *J Pediatr Orthop*. Feb 2020;40(2):e91-e95. doi:10.1097/BPO.0000000000001403
30. Schepers SA, Sint Nicolaas SM, Haverman L, et al. Real□world implementation of electronic patient□reported outcomes in outpatient pediatric cancer care. *Psycho□oncology*. 2017;26(7):951-959. doi:10.1002/pon.4242
31. Gulliver A, Griffiths KM, Christensen H. Perceived barriers and facilitators to mental health help-seeking in young people: a systematic review. *BMC Psychiatry*. Dec 30 2010;10:113.
32. Bele S, Chugh A, Mohamed B, Teela L, Haverman L, Santana MJ. Patient-Reported Outcome Measures in Routine Pediatric Clinical Care: A Systematic Review. Systematic Review. *Frontiers in Pediatrics*. 2020-July-28 2020;8doi:10.3389/fped.2020.00364
33. Berenblum Tobi C, Vu M, Díaz-González de Ferris ME, Semerzier S, Kappelman MD, deJong NA. Adolescent and caregiver perceptions of addressing mental health in inflammatory bowel disease. *Journal of Child Health Care*. 2024:13674935241241350. doi:10.1177/13674935241241350
34. Baumhauer JF, Bozic KJ. Value-based Healthcare: Patient-reported Outcomes in Clinical Decision Making. *Clinical Orthopaedics and Related Research®*. 2016/06/01 2016;474(6):1375-1378. doi:10.1007/s11999-016-4813-4

35. Kim AH, Roberts C, Feagan BG, et al. Developing a Standard Set of Patient-Centred Outcomes for Inflammatory Bowel Disease-an International, Cross-disciplinary Consensus. *Journal of Crohn's and Colitis*. Mar 28 2018;12(4):408-418. doi:10.1093/ecco-jcc/jjx161
36. Steinmann G, Delnoij D, van de Bovenkamp H, Groote R, Ahaus K. Expert consensus on moving towards a value-based healthcare system in the Netherlands: a Delphi study. *BMJ Open*. Apr 12 2021;11(4):e043367. doi:10.1136/bmjopen-2020-043367
37. Pasma A, van Lint C, Hollander MA-d, Bruinsma SM, Peters IA. Healthcare providers' use of dashboards with patient reported outcomes reinforces patients to fill out patient reported outcome measures. *Digital health*. 2024;10:20552076241293975. doi:10.1177/20552076241293975
38. de Wit M, Versloot J, Zenlea I, Goethals ER. Using Person-Reported Outcomes (PROs) to Motivate Young People with Diabetes. *Current Diabetes Reports*. 2020/05/16 2020;20(7):23. doi:10.1007/s11892-020-01305-z

SUPPLEMENTAL MATERIAL

Questionnaire on patients' experiences with value-based healthcare (translated from Dutch)

You have completed questionnaires about your mental and physical well-being twice now. We are interested to hear from you how you experienced this. Your answers help us to improve healthcare for children and adolescents with an inflammatory bowel disease (Crohn's disease or ulcerative colitis). There are no right or wrong answers. Try to be as elaborate as possible.

Experiences related to completing the questionnaires

1. What was it like for you to complete the questionnaires?
2. How long did it take to complete the questionnaire?
3. Please rate whether you agree with the following statement: I had to complete questionnaires too often.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

4. Please rate whether you agree with the following statement: I did not have to complete questionnaires frequently enough.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

5. Please rate whether you agree with the following statement: The questionnaires were too short.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

6. Please rate whether you agree with the following statement: The questionnaires were too long.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

7. Please rate whether you agree with the following statement: I was able to understand the questionnaires well.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

8. Please rate whether you agree with the following statement: It was easy to complete the questionnaires online.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

9. How would you prefer to complete the questionnaires?

0 Online (like you are doing now) 0 In the hospital waiting area 0 On paper 0 Other:...

10. What did you like about the questionnaires?
11. What did you dislike about the questionnaires and what can we improve?
12. How satisfied are you with the questionnaires?

0 Very unsatisfied 0 Unsatisfied 0 Neutral 0 Satisfied 0 Very satisfied

Experiences related to discussing the questionnaires during the outpatient clinic appointment

13. Please rate whether you agree with the following statement: My doctor or nurse specialist was able help me better because of the questionnaires.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

14. Please rate whether you agree with the following statement: My doctor or nurse specialist understands me better because of the questionnaires.

0 Completely disagree 0 Disagree 0 Neutral 0 Agree 0 Completely agree

15. Were the questionnaires discussed with you during your appointment in the hospital?

0 Yes 0 No

If yes: continue with question 16
If no: continue with question 19

16. What did you like about the way the questionnaires were discussed with you?
17. How can we improve the way the questionnaires are discussed?

18. How satisfied are you with the way the questionnaires were discussed with you?

0 Very unsatisfied 0 Unsatisfied 0 Neutral 0 Satisfied 0 Very satisfied

Experiences related to the patient dashboard

19. Did you check your answers to the questionnaires on the dashboard in your online patient record?

0 Yes 0 No

If yes: continue with question 20
If no: continue with question 25

20. How well-organized do you think the dashboard was?

0 Not at all 0 Not really 0 Neutral 0 Somewhat 0 Very much

21. How informative did you think the dashboard was?

0 Not at all 0 Not really 0 Neutral 0 Somewhat 0 Very much

22. What did you like about the dashboard?
23. How can we improve the dashboard?
24. How satisfied are you with the dashboard?

0 Very unsatisfied 0 Unsatisfied 0 Neutral 0 Satisfied 0 Very satisfied

25. What is the reason you have not looked at the dashboard?
26. Is there anything you wish to add to your feedback?

Description of PROM outcomes and referrals to psychological care

This section describes the outcomes of the PROMs, both medical and psychological. The outcomes were studied exploratory and any results should be interpreted as such.

Methods

Participants

PROMs were distributed to all patients aged 8 to 18 and their parents. Patients had to be diagnosed with Inflammatory Bowel Disease (IBD) and have a scheduled appointment at the outpatient clinic of the Department of Paediatric Gastroenterology of the Erasmus MC Sophia Children's Hospital. No preselection was made based on IQ or language barriers. Recruitment took place from November 2022 until the end of July 2023.

Methods

As part of routine care, patients and their parents received the PROMs seven days prior to their scheduled appointment at the outpatient clinic, with a time interval of three months. This was done through an automated process, linked to the electronic patient records. Patients or parents received an email notification that there were questionnaires for them to complete. They could then log into the electronic patient portal to complete the questionnaires in a secured environment.

In case questionnaires were not completed the day before the visit, patients were contacted through a safe messaging application (*BeterDichtbij*), also used for easy accessible contact between patients/parents and the healthcare providers. If the questionnaires were not completed before the visit, patients and parents were given the option to complete them on a tablet while waiting for their appointment. They could also receive help with logging into the patient portal.

Patients aged 8 and older completed all PROMIS measures, but not the IMPACT-III. Patients aged 9 and older completed all the PROMIS measures and the IMPACT-III. Consent to use the data for research purposes was provided digitally, as an additional question to the PROMs.

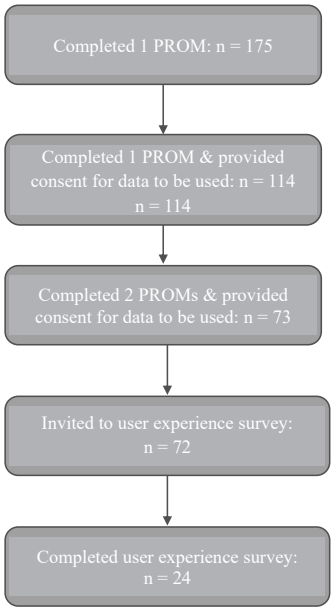
Analysis

Answers on the PROMIS questionnaires were converted to T-scores. Consequently, means and standard deviations were calculated for all questionnaires. To explore whether mean scores deviated from norm scores, one sample t-tests

were conducted. As comparison, Dutch norm scores from the literature were used. When these were unavailable, scores were compared to general norm scores (standardised to T = 50 and SD = 10). To correct for multiple testing, an FDR correction as described by Benjamini and Hochberg [1] was applied.

RESULTS

Figure S3: Flowchart of patients' data



Screening

Overall, 90.7% of patients and parents provided consent for their data to be used in this study. A total of 114 patients completed PROMs on time point 1, of whom 73 completed the PROMs also on time point 2, resulting in 187 instances of completed questionnaires. The 24 patients that completed the user experience survey are included within the 73 patients that completed two time points. A flowchart is shown in Figure S1.

On average across the 187 questionnaires, participants were 14.8 years old (SD = 2.4) and 50.3% was male. In total, 91 questionnaires were completed by patients with Crohn's disease, 91 with ulcerative colitis and 5 with IBD-U. Most patients were either in remission (65.4%) or had mild disease activity (26%). The most

common treatment was biologics (56.8%), followed by 5-aminosalicylates (37.3%) and immunomodulators (30.3%). On average, patients were diagnosed 2.3 years prior to completing the PROMs. More information is shown in Table S1.

The mean scores of all PROMs are shown in Table S2. The distribution of patients scoring across the predetermined cut-off scores is shown in Table S3. Regarding physical well-being, about half of the patients reported few complaints when it comes to general health, bowel symptoms or systemic symptoms. However, when looking at pain interference and fatigue, up to 82% of patients' scores were classified as orange or red.

When comparing to Dutch and US norm scores, patients with IBD reported significantly lower general health, both at time point 1 ($t(100) = -4.2$, $p < .001$) and time point 2 ($t(70) = -3.4$, $p = .001$), more pain interference at time point 1 ($t(100) = 17$, $p < .001$) and time point 2 ($t(70) = 15.1$, $p < .001$) , more fatigue at time point 1 ($t(100) = 4.2$, $p < .001$) and time point 2 ($t(70) = 4.2$, $p = < .001$) , more anxiety at time point 1 ($t(107) = 3.5$, $p = .006$) and time point 2 ($t(67) = 3.1$, $p = .003$) and worse cognitive functioning at time point 1 ($t(96) = -4$, $p = .001$). There were no differences with regards to depressive symptoms and peer relationships.

As to mental well-being, outcomes indicated few problems related to cognitive functioning and peer relationships. However, around 20% of patients and parents report heightened scores on symptoms of anxiety and depression that are classified as orange or red in the dashboard.

Referrals to psychological care

Overall, 23 patients scored above the cut-off for "red" scores on symptoms of anxiety or depression. Some patients had heightened scores at multiple timepoints, resulting in 27 occasions where patients report heightened scores. On four occasions, both the child and proxy-report indicated "red" scores, on six occasions only the child identified "red" scores and on 17 patients only the proxy-report identified "red" scores.

Of the 27 occasions with scores classified as red, we were unable to determine the follow-up of seven reports. Of the remaining 20, eight patients were already in contact with a psychologist, two patients were on a waiting list for psychological care and five patients were not open to contact with a psychologist. On three occasions, the psychological well-being was discussed, but no follow-up action was taken. One patient was referred to their general practitioner to ask for a referral to psychological care. In one case, the scores were not discussed.

DISCUSSION

Regarding outcomes on the PROMs, around 20% of patients scored above the cut-off for anxiety and/or depression. This is comparable to an earlier study in our outpatient clinic,[2] but higher than a meta-analysis reporting 16% or 15% of adolescents experiencing symptoms of anxiety or depression.[3] Taken together, it appears that approximately one in five children with IBD experience symptoms of anxiety and depression, underscoring the need of using PROMs to routinely monitor patients in an outpatient clinical setting. When compared to norm scores, patients with IBD score worse on general health, pain interference, fatigue, anxiety and cognitive functioning. Although exploratory, these analyses indicate that the physical and mental well-being of these patients is worse than children from the general population.

It is noteworthy that the vast majority of our sample was in remission or had mild disease activity. This could relate to the low number of patients scoring above the cut-off for a red score on symptoms of anxiety and depression. On the other hand, more than half of our sample was being treated with biologics. As this medication requires hospital visits and administration time, it is likely that this treatment has an influence on patients' lives and may also influence their scores of anxiety and depression.

Nonresponder analysis of feasibility results

To assess whether the participants that completed the user experience survey differ from nonparticipants, a non-responder analysis was conducted. Out of the 114 patients that completed at least one questionnaire (see Table S1), 24 also completed the user experience survey. These patients are compared to the 89 patients that did not complete the user experience survey. One person did not have sufficient data available. Scores were compared using two-samples t-tests, in case of continuous measures, and chi-squared tests in case of categorical measures.

Results are shown in Table S4. Overall, patients that completed the user experience survey (responders) were comparable to patients that did not complete the user experience survey in terms of demographics and PROMIS scores. The only exception was disease duration, with non-responders having a slightly longer time since diagnosis (2.7years) than responders (1.5 years; $t(47.3) = 2.5$, $p = .017$).

Table S1: Demographic characteristics of patients across time points

	Time point 1 (n = 114)	Time point 2 (n = 73)	Total (n = 187)[1]
Gender, n (%)			
Male	60 (52.6)	34 (46.6)	94 (50.3)
Female	54 (47.4)	39 (53.4)	93 (49.7)
Age range	8 - 18.9	8 - 18	8 - 18.9
Age, mean (SD)	14.7 (2.5)	15.1 (2.2)	14.8 (2.4)
Diagnosis, n (%)			
Crohn's disease	57 (50)	34 (46.6)	91 (48.7)
Ulcerative colitis	54 (47.4)	37 (50.7)	91 (48.7)
IBD-unclassified	3 (2.6)	2 (2.7)	5 (2.7)
Current therapy, n (%)			
5-aminosalicylates	40 (35.7)	29 (39.7)	69 (37.3)
Immunomodulators	34 (30.4)	22 (30.1)	56 (30.3)
Biologics	62 (55.4)	43 (58.9)	105 (56.8)
Corticosteroids	9 (8.0)	8 (11)	17 (9.2)
Nutritional treatment	1 (0.9)	0 (0)	1 (0.5)
None	7 (6.3)	1 (1.4)	8 (4.3)
Disease status (PGA)[2]			
In remission	72 (63.7)	49 (68.1)	121 (65.4)
Mild	30 (26.6)	18 (25)	48 (26)
Moderate	10 (8.9)	5 (6.9)	15 (8.1)
Severe	1 (0.9)	0 (0)	1 (0.5)
Age at onset, mean (SD)	12.14 (3.3)	12.28 (3.3)	12.19 (3.3)
Disease duration in years, mean (SD)	2.45 (2.4)	2.65 (2.3)	2.53 (2.3)

1. Combination of both time points. Some patients completed questionnaires on both time points, resulting in 114 unique patients with 187 questionnaires.

2. Physician Global Assessment: A physician rated disease activity as remission, mild, moderate or severe.

Table S2. Scores on Patient-Reported Outcome Measures (PROMs) across both time points

	Time point 1				Time point 2				Dutch norm scores
	Patient		Proxy		Patient		Proxy		Child
	N[b]	Mean (SD)	N[b]	Mean (SD)	N[b]	Mean (SD)	N[b]	Mean (SD)	Mean (SD)
General health[a]	101	44.7 (8.8)*	51	39.2 (7.8)	71	45.1 (8)*	33	41.3 (8.9)	48.3 (9.8)[4]
Pain interference[a]	101	58.8 (5.2)**	51	63.7 (5.5)	71	58.6 (4.8)**	33	65.3 (5.2)	n/a
Fatigue[a]	101	53.0 (7.3)**	51	55.1 (8.3)	71	53.3 (6.8)**	33	57.8 (6.6)	n/a
Bowel symptoms	106	27.5 (4.7)	-	-	68	27.8 (5.2)	-	-	n/a
Systemic symptoms	106	10.9 (2.7)	-	-	68	10.8 (2.7)	-	-	n/a
Anxiety[a]	108	47.5 (10.4)*	49	47.5 (10.2)	68	47.9 (10.3)*	35	48.1 (10.1)	44 (10.5)[5]
Depressive symptoms[a]	108	45.7 (10.1)	49	48 (10.8)	68	46.4 (9.8)	35	47.2 (9.8)	45 (11.2)[5]
Cognitive functioning[a]	97	48.3 (8.7)**	50	46.4 (10.2)	67	49.6 (8.6)	31	46.8 (8.3)	n/a
Peer relationships[a]	97	47.3 (6.7)	50	49.4 (7.8)	67	48.3 (6.9)	31	50.9 (8.2)	n/a

a. T-score is reported. These are standardised to reflect a mean of T = 50 and an SD of T = 10. Higher scores reflect more of the measured construct.

b. Number of questionnaires.

* *Significantly different from Dutch norm scores after FDR correction. ** Significantly different from US norm scores (T = 50) after FDR correction.*

Table S3: Number of questionnaires with scores across predetermined cut-off values, reported as N (%)

	Patients			Proxy		
	Green	Orange	Red	Green	Orange	Red
General health	132 (70.6)	50 (26.7)	3 (1.6)	44 (46.8)	40 (42.6)	10 (10.6)
Pain interference	98 (52.4)	83 (44.4)	6 (3.2)	49 (52.1)	43 (45.7)	2 (2.1)
Fatigue	32 (17.1)	140 (74.9)	15 (8.0)	13 (13.8)	72 (76.6)	9 (9.6)
Bowel symptoms	175 (91.2)	16 (8.3)	1 (0.5)	-	-	-
Systemic symptoms	160 (83.3)	24 (12.5)	8 (4.2)	-	-	-
Anxiety	146 (74.9)	42 (21.5)	7 (3.6)	73 (76.8)	16 (16.8)	6 (6.3)
Depressive symptoms	159 (81.5)	31 (15.9)	5 (2.6)	70 (73.7)	19 (20)	6 (6.3)
Cognitive functioning	158 (87.8)	22 (12.2)	0 (0)	84 (91.3)	8 (8.7)	0 (0)
Peer relationships	159 (88.3)	17 (9.4)	4 (2.2)	71 (77.2)	17 (18.5)	4 (4.4)

Note. Scores reflect normal (green), slightly elevated (orange, +1 SD) and highly elevated (red, +2 SD) scores.

Table S4: Nonresponder analysis

	Responders (n = 24)		Non responders (n = 89)		p-value[1]
	N	Mean (SD)/%	N	Mean (SD)/%	
Gender					.163
Male	9	37.5%	50	56.2%	
Female	15	62.5%	39	43.8%	
Age	24	14.5 (2.3)	89	14.7 (2.5)	.754
Diagnosis					.545
Crohn's disease	14	58.3%	43	48.3%	
Ulcerative colitis	9	37.5%	44	49.4%	
IBD-unclassified	1	4.2%	2	2.3%	
Current therapy[2]					---
5-aminosalicylates	7	30.4	32	36.4	
Immunomodulators	9	39.1	25	28.4	
Biologics	10	43.5	52	59.1	
Corticosteroids	2	8.7	7	8	
Nutritional treatment	0	0	1	1.1	
None	2	8.7	5	5.7	
Disease status (PGA)[2]					.778
In remission	15	65.2%	56	62.9%	
Mild	5	21.7%	25	28.1%	
Moderate	3	13%	7	7.9%	
Severe	0	0%	1	1.1%	
Age at onset	24	13 (2.8)	89	11.9 (3.5)	.125
Disease duration in years	24	1.5 (1.8)	89	2.7 (2.4)	.017
General health	22	43.1 (9.3)	73	45.4 (8.8)	.318
Pain interference	22	57.7 (6.8)	73	59.5 (4.2)	.273
Fatigue	22	52.8 (7.5)	73	53.3 (7.2)	.764
Bowel symptoms	22	27.1 (5.7)	80	27.6 (4.2)	.682
Systemic symptoms	22	10.7 (2.9)	80	10.9 (2.6)	.765
Anxiety	22	50.5 (9.1)	82	46.8 (10.6)	.121
Depressive symptoms	22	48 (10.4)	82	45 (10.1)	.223
Cognitive functioning	21	49.1 (6.8)	70	47.3 (6.6)	.295
Peer relationships	21	48.6 (8.3)	70	48 (8.9)	.783

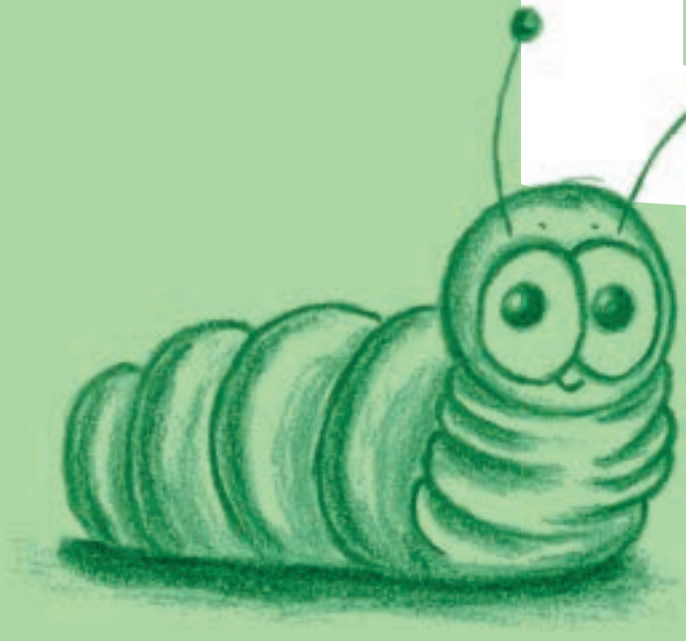
1. Two-samples t-test for continuous variables and chi-squared tests for categorical variables

2. A chi-square test was not conducted, as these categories are not mutually exclusive and thus violate basic test assumptions

REFERENCES

1. Benjamini Y, Hochberg Y. Controlling the False Discovery Rate: A Practical and Powerful Approach to Multiple Testing. *Journal of the Royal Statistical Society: Series B (Methodological)*. 1995;57(1):289-300. doi:10.1111/j.2517-6161.1995.tb02031.x
2. van den Brink G, Stapersma L, Vlug LE, et al. Clinical Disease Activity is Associated with Anxiety and Depressive Symptoms in Adolescents and Young Adults with Inflammatory Bowel Disease. *Alimentary Pharmacology & Therapeutics*. 2018;48(3):358-369. doi:10.1111/apt.14832
3. Stapersma L, van den Brink G, Szigethy EM, Escher JC, Utens EMWJ. Systematic Review with Meta Analysis: Anxiety and Depression in Children and Adolescents with Inflammatory Bowel Disease. *Alimentary Pharmacology & Therapeutics*. 2018;48(5):496-506. doi:10.1111/apt.14865
4. Luijten MAJ, Haverman L, van Litsenburg RRL, Roorda LD, Grootenhuis MA, Terwee CB. Advances in measuring pediatric overall health: the PROMIS® Pediatric Global Health scale (PGH-7). *European Journal of Pediatrics*. 2022:1-9. doi:10.1007/s00431-022-04408-9
5. Klaufus LH, Luijten MAJ, Verlinden E, et al. Psychometric properties of the Dutch-Flemish PROMIS® pediatric item banks Anxiety and Depressive Symptoms in a general population. *Quality of Life Research*. 2021;30(9):2683-2695. doi:10.1007/s11136-021-02852-y

B



Part B

The Quest of Making Success of Transition Measurable


5

Chapter 5

Self-efficacy did not Predict the Outcome of the Transition to Adult Care in Adolescents with Inflammatory Bowel Disease

G van den Brink, MAC van Gaalen, M Zijlstra, L de Ridder,
CJ van der Woude, JC Escher

Acta Paediatrica 2019;108:333–8.

ABSTRACT

Aim

It can be difficult for adolescents with inflammatory bowel disease (IBD) to make the transition from paediatric to adult care. We studied the outcomes of this process and defined what constituted a successful transition.

Methods

In 2008, 50 adolescents who attended our IBD transition clinic completed IBD-yourself, a self-efficacy questionnaire that we had previously developed and validated. We approached the subjects in 2014, two to six years after they transferred to adult care, and 35 agreed to take part in the current study. The outcome of transition was assessed by our newly developed Transition Yourself Score. In addition, the relationship between self-efficacy and the outcome of the transition was measured.

Results

The mean age of the patients was 21.8 years and 69% suffered from Crohn's disease. The transition process was successful in 63% of cases, moderately successful in 31% and failed in 6%. A successful transition was associated with effective use of medication and clinical remission at the time of transfer, but could not be predicted by self-efficacy. The Transition Yourself Score will be validated in future studies.

Conclusion

Nearly two-thirds (63%) of the adolescents who attended the IBD transition clinic had a successful transition to adult care.

INTRODUCTION

Inflammatory bowel disease (IBD) is a chronic relapsing inflammatory disorder of the intestine and manifests in adolescence in about 25% of cases.[1, 2] As IBD is a lifelong disease, all paediatric patients will need to undergo the transfer to adult care. It is advisable to have a transition period to prepare patients and parents for the transfer, which refers to the actual handover of the patient to adult healthcare.[3] A failed transition can adversely affect IBD related outcomes. It can increase non-adherence, non-attendance, hospitalisation rates and the need for surgery. [4, 5] During the transition process, patients, their parents and the paediatric and adult gastroenterologist have specific tasks. Patients should acquire disease knowledge, autonomy and self-management [6, 7], while parents need to stimulate their child's independence and physicians should be knowledgeable about adolescents' developmental and health issues and prepare them for transfer.[3] Transition programmes are designed to facilitate the transition process and increase knowledge and self-management.[3, 8] Self-efficacy, a person's belief in their capability to organise and execute the actions required to deal with prospective situations, is thought to be a prerequisite for self-management.[9, 10] It reflects self-care responsibility and has been shown to be a predictor of readiness to transfer.[11] We previously developed and validated IBD-yourself, a specific IBD knowledge and self-efficacy questionnaire.[12]

The optimal model for IBD transitional care is currently unknown, as is the definition of a successful transition, due to the scarcity of outcome research. We aimed to develop a tool, the Transition Yourself Score, to measure the success of transition. The Transition Yourself Score was applied to our IBD transition cohort to assess transition outcomes one year after transfer. In addition, we assessed the predictive value of self-efficacy for successful transition.

METHODS

Participants

In 2008, 50 patients from our IBD transition clinic participated in the validation study of the self-efficacy questionnaire, IBD-yourself.[12] Of these, 35 gave informed consent to participate in the current study in 2014. Data on patient characteristics, disease type, treatment and outpatient clinic visits after transfer were retrieved from their medical records. The study was approved by the Medical Ethics Committee of the Erasmus Medical Centre, Rotterdam, The Netherlands.

IBD transition clinic

Adolescents aged 16 to 18 years old with IBD are seen at the outpatient clinic, which is located in the adult gastroenterology department. A multidisciplinary team, consisting of a paediatric gastroenterologist, a paediatric IBD nurse specialist, an adult gastroenterologist and a family therapist, discuss all patients before the start of the clinic. Patients visit the clinic at least four times a year, where they are seen by either the paediatric gastroenterologist or the paediatric IBD nurse specialist. Once a year the adolescent patients also meet the adult gastroenterologist, to get acquainted with the adult healthcare system. During clinic visits, both the nurse and doctors check for disease knowledge, self-efficacy and self-management skills. Around the age of 18, patients are transferred, either within our centre or to a hospital closer to home. The choice of the adult care team depends primarily on the complexity of the disease and secondarily on the patient's preference and where they live. As part of the routine care for all patients, irrespective of their disease severity, an appointment at the gastroenterology department is made within three to six months after transfer. At this time an extensive transfer letter, including the patient's medical history, is sent to the adult medical healthcare provider, with a copy to the patient.

Measurements

Transition Yourself Score

A score reflecting the outcome of the transition process was developed based on previous literature [13] and the outcome of a focus group meeting with IBD experts. The Transition Yourself Score was applied to participants one year after their transfer to adult care. The following items were rated: adherence to visits at the gastroenterology outpatient clinic, adherence to medication and qualitative evaluation of transition by the patient. A pre-test of the score with a representative group of 10 patients resulted in minor adjustments in the language.

We scored the adherence to the visits to the adult gastroenterologist by determining when the patients first appeared in the gastroenterology outpatient clinic. The percentage of missed scheduled outpatient visits within the first year was calculated by dividing the number of missed visits by the number of planned visits times 100. Rescheduled visits were not counted as non-attendance. Adherence to medication was determined by an adherence interview (Table S1).[14] The interview contained 10 questions regarding medication use, missed medication and the experienced burden of the medication regimen. Patients could receive one point per question, with a maximum score of 10. Prescriptions for supplements, such as calcium or iron, were not included in the assessment of medication adherence. Low

adherence was indicated by a score below six, medium by a score of six to eight and high by a score of eight or more. At enrolment, patients retrospectively graded the transition process on a 10-point scale, where one was very poor and 10 was excellent and these scores are recorded in Table 1. A total score below five indicated a failed transition, five or six was moderately successful and above six was successful. For the analysis we combined failed and moderately successful transition into the unsuccessful transition category, to reflect that improvements were needed in this group. We then compared those findings with the group of subjects who had a successful transition.

