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Efficacy of online group interventions for adolescents and parents

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Publication date
2023

Document Version
Final published version

Citation for published version (APA):
Douma, M. (2023). *Coping with pediatric chronic illness: Efficacy of online group interventions for adolescents and parents*. [Thesis, fully internal, Universiteit van Amsterdam].

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Miriam Douma

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Cover: Maya Schutte | www.mayaschutte.com
Layout: Ilse Modder | www.ilsemodder.nl
Printed by: Gildeprint Enschede | www.gildeprint.nl

ISBN: 978-94-6419-953-6

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ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad van doctor
aan de Universiteit van Amsterdam
op gezag van de Rector Magnificus
prof. dr. ir. P.P.C.C. Verbeek
ten overstaan van een door het College voor Promoties ingestelde commissie,
in het openbaar te verdedigen in de Agnietenkapel
op vrijdag 1 december 2023, te 13.00 uur

door Miriam Douma
geboren te ZWOLLE

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General introduction

An increasing number of children (age 0-18 years) in the Netherlands are growing up with a Chronic Illness (CI) (1). Due to increased medical knowledge and improved treatments, children and adolescents with a serious CI live longer (2, 3). Besides physical consequences, a pediatric CI causes psychological challenges. This requires the monitoring of psychosocial wellbeing for those growing up with a CI, and appropriate psychosocial care. Interventions to support children and their parents cope with the illness are essential to increase resilience and to avoid possible negative consequences, such as social or academic problems or the development of psychopathology (4-6).

The objectives of the studies described in this thesis were to develop and evaluate the '*Op Koers Online*' group intervention (in English: '*On Track Online*') for adolescents with a CI and parents of a child with a CI. This general introduction first describes psychosocial adjustment in adolescents dealing with a CI and in their parents, and discusses psychosocial interventions for them. Next, the *Op Koers Online* interventions for adolescents and parents and an outline of the thesis are presented.

PEDIATRIC CHRONIC ILLNESS AMONG CHILDREN AND ADOLESCENTS IN THE NETHERLANDS

A CI is defined according to the following criteria set forth by Van der Lee et al (2007) (1): 1) onset between ages 0 and 18; 2) diagnosis based on medical scientific knowledge; 3) the illness is not (yet) curable; and 4) the illness has been present for at least 3 months, or at least three episodes have occurred in the last year (1). Approximately 14% of the children in the Netherlands suffer from a CI (7). In the Netherlands, the most common diagnosed illnesses in childhood are asthma (10% of all CI) and eczema (6%). Other moderately prevalent illnesses (<1%) are diabetes mellitus type 1, epilepsy, juvenile idiopathic arthritis (JIA), sickle cell anemia, cystic fibrosis, hemophilia and inflammatory bowel diseases. There are also many rare diseases, such as kidney diseases, metabolic diseases and congenital heart diseases (8). Many different CI diagnoses can be distinguished, with different physical symptoms and complaints. However, the psychosocial consequences for children, adolescents and their parents appear to be comparable across illnesses (3, 5, 9). Families dealing with a pediatric CI may benefit from psychosocial support in coping with the CI. This is provided in pediatric care and concerns a child life specialist to offer support to children at the hospital (e.g. prepare for medical treatments), social work to support parents with practical issues and medical psychologists for psychological support for both children and parents when necessary.

PSYCHOSOCIAL ADJUSTMENT AND INFLUENCING FACTORS

Adjustment and theoretic model

A pediatric CI affects not only the child with the illness, but the whole family. In the first place, children and adolescents with a CI themselves have to face difficulties related to their illness, such as hospitalization, the use of medication, restrictions in activities and stressors related to the course of the illness and the future (10). Growing up with a CI influences psychosocial wellbeing and the development of cognitive and social skills (11, 12). Children and adolescents growing up with a CI do not necessarily develop psychopathology, but they are constantly confronted with the stressors and related restrictions such as not being able to join peers and insecurity about the course of the illness. Especially during adolescence (12-18 years), with the formation of identity, self-image and self-esteem, a CI constitutes a major challenge (6, 13). Adolescents growing up with a CI are therefore at risk for emotional- and social problems such as feeling down and isolating oneself (12, 14). Several meta-analyses have found elevated levels of internalizing and externalizing problems in children and adolescents growing up with a CI (10, 15-17). Considering this, interventions that support adolescents with a CI and teach them how to cope with their illness are essential.

Research shows that pediatric CI influences psychosocial wellbeing in parents as well (18, 19). Hatzmann et al (2009) (20) showed that 45% of the 533 participating parents were at risk for health-related quality of life impairment. For parents, learning that their child has a chronic and potentially life-threatening illness is a very stressful and potential traumatic event (21-23). Parents can face several emotional struggles such as insecurity about the future and feelings of guilt and sorrow (23, 24). When children grow up, parents are predominantly responsible for managing the child's illness. They are confronted with stressors about their child's health as well as logistical and practical factors such as managing daily routines, relationships with other family members, the balance between family and work and possible financial problems (18, 25). Parents, mothers in particular, are disadvantaged in society probably due to the challenge of combining a child with work and leisure time (26). As a consequence, parents are at risk for sorrow and psychosocial distress (25). Parents who face stress are less able to manage the child's illness effectively (18, 23, 27). Moreover, parental depressive symptoms have found to be correlated to negative parenting practices (28), poor adherence (29) and an increase in children's illness symptoms over time (30-34).

Transactional models of child adaption to CI recognize the importance of numerous psychosocial risk- and protective factors that could be targeted and modified in interventions. The disability-stress-coping model of Wallander and Varni (1998) implies that the stressors faced by children and parents dealing with pediatric CI are multifaceted (e.g. restriction of activities, responsibility of treatment), and that the links between illness related stressors and adjustment are moderated by appraisals and coping strategies on which several personal and family risk- and protective factors are of influence (21, 35). Someone's coping has a central place in Wallander's model, moderating the effects of influencing factors. In Figure 1 the model is described in the context of our studies. In this thesis, we will focus on psychosocial functioning of **adolescents with a CI (aged 12-18 years)** and **parents of children with a CI (aged 0-18 years)**.

Coping

Coping consists of cognitive and behavioral efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of a person (36, 37). Someone's coping is known as the way people react to stressful situations. There is a distinction between engaged and disengaged strategies. Engagement refers to approach strategies (e.g. problem solving, cognitive restructuring), whereas disengagement refers to avoidant strategies (e.g. self-criticism, social withdrawal) (38). There is considerable evidence that engaged coping is related to better adjustment to CI. For example, problem solving and cognitive restructuring have been associated with better psychological outcomes (39, 40). Second, there is evidence that disengaged or passive coping is related to poorer adjustment. For instance, self-criticism and social withdrawal are strongly related to anxiety or sadness among children with a CI (11).

Coping style is not fixed within an individual and can vary in expression across situations, however people tend to apply the same strategies. When engaged coping can be reached, parents and adolescents will likely have better psychosocial adjustment and outcomes and will likely be more capable of managing their or their child's illness effectively (11, 34, 41). As shown in Figure 1, coping has a central place in the model. According to the model, someone's coping strategies can moderate the effect of influencing factors on the outcome psychosocial adjustment. Therefore, it is important to pay attention to coping of adolescents and parents dealing with a CI.

Influencing factors

According to the model of Wallander and Varni (Figure 1), personal, family and environmental factors are, via coping strategies, of influence on psychosocial adjustment for adolescents and parents dealing with a CI.

First, *personal factors* are characteristic factors of the adolescent or the parent, such as temperament and competence. For example, an introvert person will be more likely to worry a lot (42, 43).

Second, *family factors* can impact the psychosocial adjustment of both adolescents and parents to the CI, directly and indirectly via coping. Adolescents who grow up in a warm and supportive family will likely use more engaged coping strategies in situations concerning their CI compared to adolescents living in an unsecure family (34, 44). Within family factors, the parent-child relationship is particularly important (45). For parents, practical stressors such as managing daily routines in the family and taking care of possible other children are of influence on parental psychosocial adjustment to the CI. Dividing attention between the child with a CI and healthy sibling(s) in the family is known as a difficult challenge for parents. Furthermore, relationship with the (ex-)partner can play a role in how well parents adjust to the situation. Parents who feel supported in their relationship will likely be more resistant to psychosocial distress (20, 46).

Third and last, *environmental factors* are of influence on psychosocial outcomes. For adolescents, contact with peers and peer support are important for development and psychosocial adjustment (31). When support from peers is lacking, adolescents can feel different, lonely and demotivated which is a risk factor for passive coping (12, 31, 47). For parents, being and feeling supported is a protective factor for good psychosocial adjustment. Another environmental factor is for example communication with parent's work and school of the children. When parents do not feel understood and supported at work, it can be a stress factor in addition to the already existing distress (26, 46, 48).

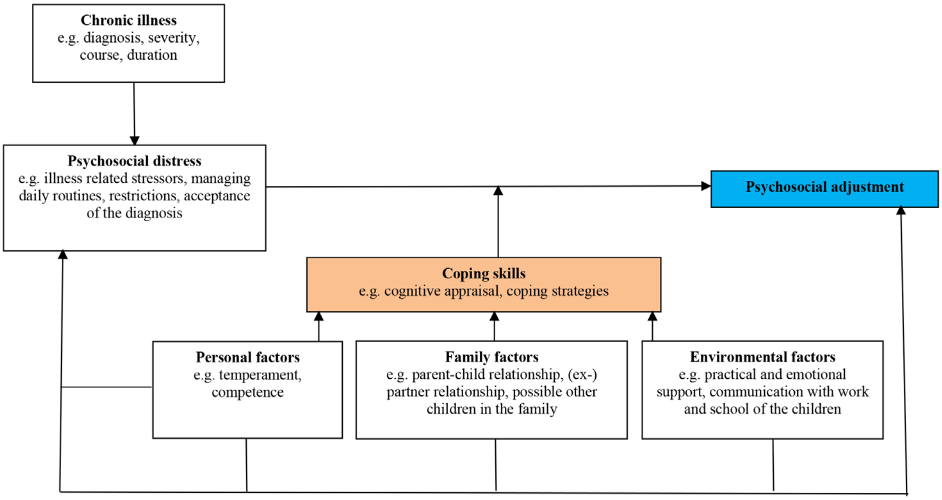


Figure 1 Adapted version of the disability-stress-coping model of Wallander and Varni (1998) to explain psychosocial adjustment in adolescents with a CI and parents of a child with a CI.

PSYCHOSOCIAL INTERVENTIONS FOR ADOLESCENTS AND PARENTS DEALING WITH CI

Content

As stated above, adolescents and parents confronted with the adolescents' CI are at risk for psychosocial problems. To prevent and/or reduce these problems in adolescents as well as parents, interventions focusing on how to cope with stressors caused by the CI are needed. Over the past few years, numbers of psychosocial interventions for children with a CI and their families have been developed and evaluated (49-51). Most interventions described in the literature are psycho-educational in character, usually involving combinations of information- and coping skill-training interventions (52-54).

Cognitive behavioral therapy (CBT) and acceptance and commitment therapy (ACT) are examples of therapies to teach the use of active coping strategies. CBT and ACT as individual therapies have been shown effective in improving psychological outcomes such as stress- and health-related quality of life (HRQoL) of children and adolescents with a CI and their families (55-57). CBT involves identifying unhelpful thoughts, challenging them, and replacing them with helpful thoughts. It also teaches coping and problem-solving skills (58). ACT, as a part of CBT, aims to reduce the influence of negative thoughts on daily life by accepting certain thoughts or situations. Exercises such as mindfulness and values elicitation to orientate participants to thoughts and activities which are in line with personally meaningful values are often used (59-61). ACT could be helpful for parents of a child with a CI, since those parents often face situations which cannot be changed and ACT provides helpful tools to learn how to deal with such situations.

Intervention formats

Interventions can be offered in individual and group format. Sharing experiences with others in a similar situation is associated with a decrease of distress and improvement of mental health for both adolescents and parents (62, 63). Offering group interventions has the additional benefit of treating more patients simultaneously and is therefore an (cost-)effective way to offer support (Heath et al, 2018). Moreover, group interventions are proven to be effective in teaching coping skills and improving knowledge about symptom reduction and disease-related problem solving (64, 65).

Most existing (CBT) group interventions for adolescents and parents are focused on a specific illness (49, 51, 66), such as epilepsy (67) or diabetes (68, 69). However, as stated before, most of the psychosocial problems are the same across illnesses. A generic approach that focuses on psychosocial problems associated with the CI rather than the CI itself is therefore suitable and would allow for patients and parents with rare illnesses to participate in a group intervention. Furthermore, most existing interventions which involve parents are child-focused: the parents learn how to support their child in managing the child's illness (35, 55). During the past years, there is more attention to offer psychosocial support to parents themselves (70).

Online based interventions are upcoming and recent results on feasibility and efficacy are promising (59, 66, 71-74). An online intervention format eliminates barriers that adolescents and parents can experience for participation in a face-to-face intervention. Adolescents can face difficulties with visiting the hospital for an intervention in addition to their regular hospital visits. Parents have to deal with practical barriers such as managing daily routines at home, work and taking care of the child or multiple children. Online interventions can overcome these barriers: participants do not need to be physically present to participate in an intervention but can stay at home, which is less time consuming. In some online interventions, it is possible to participate anonymous which can be extra appealing to participants.

To our knowledge, accessible internet-based group interventions for adolescents and parents dealing with different diagnosis of CI were not available until now. The already existing face-to-face intervention *Op Koers* was translated into an online version to fill this gap. However, information about efficacy is lacking. Most existing studies focus on for example a specific CI or on different groups of patients/people (73, 75). In the current thesis, we focused on the development and efficacy of two unique online CBT- and ACT-based psychosocial group interventions called *Op Koers Online* for adolescents and parents dealing with CI.

DEVELOPMENT OF THE OP KOERS ONLINE INTERVENTIONS

In the past 25 years, different modules of the *Op Koers* program were developed and evaluated. *Op Koers* was first developed for children and adolescents in a face-to-face format. *Op Koers* uses CBT techniques to teach participants to use engaged coping skills to prevent and/or reduce psychosocial problems (35, 76). A randomized controlled trial (RCT) of *Op Koers* face-to-face showed that the intervention had a positive effect on children's psychosocial adjustment and wellbeing (35). Involving parents in the intervention (called *Together Op Koers)* for the children and adolescents contributed to the persistence of the effects. The intervention for parents focused on the child: parents participated in parallel groups and learned what their children were learning to support them in daily life.

Op Koers face-to-face delivery involves weekly sessions at the hospital. Scholten et al (2013) noticed that adolescents were somewhat hesitant to participate in the intervention in the hospital. Coming to the hospital for an intervention in addition to their regular appointments in the hospital was a barrier for them. This outcome, and today's digital possibilities, have led to the development of *Op Koers Online*. The intervention was first designed for adolescent survivors of childhood cancer (*Op Koers Online Oncology).* A feasibility study shows promising results: participants and course leaders reported high levels of satisfaction and the dropout rate was very low (77). Most participants considered chatting appropriate for the intervention and reported to prefer *Op Koers Online* above *Op Koers face-to-face*. After the feasibility study, the intervention was optimized based on feedback from participants and course leaders. Changes during this optimization include increasing the number of sessions from six to eight, composing groups with participants in the same age category (as much as possible, depending on applicants) and excluding individuals with severe learning difficulties (77).

After the optimization of *Op Koers Online Oncology, Op Koers Online* for adolescents (aged 12 to 18 years) with CI was developed. With this internet-based intervention more flexible participation in a group intervention without an additional hospital visit (35) was offered. However, it was unknown whether the online intervention would be feasible and efficacious, in line with the face-to-face intervention. Research concerning those questions is presented in the current thesis.

Following up on the study of Scholten et al (2013) there was a wish to develop an accessible internet-based intervention for parents as well. To design an intervention according to parental support needs, insight in their needs such as the themes to address and preferences for intervention format was necessary. This thesis provides a support needs study which was conducted in order to develop the intervention for parents, following by a study to evaluate efficacy.

RESEARCH QUESTIONS, DESIGNS AND HYPOTHESES

This thesis aims to answer different research questions focusing on *Op Koers Online* for adolescents and parents separately. The first research question was: **is *Op Koers Online* for adolescents effective in enhancing engaged coping and in preventing and/or reducing emotional- and behavioral problems?** This question is answered with two studies, first of which the pilot study. With a pretest-posttest questionnaire design without a control group we evaluated the feasibility and explored the preliminary effectiveness of *Op Koers Online* for adolescents. Adolescents participating in *Op Koers Online* were asked to complete questionnaires at baseline and post-intervention. The hypothesis was that *Op Koers Online* for adolescents was feasible and preliminary effective. Second, an RCT was conducted to assess the efficacy of *Op Koers Online* for adolescents. The RCT had an intervention group and a waitlist (control) group. When assigned to the waitlist group, participants received care-as-usual and were for ethical reasons not prevented to seek for individual psychosocial treatment. Additional psychosocial care was monitored in both study groups and controlled for in the analyses. Participants in both study groups completed questionnaires at baseline, six- and twelve months follow-up. After completing the study, participants in the waitlist group were given the opportunity to participate in the intervention. The hypothesis was that *Op Koers Online* for adolescents has a positive effect on emotional- and behavioral problems and the use of engaged coping skills.

The second research question focused on the development of the intervention for parents: **what are parental support needs?** An explorative support needs study was conducted to 1) explore which themes are important for parents to address in an intervention, 2) determine what type of psychosocial intervention parents would like and 3) assess parents' practical preferences for an online group intervention. To require this information, parents were asked to fill out a support needs questionnaire. Additional focus groups were held to receive more in-depth information of what an intervention should look like according to parents. The ultimate goal was to develop a suitable intervention for parents based on their support needs. The hypothesis was that parents would be interested in a group intervention focusing on their own functioning (different themes) instead of focusing mainly on their child's functioning, and that parents would prefer an online intervention above a face-to-face format due to accessibility.

The third research question was: **is *Op Koers Online* for parents effective in enhancing engaged coping skills and in preventing and/or reducing emotional problems?** An RCT with an intervention and a waitlist (control) group was conducted to assess the efficacy of *Op Koers Online* for parents. Similar to the RCT for adolescents, participants assigned to the waitlist group received care-as-usual and were for ethical reasons not prevented to seek for individual psychosocial treatment. Additional psychosocial care was monitored in both study groups and controlled for in the analyses. Participants in both study groups completed questionnaires at baseline, six- and twelve months follow-up. After completing the study, participants in the waitlist group were given the opportunity to participate in the intervention. The hypothesis was that *Op Koers Online* for parents has a positive effect on symptoms of anxiety and depression and in enhancing engaged coping skills.

The final question was; **what are the lessons learned after 25 years of intervention development?** This study aimed to share the important lessons in the process of development, research and implementation of *Op Koers (Online)* group interventions for children with illness and their family members. Using the National Institutes of Health Stage Model for Behavioral Intervention Development the activities in the different stages were critically appraised.

Op Koers Online for adolescents and parents provided in the RCTs

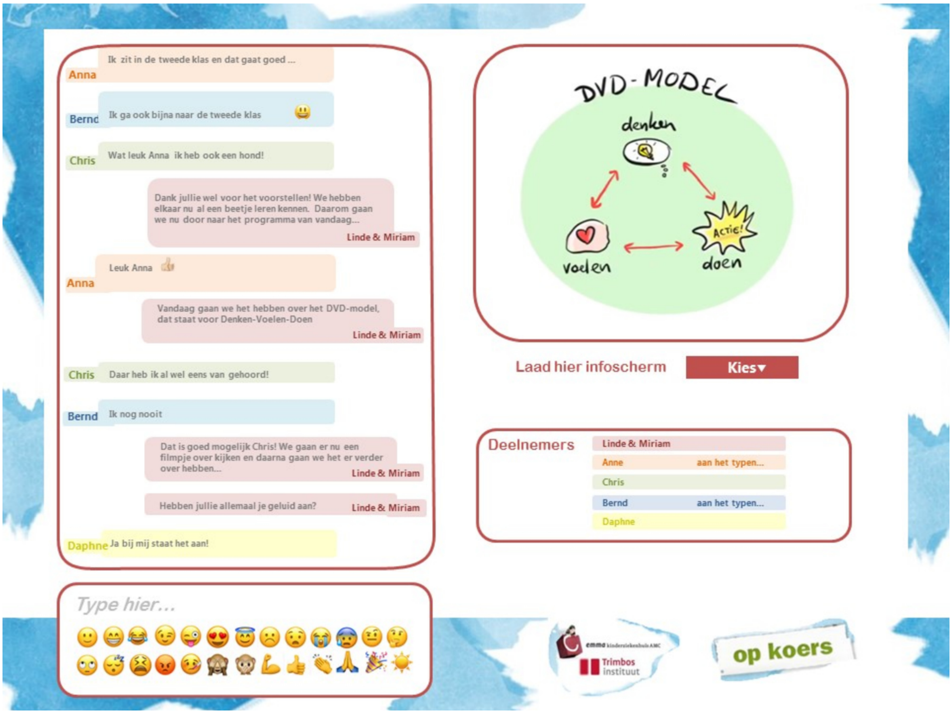
In *Op Koers Online*, weekly sessions take place at a scheduled time in a secured chatroom (Figure 2) with three to five participants under supervision of two psychologists (course leaders) who carry out the protocolled intervention. The interventions were intentionally designed without a webcam to ensure anonymity and keep the threshold for participation as low as possible. CBT and ACT techniques such as relaxation, cognitive restructuring and exercises focusing on accepting the (child's) diagnosis, are used. Central in the interventions is the Thinking-Feeling-Doing model (TFD model). With this model, course leaders teach participants the relationship between what people think, feel and how they act, and how they can influence their thoughts feelings and behaviors. Another important part of *Op Koers Online* is sharing experiences with others in a similar situation.

The intervention for adolescents consists of eight weekly 90-minutes sessions and a booster session four months after the last regular session. In the first session there is time for getting to know each other and explanation about the intervention. During the following sessions, adolescents lean how to use these five adaptive coping skills: 1) information seeking and providing about the illness, 2) use of relaxation techniques in stressful situations, 3) increasing knowledge of self-management and medical compliance, 4) improving social competence and 5) positive thinking (cognitive restructuring). The learning goals of the intervention and examples of learning activities (e.g. homework and group discussion) are shown in Table 1.

The intervention for parents consists of six weekly 90-minutes sessions and a booster session four months after the last regular session. In the first session parents get to know each other and receive explanation about the intervention. Figure 3 is introduced to parents in the first session and illustrates the parent (in the middle) and their environment around them (different layers). The figure is shown in every session to show which theme/layer is central in that session and to show parent an overview of the intervention content. The following sessions each focus on a theme. Table 2 shows the themes, group discussion subjects and examples of homework assignments of *Op Koers Online* for parents. In the sixth and last session there is time to repeat topics or to resume discussions that have not been finished due to lack of time. The coping skills, which were addressed in every session through the different themes, are 1) use of relaxation during stressful situations, 2) positive thinking, 3) social support (seeking for and accepting support) and 4) open communication about the illness.

OUTLINE THESIS

In the next chapter, **Chapter 2** the pilot phase of *Op Koers Online for adolescents* is described. In **Chapter 3** the development of *Op Koers Online for parents*, including a support needs study among parents, is presented. **Chapter 4** presents the study protocol of both RCT's (adolescents and parents) including the rationales, contents and designs of the studies. The results of the RCT on the efficacy of *Op Koers Online for adolescents* is presented in **Chapter 5**, followed by the results of the RCT on the efficacy of *Op Koers Online for parents* in **Chapter 6**. **Chapter 7** provides an overview of the whole *Op Koers project* starting twenty-five years ago, including critically reflections and lessons learned. This thesis ends with a general discussion in **Chapter 8**. Reflections on the results, recommendations for future studies and clinical implications are given.



Anna Ik zit in de tweede klas en dat gaat goed ...

Bernd Ik ga ook bijna naar de tweede klas 😃

Chris Wat leuk Anna ik heb ook een hond!

Dank jullie wel voor het voorstellen! We hebben elkaar nu al een beetje leren kennen. Daarom gaan we nu door naar het programma van vandaag... **Linde & Miriam**

Anna Leuk Anna 👍

Vandaag gaan we het hebben over het DVD-model, dat staat voor Denken-Voelen-Doen **Linde & Miriam**

Chris Daar heb ik al wel eens van gehoord!

Bernd Ik nog nooit

Dat is goed mogelijk Chris! We gaan er nu een filmpje over kijken en daarna gaan we het er verder over hebben... **Linde & Miriam**

Hebben jullie allemaal je geluid aan? **Linde & Miriam**

Daphne Ja bij mij staat het aan!

Deelnemers

Linde & Miriam	
Anne	aan het typen...
Chris	
Bernd	aan het typen...
Daphne	

Figure 2 Chatroom of *Op Koers Online*. Left screen: chat text; right screen: information screens/ videos/exercises; strip below (left): field where participant writes their text with possible use of the emojis; strip below (right): names of participants who are present in the chatroom. Note: These participants and text in this chat room are fictitious.

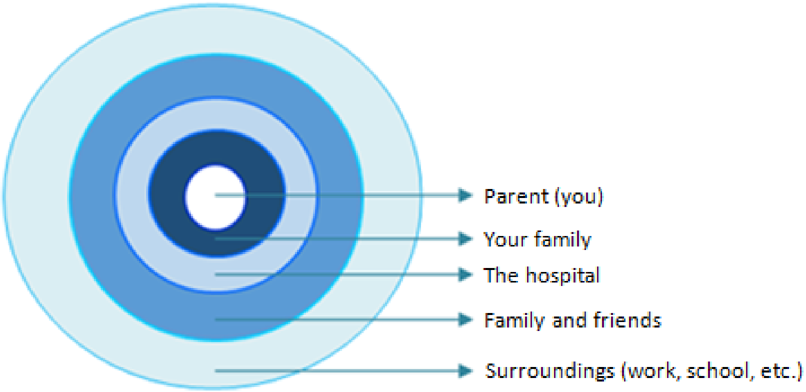


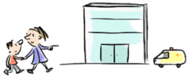
Figure 3 Illustration of the parent in the middle, and layers around the parent starting with their family (close by), the hospital, family and friends and environment (work, school).

Table 1 Learning goals and examples of learning activities and reinforcement/practice of *Op Koers Online* for adolescents

	Learning goals				
	Information seeking and giving about the illness	*Use of relaxation during stressful situations*	*Increase knowledge of self-management and compliance*	*Enhancement of social competence*	*Positive thinking (cognitive restructuring)*
Session	Session 2	Session 3	Session 4	Session 5 and 6	Session 1, 7 and 8
					
Instruction/ modelling	Relaxation exercise (audio fragment)	Education about sources of information	Group discussion about own treatment and (non-)compliance	Video and group discussion: how and what do you tell others about your illness	Thinking-Feeling-Doing game
Reinforcement/ practice (homework)	Practice the relaxation exercise	Write down questions you have, and look for answers	Write down situations for non-compliance and how to improve compliance	Think of what CAN you do (instead of CANNOT) and write down your story for the other group members	Write down positive adjustments for negative thoughts

[1] The session that focused on the specific learning goal was mentioned here. However, learning goals are applied throughout the whole course (for example: use of relaxation techniques and Thinking-Feeling-Doing are discussed in multiple sessions).

Table 2 Themes, group discussion and examples of homework assignments of *Op Koers Online* for parents.

	Theme [1]					
	Getting to know each other	*The CI of the child*	*Relationship within the family*	*Taking care of yourself besides caregiving tasks*	*Relationship with others and practical support*	*Time for repetition or resumptions*
Session	Session 1 *"Getting to know each other"*  	Session 2 *"The hospital"* 	Session 3 *"The family"* 	Session 4 *"Taking care of yourself"* 	Session 5 *"Extended family and friends"*  	Session 6 *"In retrospect"* 
Group discussion [2]	Getting to know each other, explanation of Thinking-Feeling-Doing model	Accepting the diagnosis, how to support the child, ssuccesses and struggles in medical treatment of the child and how to handle difficult situations in the hospital	What is the impact of the CI on the child with CI, siblings, the relationship with your partner? What are successes and struggles in your family?	What is the impact of the CI on your own life and emotional functioning? How do you take care of yourself?	What kind of support would you like to receive/do you receive? What kind of reactions do you get and how does that feel? What works or could be better in the communication with work/school?	Looking back at the previous sessions, time to repeat topics or to resume discussions that have not been finished due to lack of time
Examples of homework assignments	Fill out the Thinking-Feeling-Doing model for specific situations in daily life	Reading a story about how to cope with the diagnosis & together with your child, make a list of situations for adherence/ non-adherence and discuss how to reach adherence	Talk with siblings about worries they have, make a list of things to do with siblings and your partner and have quality time with them each	Practice with the relaxation exercise, take time for yourself, give yourself a compliment daily	Together with your partner, write down difficult questions/ reactions you get from others and think of possible ways to react on that	None

[1] All themes are linked to a session, however, specific content is determined by parents in every session (what they want to discuss).

[2] CBT techniques are used throughout the whole intervention

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2

Online cognitive-behavioral group intervention for adolescents with chronic illness: A pilot study

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Clinical Practice in Pediatric Psychology

ABSTRACT

Objective To assess feasibility and explore preliminary effectiveness of an online cognitive-behavioral group intervention (*Op Koers Online*) to prevent and/or reduce psychosocial problems by teaching active use of coping skills to adolescents (ages 12-18) with chronic illness.

Methods Adolescents who signed up for the chat intervention were asked to complete online questionnaires at baseline and post-intervention. Feasibility was evaluated based on attendance (missed sessions, dropout rate, homework completion), technological issues and with an evaluation questionnaire. Preliminary effectiveness was evaluated with standardized questionnaires: *Op Koers Online Questionnaire* (disease-related coping skills), *Youth Self Report* (emotional and behavioral functioning), *PedsQL* (Health-Related Quality of Life, HRQoL). Mean scale scores post-intervention were compared to baseline with paired-samples t-tests. Effect sizes were calculated.

Results In total, 33 adolescents participated in the intervention, 29 adolescents completed the baseline questionnaires. Regarding post-intervention questionnaires, 25 adolescents completed the evaluation questionnaires, 23 adolescents completed all questionnaires post-intervention. Dropout rate was 6%. In one session (2 %), there were technological issues that caused the session to stop. Participants' overall satisfaction was high. Regarding effectiveness, participants improved significantly in the use of total coping skills and the coping skills "information seeking and giving" and "social competence" after the intervention compared to baseline. Participants also reported significantly fewer externalizing problems and scored significantly better on total quality of life and emotional and psychosocial HRQoL after following the intervention.

Conclusions In conclusion, *Op Koers Online* is feasible and potentially effective. Further research (an RCT) is needed to establish the effects of the intervention.