IBD-yourself

IBD-yourself is questionnaire that was previously developed and validated by our research group to explore knowledge about IBD and self-efficacy.[12] It covers a number of domains, such as knowledge of IBD, diagnostic tests and medication

Table 1: Transition Yourself Score

Items	Score	Frequency (n=35), n (%)
Time to first outpatient visit to adult gastroenterologist		
Unknown or after 12 months	0	1 (2.9)
After 6-12 months	1	3 (8.6)
Within 3-6 months	2	31 (88.6)
Non-attendance rates at outpatient clinic < 12 months after transfer		
>25%	0	2 (5.7)
10-25%	1	3 (8.6)
None / <10%	2	30 (85.7)
Medication adherence		
Low adherence	0	4 (11.4)
Medium adherence	1	17 (48.6)
High adherence	2	5 (14.3)
No medication prescribed[A]		9 (25.7)
Quality of transition (grade 0-10) [B]		
< 5.5	0	0
5.5-7	1	11 (31.4)
>7	2	24 (68.6)
Total score:		
Failed transition	<4	2 (5.7)
Moderate successful transition	5 or 6	11 (31.4)
Successful transition	>6	22 (62.9)

[A] These patients were given two points in total transition score. [B] as experienced by the transferred patient

use. Higher scores indicated higher overall levels of self-efficacy. As the number of questions could differ between patients, we just provided scores for each domain and no total score (Table S2).[12]

Clinical outcome one year after transfer

Data regarding relapses, disease related complications, admissions, surgery and pregnancies were retrieved from medical charts. Clinical relapse was defined as a relapse with a change in treatment strategy and, or, hospital admission.

Statistical analysis

Statistical analyses were completed using SPSS 18.0 for Windows (SPSS Inc, Chicago, USA). Significance was set at $p<0.05$. Descriptive statistics were calculated as percentages for discrete data and medians with interquartile ranges (IQR) for continuous data. The chi-square test or Fisher's exact test was used to analyse categorical data. Correlation between the IBD-yourself and transition success was examined using Spearman's correlation coefficient and the Mann-Whitney *U* test.

RESULTS

Patients' characteristics

There were 35 adult patients who gave their informed consent, with a median age of 21.9 years and interquartile range (IQR) of 21.1-22.6. Of the 50 patients who took part in the 2008 study, two declined participation, one patient had died from a metastasised rectal adenocarcinoma due to Hermansky Pudlak syndrome and 12 patients had been lost to follow up. The clinical and demographic characteristics are shown in Table 2, in the column total. The baseline characteristics of the 35 participants and 15 non-participants did not differ (data not shown), except for educational status. Of the non-participants, 80% had low educational status compared to 46% of the participants.

Outcome of transition at one year after transfer

Almost 90% of the patients visited the adult outpatient clinic in the first three to six months after transfer. In addition, 85% of the patients missed less than 10% of their outpatient visits in the first year after transfer. Most patients had medium medication adherence. Almost 70% of patients valued the quality of their transition as good, scoring it above a seven. When we used the total Transition Yourself Score, the transition was successful in 22/35 (63%) patients, moderately successful in 11 (31%) and failed in two patients (6%) (Table 1). A female patient with Crohn's disease failed the transition as she did not adhere to her medication, did

Table 2: Patient characteristics

	Total (n=35)	**Unsuccessful transition¥ (n=13)**	**Successful transition (n=22)**	**p value§**
Age at diagnosis (years), (median, IQR)	13 (12-15)	14 (11.5-15.0)	13 (12.0-15.0)	0.875
Months in transition clinic before transfer (median, IQR)	13 (5-18)	10 (5.0-18.0)	13 (4.8-17.8)	0.811
Disease duration at transfer (median, IQR)	7 (7-10)	4.1 (3.0-6.9)	4.5 (2.9-6.1)	0.864
Years after transfer at inclusion in study				1.0
<2	2 (5.7%)	1 (7.7%)	1 (4.5%)	
2-4	20 (57.1%)	7 (53.8%)	13 (59.1%)	
4-6	13 (37.1%)	5 (38.5%)	8 (36.4%)	
Gender (male), no (%)	15 (42.9%)	3 (23.1%)	12 (54.5%)	0.070
Disease type, CD, no (%)	24 (68.6%)	9 (69.2%)	15 (68.2%)	1.0
Medication at transfer‡				0.840
Aminosalicylates	9 (25.7%)	3 (23.1%)	6 (27.2%)	
Immunomodulators	25 (71.4%)	9 (69.2%)	16 (72.7%)	
Anti-TNF	12 (34.2%)	5 (38.5%)	7 (31.8%)	
Prednisone	2 (5.7%)	1 (7.7%)	1 (4.5%)	
No medication	1 (2.9%)	0	1 (4.5%)	
Active disease during transfer	5 (14.3%)	4 (30.8%)	1 (4.5%)	0.052
Relapse within first year after transfer	10 (28.6%)	5 (38.5%)	5 (22.7%)	0.444
Educational level				0.467
Low	16 (45.7%)	7 (53.8 %)	9 (40.1%)	
Medium	13 (37.2%)	3 (23.1%)	10 (45.5%)	
High	6 (17.1%)	3 (23.1%)	3 (13.6%)	

‡ Some patients were prescribed more than one kind of medication ¥ moderately successful and failed transition § successful versus unsuccessful transition

Note: percentages are displayed as column percentages

not appear for her first appointment and presented more than one year after transfer in the emergency department with a clinical relapse. The other patient was a male patient with Crohn's disease who was transferred to a community hospital, but was lost to follow up after transfer.

Factors influencing outcome of transition

The clinical and demographic characteristics did not differ significantly between the group with successful and unsuccessful transition (Table 2). In the group with unsuccessful transition. 80% had active disease before their transfer to adult care and this approached significance (p=0.052). Female patients were more likely than males to have an unsuccessful transfer (p=0.069) (data not shown).

Relation between adolescent self-efficacy and successful transition

Spearman's correlation showed a significant correlation for the outcome of transition and the IBD-yourself domain that covered actual behaviour in medication use (r=0.397, p=0.025, n= 35). In parallel, adolescents with a successful transition had significantly higher scores in the domain of actual behaviour in medication use (Mann Whitney *U* test: U=60.0, z=-2.208, p=0.027). As shown in Table 3, the scores of each IBD-yourself domain were divided in three groups, namely low, medium and high. A non-significant trend was seen in seven of the domains, with more patients having a high score in the successful transition group.

Clinical outcome at one year after transfer

Of the 35 patients, 10 patients (29%) had a relapse with clinical consequences in the first year after transfer. In 50% of these patients, the outcome of their transition was scored as either moderately successful or unsuccessful. In contrast, the transition was unsuccessful in 30% of patients without relapsing disease. No significant differences were found between the patients with or without relapse with respect to the outcome of their transition, therapy adherence and missed outpatient clinic visits (data not shown). Of the 35 patients, two underwent surgery, namely resection of the remaining colon after hemi-colectomy and resection of perianal skintags, one Crohn's disease patient developed a perianal fistula and received antibiotics and two patients developed a new extra intestinal manifestation (arthralgia). One patient became pregnant during anti-tumour necrosis factor treatment and delivered a healthy baby. None of the patients developed a malignancy during the study period.

DISCUSSION

Our study showed that in our IBD transition clinic, transition was successful in 63% of patients and moderately successful in 31% of patients. This modest success ratio indicates that there is room for improvement in our IBD transition strategy, both in general and in individual patients.

To our knowledge, only one previous study has described the outcome of transition in IBD care.[15] Non-adherence rates were in accordance with our cohort, but higher rates of hospitalisation and disease complications were found after transfer. However, it is not clear at what point in the transition process this was assessed.

The outcome of transition has also been described in paediatric patients with other chronic diseases with success varying from 42-53% of patients.[16-19] All these

Table 3: IBD- yourself and outcome of transition

Domains IBD Yourself	Unsuccessful transition (failed & moderate) (n=13), n (%)	Successful transition (n=22), n (%)
Self-efficacy in knowledge of IBD (n=35)		
12-14 points	2 (15.4%)	1 (4.5%)
15-17 points	6 (46.2%)	12 (54.5%)
18-20 points	5 (38.5%)	9 (40.9%)
Self-efficacy in knowledge of diagnostic tests (n=35)		
14-17 points	2 (15.4%)	4 (18.2%)
18-21 points	7 (53.5%)	8 (36.4%)
22-24 points	4 (30.8%)	10 (45.5%)
Self-efficacy in knowledge of medication (n=27)		
17-22 points	4 (40.0%)	4 (23.5%)
23-27 points	4 (40.0%)	6 (35.3%)
28-32 points	2 (20.0%)	7 (41.2%)
Actual behaviour medication use (n=32)*		
7-10 points	1 (10.0%)	2 (9.1%)
11-14 points	7 (70.0%)	3 (13.6%)
15-17 points	2 (20.0%)	17 (77.3%)
Self-efficacy in skills for independent outpatient clinic visits (n=35)	2 (15.4%)	3 (13.6%)
12-20 points	5 (38.5%)	9 (40.9%)
21-28 points	6 (46.2%)	10 (45.5%)
29-36 points		
Actual behaviour outpatient clinic (n=26)		
4-6 points	10 (90.9%)	9 (60.0%)
7-8 points	1 (9.1%)	6 (40.0%)
Self-efficacy in coping with IBD (n=34)		
5-8 points	1(8.3%)	1 (4.5%)
9-12 points	4 (33.3%)	6 (27.3%)
13-16 points	7 (58.3%)	15 (68.2%)
Self-efficacy in knowledge of transition process (n=31)		
32-42 points	3 (25.0%)	2 (10.5%)
43-52 points	4 (33.3%)	8 (42.1%)
53-62 points	5 (41.7%)	9 (47.4%)
Self-efficacy in transfer readiness (n=34)		
4-6 points	10 (76.9%)	14 (66.7%)
7-8 points	3 (23.1%)	(33.3%)

$p<0.05$

studies used a restricted definition of success, namely attending the first one or two visits in adult care. In our clinic the transition process starts early, at the age of 16, with early involvement of the adult gastroenterologist. This could explain the relatively higher rates of successful transition in our cohort compared to the cohorts of patients with other chronic diseases.

A scoring system for the outcome of transition in IBD is currently not available and we decided to develop such an instrument, the Transition Yourself Score. In 2015, a ranking list with key elements and indicators of successful transition in general was published.[13] This list, together with previous literature, supports the elements of the Transition Yourself Score.[13, 19-21] A qualitative evaluation by the patient was included because patient experiences are of essential importance, reflect continuity of care and should be taken into account.[13]

We hypothesised that patient or disease related factors can influence the outcome of transition as well as the actual transition clinic. Our study indicated that female patients and patients with an active disease before transfer might be at risk for unsuccessful transition.

Unsuccessful transition may have serious consequences, such as non-adherence, hospitalisation or surgery. To our knowledge, there have not been any studies that have measured successful transition and correlated these to the clinical outcomes after transfer. Unfortunately, because of the limited size of our study, we were not able to do that either.

As discussed earlier, it has been suggested that self-efficacy is a prerequisite for self-management and transfer readiness.[10, 11] Whitfield et al.[22] investigated self-efficacy in adolescent IBD patients and found that mean self-efficacy scores were higher in older patients, but not in patients with longer disease duration. Remarkably, communication with the doctor did not improve with age and about 80% of patients above 18 years of age reported that they were not independent when it came to disease management tasks. Unfortunately, this study did not assess if patients actually used the skills they claimed to possess. It could be that self-efficacy does not always correlate with actual behaviour. Our study showed that actual behaviour could be associated with transition success. Since the publication of the IBD-yourself, another self-efficacy scale for adolescents and young adults has been developed and validated, but studies using this scale have not yet been published.[23]

This study had several limitations: the recruitment from a single centre, the retrospective nature and the small sample size limited the statistical analysis and generalisability. In addition, recall bias could have influenced the patients' recollections of the item quality of the transition process, since some patients participated several years after their transfer. Moreover, socioeconomic status was higher in participants than non-participants, which could have influenced the results of our study. Furthermore, the Transition Yourself Score needs to be vali-

dated and IBD-yourself needs to be compared to other measures of self-efficacy for further validation. Lastly, the generalisability may be limited because of country specific transition approaches, but the general elements chosen to classify the success of transition can be tailored to each local situation.

CONCLUSION

Despite these limitations, this study was the first to evaluate the outcome of transition in IBD by using a new score, the Transition Yourself Score. We showed that, after attending our IBD transition clinic, 63% of patients had a successful transition. The IBD-yourself, which measured self-efficacy, did not predict a successful transition, which can question the importance of self-efficacy. We stress the importance of not only assessing self-efficacy or self-management, but also the outcome of transition and clinical disease course after transfer. If predictors for a successful transition can be identified, then transition programmes can be optimised. The Transition Yourself Score is a simple tool to assess the efficacy of transition programmes, but it needs to be validated.

REFERENCES

1 Baumgart DC, Bernstein CN, Abbas Z, et al. IBD Around the world: comparing the epidemiology, diagnosis, and treatment: proceedings of the World Digestive Health Day 2010--Inflammatory Bowel Disease Task Force meeting. *Inflamm Bowel Dis*. 2011;17(2):639-644.

2 Griffiths AM. Specificities of inflammatory bowel disease in childhood. *Best Pract Res Clin Gastroenterol*. 2004;18(3):509-523.

3 Goodhand J, Hedin CR, Croft NM, Lindsay JO. Adolescents with IBD: the importance of structured transition care. *J Crohns Colitis*. 2011;5(6):509-519.

4 Bollegala N, Brill H, Marshall JK. Resource utilization during pediatric to adult transfer of care in IBD. *J Crohns Colitis*. 2013;7(2):e55-60.

5 Cole R, Ashok D, Razack A, Azaz A, Sebastian S. Evaluation of Outcomes in Adolescent Inflammatory Bowel Disease Patients Following Transfer From Pediatric to Adult Health Care Services: Case for Transition. *J Adolesc Health*. 2015;57(2):212-217.

6 Trivedi I, Keefer L. The Emerging Adult with Inflammatory Bowel Disease: Challenges and Recommendations for the Adult Gastroenterologist. *Gastroenterol Res Pract*. 2015;2015:260807.

7 Wright EK, Williams J, Andrews JM, et al. Perspectives of paediatric and adult gastroenterologists on transfer and transition care of adolescents with inflammatory bowel disease. *Intern Med J*. 2014;44(5):490-496.

8 Escher JC. Transition from pediatric to adult health care in inflammatory bowel disease. *Dig Dis*. 2009;27(3):382-386.

9 Bandura A. Self-efficacy. In: Ramachaudran ed. Encyclopedia of human behavior, Vol. Vol. 4. New York: Academic Press, 1994:71-81

10 Marks R, Allegrante JP, Lorig K. A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part II). *Health Promot Pract*. 2005;6(2):148-156.

11 van Staa A, van der Stege HA, Jedeloo S, Moll HA, Hilberink SR. Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors. *J Adolesc Health*. 2011;48(3):295-302.

12 Zijlstra M, De Bie C, Breij L, et al. Self-efficacy in adolescents with inflammatory bowel disease: a pilot study of the "IBD-yourself", a disease-specific questionnaire. *J Crohns Colitis*. 2013;7(9):e375-385.

13 Suris JC, Akre C. Key elements for, and indicators of, a successful transition: an international Delphi study. *J Adolesc Health*. 2015;56(6):612-618.

14 Mackner LM, Crandall WV. Oral medication adherence in pediatric inflammatory bowel disease. *Inflamm Bowel Dis*. 2005;11(11):1006-1012.

15 Bennett AL, Moore D, Bampton PA, Bryant RV, Andrews JM. Outcomes and patients' perspectives of transition from paediatric to adult care in inflammatory bowel disease. *World J Gastroenterol*. 2016;22(8):2611-2620.

16 Reid GJ, Irvine MJ, McCrindle BW, et al. Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics*. 2004;113:e197-205.

17 Gleeson H, Davis J, Jones J, O'Shea E, Clayton PE. The challenge of delivering endocrine care and successful transition to adult services in adolescents with congenital adrenal hyperplasia: experience in a single centre over 18 years. *Clin Endocrinol (Oxf)*. 2013;78(1):23-28.

18 Hazel E, Zhang X, Duffy CM, Campillo S. High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. *Pediatr Rheumatol Online J*. 2010;8:2.

19 Jensen PT, Karnes J, Jones K, et al. Quantitative evaluation of a pediatric rheumatology transition program. *Pediatr Rheumatol Online J*. 2015;13:17.

20 Fredericks EM, Dore-Stites D, Well A, et al. Assessment of transition readiness skills and adherence in pediatric liver transplant recipients. *Pediatr Transplant*. 2010;14(8):944-953.

21 Andemariam B, Owarish-Gross J, Grady J, Boruchov D, Thrall RS, Hagstrom JN. Identification of risk factors for an unsuccessful transition from pediatric to adult sickle cell disease care. *Pediatr Blood Cancer*. 2014;61(4):697-701.

22 Whitfield EP, Fredericks EM, Eder SJ, Shpeen BH, Adler J. Transition readiness in pediatric patients with inflammatory bowel disease: patient survey of self-management skills. *J Pediatr Gastroenterol Nutr.* 2015;60(1):36-41.
23 Izaguirre MR, Taft T, Keefer L. Validation of a Self-efficacy Scale for Adolescents and Young Adults With Inflammatory Bowel Disease. *J Pediatr Gastroenterol Nutr.* 2017;65(5):546-550.

SUPPLEMENTARY TABLES

Table S1: Questions adherence interview

1.	Did you forget to take your IBD medication (pills, tablets, enemas or suppositories) in the past two weeks or did you forget your last planned infusion/injection? (Yes=0, No=1)
2.	Did you skip your medication in the past two weeks because you think they do not help or even make you feel worse? (Yes=0, No=1)
3.	When you are not at home, do you bring along your IBD medication? (Yes=1, No=0)
4.	Sometimes medication is not taken on purpose (for other reasons than forgetting), did that happen to you past two weeks? (Yes=0, No=1)
5.	Did you take your medication(s) for IBD yesterday? And (if you get infusions or take injections): did you receive your most recent planned infusion/injection? (Yes=1, No=0)
6.	Do you sometimes skip your medication(s) when you feel good and have no symptoms of your disease? (Yes=0, No=1)
7.	Are you bothered by the fact that you have to take medication? (yes=0, no=1)
8.	Do you find it hard to remember to take or inject your medication or go to the hospital for your infusion? (Yes=0, No=1)
9.	Do you receive help from somebody else in remembering to take your medication or plan your infusions? (yes=0, no=1)
10.	When you run out of medication, do you actively arrange a new prescription (yes=1, no=0)

Table S2: Domains and scoring IBD-yourself (20)

	Domain	Number of questions	Score range
1	VAS on general independency	1	0-100
2	VAS on perceived disease burden	1	0-100
3	Self-efficacy in knowledge of IBD	5	0-20
4	Self-efficacy in knowledge of diagnostic tests	6	0-24
5	Self-efficacy in medication use	8	0-32
6	Actual behaviour regarding medication use	4	0-20
7	Self-efficacy in skills for independent outpatient clinic visits	9	0-36
8	Actual behaviour at the outpatient clinic	4	0-8
9	Self-efficacy in coping with IBD	4	0-16
10	Self-efficacy in knowledge of transition process	14	0-70
11	Self-efficacy in transfer readiness	2	0-8


6

Chapter 6

Health Care Transition Outcomes in Inflammatory Bowel Disease: A Multinational Delphi Study

G van den Brink G, MAC van Gaalen, L de Ridder, CJ van der Woude, JC Escher.

J Crohns Colitis 2019;13:1163–72.

ABSTRACT

Background

Transition programs are designed to prepare adolescent Inflammatory Bowel Disease (IBD) patients for transfer to adult care. It is still unclear which outcome parameters define 'successful transition'. Therefore, this study aimed to identify outcomes important for success of transition in IBD.

Methods

A multinational Delphi study in patients, IBD-nurses and paediatric and adult gastroenterologists was conducted. In Stage 1, panellists commented on an outcome list. In Stage 2, the refined list was graded from 1-9 (least-very important), by an expert and a patient panel. In stage 3, the expert panel ranked important outcomes from 1 to 10 (least-most important). Descriptive statistics and Mann-Whitney-U tests were performed.

Results

The final item list developed in Stage 1 was tested by the expert (n=74 participants,52.7% paediatrics) and patient panel (n=61, aged 16-25 y, 49.2% male). Respectively, 10 and 11 items were found to be important by the expert and patient panel. Both panels agreed on 8 of these items, of which 6 reflected self-management skills. In Stage 3, the expert panel formed a top-10 list. The three most important items were: decision making regarding IBD (mean score 6.7), independent communication (mean score 6.3) and patient satisfaction (mean score 5.8).

Conclusion

This is the first study identifying outcomes that IBD-health care providers and patients deem important factors for successful transition. Self-management skills were considered more important than IBD-specific items. This is a first step to further define success of transition in IBD and subsequently evaluate the efficacy of different transition models.

INTRODUCTION

In up to 25% percent of patients inflammatory bowel disease (IBD; Crohn's disease [CD] and ulcerative colitis [UC]) manifests during late childhood or adolescence.[1,2] As IBD is a lifelong disease all of these patients will need to undergo transfer of paediatric to adult care. To optimise this transfer and minimise adverse outcomes, it is advised to have a transition period where patients (and parents) are prepared for the actual transfer.[3,4] Transition is defined as the purposeful planned movement and preparation of adolescents and young adults with chronic medical conditions from child-centred to adult-oriented healthcare systems.[5] In the transition process the patient, parent, paediatric gastroenterologist, adult gastroenterologist and IBD-nurse have specific tasks.[3,6] Patients should acquire (disease) knowledge, autonomy and self-management.[7-11] Parents need to allow their adolescent child more independence. Physicians and nurses should support the transition process, be knowledgeable of adolescents' developmental and health issues and prepare adolescents for the changes that will be encountered in the adult health care system.[6,12,13] Transitional programs are designed to facilitate all these processes [12,14,15] and prepare the individual patient for his/her transfer by helping to increase knowledge as well as to reach a higher level of self-management.

As summarised in the UK guideline on transition in patients with chronic digestive diseases, inadequate transition arrangements have been associated with adverse outcomes across several medical conditions, such as diabetes [16], heart disease[17] and sickle cell disease.[18] In IBD, studies investigating the impact of structured transition are scarce. Studies showed that the lack of a structured transition service negatively impacted adherence [19,20] and attendance [19,20], and was associated with a higher hospitalisation and surgery rate.[19] On the other hand, structured transition programmes have been shown to result in better disease related outcomes[21,22], improved self- and disease knowledge and improved quality of life.[22,23]

Although many different models for transitional care have been proposed in IBD (e.g. [4,12,14,24]), there is no evidence that one particular model is more effective than others.[3] In addition, a clear definition on success of transition in IBD is lacking.[12,19,25] Two recent studies identified general, non-disease specific indicators for success of transition in adolescent medicine. Outcomes such as quality of life, continuity of care, self-management, therapy knowledge and adherence were recognised as important outcomes for success of transition.[26,27] Continuity of care is considered a core issue[27], this was also emphasised in a systematic review that showed engagement in adult care (attending first (two) visits) and retention in

adult care (continuing to attend scheduled clinic appointments) were often used in studies investigating transition in chronically ill adolescents.[28]

In another recent study, in IBD patients, their parents and paediatric health care providers, were asked to select 5 of items from the Transition Readiness Assessment Questionnaire (TRAQ), thought to be important for successful transition. All three stakeholders had a different selection of items[29], but all selected items related to adherence, communication with the doctor, calling in case of problems or adverse reactions to medication.[29]

As emphasised in the European Crohn's and Colitis Organisation (ECCO) topical review on transitional care in IBD, it is important to identify objective outcome measures that can be used to define successful transition in IBD.[3] Therefore, the primary aim of this study was to identify outcomes that health care providers working with IBD patients think are important for success of transition in IBD, using a Delphi procedure in 3 types of health care providers working with IBD patients. Our secondary aim was to compare the outcomes identified by health care providers to outcomes selected by a patient panel, which was recruited in second instance.

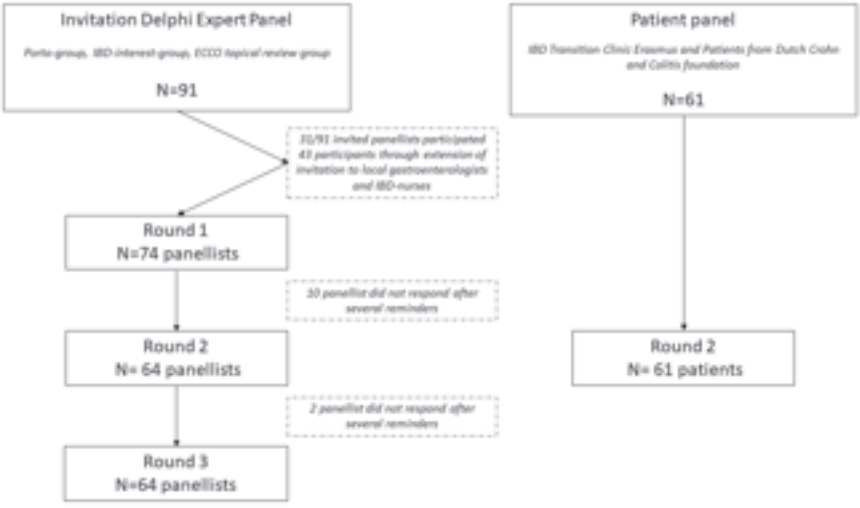
MATERIALS AND METHODS

To identify health care transition outcomes for IBD, we conducted a three-stage Delphi[30-33] process, a commonly used method for reaching consensus. The survey consisted of three rounds, which were designed and distributed using an online survey programme (SurveyMonkey). At each stage, all experts were contacted via e-mail explaining the task to be done, and a web link was included to complete the questionnaire. At each round, participants were given 2 weeks to send in their reply. Every two weeks a reminder was sent to all participant who had not yet replied. After 3 reminders, the web link was closed. We decided to give only factual feedback after each round, to avoid influencing panellists' opinion.[34] The study started July 2016 and ended March 2018.

Delphi Panel

The Delphi panel was composed based on a practical approach. To achieve international consensus, experts in the field of IBD from around the world were invited to participate. Our main aim was to create a balanced panel of all health care providers working with IBD patients in the transition process. Therefore, paediatric gastroenterologists, (adult) gastroenterologists, paediatric and adult IBD nurses

Figure 1 Flowchart composition Delphi panel



were invited. The first step in composing the Delphi Panel was inviting all members of the 'Paediatric IBD Porto and Interest Groups of European Society for Paediatric Gastroenterology Hepatology and Nutrition (ESPGHAN)' and the authors of the European Crohn's and Colitis Organisation Topical Review on transitional care in IBD.[3] The 'Paediatric IBD Porto Group' is a group of 36 paediatric IBD experts from the (ESPGHAN) whose goals are to generate collaborative international research and to provide a leadership role concerning current diagnosis and management of IBD in children. The IBD Interest Group is an open growing group of 48 ESPGHAN members at the time of the study who participate in all activities generated by the Porto group such as collaborative studies and guidelines preparation. Both groups mostly consist of paediatric gastroenterologist from Europa as well as Israel and some from North America. A total of 91 panellists were invited and were asked to participate as well as invite a paediatric IBD-nurse, adult gastroenterologist and adult IBD nurse, from their own hospital. From the 91 invited panellists 31 (34%) agreed to participate. The 31 panellists invited another 43 physicians or nurses and vouched for their credentials. In addition, the website of the hospital where they worked was also checked to double check their credentials. This resulted in 74 panellists (here-after 'Expert panel or panellists'; Figure 1).

After completing the Delphi stages in the expert panel, we concluded that also including the perspective of the adolescent and young adult patients would be of great added value. Therefore, we proceeded to include adolescent and young adult patients, hereafter defined as 'patient panel or patients'. The patients were

recruited from two sources: (a) an ongoing study into transition at the IBD outpatient transition clinic in the Erasmus Medical Centre in Rotterdam (the study was approved by the Medical Ethics Committee and patients provided informed consent), and (b) young adult IBD patients from the Dutch Crohn and Colitis patient organisation. All patients were given three weeks to complete the survey and provided information with regard to their sex, age, disease duration and disease type. The patient panel participated in stage 2 only, but were asked after completing stage 2 if they thought an item was missing.