Implication for impact statement This study evaluates the feasibility and preliminary effectiveness of a new and unique internet-delivered intervention for adolescents with chronic illness: *Op Koers Online*. The findings indicate that this intervention is feasible (based on good attendance, limited technological issues and positive overall evaluation of the intervention) and show preliminary effectiveness (based on improvements in disease-related coping skills and psychosocial functioning). In conclusion: *Op Koers Online* is a promising intervention. Our findings emphasize the growing evidence for internet-based interventions and their suitability for adolescents. The importance of teaching adolescents how to use active coping skills with CBT techniques is also highlighted.

INTRODUCTION

The number of children and adolescents living with chronic illness (CI) continues to grow due to increased medical knowledge and improved treatments (1, 2). In the Netherlands, 15-20% of children (ages 0-18) are living with a CI such as diabetes or asthma (3). Children and adolescents with CI have to live with physical consequences and face difficulties such as hospital visits and/or hospitalizations, activity restrictions and illness-related stressors such as uncertainty about the course of their illness. The stressors are multifaceted and mostly for a lifetime (4).

Children and adolescents with CI show higher levels of stress, internalizing behavior problems and somewhat elevated levels of externalizing behavior problems compared to healthy peers (4-6)[4, 5]. They do not necessarily develop psychopathology, but are constantly confronted with the stressors. Especially during adolescence, with the formation of identity, self-image and self-esteem, a CI constitutes a major challenge (7, 8). In recent years, psychosocial interventions have become increasingly important in the treatment of psychosocial problems (social, emotional and behavioral problems such as loneliness, depression, aggressive behavior) in adolescents with CI (4, 9). The psychosocial interventions discussed in the literature are mostly disease-specific (10). For example, a cognitive-behavioral therapy for adolescents with inflammatory bowel disease (IBD) showed decreased depressive symptoms in participants after treatment (11) and a psychosocial group intervention for young people with epilepsy (PIE) was effective in teaching participants about their illness, how to talk about it and how to cope with difficult feelings (12). Interventions are available in face-to-face as well as online format. Online interventions are upcoming due to new technologies (9, 13). An online psychosocial intervention could be a cost-effective way to offer therapy (9, 14, 15).

According to the disability-stress-coping model (16), stressors related to illness and psychosocial adjustment of the child are moderated by coping strategies and cognitive appraisals. The model states that the use of more effective coping strategies can prevent or reduce psychosocial problems in children with CI. Moreover, effective use of coping skills increases patients' abilities to manage illness by improving medical compliance and psychosocial functioning (4, 17-19). Active coping strategies (e.g. cognitive restructuring, relaxation), learned by transforming negative thoughts into positive, more proactive ones, are proven to be more effective than passive, avoidant coping strategies (19). To prevent and/or reduce psychosocial problems, appropriate interventions to teach adolescents how to cope with stressors caused by the CI are essential.

Interventions can be offered in individual or group format. Results on the effectiveness of group interventions are promising, particularly on learning to use more active coping skills and improving knowledge about symptom reduction and disease-related problem-solving (20). Most group interventions are focused on a specific illness, such as diabetes (21). Compared to individual interventions, psychosocial group interventions enable participants to share emotions and experiences and therapists can treat more patients simultaneously (22). Sharing emotions and experiences is helpful for adolescents with CI, as peer relationships can positively affect social adjustment and adaption to the disease (7, 20). This is illustrated by the iPeer2Peer program, where adolescents with Juvenile Idiopathic Arthritis (JIA) are matched to a trained 'peer mentor' for receiving peer support via Skype video calls. Thanks to the online format, the intervention is easily accessible and participants show improvements in perceived ability to manage JIA (23). In summary, most psychosocial group interventions for children with CI focus on a specific illness. However, even though different diagnoses may have different medical treatments, several of the psychosocial problems are the same (20). With a generic approach, it is easier for patients with rare illnesses to participate in a group intervention. The group intervention *Op Koers* (in English: "*On Track*") was designed with this in mind (24, 25).

Op Koers was primarily developed in a face-to-face format. Through cognitive-behavioral therapy (CBT) techniques, participants are taught to use active coping skills to prevent and/or reduce psychosocial problems (24-26). CBT focuses on recognizing cognitive distortions and on teaching coping and problem-solving skills (27). Sharing experiences with fellow patients is an important part of the intervention (20, 28, 29). In a Randomized Controlled Trial (RCT) of *Op Koers* face-to-face, positive effects were found on psychosocial functioning (26). The intervention has weekly sessions at the hospital, which can cause logistical barriers for potential participants.

Online intervention programs eliminate logistical barriers such as travel time and distance (30, 31) and connect to the digital environment in which adolescents live. Offering online interventions has additional advantages of improved accessibility, independence (participation from home) and anonymity (possibility to participate with a nickname). Online interventions without use of a webcam adds the benefit that appearance plays no role which might make it easier to talk about problems (32, 33). Research has also shown that e-health interventions eliminate the stigma related to participating in mental health services and therefore lower the threshold for participation (34). To this end, *Op Koers* face-to-face was translated into a chat version: *Op Koers Online*. The intervention was first designed for adolescent survivors of childhood cancer (*Op Koers Online Oncology).* A feasibility study shows promising results: participants and course leaders reported high levels of satisfaction and the dropout rate was very low. Most participants considered chatting appropriate for the intervention and reported to prefer *Op Koers Online* above *Op Koers* face-to-face. The intervention was optimized based on feedback from participants and course leaders, the number of sessions was expanded from six to eight, arranging the online intervention by age was recommended and severe learning difficulties became an exclusion criteria for participation (33). *Op Koers Online* for adolescents (ages 12-18) with CI was subsequently developed.

The aim of this pilot study was to assess feasibility and explore preliminary effectiveness based on disease-related coping skills and psychosocial functioning (emotional/behavioral problems and Health-Related Quality of Life; HRQoL) of *Op Koers Online* for adolescents with CI.

METHODS

Study design

This quasi-experimental, pre-post intervention pilot study was conducted between October 2013 and September 2015. Participants were asked to complete online questionnaires before the intervention (baseline; T0) and directly after the intervention (after eight weeks; T1). Participants received an e-mail with a hyperlink to the questionnaires and, when necessary, electronic and/or telephone reminders.

Procedure

Participants were recruited via; 1) healthcare providers, 2) folders at the hospital, and 3) online advertising. A pediatric psychologist informed interested adolescents and parents about the procedure and intervention by phone. Adolescents and parents willing to participate were asked to return the signed Informed Consent form sent by mail. Approval of the Medical Ethical Committee of the Amsterdam University Medical Centers was obtained to conduct this pilot study.

Participants

Inclusion criteria were: age between 12-18 years; CI diagnosis according to the definition of Van der Lee et al.: 1) onset between ages 0 and 18, 2) diagnosis based on medical scientific knowledge, 3) the illness is not (yet) curable, and 4) the illness has been present for at least three months, or at least three episodes have occurred in the last year (3), and receiving treatment in the Emma Children's Hospital (Amsterdam University Medical Centers). Participants also had to have access to a computer with internet connection, be able to read and write in Dutch, and independently complete the questionnaires. Adolescents with severe learning difficulties were excluded.

Intervention

Goal of the intervention is to prevent and/or reduce psychosocial problems by teaching the use of active coping skills (Table 1). Five coping skills are taught with CBT techniques (e.g. relaxation, cognitive restructuring and social skills) (35, 36): 1) information seeking and giving about the illness, 2) use of relaxation techniques in stressful situations, 3) increasing knowledge of self-management and medical compliance, 4) improving social competence and 5) positive thinking (24, 25). The Thinking-Feeling-Doing (TFD) model is used to explain how thought influences feelings and behavior. The focus lies on restructuring negative thoughts about the illness such as worrying about participating in or missing school/sports activities, worrying about what others think of you, etc.. Lastly, psychoeducation is used to expand participants' knowledge about the topics used in the intervention, e.g. about sources of information and compliance/noncompliance.

The intervention consists of eight weekly 90-minute sessions that take place at a set time in a secured chatroom (www.opkoersonline.nl) with groups of three to six participants. Participants log on to the website to enter the chatroom (Figure 1) and their personal online environment, where they can view intervention material and submit homework assignments. Sessions are led by two pediatric psychologists (course leaders), who received extensive training in carrying out the intervention based on a detailed manual. To improve adherence, participants receive a small gift (like a memory game) after the last session for participating and completing homework assignments. Four months after the last session, there is a booster session.

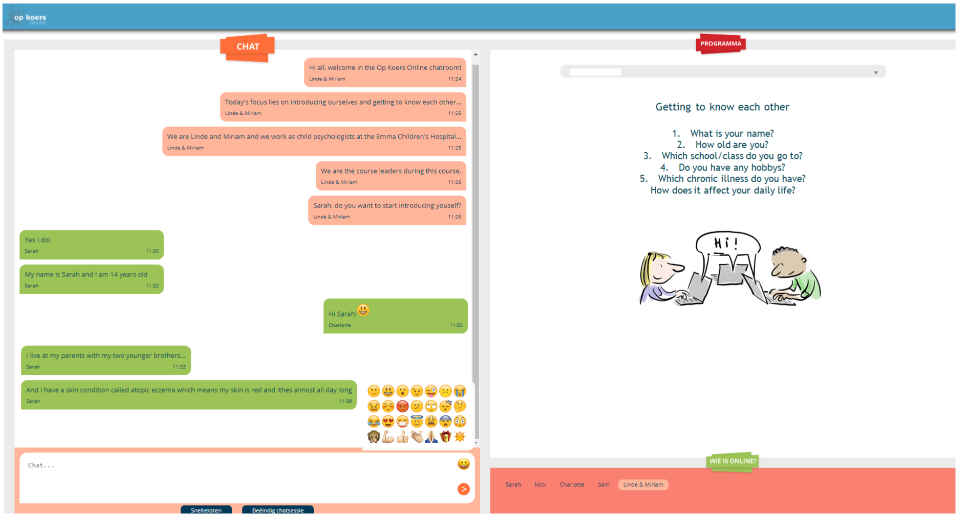
The protocol of *Op Koers Online* protects anonymity. First, participants are asked not to share contact details with each other until the last session. This way, communication between participants during the intervention elapses in the presence of the course leaders. Second, the intervention is designed without a webcam for purposes of anonymity and to keep a low threshold for participation (as participants do not need a webcam).

Table 1 The five basic learning goals of '*Op Koers Online*' and examples of learning activities

		Examples of learning activities	
	Learning goals	**Instruction/modelling**	**Practice**
1	***Information seeking and giving about the illness***	Education about sources of information	Write down questions you have and look for answers
2	***Use of relaxation during stressful situations***	Relaxation exercise (MP3 fragment)	Practice the relaxation exercise
3	***Increase knowledge of self-management and medical compliance***	Group discussion about own treatment and compliance/ noncompliance	Write down situations of noncompliance and how to improve compliance
4	***Enhancement of social competence***	Video and group discussion: how and what do you tell others about your illness	Think of what you CAN (instead of CANNOT) do and write down your story for the other group members
5	***Positive thinking***	Thinking-Feeling-Doing game	Write down positive adjustments for negative thoughts

Measures

Sociodemographic (e.g. gender, age, school-related) and medical information was obtained from adolescents via an online questionnaire. First, feasibility was measured based on attendance (dropouts, missed sessions and homework completion) and technological issues. Online presence was recorded based on entering the chatroom during the session. Participants who explicitly quit the intervention or were absent for four or more sessions were considered dropouts. Course leaders checked if participants completed their homework assignments and recorded technological issues and other particularities in every session log.



Hi all, welcome in the Op Koers Online chatroom!
Linde & Miriam 11:24

Today's focus lies on introducing ourselves and getting to know each other...
Linde & Miriam 11:25

We are Linde and Miriam and we work as child psychologists at the Emma Children's Hospital...
Linde & Miriam 11:25

We are the course leaders during this course.
Linde & Miriam 11:26

Sarah, do you want to start introducing youself?
Linde & Miriam 11:26

Yes I do!
Sarah 11:30

My name is Sarah and I am 14 years old
Sarah 11:32

Hi Sarah! 😃
Charlotte 11:32

I live at my parents with my two younger brothers...
Sarah 11:33

And I have a skin condition called atopic eczema which means my skin is red and ithes almost all day long
Sarah 11:39

Getting to know each other

1. What is your name?
2. How old are you?
3. Which school/class do you go to?
4. Do you have any hobbys?
5. Which chronic illness do you have?
How does it affect your daily life?

Figure 1 Example of a session in the chatroom. Left: chat screen with chat-talk (every participant has his/her own color), smileys to express feelings. Middle: information screen where course leaders can provide information to the participants. At the bottom: menu where course leaders can select information for the information screen. On the far right: listing of the names of participants and trainers.

Second, an evaluation questionnaire focusing on satisfaction with the content, design and course leaders was completed by participants at T1. The questionnaire consists of two parts with a total of 41 items. The first part has 25 questions (e.g. "What is your opinion about the design of the chatroom?") with different multiple-choice answer options. The second part has 16 statements (e.g. "I liked following the course via a chatroom") with a five-point Likert scale (ranging from 1 "totally agree" to 5 "totally disagree").

Outcome measures of preliminary effectiveness are disease-related coping skills and psychosocial functioning (emotional/behavioral problems and HRQoL), assessed with three specific validated questionnaires. Disease-related coping skills were assessed with the *Op Koers* questionnaire (24, 33). Adolescents were asked if they agreed with 26 statements (four-point Likert scale ranging from 1 "always/almost always" to 4 "almost never/never") on the use of coping skills taught in *Op Koers Online* (e.g. "I know how to get answers to questions about my disease"). The items are divided into five subscales: *information-seeking* (6 items), *relaxation* (3 items), *social competence* (6 items), *positive thinking* (3 items) and *medical compliance* (8 items). All items together form a total scale score. Mean item scores were calculated for the subscales and the total score (range 1-4). Higher scores reflect use of more active coping skills. Subscales had moderate to good internal consistencies (Chronbach's α=0.46 for *social competence* T0 to α=0.71 for *relaxation* T1). The total scale had good internal consistency (T0 α=0.70, T1 α=0.89). The subscale *medical compliance* was not used in the analyses because of insufficient internal consistency (T0 α=0.10, T1 α=0.40), but the items of that subscale were included in the total scores.

Emotional and behavioral problems were assessed with the Youth Self Report (YSR) (37) consisting of 119 problem items (three-point Likert scale ranging from 0 "not at all" to 2 "often/a lot"). The YSR has two broadband scales each consisting of subscales: *internalizing problems* (31 items, range: 0-62), including the subscales *anxious/depressed* (13 items), *withdrawn/depressed* (8 items) and *somatic complaints* (10 items), and *externalizing problems* (32 items, range: 0-64) including the subscales *rule-breaking behavior* (15 items) and *aggressive behavior* (17 items). We excluded the subscale *somatic complaints* from *internalizing problems*, since all participants have somatic complaints due to their illness (38). On this questionnaire, higher scores indicate more problems. Cronbach's alphas for the YSR (sub)scales at T0 and T1 were moderate to good (α=0.61 for *aggressive behavior* T0 to α=0.86 for *internalizing problems* T0) except for the subscale *rule-breaking behavior* which was therefore excluded from further analysis (α=0.36, T1). T-scores were used to assess whether participants reported subclinical/clinical symptoms; T-scores in the 90th percentile or higher in the norm population are considered subclinical/clinical, indicating that the adolescent has clinically relevant symptoms and may need professional help (37). To indicate the percentage of participants scoring within the subclinical/clinical range (T-score 63 or higher), we computed T-scores from the raw scale scores.

HRQoL was measured with the Pediatric Quality of Life Inventory – self report (PedsQL 4.0) (39). All items state a problem (e.g. "difficulty walking"), and participants indicate to what extent they had difficulties with that problem in the past month (five-point Likert scale ranging from 0 "never" to 4 "always"). The PedsQL 4.0 contains 23 items divided into four subscales: *physical functioning* (8 items), *emotional functioning* (5 items), *social functioning* (5 items) and *school functioning* (5 items). The *psychosocial functioning* scale score is the combined score of *emotional*, *social* and *school functioning,* and the total score is the sum of all subscales. Higher scores indicated a better HRQoL (range 0-100). Cronbach's alpha of the PedsQL (sub)scales were moderate-to-good (lowest α=0.66 for *physical functioning* T0 and highest α=0.89 for *physical functioning* T1).

Statistical Analyses

Descriptive analyses were performed to characterize the participants. To assess feasibility, we calculated percentages of attendance and analyzed the results on the evaluation questionnaire descriptively. To assess preliminary effectiveness, mean scale scores on disease-related coping skills, emotional/behavioral problems and HRQoL at T1 were compared to T0 by paired sample t-tests (p<.05) using SPSS. Given the explorative character of these analyses, we decided not to correct for multiple testing. Between-subject effect size, Cohen's *d,* was calculated by dividing the difference in mean scores T1-T0 by the standard deviation at T0. Effect sizes *d* of .2 were considered small, .5 medium and .8 large (40). The use of between-subject effect size was preferred over within-subject effect size because in small samples as in our pilot study, calculation of within-subject effect size may result in overestimation of the effect size. In addition, the use of between-subject effect sizes is common, which benefits the interpretation of the results (41).

RESULTS

Participants

Divided over six groups, 33 adolescents participated during the pilot period. A total of 29 participants (88%) completed the questionnaires at T0. At T1, 25 (76%) participants completed the evaluation questionnaire and 23 (69%) completed the other questionnaires too.

Table 2 presents the participant characteristics. A majority of the participants was female (62.1%), mean age was 15.1 years. There was a large variability in diagnoses, 31% occurred once. Most common diagnoses were bowel disease (20.7%) and Cystic Fibrosis (17.2%). Ten percent had subclinical/clinical scores on internalizing problems at T0. There were no subclinical/clinical scores on externalizing problems.

Table 2 Characteristics of participants of *Op Koers Online* (N=29)

	M	**SD**	**Range**	**N**
Age at T0 (years)	15.1	1.85	12.3-18.9	29
Age at diagnosis (years)	5.4	6.44	0-17	25
	N	**%**		
Female	18	62.1		
Diagnosis [a]				
Bowel disease	6	20.7		
Cystic Fibrosis (CF)	5	17.2		
Rheumatological disease	4	13.8		
Metabolic disease	3	10.3		
Heart disease	2	6.9		
Other [b]	9	31.0		
Education (current)				
Elementary school (last year)	1	3.4		
Secondary education	26	89.7		
Secondary vocational education	2	6.9		

Table 2 Continued.

	N	%
Treatment [a]		
Use of medication	20	68.9
Regular hospital visits	28	96.5
Surgery	4	13.7
Diet	1	3

[a] More than one answer is possible

[b] Other diagnoses occurred once and were: eczema, epilepsy, Graves' disease, IL12 deficiency, hereditary motor and sensory neuropathies (HMSN), spherocytosis, Alagille Syndrome and endometriosis.

Feasibility

Attendance Fourteen out of 33 participants (42%) attended all eight regular chat sessions, 14 participants (42%) missed one session and three participants (10%) missed two sessions, mostly due to illness or hospitalization and sometimes school (homework). Dropout rate was 6%; two participants decided to quit during the intervention (one due to illness/hospitalization, one due to school-related lack of time). They attended less than three sessions. Attendance at the booster session was 88% (two participants who finished the entire intervention did not attend the booster session). Fourteen participants (45%) completed all homework assignments. Six participants (19%) failed to complete one assignment, five participants (16%) failed to complete two assignments and six participants (16%) failed to complete three or more assignments.

Technological issues In 96% of the sessions, no technological issues occurred. In one session (2%) there was a technological breakdown of the website so that the session had to stop and resume later. In one other session (2%) the chatroom was interrupted a few times, but the session could go on. Sometimes a participant reported technological issues (6%; e.g. interruption of the chatroom, seeing messages multiple times). Course leaders could join the session, and kept contact with participants on how to deal with an issue (e.g. press F5/CMD+R, restart the chatroom).

Evaluation questionnaire According to the first part of the questionnaire, a majority of participants had previously used chat as a communication tool (52% regularly or often, 20% sometimes). The provided information about the content of the intervention before the start was "enough" according to 84% of participants. Most participants were satisfied with the usability of the chatroom, 64% rated it as "good". Some participants (12%) thought the design of the chatroom was not attractive. Most participants (84%) were positive about the course leaders. Regarding duration, 20% of participants thought chat sessions were too short and 24% thought they were too long. Most participants (76%) said the number of sessions was enough. Homework assignments were rated as "good" by most participants

(92%). All participants rated the intervention as understandable. The intervention was found "quite useful" by 68% of participants and "useful" by the other 32%.

On a scale of one to ten, 92% of participants rated a seven or higher for overall satisfaction with the intervention (mean: 8.2), 16% of them rated a ten. More than half of participants (52%) would definitely recommend the intervention to others, some participants (32%) would likely do that and a minority of participants might not (16%). A majority of participants (84%) rated the design of the chatroom a seven or higher. In the end, 72% participants said that given the choice, they would prefer an online group intervention over a face-to-face group intervention.

The results of the second part of the evaluation questionnaire are presented in Table 3. A majority of the participants thought a chatroom is a good format for this intervention and reported that chatting is a good way for them to talk about difficulties related to the illness. Most participants felt understood by other participants. Opinions were divided about the use of smileys (faces with expressions of emotions to use in the chatroom), anonymity and webcam use.

Table 3 Statements about *Op Koers Online*, evaluation questionnaire (N=25).

	(totally) agree	don't agree/ don't disagree	(totally) disagree
The chatbox	%	%	%
A chatbox is a good format for this intervention	88	12	0
I liked taking part in the intervention via a chatbox	76	12	12
I found it hard to take part in the intervention via a chatbox [a]	20	8	72
Chatting is a good way for me to talk about the difficulties I have in relation to the consequences of my illness	80	4	16
During the chat sessions…			
… *it was hard for me to follow the subject* [a]	8	12	80
… a lot of messages appeared on the screen at once	40	32	28
… it was (mostly) clear who responds to whom	68	24	8
… I could (mostly) say what I wanted to say	84	8	8
Interaction	%	%	%
The course leaders responded to what I said	96	0	4
I felt understood by the course leaders	88	8	4
I felt understood by the other participants	92	0	8
Tool for expression of feelings	%	%	%
Smileys helped me express my feelings	28	32	40
Smileys helped me understand participants' feelings	40	32	28
Privacy	%	%	%
I liked the fact that participation was anonymous	36	32	32
I would have liked to see other participants via webcam	36	24	40
I would have liked to see course leaders via webcam	32	12	56

[a] Statement is negative

Preliminary Effectiveness

Table 4 shows significant improvement in coping skills: total scale, t(21)=-2.83); information-seeking, t(21)=-3.07; and social competence, t(21)=-2.68. Significant decrease in emotional/behavioral problems was found for: externalizing, t(22)=2.36; withdrawn/depressed behavior, t(22)=3.27; attention problems, t(22)=2.21; and aggressive behavior, t(22)=2.47. HRQoL improved on: total scale, t(21)=-2.58; emotional functioning, t(21)=-4.06; and psychosocial functioning, t(21)=-3.42.

Table 4 Effectiveness, T1 versus T0: disease-related coping skills (*Op Koers questionnaire)* and psychosocial functioning (emotional and behavioral functioning; *YSR* and HRQoL; *PedsQL*)

	T0	T1		
	Mean (SD)	Mean (SD)	*p*-values	Effect size (d)
Op Koers questionnaire[1] – N=22				
Seeking/giving information about the illness	2.72 (0.48)	3.01 (0.53)	**<0.01**	0.60
Relaxation during stressful situations	2.41 (0.65)	2.61 (0.73)	0.16	0.31
Social competence	2.55 (0.48)	2.77 (0.45)	**0.01**	0.46
Positive thinking	2.33 (0.58)	2.62 (0.73)	0.06	0.50
Total	2.83 (0.29)	3.00 (0.43)	**0.01**	0.59
Youth Self Report [2] (YSR) – N=23				
Internalizing problems [3]	11.83 (6.67)	10.61 (6.27)	0.17	0.18
Anxious/Depressed	6.13 (4.40)	6.00 (4.25)	0.84	0.03
Withdrawn/Depressed	5.70 (3.01)	4.61 (2.64)	**<0.01**	0.36
Thought problems	4.74 (3.24)	4.30 (3.36)	0.20	0.14
Externalizing problems	5.65 (3.59)	4.30 (3.40)	**0.03**	0.38
Social problems	3.74 (3.11)	3.61 (3.01)	0.81	0.04
Attention problems	6.04 (3.30)	5.09 (3.15)	**0.04**	0.29
Aggressive behavior	3.52 (2.66)	2.26 (2.36)	**0.02**	0.47
Pediatric Quality of Life Inventory – self report [1] (PedsQL) – N=22				
Total score	55.93 (14.23)	61.07 (15.19)	**0.02**	0.36
Physical functioning	50.99 (21.87)	54.26 (22.81)	0.37	0.15
Emotional functioning	56.36 (23.41)	67.95 (20.51)	**<0.01**	0.50
Social functioning	69.31 (19.66)	71.59 (15.54)	0.43	0.12
School functioning	50.00 (17.18)	54.55 (18.19)	0.18	0.26
Psychosocial functioning	58.56 (15.13)	64.70 (15.14)	**<0.01**	0.41

Significant differences are bolded.

[1] Higher scores indicate more use of coping skills or better HRQoL

[2] Higher scores indicate more problems

[3] Without Somatic Complaints subscale.

DISCUSSION

The aim of this pilot study was to assess feasibility and explore preliminary effectiveness of *Op Koers Online*. Regarding feasibility, we found good attendance: the dropout rate was low (6%) compared to dropout rates of other internet-based interventions for adolescents (42). The technological quality was good; small issues were fixed by course leaders and/or the web developer. Only one time did a technological issue cause the end of a session. Participants reported positive overall satisfaction with the intervention, indicating its feasibility for this population with CI. Regarding effectiveness, we found improvement on disease-related coping skills and HRQoL, and decrease of emotional/behavioral problems.

Feasibility

According to the evaluation questionnaire, participants' opinion about taking part in the intervention anonymously is divided. Regarding webcam use, the difference in percentages between participants who did and did not want to see other participants and course leaders via a webcam is small, which indicates that a considerable portion of participants would have liked more openness. In the pilot study *Op Koers Online* Oncology, opinion about anonymity was divided too (33). As discussed, *Op Koers Online* is intentionally designed without a webcam, and the protocol is set up to ensure anonymity. Furthermore, results of the evaluation questionnaire showed that for a majority of participants the smileys were not specifically helpful to express personal feelings. This could be due to the type of smileys, which are a little outdated. Also, a majority of participants found that the design of the chatroom was not particularly attractive. When optimizing the intervention, a renewed, more attractive design and updates of smileys should be considered.

Among the advantages of offering this intervention online is improved accessibility. The disadvantage is the risk of technological issues which can interrupt with the intervention. Recommendations on what to do when that happens were included in the manual. For example, course leaders can call the web developer for help, and course leaders and participants can press F5 (CMD + R for Apple) to reset the chatroom. Course leaders are advised to call participants when they lose online contact to assist them with resuming the chat.

Preliminary effectiveness

Most results seemed in line with findings on efficacy of the *Op Koers* face-to-face intervention (26). However, given the different study designs actual comparison of this pilot study with the RCT is not workable. The coping skill 'use of relaxation' did not improve significantly in participants after following *Op Koers Online*. This could be explained by the way the relaxation exercise is taught. Participants had to practice with a sound fragment themselves. Though the course leaders asked questions to monitor the performance, it was difficult to check whether participants were performing the exercise correctly. To make the relaxation exercise more attractive for adolescents and increase participant willingness to perform the exercise, we recommend adding a movie to the sound fragment.

So far, studies focusing on online group interventions for chronically ill adolescents in the Netherlands are limited. Studies abroad show promising results on the efficacy of internet-delivered cognitive-behavioral interventions for youth with CI (43-45), but much work remains to be done (46). The present study's contribution to the field is an evaluation of an online group intervention (chat) for adolescents. Especially the fact that we include adolescents with all kinds of diagnoses is new. In terms of feasibility and preliminary effectiveness, we find comparable results with former research: positive results on preliminary effectiveness and good feasibility (10-12, 21, 23). The pilot study had some limitations. First, recruitment and enrollment rates are not known because an open recruitment strategy was followed. Second, the data of the assessment after the booster session could not be used for analysis because of too low response rate (10%). Third and fourth limitations are the rather small sample size and a one-group pre/post design. Although the sample size is appropriate for a pilot study, a larger sample and a control group to compare with the results of the intervention group would have expanded our capacity to find evidence for feasibility and potential effectiveness. Notably, effect sizes found in pilot studies should be interpreted with caution as the meaning of hypothesis testing is limited in pilot studies (47, 48). Fifth, because of the explorative nature of the analyses, we did not control for multiple testing. Because of these limitations, our findings should be interpreted with caution.

CONCLUSION

First steps into assessing feasibility and effectiveness of *Op Koers Online* for adolescents with CI were taken. Results are promising; the use of coping skills and psychosocial functioning has improved. The current study shows that an online CBT intervention is feasible for adolescents with CI and that they benefit from the therapeutic techniques used. It also shows that a generic approach is appropriate for these adolescents, which is contributing to the existing knowledge on psychosocial group interventions. As the results of a pilot study should be interpreted with caution, results should be validated in an RCT.

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3

Matching psychosocial support needs of parents of a child with a chronic illness to a feasible intervention

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Maternal and Child Health Journal

ABSTRACT

Objectives Parents of children with a chronic illness (CI) are at risk for psychosocial problems. The final aim of this study is to develop an online psychosocial group intervention for parents by 1) exploring which themes are important, 2) determine what type of intervention parents would like and 3) assess parents' practical preferences.

Methods Parents of children with a CI (0-18 years) were invited to complete an online questionnaire. To acquire more in-depth information, focus groups and telephone interviews were conducted. Descriptive statistics were used.

Results 272 parents (mean age=43.1 years, 85% female) participated. Three focus groups (15 parents) and seven telephone interviews were conducted. Most important themes were: the CI of the child, family functioning, taking care of yourself, relationships with others and practical support. Parents preferred a group with parents of children in the same age category. At first, parents preferred face-to-face contact. After an explanation and demonstration of an online intervention, parents became more positive about online support, mostly because they could participate from home.

Conclusions for practice Parents have a need for psychosocial support focusing on different themes. It is important that professionals explain and demonstrate an online intervention to parents. Based on these results, *Op Koers Online for parents* was developed. An RCT to assess feasibility and effectiveness of the intervention is currently running.