Stage 1

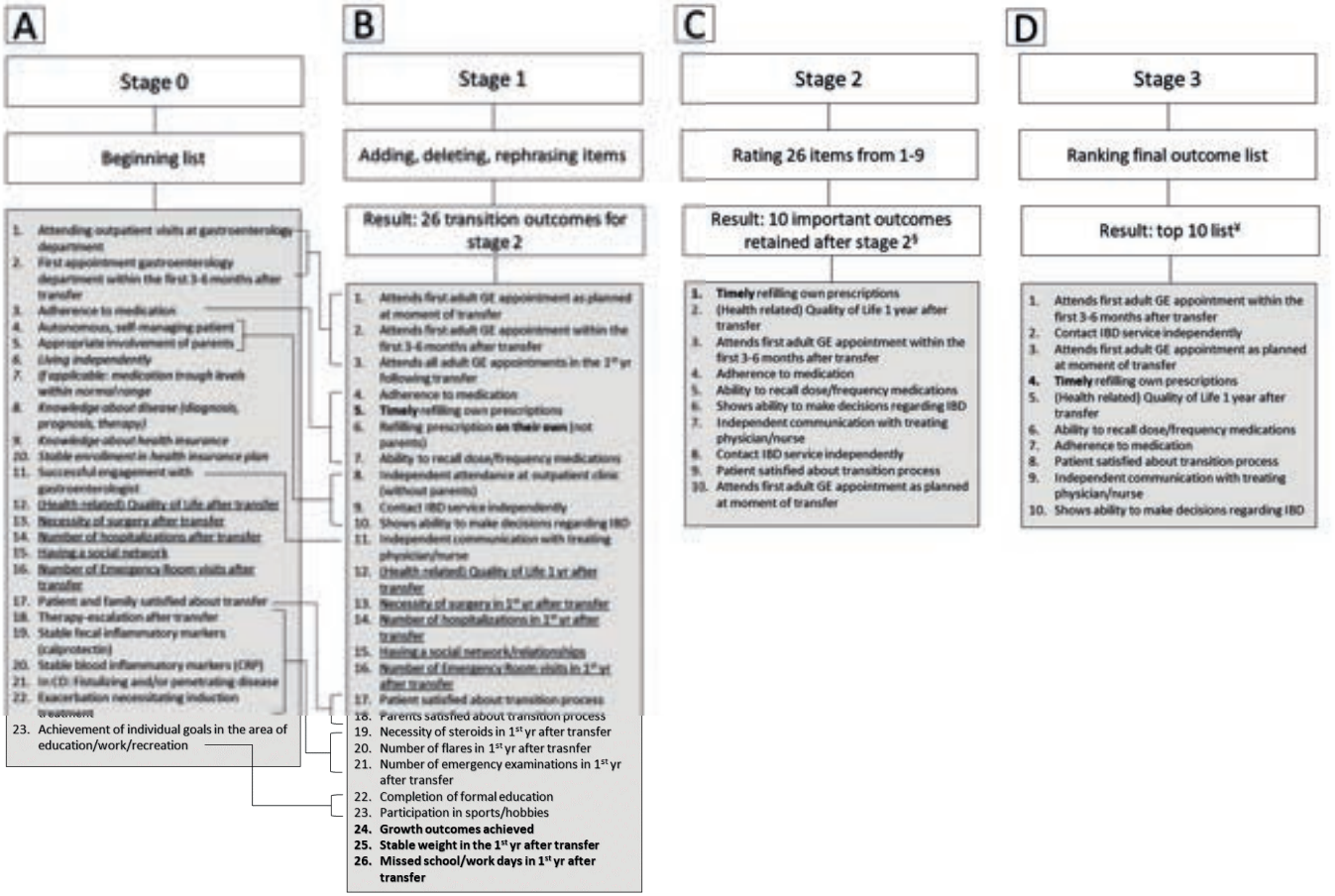
In this first stage, a literature review[4,19,20,26-28,35,36] was performed and a list with items related to outcome of transition was created. This list was sent to the research team, and was discussed in a joined meeting. The 23-item list was sent to all 74 participants of the expert panel (Figure 2A). In stage 1, participants were asked to comment on the list, for example to state if they thought an item should be removed from the list (for reasons of not being associated to outcome of transition), merged with another item, or rephrased. Additionally, participants were invited to add new items to the list. Lastly, all panellists were asked to complete a short form to collect demographic characteristics, such as their name, academic degree(s), department, position and details about the hospital where they work (name, city, country and hospital type (community vs tertiary hospital)).

The research team analysed all responses from stage 1, at first each member evaluated the responses individually, and in a meeting consensus was reached. Criteria to accept items were (a) suggestions to refine or specify items if it improved clarity or (b) every new suggested item related to outcome of transition. Items were rejected or deleted if (a) they were not related to outcome of transition (but to for example organisation or availability or the IBD transition clinic) or (b) showed large similarity with an item already on the list. Similar outcomes were categorised into themes. Country-specific items were deleted, as our aim was to achieve international consensus.

Stage 2

In the second stage, participants were given a brief summary of the results of stage 1, indicating that some items were deleted, rephrased or reformulated, and explaining that the new item list with outcomes of transition consisted of 26 items (Figure 2B). In stage 2, the panellists were asked to rate each item on a scale from 1 (least important outcome of transition) to 9 (very important outcome of transition). At all times, participants could contact the research team to comment or clarify. Before start of the study, the research team agreed to use the ‘Rand

Figure 2 Summary 3 stage Delphi procedure



Note: panel A: italic: removed items, underlined: items not changed in stage 1. Panel B: bold items are newly suggested items. Panel C:§ order of the items based on lowest (1) to highest (10) mean score Table 3. panel D:¥ 10 reflecting most important, 1 least important.

UCLA criteria for agreement', often used in Delphi studies [26,37], to categorize the outcomes as important, equivocal or not important. A threshold for retaining transition outcomes was established, based on the overall level of agreement among participants. Outcomes were labelled important when they had a mean of 7-9 without disagreement, outcomes rated 4 to 6 were considered equivocal, and outcomes rated 1 to 3 were rated as not important. Disagreement was defined as 30% of ratings are in lower third (rating 1-3) and 30% upper third (7-9).[38] Two members of the research team (GB and JCE) analysed the responses and calculated means for each outcome and determined whether disagreement was present. This stage was also completed by the adolescent and young adult patient panel, recruited in second instance.

Stage 3

In the third stage, the expert panel was given a brief summary of the results of stage 2, indicating that using the Rand UCLA criteria, the item list with 26 items was reduced to ten. In stage 3, this list with 10 important outcomes was sent to the panel with the request to rank the items from 1 to 10, with '1' meaning least important outcome of transition, and '10' reflecting essential outcome of transition. It was emphasised that each item could receive only 1 position between 1-10. Thus, the expert panel was now instructed to rank the important items from stage 2 in a top 10 list, forcing them to re-prioritise the items and state which ones they consider most important.

Statistical analysis

Descriptive statistics were used to summarise the panellists' opinions for closed questions at each round. Data were analysed with SPSS 23 (IBM) and were conducted blind to the names of the participants. Open comments were analysed qualitatively and clustered into main themes. For stage 2, according to the 'Rand UCLA criteria for agreement' mean scores were calculated per item, and proportions were given to determine disagreement. Because of a non-normal distribution of the data, subgroups (e.g. patients panel vs expert panel or paediatric vs adult providers) were compared using a Mann Whitney U test. Holms correction for multiple testing was used[39], a corrected p-value below $p<0.05$ was considered significant.

Table 1: Demographic characteristics expert panel (n=74)

		N. %
Gender	Male, %	40.5
Hospital Type	Community Hospital	6 (8.1)
	Tertiary Hospital	68 (91.9)
Department	Paediatrics	39 (52.7)
	Gastroenterology	29 (39.2)
	Internal Medicine	6 (8.1)
Position	Paediatric Gastroenterologist	28 (37.8)
	Paediatric IBD nurse	10 (13.6)
	Gastroenterologist	22 (29.7)
	Adult IBD nurse	11 (14.9)
	Clinical research fellow	1 (1.4)
	Transition manager	1 (1.4)
	Fellow paediatric Gastroenterology	1 (1.4)
Continent of origin	Europe	57 (77.0)
	North America	9 (12.2)
	Asia	8 (10.8)

Table 2 Demographics patient panel (n=61)

		N. % or Median (IQR)
Recruited from	Dutch Crohn and Ulcerative Colitis Patient organisation	20 (32.8)
	IBD Transition Clinic Erasmus Medical Centre	41 (67.2)
Age (years)	Range	16.5-24.7
	Median (IQR)	18.7 (18.1-20.1)
Gender	Male	30 (49.2)
Disease duration (years)	Range	1-18
	Median (IQR)	5.0 (3.0-7.0)
Disease type	Crohn's disease	34 (55.7)
	Ulcerative Colitis	23 (37.7)
	IBD unclassified	4 (6.6)

IQR: interquartile range

RESULTS

Delphi Panel

A total of 74 participants, from 17 countries, agreed to participate in the Delphi expert panel. Seventy seven percent of the experts came from Europe (n=57; Austria n=2, Croatia n=1, Czech Republic n=2, Denmark n=3, England n=18, Finland n=1, Germany n=2, Greece n=2, Hungary n=2, Italy n=1, Lithuania n=1, Scotland n=7, Spain n=1, the Netherlands n=14), the other participants came from Israel (n=8), Canada (n=3) and the United States (n=6) (Table 1). Participants belonged to one of the four core groups: paediatric gastroenterologist, paediatric IBD nurse, gastroenterologist or adult IBD nurse. The clinical research fellow was a medical doctor (MD) working in paediatrics ('paediatric gastroenterologist group'), the transition manager had a Bachelor of Science in adolescent care ('paediatric nurse group') and the fellow paediatric gastroenterology was an MD working in paediatrics ('paediatric gastroenterologist group'). Of the 74 panellists, 40.5% were male, 91.9% worked in a tertiary hospital, 52.7% worked in the paediatric department and 30% of panellists were IBD nurses (Table 1).

Patient Panel (only participating in stage 2)

A total of 61 adolescent and young adult patients were recruited. 67.2% originated from the IBD-transition clinic in the Erasmus Medical Centre, 49.2% was male and mean age was 18.7 years (Table 2).

Stage 1:

All 74 panellists included in the expert panel responded to stage 1. Many panellists responded with suggestions for rephrasing of items already on the list, such as timely medication refill, refilling own prescriptions. Using the suggestions from the panellists, several items were rephrased, split into more parts or specified (see Figure 2A and B). Five items were removed from the list (italic items Figure 2A) for the following reasons: Items 7, 9 and 10 were removed because of their country- and patient-specific nature. In addition, items 6 and 9 were removed because they were not considered to reflect outcome of transition, and other items reflecting autonomy and knowledge were already on the list. Three new items were suggested by the panel and added to the list: growth target achieved, stable weight and missed school/work days (items 24-26 Figure 2B). The new list consisted of 26 items (Figure 2B). Some panellists also suggested items only reflecting disease related knowledge and organisation of the transition process (e.g. good collaboration of paediatric and adult gastroenterologist), and these were not used to refine the list with outcomes.

Stage 2

Of the 74 panellists in the expert panel, 64 (86.5%) responded to stage 2, rating each item with a number from 1-9 (9=most important outcome for transition). Of the 64 remaining participants, 14 were IBD-nurses (21.8%), 28 (43.8%) paediatric gastroenterologists and 22 (34.4%) gastroenterologists. Nine out of 10 non-responders worked in a tertiary hospital (one paediatric gastroenterologist, seven IBD nurses, one research fellow, one transition manager).

Table 3 shows the mean ratings for each of the 26 items for both the expert and the patient panel. For the expert panel, 10 items had a mean score above 7 without disagreement, indicating important outcomes. Top-5 outcomes at this stage were (starting with the most important item): Attends first adult GE appointment as planned (mean 7.92, Standard Deviation [SD] 1.4), patient satisfied about transition process (mean 7.89, SD 1.5), contacting IBD service independently (mean 7.81, SD 1.1), independent communication (mean 7.79, SD 1.2) and shows ability to make decisions regarding IBD (mean 7.59, SD 1.2). The least important outcomes were 'necessity surgery', 'independent attendance outpatient clinic', 'participation sports/hobbies', 'necessity steroids', and 'stable weight'. Continent of origin did not influence grading. Female members of the expert panel gave a significantly higher grade to the items 'patient satisfaction' (mean males 7.2(SD 1.6); mean females 8.4 (SD 1.0); p 0.008), 'Parental satisfaction' (mean males 6.1(SD 1.9); mean females 7.6(SD 1.3); p 0.008), and 'Growth outcomes achieved' (mean males 5.1(SD 2.3); mean females 7.0(SD 2.6); p 0.024).

For the patient panel, 11 of the 26 items had a mean score above 7 without disagreement. The top five outcomes were (starting with the most important item): independent communication (mean 8.39, SD 0.8), shows ability to make decisions regarding IBD (mean 8.16, SD 1.3), adherence to medication, (mean 8.08, SD 1.6), ability to recall dose/frequency medication (mean 7.92, SD 1.5) and timely refilling own prescriptions (mean 7.82, SD 1.7). Least important outcomes were: 'necessity of surgery', 'necessity of steroids', number of 'hospitalisations', 'ER visits' or 'flares' (Table 3). No additional items related to success of transition were suggested by the patient panel. Sex did not influence grading within the patient panel (data not shown).

Comparing the grades from expert (n=64) and the patient (n=57) panel (Table 3), showed that both the patient and the expert panel identified the same 8 items as important (mean grades for these items were quite similar (<1 point difference between both groups). Additionally, the patient panel found 'attends all GE appointments in first year following transfer', 'parents satisfied with transition'

Table 3: Mean importance ratings# of all 26 items from stage 2

Item number		Mean score (SD) expert panel (n=64)	% with a score 1.2 or 3‡	Mean score (SD) patient panel (n=61)	% with a score 1.2 or 3‡	Corrected p-value
1.	Attends first adult GE appointment as planned at moment of transfer	**7.92 (1.4)**	1.6	6.98 (2.0)	4.9	0.036*
2.	Attends first adult GE appointment within the first 3-6 months after transfer	**7.12 (2.0)**	7.8	6.62 (2.1)	11.5	1.000
3.	Attends all adult GE appointments in the 1st year following transfer	6.76 (1.9)	6.3	**7.23 (2.1)**	6.6	0.649
4.	Adherence to medication	**7.48 (1.8)**	3.1	**8.08 (1.6)**	3.3	0.09
5.	Timely refilling own prescriptions	**7.02 (1.5)**	1.6	**7.82 (1.7)**	3.3	0.002*
6.	Refilling prescription on their own (not parents)	6.91 (1.6)	4.7	**7.33 (2.0)**	6.6	0.390
7.	Ability to recall dose/frequency medications	**7.56 (1.5)**	3.1	**7.92 (1.5)**	1.6	0.480
8.	Independent attendance at outpatient clinic (without parents)	5.94 (2.2)	17.2	5.20 (2.8)	32.8	1.000
9.	Contact IBD service independently	**7.81 (1.1)**	0	**7.77 (1.6)**	1.6	1.000
10.	Shows ability to make decisions regarding IBD	**7.59 (1.2)**	1.6	**8.16 (1.3)**	0	0.055
11.	Independent communication with treating physician/nurse	**7.79 (1.2)**	0	**8.39 (0.8)**	0	0.036*
12.	(Health related) Quality of Life 1 year after transfer	**7.11 (1.5)**	1.6	**7.39 (1.9)**	6.6	1.000
13.	Necessity of surgery in 1st year after transfer	5.56 (2.4)	23.4	2.66 (2.1)	72.1	< 0.0001*
14.	Number of hospitalizations in 1st year after transfer	6.36 (2.3)	12.5	3.08 (2.3)	62.3	< 0.0001*
15.	Having a social network/relationships	6.39 (1.9)	10.9	6.46 (2.8)	19.7	1.000
16.	Number of Emergency Room visits in 1st year after transfer	6.59 (2.2)	10.9	3.05 (2.5)	65.6	< 0.0001*
17.	Patient satisfied about transition process	**7.89 (1.5)**	1.6	**7.56 (1.9)**	6.6	1.000
18.	Parents satisfied about transition process	6.92 (1.8)	6.3	**7.28 (1.9)**	6.6	1.000
19.	Necessity of steroids in 1st year after transfer	6.02 (2.5)	18.8	2.64 (2.1)	68.9	< 0.0001*
20.	Number of flares in 1st year after transfer	6.43 (2.3)	14.1	3.15 (2.4)	63.9	< 0.0001*
21.	Number of emergency examinations in 1st year after transfer	6.36 (2.1)	10.9	3.21 (2.2)	59.0	< 0.0001*
22.	Completion of formal education	6.17 (2.2)	14.1	6.70 (2.9)	19.7	0.238
23.	Participation in sports/hobbies	5.97 (2.0)	14.1	5.90 (2.8)	23.0	1.000
24.	Growth outcomes achieved	6.16 (2.6)	18.8	5.41 (2.8)	27.9	1.000
25.	Stable weight in the 1st year after transfer	6.06 (1.9)	9.4	5.72 (2.5)	23.0	1.000
26	Missed school/work days in 1st year after transfer	6.67 (1.8)	6.3	4.10 (2.7)	47.5	< 0.0001*

GE: gastroenterology

 # participants rated each item from 1-9‡ No disagreement was found (30% of ratings in lower third (rating 1-3) and 30% upper third (7-9)). Bold represents mean score 7-9 without disagreement which reflect 'important outcomes'. * corrected p-value using Holms correction for multiple testing < 0.05*

Table 4: Top 10 ranking of stage 3 outcomes all 62 participants and per provider type

Top 10- all 62 participants	Mean (SD)	Top 10- pediatric gastroenterologists (n=28)	Mean (SD)	Top 10-gastroenterologists (n=22)	Mean (SD)	Top 10- IBD- Nurses (n=12)	Mean (SD)
Attends first adult GE appointment within the first 3-6 months after transfer	4.37 (3.2)	Attends first adult GE appointment within the first 3-6 months after transfer	4.11 (3.2)	Attends first adult GE appointment within the first 3-6 months after transfer	4.14 (2.7)	Adherence to medication	4.17 (3.2)
Contact IBD service independently	5.15 (2.4)	Timely refilling own prescriptions	5.04 (2.8)	Attends first adult GE appointment as planned at moment of transfer	4.5 (3.5)	Timely refilling own prescriptions	4.42 (1.9)
Attends first adult GE appointment as planned at moment of transfer	5.40 (3.6)	Ability to recall dose/ frequency medications	5.39 (2.6)	Contact IBD service independently	4.91 (1.9)	Contact IBD service independently	4.58 (3.0)
Timely refilling own prescriptions	5.42 (2.7)	(Health related) Quality of Life 1 year after transfer	5.43 (3.1)	Adherence to medication	5.27 (2.4)	Attends first adult GE appointment within the first 3-6 months after transfer	5.42 (3.8)
(Health related) Quality of Life 1 year after transfer	5.44 (3.3)	Contact IBD service independently	5.57 (2.5)	(Health related) Quality of Life 1 year after transfer	5.32 (3.7)	Ability to recall dose/ frequency medications	5.50 (1.7)
Ability to recall dose/ frequency medications	5.53 (2.8)	Attends first adult GE appointment as planned at moment of transfer	5.79 (3.7)	Patient satisfied about transition process	5.41 (2.7)	(Health related) Quality of Life 1 year after transfer	5.67 (3.2)
Adherence to medication	5.55 (2.3)	Patient satisfied about transition process	5.93 (2.4)	Ability to recall dose/ frequency medications	5.77 (2.3)	Patient satisfied about transition process	6.00 (2.1)
Patient satisfied about transition process	5.76 (2.4)	Independent communication with treating physician/nurse	6.00 (2.9)	Independent communication with treating physician/nurse	6.27 (2.9)	Shows ability to make decisions regarding IBD	6.00 (2.7)
Independent communication with treating physician/nurse	6.31 (2.8)	Adherence to medication	6.32 (2.7)	Timely refilling own prescriptions	6.45 (2.7)	Attends first adult GE appointment as planned at moment of transfer	6.17 (3.8)
Shows ability to make decisions regarding IBD	6.65 (2.7)	Shows ability to make decisions regarding IBD	6.68 (2.6)	Shows ability to make decisions regarding IBD	6.95 (2.7)	Independent communication with treating physician/nurse	7.08 (2.4)

Note: most important outcomes, with highest importance rank are on the lower part of the list.
GE: gastroenterology

and 'refilling prescriptions on own' important, and the expert panel considered 'attend first GE appointment as planned' and 'within the first 3-6 months after transfer' important. For some of the 'non-important' items (mean < 7) differences between both panels were large: disease related outcomes (Items 13,14,16,19-21) and 'school/work absence' (item 26) received a significantly lower mean score by the patient panel (range 2.6-3.3) compared to the expert panel (range 5.5-6.6). Comparing the different providers within the expert panel (adult (n=32) vs paediatric (n=32) health care providers; paediatric (n=28) vs adult (n=22) gastroenterologists; nurses (n=13) to physicians (n=50)) did not show significant differences after correction for multiple testing.

Stage 3

Of the 64 experts, 62 responded to stage 3. Table 4 and Figure 2D display the top-10 ranking of the important outcomes from stage 2. For the panel as a whole 'ability to make decisions regarding IBD' (mean 6.7, SD 2.7), 'independent communication' (Mean 6.3, SD 2.8) and 'patient satisfaction' (mean 5.8, SD 2.4) were the top-3 outcomes, whereas 'attends first GE appointment within the first 3-6 months after transfer' (mean 4.4, SD 3.2), 'contact IBD service independently' (mean 5.2, SD 2.4) or 'attends first GE appointment as planned' (5.4, SD 3.6), received lower scores (Table 4). Differences for the 3 provider types are shown in Table 4. Comparing the different providers within the expert panel (adult (n=32) vs paediatric (n=28) health care providers (see supplementary Figure 1); paediatric (n=28) vs adult (n=22) gastroenterologists; nurses (n=12) to physicians (n=50)) did not show significant differences after correction for multiple testing.

DISCUSSION

The primary objective of this study was to identify outcomes that three types of health care providers responsible for the care of IBD patients (gastroenterologists, paediatric gastroenterologists, and IBD-nurses) thought were important for success of transition in IBD patients. Our secondary aim was to compare these outcomes to the outcomes selected by a patient panel recruited in second instance, i.e. who only participated in stage 2 of this Delphi study. In stage 2, ten and 11 out of 26 items were identified as important by the expert (n=64) and the patient panel (n=61), respectively (Stage 2, Table 3). Surprisingly, results show that both the expert and patient panel thought the same items were important for success of transition: 8 items were identified as important in both the patient and the expert panel. Of these, 6 items concerned self-management skills and autonomy (e.g. independent communication, medication adherence), while the other 2 items

were more general: Health related Quality of Life and patient satisfaction about transition process. In addition, both panels thought that attendance to adult GE appointments was important, but gave slightly different grades (difference < 1 point) to the relevant items (items 1-3, Table 3). Similarly, 'satisfaction of parents about transition process' was considered important in both panels, and almost reached the threshold for importance in the expert panel. Likewise, 'independent attendance at outpatient clinic' received a low grade (<6) by both the expert and the patient panel, possibly reflecting that both value or at least do not disapprove the presence of parents.

Only the expert panel provided a top-10 ranking (=stage 3) of the important items, and this showed that from the 10 important items, decision-making, independent communication and patient satisfaction were considered most important for success of transition in IBD patients. Comparing the top-10 of different providers did not show significant differences.

Remarkably, not one of the IBD/disease-specific items (e.g. surgery, inflammatory markers) were found to be important. The patient panel gave even lower grades than the expert panel. This is in accordance with previous studies discussing successful transition in other chronic diseases such as congenital heart defects[40], rheumatological diseases[41,42], and congenital adrenal hyperplasia.[43] In all these studies successful transition was not defined by disease-specific items, but by attendance to the first (one or two) visits of the adult health care provider. This seems a restricted definition of success. However, continuity of care is seen as a core outcome of transition[27], and is often studied as outcome of transition[28]. Incorporating disease outcomes in the definition of successful transition can be complicated considering the heterogeneous course of chronic diseases such as IBD as well as the case mix that occurs when patients with a severe course are seen in (academic) centers.[6] Philpott et al. (2018) therefore plea for including patient-driven outcomes in the definition of successful transition, such as trust in the adult health care system and autonomy.[6]

In IBD, several studies investigated outcomes of transition. A study by Bollegala *et al.* retrospectively compared outcomes 1 year before vs 1 year after transfer (n=95, no structured transition program) and report fewer outpatient clinic visits and more non-compliance, but no differences in other aspects of health care utilisation.[20] Furthermore, a survey by Bennett *et al.* showed no differences in compliance, complications, surgery, hospitalization rate or number of flares between 46 IBD patients who had transferred to adult care (without a structured transition program) and 36 age-matched patients who received care in adult setting from

the beginning.[44] At last, Cole *et al.* showed that patients who did not attend a transition service, more often needed surgery, hospitalization, and had higher non-attendance and lower treatment adherence than patients that did attend a transition service.[19] These studies suggest that clinical outcomes might be different for patients that followed a structured transition program and those who did not, with possibly better outcomes after structured transition. However, at this point, it is unknown whether absence from the transition program itself is a risk factor for adverse outcomes or if absence is just a surrogate marker of patients that are not able to attend the transition program due to a complicated course of IBD.

So far, a definition of successful transition in IBD has not been formulated. Previously, our research group designed a score measuring success of transition in IBD (the Transition Yourself score).[45] The score comprised four elements: time to first outpatient visit to adult gastroenterologist, adherence to visits at the gastroenterology outpatient clinic, adherence to medication and qualitative evaluation of transition by the patient. The Transition Yourself Score was developed based on literature review and a focus group review with IBD experts, but has not yet been validated. In addition to validating the score, our research group considered it important to ask the opinion of a larger IBD expert panel in identifying items reflecting success of transition and also ask patients' opinion.

Two recent studies used a Delphi study to identify general, non-disease specific indicators for success of transition in adolescent medicine.[26,27] First, Fair *et al.* included 117 experts, mainly from the US (88%), and 70% paediatric professionals. In the final stage, 10 important outcomes were found: Achieving optimal QoL, self-managing own condition, understanding characteristics and complications of condition, knowing names and purposes of medication, adherence to medication, attending most medical appointments, having a medical home, avoidance unnecessary hospitalizations, understanding health insurance, and having a social network. Second, Suris and Akre included 37 adolescent health workers (mainly physicians) from 15 countries. Items found to be important for success of transition were: patient not lost to follow up, no missed consultations, trusting relationship with provider, attention for self-management, first visit adult care within the first 3-6 months after transfer, number of ER visits, patient/family satisfied transfer, maintaining stable disease or improvement.[26]

The selected items in both studies partly resemble the important items identified in this study. However, some of the items are described in general words, e.g. 'managing your own condition', which covers several items from our item list. Due

to this lack of specificity, it remains unclear which specific items are valued most by the panellists in the previous studies. Furthermore, only Suris and Akre's list included a disease related item: stable disease or disease improvement. In our expert and patient panel disease specific parameters received low grades. Moreover, disease knowledge was included in the final outcome list by Fair *et al.* We chose not to include knowledge of disease in our refined list for stage 1, because items implying disease knowledge were already on the list.

Strengths and limitations

This study was strengthened by the appropriate use of the Delphi procedure, and the use of a large multinational expert panel, including health care providers from 17 countries. Secondly, formation of the Delphi panel was initiated by inviting two pre-existing expert groups with a leadership role and expertise in the care of adolescent IBD patients to form the Delphi Panel. Extension of this invitation eventually led to the inclusion of 74 paediatric gastroenterologists, gastroenterologists and IBD nurses from both the paediatric and adult department. Thirdly, in second instance also a patient panel was composed to provide the patient perspective in grading the 26 items in stage 2. Lastly, the expert panel was balanced with 50% of panellists from both the paediatric and adult department. The study was limited by a low response rate in the first invitation round (34%), although 31 opinion leaders in IBD with great experience in transition did participate, and the final panel consisted of 74 members. In addition, the majority of experts worked in Europe, which may reflect a Western perspective. Second, 90% of panellists worked in tertiary hospitals, although this is a limitation, it makes sense considering the fact that most paediatric IBD patients are treated in tertiary hospitals.[46] Third, although using the widely accepted RAND UCLA criteria for agreement, items with a mean below 7 were now labelled 'not important', which could be judged as too stringent. Fourth, it would have been better if the patient panel was included from the beginning of the study, so that all three Delphi stages would have been completed by all participants at the same time. Fifth, the subgroup analyses performed within the expert panel had relatively low number of participants. Finally, to assure clarity of all items in the list, we chose to specify the items as much as possible. As a consequence, sometimes several items concerned the same topic, but had a different emphasis (e.g. 3 items about medication adherence (item 4-6 Figure 2B).

Conclusion

This is the first study investigating outcomes reflecting successful transition in IBD patients using a multinational expert panel and comparing the results to a patient panel. Experts and patients agreed to a great extent: 8 out of 26 items

were found to be important for success of transition, of which 6 items concerned self-management skills/autonomy. Remarkably, no IBD-specific item was found important. The three most important outcomes in the top-10 list from the expert panel were independent decision-making, independent communication and patient satisfaction and did not differ between paediatric gastroenterologists, gastroenterologist and IBD-nurses. Identifying these outcomes can facilitate the definition of successful transition and subsequently the construction of an objective score measuring success of transition. After validation, this score could be used to test the efficacy of the different transition programs, in order to improve transitional care worldwide.