INTRODUCTION

Parents raising a child with a chronic illness (CI; e.g. asthma, diabetes) are predominantly responsible for managing the child's illness. Parents are confronted with stressors about their child's health including practical stressors (e.g. managing daily medical routine) as well as emotional challenges (e.g. worrying) (1, 2). Therefore, these parents are at risk for psychosocial problems (3) and elevated levels of distress (4, 5), which can have a negative impact on parents' coping with illness-related stressors, emotional availability for their children and their ability to manage the child's illness effectively (1). Extra attention for these vulnerable parents is necessary, to prevent and/or reduce psychosocial problems and to help them support their children adequately (2, 6).

Psychosocial support of parents became more relevant in the past few years (7-9). Emotional, informational and peer support interventions for parents themselves are suggested (10, 11). A way to support parents is by using cognitive-behavioral therapy (CBT), which focusses on recognizing cognitive distortions and on teaching parents how to use active coping skills for illness-related problems (12). There are several CBT intervention programs available that involve parents. However, those interventions are often primarily focused on the child's functioning; parents learn how to support their child with managing the illness (13, 14).

Studies have shown that sharing experiences with others in a similar situation is associated with a decrease of distress and improvement of mental health for children as well as parents (15, 16). CBT interventions can be offered in group format. Little is known about the effectiveness of group CBT interventions for parents, but recent research shows promising results on feasibility and effectiveness of this type of intervention for youth with a CI (15, 17-20). A CBT group intervention could be helpful for parents of children with a CI as well.

Over the past few years, a face-to-face CBT group intervention program called *Op Koers* (in English: *On Track*) was developed in the Emma Children's Hospital (Amsterdam University Medical Centers) and was proven to be effective (19). The intervention has different modules for children and adolescents with CI, their parents (21, 22) and siblings. Patients with all kinds of CI and their family members are eligible for the intervention: research shows that even though different diagnoses may have different medical treatments, several of the psychosocial problems are the same (17). Besides, a generic approach allows for patients with rare illnesses and their family members to participate in a group intervention. *Op Koers* was designed with this in mind (22). The face-to-face parent intervention runs parallel to the child intervention: parents learn what the children learn in order to support their child better in daily life. The goal of *Op Koers* is to prevent and/or reduce psychosocial problems by teaching the use of active coping skills. Sharing emotions and experiences with others in the group is an important part of the intervention. All modules have weekly 90-minute sessions, for eight weeks, guided by two psychologists (course leaders).

A face-to-face intervention requires participants to visit the hospital. Logistical problems (e.g. travel time) and practical issues (e.g. time of onset of the sessions) have frequently been identified as barriers for participation in face-to-face interventions (23, 24). Online interventions eliminate these logistical barriers (25, 26) and practical issues are reduced when participation from home is possible (27). Moreover, for some parents is easier to type about difficult topics in an online environment instead of talking about it in real life (24). Research shows that outcomes of online interventions are comparable to face-to-face interventions (28). Online interventions will not likely replace face-to-face care completely. However, because of the benefits mentioned above, the need for online interventions continues to grow (29, 30). To increase participation in *Op Koers*, the adolescent group intervention was translated into an online version (20, 31). A pilot study shows promising results on feasibility and preliminary effectiveness (18). For parents however, there was a need to develop a new online intervention that focuses on the parents themselves.

Patient participation becomes more important in intervention development and improves adherence and patient outcomes (32, 33). Therefore, it is highly important to involve parents in the development of an intervention. The final aim of this study is to develop a feasible online group intervention for parents by 1) exploring which themes are important to address in the intervention, 2) determine what type of psychosocial intervention parents would like, and 3) assess parents' practical preferences for an online group intervention.

METHODS

Recruitment and data collection

A mixed method approach, both quantitative (questionnaire) and qualitative (focus groups/ interviews), was used. The only inclusion criterion was being a parent of a child between the ages of 0 and 18 years with a CI diagnosis according to the following criteria 1) onset between aged 0-18, 2) diagnosis based on medical scientific knowledge, 3) the illness is not (yet) curable, and 4) the illness has been present for at least three months, or at least three episodes have occurred in the last year (34).

In order to recruit parents, 57 patient associations were invited to publish a link to the open access questionnaire on their website, social media and/or in their newsletter. Fourteen patient associations (25%) agreed. In addition, the questionnaire was announced on several websites and social media accounts managed by the psychosocial department of the Emma Children's Hospital. Hardcopy flyers were spread out in the (outpatient) clinic of this hospital.

Parents who were willing to complete the online questionnaire used the open access link. At the end of the questionnaire, parents could indicate if they were interested in participating in a focus group and if so, they could leave their contact details. Interested parents were called by the researcher to schedule the focus groups. Completed questionnaires were anonymously stored in a (secured) website. The focus groups were recorded and transcribed verbatim. When parents were not able to join the focus groups, the researcher offered a telephone interview. During these interviews, extensive notes were taken.

Approval of the Medical Ethical Committee was obtained to conduct this psychosocial support needs study. Parents gave informed consent prior to participation in the focus group/interview.

Questionnaire

Background characteristics

Background characteristics of participating parents (age, sex, marital status, number of children, prior psychosocial support, need for psychosocial support now or in the future for themselves, their child with a CI and possible siblings) and of their child with a CI (age, sex, CI, presence of a second diagnosis) were collected.

Support needs

Parental support needs were assessed with 27 questions, including open and multiple-choice questions. The questions concerned 1) which themes are important to address in an intervention, 2) what type of psychosocial intervention parents would like, and 3) practical preferences for an online group intervention.

Focus groups and telephone interviews

The focus groups with parents were held in the Emma Children's Hospital and led by two researchers using semi-structured interview techniques. The goal of the focus groups and telephone interviews was to acquire more in-depth information in addition to the questionnaire. The same sequence of topics was discussed.

Data analyses

SPSS version 24.0 (IBM Corp, 2016) was used for all quantitative analyses. To indicate support needs, descriptives and percentages were computed. The transcript verbatim of the focus groups and notes from the telephone interviews were read carefully to detect any themes or important aspects of an intervention that were not found with the questionnaire.

Developing a psychosocial group intervention for parents

Based on the results of the current support needs study, previous studies and the experiences of *Op Koers* developers and course leaders, a feasible online psychosocial group intervention for parents was developed.

RESULTS

Participants

A total of 272 parents (mean age = 43.1 years, SD = 7.3 years, 86% female) completed the support needs questionnaire (Table 1). Most parents were married and/or living together (90%) and had more than one child living in their family (87%). More than half of the parents (55%) has had prior psychosocial support from one or more of the following professionals: psychologist (35%), a (medical) social worker (19%) and/or a child life specialist (10%). The mean age of the children (46% female) was 10.7 years (SD = 5.8 years). Parents reported over 60 different types of CI. Half of the parents (51%) indicated a current or future need for psychosocial support. A majority (68%) thought their child with a CI has a current or future need for psychosocial support and one-third of the parents (33%) indicated a current or future need for psychosocial support for siblings. Approximately one-third of the parents (35%) indicated that they would like to have contact with other parents with a chronically ill child for support.

A total of 85 parents (31% of all parents) left their contact details for participation in a focus group, of whom 15 parents (18% of the parents who left contact details) participated in three focus groups. The researcher conducted telephone interviews with seven parents (8%). The other 63 parents (74%) could not participate due to several logistical and practical reasons.

Table 1 Background characteristics of parents and their children with a chronic illness (N=272)

Characteristics of parents	N	Mean (SD) or %
Age in years	272	43.1 (7.3)
Sex (Female) [1]	235	86
Married and/or living together	246	90
Number of (step)children living in your family		
1	34	13
2	148	54
3	63	23
>3	26	10
Prior psychosocial support, yes	150	55
… from a psychologist	94	35
… from a (medical) social worker	51	19
… from a child life specialist	27	10
Characteristics of children		
Age in years	272	10.7 (5.8)
Sex (female)	125	46
Chronic Illnesses (main diagnosis)		
Epilepsy	68	25
Neurofibromatosis Type 1/2	57	21
Diabetes Type 1	36	13

Table 1 Continued.

Characteristics of children	N	Mean (SD) or %
Cystic Fibrosis	21	8
Cancer	12	4
Neurological disease (other than epilepsy)	11	4
Migraine	10	4
Other	51	19
Second diagnosis	91	33

[1] Three females filled out the questionnaire together with their partner (male)

Questionnaire and focus groups/telephone interviews

Information from the questionnaire and the focus groups/telephone interviews was used to assess parental support needs. Hereafter, our final aim to develop an online psychosocial intervention was reached. The aims will now be discussed one-by-one.

Which themes are important to address in the interventio*n*

Questionnaire

The parents who indicated a need for psychosocial support for themselves (51% of all parents), reported that they would like a focus on their own (emotional) functioning (76%), on how to support their child in living with a CI (70%) and on family functioning (60%). Other themes suggested in the open question were: how to support the child/adolescent in achieving independence, autonomy and self-esteem, and guidance with special (financial) arrangements and different agencies. (e.g. insurances).

Focus groups/telephone interviews

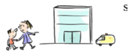
Parents indicated that they needed support in accepting the diagnosis and how to cope with several difficult situations while raising a child with a CI (e.g. coping with different future perspectives). Parents also reported that they needed support concerning the impact of the child's CI on the family and the partner relationship. Furthermore, parents would like to discuss how to take care of themselves next to all parenting responsibilities. Finally, parents would like a focus on how to communicate with their work/school of the child and on practical information (e.g. financial resources). According to parents, an intervention needs to be solution-focused. Table 2 presents parental support needs and how these are categorized into four themes to apply in the intervention.

What type of psychosocial intervention parents would like

Questionnaire

In Table 3, results on what type of psychosocial intervention parents would like are presented. More than half of the parents would like information on a website. Individual counseling with a therapist, face-to-face in the hospital was attractive to a majority of the parents. Regarding a group intervention, parents preferred a face-to-face setting in the hospital. Almost a quarter would like a group intervention in a secured chat with the same therapist. When asked about an individual e-learning, most parents preferred a website with online assistance of a therapist.

Table 2 Overview of parental support needs, how these are categorized into themes and applied in the sessions of *Op Koers Online for parents* including group discussions and homework assignments

What do parents want?	How is this applied in *Op Koers Online for parents*?		
Parental needs	**Theme***	**Group discussion**	**Homework assignments**
Guidance in accepting the diagnosis, coping with a different future perspective, adherence/non-adherence and puberty	The CI of the child *(session 2: "The hospital")* 	Accepting the diagnosis, how to support the child, successes and struggles in medical treatment of the child and how to handle difficult situations in the hospital	Reading a story about how to cope with the diagnosis & together with your child, make a list of situations for adherence/non-adherence and discuss how to reach adherence
The impact of a CI on family functioning, partner relationship and keeping balance in dividing attention between siblings	Relationships within the family *(session 3: "The family")* 	What is the impact of the CI on the child with CI, siblings, the relationship with your partner? What are successes and struggles in your family?	Talk with siblings about worries they have, make a list of things to do with siblings and your partner and have quality time with them each
Taking care of your own body and mind, paying attention to own emotions	Taking care of yourself besides caregiving tasks *(session 3: "Taking care of yourself")* 	What is the impact of the CI on your own life and emotional functioning? How do you take care of yourself?	Practice with the relaxation exercise, take time for yourself, give yourself a compliment daily
Communication with bosses/colleagues and teachers of the child about the impact of the disease, practical information about insurances etc.	Relationships with others and practical support *(session 4: "Extended family and friends")* 	What kind of support would you like to receive/do you receive? What kind of reactions do you get and how does that feel? What works or could be better in the communication with work/school?	Together with your partner, write down difficult questions/reactions you get from others and think of possible ways to react on that

* All themes are linked to a session, however, specific content is determined by parents in every session (what they want to discuss).

Focus groups/telephone interviews

All parents preferred a group intervention where they can share experiences and tips with other parents in a similar situation. As parents want to discuss different stages in the life of their child, intervention groups should be composed based on the (developmental) age of the child instead of the CI. Parents expressed a preference for face-to-face support, however, a combination of face-to-face and online sessions would also be appropriate. Most parents preferred to have the first session face-to-face, followed by online sessions. It was important for parents that the intervention is guided by professionals.

Next to the type of intervention, the discussion about timing of the intervention came up in the focus groups. Parents indicated that they received a lot of information when their child was diagnosed with a CI. They experienced trouble finding their way into psychosocial support and/or contact with other parents. Moreover, parents found it hard to seek for and accept psychosocial support. They considered consulting a psychosocial healthcare specialist as a failure and felt like they had to solve the problems themselves. Considering this, parents suggested a standard consultation with a psychosocial healthcare provider (e.g. psychologist, social worker) a few months after the child's diagnosis and they emphasized that an intervention should be easily accessible.

Practical preferences for an online group intervention

Questionnaire

A majority of the parents would like to participate in a group of parents of children in the same age category (Table 3). More than half of the parents prefers sessions planned in the evening, almost one-fifth prefers morning sessions.

Table 3 Parents' answers on what type of psychosocial intervention they would like and their practical preferences for an online group intervention (N = 272)

Preferences for type of intervention	**N**	**%**
Information on a website	148	54
Information in a folder	85	31
Individual counselling of a therapist		
Face-to-face in the hospital	176	65
Online individual counselling (the same therapist in all sessions)	92	34
Via an open chat (openly accessible, each time a different therapist)	16	6
Group intervention with other parents and a therapist		
Face-to-face in the hospital	123	45
Via a secured chat (each time the same therapist)	63	23
Via an open chat (openly accessible, each time a different therapist)	16	6
E-learning		
On a website with online assistance	105	39
On a website without online assistance	70	26
Practical preferences for an online group intervention		
Group composition		
Parents of children in the same age category	179	66
Parents of children aged 0-18	52	19
Parents of children with the same CI	9	3
Other (e.g. matched on parent's own age, no preference)	32	12
Time of sessions		
Evening	153	56
Morning	51	19
Afternoon	13	5
Flexible	14	5
During the weekend	5	2
Other (e.g. no preference, I don't know)	36	13

Focus groups/telephone interviews

Parents acknowledged the advantages of an online intervention in terms of logistical and practical factors. They mentioned that the possibility to participate from home is a big advantage because it improves accessibility. However, in the beginning, parents were reluctant about an internet intervention and preferred face-to-face contact. After an explanation of an online intervention and a demonstration of what a chatroom would look like, parents became more enthusiastic. In hindsight, parents told they had preconceptions about an online intervention (e.g. difficult to log on) which appeared to be incorrect.

Development of an online psychosocial group intervention for parents

Based on the results of the current support needs study, knowledge from former literature and the experiences of *Op Koers* developers and course leaders, an online CBT group intervention for parents was developed: *Op Koers Online* for parents. The intervention consists of six weekly morning or evening sessions of 90 minutes and one booster session four months after the last session. A group of three to five parents chats under supervision of two psychologists (course leaders) in a secured chatroom.

The support needs of parents were categorized into four themes (Table 2): 1) the CI of the child, 2) relationships within the family, 3) taking care of yourself besides all caregiving tasks and 4) relationships with others and practical support (e.g. school, work). Corresponding topics for group discussion were added. Specific content of each session is determined by parents: what they want to discuss about that theme. The first session is used for introductions and explanation about the intervention. In the last session, there is time to repeat topics, to address matters that have not been discussed due to lack of time and to reflect on the intervention. In addition to the chat sessions, parents can log on to their own personal environment where they can submit weekly homework assignments and view supplementary background information (Table 2).

DISCUSSION

The final aim of the current study was to develop an intervention for parents of children with a CI based on their support needs. Support needs were assessed by combining the results from both quantitative as well as qualitative research.

First, important themes to address in an intervention were explored. A majority of the parents with a current or future need for psychosocial support would like an intervention that focuses on their own (emotional) functioning, how to support their child in living with a CI, family functioning, taking care of themselves, relationships with others (outside the family) and/or practical support. These themes are in line with important themes found in previous research (7, 8, 35).

The second aim was to determine which type of psychosocial intervention parents would like. The parents in our study preferred a face-to-face intervention over an online intervention. This is contradictory to parents' practical possibilities, which was underlined by the low percentage of parents able to attend the focus group they signed up for. A potential explanation could be that parents have an incomplete understanding of what an online intervention entails. Unfamiliarity with online interventions may lead to negative preconceptions and may cause parents to prefer the more conventional face-to-face setting. In the end, it is expected that practical and logistical advantages of an online format will overweigh parent's wish for a face-to-face setting. Adequate explanation and demonstration will make an online format more feasible. Finally, parents indicated trouble finding their way into psychosocial support and/or contact with other parents of a child with a CI. This is corresponding with former research which shows that parents of an ill child can feel isolated, the information they find online is lacking and peer support is desirable (24). The suggestion of a standard psychosocial consultation after the child's diagnosis that parents made is valuable.

The third aim was to assess practical preferences of an online group intervention. Parents preferred to participate in a group with parents of children from the same age category. When composing a group, attention should be paid to the developmental age of the child as well as the calendar age. Furthermore, most parents preferred sessions planned in the evening. Some parents indicated not to know their practical preferences for time of onset of the sessions. A possible explanation could be the unfamiliarity with online interventions which can make it difficult to indicate a preference.

Based on the results of the support needs study, an online psychosocial group intervention *Op Koers Online* for parents was developed. Parents' preferences were mostly met, for example: the themes were established by the parents, the day and time of onset of the sessions is planned in consultation with participants, group composition is based on the age category of the children, a manual to support parents by logging on to the website and entering the chatroom was developed and the intervention is easily accessible (easily accessible website, participation possible from home). However, not all parents' wishes can be granted. For example, parents preferred one face-to-face session followed by online sessions. However, to ensure anonymity and to eliminate practical and logistical barriers, a face-to-face session was not included. Instead, the course leaders speak to all participants separately on the phone before the first session to introduce themselves, explain the chatroom and answer possible questions of participants. Finally, there will still be a group of parents who prefer face-to-face over an online setting. For those parents, there is always the option for face-to-face care in the hospital.

This study has some limitations. An open recruitment strategy was used, which eliminates the possibility to acquire and discuss information about response rates and differences between non-respondents and respondents. Furthermore, fathers were underrepresented. As a result we can not comment clearly on the representativeness of the results.

CONCLUSIONS FOR PRACTICE

Based on parental support needs and the themes parents considered as important to address in an intervention, an innovative parent-focused intervention *Op Koers Online* for parents was developed to use in clinical practice. *Op Koers Online* for parents can be offered to parents after receiving the child's diagnosis. It is an important contribution to the field, because the focus is on parents themselves, as opposed to existing parental interventions that focus on teaching parents how to support their children. Furthermore, because the intervention has a generic approach, parents of children with rare illnesses have the opportunity to participate in a group intervention. An important finding is the fact that parents are reluctant about the online aspect of the intervention. Caregivers should be aware that it is important to explain and demonstrate the online intervention to parents as well as to discuss possible preconceptions and/or misconceptions. For *Op Koers Online* for parents, information booklets and an extensive login manual for (potential) participants were made. Another important finding is that parents suggest a standard psychosocial consultation after receiving the child's diagnosis, to overcome the barriers for seeking for psychosocial support. We suggest that medical staff direct parents to the psychosocial department of the hospital for a standard consultation when their child is diagnosed with a CI.

Future research should examine the effects of *Op Koers Online* for parents. An RCT to assess feasibility and effectiveness of the intervention is currently running (31).

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4

Online cognitive-behavioral based group interventions for adolescents with chronic illness and parents: Study protocol of two multicenter randomized controlled trials

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ABSTRACT

Background Adolescents with chronic illness (CI) and parents of a child with CI are at risk for psychosocial problems. Psychosocial group interventions may prevent these problems. With the use of cognitive-behavioral therapy, active coping strategies can be learned. Offering an intervention online eliminates logistic barriers (travel time and distance) and improves accessibility for participants. Aim of this study is to examine the effectiveness of two cognitive-behavioral based online group interventions, one for adolescents and one for parents: *Op Koers Online*. The approach is generic, which makes it easier for patients with rare illnesses to participate.

Methods/design This study conducts two separate multicenter randomized controlled trials. Participants are adolescents (12 to 18 years of age) with CI and parents of children (0 to 18 years of age) with CI. Participants are randomly allocated to the intervention group or the waitlist control group. Outcomes are measured with standardized questionnaires at baseline, after 8 (adolescents) or 6 (parents) weeks of treatment, and at 6- and 12-month follow-up period. Primary outcomes are psychosocial functioning (emotional and behavioral problems) and disease-related coping skills. Secondary outcomes for adolescents are self-esteem and quality of life. Secondary outcomes for parents are impact of the illness on family functioning, parental distress, social involvement and illness cognitions. The analyses will be performed according to the intention-to-treat principle. Primary and secondary outcomes will be assessed with linear mixed model analyses using SPSS.

Discussion These randomized controlled trials evaluate the effectiveness of two online group interventions improving psychosocial functioning in adolescents with CI and parents of children with CI. If proven effective, the intervention will be optimized and implemented in clinical practice.

Trial registration ISRCTN ISRCTN83623452. Registered 30 November 2017. Retrospectively registered.

Keywords chronic illness, psychosocial functioning, coping, cognitive-behavioral therapy, online psychosocial intervention, group intervention, e-health, adolescents, parents

INTRODUCTION

Children and adolescents with a chronic illness (CI) have to face difficulties related to their illness, such as hospitalization, the use of medication, restrictions in activities and stressors related to the course of the illness and the future (1-3). In the Netherlands, 14% of children and adolescents is growing up with a CI (for example diabetes, asthma or Cystic Fibrosis) (4). Growing up with a CI influences psychosocial wellbeing and the development of cognitive and social skills (2, 5-7). Especially during adolescence, with the formation of identity, self-image and self-esteem, a CI constitutes a major challenge (8, 9).

Research shows that pediatric CI influences psychosocial wellbeing in parents as well (10, 11). Parents are predominantly responsible for managing the child's illness. They are confronted with stressors about their child's health as well as logistical and practical factors such as managing daily routines, relationships with other family members, the balance between family and work and possible financial problems (11, 12). Therefore, parents are at risk for sorrow and psychosocial distress (10, 12). Parents who face stress are less able to manage the child's illness effectively (11). To prevent and/or to reduce psychosocial problems in parents as well as adolescents, interventions focusing on how to cope with stressors caused by the CI are needed (13).

The disability-stress-coping model states that stressors related to the illness and psychosocial adjustment of children and mothers are moderated by coping strategies and cognitive appraisals (14). Moreover, research has shown that effective use of coping skills increases adolescents' medical compliance, improves their psychosocial functioning (2, 15-20) and reduces distress and anxiety in parents (21, 22). Results on the effectiveness of cognitive-behavioral based psychosocial group interventions to learn children and adolescents how to use more effective coping skills are promising (23-25). Research shows that including parents in a psychosocial intervention for children with chronic pain is effective in reducing child's pain (22). There is some evidence of effectiveness of interventions focusing on parents themselves: coping support interventions reduce parental psychological problems during acute hospitalization (21) and problem solving therapy for parents improves parental mental health (22). However, little is known about the effectiveness of cognitive-behavioral based psychosocial group interventions for parents focusing on themselves.

In 2003, the face-to-face cognitive-behavioral based group intervention *Op Koers* (English: *On Track)* for children and adolescent with CI was developed in the Emma Children's Hospital (Academic Medical Center Amsterdam). Over the years, the program was expanded with courses for siblings and parents, and a similar *Op Koers* program for pediatric oncology patients. Goal of *Op Koers* is to prevent and/or to reduce psychosocial problems of participants by teaching active coping skills with the use of cognitive-behavioral therapy (CBT) techniques (26). The approach is generic, which means that patients with every type of CI can participate. This has the benefit of giving more patients at once the opportunity to participate and to include patients with rare illnesses in group interventions (23, 26). Sharing experiences with others in a similar situation had been associated with a decrease of distress and improvement of social health (23, 27-29) and is therefore an important part of *Op Koers*. There have been different studies about this intervention program (26, 30, 31). Part of the research has been an RCT on the efficacy of *Op Koers* for children and adolescents with CI. This study showed positive short- (half year) and long-term (one year) effects on the use of coping skills and psychosocial functioning. For children and adolescents, there was an additional positive effect of parental involvement, especially on long-term and in social-emotional vulnerable children (32-34).

The face-to-face setting of *Op Koers* requires participants to regularly come to the hospital, in addition to other medical appointments. An online intervention eliminates logistical barriers such as travel time and distance (35, 36) which makes the intervention more easily accessible (35, 37, 38). Participating in an intervention online connects to the digital environment in which people live nowadays. Besides, an online environment without use of a webcam increases anonymity: appearance plays no role and this might make it easier to talk about problems (38-40). Therefore, *Op Koers* was translated into an online intervention: *Op Koers Online*.

First, the intervention for survivors of childhood cancer was developed. A pilot study on the feasibility of *Op Koers Online Oncology* for adolescent survivors showed promising results (39). The intervention was optimized based on feedback from participants and course leaders (for example: expanding the sessions from six to eight). After that, *Op Koers Online* for adolescents with CI was developed. Similar to the face-to-face intervention, goal is to prevent and/or reduce psychosocial problems by teaching the use of active coping skills with CBT techniques. Sharing experiences with other chronically ill adolescents is an important part of the intervention. First pilot results on the feasibility and potential effectiveness of *Op Koers Online* for adolescents with CI are promising (41).

For parents, most existing interventions focus on the child's functioning (13, 22). The same applies to *Op Koers* face-to-face for parents, where participating parents learn what their child learns and how to support their child in implementing coping skills in daily life (32, 34). However, research suggests the need of emotional, informational and peer support for parents (42, 43). For the development of *Op Koers Online* for parents, the Emma Children's Hospital (Academic Medical Center, Amsterdam) conducted a survey among parents on their specific wishes and needs. The need for an intervention focusing on parental functioning instead of focusing only on the child emerged from the survey. Based on the results of this survey, *Op Koers Online* for parents of children with CI was designed.

This paper describes the rationale and the design of two separate multi-center randomized controlled trails aimed to assess the extent to which *Op Koers Online* is effective in preventing and/or reducing psychosocial problems (emotional/behavioral problems and quality of life) and improving the use of disease-related coping skills in adolescents with CI

(12-18 years) and in parents of children and adolescents (0-18 years) with CI.

METHODS

Procedure

Figure 1 shows the different phases of the study procedure. There is one coordinating hospital (Emma Children's Hospital, Academic Medical Center, Amsterdam) and eight participating hospitals across the Netherlands (one academic, seven non-academic). The researcher of the coordinating hospital coordinates overall recruitment and administers inclusion of all participants. Local recruitment is coordinated by local investigators of each participating hospital. Adolescents and parents from the outpatient clinics from the nine hospitals receive an information letter from their pediatrician. To improve inclusion of adolescents and parents for the study, we asked permission from the Medical Ethical Committee (METC) to make the procedure open accessible and permission was obtained. Besides the information letters, pamphlets are available in the participating hospitals and other interested hospitals (approached randomly by the coordinating researcher). Recruitment is done via internet (websites and social media) and via patient associations.

Interested adolescents and parents are asked to return the application form added to the pamphlet or to send an e-mail. A telephonic interview is used to screen inclusion criteria, discuss the information about the intervention and the study and to discuss the informed consent. Potential participants can ask questions and get one week to overthink participation. When willing to participate, an informed consent form is sent to the participant to sign and return. As soon as the informed consent form is signed by the researcher as well, the researcher registers the participant online (www.opkoersonline.nl). Participants receive an e-mail with a link to create personal login codes, with which they can login to the secured website.

Every registered participant is in the virtual 'waiting room' until randomization. They are informed about the result of the randomization by e-mail. When randomized in the intervention group, the researcher calls the participant to determine the dates of the intervention. At four time-points, all participants and parents of participating adolescents are invited to complete questionnaires via an e-mail with a personal link to the questionnaire. Total duration time for completion is estimated at 45 minutes for adolescents and parents and 30 minutes for the parents of participating adolescents. After completing all assessments, participants receive a financial reward (€20 voucher for an online book/game store).

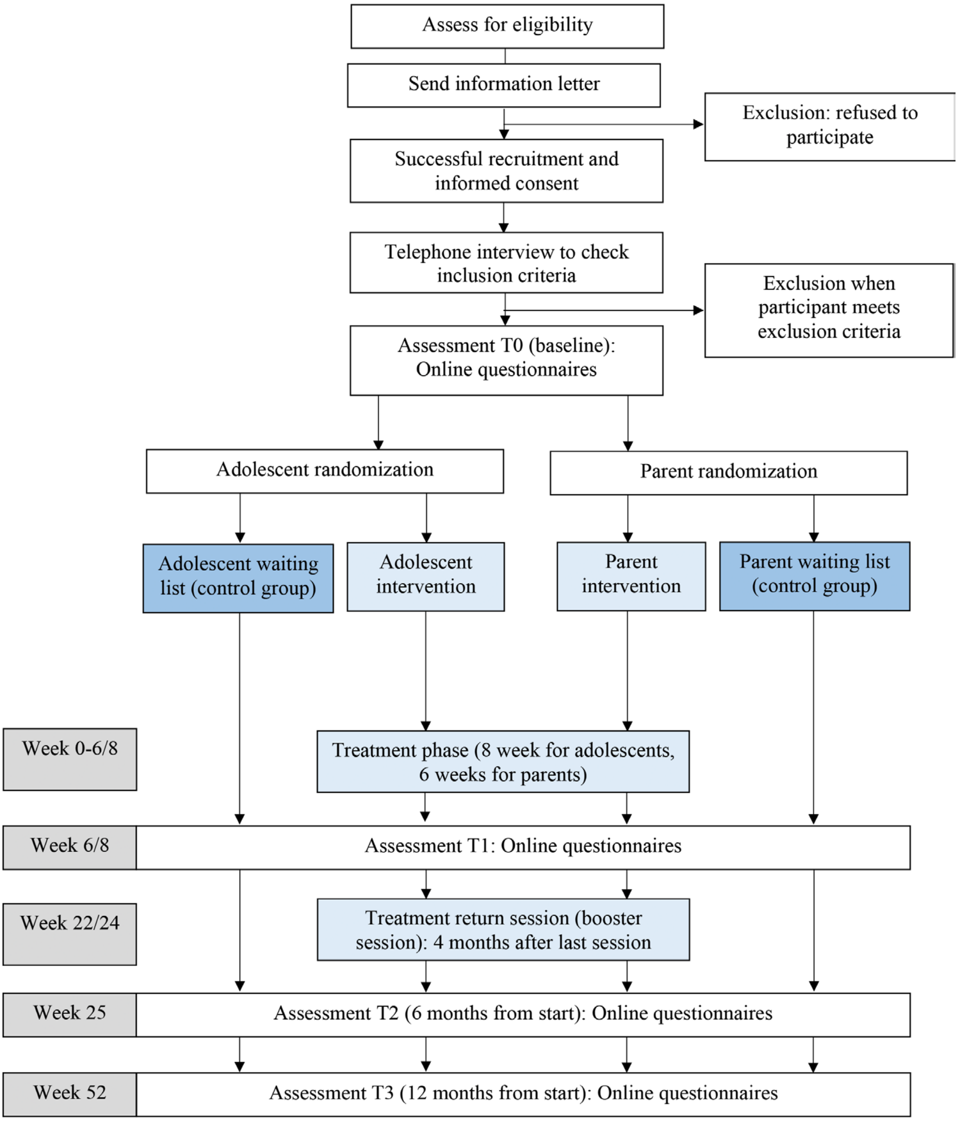

Figure 1 Study procedure in flow diagram

Interventions

The interventions consist of eight (for adolescents) or six (for parents) weekly sessions of 90 minutes, which take place in a secured chatroom (Figure 2) with groups of three to six participants. The interventions are guided by two course leaders, one specialized health-care psychologist and one co-therapist (mostly a psychological assistant), who are trained and use a detailed manual. The training consists of three parts: 1) teaching the main principles of cognitive-behavioral group therapy and the history of the *Op Koers* courses, 2) giving more specific information on the procedures and goals related to the different sessions using examples from former chat sessions and the extensive manual for psychologists, 3) practicing in online subgroups. To ensure treatment integrity, the researcher of the coordinating hospital randomly checks the chats of participating hospitals with the manual. All participants and course leaders log on at the same time every week. Participants can log on to the homework site to view the intervention material (information sheets and videos), submit homework before every session and view additional information. Four months after the last session, there is a booster session.

Central in the interventions is the Thinking-Feeling-Doing model (TFD model). With this model, course leaders teach participants the relationship between what people think, feel and how they act, and how they can influence their thoughts feelings and behaviors. Every intervention group starts the first session with an extensive acquaintance (questions such as: who are you, what do you do, which illness do you/does your child have, what are your expectations of the course, etc.) to create a feeling of safety within the group and in the chat box. No webcams are used in the interventions.

'Op Koers Online' for adolescents

The aim of the intervention for adolescents (12 to 18 years) is to prevent and/or to reduce psychosocial problems, by teaching the use of active coping strategies. These strategies are taught by recognizing negative thoughts and transforming them into more positive and proactive ones, with the use of CBT techniques (TFD-model) (25, 44, 45).

Learning goals of the adolescent intervention are increasing the use of five coping skills taught with the CBT techniques (e.g. relaxation, cognitive restructuring and open communication) (44, 46, 47): 1) information seeking and giving about the illness, 2) use of relaxation during stressful (medical) situations, 3) increase knowledge of self-management and medical compliance, 4) improvement of social competence and 5) positive thinking (26, 34). See Table 1 for learning goals and corresponding instructions and reinforcement techniques. Each coping skill is taught during one specific session, but elements of the coping skills are also addressed in the subsequent sessions. The skills are taught by psycho-education (e.g. video's, group discussions), through exercises (e.g. virtual board games) and homework assignments (e.g. practicing relaxation exercise in daily life).

Table 1 Learning goals and examples of learning activities of *Op Koers* Online for adolescents and parents

		Learning goals						
		Information seeking and giving about the illness	*Use of relaxation during stressfull situations*	*Increase knowledge of self-management and compliance*	*Enhancement of social competence*	*Positive thinking*	*Positive parenting*	*Open communication & seeking and accepting support*
*Examples of learning activities of the **adolescent** intervention*	*Instruction/ modelling*	Education about sources of information	Relaxation exercise (MP3)	Group discussion about own treatment and (non-)compliance	Video and group discussion: how and what do you tell others about your illness	Thinking-Feeling-Doing game		
	Reinforcement/ practice (homework)	Write down questions you have, and look for answers	Practice the relaxation exercise	Write down situations for non-compliance and how to improve compliance	Think of what CAN you do (instead of CANNOT) and write down your story for the other group members	Write down positive adjustments for negative thoughts		
*Examples of learning activities of the **adolescent** intervention*	*Instruction/ modelling*		Group discussion: take care of yourself and relaxation exercise (MP3)	Group discussion: medical treatment and compliance		Discussing the Thinking-Feeling-Doing model	Group discussion: how to divide your attention between different family members	Group discussion: what kind of support do you receive, like and want (practical/ emotional)
	Reinforcement/ practice (homework)		Practice the relaxation exercise	Together with your child (when possible): make a list concerning medical treatment and discuss (non-) compliance		Learning about negative thoughts and how to replace them for positive ones	Make a list of activities to do with different family members (apart)	Think of people who could help and support you and write down what you would ask them

'Op Koers Online' for parents

Aim of the intervention for parents is also to prevent and/or to reduce psychosocial problems by teaching the use of active coping strategies. Strategies to help parents focusing on elements they think are important in life, and to act conform these elements, are taught with the use of CBT techniques and Acceptance and Commitment Therapy (ACT). ACT, part of CBT, is an intervention strategy to learn participants how to accept a new situation (such as: having a child with CI) and to establish new routines. Goal is to increase or create psychological flexibility. This is done with relaxation exercises and reflection which helps participants to remind and recognize what barriers they face in achieving goals and living consistent with their values, and how to adjust behavior in these situations (48-50). There is growing evidence for the effectiveness of ACT (48, 51-53).

Learning goals of the parent intervention are increasing the use of five coping skills taught with CBT and ACT techniques: 1) use of relaxation during stressful situations, 2) increase knowledge of self-management and compliance of their child, 3) positive thinking, 4) positive parenting and 5) open communication about the illness and seeking and accepting support. See Table 1 for learning goals and corresponding instructions and reinforcement techniques.

Every session has a subject. However, specific content of each session is determined by parents: what they want to discuss. Subjects are: the parent (e.g. taking care of yourself), the family (e.g. positive parenting), the hospital (e.g. child's compliance), extended family and friends (e.g. seeking and accepting support), and daily life (e.g. work, school; open communication). Participants are asked to answer questions concerning the subject of the session (for example the following question about the subject 'the family': "How does the illness affect your child his/her siblings/you and your (ex-)partner?") and to react on each other (giving tips, asking questions, sharing experiences). The questions are displayed in the right screen of the chat box. An important part is sharing experiences with other group members and giving and receiving social/emotional support. Compared to the intervention for adolescents, the intervention for parents is less protocolled. There is more room for personal input and (spontaneous) group discussions, and there are less video's, games and exercises during the sessions.

Inclusion and exclusion criteria

Adolescents between 12 and 18 years old with CI, and parents of children between 0 and 18 years old with CI are included. The term CI refers to an illness that requires at least six months of continuous medical care, permanent life style changes and continuous behavioral adaptation to the unpredictable course of the illness (4). Participants (for parents: their child) have to be treated by a pediatric specialist in a pediatric hospital in the Netherlands. Adolescents and parents of participating adolescents should be able to fill out Dutch questionnaires and to follow the chat intervention in Dutch. A computer or tablet with internet connection to enter the website and chat box is necessary. Adolescents and parents with severe learning difficulties are excluded from the intervention. For them, an adapted or individual program might fit better to their individual cognitive needs.

Study design

The current study consists of two separate multi-center randomized controlled trails: one for adolescents and one for parents. Both trials have two conditions: the intervention group (*Op Koers* Online) and the waitlist control group. An adolescent and a parent can both participate, but this is not required. When both parents want to apply for the study, they can participate separately. Participants assigned to the waitlist control group receive care-as-usual and are not prevented to seek individual psychosocial treatment. If a participant needs psychosocial care, this will be approved. When participants from either the intervention or the control group receive (additional) psychological treatment during the study period, it will be extensively documented and controlled for in the analyses. When the study is finished, participants from the waitlist group have the opportunity to participate in the intervention.

Assessment of outcome measures takes place with online questionnaires at baseline (before randomization; T0), directly after the intervention period (eight weeks for adolescents, six weeks for parents; T1), six months (T2) and twelve months (T3) after baseline. For adolescents participating in the study, one of their parents is asked to complete questionnaires as well.

This study was approved by the METC of the Academic Medical Center Amsterdam and of the eight participating hospitals.

Randomization

With an average of five participants per intervention group, a total of ten intervention groups for both adolescents and parents will be given. Interventions are organized at different time points (in four to six cohorts, dependent on inclusion rates). In each cohort, participants are randomly allocated to the conditions resulting in an equal number of participants in the intervention and waitlist control condition. The randomization is carried out by an independent IT worker from a company for e-health development who administers the website for *Op Koers Online*, using block randomization software.

Sample size

Earlier studies on the effectiveness of *Op Koers* and comparable effect studies showed effects of medium size (32, 54). Based on a design with four repeated measurements and a within subject correlation of .5, 84 adolescents and 84 parents are needed – 42 in each condition – to detect an intervention effect of medium size (d=.05) over time, at a two-sided .05 significance level and 80% power. Taking into account a dropout of 15% over time, 96 adolescents and 96 parents are needed to achieve the intended power.

OUTCOME MEASURES

Questionnaires

The outcome measures will be assessed by standardized questionnaires with good psychometric qualities and available normative data (Table 2) (26, 55-64). To assess participants' satisfaction with the intervention, content, design and course leaders, participants in the intervention group complete an evaluation questionnaire at the end of the intervention period (T1).

Statistical analyses

Analyses will be performed according to the intention-to-treat principle. Primary and secondary outcomes will be assessed with linear mixed model analyses using SPSS. The intervention is qualified as effective if the intervention group improved more over time on one of the primary outcomes than the control group, at a significance level of 0.05 and at small to medium effect size d (0.2-0.5).

Table 2 Primary/secondary outcome measures, measurements and informant for the adolescent and parent intervention

Primary outcome measures	Measurements	Informant
Adolescents		
Psychosocial functioning	Child Behavior Checklist (CBCL) and Youth Self Report (YSR)	Parent and adolescent
Disease-related coping skills	*Op Koers* Questionnaire (OKQ)	Adolescent
Parents		
Psychosocial functioning	Hospital Anxiety and Depression Scale (HADS)	Parent
Disease-related coping skills	*Op Koers* Questionnaire (OKQ)	Parent

Secondary outcome measures	Measurements	Informant
Adolescents		
Self-esteem	Perceived Competence Scale for Adolescents (CBSA)	Adolescent
Quality of Life	Pediatric Quality of Like Inventory - self report (PedsQL)	Adolescent
Parents		
Impact of the illness on family functioning	Pediatric Quality of Life Inventory - Family Impact Module (PedsQL-FIM)	Parent
Parental distress	Distress Thermometer for Parents (DTP)	Parent
Social involvement	Inventory Social Involvement (ISI)	Parent
Illness cognitions	Illness Cognition Questionnaire for Parents (ISQ)	Parent

DISCUSSION

This paper outlines the study protocol for two multicenter randomized controlled trials on the effects of two cognitive-behavioral based online group interventions: one for adolescents with CI and one for parents of children and adolescents with CI. Earlier studies showed that psychological interventions for children and adolescents with CI, and for parents, can improve psychosocial functioning (22, 23, 32). Also, studies on effectiveness of online interventions showed promising results (40, 65-67). Online interventions are easily accessible and, when not using a webcam, anonymous (38-40). These factors can increase possibility and willingness from participants to apply for a psychosocial intervention. There is a lack of evidence-based online group interventions for adolescents with CI and for parents. Studies in this field are limited. Therefore, this study is unique in focusing on an online cognitive-behavioral group intervention for these populations.

This study has several strong points. First, participation in the intervention and the study are completely online, which eliminates logistical barriers for participants and therefore keeps drop-out rate low. Second, we include nationwide with focus on a heterogeneous group of different medical chronic diagnosis. This way, is easier to achieve a relatively large study sample, which is beneficial for the statistical power. Third strong point is that participants in the intervention group can be divided over treatment groups independent of the hospital, which benefits the feasibility of the study (it is easier to create intervention groups on different time points, this will overcome drop-out due to availability). Finally, the relatively long term follow-up period promotes stronger long-term results.

Some vulnerabilities have also to be taken into account. First, since recruitment of adolescents for health studies is challenging (68, 69), the intervention for adolescents is at risk for recruitment problems or delay. This could be a threat to the inclusion rates and the statistical power of the study. Second, due to the relatively long follow-up period it is possible that participants will seek other psychosocial support in the study period. This could bias the results. Lastly, since we include nationwide, it is impossible to identify the approached group and to determine non-response.

In conclusion, adolescents with CI and parents of children and adolescents with CI are at risk for developing psychosocial problems. Easily accessible online evidence-based interventions are needed. This study aims to contribute to research on effective interventions for adolescents with CI and parents of children and adolescents with CI by investigating two separate group interventions, for adolescents and for parents. If this study shows significant effects of the interventions on improving psychosocial wellbeing and disease related coping skills in adolescents and/or parents, *Op Koers Online* will be implemented in clinical practice.

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5

Online psychosocial group intervention for adolescents with a chronic illness: A randomized controlled trial

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Internet Interventions

ABSTRACT

Objective The present study aimed to evaluate the efficacy of *Op Koers Online*, a protocolled online psychosocial group intervention for adolescents with a chronic illness (CI).

Methods Adolescents (12-18 years) with different types of CI (N=59; Mean age=15.1 years, SD=1.7; 54% female) participated in a parallel multicenter randomized controlled trial comparing *Op Koers Online* (N=35) with a waitlist control group (N=24). Assessments (online questionnaires) took place at baseline (T0), 6-months (T1) and 12-months follow-up (T2). Primary outcomes were internalizing and externalizing behavioral problems and disease-related coping skills. Health-Related Quality of Life was secondary. Efficacy was tested with linear mixed models.

Results Compared to the control group, the intervention had a significant positive effect ($p<.05$) on disease-related coping skills T1 vs T0 (use of relaxation, $\beta=.68$; social competence, $\beta=.57$) and T2 vs T0 (information seeking, $\beta=.61$), and on HRQoL (social-, school-, psychosocial functioning and total HRQoL) T1 vs T0 ($\beta=.52$ to $\beta=.60$). No intervention effects on internalizing and externalizing behavioral problems were found.

Conclusion The results of this randomized controlled trial indicate a positive effect of *Op Koers Online*. The intervention had beneficial effects on disease-related coping skills and HRQoL.

Practice Implications The next step is to implement *Op Koers Online* for adolescents in clinical practice.

INTRODUCTION

Children and adolescents growing up with a chronic illness (CI; e.g. asthma, diabetes) are at risk for emotional- and social problems such as feeling down and isolating oneself (1-3). When untreated, these problems can cause mental disorders such as depression. During adolescence, patients have the challenge of becoming autonomous, which is extra demanding for those with a CI (4). Considering this, interventions that support adolescents with a CI and teach them how to cope with their illness are essential.

Cognitive Behavioral Therapy (CBT) is known as an effective evidence-based psychological treatment for youth with a CI (5, 6) in improving psychosocial wellbeing (7) and in teaching positive coping skills (8). Coping skills are important mediators of effects of a CI on adaptive psychosocial functioning (9).

Offering CBT interventions via the internet is upcoming because of the logistical and practical benefits (10, 11). For example, patients do not need to visit the hospital because they can participate from home, which is also a great advantage for countries with sparse populations. Moreover, an online environment is appealing to adolescents specifically, since they are generally used to being online (12, 13). Results of online CBT interventions on improving psychological functioning and decreasing disease-related impact are promising. A meta-analysis showed reduction of depressive symptoms, anxious symptoms and general distress (effect sizes from different studies ranging from 0.17 to 0.21) (12).

Online CBT interventions can be offered in individual or group format. Group interventions have important advantages, such as the possibility to share emotions and experiences with others in a similar situation and the fact that therapists can treat more patients simultaneously (9, 14). Moreover, group interventions are proven to be effective in teaching coping skills and improving knowledge about symptom reduction and disease-related problem-solving (9, 15). However, studies that evaluate online group therapies for adolescents with a CI are scarce (16).

Most existing (CBT) group interventions for adolescents with a CI, face to face as well as online interventions, are focused on a specific illness (17, 18), such as epilepsy (8). However, most of the psychosocial problems (e.g. anxiety and depressive symptoms) are the same across illnesses. Generic consequences of having a CI (e.g. feeling different, dealing with food or social restrictions, taking medication) cause psychosocial problems (9). A generic approach that focuses on psychosocial problems associated with the CI rather than the CI itself is therefore suitable and would allow for patients with rare illnesses to participate in a group intervention. Besides, it offers therapists the possibility to treat more patients simultaneously which can be cost-effective.

The intervention in the current study, *Op Koers Online* (English: *On Track Online*) is an online CBT group intervention (chat, without use of video) for adolescents with different types of CI based on the already existing face-to-face interventions *Op Koers* for children, adolescents and parents (19, 20). *Op Koers* face-to-face showed positive results on improving psychosocial functioning (parent-reported internalizing problems and child-reported externalizing problems) and on use of the coping skills information seeking, social competence and positive thinking (20). Pilot studies of *Op Koers Online* had promising results on feasibility and efficacy for adolescents who survived cancer respectively with a CI. Satisfaction rates of both course leaders and participants were high (21). The study of Douma et al (22) showed that participant's use of several coping skills and aspects of Health-Related Quality of Life (HRQoL) improved after following the intervention (21, 22). The pilot studies did not include a control group, therefore more research was needed to establish the effects of the intervention.

In a randomized controlled trial (RCT) we aimed to answer the following research question: Is *Op Koers Online* for adolescents with a CI an efficacious intervention? We hypothesized that *Op Koers Online* for adolescents had a positive effect on adolescent's internalizing and externalizing behavioral problems, disease-related coping skills and HRQoL.

METHODS

Study Design

A multicenter parallel RCT comparing the intervention to a control group (waitlist control group) was designed in accordance with the Standard Protocol Items: Recommendations for Intervention Trials Checklist (CONSORT-Checklist). In this RCT we used the assessments (online questionnaires) that were completed at baseline (before randomization; T0), at 6-months (T1) and 12-months (T2) follow-up from baseline. Full details of the study protocol and the intervention were reported previously by Douma et al (23) (registry number ISRCTN83623452). Approval of the Medical Ethical Committee of the Amsterdam University Medical Centers was obtained for this study. Participants from both the control and intervention group received care-as-usual and were not prevented to seek individual psychosocial treatment.

Procedure

The study was conducted between July 2016 and April 2019. Participants were recruited between September 2016 and June 2018 from outpatient clinics of nine participating hospitals via information letters and pamphlets. Healthcare professionals were asked to invite adolescents in person to participate in the study. Additional nationwide recruitment was done via patient associations and online advertisement. Interested adolescents could use a reply form or send an e-mail. After a positive reply, adolescents (and their parents) were phoned to assess eligibility and to obtain informed consent. Inclusion criteria were aged between 12 and 18 years with a physical CI diagnosis, according to the following criteria set forth by Mokkink et al: 1) onset between ages 0 and 18; 2) diagnosis based on medical scientific knowledge; 3) the illness is not (yet) curable; and 4) the illness has been present for at least three months or at least three episodes have occurred in the last year (24). Furthermore, having access to a laptop/computer/tablet with internet connection was necessary to participate in the intervention and to complete questionnaires. Exclusion criteria were having cognitive disabilities or language problems which limited the ability to participate in the intervention and/or to fill out questionnaires.

Randomization

The randomization into intervention and control group was carried out by an independent IT worker from a company for e-health development who administers the website for *Op Koers Online*. Block randomization with block size four was performed, based on a previously generated randomization schedule with allocation ratio 1:1. Because the recruitment period was spread out over time, randomization was done at five time points. In case of an incomplete randomization block, participants were assigned to the intervention group, to assure enough participants to give the group intervention. When randomized in the intervention group, the researcher called the participant to schedule the intervention. Participants in the control group were given the opportunity to participate in the intervention after the final follow-up assessment. The researchers were not blinded to group assignment.

Intervention

Op Koers Online consists of eight weekly 90-minutes sessions and a booster session 4 months after the last session. The goal of the intervention is to prevent and/or reduce psychosocial problems by teaching the use of engaged coping skills using CBT techniques, such as cognitive restructuring and relaxation. Five coping skills were taught; 1) information seeking and providing information about the illness, 2) use of relaxation techniques in stressful situations, 3) increasing knowledge of self-management and medical compliance, 4) improving social competence and 5) positive thinking (cognitive restructuring) (19, 23). The learning goals of the intervention and examples of learning activities are shown in Table 1.

Sessions took place at a scheduled time in a secured chatroom (Figure 1) with three to six participants and two qualified psychologists (course leaders) who carry out the protocolled intervention. The intervention was designed without the use of a video camera to ensure anonymity as much as possible and keep the threshold for participation low. Turn taking during the sessions was managed by the course leaders. After each session, course leaders filled out a log providing information about the session: particularities of participant's absence or situation, any technological issues and whether or not course leaders followed the intervention protocol. The log was checked by the coordinating researcher. Assessment of the log did not show any major deviations of the intervention protocol.

Table 1 Learning goals and examples of learning activities of Op Koers Online for adolescents

		Learning goals				
		Information seeking and giving about the illness	*Use of relaxation during stressful situations*	*Increase knowledge of self-management and compliance*	*Enhancement of social competence*	*Positive thinking (cognitive restructuring)*
Examples of learning activities of Op Koers Online for adolescents	*Instruction/modelling*	Education about sources of information	Relaxation exercise (MP3)	Group discussion about own treatment and (non-)compliance	Video and group discussion: how and what do you tell others about your illness	Thinking-Feeling-Doing game
	Session[1]	Session 3	Session 2	Session 4	Session 5 and 6	Session 1, 7 and 8
	Reinforcement/practice (homework)	Write down questions you have, and look for answers	Practice the relaxation exercise	Write down situations for non-compliance and how to improve compliance	Think of what CAN you do (instead of CANNOT) and write down your story for the other group members	Write down positive adjustments for negative thoughts

[1] The session that focused on the specific learning goal was mentioned here. However learning goals are applied throughout the whole course (for example: use of relaxation techniques and Thinking-Feeling-Doing are discussed in multiple sessions).

Hi all, welcome in the Op Koers Online chatroom!
Linde & Miriam 11:24

Today's focus lies on introducing ourselves and getting to know each other...
Linde & Miriam 11:25

We are Linde and Miriam and we work as child psychologists at the Emma Children's Hospital...
Linde & Miriam 11:25

We are the course leaders during this course.
Linde & Miriam 11:26

Sarah, do you want to start introducing youself?
Linde & Miriam 11:26

Yes I do!
Sarah 11:30

My name is Sarah and I am 14 years old
Sarah 11:32

Hi Sarah!😃
Charlotte 11:32

I live at my parents with my two younger brothers...
Sarah 11:33

And I have a skin condition called atopic eczema which means my skin is red and ithes almost all day long
Sarah 11:39

Getting to know each other

1. What is your name?
2. How old are you?
3. Which school/class do you go to?
4. Do you have any hobbys?
5. Which chronic illness do you have?
How does it affect your daily life?

Figure 1 Chat room of *Op Koers Online*. Left screen: chat text; right screen: information screens/videos/exercises; strip below (left): field where participant writes their text with possible use of the emojis; strip below (right): names of participants who are present in the chatroom. *Note: this participants and text in this chat room are fictitious.*

All course leaders (N=22 in total; all female and psychologist) from nine participating hospitals are extensively trained during an 8-hour workshop in using the detailed intervention protocol. The workshop was led by the coordinating researchers of the study and took place in the Emma Children's Hospital. It included a theoretical part and exercises to learn how to use the website.

Participants log on to the website (www.opkoersonline.nl) to enter the chatroom and their own personal environment to complete homework assignments. To improve adherence, participants received a small gift (e.g. memory game) after the last session as a reward for participating and completing homework assignments.

Measures

Primary outcomes

Behavioral problems were assessed with the Child Behavior Checklist (CBCL; 6-18 years, parent report) and the Youth Self Report (YSR; 11-18 years) (25, 26), consisting of items about problem behavior during the past six months. Two broadband scales were used in the present study: internalizing problems and externalizing problems. Internalizing problems (range 0-42) included the subscales anxious-depressed and withdrawn-depressed. Items from the subscale somatic complaints were disregarded in the analyses (27). Externalizing problems (CBCL range 0-70, YSR range 0-64) included the subscales rule-breaking behavior and aggressive behavior. Higher scores indicate more problem behavior. The internal consistency of the internalizing and externalizing scales was satisfactory across time points, with Cronbach's α=.78 to α=.99.

Disease-related coping skills were assessed with the Op Koers Questionnaire (OKQ) (19, 22). Adolescents were asked to what extent (1 “always/almost always” to 4 “almost never/never”) they agree with statements about the use of coping skills taught in *Op Koers Online* (e.g. “I know how to get answers about my disease”). The items are divided into five subscales: information seeking (6 items), relaxation (3 items), social competence (6 items), positive thinking (3 items) and medical compliance (8 items). Higher scores reflect more use of engaged coping skills. Internal consistency was moderate to satisfactory across time points, with Cronbach’s α=.61 to α=.92, except for relaxation at T0 (α=.51) and medical compliance at T0 (α=.45). The last was excluded from the analyses.

Secondary outcomes

HRQoL was assessed with the Pediatric Quality of Life Inventory version 4.0 Generic Core Scales (PedsQL™4.0) (28). Items are divided into 4 domains: physical health, emotional functioning, social functioning and school functioning. Two summary scores are computed: psychosocial health (emotional, social and school functioning) and total score (all domains). Scores range from 0 to 100, with higher scores indicating better HRQoL. Internal consistency for the PedsQL was satisfactory across time points, with Cronbach’s α=.69 to α=.95.

Socio-demographic characteristics were obtained from parents via an online socio-demographic questionnaire at baseline: family composition, socioeconomic status (income), gender and ethnicity. Adolescent’s stressful life-events and the use of psychological care besides *Op Koers Online* were also obtained.

Within the same questionnaire, *illness characteristics* (illness type, duration, severity) were obtained. Parents rated illness severity using a proxy measure based on the occurrence of the following 13 possible consequences of CI in the past year (scale 0-13): doctor visits, hospitalization, surgery, use of medication, dietary consequences, visible malformations, non-visible malformations, use of appliances, limitations in movements, problems with hearing, vision and speech (0=no, 1=yes) and course of the disease (0=improving/stable, 1=deterioration/unstable).

Statistical Analysis

A priori power analysis based on the detection of an intervention effect of medium size with d≈.50, indicated a required number of 84 study participants. Post-hoc power calculations based on the inclusion rates of the current study with three time points indicated that differences of medium size (*d*=.65) between study groups over time at a significance level of .05 with a power of .80 could be detected (29). Preliminary analyses examined baseline differences between the intervention and control group on socio-demographic and illness characteristics and on the outcome variables. To characterize the sample, externalizing problems and HRQoL at T0 were compared to Dutch norms (25, 26, 30) with one-sample tests. Comparison of internalizing problems was not possible because norm scores were not available for internalizing problems without somatic complaints.

Linear mixed model analyses were performed to examine efficacy of the intervention accounting for dependency of data within participants. Correcting the analyses for dependency within study groups was not necessary, as the intra-class correlation coefficients were not significant (or below .05) (31). Outliers on outcome measures were rescaled according to Tabachnick and Fidell (32). Intention-to-treat analyses were performed based on the random allocation, using the mixed-model procedure in SPSS (19.0) with Full Maximum Likelihood estimation. Participants were included in the efficacy analyses if data at baseline (T0) were available as well as data at T1 and/or T2. Missing data were not imputed. To facilitate interpretation of regression coefficients, all continuous scores were standardized, expressing deviations from the mean at T0. For binary coded variables, standardized regression coefficients of 0.2 were considered small, 0.5 medium and 0.8 large (33).

Dependent variables were parent- and self-reported internalizing and externalizing problems disease-related coping skills and HRQoL. Linear mixed models were fitted with a random intercept and fixed slopes for study group (intervention vs control), time (T1 vs T0 and T2 vs T0) and the interaction term study group x time. This interaction tested the effects of the intervention. Potential differences between intervention and control group on outcome measures at T0 were controlled by the random intercept. An alpha of .05 was used to test the statistical significance of the effects.

RESULTS

Sample Characteristics

Figure 2 shows the participant flow from recruitment to follow-up. Most applicants (56%) applied after seeing online advertisement. Of those who received an invitation letter, 3% applied. Of the applicants, 23% dropped out before randomization. Main reason for drop-out was that they were not available at the times scheduled for the sessions.

Regarding the intervention, course leaders reported no study-related adverse events. As recorded by the course leaders in the log, 88% of all sessions was carried out according to the protocol. The sessions that were not, concern the first session in which a lot of time was spent on explaining the intervention and getting to know each other. Course leaders were advised to do so for creating group cohesion and, when necessary, move some treatment elements to the next session.

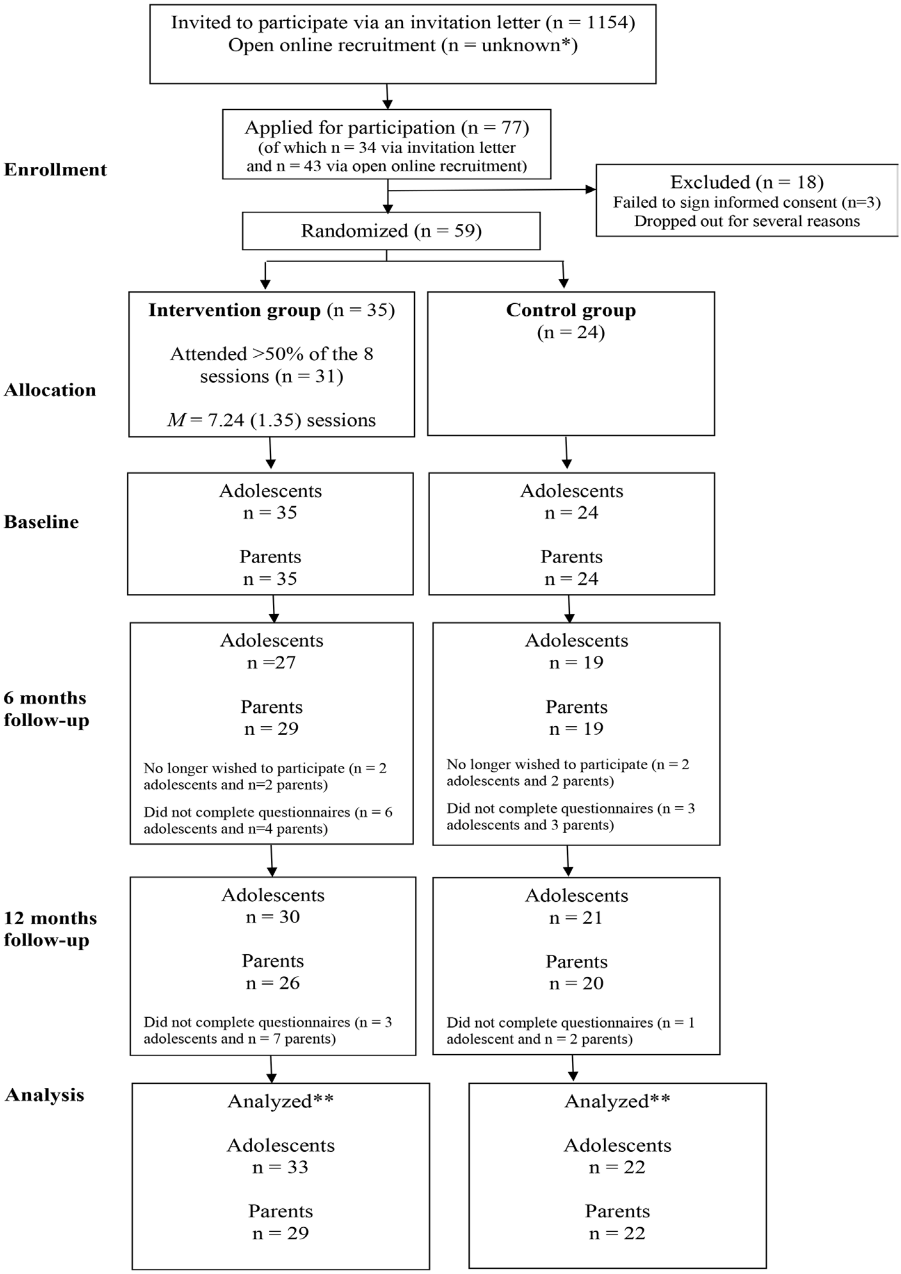

Figure 2 Participant flow through the study.
* Open recruitment was done via patient associations, social media and advertisements on websites. The number of adolescents reached online is unknown. ** Linear mixed model analyses included all available data from each subject with a baseline and at least one follow-up assessment up to withdrawal or study completion.