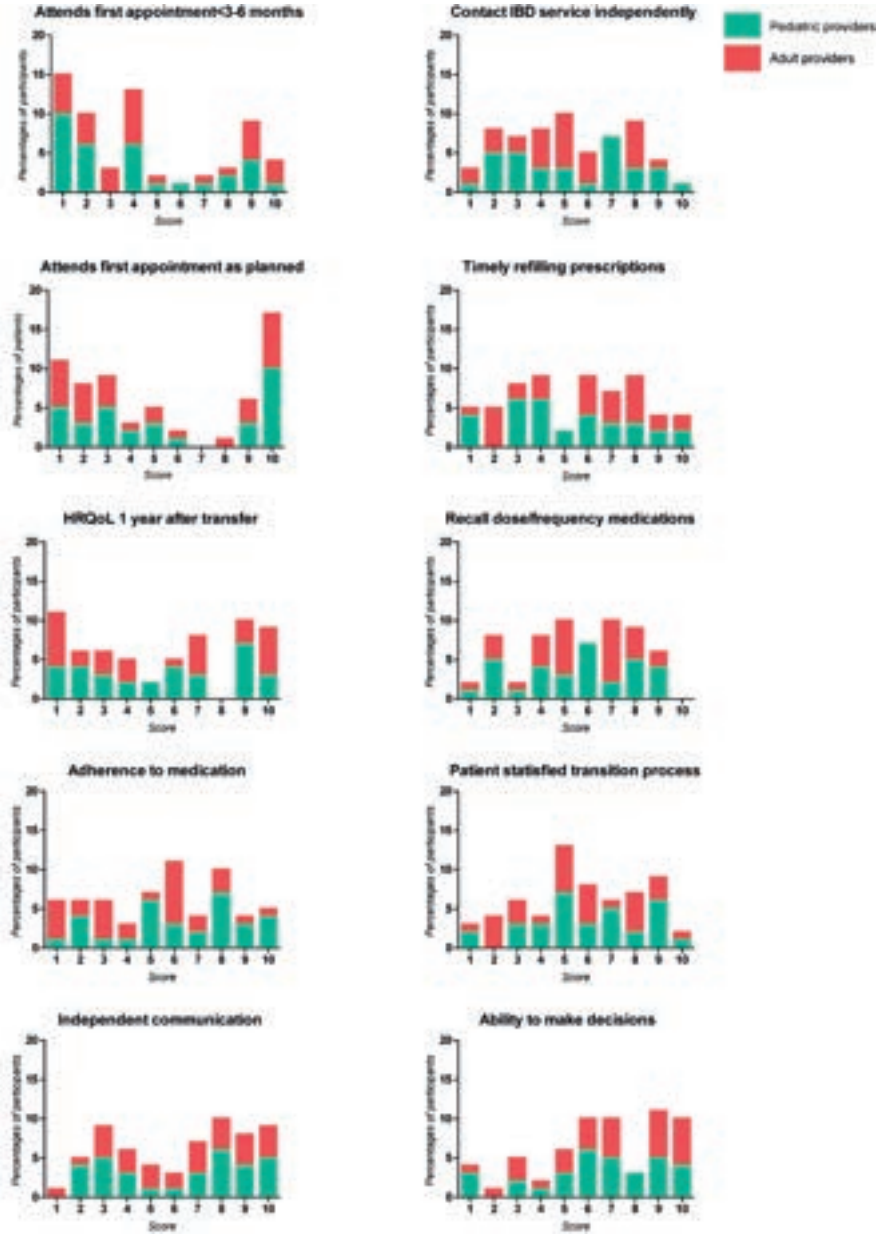
REFERENCES

1. Griffiths AM. Specificities of inflammatory bowel disease in childhood. *Best Pract Res Clin Gastroenterol* 2004;**18**:509-23.
2. Adamiak T, Walkiewicz-Jedrzejczak D, Fish D*, et al.* Incidence, clinical characteristics, and natural history of pediatric ibd in wisconsin: A population-based epidemiological study. *Inflamm Bowel Dis* 2013;**19**:1218-23.
3. van Rheenen PF, Aloi M, Biron IA*, et al.* European crohn's and colitis organisation topical review on transitional care in inflammatory bowel disease. *J Crohns Colitis* 2017;**11**:1032-8.
4. Brooks AJ, Smith PJ, Cohen R*, et al.* Uk guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. *Gut* 2017;**66**:988-1000.
5. Blum RW, Garell D, Hodgman CH*, et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the society for adolescent medicine. *J Adolesc Health* 1993;**14**:570-6.
6. Philpott JR, Kurowski JA. Challenges in transitional care in inflammatory bowel disease: A review of the current literature in transition readiness and outcomes. *Inflamm Bowel Dis* 2018.
7. van Groningen J, Ziniel S, Arnold J, Fishman LN. When independent healthcare behaviors develop in adolescents with inflammatory bowel disease. *Inflamm Bowel Dis* 2012;**18**:2310-4.
8. Fishman LN, Ziniel SI, Adrichem ME, Fernandes SM, Arnold J. Provider awareness alone does not improve transition readiness skills in adolescent patients with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2014;**59**:221-4.
9. Trivedi I, Keefer L. The emerging adult with inflammatory bowel disease: Challenges and recommendations for the adult gastroenterologist. *Gastroenterol Res Pract* 2015;**2015**:260807.
10. Fishman LN, Barendse RM, Hait E, Burdick C, Arnold J. Self-management of older adolescents with inflammatory bowel disease: A pilot study of behavior and knowledge as prelude to transition. *Clin Pediatr (Phila)* 2010;**49**:1129-33.
11. Wright EK, Williams J, Andrews JM*, et al.* Perspectives of paediatric and adult gastroenterologists on transfer and transition care of adolescents with inflammatory bowel disease. *Intern Med J* 2014;**44**:490-6.
12. Goodhand J, Hedin CR, Croft NM, Lindsay JO. Adolescents with ibd: The importance of structured transition care. *J Crohns Colitis* 2011;**5**:509-19.
13. Brooks AJ, Smith PJ, Lindsay JO. Monitoring adolescents and young people with inflammatory bowel disease during transition to adult healthcare. *Frontline Gastroenterol* 2018;**9**:37-44.
14. Escher JC. Transition from pediatric to adult health care in inflammatory bowel disease. *Dig Dis* 2009;**27**:382-6.
15. Dabadie A, Troadec F, Heresbach D*, et al.* Transition of patients with inflammatory bowel disease from pediatric to adult care. *Gastroenterol Clin Biol* 2008;**32**:451-9.
16. Nakhla M, Daneman D, To T, Paradis G, Guttmann A. Transition to adult care for youths with diabetes mellitus: Findings from a universal health care system. *Pediatrics* 2009;**124**:e1134-41.
17. Wray J, Frigiola A, Bull C, Adult Congenital Heart disease Research N. Loss to specialist follow-up in congenital heart disease; out of sight, out of mind. *Heart* 2013;**99**:485-90.
18. Andemariam B, Owarish-Gross J, Grady J*, et al.* Identification of risk factors for an unsuccessful transition from pediatric to adult sickle cell disease care. *Pediatr Blood Cancer* 2014;**61**:697-701.
19. Cole R, Ashok D, Razack A, Azaz A, Sebastian S. Evaluation of outcomes in adolescent inflammatory bowel disease patients following transfer from pediatric to adult health care services: Case for transition. *J Adolesc Health* 2015;**57**:212-7.
20. Bollegala N, Brill H, Marshall JK. Resource utilization during pediatric to adult transfer of care in ibd. *J Crohns Colitis* 2013;**7**:e55-60.
21. Crowley R, Wolfe I, Lock K, McKee M. Improving the transition between paediatric and adult healthcare: A systematic review. *Arch Dis Child* 2011;**96**:548-53.
22. Mackie AS, Islam S, Magill-Evans J*, et al.* Healthcare transition for youth with heart disease: A clinical trial. *Heart* 2014;**100**:1113-8.
23. McDonagh JE, Southwood TR, Shaw KL, British Society of P, Adolescent R. The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)* 2007;**46**:161-8.

24. de Silva PS, Fishman LN. Transition of the patient with ibd from pediatric to adult care-an assessment of current evidence. *Inflamm Bowel Dis* 2014;**20**:1458-64.
25. Leung Y, Heyman MB, Mahadevan U. Transitioning the adolescent inflammatory bowel disease patient: Guidelines for the adult and pediatric gastroenterologist. *Inflamm Bowel Dis* 2011;**17**:2169-73.
26. Fair C, Cuttance J, Sharma N*, et al.* International and interdisciplinary identification of health care transition outcomes. *JAMA Pediatr* 2016;**170**:205-11.
27. Suris JC, Akre C. Key elements for, and indicators of, a successful transition: An international delphi study. *J Adolesc Health* 2015;**56**:612-8.
28. Rachas A, Lefeuvre D, Meyer L*, et al.* Evaluating continuity during transfer to adult care: A systematic review. *Pediatrics* 2016;**138**.
29. Gray WN, Reed-Knight B, Morgan PJ*, et al.* Multi-site comparison of patient, parent, and pediatric provider perspectives on transition to adult care in ibd. *J Pediatr Nurs* 2018;**39**:49-54.
30. Hsu CC, Sandford BA. The delphi technique: Making sense of consensus. *Practical Assessment Research and Education.* 2007:**12**:1-8.
31. Dewa LH, Murray K, Thibaut B*, et al.* Identifying research priorities for patient safety in mental health: An international expert delphi study. *BMJ Open* 2018;**8**:e021361.
32. Russell D, Atkin L, Betts A*, et al.* Using a modified delphi methodology to gain consensus on the use of dressings in chronic wounds management. *J Wound Care* 2018;**27**:156-65.
33. Snelson E, Ramlakhan S. Which observed behaviours may reassure physicians that a child is not septic? An international delphi study. *Arch Dis Child* 2018: **103:** 864-867.
34. Hasson F, Keeney S, McKenna H. Research guidelines for the delphi survey technique. *J Adv Nurs* 2000;**32**:1008-15.
35. Oswald DP, Gilles DL, Cannady MS*, et al.* Youth with special health care needs: Transition to adult health care services. *Matern Child Health J* 2013;**17**:1744-52.
36. Sattoe JNT, Hilberink SR, van Staa A. How to define successful transition? An exploration of consensus indicators and outcomes in young adults with chronic conditions. *Child Care Health Dev* 2017;**43**:768-73.
37. Elwyn G, O'Connor A, Stacey D*, et al.* Developing a quality criteria framework for patient decision aids: Online international delphi consensus process. *BMJ* 2006;**333**:417.
38. Fitch K, Bernstein S, Aguilar MD*, et al.* The RAND/UCLA appropriateness method user's manual. Santa Monica: Rand; 2001. https://www.rand.org/pubs/monograph_reports/MR1269.html. Accessed February 2019.
39. Holms S. A simple sequential rejective method procedure. *Scandinavian Journal of Statistics* 1979;**6**:65-70.
40. Reid GJ, Irvine MJ, McCrindle BW*, et al.* Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics* 2004;**113**:e197-205.
41. Hazel E, Zhang X, Duffy CM, Campillo S. High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. *Pediatr Rheumatol Online J* 2010;**8**:2.
42. Jensen PT, Karnes J, Jones K*, et al.* Quantitative evaluation of a pediatric rheumatology transition program. *Pediatr Rheumatol Online J* 2015;**13**:17.
43. Gleeson H, Davis J, Jones J, O'Shea E, Clayton PE. The challenge of delivering endocrine care and successful transition to adult services in adolescents with congenital adrenal hyperplasia: Experience in a single centre over 18 years. *Clin Endocrinol (Oxf)* 2013;**78**:23-8.
44. Bennett AL, Moore D, Bampton PA, Bryant RV, Andrews JM. Outcomes and patients' perspectives of transition from paediatric to adult care in inflammatory bowel disease. *World J Gastroenterol* 2016;**22**:2611-20.
45. van den Brink G, van Gaalen MAC, Zijlstra M*, et al.* Self-efficacy did not predict the outcome of the transition to adult care in adolescents with inflammatory bowel disease. *Acta Paediatr* 2019: **108**(2):333-338 .
46. de Bie CI, Buderus S, Sandhu BK*, et al.* Diagnostic workup of paediatric patients with inflammatory bowel disease in europe: Results of a 5-year audit of the eurokids registry. *J Pediatr Gastroenterol Nutr* 2012;**54**:374-80.

SUPPLEMENTARY FIGURES

Supplementary Figure 1 Top-10 outcomes stage 3: Paediatric versus adult providers



Note: Ranking 1 (least important) to 10 (most important); Providers includes physicians as well as nurses

Chapter 7

Effectiveness of Transitional Care in Inflammatory Bowel Disease; Development, Validation, and Initial Outcomes of a Transition Success Score

MAC van Gaalen, M van Pieterson, P Waaijenberg, A Kindermann, V Wolters, A Dijkstra, H van Wering, M Wessels, L de Ridder, D Rizopoulos, CLAAP. Derikx, JC. Escher, on behalf of the Kids with Crohn's, Colitis (KiCC) Working Group for Collaborative Paediatric IBD Research in the Netherlands, the Dutch Initiative on Crohn and Colitis (ICC) and Dutch Nurses Network Inflammatory Bowel Disease (NIBD).

J Crohn's Colitis 2024; Nov 2:jjae166. doi: 10.1093/ecco-jcc/jjae166. Online ahead of print

ABSTRACT

Background and Aims

The effectiveness of transition programs from paediatric to adult healthcare in adolescents with inflammatory bowel disease is not clear, as prospective studies using validated outcome measures for transition are lacking. This study aimed to develop and validate a quantitative Transition Success Score, and to apply it in a multicenter setting to assess the effectiveness of transitional care.

Methods

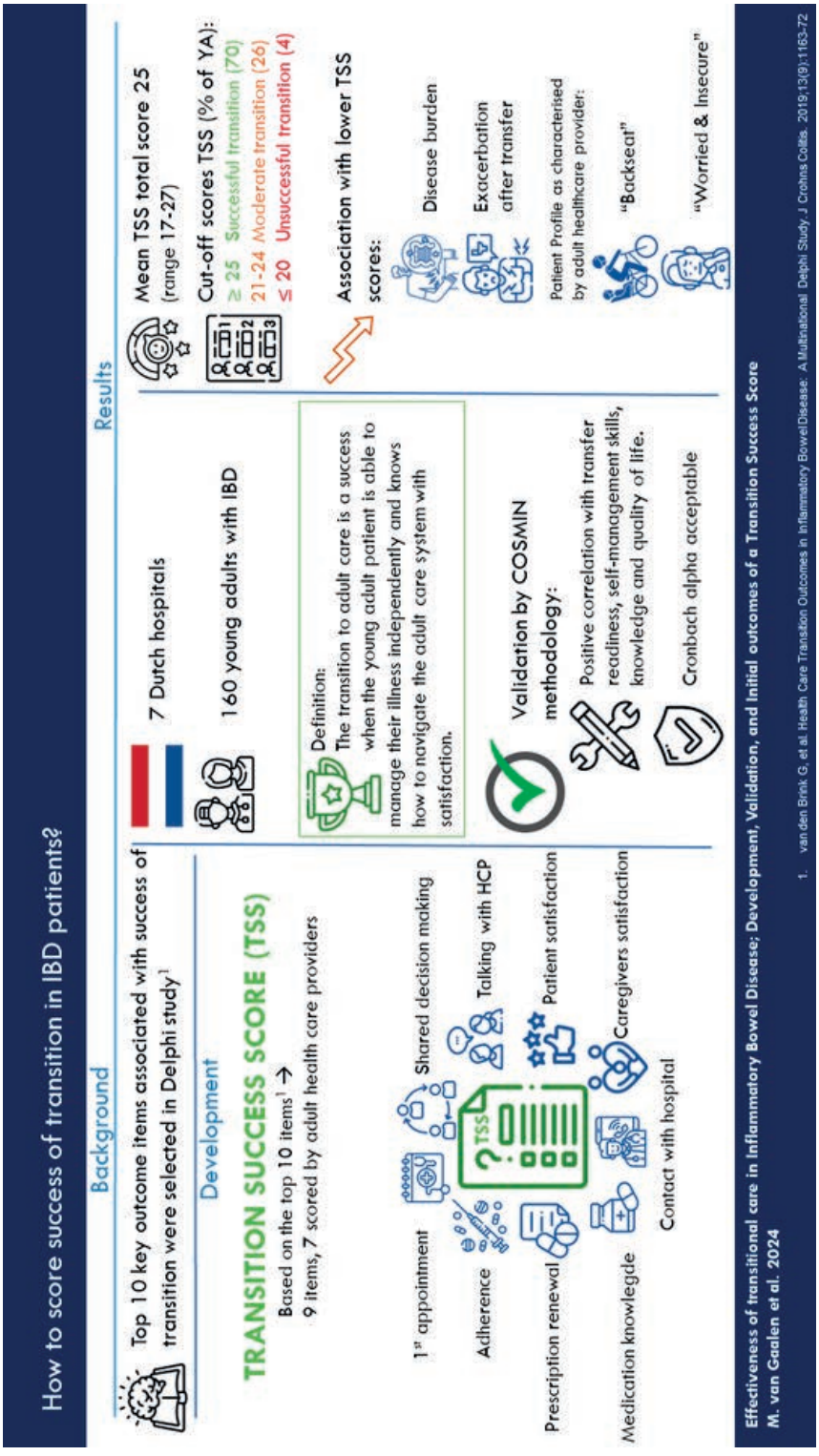
The Top 10 outcome items related to successful transition, identified through an international Delphi study with IBD stakeholders, were integrated into a generic questionnaire, the Transition Success Score. In a prospective, multicenter study, Transition Success Score was scored by adult healthcare providers, young adult patients and caregivers, 9-15 months after transfer of care.

Results

In seven Dutch hospitals, 160 patients completed the Transition Success Score. The mean score was 25 (range 17-27), 25.6% of patients achieving maximum score. Hypothesis testing for construct validity revealed significant associations with characteristics related to transitional care, such as knowledge, independence, and quality of life ($p < 0.005$). Structural validation indicated the score was most effective at discerning lower levels of transition success. Internal consistency was acceptable (0.64). High disease burden, exacerbation during or after transfer, and certain personality profiles were associated with lower scores.

Conclusions

The Transition Success Score serves as a quantitative tool to evaluate the effectiveness of transitional care interventions and to identify inflammatory bowel disease patients at risk of encountering challenges during the transition to adult healthcare.


How to score success of transition in IBD patients?
Background
Top 10 key outcome items associated with success of transition were selected in Delphi study[1]
Development
TRANSITION SUCCESS SCORE (TSS)
Based on the top 10 items[1] →
9 items, 7 scored by adult health care providers
1st appointment
Shared decision making
Adherence
Talking with HCP
Prescription renewal
Patient satisfaction
Medication knowlegde
Caregivers satisfaction
Contact with hospital
TSS
Results
7 Dutch hospitals
160 young adults with IBD
Definition:
The transition to adult care is a success when the young adult patient is able to manage their illness independently and knows how to navigate the adult care system with satisfaction.
Validation by COSMIN methodology:
Positive correlation with transfer readiness, self-management skills, knowledge and quality of life.
Cronbach alpha acceptable
Mean TSS total score 25
(range 17-27)
Cut-off scores TSS (% of YA):
≥ 25 Successful transition (70)
21-24 Moderate transition (26)
≤ 20 Unsuccessful transition (4)
Association with lower TSS scores:
Disease burden
Exacerbation after transfer
Patient Profile as characterised by adult healthcare provider:
"Backseat"
"Worried & Insecure"
Effectiveness of transitional care in Inflammatory Bowel Disease; Development, Validation, and Initial outcomes of a Transition Success Score
M. van Gaalen et al. 2024
1. van den Brink G, et al. Health Care Transition Outcomes in Inflammatory Bowel Disease: A Multinational Delphi Study J Crohns Colitis. 2019;13(9):1163-72

INTRODUCTION

Successful transition from pediatric to adult healthcare is crucial for the health and well-being of young adults (YAs, aged 18-25 years) with chronic conditions such as inflammatory bowel disease (IBD), including Crohn's disease and ulcerative colitis [1,2]. However, ensuring structured and high-quality transitional care for all patients to achieve successful transition poses challenges. It requires strong commitment of healthcare providers to support YA patients in navigating the changes they will encounter in adult healthcare while considering their physical, psychological, and social health status. [1,3,4]. Additionally, caregivers must allow their adolescent children to develop independence gradually. Moreover, empowerment of YA patients with disease knowledge, autonomy, self-efficacy, and self-management skills is crucial [2,5-7]. These aspects can be particularly challenging as they coincide with a developmental period filled with changes in many areas, such as school, work, housing, and relationships.

The implementation of a structured transition program in IBD has been reported to improve the YA's self-management skills, disease knowledge, and quality of life [1,2,8-10]. There is however no evidence favouring 1 transitional care model over others due to lack of prospective studies using a clear definition of transition success [11,12] . Studies evaluating transition success often rely on non-validated, or qualitative tools based on patients› experience or satisfaction [11-15].

Previously, our research group conducted a Delphi study identifying key outcomes of transition success for IBD patients [16]. Table 1 displays the Top 10 items determining transition success (of increasing importance, so number 10 as most important item), selected by an international panel of adolescent and YA IBD patients, pediatric and adult gastroenterologists, and specialized nurses. The most crucial factor was *"demonstration of decision-making skills in relation to IBD"* as well as other self-management skills that were prioritized over IBD-specific factors such as *"frequency of exacerbations"* and "*surgery*". In the absence of a clear definition, the following definition of transition success was used for this study, based on the findings of this top 10 list: The transition to adult care is a success when the YA patient is able to manage their illness independently and knows how to navigate the adult care system with satisfaction.[16,17]

Table 1: Top 10 items associated with transition success (16)

1.	Attends first adult healthcare appointment within the first 3-6 months after transfer.
2.	Contacts IBD service independently
3.	Attends first adult healthcare appointment as planned at moment of transfer.
4.	Fills own prescriptions on time.
5.	(Health related) Quality of Life 1 year after transfer.
6.	Ability to recall dose/frequency of medication.
7.	Medication adherence
8.	Patient satisfaction with transition process
9.	Independent communication with treating physician/nurse
10.	Shows ability to make decisions regarding IBD

IBD: inflammatory bowel disease.

Our Delphi study was the starting point of the development of an outcome measure to assess transitional care effectiveness. The present multicentre study in YAs with IBD aimed to (1) create and validate a Transition Success Score (TSS) based on the Top 10 list from our Delphi study and (2) prospectively use this TSS score to evaluate outcome of transitional care in a multicentre setting.

MATERIALS AND METHODS

The COSMIN methodology [18] guided the development and validation of a Patient Report Outcome Measurement (PROM). An international consensus procedure was utilized to create the Transition Success Score (TSS), which underwent initial testing in a pilot study. Subsequently, a multicentre prospective validation study was conducted using the finalized TSS.

The Research Ethics Review Board of the Erasmus University Medical Centre approved this study (MEC-2017-459). Informed consent was obtained from all patients before recruitment into the study.

Development of the TSS

All 64 healthcare experts who participated in our previous Delphi study [16] were contacted to be involved in the development of the TSS, based on the outcomes of our previous Delphi study (Table 1). Of 64 international healthcare experts, 60 confirmed their availability (see supplemental Table 1 for demographic characteristics).

The initial version of the English language TSS was developed by our research team (MvG, JCE). For each item on the Top 10 item list, we formulated a single question with three answer options. Four rounds of the Delphi process were

conducted between October 2019 to July 2020 using the online survey platform SurveyMonkey. These rounds aimed to collect feedback (agreement or disagreement) from participating experts regarding the formulated questions and answer options. Each round allowed a four-week timeframe for participants to submit their responses, with reminders sent every week to non-respondents. The survey link was closed after three reminders were sent or when response rate reached 80%.

Figure 1 shows that consensus (agreement above the 80%) was reached on nine items of the TSS after four rounds. During the first round, the experts discussed the complexity of measuring and establishing a relationship between quality of life (QoL) and transition success. It was then decided not to include QoL in the TSS, but to use it as a validation measure, as previous studies have shown that good quality transitional care can lead to better QoL [15,19,20].

The first version of the TSS
Following the four Delphi rounds, the TSS included seven items regarding patient's disease management (assessed by healthcare providers) and two items related to patient satisfaction (assessed by both the patient and caregiver). The maximum cumulative score of the TSS is twenty-seven, with higher scores indicating a more successful transition.

Pilot study with the first version of the TSS
From August to October 2020, we performed a pilot study to assess reliability and ease of completing the first version of the TSS among 44 IBD patients (age range 18.7-20.8 years) who had 9 to 15 months earlier transitioned to adult care in Erasmus MC, Rotterdam, The Netherlands. YA also completed the Rotterdam Transition Test (RTT) to assess disease knowledge [6], the Transition Readiness Assessment Questionnaire (TRAQ-NL) to assess self-management skills [5], and the Inflammatory Bowel Disease Questionnaire (IBD-Q) to assess QoL [21]. Efficient completion of the TSS within five minutes was demonstrated, and its reliability was deemed satisfactory (Cronbach's alpha = 0.78). Since not all questions achieved a Cohen's kappa score exceeding 0.61 (ranging from 0.44 to 1.00), modifications were made to enhance the differentiation between response options and provide additional clarification where necessary (Table 2; final version of the TSS).

Validation of the TSS

Validation was carried out using the COSMIN method, which involves construct validation, structural validation, and measurement of reliability.

Figure 1: overview Delphi study; consensus development TSS

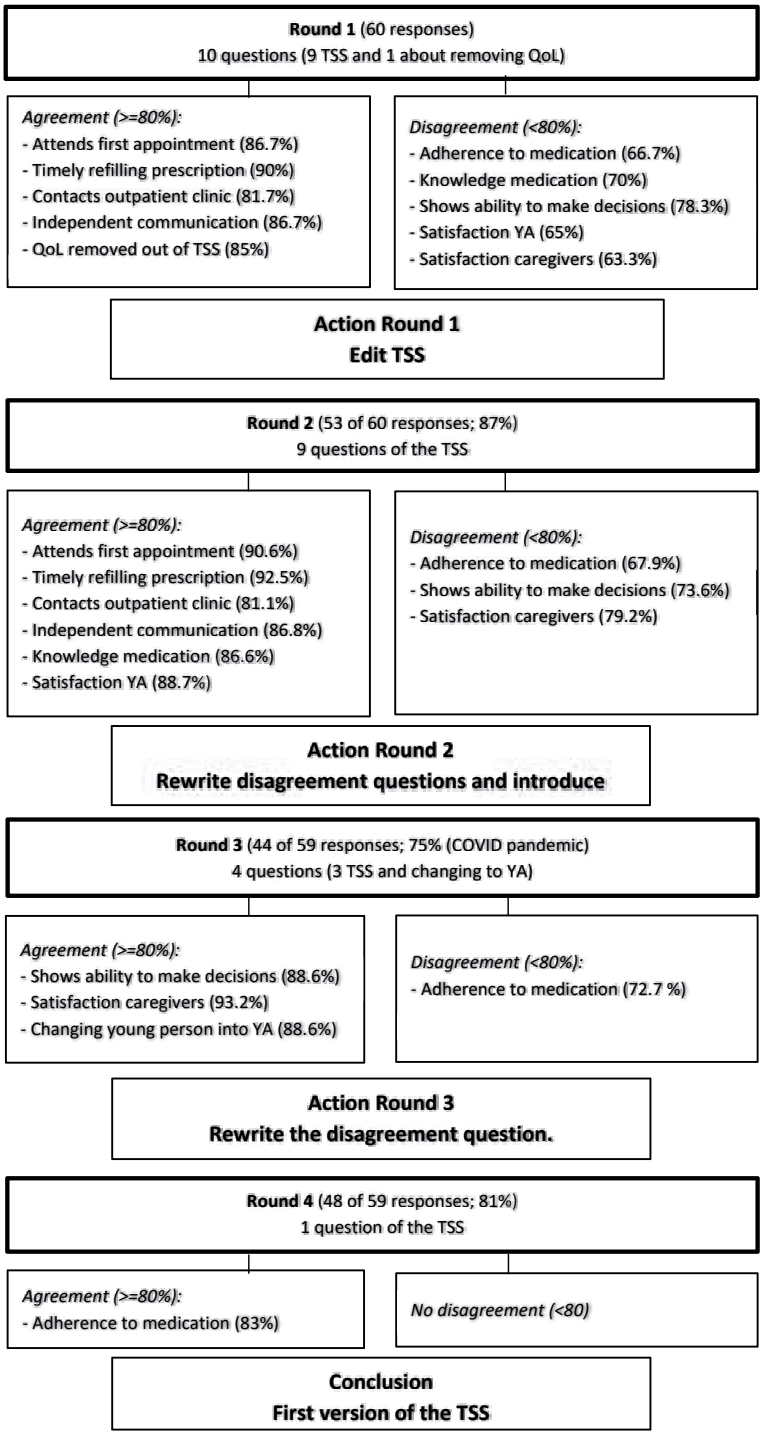


Table 2: Final version of the Transition Success Score with the total scores of the YA

Question (by who)	**Answer options (how to score)** {awarded points}	**Score of all YA (n=160)** n (%)
Q1 (by professional) Has the YA (adolescent/ young adult) attended their first appointment at the adult GI department?	**A1 (chart review)**	
	· Yes, YA attended at the first appointment at the adult GI department *(also applies if the YA has rescheduled, in short term, the appointment for legitimate reasons)* {3}	142 (89)
	· No, the YA did not attend the first appointment, but the YA did attend a new appointment within 6 months after the last visit in pediatric care. {2}	14 (9)
	· No, YA did not attend any visit at the adult GI department within 6 months. {1}	4 (2)
Q2 (by professional) Has the YA adhered correctly to their medical treatment (oral, rectal, infusion, injections) since last visit?	**A2 (Estimate of the professional, checking during visit)**	
	· Good adherence; took medication always or most of the time *(> 80% of doses)* {3}	143 (90)
	· Partial adherence: did not take medication at all times or did not take all types of medication *(e.g. good adherence to oral, but not rectal medicatio*n). {2}	12 (7)
	· Significant non-adherence: stopped all or most medication or did not show up for infusions. {1}	5 (3)
Q3 (by professional) Does the YA show that he/she can organize a renewal of their prescription(s) by themselves? Or in case of infusion medication: Has the YA planned their infusions independently?	**A3 (chart review on request renewal of prescription OR check by YA if he/she know how to arrange)**	133 (83)
	· Yes, YA shows that he/ she can organize a renewal of their prescription/ infusion appointment by themselves{3}	
	· YA organized a renewal of their prescription/infusion appointment together with caregiver(s). {2}	21 (13)
	· No, renewal of their prescription/ infusion appointment always organized by the caregiver(s). {1}	6 (4)
Q4 (by professional) Has the YA been able to recall name/ dose/ frequency of his/her medication?	**A4 (check during visit)**	
	· Yes, YA was able to recall name and dose and frequency correctly *(in case of infusion therapy: able to recall name and dosing interval only).* {3}	147 (92)
	· No, YA could only recall name or dose/frequency correctly. {2}	12 (7)
	· No, YA was not able to give correct answers regarding any medication {1}	1 (1)
Q5 (by professional) When the YA has a question relating to their disease, does they contact the hospital and health care professionals themselves?	**A5 (chart review on who contacts OR check by/ assess during visit)**	
	· Yes, the YA will/is able to contact the hospital by themselves. {3}	131 (82)
	· Sometimes caregivers make contact or YA needs help from caregivers to contact the hospital. {2}	25 (16)
	· No, the YA is unable to contact the hospital or caregiver(s) have always contacted the hospital and health care professionals. {1}	4 (2)

Q6 (by professional) Does the YA talk independently with the treating physician/nurse?	**A6 (check during visit)** • Yes. Caregivers are allowed to be present during the appointment but ask almost no questions (*comparable to a partner who is present during an appointment with an adult patient*). {3} • YA needs help from caregivers to ask/ answer certain questions or to be reminded to ask something. {2} • No, most (> 50%) of the communication went through the caregiver(s). {1}	 145 (91) 13 (8) 2 (1)
Q7 (by professional) Do you feel that the YA is involved in shared decision making, and that the YA can discuss independently with the health care provider about treatment, tests and other disease-related issues?	**A7 (assessment of the professional, during visit)** • Yes, the YA is independently involved in shared decision making and contributes in the care process. If necessary, the YA consults with a caregiver on an equivalent level (*comparable to a partner who is present during an appointment with an adult patient*). {3} • Yes, but the YA needs help/guidance from the caregiver(s) in shared decision making (*for example, in the final decision making*) or caregivers lead a major part of the conversation. {2} • No, there is no shared decision making with the YA *(no share decision making at all or only by the caregiver(s))* {1}	 131 (82) 28 (17) 1 (1)
Q8 (by YA) How satisfied are you with the transition process? Give a score (1 = completely unsatisfied and 10 = completely satisfied)	**A8 (ask the YA)** • Satisfied (score 8-10) {3} • Neutral (score 5-7) {2} • Unsatisfied (score 1-4) {1}	 99 (62) 57 (36) 4 (2)
Q9 (by caregiver) How satisfied are you, as a caregiver, with the transition process? Give a score (1 = completely unsatisfied and 10 = completely satisfied)	**A9 (ask the caregiver)** · Satisfied (score 8-10) {3} · Neutral (score 5-7) {2} · Unsatisfied (score 1-4) {1}	 85 (53) 71 (44) 4 (3)

TSS: Transition Success Score, Q: Question, A: Answer option, YA: Young Adult

Patients

Between May 2021 and April 2023, seven Dutch hospitals (four academic and three non-academic hospitals) enrolled YA with IBD who had transitioned to adult healthcare 9-15 months before. This time frame was chosen to ensure that YAs were used to the responsibilities and routines of adult care and could still recall the transfer experience. Following the COSMIN methodology, our goal was to collect 150 completed TSS, which is deemed sufficient for validation purposes. [18] All eligible YAs were invited to participate, and in cases of refusal, reasons for declining were documented. Although transition programs varied between the hospitals, most had a transition protocol or clinic to focus on transitional care within the local setting. Following standard practice in the Netherlands, patients usually transferred to adult care around 18 years of age.

Data collection

The receiving adult healthcare providers were responsible for reporting on 7 of the 9 items of the TSS during an outpatient visit. Concurrently, YAs and their caregivers were instructed to digitally respond to the two satisfaction-related questions, translated into Dutch. YA also completed validated questionnaires related to transition; the RTT (knowledge)[6], TRAQ-NL (self-management)[5], and the IBD-Q (QoL)[21]. Transfer readiness assessments were collected from YA, pediatric and adult healthcare providers using a 100-point visual analogue scale (VAS). In addition, YA self-assessed their level of independence using a VAS. All questionnaires were requested to be completed within a four-week period, encompassing two weeks before a routine outpatient visit to two weeks after. The questionnaires were distributed using Lime Survey version 3.1.1 (LimeSurvey GmbH, Hamburg, Germany).

To evaluate the construct validity of the TSS, we examined the correlation between the TSS total score and self-management skills and transition readiness characteristics (Table 3). Since no major changes were made to the TSS after the pilot phase, the final validation analysis included the TSS outcomes as along with the results of the TRAQ, RTT, and QoL assessments from the pilot study.

Predictors of successful transition

To identify predictive factors regarding success of transition, demographic information, disease behaviour, disease activity assessment at the time of transfer and at the time of completion of the TSS were collected through chart review and a supplementary questionnaire for YA on education level, family composition, patients' and caregivers' country of birth. YA conducted self-assessments on disease acceptance using a 4-point Likert scale response to the question, "*I can accept that*

I will have this disease for the rest of my life." Additionally, perceived disease burden was self-assessed on a 4-point Likert scale in response to the question, "*How much do you currently suffer from your illness?"*. Identification of IBD exacerbation between transfer and completion of the TSS was based on initiation of or changes in medication due to increased symptoms or the presence of endoscopic disease activity. Disease activity was scored according to the Physician Global Assessment (PGA), which categorizes disease activity as normal, mild, moderate or severe.

A previous study using Q methodology categorized a group of chronically ill YA into four personality and behavioral profiles.[22] The «*Conscious & Compliant*" profile denoted individuals with a high level of involvement in disease knowledge and management. The "*Backseat Patient*" profile characterized YA who were less mature and relied more on their caregivers. Patients expressing a strong desire to be transparent about their disease, not concealing it but embracing it in their daily lives, were classified as *"Self-confident & Autonomous"*. The *"Worried & Insecure"* profile encapsulated patients primarily concerned about their disease. In the present study, both pediatric and adult healthcare providers utilized these profiles to determine the patient type for each YA within the context of transition.

Statistical analysis

Statistical analyses were performed conducted SPSS for Windows, version 28.0.1.0 (IBM SPSS Statistics for Windows, Armonk, NY, USA). Significance was determined at a p-value <0.05. Only fully completed TSS questionnaires were included in the analysis. Incompletely answered supplementary questionnaires were excluded from the analysis, leading to fewer participants for the corresponding analysis.

Validation phase

To compare the independent variables of the hypothetical constructs in Table 3, t-tests were used for dichotomous variables and Pearson's correlation coefficients for continuous variables. Structural validation was conducted using Rasch analysis, a method that assesses question characteristics, identifies the need for question modification, and evaluated the questions' ability to distinguish between different levels of success of transition, which is the latent variable. This latent variable is normally distributed with a mean of 0 and a variance of 1. A value of 0 represents the mean transition success, and the range of all values is from -4 to 4.[23]. The reliability of the entire TSS was evaluated using Cronbach's alpha.

Table 3: Construct validation of the TSS with hypothesis testing

Association of total TSS scores of patients with IBD with specific variables:	Measurement	Pearson correlation	p-value
TSS should correlate positively with higher VAS self-management scored by YA	VAS self-management YA	0.348	**<0.001**
TSS should correlate positively with higher VAS transfer readiness scored by YA	VAS transfer readiness YA	0.317	**<0.001**
TSS should correlate positively with higher VAS transfer readiness scored by paediatric health care providers	VAS transfer readiness paediatric HCP	0.279	**<0.001**
TSS should correlate positively with higher VAS transfer readiness scored by adult health care providers	VAS transfer readiness adult HCP	0.584	**<0.001**
TSS should correlate positively with higher level of self- management	TRAQ-NL (5)	0.444	**<0.001**
TSS should correlate positively with higher level of disease knowledge	RTT (6)	0.308	**<0.001**
TSS should correlate positively with higher level of quality of life	IBDQ (18)	0.242	**0.004**

HCP: health care providers, TRAQ: Transition Readiness Assessment Questionnaire, QoL: Quality of Life, RTT: Rotterdam Transition Test, TSS: Transition Success Score, VAS: Visual Analogue Scores, YA: Young Adult, ***Significant difference; p=<0.005***

Predictors of successful transition

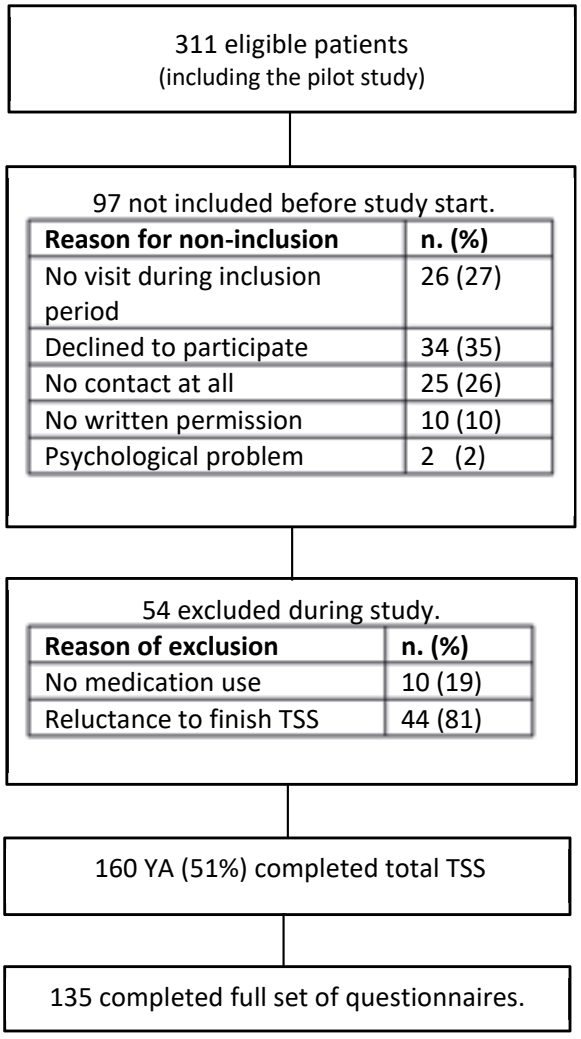
Associations between various predictor variables and total TSS scores were assessed using univariable and multivariable analyses. Due to low response frequencies, the estimation of the model's coefficients was not stable, the questions concerning disease activity (PGA), disease burden and acceptance of disease, all rated on a 4-point scale, were merged into two categories (present versus absent). An analysis of variance was carried out to explore the relationship between education level and TSS score as well as family composition and TSS score. The dependent variable was TSS, while the categorical predictor was either education level or number of siblings. For comparing Q-patient profiles, the Fisher-Freeman-Halton exact test was used due to the nominal nature and low frequencies in some profiles.

Cut-off scores

To enhance the practicality of the TSS in routine clinical care, cut-off scores need to be determined. In the absence of a gold standard, these scores were determined by an expert panel. Most importantly, the cut-off scores should enable identification the YAs who had a problematic transition and transfer of care, and who are

in most need of individualised transitional care. In light of the aforementioned premises and the previously described definition of transition success, along with the questions posed by the TSS, the following score distribution was established: Transition was successful with total TSS scores ≥ 25, moderately successful with scores of 21-24, and scores ≤ 20 were indicative for unsuccessful transition.

Figure 2: Inclusion procedure TSS validation study



TSS: Transition Success Score, YA: Young Adult

RESULTS

Young adult patients

One hundred and sixty of the 311 IBD eligible patients (meeting the inclusion criteria) completed the TSS (response percentage 51%), of whom 49% were male and 56% had Crohn's disease. Mean age at diagnosis was 14 years (IQR 3.56). One hundred and thirty-five participants completed the full set of questionnaires (Figure 2). Table 4 shows the demographics of participating patients as well as patients who were not included. Reasons for not willing to participate are reported in Figure 2. A significantly higher proportion of non-included patients were male (p=0.02), patients with disease in clinical remission (p=0.04) and had a significantly lower transfer readiness score as reported by their paediatrician (p=0.005). Additionally, non-included patients were significantly (p=0.03) more often identified as *"Self-confident & Autonomous"*. After transition and transfer, adult healthcare providers rated the YAs more often as *"self-confident and autonomous*" (n=106, 80.3%) compared to the paediatricians before the transition (n=90, 68.2%).

Table 4: Demographics of included (n=160) and excluded patients (n=151)

	N.(%) or Mean **Inclusion patients**	N. (%) or Mean (SD) **Exclusion patients**	p-value
Sex			
Male, %	78 (49)	94 (61)	**0.02**
IBD diagnosis			ns
Crohn's disease	89 (55.6)	87 (56.6)	
Ulcerative colitis	66 (41.3)	61 (39.6)	
IBD-U	5 (3.1)	6 (3.9)	
Disease activity at moment of transfer			
Remission	96 (60)	107 (71.3)	**0.04**
Q-Profile YA (as reported by paediatric healthcare provider)			Versus profile '*Conscious & Compliant'*
'Conscious & Compliant'	90 (68.2)	68 (49.6)	
'Backseat patient'	14 (10.6)	25 (18.2)	ns
'Self-confident & Autonomous'	15 (11.4)	30 (21.9)	**0.03**
'Worried & Insecure'	13 (9.8)	14 (10.2)	ns
VAS transfer readiness (as reported by paediatric healthcare provider)	Mean 82.95 (12.59) IQR 15	Mean 78,2 (13.3) IQR 20	**0.005**

IBD: inflammatory bowel disease, IQR: interquartile range, SD; standard Deviation, ns: not significant, r: Pearson correlation, VAS: Visual Analogue Scores, YA: Young Adult.

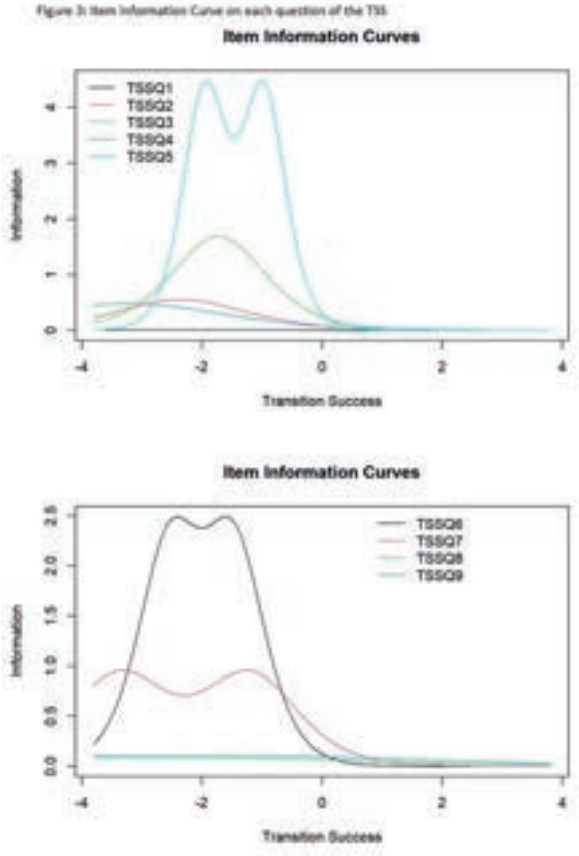
Significantly difference; p=<0.05

Validation of TSS

Structural validation of the TSS

Figure 3 depicts the results of the Rasch model per question, which ranges from -4 to 4. The figure illustrates that the TSS is most effective in discerning between low (-4) and moderate (0) levels of transition success. This implies that the TSS can best identify patients who did not have a successful transition. Questions with the lowest discriminative value were those on *"attended first appointment"* (TSSQ1) and *"satisfaction scored by patient and the caregiver"* (TSSQ8 and TSSQ9).

Figure 3: Item information curves



Test Information Function showing per question how much information it provides (x-axis) at the transition success level (y-axis). -4 totally failed transition to 4 perfectly successful transition
TSS: Transition Success Score, Q; Question

Construct validity with hypothesis testing
All questionnaires pertaining to transition readiness and self-management skills, including TRAQ-NL, RTT and QoL, correlated significantly, positively with the TSS (Table 3). Transfer readiness as scored by the adult healthcare providers using VAS had highest correlation, albeit still relatively weak (r 0.584; p<0.001). The most robust relationship was observed between the TSS and a questionnaire was self-management skills assessed by TRAQ-NL (r 0.444; p<0.001).

Reliability of the TSS
Cronbach's alpha for internal consistency of the TSS was 0.64, demonstrating acceptable reliability of the total TSS.

Outcome and predictors of the TSS

The mean TSS score was 25 (score range 17-27), with a quarter of YA patients achieving the maximum score. Table 2 indicates that 92% of the participants achieved the maximum score on question 4 about recalling medication. Of the caregivers, 53% scored a maximum VAS score (of 10) for their satisfaction concerning the transition process. Caregivers and YAs were satisfied with the transition care program (mean 7.4; SD. 1.5 vs. 7.8; SD. 1.5).

Cut-off scores
Transition was successful (total TSS-score ≥ 25) in 112 patients (70%). Transition success was moderate in 42 patients (26.2%) who had a total TSS score between 21-24. Transition was unsuccessful (TSS ≤ 20) in 6 patients (3.8%).

Predictors of success of transition
The results of the univariable analysis indicated that a higher disease activity (as assessed with PGA) and perceived disease burden (as reported by patients) at the time of TSS completion, along with experiencing disease exacerbation after transfer, were associated with lower TSS scores (Table 5). Additionally, patients categorized in the *"Backseat"* group (n=7, 5.3%; mean 20.86) and *"Worried & Insecure"* group (n=6,.5%; mean: 22.83) demonstrated significantly (p=<0.001) lower TSS scores compared to patients in the "*Conscious & Compliant"* group (n=106, 80.3%; mean 25.45). No significant relationships were found between TSS scores and surgery, time between transfer and first adult care appointment, country of birth, divorced caregivers, independent living, or transferring to an external hospital.

Table 5: Patient demographics in relation with total score TSS

		Relation with the TSS		
	Inclusion patients N (%) or Mean (SD)	TSS mean (SD)	Pearson correlation	p-value
Sex				
Male, %	78 (49)	Male; 24.9 (2.2) Female; 25.2 SD. 1.9		0.43
IBD diagnosis				
Crohn's disease	89 (55.6)	25.0 SD. 2.0		0.91
Ulcerative colitis	66 (41.3)	25.1 SD. 2.1		
IBD-U	5 (3.1)	24.8 SD. 1.8		
Age at filling in TSS (years)	Mean 19.1 (0.3) IQR 0.42		-0.076	0.35
Age at diagnosis (years)	Mean 14.0 (3.2) IQR 3.56		0.008	0.92
Age at transfer (years)	Mean 18.0 (0.3) IQR 0.17		-0.097	0.22
Educational level (n=150)				0.09
Low (lower secondary)	13 (8.7)	24.31 (2.2)		
Medium (upper secondary)	92 (61.3)	24.93 (2.0)		
High (pre-university)	45 (30)	25.48 (1.7)		
Duration (days) between transfer and completing TSS (n=154)	Mean 369.8 (82.1) IOR 106.5		-0.007	0.94
Disease activity (PGA) at moment of transfer		Remission 25.2 (2.0) Disease activity 24.8 (2.0)		0.268
Remission (PGA; Normal)	96 (60)			
Disease activity (PGA) at moment of filling in the TSS (n=132)		Remission 25.2 (1.8) Disease activity 24.2 (2.3)		**0.02**
Remission (PGA; Normal)	97 (73.4)			
Disease exacerbation after transfer (n=132)		Yes; 24.4 (2.0) No; 25.3 (1.9)		**0.02**
Yes	37 (28)			
Disease burden YA at moment of filling in TSS (n=152)		no complains; 25.7 (1.8) disease burden; 24.6 (2.1)		**<0.001**
Disease burden	97 (63.8)			
Q-Profile YA by paediatric HCP				0.151
'Conscious & Compliant'	90 (68.2)	25.3 (1.7)		
'Backseat patient'	14 (10.6)	24.2 (2.5)		
'Self-confident & Autonomous'	15 (11.4)	24.5 (2.7)		
'Worried & Insecure'	13 (9.8)	24.6 (1.9)		
Q-Profile YA as scored by adult HCP (n=132)				
'Conscious & Compliant'	106 (80.3)	25.5 (1.5)		
'Backseat patient'	7 (5.3)	20.9 (3.5)		
'Self-confident & Autonomous'	13 (9.8)	24.7 (1.2)		
'Worried & Insecure'	6 (4.5)	22.8 (2.0)		
Family composition (n=152)				
No siblings	8 (5.3)	24.8 (3.0)		
1-2 siblings	117 (77)	25.1 (2.1)		
3-4 siblings	22 (14.5)	24.5 (1.7)		
More than 4 siblings	5 (3.3)	26.0 (0.7)		
Acceptance of disease (n=111)				0.215
Yes, always or often	91 (82)	Yes; 25.2 (1.9) No; 24.4 (2.7)		

IQR: interquartile range, PGA: Physician Global Assessment, SD; standard Deviation, TSS: Transition Success Score, YA: Young Adult, ***Significantly difference; p=<0.05***

DISCUSSION

In this multicentre validation study conducted in the Netherlands we developed and validated the TSS, the first instrument for assessing the effectiveness of transition in YA IBD patients. We have demonstrated that the TSS is a reliable and valid tool to measure transition success. YAs who participated in the study achieved high scores, suggesting a high level of successful transition in this population, with a mean score of 25 (range 17-27) and a quarter of the participants even achieving maximal scores (27). Specific patient profiles such as *"Backseat"* and *"Worried & Insecure"* as well as disease activity and burden at the time of TSS completion and exacerbation after transfer, were identified as predictors of unsuccessful transition.

Discussion of the development of the TSS

Our previous Delphi study revealed that stakeholders valued generic outcomes such as self-management over disease-specific characteristics [16]. Consequently, the TSS was developed as a generic tool, not tied to any specific disease. The TSS items are highly similar to the items identified as relevant in Delphi studies on transition outcomes in other diseases, which identified key items related to independence, adherence, appearance in adult care, and satisfaction [24,25]. In a patient-based study, 518 YAs with different chronic diseases identified 5 indicators of transition success. These 5 indicators (patient not lost to follow-up, attending scheduled visits in adult care, patient building a trusting relationship with adult provider, continuing attention for self-management, patient satisfaction with transfer of care) are all included in the TSS in some way [17]. For diseases such as haemophilia [24,26] and diabetes [27], disease-specific items that monitor disease control such as annual bleeding rate or HbA1c have been suggested to reflect transition success. In IBD, faecal calprotectin is a validated marker of disease activity, but not associated with transition success. A Delphi study on sickle cell disease prioritized general items such as Quality of life (QoL) [28]. Our Delphi group decided to not include QoL in the TSS, but we validated the TSS using QoL as a measure and found a significant correlation between a higher TSS score and better QoL ($r=0.242$, $p=0.004$).

Questionnaires have been developed that advocate measuring success of transition for YAs with diabetes [29] and another for crossover youth [30] by measuring their self-management skills across various domains to assess transition success. These questionnaires assume that failure to master self-management skills means that the transition was not successful [30]. The mastery of pre-transfer skills, such as those measured by the TRAQ, can be compared with these questionnaires [29].

In consideration of the aforementioned factors, success of transition measured by the TSS is defined as: The transition to adult care is a success when the young adult patient is able to manage their illness independently and knows how to navigate the adult care system with satisfaction. To the best of our knowledge, the TSS is the first instrument that quantitatively measures success of transition.

Discussion of the validation of the TSS

As is typical of a well-designed questionnaire, the questions from the TSS elicit information at varying levels of knowledge. Questions 1, 8, and 9 provide minimal information, yet the study group, supported by literature[15,16,19,20,31], deemed them to be of such relevance, based on the holistic view of transition (satisfaction YA and their caregivers), that they were retained in the TSS. Furthermore, the removal of these questions did not result in significantly altered values in the validation analyses conducted with Cronbach's alpha and Pearson's correlations. We demonstrated that the TSS effectively identifies YAs who did not have a successful transition (SD -4- 0; Rasch range -4- 4). This is of major importance for routine clinical use, as it can identify at risk patients who require heightened guidance and care, likely benefitting from a tailored transition program or intervention. Correlation analyses provided support for all hypotheses regarding construct validation, although correlations were predominantly low or moderate, suggesting that the success of transition is influenced by multiple factors. Consistent with prior research findings, a holistic view of the transition period is crucial, with care strategies tailored to individual patients [1-3,9,31-33] Conversely, it can be argued that QoL, self-management and knowledge are influenced not only by the manner in which the transition occurs, but also by factors such as disease course and activity and specific patient characteristics, for example gender and age [5,6,21].A notable distinction between pediatric and adult healthcare lies in the shift of responsibility from caregivers to the patient in the management of their disease and treatment regimen. As such, the development of self-management skills such as taking medication is important during a transition program [2,9,32]. Notably, the correlation between the TSS and the self-management tool TRAQ-NL emerged as the strongest, underscoring the importance of enhancing self-management skills within structured transition programs.

Discussion of the patient outcomes

Lower TSS scores were found in patients characterized as *"Backseat patients",* who prefer to be guided and cared for by their caregivers and often show less interest in managing their illness [22]. Such coping behaviour can diminish independence and impact transition success negatively. Additionally, patients experiencing greater disease burden and more disease exacerbations after transfer exhibited sig-

nificantly lower TSS scores. This may reflect the consequences of an unsuccessful transition. Or, vice versa, active disease during transfer has been reported to lead to a less successful transition[15].

Strengths and Limitations

A strength of this study is the multicentre prospective design and the robust development of a novel quantitative tool that evaluates effectiveness of transitional care. All validation steps have been carried out according to the COSMIN methodology with a sufficient number of questionnaires that enables us to draw clear conclusions. Nevertheless, some limitations of this study should be mentioned. Firstly, the absence of a gold standard to measure the success of transition complicates validation of the TSS and necessitated the use of measurement tools that appear to be scientifically related to the skills needed for successful transition; self-management, knowledge and Qol. This underscores, the critical need for establishing such a standard to accurately measure success. Second, selection bias may apply since all included patients scored high on the TSS. This might be the consequence of the high quality of transitional IBD care in the Netherlands, even though it is not uniform between hospitals. Four hospitals (representing 85% of the included patients) had established multidisciplinary transition clinics or protocols. Alternatively, a considerable proportion of eligible YA (49%) could not be included into the study, with 26% being unable to be contacted and 35% declining to participate (Figure 2). This group of patients differed significantly from those who did participate, primarily comprising patients with less severe disease activity at the time of transfer, often categorized by pediatricians as *"Self-confident & Autonomous"* for whom transition may have been less successful. Therefore, TSS scores from this study might not be representative for the IBD population at large. Further research is required to gain a deeper understanding of the cut-off scores. It is crucial to have a representative sample of the entire IBD patient population, ideally from multiple countries. One potential approach is to obtain consent from patients to participate in the study at an early stage, before the transfer. This way, those patients who will potentially drop out of care later (lost to follow-up), thus having unsuccessful transition, will still be part of the study sample. Third, VAS scores from both patients and caregivers measuring transition success might be inflated since the survey has been conducted by their own healthcare team. To mitigate this effect, patients and caregivers were asked to fill in the questionnaire online.

CONCLUSIONS

In this study, we developed the first validated quantitative measure of healthcare transition outcomes for YA with IBD. The TSS allows healthcare professionals in adult care to identify YA in whom transfer of care was unsuccessful in order to provide additional tailored transitional care in response. Moreover, the TSS aids in generating risk profiles to pinpoint patients at risk of transition failure and assists in discerning effective interventions or transition programs.

As this TSS is non-disease specific, validation in cohorts of patients with other chronic diseases would be insightful after TSS validation in these disorders as well. We encourage all healthcare providers involved in IBD transitional care to adopt the TSS to monitor in order to evaluate and improve their transition programs.