Note: parents were not involved in the intervention. They were asked to fill out a socio-demographic questionnaire once and a questionnaire about their child at each time-point.

In total, 59 participants were randomized, of which 35 were assigned to the intervention group and 24 to the control group. A total of 31 (89%) participants from the intervention group attended more than 50% of the sessions. Mean attendance was 7.2 (SD=1.4) sessions. In the end, 55 participants were included in the analyses, of which 33 in the intervention group (Mean age=15.1 SD=1.7; 45.5% female) and 22 controls (Mean age=15.0 SD=1.8; 63.6% female). Compared to the Dutch norms (p<.05), our sample reported significantly fewer problems on baseline self-reported externalizing problems (26) and worse HRQoL (physical, psychosocial, emotional, social, school and total) (30). At baseline, 13% of participants scored in the clinical range for internalizing problems; 5% of the participants scored in the clinical range for externalizing problems. There were no significant differences in socio-demographic characteristics at baseline between the intervention and control group (Table 2). The intervention group had significantly worse HRQoL (physical, social, school, psychosocial and total) than the control group at baseline.

During the intervention period, 11% (n=2) of the adolescents in the control group received alternative psychosocial care (individual psychological treatment). At 6-months follow-up, 11% in the control group and 14% in the intervention group received additional psychosocial care. At 12-months follow-up, this was 14% in the control group and 28% in the intervention group; these percentages did not differ significantly (*p*=.26).

Table 2 Sample socio-demographic characteristics.

	Intervention group (N = 33)	Control group (N = 22)	*p* [1]
Age *(in years)*	15.1 (1.7)	15.0 (1.8)	.824
Gender			.186
Male	54.5%	36.4%	
Female	45.5%	63.6%	
Income			.390
< Modal	24.1%	14.3%	
≥ Modal	75.9%	85.7%	
Ethnicity *(based on country of birth of the parent of the participant)*			.230
Dutch	72.7%	86.4%	
Non-Dutch	27.3%	13.6%	
Diagnosis			
Asthma	18%	23%	
Type 1 Diabetes	18%	14%	
IBD	3%	9%	
Auto-immune disease (JIA)	6%	5%	
Hashimoto's disease	3%	9%	
Other [2]	52%	40%	
Illness Duration (*in years)*	8.66 (5.79)	7.95 (5.22)	.651
Illness Severity *(scale 0-13)*	4.06 (1.72)	3.55 (1.14)	.658

Table 2 Continued.

	Intervention group (N = 33)	Control group (N = 22)	p [1]
Former psychological help			.849
Yes	20%	17.6%	
No	80%	82.4%	

[1] Group differences tested with independent samples t-tests for continuous variables and χ2-tests for categorical variables. [2] All other diagnoses occurred once.

Primary outcomes

Internalizing and Externalizing Problems (CBCL, YSR)

The intervention had no significant effect on the change in parent-reported and self-reported internalizing or externalizing problems over time (Table 3). None of the interactions of study group x time were significant.

Disease-related coping (OKQ)

Significant beneficial effects of the intervention (study group x time *p*<.05) on use of coping skills were found at 6-months follow up (T1 vs T0) for relaxation (β .68, *p*=.0.011) and social competence (β=.57, *p*=.030; see Table 3, Figure 3 and Figure 4). The intervention group improved over time while the control group did not improve. No significant effects on relaxation and social competence were found at 12-months follow-up (T2 vs T0). The intervention effect on information seeking was marginally significant at 6-months follow up (β=.52, *p*=.063) but significant at 12-months follow up (β=.61, *p*=.026; Figure 5).

Secondary outcomes

Health-related Quality of Life (PedsQL)

Significant beneficial effects of the intervention (study group x time, *p*<.05) were found at 6-months follow up (T1 vs T0) on social functioning, school functioning and on the summary scales psychosocial health (Figure 6) and total HRQoL (Figure 7); β ranging from .52-.60, *p* ranging from .011 to .029 (Table 3). While HRQoL in the intervention group improved from T0 to T1, HRQoL of the control group did not improve from T0 to T1 or worsened.

No significant effects were found at 12-months follow-up (T2 vs T0), though the marginal significance of the regression coefficients of social functioning (β=.41, *p*=.052) and psychosocial functioning (β=0.42, *p*=0.081; Figure 5) indicated a long-term effect of the intervention at T2.

Table 3 Means, Standard Deviations at baseline (T0), 6-month (T1) and 12-month follow-up (T2) and results of linear mixed model analyses.

	Intervention group			**Control group**			**Study group*time**			
	T0	T1	T2	T0	T1	T2	T0-T1 intervention vs waitlist		T0-T2 intervention vs waitlist	
	M (SD)	M (SD)	M (SD)	M (SD)	M (SD)	M (SD)	β	*p*	β	*p*
Behavioral problems (YSR)	N = 33	N = 27	N = 30	N = 22	N = 19	N = 21				
Internalizing (without somatic complaints)	9.39 (5.29)	9.78 (7.72)	9.90 (6.82)	7.50 (6.14)	8.00 (6.46)	8.24 (7.01)	-.34	.140	-.24	.287
Externalizing	8.67 (6.37)	7.15 (6.04)	7.07 (4.47)	6.27 (4.14)	7.21 (5.82)	6.62 (4.85)	-.34	.129	-.28	.195
Behavioral problems (CBCL)	N = 29	N = 29	N = 26	N = 23	N = 19	N = 20				
Internalizing (without somatic complaints)	7.48 (7.25)	5.93 (5.41)	6.65 (6.24)	7.74 (5.31)	6.53 (5.56)	5.45 (5.40)	-.02	.928	.21	.354
Externalizing	5.17 (5.54)	4.14 (4.16)	5.08 (4.70)	5.96 (3.82)	5.89 (5.95)	4.30 (3.61)	-.05	.815	.32	.140
HRQoL (PedsQL)	N = 33	N = 27	N = 30	N = 22	N = 19	N = 21				
Physical health	59.85 (22.25)	68.40 (22.22)	65.94 (24.28)	78.27 (18.84)	77.63 (20.92)	80.21 (18.26)	.34	.056	.17	.309
Emotional functioning	69.09 (18.48)	71.67 (18.55)	71.17 (18.55)	75.91 (19.00)	70.53 (20.06)	70.95 (23.80)	.35	.212	.29	.272
Social functioning	71.82 (17.63)	76.67 (15.81)	76.17 (17.75)	87.95 (13.42)	82.89 (15.75)	85.00 (16.12)	**.56**	.011	.41	.052
School functioning	60.61 (17.22)	63.70 (17.90)	60.50 (17.14)	72.27 (15.41)	65.26 (16.20)	66.90 (17.50)	**.55**	.029	.31	.198
Psychosocial functioning	67.17 (13.66)	70.68 (14.53)	69.28 (14.04)	78.71 (12.92)	72.89 (15.12)	74.29 (16.13)	**.60**	.017	.42	.081
Total HRQoL	64.62 (15.09)	69.89 (15.55)	68.12 (16.53)	78.56 (13.66)	74.54 (16.09)	76.35 (16.06)	**.52**	.015	.33	.107
Disease-related coping skills (OKQ)	N = 33	N = 27	N = 30	N = 22	N = 19	N = 21				
Information seeking	1.97 (.60)	2.51 (.45)	2.45 (.61)	2.23 (.61)	2.41 (.63)	2.34 (.62)	.52	.063	**.61**	.026
Use of relaxation	1.77 (.71)	2.21 (.66)	2.06 (.71)	1.89 (.70)	1.84 (.83)	1.95 (.70)	**.68**	.011	.30	.232
Social competence	1.72 (.62)	2.12 (.44)	1.94 (.59)	1.79 (.61)	1.80 (.70)	1.92 (.64)	**.57**	.030	.13	.613
Positive thinking	1.73 (.74)	2.15 (.68)	2.20 (.61)	1.70 (.86)	1.95 (.75)	1.92 (.87)	.21	.521	.31	.325

Note: Significant (p<.05) intervention effects (β) are presented in bold.

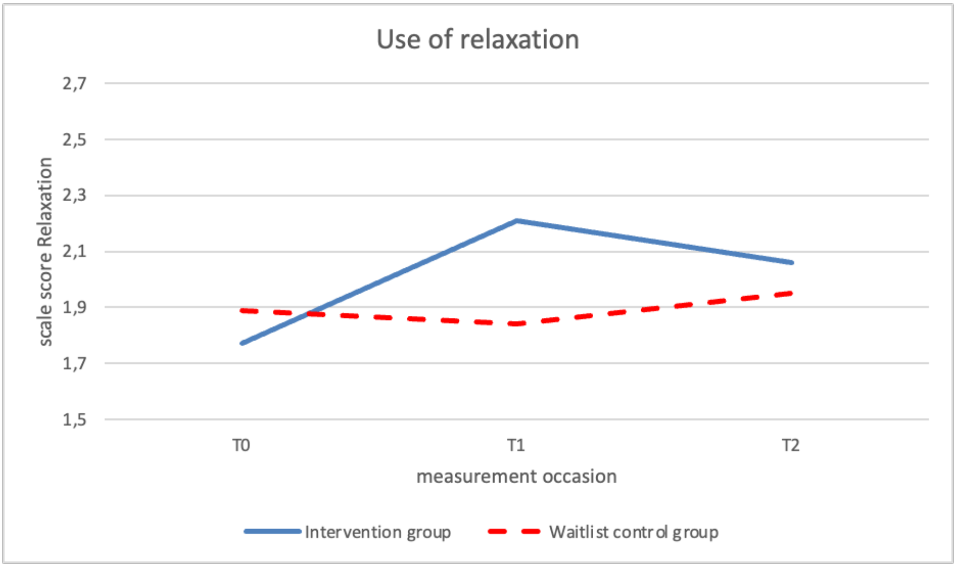

Figure 3 Use of the disease-related coping skill relaxation measured with the *Op Koers* Questionnaire, at T0 (baseline), T1 (6 months follow-up), and T2 (12 months follow-up); intervention (*Op Koers Online*) and waitlist control group.

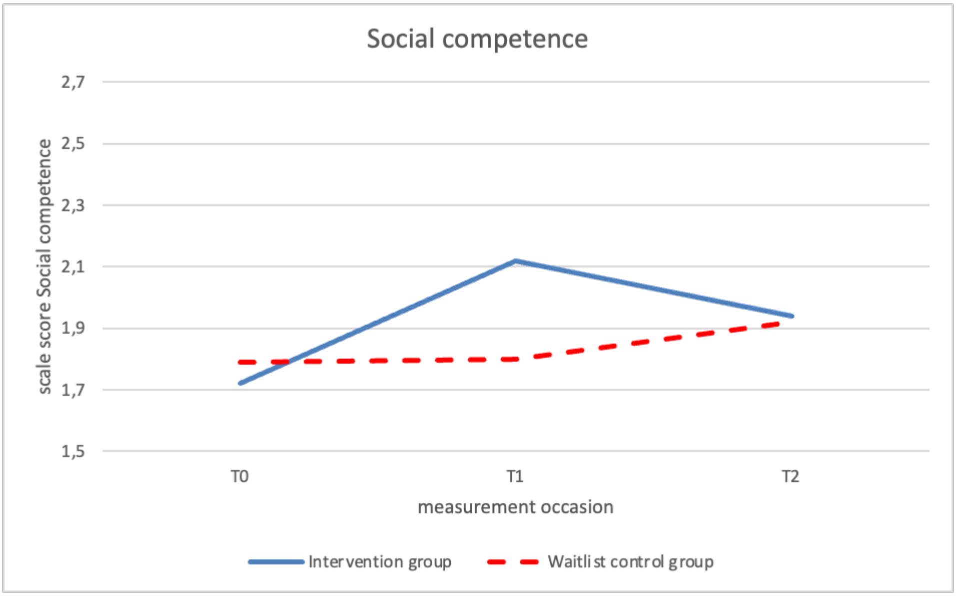

Figure 4 Use of the disease-related coping skill social competence measured with the *Op Koers* Questionnaire, at T0 (baseline), T1 (6 months follow-up), and T2 (12 months follow-up); intervention (*Op Koers Online*) and waitlist control group.

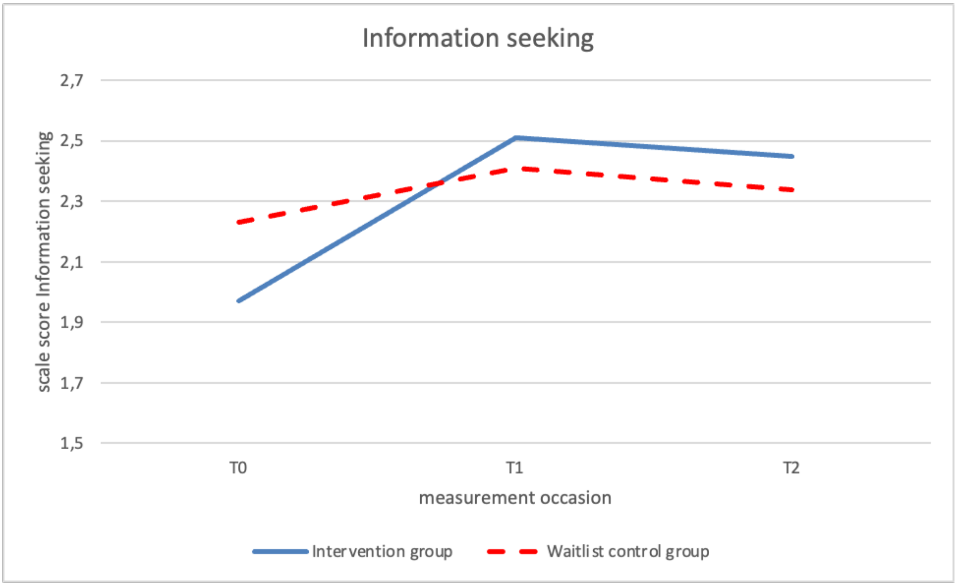

Figure 5 Use of the disease-related coping skill information seeking measured with the *Op Koers* Questionnaire, at T0 (baseline), T1 (6 months follow-up), and T2 (12 months follow-up); intervention (*Op Koers Online*) and waitlist control group.

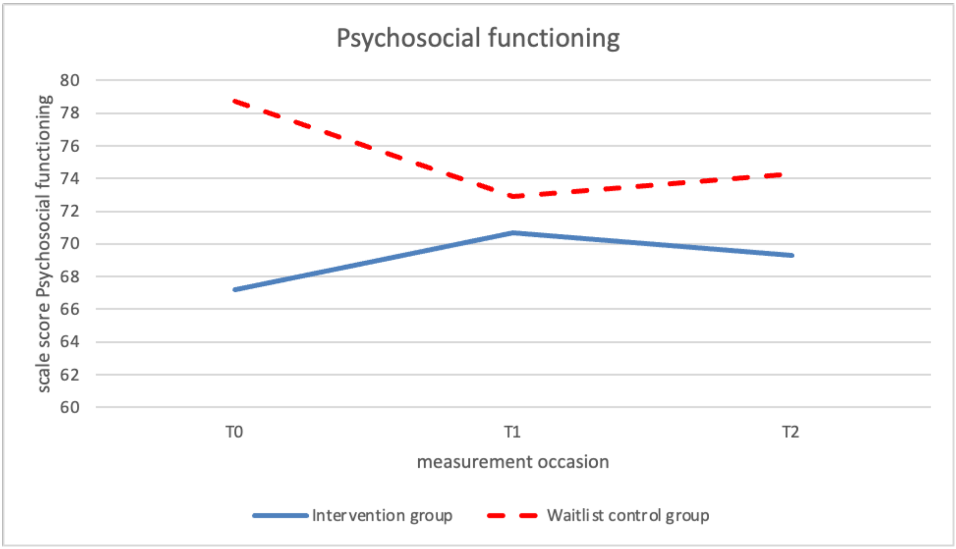

Figure 6 Psychosocial functioning measured with the Pediatric Quality of Life Inventory version 4.0 Generic Core Scales (PedsQL™4.0), at T0 (baseline), T1 (6 months follow-up), and T2 (12 months follow-up); intervention (*Op Koers Online*) and waitlist control group.

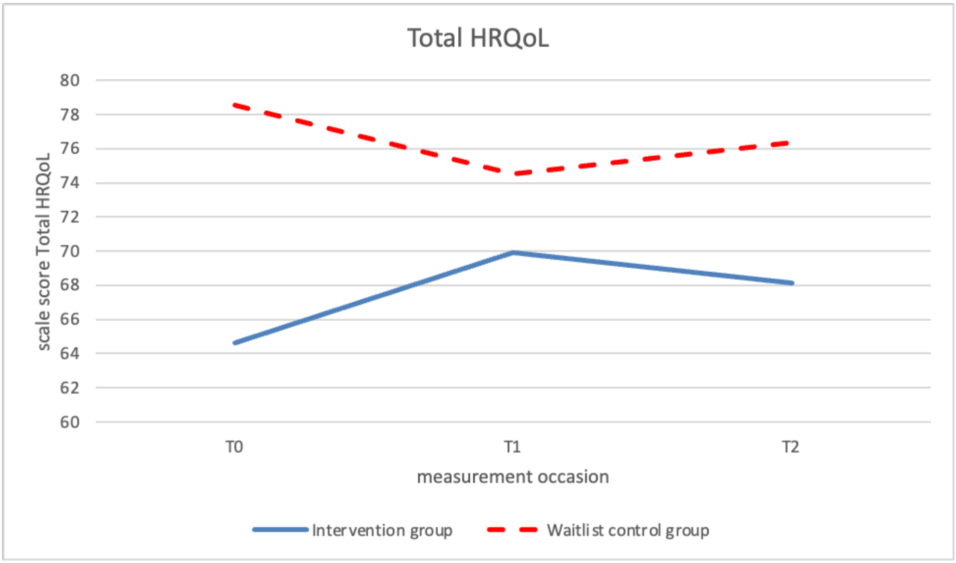

Figure 7 Total Health-Related Quality of Life measured with the Pediatric Quality of Life Inventory version 4.0 Generic Core Scales (PedsQL™4.0), at T0 (baseline), T1 (6 months follow-up), and T2 (12 months follow-up); intervention (*Op Koers Online*) and waitlist control group

DISCUSSION

The results of this RCT indicate a positive effect of *Op Koers Online* for adolescents, an online psychosocial group intervention for adolescents with different types of CI: the intervention had a positive effect on disease-related coping skills (use of relaxation, social competence and information seeking) and on HRQoL (social, school, psychosocial and total). The effect sizes were medium with standardized regression coefficient β's between .52 and .68, which was in line with earlier studies on the efficacy of psychosocial interventions for youth with a CI (20, 34).

We found no statistically significant intervention effect on parent- and self-reported internalizing and externalizing problems in the present RCT nor in the previous pilot study on *Op Koers Online for adolescents*. Possibly, this is due to the fact that *Op Koers Online* is a preventive as well as curative intervention (22) so that *Op Koers Online* is not explicitly designed to decrease psychopathology symptoms. Furthermore, having clinical levels of behavioral problems was not an inclusion criterion, thereby limiting the chance to achieve a decrease in internalizing and externalizing problems by the intervention. The absence of intervention effects on internalizing and externalizing problems in the present study is not in line with the results of the previous RCT on *Op Koers face-to-face*. Since the intervention effects of *Op Koers face-to-face* were of small size (20), just as the effects in the present RCT on *Op Koers Online*, one could assume that lack of power in the present study explained the discrepancies between the statistically significant effects of *Op Koers Online* and *Op Koers face-to-face*.

Recruitment was problematic. The response rate on information letters was lower than expected based on former research on *Op Koers* face-to-face (20). Even though we experienced that face-to-face recruitment works best, healthcare professionals had trouble motivating adolescents to participate in the study. Therefore, the intended sample size was not reached. Inclusion problems in RCT's, especially with adolescents, are common (35). Given their age and puberty, adolescents are a hard group to motivate for any intervention, possibly especially a psychosocial group intervention. However afterwards, we found that the adolescents enjoyed the intervention. Satisfaction was high and two-third of the participants would definitely recommend the intervention to peers, while a quarter reported that they maybe would recommend the intervention (data not shown). Healthcare providers should be (more) aware of this, and should pay more attention explaining to adolescents what the intervention entails and why participation could be helpful. Next to medical doctors, it is important to involve nurses in this process. Furthermore, it is important to note that engaging participants in the design and delivery of interventions and integrating their feedback may increase participants' engagement in interventions.

The intervention effect on coping skills diminished at one-year follow-up with exception of information seeking and giving about the disease, which is an important factor for good adaption to living with the disease. In the study of Scholten et al. (2013) (20) regarding *Op Koers face-to-face*, the long-term intervention effects were stronger when parents were involved. In the face-to-face intervention, parents and children/adolescents participated in separate, parallel groups and parents learned how to support their child in daily life. *Op Koers Online* for adolescents was intentionally designed without involving parents, with the argument that it keeps the threshold for participation low and gives adolescents the opportunity to participate independently from their parents. However, separately from the intervention for adolescents, the *Op Koers Online* program offers a module for parents, *Op Koers online* for parents. It aims to prevent and/or reduce psychosocial problems of parents by teaching the adaptive coping skills related to their child's disease. *Op Koers online for parents* was recently positively evaluated (36). *Op Koers Online* for parents intervention can be recommended to parents of participating adolescents, to support parents in how to cope with their child's CI. Future research should focus strengthening the long-term effects by involving parents.

With this RCT, we contribute to the literature with an evaluation of a unique online CBT group intervention for adolescents with different types of CI. The study has some limitations. First limitations are the unknown recruitment and enrollment rates and the lack of information about non-respondents, cause of partially online open recruitment. Second, we relied on self-reported outcome measurements, which has the risk for socially desirable answers or concealing of symptoms. This could have led to an overestimation of the intervention effect and should be taken into account while interpreting the results.

Furthermore, the outcomes relaxation and positive thinking had moderate internal consistency. On the one hand, the use of scales with moderate internal consistency is acceptable for group comparisons because the internal consistency is an indication of random error and has nothing to do with systematic error (bias). On the other hand, Cronbach's alphas should preferably be .7 or higher because the lower the internal consistency, the larger the random measurement error, and so, the more difficult to detect differences between groups.

A priori power analysis was based on the detection of an intervention effect of medium size with $d \approx .50$, while the post hoc power analysis revealed that with the current sample size we were still able to detect an intervention effect of medium size with $d = .65$. Furthermore, there were differences between participants in the two study groups on HRQoL at baseline. The mixed models analyses corrected for baseline differences between intervention and controls but the intervention group might have had more room for improvement.

Future efforts should focus on maintaining the effects on coping skills at one-year follow-up and it would also be interesting to investigate whether the effects of the intervention on HRQoL and are mediated by the disease-related coping skills taught during the intervention.

CONCLUSION

The results of this randomized controlled trial indicate a positive effect of an innovative online psychosocial group intervention for adolescents with all kinds of CI. After following the intervention, participant's use of adaptive coping skills and their HRQoL improved.

Practice implications

The *Op Koers Online* intervention is an important addition to the pediatric field to support adolescents with a CI, and contribute to their HRQoL. A big advantage is the possibility to participate from home, so that additional hospital visits are not necessary. Healthcare professionals (medical doctors, nurses, etc.) should be involved in recruitment, and should be aware of the necessity of motivating adolescents to participate. The *Op Koers Online for parents* intervention can be offered to parents of participating adolescents (36). The next step is to support more adolescents by using *Op Koers Online* for adolescents in regular clinical practice. At the same time, the intervention needs to be investigated more widely to keep improving the content and confirm the positive outcomes.

Financial Disclosure

The authors have no financial relationships relevant to this article to disclose.

Funding source

This study was funded by Fonds NutsOhra (FNO; project number: 100.977), a social fund for vulnerable groups in Dutch society. FNO had no role in the study design.

Declarations of Interest

None.

Clinical Trial registry name

ISRCTN

Registration number

ISRCTN83623452.

Abbreviations

Chronic illness: CI; Randomized Controlled Trial: RCT; Health Related Quality of Life: HRQoL.

Acknowledgements

We thank the adolescents and their parents who participated in this study and acknowledge the contributors of the staff at the participating centers (with special thanks to the local investigators: I. M. Visser, E. M. van Dijk-Lokkart, E. Kakes, W. Meerstra, Y. Krol and S. Jansen-Kamphorst). Finally, we acknowledge Fonds NutsOhra for funding this study.

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6

Online psychosocial group intervention for parents: Positive effects on anxiety and depression

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Journal of Pediatric Psychology

ABSTRACT

Objective To evaluate the efficacy of an online psychosocial group intervention for parents of children with a chronic illness, in terms of anxiety and depression, and disease-related coping skills.

Methods Parents (N=73) participated in a parallel multicenter randomized controlled trial comparing an intervention group to a waitlist control group. In the group intervention *Op Koers Online* (English: *On Track Online)* parents learned how to use adaptive coping strategies taught with cognitive behavioral therapy and acceptance and commitment therapy techniques. Assessments (online questionnaires) took place at baseline (T0), 6-months (T1) and 12-months (T2) follow-up. Mixed-model analyses were performed to test the difference in change in anxiety, depression, and disease-related coping skills between intervention (N=34) and waitlist control group (N=33).

Results Compared to the waitlist control group, the intervention had a significant positive effect ($p<.05$) on changes in anxiety, depression and total score T1 vs T0 ($\beta=-.47$ to $-.51$) and T2 vs T0 ($\beta=-.39$ to $-.46$), the coping skills open communication, relaxation, social support, acceptance, predictive control ($\beta=.42$ to $.88$) and helplessness ($\beta=-.47$) T1 vs T0 and relaxation and positive thinking T2 vs T0 ($\beta=.42$ to $.53$).

Conclusions Parental anxiety and depression decreased, and use of adaptive coping skills improved after the intervention. The online character, the focus on parents themselves instead of on their child and the possibility for parents of children with rare illnesses to participate, are innovative and unique aspects of *Op Koers Online* for parents. The next step is to implement the intervention in clinical practice.

INTRODUCTION

Parents of children with a chronic illness (CI) have to deal with several stressors about their child's health including practical stressors (e.g. managing daily routines) as well as emotional challenges (e.g. worrying) (1-5). Therefore, these parents are at risk for psychosocial problems (6-9) such as anxiety and depression (10, 11). Parents with psychosocial problems may have more difficulties managing their child's illness effectively (1). Moreover, parental depressive symptoms have found to be correlated to negative parenting practices (12), poor adherence (13) and an increase in children's illness symptoms over time (14). Given the negative consequences of parental psychosocial problems for parents themselves as well as their children, interventions to support parents of a child with a CI are needed (11).

Transactional models of child adaption to CI recognize the importance of numerous psychosocial risk- and protective factors that could be targeted and modified in interventions. The disability-stress-coping model of Wallander and Varni (1998) implies that the stressors faced by children and parents dealing with pediatric CI are multifaceted (e.g. restriction of activities, responsibility of treatment), and that the links between illness related stressors and adjustment are moderated by appraisals and coping strategies on which several personal and family risk- and protective factors are of influence (15, 16).

The model of Wallander and Varni (1998) was adapted for the current study to explain outcomes for parents of a child with a CI (Figure 1). Coping style, known as the way people react to stressful situations, plays an important role in the model. Engaged coping (e.g. problem solving, cognitive restructuring) is proven to be more effective for good psychosocial adjustment, including anxiety and depression, than disengaged or passive coping (e.g. self-criticism, social withdrawal) (17, 18). Several personal (e.g. temperament), family (e.g. parent-child relationship) and environmental factors (e.g. practical and emotional support from others) have an effect on parents' coping style (19) and are therefore important determinants to address in an intervention. When engaged coping can be reached, parents will likely have better psychosocial adjustment and outcomes and more capability to manage their child's illness effectively.

Cognitive behavioral therapy (CBT) and Acceptance and Commitment Therapy (ACT) are examples of therapies to teach how to use engaged coping strategies. CBT and ACT have been shown effective in improving psychological outcomes such as stress and health-related quality of life of children with a CI and their families (20-23). CBT involves identifying unhelpful thoughts, challenging them, and replacing them with helpful thoughts (24). ACT, as a part of CBT, aims to reduce the influence of negative thoughts on daily life by accepting certain thoughts or situations. Exercises such as mindfulness and values elicitation to orientate participants to thoughts and activities which are in line with personally meaningful values are often used (25). ACT could be helpful for parents of a child with a CI, since those parents often face situations which cannot be changed and ACT provides helpful tools to learn how to deal with such situations.

Studies have shown that sharing experiences with others in a similar situation is associated with a decrease of distress and improvement of mental health (26-28). Offering group interventions to parents may therefore be an effective way to support them. Most existing group interventions which involve parents are child-focused: the parents learn how to support their child in managing the illness (16, 21, 23). During the past years, there is more attention to psychosocial support focusing on parents themselves (29). However, parents often experience practical barriers for participation in interventions, such as travel time, distance and costs, taking care of the children and absence from work (30, 31). An online intervention can increase participation rates: it eliminates or decreases those barriers because participation from home is possible (32, 33).

Considering this, an online psychosocial CBT/ACT group intervention called *Op Koers Online* (English: *On Track Online*) for parents, focusing on parents themselves, was designed in close cooperation with parents. Parents were asked via an open access survey and focus groups what they would like in an intervention (34). The intervention is partly based on the already existing *Op Koers* face-to-face intervention for children, adolescents and parents, which was proven to be effective (16), and on *Op Koers Online* for adolescents of which pilot studies showed promising results on feasibility and preliminary efficacy (35, 36). *Op Koers Online* is chatroom delivered without the use of video to ensure anonymity, which can be beneficial because it could be easier to talk about problems when nobody knows or sees you (Maurice-Stam et al., 2014).

In a randomized controlled trial (RCT) we aimed to answer the following research question: Is *Op Koers Online* for parents of a child with CI an efficacious intervention? We hypothesized that *Op Koers Online* for parents had a positive effect on parental anxiety and depression and on the use of adaptive coping skills. Outcomes were symptoms of anxiety and depression, and disease-related coping skills, including skills that match the content of the intervention and more generic illness cognitions (see Figure 1).

METHODS

Study design

In a parallel RCT we used the data of the online questionnaires that were completed at baseline (before randomization; T0), at 6-months (T1) and at 12-months (T2) follow-up from baseline. Approval of the Medical Ethical Committee of the Amsterdam University Medical Centers was obtained for this study. Full details of the study protocol and the intervention content were reported previously (registry number ISRCTN83623452) (37). Participants from both the waitlist control and intervention group received care-as-usual and were not prevented to seek individual psychosocial treatment.

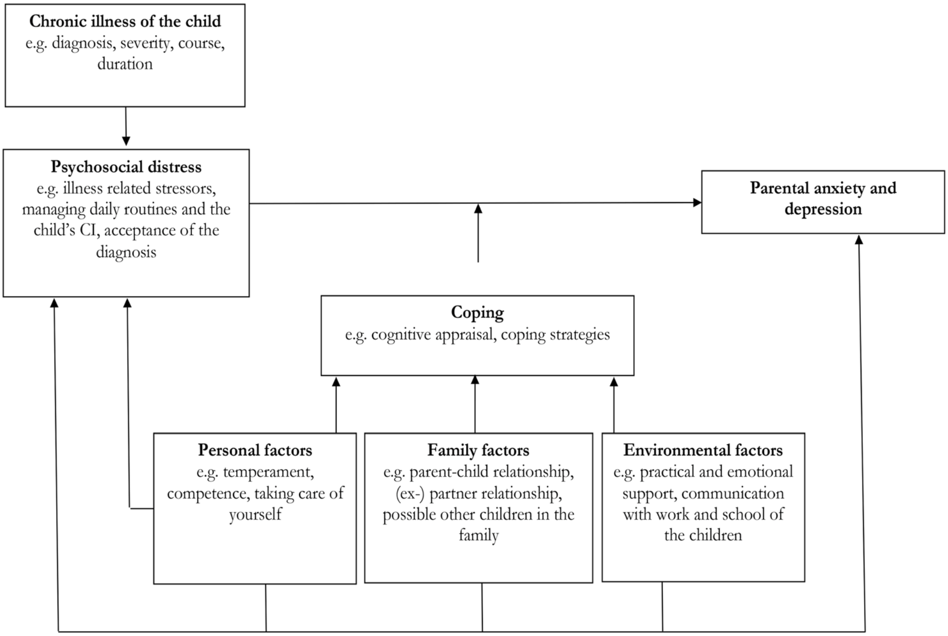

Figure 1 Adapted version of the disability-stress-coping model of Wallander and Varni (1998) to explain anxiety and depression in parents of a child with a CI.

Procedure

The study was conducted between July 2016 and April 2019. Participants were recruited between July 2016 and March 2018. Recruitment was done via the outpatient clinics of nine public hospitals all over The Netherlands with information letters and pamphlets, and across the Netherlands through patient associations with online advertisement and social media. Interested parents could use the reply form attached to the information letter or pamphlet, or could send an e-mail. After a positive reply, parents were phoned by the researcher, a psychologist, to assess eligibility and to obtain informed consent. Inclusion criteria were being caregiver of a child between the ages of 0 and 18 years with a physical CI diagnosis, according to the following criteria set forth by Mokkink et al.: 1) onset between ages 0 and 18; (2) diagnosis based on medical scientific knowledge; (3) the illness is not (yet) curable; and (4) the illness has been present for at least 3 months, or at least three episodes have occurred in the last year (38). Furthermore, having access to a laptop/computer/tablet with internet connection was necessary to participate in the intervention and to complete online questionnaires at home or at another location. Exclusion criteria were having cognitive disabilities or language problems which limited the ability to participate in the intervention and/or to fill out questionnaires. Participants received a voucher of EUR 20 after completing all assessments.

Randomization

The randomization into intervention and waitlist control group was carried out by an independent IT worker from a company for e-health development who administers the website for *Op Koers Online*. Block randomization with block size four was performed, based on a previously generated randomization schedule with allocation ratio 1:1. Because the recruitment period was spread out over time, randomization took place at four time points. When randomized in the intervention group, the researcher called the participant to schedule the intervention. Participants in the waitlist control group were given the opportunity to participate in the intervention after the final follow-up assessment. The researchers were not blinded to group assignment.

Intervention

The intervention consists of six weekly 90-minutes sessions and a booster session 4 months after the last regular session. The goal of the intervention is to prevent and/or reduce psychosocial problems by teaching the use of adaptive disease-related coping skills. In the first session parents get to know each other and receive explanation about the intervention. The following sessions each focus on a theme: session 2) the CI of the child, 3) relationships within the family, 4) taking care of yourself and 5) relationships with others and practical support (e.g. school of the child, work). In the sixth and last session there is time to repeat topics or to resume discussions that have not been finished due to lack of time (34). Coping skills were addressed in every session. Depending on the subjects that came up during the session, course leaders addressed the different coping skills accordingly. To teach coping skills, CBT and ACT techniques, such as relaxation, cognitive restructuring and exercises focusing on accepting the child's diagnosis, are used (37).

Sessions take place at a scheduled time in a secured chatroom with three to five parents under supervision of two psychologists (course leaders), who carry out the protocolled intervention. After each session, course leaders filled out a log providing information about the session: particularities of participant's absence or situation, any technological issues and whether or not course leaders followed the intervention protocol. The log was checked by the coordinating researcher. Assessment of the log did not show any major deviations of the intervention protocol.

All course leaders (N=22 in total; all female) from the participating hospitals are extensively trained during an 8-hour workshop in using the detailed intervention protocol. The workshop was led by the coordinating researchers of the study and took place in the Emma Children's Hospital. It included a theoretical part and exercises to learn how to use the website.

Groups are composed based on the age of the children as much as possible for better match within groups. Parents log on to the website (www.opkoersonline.nl) to enter the chatroom, to read additional material about the themes, and to complete homework assignments in their personal environment. An extensive login manual with an explanation in person was provided to guide the parents through logging on and using their own personal environment.

Measures

Socio-demographic and illness characteristics

Socio-demographic characteristics were obtained with an online questionnaire assessing age, gender, income, educational level, ethnicity and former psychological help. Illness characteristics (diagnosis, duration, severity) of the child (see Table 1) were assessed with this questionnaire as well. Parents rated illness severity using a proxy measure based on the occurrence of the following 13 possible consequences of CI in the past year (scale 0-13): doctor visits, hospitalization, surgery, use of medication, dietary consequences, visible malformations, non-visible malformations, use of appliances, limitations in movements, problems with hearing, vision and speech (0=no, 1=yes) and course of the disease (0=improving/stable, 1=deterioration/unstable).

Anxiety and depression

The Hospital Anxiety and Depression Scale (HADS) was used to assess anxiety and depression (39, 40). This 14-item questionnaire is divided into two seven-item scales; anxiety (e.g. “I feel tense”), depression (e.g. “I have lost interest in my appearance”) and a total sum score of all items. Items are scored on a four-point Likert scale (range 0 = not at all to 3 = very often). Higher scores indicate more anxiety or depression in the past week. A scale score of 8 or above (cut-off score) is considered as an indicator for clinically significant anxiety or depression for both men and women, which means that the parent may need professional help. Internal consistency (Cronbach’s alpha) was moderate to good (range .61 for anxiety to .88 for depression) in the current study. Norm data representative of Dutch parents between 20 and 60 years old are available (41).

Disease-related coping skills

The Op Koers Questionnaire for Parents (OKQ-P) assesses the use of disease-related coping skills taught in the intervention (16). Parents are asked if they agree with 25 statements on a four-point Likert scale (range 1 = almost never/never to 4 = always/almost always). Higher scores reflect use of more engaged coping skills. Items are divided into four subscales: open communication (9 items; e.g. “I can inform the people around me about the impact of the illness of my child myself”), relaxation (8 items; e.g. “I know how to relax myself”), social support (4 items; e.g. “I am able to ask for support to the people around me”) and positive thinking (4 items; e.g. “I know how to worry less about the consequences of the illness of my child”). Internal consistency (Cronbach’s alpha) was moderate to good (range .57 for positive thinking to .78 for relaxation) in the current study.

The Illness Cognition Questionnaire for Parents (ICQ-P) measures parental illness cognitions that reflect different ways of assigning meaning to the CI of their child (42, 43).

Parents have to indicate to what extent statements fit their situation on a four-point Likert scale (ranging from 1 = not at all to 4 = completely). Higher scores indicate that statement fits the respondent's situation to a larger degree. Items are divided over three six-item subscales; helplessness (e.g. "The illness of my child controls my life"), acceptance (e.g. "I can handle the problems related to my child's illness") and disease benefits (e.g. "Dealing with the illness of my child has made me a stronger person"). Cronbach's alphas were satisfactory (range .79 to .93) in the current study.

The Cognitive Coping Strategies Scale Parent Form (CCSS-PF) assesses to what extent parents of children with a chronic or life-threatening illness try to maintain a sense of cognitive control, by relying on cognitive coping strategies (44, 45). The predictive control strategy was used in the current study. This domain consists of five statements (e.g. "When I think about the illness of my child, I assume it will be all right"), on which parents can indicate on a four-point Likert scale (ranging from 1= totally agree to 4 = totally disagree) to what extent they agree. Item scores are recoded so that higher scores indicate that parents are more optimistic about the course of the child's disease. Cronbach's alphas were good (range .80 to .85) in the current study.

Data Analyses

Post-hoc power calculations based on the inclusion rates of the current study with three time points indicated that differences of medium size (*d*=.59) between study groups over time at a significance level of *p*=.05 with a power of .80 (46) could be detected. Preliminary analyses examined baseline differences between the intervention and waitlist control group on socio-demographic and illness characteristics and on the outcome variables. To characterize the sample in terms of anxiety and depression, percentage of parents with scores in the clinical range were compared with a Dutch norm group with Chi-squared tests.

Linear mixed models analyses were performed to examine efficacy of the intervention accounting for dependency of data within participants. The intra-class correlation coefficients indicated that correcting the analyses for dependency within treatment groups was not necessary, as they were not significant (or below .05). Outliers on outcome measures were rescaled according to Tabachnick and Fidell (2013) (47). Intention-to-treat analyses were performed based on the random allocation, using the mixed-model procedure in SPSS (19.0) with Full Maximum Likelihood estimation. Participants were included in the analyses if data at baseline (T0) were available as well as data at T1 and/or at T2. Missing data were not imputed. To facilitate interpretation of regression coefficients, all continuous scores were standardized, expressing deviations from the mean at T0. For binary coded variables, standardized regression coefficients of 0.2 were considered small, 0.5 medium and 0.8 large (48).

Dependent variables were anxiety, depression and HADS total score, and disease-related coping skills. Linear mixed models were fitted with a random intercept and fixed slopes for study group (intervention vs waitlist control), time (T1 vs T0 and T2 vs T0) and the interaction term study group x time. Potential differences between intervention and waitlist control group on outcome measures at T0 were controlled for by the random intercept. Because no differences were found between the intervention and the control group, no other variables were added to the models. An alpha of .05 was used to test the statistical significance of the effects.

RESULTS

Sample Characteristics

Figure 2 shows the participant flow from recruitment to follow-up. The exact number of invited parents was unknown and information of non-respondents was lacking, because recruitment was partly done through online advertisement and patient associations. A total of 98 parents applied for participation. The majority of all applicants (57%) applied after seeing an online advertisement. Of the 98 applicants, 25 (26%) dropped out before randomization mostly due to practical problems (most common: expecting to be unable to spend enough time on study participation and/or following the intervention due to a busy family life, work, etc.). In the end, 73 parents were randomized, of whom 67 (92%) could be included in the analyses.

Of the 67 parents who were included in the analyses, 34 were assigned to the intervention group (Mean age = 42.40 SD = 6.10; 100% female) and 33 to the waitlist control group (Mean age = 42.90 SD = 5.82; 97% female). No significant differences in socio-demographic and illness characteristics (Table 1), psychosocial problems and outcome variables at baseline were found between the intervention and the waitlist control group. Regarding psychosocial problems at baseline, 47% of the participants in the intervention group and 27% in the waitlist control group showed clinically significant anxiety symptoms. The percentages for clinically significant depression symptoms were 29% and 24% respectively. Participants in both the intervention and the waitlist control group scored significantly more often in the clinical range than the norm group ($p<.05$) (8, 41), except for the waitlist control group on anxiety ($p=.24$). No adverse events occurred during the RCT.

Anxiety and depression

Significant beneficial effects of the intervention ($p<.05$) were found at 6- and 12-months follow-up (T1 and T2 vs T0) for symptoms of anxiety (Figure 3), symptoms of depression (Figure 4) and the total score on the HADS; regression coefficients ranged from $\beta=-.39$ (anxiety T2 vs T0) to $\beta=-.51$ (total score T1 vs T0; Table 2). The intervention group improved over time compared to baseline (T0) while the waitlist control group did not.

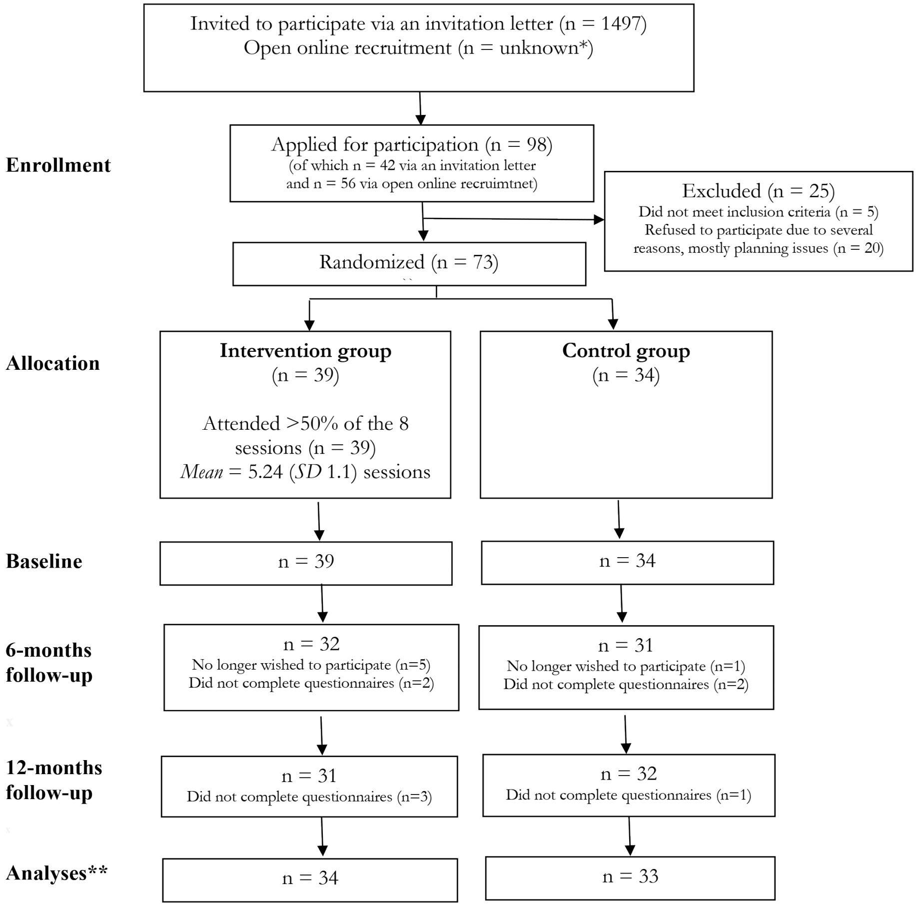

Figure 2 Participant flow through the study.
* Open recruitment was done via patient associations, social media and advertisements on websites. The number of parents reached online is unknown. ** Number of participants with baseline data and at least one follow-up assessment completed.

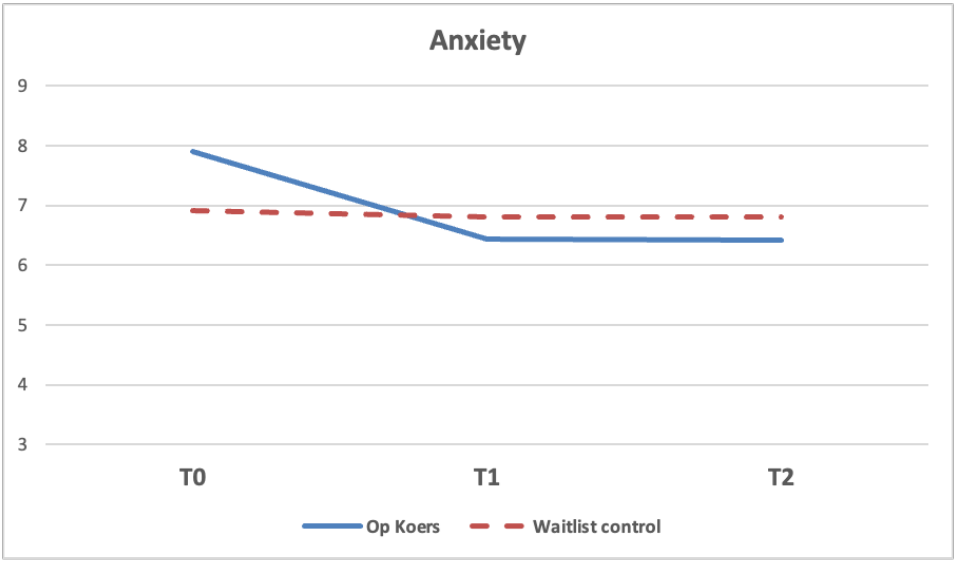

Figure 3 Symptoms of anxiety measured with the Hospital Anxiety and Depression Scale (HADS) at T0 (baseline), T1 (6-months follow-up) and T2 (12-months follow-up); intervention (*Op Koers* Online) and waitlist control group.

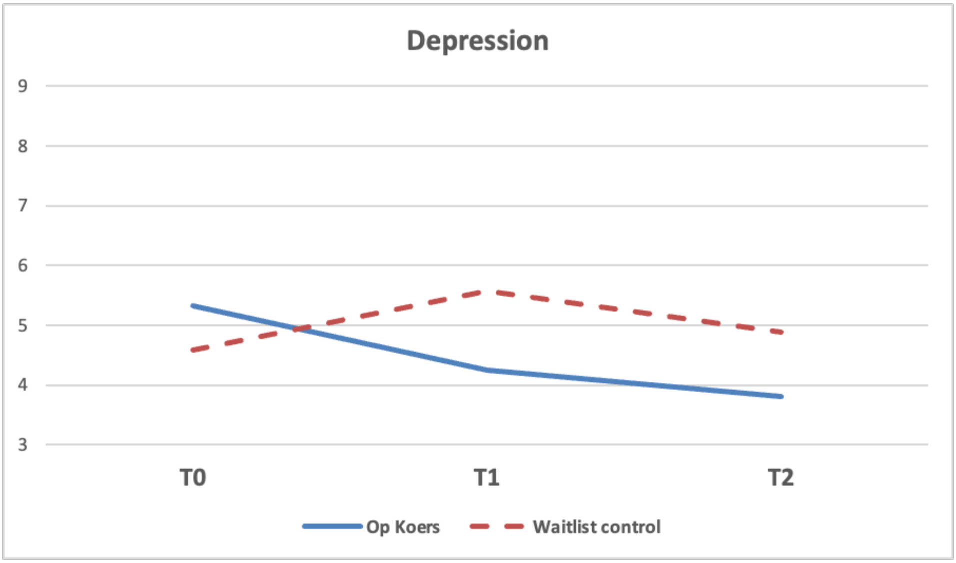

Figure 4 Symptoms of depression measured with the Hospital Anxiety and Depression Scale (HADS) at T0 (baseline), T1 (6-months follow-up) and T2 (12-months follow-up); intervention (*Op Koers* Online) and waitlist control group.

Table 1 Sample socio-demographic characteristics of parents included in the analyses.

	Intervention group (N = 34)	Control group (N = 33)	*p* [1]
Age *(in years)*	42.40 (6.10) Range 30.21 – 59.07	42.90 (5.82) Range 32.32 – 54.92	.712
Gender			.293
Male	0%	3%	
Female	100%	97%	
Income			.371
< Modal	27%	37%	
≥ Modal	73%	63%	
Ethnicity			.452
Dutch	78%	85%	
Non-Dutch	22%	15%	
Diagnosis of the child			
Asthma	5%	6%	
Epilepsy	5%	6%	
Type 1 Diabetes	11%	29%	
IBD	11%	0%	
Kidney disease	11%	6%	
Other [2]	57%	53%	
Illness Duration (*in years)*	5.95 (4.69)	6.18 (4.41)	.832
Illness Severity *(scale 0-13)*	5.16 (1.97)	5.03 (1.90)	.773
Former psychological help			.890
Yes	81%	82%	
No	19%	18%	
Educational level parents			.190
Low/intermediate [3]	43%	59%	
High [4]	57%	41%	

[1] Group differences tested with independent samples t-tests for continuous variables and χ2-tests for categorical variables [2] Other diseases occurred once [3] Primary education, lower and middle vocational education, lower and middle general secondary education, higher secondary education and pre-university education [4] Higher vocational education and university

Disease-related coping skills

Significant beneficial effects of the intervention ($p<.05$) on use of disease-related coping skills were found at 6-months follow-up (T1 vs T0) for open communication, relaxation, social support (OKQ-P), helplessness and acceptance (ICQ-P), predictive control (CCSS-PF); regression coefficients ranged from $\beta=.42$ (acceptance) to $\beta=.88$ (predictive control; Table 2). Significant effects at 12-months (T2 vs T0) follow-up were found for relaxation ($\beta=.42$) and positive thinking ($\beta=.53$), while the intervention effect on helplessness was marginally significant ($\beta=-.36$, p=.067; Table 2). Overall, the intervention group improved over time compared to baseline (T0) while the waitlist control group did not.

Table 2 Means, Standard Deviations at baseline (T0), 6-month (T1) and 12-month follow-up (T2) and results of linear mixed model analyses.

	Intervention group			Control group			Intervention*time			
	T0 N ≈ 34	T1 N ≈ 32	T2 N ≈ 31	T0 N ≈ 33	T1 N ≈ 31	T2 N ≈ 32	T0-T1 Intervention vs control group		T0-T2 Intervention vs control group	
	M (SD)	M (SD)	M (SD)	M (SD)	M (SD)	M (SD)	β	*p*	β	*p*
Anxiety and depression (HADS)										
Anxiety	7.91 (3.02)	6.44 (3.25)	6.42 (2.98)	6.91 (3.41)	6.81 (2.97)	6.80 (3.22)	**-.47**	.014	**-.39**	.043
Depression	5.32 (3.91)	4.25 (3.36)	3.81 (4.14)	4.58 (3.63)	5.58 (4.30)	4.88 (4.26)	**-.49**	.020	**-.43**	.039
Total	13.24 (6.22)	10.69 (5.94)	10.23 (6.75)	11.48 (6.50)	12.39 (6.53)	11.68 (6.75)	**-.51**	.007	**-.46**	.016
Coping (OKQ)										
Open communication	3.10 (.50)	3.39 (.40)	3.33 (.48)	3.12 (.55)	3.17 (.48)	3.18 (.54)	**.43**	.018	.28	.120
Relaxation	2.74 (.57)	2.94 (.49)	2.98 (.52)	2.73 (.53)	2.62 (.45)	2.69 (.47)	**.49**	.020	**.42**	.043
Social support	2.84 (.57)	3.09 (.55)	3.07 (.56)	2.85 (.73)	2.73 (.68)	2.88 (.74)	**.52**	.025	.25	.271
Positive thinking	2.64 (.35)	2.82 (.37)	2.89 (.41)	2.60 (.37)	2.60 (.29)	2.61 (.36)	.37	.168	**.53**	.045
(ICQ-P)										
Helplessness	5.58 (3.69)	4.13 (2.79)	3.97 (3.18)	4.39 (3.47)	4.87 (3.59)	4.41 (3.47)	**-.47**	.017	-.36	.067
Acceptance	10.21 (4.39)	12.28 (4.39)	13.06 (3.73)	12.58 (3.29)	12.58 (3.16)	13.62 (3.28)	**.42**	.037	.30	.134
Disease benefits	10.42 (4.49)	11.22 (4.05)	11.61 (5.15)	10.15 (4.51)	10.00 (5.02)	10.12 (4.62)	.16	.379	.21	.253
(CCSS-PR)										
Predictive control	2.64 (.28)	2.83 (.62)	2.83 (.52)	2.53 (.38)	2.37 (.62)	2.50 (.64)	**.88**	.025	.47	.223

Note: Significant ($p < .05$) intervention effects (β) are presented in bold.

DISCUSSION

This RCT indicated evidence for the efficacy of *Op Koers Online* for parents, an online psychosocial group intervention for parents of children with different types of CI: the intervention had a positive effect of medium size on symptoms of parental anxiety and depression and on the disease-related coping skills open communication, relaxation, social support, positive thinking, helplessness, acceptance and predictive control. Until now, suitable interventions for parents with a focus on parents' own mental health and behavior have been lacking (49). This study contributes to the field of pediatric psychology with a protocol- and evidence-based intervention for this vulnerable group of parents. The online character, the focus on parents themselves instead of on their child and the possibility for parents of children with rare illnesses to participate, are innovative and unique aspects of *Op Koers Online* for parents. Moreover, this study shows the gateway of the internet for offering interventions to parents. The possibility to participate from home, thereby limiting logistical and practical barriers, increases the opportunities for participation.

Effects of *Op Koers Online* for parents on anxiety and depression symptoms were significant and of medium size at both follow-up moments. This is an important outcome, as depression in parents can cause poorer outcomes for the child (13). According to the model of Wallander and Varni and our adapted version of that model (Figure 1), parents with better psychosocial functioning are more able to support their child with a CI adequately (1), which will likely be related to better psychosocial and illness-related outcomes for the children (12, 14). Therefore, following *Op Koers Online* benefits the parents as well as the children. *Op Koers Online* is an important addition to the toolbox of clinicians to support parents of a child with an easily accessible group intervention.

Regarding disease-related coping skills, some of the significant intervention effects of medium size at 6-months diminished at 12-months follow-up. At 12-months, only effects on the coping skills relaxation and positive thinking were found. According to the literature, coping skills could be expected to mediate the effect of the intervention on parental anxiety and depression outcomes. In our study, continued reliance on relaxation and positive thinking may explain the long-term significant outcomes on anxiety and depression. Future research should examine the working mechanism of *Op Koers Online* and the possible mediating effect of relaxation and positive thinking. Furthermore, attention should be paid to the other coping skills and maintaining the effects on the long-term.

During the recruitment period, we had problems with recruiting the desired amount of participants, which is common in RCT's (50). When implementing the intervention in clinical practice, it is important to keep in mind that professionals should invest time and effort in recruitment and guiding parents through the online environment of the intervention. Luckily, although the sample size was slightly smaller than intended, this was hardly at the expense of the power of the study. A priori power analysis was based on the detection of an intervention effect of medium size with $d \approx 50$, while the post hoc power analysis revealed that with the current sample size we were still able to detect an intervention effect of medium size with *d*=.59.

In this RCT, almost all participants were mothers. We experienced that fathers are hard to reach due to several reasons, mostly having lack of time and indicating no need for support. However, we know from the literature that fathers of children with Down syndrome do express a need for psychosocial support (51). We recognize this finding as there were some fathers that were interested in the intervention, however, they expected mostly mothers to participate and were hesitant of participating in a group with only female participants. Although we tried to encourage those fathers to participate, this was a reason not to do so for some of them. A possible solution could be to schedule separate groups for fathers once or twice a year. In the future, caregivers should offer this option and should pay attention to motivating fathers to participate.

Strengths of the current study included 1) recruitment of participants across the whole country from nine centers and via patient associations and 2) using a standardized intervention protocol which is essential for conducting sound intervention research and for implementation in clinical practice. The study had some limitations as well. First, the unknown recruitment and enrollment rates and the lack of information about non-respondents, because of the partially online open recruitment. Second, outcomes were based on mothers, which limits the representativeness of the results. Third, we relied on self-reported outcome measurements, which had a risk for socially desirable answers or concealing of symptoms. This could have led to an overestimation of the intervention effect and should be taken into account while interpreting the results. Fourth, the baseline levels of anxiety and depression seemed higher in the intervention group than in the control group. Though the differences were not significant and the mixed models analyses corrected for baseline differences between intervention and control group, the intervention group might have had more room for improvement. Finally, the internal consistency of anxiety, open communication, social support and positive thinking was moderate on one or more time points. On the one hand, the use of scales with moderate internal consistency is acceptable for group comparisons because the internal consistency is an indication of random error and has nothing to do with systematic error (bias). On the other hand, Cronbach's alphas should preferably be .7 or higher because the lower the internal consistency, the larger the random measurement error, and so, the more difficult to detect differences between groups. Considering the significant betas for anxiety, open communication, social support and positive thinking (see Table 2), we can conclude that intervention effects were not overlooked due to moderate Cronbach's alpha.

In conclusion, the results indicated that *Op Koers Online* for parents has a positive effect on maternal anxiety and depression and use of disease-related coping skills. The next step for clinical practice is to help more parents by implementing *Op Koers Online* for parents in hospitals across the country, by training more psychologists in using the manual to carry out the intervention. In the future, *Op* Koers Online can be translated for non-

Dutch speaking parents in the Netherlands.

Funding

This study was funded by Fonds NutsOhra (FNO; grant number: 100.977).

Acknowledgements

We thank the parents who participated in this study and acknowledge the contributions of the staff at the participating centers (with special thanks to the local investigators: I. M. Visser, W. Meerstra, E. M. van Dijk-Lokkart, E. Kakes, S. Jansen-Kamphorst). Finally, we would like to thank Ad Vingerhoets for providing the HADS reference data.

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7

Development, research and implementation of the psychosocial group intervention Op Koers: Lessons learned

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Submitted

ABSTRACT

Objective This paper aims to share the lessons that we have learned in the process of 25 years of development, research and implementation of a psychosocial group intervention for children with illness and their family members (called: *Op Koers*, or *On Track* in English).