REFERENCES

1. van Rheenen PF, Aloi M, Biron IA*, et al.* European crohn's and colitis organisation topical review on transitional care in inflammatory bowel disease. *J Crohns Colitis* 2017;**11**:1032-8.
2. Brooks AJ, Smith PJ, Cohen R*, et al.* Uk guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. *Gut* 2017;**66**:988-1000.
3. Philpott JR, Kurowski JA. Challenges in transitional care in inflammatory bowel disease: A review of the current literature in transition readiness and outcomes. *Inflamm Bowel Dis* 2019;**25**:45-55.
4. Hait EJ, Barendse RM, Arnold JH*, et al.* Transition of adolescents with inflammatory bowel disease from pediatric to adult care: A survey of adult gastroenterologists. *J Pediatr Gastroenterol Nutr* 2009;**48**:61-5.
5. van Gaalen MAC, van Gijn E, van Pieterson M*, et al.* Validation and reference scores of the transition readiness assessment questionnaire in adolescent and young adult ibd patients. *J Pediatr Gastroenterol Nutr* 2023.
6. van Gaalen MAC, van Pieterson M, van den Brink G*, et al.* Rotterdam transition test: A valid tool for monitoring disease knowledge in adolescents with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2022;**74**:60-7.
7. Cole R, Ashok D, Razack A, Azaz A, Sebastian S. Evaluation of outcomes in adolescent inflammatory bowel disease patients following transfer from pediatric to adult health care services: Case for transition. *J Adolesc Health* 2015;**57**:212-7.
8. Fishman LN, Ding J. Optimizing the transition and transfer of care in pediatric inflammatory bowel disease. *Gastroenterol Clin North Am* 2023;**52**:629-44.
9. Kim J, Ye BD. Successful transition from pediatric to adult care in inflammatory bowel disease: What is the key? *Pediatr Gastroenterol Hepatol Nutr* 2019;**22**:28-40.
10. Crowley R, Wolfe I, Lock K, McKee M. Improving the transition between paediatric and adult healthcare: A systematic review. *Arch Dis Child* 2011;**96**:548-53.
11. Coyne B, Hallowell SC, Thompson M. Measurable outcomes after transfer from pediatric to adult providers in youth with chronic illness. *J Adolesc Health* 2017;**60**:3-16.
12. Cleverley K, Rowland E, Bennett K, Jeffs L, Gore D. Identifying core components and indicators of successful transitions from child to adult mental health services: A scoping review. *Eur Child Adolesc Psychiatry* 2020;**29**:107-21.
13. Pearlstein H, Bricker J, Michel HK*, et al.* Predicting sub-optimal transitions in adolescents with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2020.
14. van Staa A, Sattoe JN. Young adults' experiences and satisfaction with the transfer of care. *J Adolesc Health* 2014;**55**:796-803.
15. van den Brink G, van Gaalen MAC, Zijlstra M*, et al.* Self-efficacy did not predict the outcome of the transition to adult care in adolescents with inflammatory bowel disease. *Acta Paediatr* 2019;**108**:333-8.
16. van den Brink G, van Gaalen MAC, de Ridder L, van der Woude CJ, Escher JC. Health care transition outcomes in inflammatory bowel disease: A multinational delphi study. *J Crohns Colitis* 2019;**13**:1163-72.
17. Sattoe JNT, Hilberink SR, van Staa A. How to define successful transition? An exploration of consensus indicators and outcomes in young adults with chronic conditions. *Child Care Health Dev* 2017;**43**:768-73.
18. Mokkink LB, Terwee CB, Patrick DL*, et al.* The cosmin checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: An international delphi study. *Qual Life Res* 2010;**19**:539-49.
19. Erős A, Soós A, Hegyi P*, et al.* Spotlight on transition in patients with inflammatory bowel disease: A systematic review. *Inflamm Bowel Dis* 2020;**26**:331-46.
20. Sattoe JNT, Peeters MAC, Haitsma J*, et al.* Value of an outpatient transition clinic for young people with inflammatory bowel disease: A mixed-methods evaluation. *BMJ Open* 2020;**10**:e033535.
21. Russel MG, Pastoor CJ, Brandon S*, et al.* Validation of the dutch translation of the inflammatory bowel disease questionnaire (ibdq): A health-related quality of life questionnaire in inflammatory bowel disease. *Digestion* 1997;**58**:282-8.

22. Jedeloo S, van Staa A, Latour JM, van Exel NJ. Preferences for health care and self-management among dutch adolescents with chronic conditions: A q-methodological investigation. *Int J Nurs Stud* 2010;**47**:593-603.
23. Baker F. *The basics of item response therapy.* University of Maryland, College Park: ERIC Clearinghouse on Assessment and Evaluation; 2001.
24. Suris JC, Akre C. Key elements for, and indicators of, a successful transition: An international delphi study. *J Adolesc Health* 2015;**56**:612-8.
25. Fair C, Cuttance J, Sharma N*, et al.* International and interdisciplinary identification of health care transition outcomes. *JAMA Pediatr* 2016;**170**:205-11.
26. Sun HL, Breakey VR, Straatman L, Wu JK, Jackson S. Outcomes indicators and processes in transitional care in adolescents with haemophilia: A delphi survey of canadian haemophilia care providers. *Haemophilia* 2019;**25**:296-305.
27. Pierce J, Aroian K, Schifano E, Gannon A, Wysocki T. Development and content validation of the healthcare transition outcomes inventory for young adults with type 1 diabetes. *J Patient Rep Outcomes* 2019;**3**:71.
28. Sobota AE, Shah N, Mack JW. Development of quality indicators for transition from pediatric to adult care in sickle cell disease: A modified delphi survey of adult providers. *Pediatr Blood Cancer* 2017;**64**.
29. Pierce J, Hossain J, Gannon A. Validation of the healthcare transition outcomes inventory for young adults with type 1 diabetes. *J Pediatr Psychol* 2020;**45**:767-79.
30. Dubov V, Agnihotri S, Goodman D, Patel M. Development of the successful transitions assessment tool (stat) for crossover youth (coy): Youth voice and expert feedback with a delphi approach. *Child Abuse Negl* 2022;**134**:105900.
31. Bihari A, Olayinka L, Kroeker KI. Outcomes in patients with inflammatory bowel disease transitioning from pediatric to adult care: A scoping review. *J Pediatr Gastroenterol Nutr* 2022;**75**:423-30.
32. Goodhand J, Hedin CR, Croft NM, Lindsay JO. Adolescents with ibd: The importance of structured transition care. *J Crohns Colitis* 2011;**5**:509-19.
33. Paine CW, Stollon NB, Lucas MS*, et al.* Barriers and facilitators to successful transition from pediatric to adult inflammatory bowel disease care from the perspectives of providers. *Inflamm Bowel Dis* 2014;**20**:2083-91.

SUPPLEMENTAL DATA

Supplemental table 1: Demographic characteristics of the expert panel (n=60)

		N (%)
Sex	Male, %	24 (40)
Hospital type	Non-academic hospital	5 (8.3)
	Academic hospital	55 (91.7)
Department	Paediatrics	36 (60)
	Gastroenterology	18 (30)
	Internal Medicine	6 (10
Position	Paediatric Gastroenterologist	25 (41.7)
	Paediatric IBD Nurse	9 (15)
	Gastroenterologist	19 (31.7)
	Adult IBD Nurse	5 (8.3)
	Clinical research fellow	1 (1.7)
	Transition manager	1 (1.7)
Continent	Europe	48 (80)
	North America	6 (10)
	Asia	6 (10)

C

Part C

General Discussion and Summary


8

Chapter 8

General Discussion

Adolescence is a turbulent phase in which young people search their ways to find their identity. The phase is made even more challenging if you also have a chronic illness such as IBD. Besides adolescence and dealing with all the consequences of chronic illness, the transition is complicated by the differences between pediatric and adult care. To make this phase to independent adults as smooth as possible, a team effort, collaboration between pediatric, adult care, patients and caregivers, is crucial.

The overall aim of this thesis was how healthcare providers can best guide adolescent and young adult IBD patients during the transition period to adult healthcare. The objective was to optimize the tools used for monitoring of skills building during transition (Part A), and to establish a valid method to measure success (or effectiveness) of the transition process (Part B).

To enhance the quality of transitional care, we have developed measurement tools for the different facets of transition (see Table 1)
This final chapter compares the main findings of this thesis with the current literature, including my personal perspective and, most importantly, potential future applications in clinical practice.

Table 1: overview of developed measurement tools

Measurement tool	**What it measures**	**Chapter**
Rotterdam Transition Test (RTT)	IBD related knowledge - Open-ended questionnaire - Correction model	Chapter 2
Transition Readiness Assessment Questionnaire; Dutch version (TRAQ-NL)	Self-management - Translation of TRAQ[1] - Reference scores for IBD patients	Chapter 3
Transition Yourself	Composite score of success of transition - Based on the opinion of pediatric IBD experts	Chapter 5
Transition Success Score (TSS)	Success of Transition - Based on the Top 10 item list generated by international Delphi procedure (with pediatric and adult IBD experts, and patients)	Chapter 6 & 7

Part A: Developing tools to support the transition process.

Adult healthcare is different from what IBD patients coming from paediatrics are used to. It is provided by a different team of healthcare professionals in a different part of the hospital, or even in another hospital. In addition, adult healthcare is structured differently, driven by the high number of patients and adapted to adult behaviour. This means that the young adult is expected to behave in a different way in adult healthcare compared to pediatric healthcare. In the topical review on transition by the European Crohn's Colitis Organization (ECCO), it is schematically presented that the adolescent's disease-related knowledge and self- management level increase during this period in order to learn how to navigate in adult healthcare.[2]

Adolescents[3-6], but also adults[7], may have insufficient disease-related knowledge, whereas good IBD knowledge has been associated with a number of benefits, including improved medication adherence[8-10], coping[11], disease outcomes and even reduced costs.[12] For this reason, several questionnaires have been developed to measure patients' knowledge of IBD, including those in transition. All these tools were designed as multiple-choice questionnaires. [4,13-20] This can be a problem, as adolescents will sometimes guess the answer, with a chance of 25% of getting it right (in 4 multiple choice options). For this reason, we developed an open-ended questionnaire with a response model to measure the patient's actual knowledge; the Rotterdam Transition Test (RTT), described and studied in **Chapter 2.** The RTT, perhaps because it is an open-ended questionnaire, is not the most obvious knowledge questionnaire to use in a research setting. While more than substantial agreement (K>0.61) was achieved for all questions after several adjustments, inter-rater reliability still depends on how the sometimes-vague answers are interpreted and scored by the assessor. Still, for guidance in daily practice, the open-ended nature of the RTT certainly has specific value. For example, the strangest answers to the questions will provide good insight into what the adolescent truly knows. This makes it easy to start a conversation with the adolescent, even about more sensitive issues such as smoking, alcohol and drugs, or sexuality and pregnancy.[21] The RTT study showed that girls, as well as patients of older age, with higher level of education and acceptance of the disease had significantly higher levels of knowledge. It should be noted that this does not apply to all 17-year-old girls pursuing higher education. Additionally, while some may perform well on the entire RTT, they may still lack knowledge in certain areas. Altogether, the RTT is a valuable tool that can tailor individual care. If a patient knows all the answers, there is no need to discuss the items at length and there is room to focus on the patient's individual needs. Patient questionnaires pose a significant challenge in both practice and scientific research: a substantial number of adolescents regard

the completion of questionnaires as an onerous task, primarily due to the time commitment required in the context of their already demanding schedules.[22] This is certainly true of the RTT, which can take up to 15 minutes to complete properly, due to the open-ended nature of the 25 questions for boys and 27 questions for girls. In the future, a shortened (but still complete) version of the RTT needs to be developed.

Self-management of the disease was another important skill to consider during the transition process.[2] Knowledge and self-management skills are connected. A higher level of knowledge can lead to a higher level of self-management. The adolescent can turn disease knowledge into the action of self-management. One is not possible without the other, so it is important to pay attention to both skills. A lack of either can also lead to poorer adherence to treatment.[9,23] In IBD, non-adherence can lead to disease exacerbation. **Chapter 3** describes the translation into Dutch and validation of a widely used generic self-management questionnaire, the Transition Readiness Assessment Questionnaire (TRAQ)[1]. The TRAQ is an easy and quick to complete questionnaire that gives healthcare providers insight into the patient's independence in five different areas of self-management. For daily practice, it is very important that a questionnaire is useful and that the individual patient benefits from the questionnaire.[24] That is why we developed age-specific reference scores. With these scores, healthcare providers can show patients in the consultation room where they stand in relation to their peers and where they could possibly work harder or where they are doing well. The scores increase with age, showing higher level of independence when patients become older, just as disease-related knowledge measured by the RTT increases with age (Chapter 2). It seems reasonable to suggest that this correlation may be attributable to the fact that, at a certain age, the patient has undergone the intense period of adolescence and is therefore better able to cope with whatever may arise in the context of living with IBD as a young adult.[25] With reference scores, the transition readiness of a patient can be compared with the rest of the cohort. The question remains: which score gives the best chance of a successful transition? There is a relation between a higher level of self-management scored by the TRAQ and the healthcare related outcome in the year after transfer.[26] It can be posited that the development of self-management skills is a crucial aspect of the transition process. In the concluding sections of this thesis, we undertake a more detailed examination of the utility of self-management as a metric for assessing the success of transitions.

A patient-reported outcome measure (PROM), such as the TRAQ, must not only be useful, but also easy to use for healthcare providers and patients. Digital applications can be used to properly integrate PROMs into the healthcare system.[27] In **Chapter 4**, we show how we introduced and implemented PROMs for anxiety and depression in our IBD patients in the electronic patient record. Here, we were particularly interested in how the patients experienced the use of PROMs as part of their care. The message from the patients was that they were satisfied but would like to see the questionnaire less often (not every 3 months as offered in the study). It is a difficult balance between getting the necessary information and burdening the patient. We, and others before us, learned from this study that it is important that the usefulness of the questionnaire is clear to the patient, that the results are handled by the healthcare providers, shared with the patients, and that the healthcare providers are aware that a patient has more to do in life.[28,29]

Part B: The quest of making success of transition measurable

As healthcare providers in pediatric care, we need to prepare adolescents with IBD as well as possible for the differences and expectations of adult healthcare. Several structured transition programs exist, but it is unclear which ones are most effective in facilitating a successful transition. [2,30,31] In addition to that there is still no clear definition of transition success, and many terms such as readiness and transition success are used interchangeably.[32-34] For this reason, the expert group in Chapter 7 define success of transition as follows: *'The transition to adult healthcare is a success when the young adult patient is able to manage their illness independently and knows how to navigate the adult healthcare system with satisfaction'.* The focus is on the young adult's skills in managing their illness independently, while the most important thing is that he knows where to go with a question or problem in adult health care. This does not mean that all patients are completely 'ready' at the time of transfer.[35] Our quest to measure transition success has similarities to the transition process of IBD patient. This process is also one of trial and error, providing the opportunity to learn and improve ourselves to achieve a favourable outcome. A first attempt to measure transition success was made in **Chapter 5**. In 2008, our research group developed a questionnaire measuring self-efficacy.[36] Self-efficacy, a person's belief in his ability to organize and perform the actions necessary to deal with future situations, is considered a prerequisite for self-management. [37,38] Self-efficacy was thought to be important for successful transition, in addition to self-management and disease-related knowledge.[39,40] Based on the available literature and the outcomes of a focus group with IBD experts, a score was developed (Transition-Yourself) consisting of the following items: attendance at the adult gastroenterology outpatient clinic, adherence to therapy and rating of the quality of his transition process by the

adolescent.[41] This composite score showed in a group of 35 IBD patients that 63% had a successful transition, 31% a moderate successful transition and 6% had an unsuccessful transition. Transitional care has changed over the years. At the time of this study (2014), transition care consisted of outpatient appointments at the adult care site, multidisciplinary meetings with pediatric and adult care, attention to disease knowledge by pediatric healthcare providers, and both oral and written handovers between healthcare providers. No relationship was found between self-efficacy, as measured by the IBD-Yourself, and the success of the transition. The lesson we learned from conducting this study is that ascertainment of reliability and validity of a measurement tool (such as Transition-Yourself composite score) is of utmost importance too; this was not done and therefore it is unclear whether the Transition-Yourself is an accurate reflection of the construct it is designed to assess.

Next, in order to properly measure the success of transition, we took the appropriate steps to develop a measurement tool in a scientifically valid approach. To achieve this, we performed a three-stage Delphi study in which we asked an international IBD expert panel (of 64 people: nurses, pediatric and adult gastroenterologists) and 61 IBD patients up (to the age of 25 years) what they considered to be the most important parameters of transition success (**Chapter 6**). The results of the expert panel and the patient group were very similar, with 8 items identified as important by both groups. It was noticeable that the selected items mainly focused on self-management skills and autonomy of the patient (e.g. independent communication, medication adherence), but also on quality of life and patient satisfaction with the transition process. Disease-related issues (exacerbation, inflammatory markers and surgery) were not considered important. Finally, in the Top 10, 'shows ability to make decisions regarding IBD' was seen as the most important item, ahead of 'independent communication with treating physician or nurse'. We found that the Top 10 items were broadly similar to those that have been found to be important for a successful transition in other chronic conditions.[41-46]

The Top 10 from the international Delphi study provided a good basis for the development of a new measurement tool with the same group of IBD experts from the Delphi study (**Chapter 7**). Using a consensus methodology over 4 rounds, the Top 10 were translated into a quantitative questionnaire of 9 questions with 3 response options per question: the Transition Success Score (TSS). 7 of the 9 questions are to be reported by the adult healthcare provider and concern the patient's behaviour in relation to their illness (e.g. contacting the hospital themselves, being involved in decision making). The last two questions are about

patient and caregiver satisfaction with the transition process. In order to validate the TSS questionnaire, it is important that the questionnaire measures what it is supposed to measure and that it is reliable.[24] We used the COSMIN method[47] to test whether the TSS was valid and reliable. After the lessons learned from the Transition-Yourself Score study, we conducted a multicentre study to ensure that bias in care delivery (e.g. the transition program or healthcare providers behaviour) would not affect the validation of the TSS. We also kept the time frame for completing the TSS after transfer shorter (maximum 16 months) than in the Transition-Yourself Score study. This was based on the experience that many young adults found it difficult to assess the transition process two years after transfer. As a result of all this, the TSS is construct and structurally valid with acceptable reliability and can certainly be used as a measure to identify young adults whose transition has not gone well. This is a great addition to the practice to further optimize transitional support. There is a significant correlation with the core transition skills[2]; knowledge (measured with the Rotterdam Transition Test (Chapter 2)) and independence (measured with the TRAQ (Chapter 3)), but also with quality of life (measured with the IBDQ)[48], but the correlations were not strong. This again suggests that transition in care and its success does not depend on one factor and that transition in care should be personalised.[2,30,49-53] To make the TSS usable in daily practice, we have developed cut-off scores based on expert opinion in the absence of a gold standard. These scores need further validation.

We also looked into the type of patients that scored lower on the TSS and therefore had a less successful transition. The TSS score was lower in patients described as 'backseat patients', who prefer to be guided and cared for by their caregivers and often have less interest in managing their illness.[54] Such coping behaviour can reduce independence and negatively impact on transition success. However, patients who experienced high disease burden or exacerbation after transfer also had lower TSS scores. It is unclear whether disease burden and disease activity is cause or consequence of a less successful transition. It may also be that disease in remission contributes to a successful transition, as we saw in the Transition-Yourself Score study.

Conducting research with adolescents is challenging, especially when they are asked to spend time on completing questionnaires, from which they do not directly benefit. This was a recurring theme in each study. This sometimes led to a bias towards well-behaved patients in the study, which certainly influenced the results.[22,55-57] In the studies measuring success of transition, it is conceivable that young adults who did not give consent, or who could not be reached for consent, were the ones with less successful transition. For follow-up studies, it may be

helpful to ask for consent before transfer so that there is at least permission to use the data from adult healthcare providers if the patient is not reachable.

LOOKING INTO THE FUTURE

At the Erasmus MC-Sophia Children's Hospital, the Young Adult Program was launched in 2024. This generic structured transition program prepares every adolescent with a chronic illness (beyond IBD) and their caregivers for the transfer to adult healthcare according to a standard framework comparable with the core elements of the Dutch Quality standard "Adolescents in Transition from child to adult healthcare".[58] The 5 core elements of the Erasmus MC Young Adult Program are:

- having a transition coordinator
- individual transition plan
- introduction to a specialist or nurse from the adult healthcare team around the age of 17
- a warm handover of care at the time of transfer
- at the first appointment in adult care, a longer consultation for a more in-depth introduction.

The individual transition plan is developed by the transition coordinator together with the patient and caregivers, using PROMs such as the TRAQ-NL (Chapter 3) and the 'Ready Steady Go' program.[59] This plan focusses on what the patient is doing in relation to the illness, goals that need work and guidance on their caregivers needs. The patient takes the lead and is guided by a transition coordinator and everything is evaluated and adjusted every year, or more often if they want or need it. In a warm handover of care consultation, everyone involved (patient, caregivers, healthcare providers, pediatric and adult services) sits together to transfer care as completely as possible to adult services.

My expectation is that the Young Adult Program will be a success for a large group of patients. There is however a certain type of patient, e.g. the 'backseat patient', who does not care that much and who remains very attached to their caregivers. And there will be caregivers who still find it difficult to give their adolescent the space to develop their skills.

With further research, we can look more closely at patient and caregivers' characteristics that might be associated with a higher risk of a less successful transition. There is also a healthcare provider barrier: doctors and nurses must make time

and continue to have the motivation to read and interpret the questionnaires as well as share and discuss the results with the patients, so that the patient knows that filling in a questionnaire is important.

Standard health insurance reimbursement for transitional care, for a nurse transition coordinator, will make it easier to provide that care. Hopefully, with the advent of the TSS, it will now be possible to demonstrate that a program such as the Young Adult Program makes a difference, and patients, healthcare providers and health insurers will be motivated to do their bit. This will facilitate the implementation of more appropriate and flexible transitional care, thereby reducing the probability that IBD patients are lost to follow-up and subsequently present with an exacerbation or complication of an untreated disease in the emergency room. Ultimately, this approach will result in more trusting caregivers and more satisfied patients who are able to navigate their own way to adult healthcare, even in situations where the transition process is difficult during the turbulent adolescent phase.

Remaining questions

In light of the preceding discussion, a number of questions remain unresolved. Hopefully, further research will answer these questions.

- There are many PROMs that provide useful information about the adolescent IBD patient and at what point the quality of care can be improved. But where is the balance so that we do not overburden the patient?
- Given that both knowledge and self-management are increasing with age, does this mean that transition will be more successful when patients simply transfer at a later age (e.g. 25 years)?
- The Transition-Yourself is shorter than the TSS and possibly even easier to complete, but is it also a reliable and valid tool?
- What is the role of disease activity and burden of disease on the success of transition? Is this cause or consequence of a less successful transition? Or should we focus on the fact that transition is more likely to be successful when the disease is in remission?
- How can we motivate 'backseat patients' to actively participate in their transition process, for example by filling in the questionnaires?

REFERENCES

1. Wood DL, Sawicki GS, Miller MD*, et al.* The transition readiness assessment questionnaire (traq): Its factor structure, reliability, and validity. *Acad Pediatr* 2014;**14**:415-22.
2. van Rheenen PF, Aloi M, Biron IA*, et al.* European crohn's and colitis organisation topical review on transitional care in inflammatory bowel disease. *J Crohns Colitis* 2017;**11**:1032-8.
3. Fishman LN, Barendse RM, Hait E, Burdick C, Arnold J. Self-management of older adolescents with inflammatory bowel disease: A pilot study of behavior and knowledge as prelude to transition. *Clin Pediatr (Phila)* 2010;**49**:1129-33.
4. Benchimol EI, Walters TD, Kaufman M*, et al.* Assessment of knowledge in adolescents with inflammatory bowel disease using a novel transition tool. *Inflamm Bowel Dis* 2011;**17**:1131-7.
5. Day AS, Mylvaganam G, Shalloo N*, et al.* Assessment of disease-specific knowledge in australian children with inflammatory bowel disease and their parents. *J Paediatr Child Health* 2017;**53**:778-81.
6. Hait EJ, Barendse RM, Arnold JH*, et al.* Transition of adolescents with inflammatory bowel disease from pediatric to adult care: A survey of adult gastroenterologists. *J Pediatr Gastroenterol Nutr* 2009;**48**:61-5.
7. Buerkle KS, Vernon-Roberts A, Ho C, Schultz M, Day AS. A short knowledge assessment tool is valid and acceptable for adults with inflammatory bowel disease. *Dig Dis Sci* 2022;**67**:2049-58.
8. Cervený P, Bortlík M, Kubena A*, et al.* Nonadherence in inflammatory bowel disease: Results of factor analysis. *Inflamm Bowel Dis* 2007;**13**:1244-9.
9. Hommel KA, Greenley RN, Maddux MH, Gray WN, Mackner LM. Self-management in pediatric inflammatory bowel disease: A clinical report of the north american society for pediatric gastroenterology, hepatology, and nutrition. *J Pediatr Gastroenterol Nutr* 2013;**57**:250-7.
10. Lim JK, Lee YJ, Park JH. Medication-related knowledge and medication adherence in pediatric and adolescent patients with inflammatory bowel disease. *J Korean Med Sci* 2020;**35**:e92.
11. Moradkhani A, Kerwin L, Dudley-Brown S, Tabibian JH. Disease-specific knowledge, coping, and adherence in patients with inflammatory bowel disease. *Dig Dis Sci* 2011;**56**:2972-7.
12. Colombara F, Martinato M, Girardin G, Gregori D. Higher levels of knowledge reduce health care costs in patients with inflammatory bowel disease. *Inflamm Bowel Dis* 2015;**21**:615-22.
13. Haaland D, Day AS, Otley A. Development and validation of a pediatric ibd knowledge inventory device: The ibd-kid. *J Pediatr Gastroenterol Nutr* 2014;**58**:313-9.
14. Tung J, Grunow JE, Jacobs N. Pilot development of an electronic pediatric inflammatory bowel disease quiz game. *J Pediatr Gastroenterol Nutr* 2015;**61**:292-6.
15. Eaden JA, Abrams K, Mayberry JF. The crohn's and colitis knowledge score: A test for measuring patient knowledge in inflammatory bowel disease. *Am J Gastroenterol* 1999;**94**:3560-6.
16. Keegan D, McDermott E, Byrne K*, et al.* Development, validation and clinical assessment of a short questionnaire to assess disease-related knowledge in inflammatory bowel disease patients. *Scand J Gastroenterol* 2013;**48**:183-8.
17. Jones SC, Gallacher B, Lobo AJ, Axon AT. A patient knowledge questionnaire in inflammatory bowel disease. *J Clin Gastroenterol* 1993;**17**:21-4.
18. Casellas F, Navarro E, Amil P*, et al.* Development and validation of the quecomiicat questionnaire: A tool to assess disease-related knowledge in patients with inflammatory bowel disease. *Rev Esp Enferm Dig* 2019;**111**:586-92.
19. Yoon H, Yang SK, So H*, et al.* Development, validation, and application of a novel tool to measure disease-related knowledge in patients with inflammatory bowel disease. *Korean J Intern Med* 2019;**34**:81-9.
20. Danion P, Buisson A, Roblin X*, et al.* Ibd-info questionnaire: A multicenter french up-to-date survey of patient knowledge in inflammatory bowel disease. *Inflamm Bowel Dis* 2018;**24**:943-52.
21. Jenn NC. Designing a questionnaire. *Malays Fam Physician* 2006;**1**:32-5.
22. Bidonde J, Meneses-Echavez JF, Hafstad E, Brunborg GS, Bang L. Methods, strategies, and incentives to increase response to mental health surveys among adolescents: A systematic review. *BMC Med Res Methodol* 2023;**23**:270.
23. Vernon-Roberts A, Rouse E, Gearry RB, Day AS. Systematic review of self-management assessment tools for children with inflammatory bowel disease. *JPGN Rep* 2021;**2**:e075.

24. Boparai JK, Singh S, Kathuria P. How to design and validate a questionnaire: A guide. *Curr Clin Pharmacol* 2018;**13**:210-5.
25. Crone EA, Dahl RE. Understanding adolescence as a period of social-affective engagement and goal flexibility. *Nat Rev Neurosci* 2012;**13**:636-50.
26. Nardone OM, Martinelli M, de Sire R*, et al.* Time to grow up: Readiness associated with improved clinical outcomes in pediatric inflammatory bowel disease patients undergoing transition. *Therap Adv Gastroenterol* 2024;**17**:17562848241241234.
27. Huberts AS, Koppert LB, Benschop JAM*, et al.* Facilitators and barriers in the implementation and adoption of patient-reported outcomes measurements in daily practice. *Value Health* 2024;**27**:1235-42.
28. McCabe E, Rabi S, Bele S, Zwicker JD, Santana MJ. Factors affecting implementation of patient-reported outcome and experience measures in a pediatric health system. *J Patient Rep Outcomes* 2023;**7**:24.
29. Marshall G. The purpose, design and administration of a questionnaire for data collection. *Radiography* 2005;**11**:131-6.
30. Brooks AJ, Smith PJ, Cohen R*, et al.* Uk guideline on transition of adolescent and young persons with chronic digestive diseases from paediatric to adult care. *Gut* 2017;**66**:988-1000.
31. Chan P, McNamara J, Vernon-Roberts A*, et al.* Systematic review: Practices and programs in inflammatory bowel disease transition care. *Inflamm Bowel Dis* 2024.
32. Sattoe JNT, Hilberink SR, van Staa A. How to define successful transition? An exploration of consensus indicators and outcomes in young adults with chronic conditions. *Child Care Health Dev* 2017;**43**:768-73.
33. Cleverley K, Rowland E, Bennett K, Jeffs L, Gore D. Identifying core components and indicators of successful transitions from child to adult mental health services: A scoping review. *Eur Child Adolesc Psychiatry* 2020;**29**:107-21.
34. Coyne B, Hallowell SC, Thompson M. Measurable outcomes after transfer from pediatric to adult providers in youth with chronic illness. *J Adolesc Health* 2017;**60**:3-16.
35. Gumidyala AP, Greenley RN, Plevinsky JM*, et al.* Moving on: Transition readiness in adolescents and young adults with ibd. *Inflamm Bowel Dis* 2018;**24**:482-9.
36. Zijlstra M, De Bie C, Breij L*, et al.* Self-efficacy in adolescents with inflammatory bowel disease: A pilot study of the "ibd-yourself", a disease-specific questionnaire. *J Crohns Colitis* 2013;**7**:e375-85.
37. Escher JC. Transition from pediatric to adult health care in inflammatory bowel disease. *Dig Dis* 2009;**27**:382-6.
38. A. B. Self-efficacy in ramachaudran vs, editor. *Encyclopedia of human behavior New York, NY: Academic Press* 1994:71-81.
39. van Staa A, van der Stege HA, Jedeloo S, Moll HA, Hilberink SR. Readiness to transfer to adult care of adolescents with chronic conditions: Exploration of associated factors. *J Adolesc Health* 2011;**48**:295-302.
40. Carlsen K, Haddad N, Gordon J*, et al.* Self-efficacy and resilience are useful predictors of transition readiness scores in adolescents with inflammatory bowel diseases. *Inflamm Bowel Dis* 2017;**23**:341-6.
41. Suris JC, Akre C. Key elements for, and indicators of, a successful transition: An international delphi study. *J Adolesc Health* 2015;**56**:612-8.
42. Fair C, Cuttance J, Sharma N*, et al.* International and interdisciplinary identification of health care transition outcomes. *JAMA Pediatr* 2016;**170**:205-11.
43. Gleeson H, Davis J, Jones J, O'Shea E, Clayton PE. The challenge of delivering endocrine care and successful transition to adult services in adolescents with congenital adrenal hyperplasia: Experience in a single centre over 18 years. *Clin Endocrinol (Oxf)* 2013;**78**:23-8.
44. Jensen PT, Karnes J, Jones K*, et al.* Quantitative evaluation of a pediatric rheumatology transition program. *Pediatr Rheumatol Online J* 2015;**13**:17.
45. Hazel E, Zhang X, Duffy CM, Campillo S. High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. *Pediatr Rheumatol Online J* 2010;**8**:2.
46. Reid GJ, Irvine MJ, McCrindle BW*, et al.* Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics* 2004;**113**:e197-205.

47. Mokkink LB, Terwee CB, Patrick DL*, et al.* The cosmin checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: An international delphi study. *Qual Life Res* 2010;**19**:539-49.
48. Russel MG, Pastoor CJ, Brandon S*, et al.* Validation of the dutch translation of the inflammatory bowel disease questionnaire (ibdq): A health-related quality of life questionnaire in inflammatory bowel disease. *Digestion* 1997;**58**:282-8.
49. Philpott JR, Kurowski JA. Challenges in transitional care in inflammatory bowel disease: A review of the current literature in transition readiness and outcomes. *Inflamm Bowel Dis* 2019;**25**:45- 55.
50. Kim J, Ye BD. Successful transition from pediatric to adult care in inflammatory bowel disease: What is the key? *Pediatr Gastroenterol Hepatol Nutr* 2019;**22**:28-40.
51. Bihari A, Olayinka L, Kroeker KI. Outcomes in patients with inflammatory bowel disease transitioning from pediatric to adult care: A scoping review. *J Pediatr Gastroenterol Nutr* 2022;**75**:423- 30.
52. Goodhand J, Hedin CR, Croft NM, Lindsay JO. Adolescents with ibd: The importance of structured transition care. *J Crohns Colitis* 2011;**5**:509-19.
53. Paine CW, Stollon NB, Lucas MS*, et al.* Barriers and facilitators to successful transition from pediatric to adult inflammatory bowel disease care from the perspectives of providers. *Inflamm Bowel Dis* 2014;**20**:2083-91.
54. Jedeloo S, van Staa A, Latour JM, van Exel NJ. Preferences for health care and self- management among dutch adolescents with chronic conditions: A q-methodological investigation. *Int J Nurs Stud* 2010;**47**:593-603.
55. Bishop K. Challenging research: Completing participatory social research with children and adolescents in a hospital setting. *Herd* 2014;**7**:76-91.
56. Steinbeck K, Baur L, Cowell C, Pietrobelli A. Clinical research in adolescents: Challenges and opportunities using obesity as a model. *Int J Obes (Lond)* 2009;**33**:2-7.
57. Singh JA, Siddiqi M, Parameshwar P, Chandra-Mouli V. World health organization guidance on ethical considerations in planning and reviewing research studies on sexual and reproductive health in adolescents. *J Adolesc Health* 2019;**64**:427-9.
58. Specialisten FM. Kwaliteitsstandaard - jongeren in transitie van kinderzorg naar volwassenenzorg. https://richtlijnendatabase.nl/richtlijn/kwaliteitsstandaard_-_jongeren_in_transitie_van_kinderzorg_naar_volwassenenzorg/startpagina_-_jongeren_in_transitie_van_kinderzorg_naar_volwassenenzorg.html Accessed 10-10-2024, 2022.
59. Nagra A, McGinnity PM, Davis N, Salmon AP. Implementing transition: Ready steady go. *Arch Dis Child Educ Pract Ed* 2015;**100**:313-20.

Chapter 9

Summary

For our young patients, having chronic inflammatory bowel disease (IBD: Crohn's disease and ulcerative colitis) is an uncertain, lifelong journey. Symptoms experienced by the patient depend on the location of the inflammation in the gastrointestinal system, but are often characterized by decreased appetite, abdominal pain, diarrhea and fatigue. Whether the disease responds well to treatment and how long the disease stays in remission can vary from patient to patient. The disease course can have ups and downs: periods with a flare-up (when a patient can feel very sick) and remission (with no or little abdominal symptoms) may alternate. Even when the disease is in remission, patients may still have abdominal pain and fatigue symptoms that can be disabling. IBD patients have a lower quality of life, a higher prevalence of anxiety and depression symptoms and often experience problems at school and in the family. On top of this, a young person with IBD, like their peers, goes through all the stages of adolescence, with its challenges, on their way to adulthood both inside and outside of the hospital world. Within the hospital, this transition process includes the transfer to adult healthcare. This is challenging given the differences between pediatric and adult care and the associated patient's behaviour, such as the development of a certain level of independence. These challenges must be overcome as the consequences of a failing transition can be substantial, including loss of disease follow-up with an increased risk of comorbidities such as intestinal stenosis and colon cancer. The overall aim of this thesis was to investigate how healthcare providers can best guide adolescent and young adult IBD patients during the transition period to adult healthcare. The objective was to optimize the tools used for monitoring of skills building during transition (Part A), and to establish a valid method to measure success (or effectiveness) of the transition process (Part B).

Chapter 1 provides a general overview of this thesis, will introduce the reader to the adolescent's life with IBD and discusses the meaning of transition and transitional care.

Knowledge about their own disease and treatment is an important factor that will contribute to a patient becoming more independent (from their caregivers and/or healthcare providers) in the management of their disease. Measuring instruments concerning knowledge are already available as multiple-choice questionnaires. We wanted to map the adolescents' true knowledge using open-ended questions, leaving no room for guessing answers. We therefore developed a new questionnaire, the Rotterdam Transition Test (RTT).

Chapter 2 describes its development including a correction model and validation process using the COSMIN method. The RTT has 25 open questions for boys and 27 open questions for girls (2 additional questions on pregnancy and childbirth). The questions cover the disease and treatment, but also life with IBD such as smoking, alcohol and drugs, fertility, and the transition of care. From our IBD centre, 111 adolescents completed 207 RTTs. When we tested the RTT, we found that the test was a good and reliable test. We learned that adolescents with IBD who were treated with prednisolone in the past 3 months or had flare- ups of the disease in the past year scored significantly higher on the RTT, meaning they had better disease knowledge. Patients who received so-called 'biological' treatment (infliximab, adalimumab, vedolizumab) scored significantly lower on the RTT, so they had a lower level of knowledge.

Besides increasing knowledge, it is important to increase the independence of adolescents with IBD during the transition process. The Transition Readiness Assessment Questionnaire (TRAQ) is an internationally widely used list to assess the level of independence of chronically ill adolescents during the transition phase. The TRAQ consists of 20 questions on independence divided over 5 domains but was not yet available in Dutch. For this reason, in **Chapter 3** we translated the TRAQ from English to Dutch using the back-to-back method and validated this Dutch version (TRAQ-NL) by COSMIN method. 136 adolescents with IBD completed 250 TRAQs during their visit to our IBD transition clinic. TRAQ-NL discriminates well between different levels of independence but mainly between no/low and high levels of independence. This implies that TRAQ-NL is especially good to use to find adolescents with low levels of self-management. For construct validation, hypothesis testing was used where significant correlation was found with the TRAQ-NL and the pre-set hypotheses such as age, completing the questionnaire more often, completing it after transition to adult care and self-management. For usability in practice, we developed TRAQ-NL reference scores for all 5 domains for IBD patients. Since age plays an important role in the level of independence, we calculated age-dependent reference scores: for patients aged 16 years, 17 years and over 18 years.

Patient Reported Outcome Measures (PROMs), such as the TRAQ, are increasingly used in day-to-day care delivery. However, it is not known how to best implement these PROMs in daily practice. As is well known, IBD patients have a higher risk of developing anxiety and depression symptoms and often have a lower quality of life. Monitoring mental health is therefore advised, but it is difficult to discussing these psychosocial issues in the consulting room.

In **Chapter 4** , we evaluated the experiences of patients and their caregivers with PROMS that assessed quality of life (generic and disease-specific), anxiety and depression symptoms, cognitive functioning and social relationships. Prior to this study, all PROMs were incorporated into the electronic patient record (EPR). A dashboard in each patient's EPR visualized the score level, making it easier to discuss the results with patients and caregivers. We asked patients who had completed the PROMs twice to complete a Patient Reported Experience Measure (PREM), about their experience of completing the PROMs, the healthcare provider's discussion of the PROMs and the patient dashboard. Out of 71 patients (who completed the PROM twice), 25 IBD patients (mean age 15 years) completed the PREM. Satisfaction with the patient dashboard and healthcare provider's discussing the results was neutral to good. However, 40% indicated that the results of the questionnaire were not discussed. When asked about the PROMs, there was some negative feedback such as the questionnaire being too long and offered too often (every 3 months in this study).
These are good points that will be taken into account when further implementing PROMs in the care of children and adolescents with IBD.

Besides disease-related knowledge and self-management, self-efficacy is seen as an important factor for successful transition. Self-efficacy is a person's belief in his or her ability to organize and carry out the actions needed to deal with future situations and is considered a prerequisite for self-management. In 2008, our research group had developed a questionnaire on self-efficacy of the IBD patient; IBD-Yourself. In **Chapter 5**, we examined the potential of the questionnaire outcome to serve as a predictor of transition success. In the absence of a pre-established definition or scoring system for transition success, our team, in collaboration with an expert panel comprising pediatric IBD specialists and transition experts, devised the Transition Yourself Score. The score was comprised of three elements: appearance at appointments at the adult gastroenterology outpatient clinic, therapy adherence, and a rating of patient satisfaction. Of the 35 IBD patients included in the 2014 study, 63% were found to have undergone a successful transition, 31% had undergone an average transition, and 6% had undergone a failed transition. No correlation was identified between self-efficacy and the success of transition, as measured by the Transition Yourself Score. One potential explanation for this discrepancy is that self-efficacy may not always align with the actual behaviour of the adolescent.

It is crucial to demonstrate the efficacy of specific interventions during the transitional care phase. The absence of a universally accepted standard for measuring the success of a transition and a clear definition of what constitutes a successful transition became evident during the research described in Chapter 5. In **Chapter 6**, we employed a Delphi procedure to advance the definition of transition success. This study compiled a list of outcomes that reflect transition success. The opinions were asked of a large group of international IBD experts from both pediatric and adult care (n=74) and a patient panel (n=61). Analyses, using RAND UCLA criteria, showed agreement of 10 (by IBD experts) and 11 outcomes (by patients) considered to be indicative for transition success. Both panels agreed with each other on 8 of these items. Interestingly, 6 of these 8 items concerned self-management skills and did not include any disease-specific characteristics. Based on these items, a Top 10 was created in phase 3 where 'making independent decisions regarding IBD' was seen as the most important. All this formed the basis for the Transition Success Score (TSS), the measurement instrument whose development, validation and initial outcomes are described in Chapter 7. The same international group of IBD experts agreed through 4 rounds on the 9 questions and 3 answer options per question based on the Top 10 list. Seven questions were devised for healthcare providers in adult care settings to assess the degree of independence exhibited by patients in their behaviour regarding their disease, medication, and conduct in the consulting room. The final two questions pertained to satisfaction, one for the patient and one for their caregiver. Based on the TSS, a definition of success for transition was formulated: '*The transition to adult healthcare is a success when the young adult patient is able to manage their illness independently and knows how to navigate the adult healthcare system with satisfaction'.*

Chapter 7 describes the multicentre national prospective study where the TSS was administered to 160 young adult IBD patients from 7 different hospitals in the Netherlands, 9-15 months after their transfer to adult care. Using COSMIN method, the validation and reliability of the TSS were examined. For construct validation, the TSS was compared with validated questionnaires related from the literature to transition such as Rotterdam Transition Test (RTT; Chapter 2), TRAQ-NL (Chapter 3) and quality of life. A significant correlation was found between the questionnaires, especially between the TRAQ-NL (self-management skills) and TSS. Moreover, the TSS was observed to primarily identify patients in whom the transition was unsuccessful. To enhance the TSS's utility in clinical practice, cut-off values were established. We found that 70% of young adult IBD patients had experienced a successful transition and that in 3.8% the transition had failed. More disease activity, more disease burden and exacerbation after transfer were related to a lower TSS score, i.e., less successful transition. This was also the case

for patients who were seen as 'backseat' patients. One issue that emerged in the process of measuring transition success was that the results may have been overestimated due to the fact that the study population consisted primarily of willing, therapy-compliant patients.

Chapter 8 reflects on the main findings of our studies and makes recommendations for practice and future research. The utilization of questionnaires such as the TRAQ-NL and the RTT can facilitate the identification of areas where patients may require additional assistance in their transition towards an independent, informed young adult with a comprehensive understanding of their disease and treatment. While it is essential to tailor care to the individual patient, it is also crucial to ensure that the time investment required by the patient is commensurate with the utility and objective of the questionnaires. Our research on measuring transition success has resulted in the development of the TSS, a valid and reliable instrument for measuring transition success. The TSS has been developed in a well- established manner. It is recommended that our findings be confirmed in larger, international patient groups, with a particular focus on young adults who are either unwilling or unable to participate in the study. This is because these patients are potentially more likely to have disappeared from the healthcare system, indicating a failed transition. Consequently, our results may be biased towards more successful transitions.


10

Chapter 10

Samenvatting

Het hebben van chronische darmontsteking (inflammatory bowel disease, IBD) is een onzekere, levenslange reis. Onder IBD vallen de ziekte van Crohn en colitis ulcerosa. Welke klachten de patiënt ervaart is afhankelijk van de locatie van de ontsteking in het maagdarmstelsel, maar vaak zijn het klachten van verminderde eetlust, buikpijn, diarree en vermoeidheid. Of de ziekte goed reageert op een behandeling en hoelang de ziekte dan rustig blijft kan per patiënt verschillen. De ziekte gaat gepaard met rustige fases van remissie (geen of heel weinig klachten), en opvlammingen (opspelen van klachten). Ook wanneer de ziekte rustig is kan een patiënt nog buikpijn en vermoeidheidsklachten hebben welke invaliderend kunnen zijn. IBD-patiënten hebben een lagere kwaliteit van leven gecombineerd met een hogere kans op angst en depressieklachten en problemen op school en in het gezin. Hier komt nog bij dat een jongere met IBD net als zijn leeftijdsgenoten door alle fases van adolescentie gaat, met de daar bijhorende uitdagingen, op weg naar volwassenheid zowel binnen als buiten het ziekenhuis. Binnen het ziekenhuis hoort bij dit transitieproces de overstap (of transfer) naar de zorg voor volwassenen. Dit is een uitdaging gezien de verschillen in zorg tussen kinderzorg en zorg voor volwassen en het bijbehorend gewenste gedrag van de patiënt, zoals de ontwikkeling van een bepaalde mate van onafhankelijkheid. Deze uitdagingen moeten worden overwonnen omdat de gevolgen van een mislukt transitieproces aanzienlijk kunnen zijn, waaronder verlies van ziektecontrole met een verhoogd risico op complicaties zoals darmvernauwing en darmkanker. Het algemene doel van dit proefschrift was om te onderzoeken hoe zorgverleners adolescente en jongvolwassen IBD-patiënten het beste kunnen begeleiden tijdens de transitieperiode naar de gezondheidszorg voor volwassenen. Hiertoe hebben we meetinstrumenten ontworpen die de vaardigheidsontwikkeling tijdens de transitie vervolgen (Deel A), en een valide test ontwikkeld waarmee het succes (of de effectiviteit) van het transitieproces kan worden gemeten (Deel B).

In **Hoofdstuk 1** wordt naast een overzicht van de inhoud van dit proefschrift ingegaan op het leven van de adolescent met IBD en de betekenis van transitie en transitiezorg.

Kennis over de eigen ziekte en de behandeling is een belangrijke factor die bijdraagt aan het zelfstandig zorg dragen over deze ziekte. Meetinstrumenten voor kennis zijn reeds beschikbaar, maar hierbij werd altijd gebruikt gemaakt van meerkeuze vragenlijsten. Wij wilden middels open vragen, zodat er geen ruimte is voor het gokken van antwoorden, de “echte” kennis van de jongeren in kaart brengen. Daarom hebben wij een nieuwe vragenlijst ontwikkeld, de Rotterdamse Transitie Toets (RTT). In **Hoofdstuk 2** wordt de ontwikkeling van de RTT en het daar bijhorende correctiemodel en validatieproces middels de COSMIN- methode

beschreven. De RTT bestaat uit 25 open vragen voor jongens en 27 open vragen voor meisjes (2 extra vragen over zwangerschap en bevalling). De vragen gaan over de ziekte en behandeling, maar ook over het leven met IBD zoals roken, alcohol en drugs, vruchtbaarheid en de transitiefase. Uit ons IBD-centrum hebben 111 adolescenten 207 RTTs ingevuld. De analyses laten zien dat de RTT, zoals bij een goede vragenlijst hoort, verschillende soorten vragen bevat en een onderscheid kan maken tussen de verschillende niveaus van kennis. Om te bepalen of de RTT meet wat hij moet meten (construct validatie) is er middels testen van hypotheses onderzocht of er een significante correlatie is tussen de RTT-totaalscore en vooraf gestelde hypotheses zoals hogere scores op de RTT bij vaker de vragenlijst invullen, oudere leeftijd en meer zelfstandigheid. Dit was inderdaad het geval. De RTT laat zien dat adolescenten met IBD die de afgelopen 3 maanden met prednison werden behandeld of het afgelopen jaar een opvlamming van de ziekte hadden, significant hoger scoren op de RTT, dus meer kennis hadden. Patiënten die een zogenoemde “biological” behandeling (infliximab, adalimumab, vedolizumab) kregen scoorden significant lager op de RTT, dus hadden een lager kennisniveau.

Naast vergroten van kennis is het belangrijk dat de zelfstandigheid van de adolescenten met IBD toeneemt tijdens het transitieproces. De *Transition Readiness Assessment Questionnaire (TRAQ)* is een internationaal veel gebruikte lijst om inzicht te krijgen in de mate van zelfstandigheid van chronisch zieke adolescenten tijdens de transitiefase. De TRAQ bestaat uit 20 vragen over zelfstandigheid verdeeld over 5 domeinen, maar was nog niet beschikbaar in het Nederlands. Om deze reden hebben we in **Hoofdstuk 3** de TRAQ middels de back-to- back methode vertaald vanuit het Engels naar het Nederlands en deze Nederlandse versie (TRAQ-NL) gevalideerd middels COSMIN-methode. 136 adolescenten met IBD hebben 250 TRAQ lijsten ingevuld tijdens hun bezoek aan ons IBD-transitiespreekuur. TRAQ-NL discrimineert goed tussen de verschillende niveaus van zelfstandigheid maar voornamelijk tussen geen/laag en hoog niveau van zelfstandigheid. Dit impliceert dat de TRAQ-NL vooral goed te gebruiken is om adolescenten te vinden met een laag niveau van zelfmanagement. Voor constructvalidatie werd er gebruikt gemaakt van hypothese testen waarbij er een significante correlatie gevonden werd met de TRAQ-NL en de voor afgestelde hypotheses zoals leeftijd, vaker invullen van de vragenlijst, invullen na de overstap naar de zorg voor volwassenen en zelfstandigheid. Voor bruikbaarheid in de praktijk hebben we TRAQ-NL referentie scores voor alle 5 de domeinen ontwikkeld voor IBD-patiënten. Aangezien leeftijd een belangrijke rol speelt in de mate van zelfstandigheid hebben we de leeftijdsafhankelijke referentiescores berekend: voor patiënten van 16 jaar, 17 jaar en ouder dan 18 jaar.

Patiënt Reported Outcome Measures (PROMs), zoals de TRAQ, worden steeds vaker gebruikt in de dagelijkse zorgverlening. Het is echter niet bekend hoe je deze PROMs het beste kan inzetten in de dagelijkse praktijk. Zoals bekend hebben IBD-patiënten een hogere risico op het ontwikkelen van angst en depressieklachten en hebben ze vaak een lagere kwaliteit van leven. Hoewel wordt geadviseerd om de mentale gezondheid van jongeren met IBD frequent dit te monitoren, is het bespreken van deze psychosociale onderwerpen lastig in de spreekkamer. In **Hoofdstuk 4** beschrijven we hoe de ervaringen van patiënten en hun verzorgers met PROMS ten aanzien van kwaliteit van leven (generiek en ziekte-specifiek), angst en depressieklachten, cognitieve functioneren en sociale relaties zijn. Voorafgaande aan dit onderzoek waren alle PROMs ingebouwd in het elektronisch patiëntendossier. Middels een dashboard is daardoor inzichtelijk wat de gescoorde score betekent waardoor het makkelijker is om de resultaten samen met de patiënt en verzorgers te bespreken. We vroegen aan de patiënten die 2 keer de PROMs hadden ingevuld om een vragenlijst, een PREM; Patiënt Reported Experience Measure, naar hun ervaring met het invullen van de PROMs, het bespreekbaar maken van de PROMs door de zorgverlener en het patiënten dashboard in te vullen. Van de 71 patiënten (die 2 keer de PROMs hadden ingevuld) hebben 25 IBD patiënten (gemiddelde leeftijd 15 jaar) de PREM ingevuld. De tevredenheid over het patiënten dashboard en het bespreekbaar maken van de uitslagen door de zorgverlener was neutraal tot goed. In 40% bleek echter dat de uitslag van de vragenlijst niet met hen besproken was. Betreffende de PROMs zelf kwam wel enige negatieve feedback zoals te lange vragenlijsten en te vaak moeten invullen (in deze studie was dit elke 3 maanden). Dit zijn goede punten die meegenomen worden bij het verder implementeren van PROMs bij kinderen en adolescenten met IBD.

Naast ziekte-gerelateerde kennis en zelfmanagement wordt zelfredzaamheid als een belangrijke factor gezien voor het slagen van transitie. Zelfredzaamheid is het geloof van een persoon in zijn of haar vermogen om de acties te organiseren en uit te voeren die nodig zijn om met toekomstige situaties om te gaan en wordt beschouwd als een voorwaarde voor zelfmanagement. In 2008 had onze onderzoeksgroep een vragenlijst ontwikkeld om te kijken naar zelfredzaamheid van de IBD-patiënt; IBD-Yourself. In **Hoofdstuk 5** onderzochten we of de uitslag van deze vragenlijst een voorspeller kon zijn voor succes van transitie. Gezien er nog geen definitie of score voor succes van transitie bestond, ontwikkelden we, na een focusgroep bijeenkomst, met een expertpanel een nieuwe score, de Transition Yourself Score. Deze score bestond uit de volgende items: verschijnen op afspraken op de Maag Darm Lever (MDL) polikliniek voor volwassenen, therapietrouw en een rapportcijfer ten aanzien van de tevredenheid van

de patiënt. Van de 35 IBD-patiënten (geïncludeerd in 2014) bleek 63% een succesvolle transitie, 31% een gemiddelde transitie en 6% een mislukte transitie te hebben doorlopen. Er werd geen relatie gezien tussen zelfredzaamheid, en succes van transitie, gemeten met Transition Yourself. Een mogelijke verklaring is dat zelfredzaamheid niet altijd overeenkomt met het werkelijke gedrag van de adolescent.

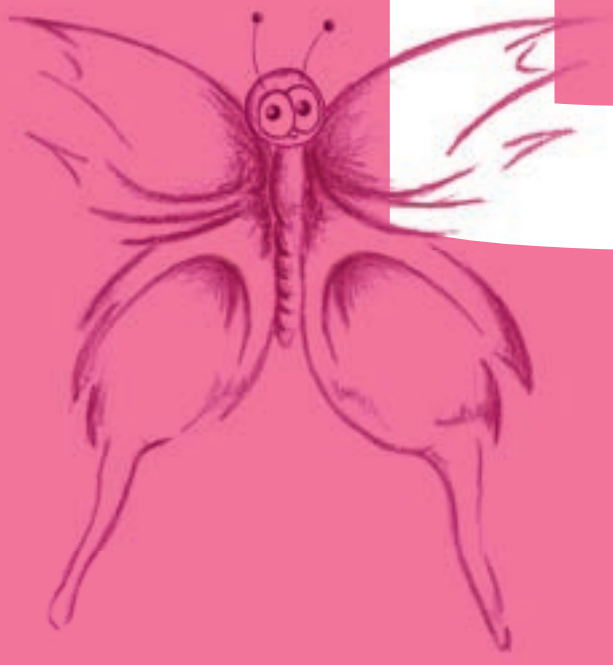
Het gemis van een gouden standaard voor het meten van succes van transitie en een heldere definitie van wanneer transitie succesvol is werd duidelijk tijdens het onderzoek zoals beschreven in Hoofdstuk 5. Om aan te tonen dat bepaalde interventies tijdens transitiezorg zinvol zijn is het van belang om het nut van deze interventies aan te tonen. In **Hoofdstuk 6** maakten we middels een Delphi procedure de volgende stap in het definiëren van het succes van transitie. In deze studie werd een lijst opgesteld van uitkomsten die het succes van transitie weerspiegelen. De mening werd gevraagd van een grote groep internationale IBD- experts vanuit zowel de kinderzorg als de zorg voor volwassenen (n=74) en een patiënten panel (n=61). Analyses toonde met behulp van de RAND UCLA criteria overeenstemming van 10 (door IBD-experts) en 11 items (door patiënten) die belangrijk werden geacht voor het succes van transitie. Beide panels waren het met elkaar eens over 8 van deze items.
Opvallend was dat 6 van deze 8 items zelfmanagementvaardigheden betroffen en er geen enkel ziekte-specifiek kenmerk bij zat. Zo werd in fase 3 op basis van deze items een Top 10 gevormd waarbij 'zelfstandig beslissingen maken omtrent IBD' als belangrijkste werd gezien. Dit alles heeft de basis gevormd voor de Transitie Succes Score (TSS), het meetinstrument waarvan de ontwikkeling, validatie en eerste uitkomsten zijn beschreven in Hoofdstuk 7.

Dezelfde internationale groep van IBD-experts heeft middels 4 rondes overeenstemming bereikt over de 9 vragen en 3 antwoordmogelijkheden per vraag gebaseerd op de Top 10 lijst. Er werden 7 vragen geformuleerd voor de zorgverlener in de zorg voor volwassenen betreffende de mate van zelfstandigheid van de patiënt rondom zijn ziekte, medicatie en gedrag in de spreekkamer. De laatste 2 vragen gaan over tevredenheid; 1 voor de patiënt en 1 voor hun verzorger. Op basis van de TSS werd de volgende definitie gemaakt voor succes van transitie: *De overstap naar de zorg voor volwassenen is geslaagd wanneer de jongvolwassen patiënt in staat is om zijn ziekte zelfstandig te managen en zijn weg in de zorg voor volwassenen weet te vinden met tevredenheid.*
Hoofdstuk 7 beschrijft de grote nationale studie waar bij 160 jongvolwassen IBD-patiënten vanuit 7 verschillenden ziekenhuizen in Nederland, 9-15 maanden na hun overstap naar de zorg voor volwassenen, de TSS werd afgenomen. Middels

COSMIN-methode werd gekeken naar de validatie en betrouwbaarheid van de TSS. Voor construct validatie werd de TSS vergeleken met gevalideerde vragenlijsten die gerelateerd zijn vanuit de literatuur met transitie zoals de Rotterdam Transitie Toets (RTT; Hoofdstuk 2), TRAQ-NL (Hoofdstuk 3) en kwaliteit van leven. Tussen al deze meetinstrumenten werd een significante correlatie gevonden, vooral tussen de TRAQ-NL (zelfstandigheid) en TSS. Verder zagen we dat de TSS voornamelijk de patiënten detecteert waarbij de transitie niet succesvol verlopen is. Om de TSS meer bruikbaar te maken in de dagelijkse praktijk werden er afkapwaardes vastgesteld. Zo bleek dat 70% van de jongvolwassen IBD-patiënten een succesvolle transitie had doorgemaakt en dat bij 3.8% de transitie mislukt was. Meer ziekteactiviteit, meer ziektelast en een opvlamming na transfer waren gerelateerd met een lagere TSS-score, dus minder succesvolle transitie. Dit was ook het geval voor patiënten die werd gezien als “backseat” patiënts. Een probleem dat we tegenkwamen bij het meten van transitiesucces is dat de resultaten mogelijk vertroebeld zijn doordat hoofdzakelijk bereidwillige, therapietrouwe, patiënten deel hebben genomen.