Methods Using the National Institutes of Health Stage Model for Behavioral Intervention Development, we critically appraised our activities in the different stages**.** Over the years, we used basic research to create and modify intervention modules in several populations. *Op Koers* was pilot-tested several times and found effective in improving coping skills and emotional functioning in larger efficacy studies. Finally, the intervention is part of usual care in the authors' institutions and was disseminated throughout the Netherlands.

Results Important lessons were learned in the domains of participants and health care providers (engagement, recruitment), research methodology (study design, outcome measures) and environmental conditions (technological advances, funding). Future efforts may be directed towards continuous improvement of the intervention and successful lasting implementation.

Conclusions *Op Koers* is one of few psychosocial interventions that has been both extensively studied and implemented. The main factor for this achievement is the close collaboration between, and the perseverance of the clinical care and research departments.

INTRODUCTION

As a result of advances in pediatrics, an increasing number of children is growing up with chronic illness (CI). In the Netherlands, around 500.000 children have a CI, including approximately 600 children newly diagnosed with cancer each year (1). Children and their families face multiple consequences of the illness and treatment, including changes in daily routines and stressful situations, that impact their psychosocial well-being (2-6).

Children, parents and siblings may benefit from psychosocial support in coping with CI. In the Netherlands, this concerns support from a child life specialist, psychologist and/or a social worker, which is provided in the pediatric setting.

The senior authors of this paper (MG and LS), at the time employed at the psychosocial department of Emma Children's Hospital Amsterdam UMC, developed a psychosocial intervention that aimed to improve psychosocial well-being of families coping with pediatric CI using cognitive behavioral therapy (CBT) techniques: *Op Koers* (in English: On Track). From a preventative point of view, the previous clinical staff thought a group intervention could be beneficial. With a generic approach many children could be reached, which is important in pediatric illnesses with a small number of patients (7). This group intervention teaches active coping skills by giving information, using relaxation techniques, and encouraging self-management, social competence and positive thinking. After initial development for siblings of children with cancer, the intervention was adapted to fit other target populations as well, that is children with CI, of different ages, and their parents. *Op Koers* is currently part of standard care in several health care institutions in the Netherlands.

Although there is a clear need for psychosocial interventions in families with an ill child, and many interventions that have been proven to be effective exist, they often do not reach the targeted population because of barriers that impede their implementation (8, 9). Publicly sharing implementation experiences could inform and promote future implementation of psychosocial interventions, but publications on the implementation of developed and researched interventions in clinical practice are scarce (10, 11).

The use of an established model can be helpful in structurally observing and reporting the development of an intervention. The National Institutes of Health (NIH) Stage Model for Behavioral Intervention Development (further referred to as the stage model) is such a model, and it was developed to identify, define, and clarify the activities involved in behavioral intervention development to facilitate the scientific development of potent and implementable interventions (9). The stage model arranges activities in several stages, that do not occur linearly but in an iterative, recursive manner, and seemed useful to structure our reflections on the development of *Op Koers.* The stage model describes basic science activities as research that occurred before and informs intervention generation or refinement. Feasibility and pilot testing includes preliminary evaluations of a developed or refined intervention, thus providing valuable information for further development, efficacy testing or implementation. If pilot results are promising, research into efficacy, in research or community settings, is the next step. Ultimately, effectiveness research evaluates an intervention while maximizing external validity. Since *Op Koers* was developed in real-life community settings of pediatric hospitals, implying a high degree of external validity, we chose to classify our studies as efficacy research in the present paper. The final stage of the model describes implementation and dissemination, the adoption of a new intervention in clinical settings, and the distribution of information and material to relevant groups.

Our psychosocial research and care staff started with *Op Koers* more than 25 years ago. Over the years we have gained experience with the complex process of development, feasibility and efficacy research and implementation, which resulted in a large number of peer reviewed publications. See Table 1 in the supplementary material for an overview. To enable others to learn from our experiences, the current paper aims to critically appraise our efforts guided by the NIH stage model and discuss the lessons that we have learned along the way. We describe the intervention as it is now, followed by our activities in development, evaluation and implementation of the intervention. We finish with the lessons we learned.

THE INTERVENTION: *ON TRACK* (*OP KOERS*)

The *Op Koers* program includes specific modules for children with CI or cancer, in different age groups, for different family members, for different diagnosis groups, and provided either face-to-face or online (Figure 1). Currently, 15 different modules are available.

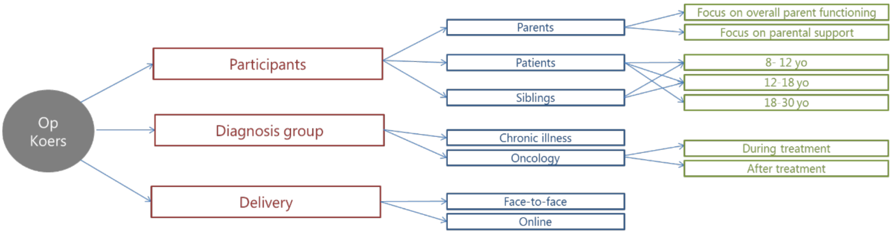

Figure 1 Different modules of the *Op Koers* program

Summary of general components of The *Op Koers* Intervention program

- Courses consist of five to eight 90-minute group sessions that take place face-to-face (in the hospital or online (in a secured chat room, no audio or video) on a set time and day of the week.
- Six months after the start of the intervention, a booster session takes place.
- Every group consists of 3-6 participants and two course leaders, one of whom is a

specialized health care psychologist and the other can be a junior psychologist or social worker.

- In between the sessions, participants complete homework assignments. Additional in-depth reading material is available in the parents and young adults modules
- The first session is meant for the participants to get to know each other. In the last session, participants look back on the course and what they have learned.
- Every session in between focusses on a specific coping skill or a theme, both during the courses and in the in-depth information and homework assignments.

o *Child with illness and siblings*

Coping skills are translated into five learning goals: 1) information seeking and information giving about the disease (‘good to know better’ principle), 2) use of relaxation during stressful situations, 3) increasing knowledge of self-management and compliance (not applicable for siblings), 4) enhancing social competence (group discussions, role playing), and 5) positive thinking (use of the Thinking-Feeling-Doing model; replacement of inaccurate thoughts)

o *Young adults and parents*

Themes are structured around different environments of the participants (Figure 2). Learning goals are either discussed in the chat session, and/or are a part of the homework assignments and the in-depth reading material. Learning goals are: 1) use of relaxation during stressful situations; 2) increasing knowledge of self-management and compliance (of their child); 3) positive thinking; 4) positive parenting (not applicable for young adults); 5) open communication about the illness and seeking and accepting support.

- During the group sessions, psycho-education is used to reach the learning goals (through, among others, informative videos and group discussions), and reinforced and practiced through exercises (such as role-play in the face-to-face sessions and games in the online sessions).
- In the courses for patients and siblings in pediatric oncology, an oncologist joins for one session to answer participants’ questions about the illness or treatment.

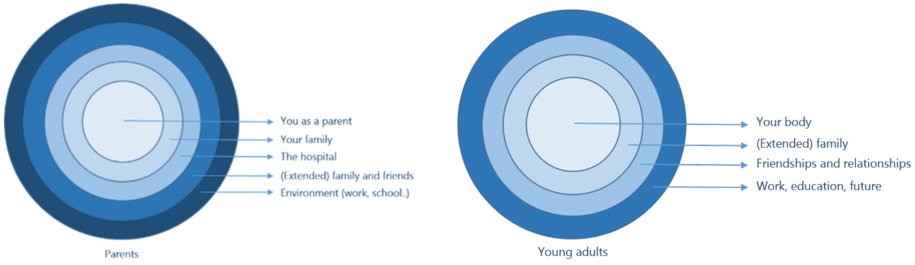

Figure 2 Environments in which the themes of the *Op Koers* interventions for parents and for young adults were structured.

Intervention development, evaluation and implementation activities

In this section we describe our activities in intervention development, evaluation and implementation. These activities occurred non-linearly, but were clustered in a) basic research, b) generation and refinement of the intervention, c) feasibility and pilot testing, d) efficacy research, and finally e) implementation and dissemination. All studies were approved by the medical ethics board of the concerned hospitals and in*formed consent was obtained from all participants.*

a) Basic Research

Op Koers finds its basis in knowledge from the field of pediatric psychology, which focuses on the impact of pediatric illness, in particular on psychosocial outcomes and risk- and protective factors in children and their family members. Since the late 1990s our research groups have conducted a large number of studies focusing on the health-related quality of life and psychosocial functioning of children with CI including pediatric oncology (12-15), as well as their siblings (16) and parents (17-20). Overall, these studies, in line with literature, revealed that families can experience less favorable health-related quality of life and psychosocial functioning than reference groups. Although most families are resilient and only a small proportion is at risk for developing (severe) psychosocial problems (21), standards of care for children with a CI or cancer recommend access to psychosocial interventions for children and family member to facilitate their wellbeing (22, 23).

Wellbeing is impacted by the multiple stressors that families of a child with an illness have to face according to the model presented by Wallander and Varni (24). The relation between stressor and wellbeing is mediated by coping skills, which in itself are impacted by personal, family and environmental factors. Coping is central in the model because it plays a crucial role in the adaptation to stressful situations such as illness of the child. Coping consists of cognitive and behavioral efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of a person (25). These theories were translated into a psychosocial support model (26), based on emotions and coping strategies of children and parents who were confronted with childhood cancer.

The foregoing provided the basis for the further development of the intervention. See Figure 3 for an adaptation of the model of Wallander and Varni including outcomes and coping skills that we considered relevant for the *Op Koers* research. Teaching coping skills became the central element of *Op Koers*. Coping skills are taught using cognitive behavioral therapy (CBT) techniques, because CBT was previously found to be effective in reducing psychosocial problems (27). CBT focusses on recognizing and acting upon cognitive distortions and on teaching how to use active coping skills for psychosocial problems. Over the years, elements of acceptance and commitment therapy (ACT) were integrated in the *Op Koers* program. ACT is a part of CBT that strives for acceptance of thoughts or situations to reduce their impact on daily life. ACT is an effective psychological intervention for mental health disorders (28). It can be useful for children with an illness and their family members, to teach them to cope with the situation they are confronted with.

Sharing experiences combined with teaching coping skills became the back bone of The *Op Koers* intervention. The group format, disease-generic use and possibility for online delivery of *Op Koers* have benefits. Sharing experiences with people that are in a comparable situation is found to be helpful in decreasing distress and is effective in teaching coping skills and disease-related problem solving (29, 30). Since psychosocial problems in children with different illnesses usually overlap (Plante 2001), *Op Koers* was developed as a disease-generic intervention. Thus, it provides an intervention also for families of children with rare illnesses. *Op Koers* has an universal approach targeting children with a CI (and their families), regardless whether psychosocial problems are present. Universal interventions like *Op Koers* have a preventative as well as a curative character (29). Besides face-to-face courses, *Op Koers* online was developed to remove known potential practical barriers for participating in face-to-face interventions such as travel distance, time and costs (31).

b) Intervention Generation and Refinement

Each new *Op Koers* module was developed through adaptations of existing modules. For example, the pediatric oncology module originated from the module for patients with a CI, adding cancer-related elements. Adaptations of existing modules relied on research into participant's and health care providers' (HCP) needs (32-34), HCP experiences or participant feedback. For example, experiences with the parent component of *Op Koers* for children with CI uncovered the need to pay attention to the psychosocial needs of parents themselves (35).

The idea for an online intervention arose when adolescents showed to be less likely to participate in a face-to-face intervention than younger children (35), while at that time the first eHealth interventions were emerging. Combining the content of *Op Koers* face-to-face with the technology of an existing Dutch chatroom intervention for participants with depression symptoms (36, 37), the first online module of *Op Koers* was developed for survivors of childhood cancer. After pilot testing this module including surveys and focus groups, we developed online modules for parents, adolescents and siblings of children with a CI, for parents and siblings of children with cancer, and young adult survivors of childhood cancer specifically.

c) Feasibility and pilot testing

The first step in testing an intervention is to study its feasibility in pilot studies. Results of pilot studies were published for four face-to-face modules (7, 38-40) and three online *Op Koers* modules (34, 41, 42). Feasibility of *Op Koers* was evaluated with participants and/or course leaders, and participation rates, e.g. attendance and drop-out, were assessed. Dropout rate was low and satisfaction with the course was high in all studies. Feasibility studies also yielded valuable information for refinement of the content *Op Koers*, as described above (see b).

Besides feasibility, most of these pilot studies also addressed preliminary efficacy in a pre-post-test design, see table 1 (7, 38-40, 42). Coping and emotional functioning were the primary and secondary outcomes, respectively, based on the adapted Model of Wallander and Varni (see Figure 3). Overall, the *Op Koers* modules showed promising results. Coping skills, such as information seeking behavior, improved in children and adolescents with CI and childhood cancer after *Op Koers* face-to-face and online (7, 34, 39, 40, 42), and feelings of helplessness in young adult survivors of childhood cancer decreased after *Op Koers Online* (34). Moreover, at least one aspect of emotional functioning improved in each module. For example, anxiety was lower in siblings after face-to-face Op Koers and young adult survivors of childhood cancer were less distressed after *Op Koers Online* (34, 38).

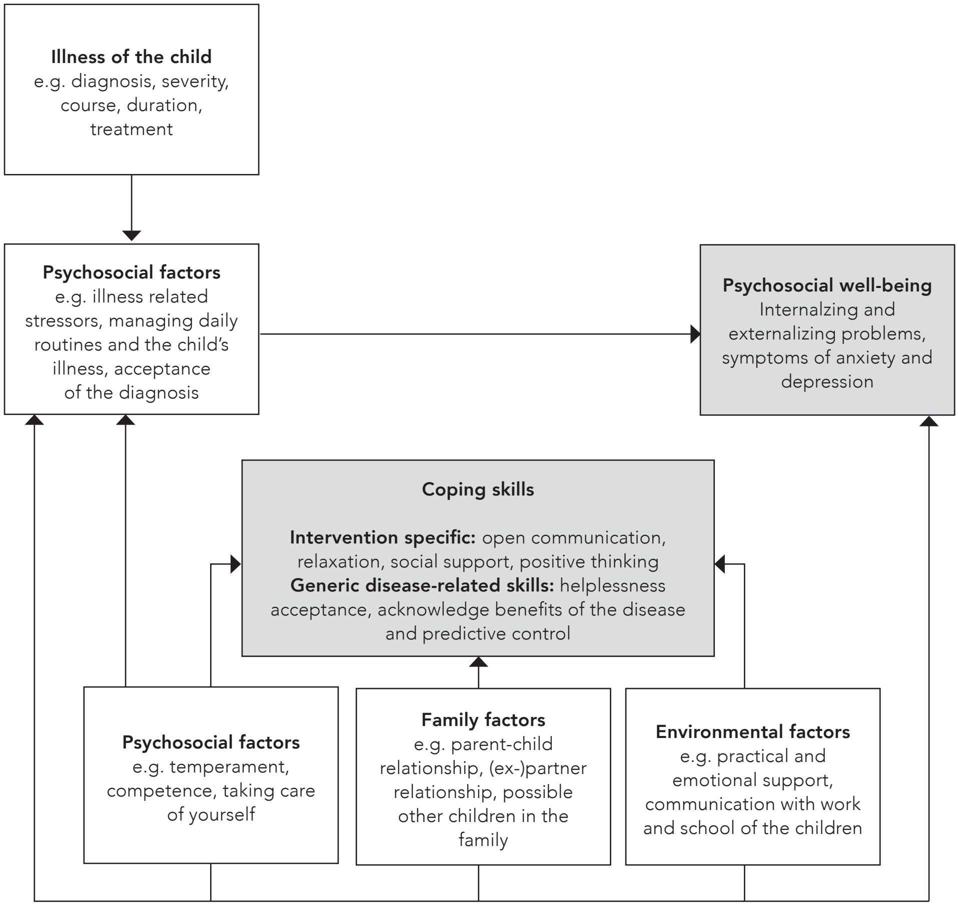

Figure 3 Adapted version of the disability-stress-coping model (24) to explain problems in psychosocial wellbeing in children with an illness and their in family members

d) Efficacy research

To verify if the positive outcomes of the pilot studies could be attributed to *Op Koers*, we conducted efficacy research with the help of large research grants. Four modules of *Op Koers* have been studied in randomized controlled trials (RCT's), see table 1 (35, 43, 44). All modules showed positive effects on both coping skills and emotional functioning, without negative effects on any outcome. Thus, the promising results from the pilot studies were confirmed in the RCT's. We conclude that *Op Koers* is effective for different family members and diagnoses.

Using the data from one RCT (35), the psychosocial characteristics of children and parents were studied as predictors, moderators and mediators of the intervention effect (45, 46).

e) Implementation & Dissemination

After the first face-to-face modules were developed and studied by researchers from the Emma Children's Hospital Amsterdam UMC in the Netherlands, *Op Koers* became part of standard care. Furthermore, it was disseminated throughout the Netherlands and the face-to-face module of *Op* Koers for children with a CI and their parents has been implemented in 30 Dutch hospitals. Thanks to project funding, HCP in those hospitals could be trained and provided with *Op Koers* materials, e.g. instruction manuals, course materials. We do not have information about the uptake of the intervention: how often the hospitals have provided the course and if these courses are still part of the standard of care. We do know that some of the hospitals who used *Op Koers* in the context of the RCT's would like to offer the course as regular care, but are unable due to lack of time and money. In the Princess Máxima Center for Pediatric Oncology, where care and research have been centralized in the Netherlands since 2015, implementation of *Op Koers* was aspired early on and supported by a grant. However, the timing of implementation was inappropriate in the first year of the opening of the hospital because the focus had to be on providing medical care adequately and forming new structures. As a result, implementation activities took longer than anticipated.

We shared our research results in peer-reviewed journals and conferences so that pediatric psychology colleagues can profit from our knowledge and experience. In a commentary on one of these publications, it was suggested that it may be relevant to disseminate *Op Koers* globally (47). In addition, over the years, our research team has been contacted several times by international colleagues who were interested in *Op Koers*. However, international dissemination has been limited so far because translating the intervention and preparing the website for international use is very costly in terms of time and funding. Nevertheless, the manual was translated to Swedish and *Op Koers* is being implemented at Drottning Silvias barnsjukhus, Göteborg.

Table 1. A Summary of all Peer reviewed Research into the *Op Koers Online* Intervention by development phase (b-d)

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
b) Intervention refinement				
Siblings of children with chronic illness (CI), online (32)	To identify siblings' online support needs in order to develop a sibling-specific module	Mixed methods study Questionnaire and semi-structured video interviews on online support needs	Siblings of children with CI (age: 12-18 years) Questionnaire: n=91 Semi-structured video interviews: n=9	Of all participants, 55% would like to initiate or increase contact with other siblings of children with a chronic condition and 46% of those were interested in an online chat course. Important identified themes were: impact on daily life, worrying about brother's/sister's future, handling other people's reactions and how attention is divided within the family.
Parents of children with CI, online (33)	To refine the existing face-to-face module into an online module for parents	Mixed methods study Questionnaire, focus groups and telephone interviews on online support needs	Parents of children with CI (age child: 0-18 years) Questionnaire: n=272 Focus groups: n=15 Telephone interviews: n=7	Important themes for an intervention were identified such as parents' own (emotional) functioning, how to support their child in living with a CI, family functioning, taking care of themselves, relationships with others and practical support. Parents preferred a group with parents of children in the same age category. At first, parents preferred face-to-face contact. After an explanation and demonstration of an online intervention, parents became more positive about online support, mostly because they could participate from home.
Young Adult childhood cancer survivors (YACCS), online (62)	To assess support needs (types and domains) and associated factors in Dutch YACCS	Questionnaire study Questionnaire on support needs	YACCS (age: 18-30 years), n=151	YACCS report the strongest need for support, in particular for concrete information, in the domains lifestyle, fertility, and physical consequences of childhood cancer. Information for YACCS should go beyond the physical consequences of childhood cancer but also focus on emotional and social consequences. Psycho-education should be at the base of survivorship care for YACCS, in order to meet their need for information as well as empower them to take control over their health during the crucial life phase of young adulthood. Survivorship care clinics could specifically consider developing and offering interventions that can be delivered online

Table 1. Continued.

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
c) Feasibility and pilot testing				
Siblings of children with cancer, face-to-face (38)	To study whether the module for siblings reduces anxiety, and to determine predictors of anxiety reduction	Pilot study Longitudinal questionnaire assessment (T0, T1 after intervention) Anxiety (STAI-C)	Siblings of children with cancer (age: 7-18 years), n=24	After group participation, siblings showed less anxiety than before. Age, sex, treatment phase, survival perspective and time since diagnosis were not related to anxiety reduction.
Children with CI) face-to-face (7)	To study the efficacy of the course for children with an illness	Pilot study Longitudinal questionnaire assessment (T0, T1 0-6 weeks after intervention, T2 6-8 months after intervention) Disease-related skills (QOK-c) Cognitive coping (CCSS) Behavioral-emotional problems (CBCL) Anxiety (STAI-C) Self-worth (SPPC) Quality of daily functioning (DUX-25)	Children 8-18 year with CI or childhood cancer (age: 8-18 years), n=109	Improvements in behavioral–emotional outcomes, social competence, information seeking, relaxation and positive thinking were found both in short and medium term.
Adolescents with inflammatory bowel disease (IBD), face-to-face (39)	To Investigate efficacy of psychoeducational group intervention for adolescents with IBD	Controlled pilot study Longitudinal questionnaire assessment (T0, T1 6-8 months after intervention) Cognitive coping (CCSS) Self-worth (SPPA) Anxiety (STAI-C) Quality of daily functioning (DUX-25)	Adolescents with IBD (age: 12-18 years), n=40 Intervention group, n=22 Control group, n=18	Positive effect on: coping (predictive control), feelings of competence (global self-worth and physical appearance), and HRQoL

Table 1. Continued.

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
Children after treatment for childhood cancer, face-to-face (40)	Pilot study into the appropriateness and efficacy of the module for children after treatment for childhood cancer	Mixed methods pilot study Longitudinal questionnaire assessment (T0, T1 0-4 weeks after intervention) and focus group Disease-related skills (QOK-c)	Children after completion of successful childhood cancer treatment (age: 8-12 years), n=11, and their parents	Positive outcomes on most items concerning disease-related skills. Remarkable improvements with respect to the intervention goals 'social competence' and 'positive thinking'. Parents reported sharing emotions and experiences of their child with other patients/survivors as an important element of the intervention.
Adolescents after treatment for childhood cancer, online (41)	Pilot study into the satisfaction with and feasibility of the online module for adolescents after treatment for childhood cancer	Mixed methods pilot study Questionnaire (adolescents) and interview (course leaders) after intervention Questionnaire on satisfaction with and feasibility of intervention	Adolescents after completion of successful childhood cancer treatment (age: 11-17 years), n=11 and course leaders (n=6)	Dropout was very low and high levels of satisfaction were reported by CCS and course leaders. The positive evaluations indicate that OK Onco Online is a promising innovative group intervention.
Adolescents with CI, online (42)	Pilot study into feasibility and efficacy of the online module for adolescents with CI	Pilot study Longitudinal questionnaire assessment (T0, T1 0-2 weeks after intervention) Disease-related skills (QOK-c) Emotional and behavioral problems (YSR) Health-related quality of life (PedsQL)	Adolescents with CI (age: 12-18 years), n=29	High overall satisfaction. Increased use of total coping skills, information seeking and giving, and social competence strategies; significantly fewer withdrawn/depressed behavior and significantly better scores on emotional and psychosocial HRQoL

Table 1. Continued.

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
Young Adult survivors of childhood cancer (YACCS), online (34)	Pilot study into feasibility and efficacy of the online module for YACCS	Pilot study Longitudinal questionnaire assessment (T0, T1 1-4 weeks after intervention) Evaluation questionnaire Distress (DT) Sense of control over changes in life (MS) Illness cognitions (ICQ) Impact of cancer (IOC-CS) Health-related quality of life (PedsQL)	YACCS (age: 18-30 years), n=10	This first, small pilot study showed short term decrease in distress and feelings of helplessness and, improvement of self-efficacy. The pilot also indicated that *Op Koers* Online is a feasible intervention, filling a gap in psychosocial services for YACCS
d) Efficacy research				
Children with CI, face-to-face (35, 63)	To investigate the efficacy of the module for children with chronic illnesses and to test the effect of an added parent component.	RCT Longitudinal questionnaire assessment (T0, T1 6 months after intervention, T2 12 months after intervention) Behavioral-emotional problems (CBCL) Disease-related skills (QOK-c, QOK-p)	Children with CI (age: 8-18 years), n=194 and their parents, n=49 Child intervention, n=71 Parent-child intervention, n=49 Wait-list control, n=74	Results show a positive effect of the intervention on parent-reported internalizing problems, child-reported externalizing problems, information seeking, social competence, and positive thinking. The additional effect of parental involvement was observed on parent-reported internalizing problems, child-reported externalizing problems, information seeking, and social competence.
Children with CI, face-to-face (45, 63)	To investigate characteristics of children and parents as predictors and moderators of the effect of the module for children with chronic illness	RCT Longitudinal questionnaire assessment (T0, T1 6 months after intervention, T2 12 months after intervention) Child disengaged coping (CSI) Self-worth (SPPC, SPPA) Emotional security of the parent-child relationship (CSS) Parenting stress (PSI)	Children with CI (age: 8-18 years), n=194 Child intervention, n=71 Parent-child intervention, n=49 Waitlist control, n=74	Children with a more disengaged coping style or lower self-worth benefited more from *Op Koers*, especially as their parents were involved as well. The benefit of parental involvement was dependent on the quality of the parent-child relationship.

Table 1. Continued.

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
Children with CI, face-to-face (46, 63)	To test tested child coping and parenting stress as underlying mechanisms of the effect of an intervention for children and an additional group intervention for parents.	RCT Longitudinal questionnaire assessment (T0, T1 6 months after intervention, T2 12 months after intervention) Behavioral-emotional problems (CBCL) Disease-related skills (QOK-c) Parenting stress (PSI)	Children with CI (age: 8-18 years), n=120 Child intervention, n=71 Parent-child intervention, n=49	Active coping skills and parenting stress were mediators of the effect of the added parent component on internalizing behavior.
Adolescents with CI, online (43, 64)	To evaluate the efficacy of the module for adolescents with a chronic illness.	RCT Longitudinal questionnaire assessment (T0, T1 6 months after intervention, T2 12 months after intervention) Behavioral-emotional problems (CBCL) Disease-related skills (QOK-c) Health-related quality of life (PedsQL)	Adolescents with CI (age: 12-18 years), n=59 Intervention, n=35 Waitlist control, n=24	The intervention had a significant positive effect on disease related coping skills T1 vs T0 (use of relaxation and social competence) and T2 vs T0 (information seeking), and on HRQoL (social-, school-, psychosocial functioning and total HRQoL) T1 vs T0.
Parents of children with CI, online (44, 64)	To evaluate the efficacy of the online module for parents of children with a chronic illness	RCT Longitudinal questionnaire assessment (T0, T1 6 months after intervention, T2 12 months after intervention) Anxiety and depression (HADS) Disease-related skills (QOK-p) Illness cognitions (ICQ) Cognitive coping (CCSS)	Parents of children with CI (child's age: 0-18 years), n=77 Intervention, n=34 Waitlist control, n=33	The intervention had a significant positive effect on changes in anxiety, depression, and total score T1 versus T0 and T2 versus T0, the coping skills open communication, relaxation, social support, acceptance, predictive control and helplessness T1 versus T0 and relaxation and positive thinking T2 versus T0.

Table 1. Continued.

Target group and reference	Focus	Methods Outcome (measures)	Sample	Main Results
Parents of children with cancer, online *Publication in preparation*	To evaluate the efficacy of the online module for parents of children with cancer	RCT Longitudinal questionnaire assessment (T0, T1 5 months after intervention, T2 12 months after intervention) Anxiety and depression (PROMIS anxiety and depression and HADS) Disease-related skills (QOK-p) Illness cognitions (ICQ) Cognitive coping (CCSS)	Parents of children with cancer, expected n=98 Intervention, expected n≈49 Waitlist control, expected n≈40	*In preparation*

Abbreviations:
CI chronic illness; YACCS young adult survivors of childhood cancer; IBD inflammatory bowel disease; CCS childhood cancer survivors; HRQoL health-related quality of life, QOK-c Questionnaire *Op Koers* for children; QOK-p Questionnaire *Op Koers* for parents; CCSS cognitive control strategies scale; STAI-C State Trait Anxiety Inventory for Children; CBCL Child behavior check list; SPPC Self-perception profile for children; SPPA Self-perception profile for adolescents; YSR Youth Self-Report; PedsQL Pediatric Quality of Life Inventory; DT Distress Thermometer; MS Mastery scale; ICQ illness cognitions questionnaire; IOC-CS Impact of cancer – childhood cancer survivors; CSI coping strategies inventory; CSS: child security scale; PSI Parenting Stress Index; HADS Hospital anxiety and depression scale; PROMIS patient reported outcome measurement information system

LESSONS LEARNED

In this section we describe the lessons we learned in the development, evaluation and implementation of the *Op Koers* program.

Health care provider engagement

Development and evaluation of interventions are often led by researchers. A strength and facilitator of the development of *Op Koers* was the collaboration from the start between pediatric psychologists and the research team, promoted by the integration of research and care in the involved university medical care centers. Notably, the researchers were positioned in the clinical department, rather than universities outside the pediatric clinic. HCP should be involved as early as possible to prevent a research-to-practice gap. Doing so results in more commitment, input and effort from clinicians and it leads to a higher chance of successful implementation of an intervention (8).

Engagement of HCP in the dissemination and implementation of *Op Koers* was a challenge. Firstly, though the multicenter approach in most RCT's was favorable for the inclusion rate and was a first step towards nationwide implementation, it was logistically complicated to prepare the pediatric psychologists for the intervention. It was helpful that we used a very detailed protocol and trained course leaders personally before they started *Op Koers*. Secondly, as a result of the high work pressure that many psychologists experienced, it was sometimes challenging to introduce this new intervention. It helped to have an 'ambassador' or 'champion' in the clinical department (48). Ideally, the ambassador has experience with the program and has gained trust from the intended course leaders, so that they will be more inclined to adopt a positive attitude towards the new intervention. The ambassador could also motivate other HCP such as pediatricians, to see the added value of the intervention, and motivate them to invest in recruitment.