In **Hoofdstuk 8** wordt stil gestaan bij de belangrijkste bevindingen van onze studies en worden er aanbevelingen gedaan voor de praktijk en toekomstig onderzoek. Het gebruik van vragenlijsten zoals de RTT en de TRAQ-NL kan patiënten en zorgverleners inzicht geven in of en waar de patiënt nog verder hulp nodig heeft in de ontwikkeling tot een zelfstandige patiënt met voldoende kennis over zijn ziekte en behandeling. De zorg moet zoveel mogelijk gepersonaliseerd worden, waarbij het belangrijk is dat er een balans is tussen de tijdsinvestering van de patiënt en het nut en doel van de vragenlijsten.
Met ons onderzoek naar het meten van transitie hebben we met de TSS een valide en betrouwbaar meetinstrument naar succes van transitie in handen die op een gedegen manier ontwikkeld is. Onze bevindingen moeten bevestigd worden in grotere, internationale, patiëntengroepen, met speciale aandacht voor de jongvolwassenen die niet willen of kunnen deelnemen aan de studie. Dit zijn namelijk de patiënten die in potentie een grotere kans hebben dat ze verdwenen zijn uit het zorgsysteem en dus een mislukte transitie hebben gehad.

D



Part D

Appendices

List of abbreviations
Authors and affiliations
List of publications
PhD Portfolio
About the author

List of Abbreviations

AYA	Adolescent and Young Adult
ANOVA	Analysis of Variance
AIH	Auto-Immune Hepatitis
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
CD	Crohn's disease
d	Days
EEN	Exclusive Enteral Nutrition;
EPR	Electronic Patient Record
ER	Emergency Room
ECCO	European Crohn´s and Colitis Organisation
ESPGHAN	European Society for Pediatric Gastroenterology and Hepatology and Nutrition
F2F	Face to Face
GE	Gastroenterology
HCP	Healthcare providers
IBDQ	IBD Questionnaire
IBD-Q	IBD questionnaire
IBD-U	IBD unclassified
IBD	Inflammatory Bowel Disease
IQR	InterQuartile Range
JCC	Journal of Crohn's and Colitis
JPGN	Journal of Pediatric Gastroenterology and Nutrition
K	Kappa
ns	not significant
n	numbers of participants
PREM	Patient Reported Experience Measure
PROM	Patient-Reported Outcome Measure
r	Pearson correlation
PCDAI	Pediatric Crohn Disease Activity Index
PUCAI	Pediatric Ulcerative Colitis Activity Index
PC	Percentile scores
PGA	Physician Global Assessment
PSC	Primary Sclerosing Cholangitis
QoL	Quality of Life
Q	Question

RTT	Rotterdam Transition Test
SD(S)	Standard Deviation (Score)
NL	The Netherlands (Dutch)
TRAQ	Transition Readiness Assessment Questionnaire
TSS	Transition Success Score
UC	Ulcerative colitis
US	United States
vs	versus
VAS	Visual Analog Scale
y	years
YA	Young Adult

Authors and Affiliations

Gertrude van den Brink	Department of Pediatric Gastroenterology, Erasmus University Medical Centre- Sophia Children's Hospital, Rotterdam, the Netherlands
Marije van Dalen	Department of Child and Adolescent Psychiatry/Psychology, Erasmus Medical Centre- Sophia Children's Hospital
Lauranne Derikx	Department of Gastroenterology, Erasmus University Medical Centre, Rotterdam, the Netherlands
Karolijn Dulfer	Department of Child and Adolescent Psychiatry/Psychology, Erasmus Medical Centre- Sophia Children's Hospital
Alie Dijkstra	Department of Pediatric Gastroenterology, University Medical Centre, Groningen – Beatrix Children's Hospital, Groningen, The Netherlands
Johanna (Hankje) C. Escher	Department of Pediatric Gastroenterology, Erasmus University Medical Centre- Sophia Children's Hospital, Rotterdam, the Netherlands
Marein M. Favejee	Department of Quality & patient care, Erasmus University Medical Centre, Rotterdam, the Netherlands
Emma van Gijn	Department of Pediatric Gastroenterology, Erasmus University Medical Centre- Sophia Children's Hospital, Rotterdam, the Netherlands
Monique S. den Hollander-Ardon	Department of Quality & patient care, Erasmus University Medical Centre, Rotterdam, the Netherlands
Angelika Kindermann	Department of Pediatric Gastroenterology, Amsterdam University Medical Centre – Emma Children's Hospital, Amsterdam, The Netherland
Merel van Pieterson	Department of Pediatric Gastroenterology, Erasmus University Medical Centre- Sophia Children's Hospital, Rotterdam, the Netherlands
Lissy de Ridder	Department of Pediatric Gastroenterology, Erasmus University Medical Centre- Sophia Children's Hospital, Rotterdam, the Netherlands
Dimitris Rizopoulos	Department of Biostatistics, Erasmus University Medical Centre, Rotterdam, the Netherlands
Petra Waaijenberg	Department of Gastroenterology, Amsterdam University Medical Centre, Amsterdam, The Netherlands
Herbert van Wering	Department of Pediatrics, Amphia Hospital, Breda, The Netherlands
Margreet Wessels	Department of Pediatrics, Rijnstate Hospital, Arnhem, The Netherlands
Victorien M. Wolters	Department of Pediatric Gastroenterology, University Medical Centre Utrecht – Wilhelmina Children's Hospital, Utrecht, The Netherlands
C. Janneke van der Woude	Department of Gastroenterology, Erasmus University Medical Centre, Rotterdam, the Netherlands
Marieke Zijlstra	Department of Pediatric Gastroenterology, Utrecht Medical Centre/ Wilhelmina Children's Hospital, Utrecht, the Netherlands

List of Publications

Included in this thesis

G van den Brink, **MAC van Gaalen,** M Zijlstra, L de Ridder, CJ van der Woude, JC Escher. Self-efficacy did not predict the outcome of the transition to adult care in adolescents with inflammatory bowel disease. Acta Paediatrica 2019;108:333–8.

G van den Brink G, **MAC van Gaalen,** L de Ridder, CJ van der Woude, JC Escher. Health care transition outcomes in inflammatory bowel disease: a multinational Delphi study. J Crohns Colitis 2019;13:1163–72.

MAC van Gaalen, M van Pieterson, G van den Brink, L de Ridder, D Rizopoulos, JC Escher. Rotterdam transition test: a valid tool for monitoring disease knowledge in adolescents with inflammatory bowel disease. J Pediatr Gastroenterol Nutr 2022;74:60–7

MAC van Gaalen, E van Gijn, M van Pieterson, L de Ridder, D Rizopoulos, CJ van der Woude, JC Escher. Validation and reference scores of the transition readiness assessment questionnaire in adolescent and young adult IBD patients. J Pediatr Gastroenterol Nutr 2023;77:381–8.

MAC van Gaalen, M van Pieterson, P Waaijenberg, A Kindermann, V Wolters, A Dijkstra, H van Wering, M Wessels, L de Ridder, D Rizopoulos, CLAAP. Derikx, JC. Escher, on behalf of the Kids with Crohn's, Colitis (KiCC) Working Group for Collaborative Paediatric IBD Research in the Netherlands, the Dutch Initiative on Crohn and Colitis (ICC) and Dutch Nurses Network Inflammatory Bowel Disease (NIBD). Effectiveness of Transitional Care in Inflammatory Bowel Disease; Development, Validation, and Initial Outcomes of a Transition Success Score. J Crohn's Colitis 2024; Nov 2:jjae166. doi: 10.1093/ecco-jcc/jjae166. Online ahead of print

M van Dalen, **MAC van Gaalen**, MM Favejee, MS den Hollander – Ardon, K Dulfer, L de Ridder, JC. Escher. Implementing routine medical and mental health screening in children and adolescents with inflammatory bowel disease. J Pediatr Gastroenterol Nutr 2025 (Accepted)

Not included in this thesis

MAC van Gaalen, JC Escher. Transitie van zorg voor jongeren met een chronische darmontsteking. Praktische pediatrie 2023

G van den Brink, CJ van der Woude, L de Ridder, **MAC van Gaalen,** JC Escher. Transition form pediatric to adult care; mangement of adolescents with inflammatory bowel disease. Ned Tijdschr Geneeskd 2016; 160:D578

PhD-Portfolio

	Year	ECTS
General Courses		
Online WMO/ GCP training	2018	1
Online WMO/ GCP training Refresher	2022	0.5
Motivational Interviewing	2018	2
Erasmus MC PhD introduction session	2023	0.2
Scientific Integrity	2023	0.3
BROK course	2024	1.5
Presentations		
Seminars and workshops		
Lab and clinic meetings (twice a month) Attending + Oral (two times)	2019-2024	1
Journal Club pediatric gastroenterology Attending (once a month) + article input twice a year	2020	1
Daycare update Oral	2020	0.2
T-Pensant Rotterdam Kinder-MDL Attending	2020	0.2
Training Praktijkopleiders VIOS MANP Attending	2020	0.2
Hot & Happening Pain Webinar Attending	2020	0.2
Zet de patiënt op 2. Zorg voor anderen begint bij jezelf Attending	2020	0.2
T-Pensant pediatric gastro-enterology Attending	2021	0.2
Regional meeting IBD- nurses (4x) Organizer + Oral	2021-2024	2
Webinar 'Denk in groei!' Attending	2021	0.2
Referral evening Erasmus MC VS Oral	2021	0.2
Referral evening Erasmus MC- Sophia (3x) Organizer + Oral	2022-2024	2
Department VWS; Value care based in PIBD Oral	2024	0.5
Inspection Gezin en Jeugd (IGJ); patient-centred care PIBD Oral	2024	0.5
National conferences		
KinderFonds Symposia Oral	2021	0.8
DDD Oral	2021	0.8

	Year	ECTS
First MDL Transition Symposium Organizer + Oral	2021	1
IBD Todag & Tomorrow Attending	2021	0.3
IBD leeft! Attending	2022	0.3
DDD Oral	2022	0.3
Second MDL Transition Symposium Organizer + Oral	2023	1
V&VN VS Oral	2023	1.1
National Voedingscongres Oral	2024	0.8
International conferences		
European Crohn's and Colitis Organisation (ECCO) Attending *Copenhagen*	2019	0.9
PIBD Oral + Poster *Budapest*	2019	1.4
European Crohn's and Colitis Organisation (ECCO) Attending *Wenen*	2020	0.9
European Crohn's and Colitis Organisation (ECCO) Attending *Online*	2021	0.9
PIBD Poster *Edinburgh*	2022	0.9
European Crohn's and Colitis Organisation (ECCO) Oral *Online*	2022	0.9
ESPGHAN Annual Meeting Attending *Copenhagen*	2023	0.9
European Symposium on Transition Oral *Rotterdam*	2023	0.6
European Crohn's and Colitis Organisation (ECCO) Attending *Copenhagen*	2023	0.6
European Crohn's and Colitis Organisation (ECCO) Oral + poster *Stockholm*	2024	0.9
Tillotts nurses IBD congres Oral *Amsterdam*	2024	0.3
ESPGHAN Annual Meeting Oral *Milan*	2024	0.6
ESPGHAN Masterclass on Transition in care Oral *Porto*	2024	0.6

	Year	ECTS	
Teaching			
Tutor 3 MANP student for 2 years	2020	2	
Annual Lecture Nurse IBD – Pediatric IBD And Transition Hogeschool Arnhem/ Nijmegen	Since 2015	1	
Supervising Master thesis medical student Nicola Abbink	2020	1	
Supervising medical student Emma Gijn	2021	0.5	
Gastro enterology course pediatric nurses (3 times a year)	2023	1	
Board Experiences			
Member of the board of nurse specialists at Sophia Children's Hospital	2019		
Chair of the board of nurse specialists at Sophia Children's Hospital	Since 2021		
Member of working group *Young Adult Program (YAP)*	2023/2024		2
Member of working group *Value Based Healthcare*	2024		1
Others			
Chapter Transition written in revised guideline "IBD bij kinderen"		2018-2019	2
"meelezer" revised guideline "IBD bij kinderen"		2024	0.5

About the Author

Martha van Gaalen was born on 12 March 1987 in Breukelen. After completing her secondary education at the Nassau (HAVO) in Breda, she started a dual study program in nursing at the Rotterdam University of Applied Sciences in cooperation with the Erasmus Medical Centre in 2004. She followed the minor child and hospital. She obtained her Bachelor of Nursing degree in 2008. In 2009, she went on to graduate as a registered pediatric nurse. After several years of experience as a nurse at the pediatric department of the Erasmus MC- Sophia children's hospital, Martha started the Master Advance Nurse Practice (MANP) at the University of Applied Sciences in Leiden in 2012, combined with a job as a nurse specialist in training in pediatric gastroenterology at the Sophia children's hospital under supervision of Prof. Dr. J.C. Escher.

From 2014 until now she has been working as a nurse specialist in pediatric gastroenterology with a focus on pediatric inflammatory bowel disease (IBD). In addition to her clinical work as a nurse specialist, she teaches IBD nurses in training at the Arnhem/Nijmegen University of Applied Sciences, organizes various training courses, participates in various project groups (Young Adult Program and Value Based Healthcare) and is, among other things, chair of the board of nurse specialists at Sophia Children's Hospital.

In recent years, Martha has been involved in several studies to improve the quality of care for young adults with inflammatory bowel disease who are transitioning from pediatric to adult care. She has developed a knowledge questionnaire, the Rotterdam Transition Test and the Dutch version of the TRAQ, and has worked on making the success of transition to adult care quantitatively measurable (TSS).

Dankwoord

Wat ben ik dankbaar dat ik dit dankwoord mag schrijven. Het is toch het meest gelezen hoofdstuk van een proefschrift. Ik had het nooit verwacht en het was ook zeker niet gelukt zonder hulp van velen.

Ik ben trots op, toch wel, mijn IBD- patiënten. Die elke dag geconfronteerd worden met de uitdagingen van deze darmziektes, de een meer dan de ander, maar allemaal op hun eigen manier omgaan met de onzekerheden en tegenslagen. Als jullie verpleegkundig specialist is het een eer dat ik jullie zo goed als mogelijk mag begeleiden met deze uitdagingen maar ook om de succesmomenten te vieren. Ik ben jullie daar allemaal dankbaar voor. Vele van jullie waren zeer bereid om deel te nemen aan de onderzoeken uit dit proefschrift. Weer meer vragenlijsten invullen, toch iets wat voor vele een belasting is naast het drukke leven. Jullie deden het en dat is geweldig. Zonder jullie is het niet mogelijk om de zorg voor jongeren met IBD naar een hoger niveau te tillen.

In de zorg voor chronisch zieke kinderen werk je in een team met ouders. Ik ben dan ook dankbaar voor het vertrouwen, de openheid en bijdragen van de ouders/verzorgers van de IBD-patiënten in de uitgevoerde onderzoeken.

Mijn promotieteam

Mijn promotor **Prof. dr. J.C. Escher**; Lieve Hankje, als er een iemand voor heeft gezorgd dat ik dit dankwoord mag schrijven ben jij het wel. Niet alleen heb je mij de mogelijkheid gegeven om verpleegkundig specialist te worden (als een van de eerste binnen de kindergeneeskunde). Jij gaf mij de ruimte en het vertrouwen om de functie zo vorm te geven dat het nu deze te gekke afwisselende baan is. Waarbij je mij uitdaagde om ook onderzoek te gaan doen op jouw interessegebied; transitie. Ik ben dankbaar voor al je uren correctiewerk van mijn teksten, door mijn dyslexie en belabberde Engels (geslaagd met een vijfje op de HAVO), had je daar een aardige kluif aan. Ik heb je nooit horen klagen en dat is jouw kracht; jouw positiviteit, welke aanstekelijk werkt en motiveert. Maar je respecteerde ook mijn behoefte om mijn eigen route te bewandelen en niet direct in een promotietraject te gaan. Ik heb zo geboft dat jij mijn promotor wilde zijn, bedankt voor alles!

Mijn copromotor **Dr. L. de Ridder**; Lieve Lissy, samen met Hankje vormen wij met z'n drieën HET basisteam voor onze IBD patiënten. Alle drie zijn wij anders, qua persoonlijkheid en manier van werken, maar daarom vullen wij elkaar zo goed aan. Wij zijn alle drie goed in multitasking, maar jij spant hierin echt de kroon. Bewonderingswaardig vind ik hoe goed jij kan prioriteren en je focus kan houden. Ik probeer hier zoveel mogelijk van te leren. Ik ben je dankbaar voor je interesse en feedback op de onderzoeksopzetten en artikelen, met altijd de focus op het doel; kwaliteitsverbetering voor de patiënt.

De leden van mijn beoordelingscommissie **Prof. dr. E.H.H.M. Rings, Prof. M van Dijk en Dr. P. van Rheenen,** hartelijk dank dat u allen deel uit wilt maken van mijn leescommissie. Ik zie er naar uit om met jullie en de rest van de commissie van gedachten te wisselen over mijn proefschrift.

Beste Monique, ik wil jou graag in het bijzonder bedanken voor het gesprek met jou van enkele jaren geleden toen mijn eerste artikel gepubliceerd was. Daarbij kwam direct de vraag op of ik niet moest gaan promoveren (iets wat ik toen nog enger vond, dan dat ik het nu vind). Jij gaf mij de tip om eerst gewoon lekker onderzoek te doen en artikelen te schrijven. Als het dan tot een promotie komt is dat leuk, maar geen verplichting. Deze route was voor mij perfect naast mijn gezin en werkzaamheden in de directe patiëntenzorg.

Kinder-MDL team

Lieve **Merel**, van samen subsidies indienen, patiënten includeren en achter hun broek aan zitten om vragenlijsten in te vullen tot de vele reisjes naar de congressen, waarbij wij goede gesprekken hadden. Bedankt voor alle hulp en tips, zowel zakelijk als privé.

Lieve **Barbara**, wat ben ik jou dankbaar voor jouw enthousiasme en je interesse in mijn werk, maar ook in mij als mens. Als ik even ergens mee zit dan ben je een heerlijke gesprekspartner. Je helpt de boel te relativeren en ik kom altijd beter uit ons gesprek.

Lieve **Marjolein**, ik denk nog regelmatig aan ons reisje samen naar de ESPGHAN in Kopenhagen met het veel te dure broodje hamburger met een wijntje op een terrasje. Maar het leven moet gevierd worden. Ik ben blij dat jij hier ons, als collega's, regelmatig aan helpt herinneren met een koffie halen of een borrel bij jou thuis.

Lieve **Leontien**, mede VS-er Kinder-MDL, kamergenoot; dank voor je interesse en het aanhoren van al mijn uitdagingen. Jij bent altijd bereid om even mee te kijken en te denken. Ik ben benieuwd of je er bent tijdens mijn verdediging of dat je al mag genieten van jullie kleine meid!?

Lieve **Sabine, Marieke, Maaike, Willemien, Yenny, Caroline, Pauline, Dagmar** en **Esther**, wat vond ik het fijn als jullie als fellow/ verdieper een IBD-blok hadden. Het betekende dat jullie even wat werk van mij overnamen en ik meer tijd kon besteden aan mijn onderzoek. Bedankt daarvoor!

Lieve **Willie**, de draaiende motor binnen de kinder-MDL. Mijn steun en toeverlaat voor advies over van alles en nog wat, maar vooral voor een heerlijk kletspraatje!

Beste medeauteurs,

Ik wil alle medeauteurs bedanken voor hun bijdragen aan de hoofdstukken in dit proefschrift.

In het bijzonder wil ik graag alle TSS onderzoekers uit de deelnemende centra bedanken. Het was een ingewikkelde multicenter studie, waarbij een goede samenwerking tussen kinderzorg, zorg voor volwassenen, artsen en verpleegkundig (specialisten) essentieel was, zoals dit ook bij transitie in de zorg van belang is. Wat gaaf dat dit met elkaar gelukt is! Bedankt voor jullie tijd en inzet.

Lieve **Gertrude**, ik ben jou dankbaar dat ik, in het begin, 2 artikelen samen met jou heb mogen schrijven. Onderzoek doen had wel mijn interesse, maar was echt nog niet bezig was met het idee van promoveren. Jij nam mij mee in het proces en ik heb mogen leren van de valkuilen van het onderzoek doen naar het meetbaar maken van succes van transitie. Het was niet zo gemakkelijk als wij dachten!

Lieve **Marije**, ik ben je dankbaar voor al je werk rondom het inzetten van PROMs in de spreekkamer voor IBD patiënten. Naar mijn mening is dit echt van toegevoegde waarde en de toekomst van de zorg.

Beste **Dimitris**, wat heb ik een hoop van je mogen leren over statistiek. Erg fijn dat ik altijd even mijn analyses bij je kon laten controleren of dat ik kon aankloppen voor een lesje statistiek voor Dummies. Bedankt voor alle hulp en je geduld.

(Master) studenten,

Lieve **Emma** en **Nicoline**; bedankt voor jullie bijdrage aan het includeren van patiënten en het verzamelen, invoeren en analyseren van data. Het was een eer om jullie te mogen begeleiden.

Lieve vrienden,

Lieve **Robin, Jeroen** en **Alex** (Ilse pak ik later nog even aan) wat ben ik jullie dankbaar voor jullie vriendschap. Robin, wat kennen wij elkaar al lang en al die jaren weet ik dat je voor mij klaar staat. Zelfs toen ik je even nodig had als leerkracht om de TransitieToets te controleren op taal en opzet van een vragenlijst deed jij dit met liefde. Met Jeroen aan jouw zijde vormen jullie een topteam voor onze "eetclub"-momentjes. Echt Jeroen, wat jij allemaal van die Greenegg tovert, verrukkelijk! Heerlijk zijn die avonden om alles te vergeten en te lachen om niks. Lieve Alex, wat ben ik blij dat jij mij na al dat eten weer op scherp zet met Kudo's en Ride on's op STRAVA en ZWIFT. Ik ben jou naast de spaarzame dansjes en vele gezellige avonden (met gin-tonic) vooral dankbaar voor onze gesprekken. Jij hebt de kracht om mensen op scherp te zetten en op een "Alex-manier" te motiveren.

Ik ben jullie ook dankbaar voor jullie prachtige kinderen; **Lotte, Fenna, Floor** en **Daan**. Wat een toppers zijn zij. Bedankt dat ik ze af en toe lekker mag verpesten!

Lieve **Evelien** en **Marleen**; 2 van de drie zusters, jullie zitten in de basis van mijn professionele leven. Wij zijn samen begonnen op de HBO-V en wij zijn alle drie totaal een andere kant op gegaan, zowel beroepsmatig als voor wat betreft onze woonplaats. We spreken elkaar echt te weinig, maar als we elkaar spreken is het weer als vanouds. Saskia, onze HBO-V coach van destijds, zal trots op ons zijn.

Lieve Peertjes van de MANP; **Anne-Wil, Kayleigh, Brigit** en **Marianne**, wat een lol hebben wij gehad tijdens de MANP opleiding. Bedankt hiervoor! Al zijn de momenten dat wij elkaar spreken schaars, zeker nu jullie één voor één het Sophia verlaten, ik ben zo trots op jullie. Allemaal een druk gezinsleven en allemaal top VS-ers! Jullie riepen het al tijdens de opleiding dat dit boekje er zou gaan komen. Ik wilde jullie niet geloven, maar jullie hebben gelijk; hier is het!

Lieve **collega verpleegkundig specialisten in het Sophia en van de N-IBD,** mede door jullie enthousiasme voor mijn onderzoeken en de bemoedigende woorden heb ik besloten om het PhD-traject in te gaan. Wij hebben het mooiste beroep ter wereld! En wat ben ik trots dat ik mag laten zien dat wij als verpleegkundig specialist zo'n divers beroep hebben en wat wij allemaal in onze mars hebben!

Lieve Paranimfen,

Lieve **Irene**, wat herinner ik mij nog goed de nachtdiensten samen op 2midden, waar er altijd wel iets gebeurde. Wij hadden hele gesprekken over de functie van verpleegkundig specialist. Dit was toch echt iets voor ons. Na pogingen bij de cardiologie kon jij starten bij de kinderchirurgie. Wat keek ik met bewondering naar je. Ik ben je, nog steeds, dankbaar voor je goede woorden over mij bij Hankje toen je hoorde dat ze een VS zochten bij de kinder-MDL. Wij werken niet op dezelfde afdeling, maar onze wegen blijven elkaar kruisen en wat is het dan heerlijk om met je samen te werken. Samen het Young Adult Programma vorm geven, VS-ers in het Sophia naar een hoger niveau tillen of samen een presentatie geven. Ook privé lopen onze levens aardig gelijk op; wij hebben ongeveer tegelijkertijd kinderen gekregen, wij hebben mannen die van klussen houden en wij kunnen zelf ook niet goed stil zitten. Het is heerlijk om tussen het werk door af en toe een bakkie te doen of lekker bij te kletsen bij een etentje. Wat ben ik ontzettend dankbaar dat jij tijdens mijn verdediging achter mij staat. Mocht ik een inzinking krijgen weet ik zeker dat jouw down-to-earth mentaliteit mij weer met beide voeten op de grond zet. Op naar nog vele jaren gezellige samenwerking en wie weet lees ik over een aantal jaren jouw dankwoord.

Lieve **Ilse**, toen ik door kreeg dat ik mijn proefschrift mocht gaan verdedigen wist ik gelijk dat jij sowieso achter mij moest komen te staan. Je bent zo'n belangrijke steunpilaar voor mij. Ik ben je dan ook zo dankbaar voor je vriendschap die als tiener is begonnen en nu zo'n stabiele basis is in mijn leven. Ik ben je dankbaar voor de avonturen die we samen hebben mogen beleven, jouw steun tijdens mooie en minder mooie momenten in het leven, de ontelbare gesprekken, vele wijntjes en dansjes. Wat ben ik ontzettend trots op jou, hoe jij alles combineert; gezin, man, werk, opleiding en dan nog zo'n geweldige vriendin zijn. Bedankt dat jij 21 mei achter mij wilt staan, dat geeft mij sowieso kracht!

Lieve familie,

Lieve Bro's en Tus; **Stijnie, Leendert** en **Anne**; onze familiemomenten zijn vaak een grote chaos, maar ik geniet ervan. Alles en iedereen bij en door elkaar. Fijn dat we nu minstens 1 keer per jaar met z'n vieren afspreken om echt bij te praten; dat moeten wij erin houden!

Ik ben jullie, samen met jullie partners **Wouter, Elzie en Hester** en **jullie kids**, dankbaar voor deze familiemomenten, maar ook jullie support en bewondering naar mij tijdens de uitvoering van dit traject.

Lieve **Yvette** en **Nils**; wat een feestje dat **Evy** er nu is, heerlijk die familiemomenten met elkaar. Bedankt voor jullie interesse in mijn promotietraject.

Lieve **Lies** en **Frans**; bedankt voor jullie oprechte interesse in mij en mijn werk. Jullie steun bij het klussen in huis en de vele oppasmomenten op de kids zijn onbetaalbaar.

Lieve **papa** en **mama**, wat ben ik dankbaar voor jullie geloof en vertrouwen in mij. Hierdoor zijn jullie op zoek gegaan naar de juiste mensen om mij heen zoals begeleiding bij mijn dyslexie. Mam, ik weet dat jij graag wilde dat de dyslexie mij professioneel niet zo in de weg zou zitten zoals het bij jou heeft gedaan. Dat hebben jullie perfect gedaan met jullie mentaliteit "waar een wil is, is een weg". Lieve papa, hoe jij mij hebt geleerd om soms dingen gewoon te doen en dan te zien waar het schip strandt heeft mij erg geholpen in deze reis. Gewoon starten met een artikel schrijven of dat praatje in het Engels houden voor een groep artsen en dan sta je "opeens" je proefschrift te verdedigen. Nog steeds voelt het erg ongeloofwaardig, maar ik heb het allemaal gedaan en kon het niet zonder jullie steun, vertrouwen en ontelbare hulp met de kids, het huis en zoveel meer. Ik ben jullie beide onwijs dankbaar wat jullie mij als ouders hebben meegegeven om de vrouw te worden die ik nu ben geworden.

Lieve **Veerle, Stef** en **Suze**; mijn kanjers! Wat ben ik ontzettend trots op jullie. Jullie hebben het niet altijd makkelijk met jullie drukke ouders. Dan zijn ze druk aan het klussen, dan weer druk aan het werk en dan weer op pad. Toch blijven jullie de leukste kinderen op aarde! Ik hou van jullie.

Lieve Veerle, wat vind ik het super lief dat je mij hebt willen helpen met het ontwerpen van de kaft van het boek. Wat bof ik toch met zo'n creatieve dochter!

Liefste **Niels**, mijn tegenpool, mijn rots in de branding, mijn thuis! Wat ben ik ontzettend dankbaar dat jij altijd aan mijn zijde staat. Jij remt mij op de juiste momenten af, maar je stimuleert mij ook om het beste uit mijzelf naar boven te halen. Ik weet dat menig mensen zich afvragen hoe wij alles voor elkaar krijgen met onze banen, dit promotietraject, jouw vrijwilligerswerk (ik noem het maar even zo, anders snappen ze het niet boven de rivieren) en ons droom (klus)huis. Maar zoals jij altijd zegt: "Gewoon stap voor stap, dan komt het vanzelf allemaal goed". Dat vind ik, best vaak, erg lastig. Wij 5-en tegen de rest van de wereld! Ik ben zo blij met jou en dankbaar dat jij mij elke dag eraan herinnert dat, ondanks het leven druk is, het belangrijk is om van het leven te genieten.

Het leven is een feestje, maar jij moet zelf de slingers ophangen!