Participant engagement

Patient and parent participation at an early stage is important in the development of an intervention, as well as for future participation in studies (49). We encountered that involving patients and parents in the development of *Op Koers* was difficult, and it was challenging to ensure representativeness. We asked patients and parents either about their support needs before (re)designing the intervention, or about their opinion after participating in the intervention. This often lead to valuable insights, for example that explaining the online course should include visual support (33). It also provided challenges when opinions, literature or clinical experience contradicted each other. For example, when we asked siblings for their ideas about an intervention, they reported to prefer a course group with siblings of children with a similar CI (32), whereas the literature showed that the psychosocial challenges of living with a CI are similar regardless of the diagnosis (30).

Recruitment

In research settings, problems in recruitment lead to problems such as study delay or smaller sample sizes and power. Lack of power makes it difficult to prove effectiveness. This is a typical barrier for psychosocial interventions (50-52) and complicates implementation. For example, in the RCT on *Op Koers* for parents of children with a CI, we did not reach the planned sample size, even after lengthening the inclusion period. Nevertheless, fortunately, power turned out to be sufficient to prove intervention effects (44). Besides proving effectiveness, it would be interesting to investigate who profits most from the intervention, to make sure the people who benefit get access to it (9). Unfortunately, the sample sizes of most our RCT's were not sufficient to conduct subgroup analyses, or to examine the working mechanism of *Op Koers*.

We often used an open recruitment strategy to increase the number of participants. We promoted Op Koers through HCP and social media, in the hospital newsletters or via patient organizations. The downside of this open recruitment strategy is that there is no information available about response rates, nor about the characteristics of non-participants. This hampers drawing conclusions about generalizability of the results. Nonetheless, we consider an open recruitment strategy the best option, using multiple recruitment methods; through social media, leaflets in the outpatient clinics, and to have the psychosocial department in the clinics refer families to *Op Koers*. To promote *Op Koers*, we also developed two videos with information for children and adolescents about the *Op Koers* program (see www.opkoersonline.nl).

In clinical settings, recruitment and scheduling of participants for *Op Koers* was challenging. *Op Koers* courses occasionally had to be cancelled due to a lack of participants. We found an effective solution in including the courses in the annual planning of psychosocial care and appointing a coordinator to manage the schedule and the waitlist of participants. When courses are planned regularly, clinicians can discuss participation with families more easily and patient organizations can add to recruitment by pointing out the course to their members.

Study design

We have conducted studies with different designs, depending on research aims and opportunities. We started with pilot studies using a pre-post design without a control condition. In addition, multiple RCT's have been conducted over the years, because an RCT is considered to be the gold standard in intervention efficacy research. A recent meta-analysis into group interventions for parents of children with cancer encourages further RCT's (53). However, attention for the downsides of RCT's is growing (54, 55), which are in line with our own experiences: since an RCT needs a control group, more participants are needed, which makes RCT's very time- and money consuming. Also, such robust but laborious research design makes it difficult to evaluate continuous improvements and adaptations to the intervention, which impacts the validity of research results. This makes

RCT's less suitable for intervention development. While some researchers believe RCT's are the only design allowing for conclusions on causality (56), others argue that valid causal inferences can also be drawn from single-case designs because these are also controlled experiments (57). To date, single-case designs are generally considered to be less valuable and therefore seemingly harder to publish. Thus, we still felt the need to use the RCT design for our efficacy studies.

In our efficacy research, we deliberately opted for a waitlist-control design instead of a control condition consisting of peer support without CBT and ACT elements because peer support is a key element of the *Op Koers* intervention. Nevertheless, it would be interesting to study the additional effect of CBT and ACT elements on peer support only, but this would require more participants. Regarding our choice to use a waitlist-control group with 'care as usual', it is important to realize that the added value of the intervention may be smaller than compared to a control condition with no care at all. On the positive side, studying *Op Koers* alongside regular care is actually more informative as this matches with the real-world situation and how it would be implemented. Also, for ethical reasons, the control condition in psychosocial research mostly receives 'care as usual'.

Outcome measures

The use of appropriate outcome measures is essential to capture intervention effects (58). Operationalization of intervention effects into outcomes and thereafter, finding sound measures (questionnaires), is crucial but often problematic. If outcome measures are generic, it can be hard to identify intervention-specific outcomes such as coping skills. If outcome measures are more specific, it can be hard to compare the results to other research projects or populations. Therefore, in our research into *Op Koers*, we used generic as well as specific outcome measures. Regarding generic outcome measures, the Patient-Reported Outcomes Measurement Information System (PROMIS) is recommended. They overcome the lack of comparability between different measurements (59) and are more reliable across a range of functioning than other generic outcomes because they are developed using item response theory (60). In the RCT with *Op Koers* for parents online in pediatric oncology that is currently being carried out, we use computer adaptive testing of the PROMIS anxiety and depression item banks. To assess *Op Koers* specific outcomes, we developed a questionnaire that covers the disease-related coping skills taught through *Op Koers*. This measure, with variants for children (Questionnaire *Op Koers* for children (QOK-c)) and parents (QOK-p) has been used in most of our studies (Table 1).

When choosing an instrument, it is important to make sure that the measures are sensitive to change. It sometimes occurs that participants say they gained a lot from the intervention while showing no improvement on the outcome measures. Another factor that complicates demonstrating effects could be the preventative character of *Op Koers*, which allowed participants to join without reporting significant clinical problems. If the participants' coping skills were sufficient and participants were not experiencing high levels of psychosocial problems from the start of an intervention, there is little room for improvement and ceiling effects on measures could occur. This is a common problem among studies on preventative interventions (61). Nevertheless, on average, we found improvements on several outcomes in all evaluation studies of *Op Koers*. In conclusion, it is necessary to make a well-considered choice of outcome measures and questionnaires for every new research project.

Technological development

When developing e-health applications, cooperation with web designers/administrators and application builders is required. Communication about what you want and what is technically possible comes with challenges that require professionals.

In an evolving field like e-health and e-intervention development, the material or research results may become somewhat outdated quickly. For example, *Op Koers Online* was designed as a chatroom intervention. At the time of development, video calls were not very common in the Netherlands. Over the past years, especially during the COVID-19 pandemic, people became more accustomed to online activities, and *Op Koers* participants seemed to be increasingly interested in an online course that is provided through video calls instead of only a chatroom. Even though we think there are advantages to typing instead of speaking (e.g. having time for reflection and anonymity), we also deem it important to consider new options and are currently experimenting with a video call format with siblings of children with cancer. Using videoconferencing could make it easier to further implement and disseminate the course internationally, since the chat platform would not have to be translated.

Funding

Over the entire process from starting with *Op Koers* 25 years ago, and still refining and studying it today, a lot of funds have been necessary. Costs are related to (research) personnel, materials and technical support. An additional challenge in developing a new intervention are the efforts in time and money that need to be made outside our field of expertise, e.g. general data protection regulation laws, designing the course materials' layout and keeping the website up to date. Over the years, *Op Koers* research was funded by multiple grants, with an overall value of 2.1 million euros. Besides providing knowledge and experience with the *Op Koers* intervention, this funding contributed to the PhD training of several young researchers.

Applying for grants is time consuming and several grant applications were rejected. A barrier for funding was the generic approach of *Op Koers*. Not focusing on a specific illness makes *Op Koers* unsuitable for many funding agencies targeting only one diagnostic group. Even though costs of research are high, it is important to provide evidence-based care. Also, we believe that preventative group interventions such as *Op Koers* may in the end be cost-effective in the real world setting, since psychologists can treat multiple participants at the same time and preventing problems may save needed treatment, and thus resources in the longer term (29).

Preferably, implementation should be evaluated structurally and guided by an implementation plan, which is grounded on theory and based on a conceptual framework (8). This requires time and effort (i.e. financial investment), which is a barrier for using such an approach. Nevertheless, the *Op Koers* team aims to keep working on refining the intervention where needed and continues to make efforts for the implementation.

Making up the balance

After carrying out so many research projects on *Op Koers*, pilot studies as well as RCT's, we ask ourselves what the endpoint should be. In all projects so far, the intervention had a positive effect on coping skills and emotional functioning of participants. Still, we keep refining and optimizing the intervention either for existing modules or new ones. Our aspirations include for example new modules for family members of a child with acquired brain injury. Should we invest our time in efficacy research for adapted or new modules or focus on wider lasting implementation of *Op Koers*? On the one hand, one could argue that efficacy studies are no longer needed, because it is justifiable to trust on the results of our previous research when a module is adapted for a new target population. On the other hand, it is only possible to make statements about the effect of an intervention when it is studied within the appropriate target population. From a researcher's point of view, we would like to continue studying the intervention, for example regarding the working mechanism of *Op Koers*, or providing the course using videoconferencing. From a more practical point of view, logistical barriers such as time and money sometimes keep us from doing further research. In combination with the often problematic recruitment, this results in having to compromise: only study a new module or refinement of Op Koers when there is a promising grant opportunity and if the number of available participants is expected to be sufficient.

CONCLUSION

In the 25 years of developing the *Op Koers* intervention, many intervention development and evaluation activities were conducted, partly described in 15 publications in international peer-reviewed journals. The efforts resulted in the availability of 15 separate *Op Koers* modules that are part of standard care in several Dutch hospitals. In the process, multiple barriers in psychosocial intervention development and research were encountered, regarding participants, HCP, funding and methodological considerations. To some of those we have found solutions, and some remain continuous challenges that we will keep trying to cope with in the coming years. *Op Koers* is one of few psychosocial interventions in pediatric psychology that has been both extensively studied and implemented. The main factor for this achievement is the close collaboration between, and the perseverance of the clinical care and research departments.

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8

General discussion

The research for this thesis was conducted to extend understanding on how to prevent and/or reduce emotional and behavioral problems in adolescents and parents who are confronted with a Chronic Illness (CI), by investigating two separate online group interventions *Op Koers Online*.

Op Koers Online is a cognitive-behavioral based program which aims to teach adolescents and parents to use adaptive coping skills that help them cope with the consequences of the CI. Peer support is also an important aspect of the intervention. The intervention is designed for adolescents and parents of children with different diagnosis, offering patients and parents of patients with rare illnesses the possibility to participate. The courses consist of eight (adolescents) and six (parents) weekly online sessions, led by two course leaders (psychologists). The *Op Koers Online* program relies on an adapted version of the disability-stress-coping model of Wallander and Varni (1998) (Figure 1) (1). The model implies that the stressors faced by children and parents dealing with pediatric CI are multifaceted and that the associations between illness related stressors and adjustment are moderated by appraisals and coping strategies on which several personal and family risk- and protective factors are of influence.

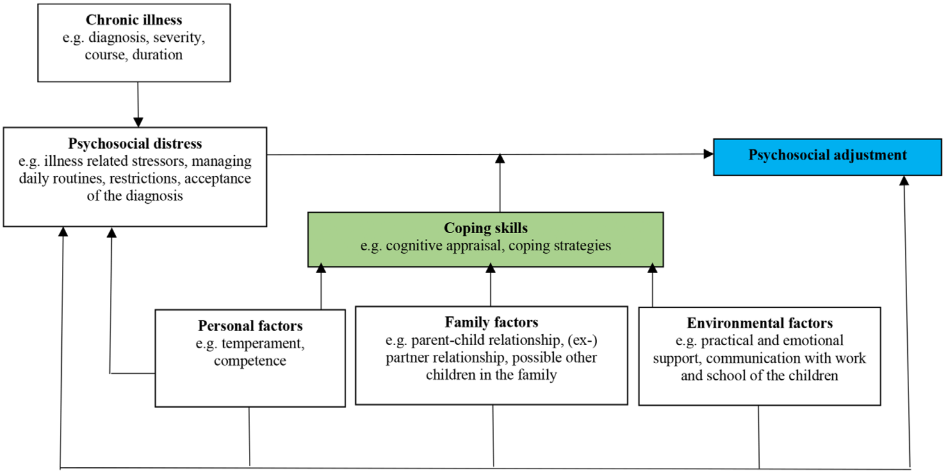

Figure 1 Adapted version of the disability-stress-coping model of Wallander and Varni (1998) to explain psychosocial adjustment in adolescents with a CI and parents of a child with a CI.

This thesis aims to answer different research questions focusing on *Op Koers Online* for adolescents and parents separately. The first research question was: **is *Op Koers Online* for adolescents effective in enhancing engaged coping and in preventing and/or reducing emotional- and behavioral problems?** This was tested in a pilot study (Chapter 2) and a Randomized Controlled Trial (RCT) to assess the efficacy of *Op Koers Online* for adolescents (Chapter 5). The second research question focused on the development of the intervention for parents: **what are parental support needs?** An explorative support needs study concerning this question is presented in Chapter 3. The third research question was: **is *Op Koers Online* for parents effective in enhancing engaged coping skills and in preventing and/or reducing emotional problems?** The RCT to assess the efficacy of *Op Koers Online* for parents is presented in Chapter 6. The fourth and last research question was; **what are the lessons learned after 25 years of intervention development?** An overview of the *Op Koers* program throughout the years is presented in Chapter 7. Objectives, characteristics and main findings off all chapters are presented in Table 1.

REFLECTION ON MAIN FINDINGS

Development of Op Koers Online

The *Op Koers Online* program runs for approximately ten years now. Experience with intervention development, effectivity research and implementation was gained over the years. *Op Koers Online for adolescents* finds its origin in the *Op Koers Online face-to-face* modules, which was proven effective on psychosocial functioning and use of adaptive coping skills (2). In the face-to-face intervention, it was difficult to include adolescents. To increase accessibility, *Op Koers Online for adolescents* was developed and rated feasible in pediatric oncology (3). Next step was the pilot test of *Op Koers Online for adolescents* with a CI (Chapter 2, this thesis), using a pre-post-test design, with promising results. To test efficacy using a control group, an RCT was conducted (Chapter 5, this thesis). Even though recruiting adolescents was still difficult, the online accessible character seemed to be appealing for the participating adolescents, which met our expectations but was not questioned beforehand. This finding confirmed what was already upcoming in literature: patient participation in order to fit interventions to patient's needs is important.

The *Op Koers face-to-face* module for parents runs parallel to the child intervention and focusses primarily on the child (called *Together Op Koers, "Samen Op Koers"* in Dutch). It became clear parents were interested in sharing their own feelings for which an intervention was lacking. To our knowledge, an online illness-generic group intervention for parents themselves was not available until now. To explore parental needs for an intervention, first a support needs study was conducted (Chapter 3, this thesis). Parents filled out a questionnaire concerning support needs and focus groups were held. Subsequently, according to the outcomes of the support needs study, *Op Koers Online for parents* was developed. The intervention focusses on specific themes, established by the parents in the study: 1) the CI of the child/the hospital, 2) relationships within the family, 3) taking care of yourself besides caregiving tasks and 4) relationships with others and practical support. Parents' practical preferences (e.g. concerning format, day and time of the intervention, grouping) for an intervention were also assessed and, where possible, met while designing the intervention. Not all parent's preferences could be granted. For example, some parents wished to have one face-to-face meeting before the start of the online intervention. To ensure anonymity and to eliminate practical and logistical barriers, this was not added to the intervention format. Instead, the course leaders speak to all participants separately on the phone to introduce themselves, explain the intervention format and answer possible questions. Finally, there will still be a group of parents for whom the intervention does not fit. For those parents, the option for face-to-face care in the hospital is always available.

Efficacy

Op Koers Online for adolescents

The pilot study with a pre-post design (see Table 1) indicated that *Op Koers Online* for adolescents was feasible and potentially effective (Chapter 2). The RCT confirmed that the intervention had beneficial effects of medium size on the use of disease-related coping skills (6-months follow up: relaxation and social competence; and 12-months follow up: information seeking) and on HRQoL (6-months follow-up) (Chapter 5). Although the coping skill positive thinking is central in the intervention and was statistically significant in the study of Scholten et al (2013) on *Op Koers face-to-face*, it was not statistically significant in the current RCT. Given the small- to medium effect sizes of the coping skill, it is possible that the stronger power in the study of Scholten et al (2013) explains the discrepancy between the two studies. However, it could also be due to the intervention format: it could be more difficult to reach participants via chatting. An online format could lead to difficulties in explaining the intervention content and checking whether or not explanation is understood with the absence of nonverbal contact.

Even though several studies present comparable effects of face-to-face versus online therapy modules (4-6), there is also evidence for less compliance and less therapist-reported treatment understanding in internet-based interventions compared to a similar face-to-face interventions (7). The *Op Koers Online* intervention contains weekly homework assignments, to which participants in the current RCT were mostly compliant to, with encouragement of course leaders. However, when discussing the homework assignments, course leaders were sometimes in doubt about the investment of participants to the assignments. It seemed that some participants made the assignments in a rush which can nullify the understanding and treatment effect. For example, to learn and integrate positive thinking, one of the key elements of *Op Koers Online*, exercise is needed. The fact that positive thinking did not improve significantly in the RCT for adolescents could be due to lack of homework attention and/or understanding. In the future, attention should be paid to the discussion of the completed homework assignments during the online sessions. This could motivate participants to accurately complete the assignments. The face-to-face format might include homework discussion more naturally. In addition, participants could feel more obligated to complete homework assignments when physically seeing course leaders and group members in the face-to-face format. In addition, to help support and alert adolescents for the exercises in the face-to-face format, a reminder by text message could be useful.

The waitlist control group received 'care as usual', in the current RCT meaning mostly no psychosocial care or treatment in primary psychosocial care. Of the adolescents in the waitlist control group, 14% of adolescents received psychosocial care during the study, compared to 74% of the children and adolescents in the control group in the study on *Op Koers* face-to-face (2). The clinical scores at baseline were lower in the *Op Koers Online* sample compared to the face-to-face sample, explaining the difference in seeking psychosocial care during the waitlist period. Also, the face-to-face sample includes children and adolescents, and the online sample includes only adolescents who are less likely to seek psychosocial care in general (8). Possibly, the motivation to participate in the online intervention differs from participant's motivation for the face-to-face intervention. Online participation can be done from home, anonymous if desired. The participants in the current RCT had relatively low scores of emotional and behavioral problems, but still wanted to participate in an online intervention, possibly to meet others in a similar situation. Consequently, finding intervention effects could be more difficult. For the face-to-face intervention, participants have to visit the hospital multiple times, in addition to their regular hospital visits. Feeling the need for a psychosocial intervention for emotional and behavioral problems seems to overcome the barrier of additional hospital visits. In addition, since adolescents participate in *Op Koers Online* without their parents (in contrary to the face-to-face intervention where children and parents participate simultaneously), it could be more difficult for parents to motivate their child to sign up for the intervention.

No statistically significant intervention effect was found on parent- and self-reported internalizing and externalizing problems in the pilot study nor in the RCT for adolescents. Possibly, this was due to the fact that *Op Koers Online* is a preventative as well as a curative intervention so that *Op Koers Online* is not explicitly designed to decrease psychopathology symptoms. Having clinical levels of behavioral problems was not an inclusion criterion. Since the vast majority of participants in the current sample had no clinical levels, there was a limited room for improvement by the intervention. The absence of intervention effects on internalizing and externalizing problems in the present studies is not in line with the results of the RCT on *Op Koers* face-to-face (2). The discrepancy in statistically significant effects of *Op Koers Online* and *Op Koers* face-to-face could be explained by the fact that more participants in the face-to-face sample had clinical levels of behavioral problems at baseline, enlarging the room for improvement by the intervention.

Table 1. Summary of Main Findings

Chapter	Title	Objectives	Participants/procedures	Design and measures	Main findings
2	Online cognitive-behavioral group Intervention for adolescents with chronic illness: A pilot study)	To assess feasibility and explore preliminary effectiveness of an online cognitive-behavioral group intervention (*Op Koers Online*) to prevent and/or reduce psychosocial problems by teaching use of active coping skills to adolescents (ages 12 to 18) with chronic illness.	N = 29 adolescents (ages 12 to 18) with various chronic illnesses Mean age 15.1 years (SD = 1.85)	• Pre-post questionnaire design • *Op Koers* Questionnaire (OKQ; disease-related coping skills) • Youth Self Report (YSR; internalizing/ externalizing problems) • Pediatric Quality of Life Inventory-Self Report (PedsQL; Health-Related Quality of Life)	• *Op Koers Online* for adolescents is feasible and potentially effective • Participants improved significantly in the use of several coping skills • Participants' overall satisfaction was high
3	Matching psychosocial support needs of parents of a child with a chronic illness to a feasible intervention)	To refine an existing face-to-face intervention into an online psychosocial group intervention for parents by 1) exploring which themes are important, 2) determine what type of intervention parents would like and 3) assess parents' practical preferences.	N = 272 parents of a child with a chronic illness (ages of the child between 0-18) (questionnaire) N = 15 parents of a child with a chronic illness (ages of the child between 0-18) (focus groups) and N = 7 (telephone interviews)	• Mixed method approach: quantitative (self-developed questionnaire) and qualitative (structured focus groups/ interviews according to COREQ criteria and the SROBE-checklist for cross-sectional research)	• Parents have a need for support focusing on different themes concerning themselves: the CI of the child, the family, taking care of yourself, extended family and friends and practical support • Parents preferred a group with parents of children in the same age category • Professionals should explain and demonstrate an online intervention to parents
4	Online cognitive-behavioral based group interventions for adolescents with chronic illness and parents: Study protocol of two multicenter randomized controlled trials	To present the study protocols of the RCT's	N.A.	N.A.	N.A.

Table 1. Continued.

Chapter	Title	Objectives	Participants/procedures	Design and measures	Main findings
5	Online psychosocial group intervention for adolescents with a chronic illness: A randomized controlled trial	To evaluate the efficacy of *Op Koers Online* for adolescents, a protocolled online psychosocial group intervention for adolescents with a chronic illness	N = 59 adolescents (ages 12 to 18) N = 35 intervention group; N = 24 waitlist control group Mean age = 15.1 years (SD = 1.7)	• Assessments (online questionnaires) at baseline, post-intervention, 6- and 12-months follow-up • Mixed model analyses • Youth Self Report (YSR) and Child Behavior Checklist (CBCL; parent report; internalizing/ externalizing problems) • *Op Koers* Questionnaire (OKQ; disease-related coping skills) • Pediatric Quality of Life Inventory (PedsQL; Health-Related Quality of Life) • Socio-demographic and illness characteristics, self-designed questionnaire	• The results of the RCT indicate a positive effect of *Op Koers Online* for adolescents • The intervention had a significant positive effect on several disease-related coping skills • The intervention had a significant positive effect on HRQoL • No intervention effects on internalizing and externalizing behavioral problems were found.
6	Online psychosocial group intervention for parents: Positive effects on anxiety and depression	The evaluate the efficacy of an online psychosocial group intervention *Op Koers Online* for parents of children with a chronic illness, in terms of anxiety and depression, and disease-related coping skills	N = 73 parents of children (ages 0 to 18) with a chronic illness N = 34 intervention group; N = 33 waitlist control group	• Assessments (online questionnaires) at baseline, post-intervention, 6- and 12-months follow-up • Mixed model analyses were performed • Hospital Anxiety and Depression Scale (HADS) • Op Koers Questionnaire for Parents (OKQ-P; disease-related coping skills), Illness Cognition Questionnaire (ICQ-P), Cognitive Coping Strategies Scale Parent Report-form (CCSS-PR)	• The RCT indicates evidence for the efficacy of *Op Koers Online* for parents • Parental anxiety and depression decreased, and use of adaptive coping skills improved after the intervention • The online character, the focus on parents themselves instead of on their child and the possibility for parents of children with rare illnesses to participate, are innovative and unique aspects of Op Koers Online for parents.

Table 1. Continued.

Chapter	Title	Objectives	Participants/procedures	Design and measures	Main findings
	Development, research and implementation of the psychosocial group intervention *Op Koers*: Lessons Learned	To share the lessons that we have learned in the process of 25 years of development, research and implementation of a psychosocial group intervention for children with illness and their family members (*Op Koers*)	N.A.	• A critical appraisal of our activities in the different stages using the National Institutes of Health Stage Model for Behavioral Intervention Development	• Important lessons were learned in the domains of participants and health care providers (engagement, recruitment), research methodology (study design, outcome measures) and environmental conditions (technological advances, funding) • *Op Koers* is one of the few psychosocial interventions that has been both extensively studied and implemented. The main factor for this achievement is the close collaboration between, and the perseverance of the clinical care and research departments

In the current RCT, the intervention effect on coping skills and HRQoL diminished at one-year follow-up with exception of information seeking and giving about the disease. In the study of Scholten et al (2013) regarding *Op Koers face-to-face,* adding a parental component to the intervention contributes to the persistence of the effects. In the face-to-face intervention, parents and children/adolescents participated in separate, parallel groups and parents learned how to support their child in daily life. *Op Koers Online* for adolescents was intentionally designed without involving parents, with the argument that it keeps the threshold for participation low and gives adolescents the opportunity to participate independently from their parents. The *Op Koers Online* module for parents (studied in Chapter 5) is stand alone and does not focus on teaching the parents to support coping skill in their children. Possibly this explains the diminishing of the effects on coping skills on the long term for adolescent in the online module. In addition, the intervention uses cognitive behavioral therapy (CBT) techniques but is not a CBT by itself. Some parts of the intervention such as cognitive restructuring is introduced and addressed shortly. This is not comparable with an extensive CBT in which there is a lot of practice in order to integrate the learned skills in daily practice. Also, the effects of the intervention could diminish when contact with and support from course leaders and group members disappears after the intervention.

Op Koers Online for parents

The RCT on *Op Koers Online* for parents indicated evidence for efficacy: the intervention had a positive effect of medium size on symptoms of parental anxiety and depression (6- and 12-months follow-up) and on disease-related coping skills (6-months follow-up: open communication, relaxation, social support, positive thinking, helplessness, acceptance and predictive control; 12-months follow-up: relaxation and positive thinking; Chapter 6). Parents with better psychosocial functioning are more able to support their child with a CI adequately (9-11), which will likely be related to better psychosocial and illness-related outcomes for the children (12-16). According to a recent systematic review, interventions that involve a significant peer support component are associated with decreases in for example parental distress, underscoring the importance of the group element of *Op Koers Online* for parents (17).

Regarding disease-related coping skills, some of the significant intervention effects of medium size at 6-months diminished at 12-months. However, at 12-months follow-up, effects on the coping skills relaxation and positive thinking were established. According to the literature, coping skills could be expected to mediate the effect of the intervention on parental anxiety and depression outcomes (18, 19). In the current study, continued reliance on relaxation and positive thinking may explain the long-term significant outcomes on anxiety and depression. Little is known about how to maintain long-term effects of online CBT group interventions. A study evaluating an online group intervention for parents of childhood cancer survivors (Cascade) showed that parents of the intervention group feel confident about the use of CBT skills after following the intervention, however this did not appear to translate in actual use of the CBT skills (20). This could also be applicable for the long-term use of some of the coping skills after following *Op Koers Online* for parents. Future efforts should focus on exploring whether for example an extra booster session at one-year follow-up could maintain the effect on the other coping skills at long-term.

Of the parents in the waitlist control group, 15% received psychosocial care during the study. This is conform our expectations, given the fact that parents indicate practical barriers for seeking and receiving psychosocial support and the wish for an online format to overcome those barriers. It is likely that parents who assign for an online intervention with a low threshold, do not seek for other (face-to-face) psychosocial support during a waitlist period.

In conclusion, overall findings from this thesis indicate that teaching adaptive coping skills promotes adjustment in adolescents with a CI and in parents. Furthermore, teaching these coping skills via an online format appears to be as feasible and effective.

Recruitment and study design

In both RCT's, recruitment was difficult. We hoped the online format as well as the multicenter approach would make participation much easier for families. Nevertheless, recruiting adolescents and parents was difficult, which could possibly be due to the fact that participants had to fill out questionnaires at multiple time-points. The response rate on information letters(3%) was lower than expected based on former research on *Op Koers face-to-face* (19%) (2). Even though we experienced that face-to-face recruitment works best, healthcare professionals had trouble motivating adolescents to participate in the study. Inclusion problems in RCT's are common (21, 22). Especially adolescents are a hard group to motivate given their age, puberty and possibly the psychosocial character of the intervention. Adolescents do not want to be 'different' and generally do not like to be confronted with their illness (23-26). Adolescents tend to avoid thinking and talking about their illness and therefore, do not sign up for an intervention to talk about living with a CI such as *Op Koers Online*. As an avoidant coping strategy is a risk factor for the development of social and emotional problems and poor compliance (27-29), an intervention that is focused on adjusting avoidant coping strategies into a more adaptive coping style, is very relevant for those with an avoidant coping style. Moreover, the study of Scholten et al (2013) showed that participants with an avoidant coping style gain more from the intervention than participants with another predominant coping style. The challenge for the *Op Koers* team and health-care providers is to include patients with an avoidant coping strategy.

To motivate adolescents to join the intervention, it could be helpful when healthcare providers pay more attention to explain what the intervention entails and why participation would be helpful for them. Healthcare providers should be aware of a patient's avoidant coping style, to emphasize the importance of participation. In addition, communicating experiences from former participants could be useful for recruitment. One could also think of a web-based explanation of the intervention with illustrations and text or a short video, which potential participants can view by themselves on their smartphone or tablet.

Recruiting parents was less difficult than recruiting adolescents, however, expected targets were not reached despite extended recruitment periods. Problems with including parents were also visible in an effectivity study of a comparable group intervention for parents of childhood cancer survivors (20). Several studies highlight that parents face many challenges from the time their child gets the CI diagnosis (30). Parents are confronted with many extra caregiving tasks and are busy managing daily routines, putting time for themselves under pressure. Parents tend to prioritize the care of their child first (30). It is imaginable that these parents are not available to participate in a weekly group intervention. Furthermore, we found out that parents need some extra explanation about the online character of the intervention. When implementing the intervention in clinical practice, it is important to keep in mind that professionals should invest time and effort in recruitment and guiding parents through the online environment of the intervention.

In the RCT for parents, almost all participants were mothers. We experienced that fathers are hard to reach due to several reasons, mostly lack of time and indicating no need for support, although it is known that fathers as primary caregiver reported elevated levels of depression (31). Furthermore, we know from the literature that for example fathers of children with Down Syndrome do express a need for psychosocial support (32). Father's need for psychosocial support seems to be dependent on their involvement in the treatment of the CI. Research shows that depressive symptoms of fathers do not mediate intervention outcomes, suggesting that fathers may have a different role from the mothers and are for example more focused on for example ensuring family income (16). Some fathers were interested in *Op Koers Online*, however, some of them indicated that they expected mostly mothers to participate and were hesitant of participating in a group with only female participants. Hopefully, in the changing society where caregiving tasks become more equally divided between mothers and fathers, more fathers will be more likely to participate in interventions like *Op Koers Online.*

STRENGTHS AND LIMITATIONS OF THE STUDIES

A strength of the current thesis is patient engagement. Patient participation at an early stage is important for the development of an intervention, as well as for future participation in studies (33). For the development of *Op Koers Online* for parents, parents were involved in the support needs study. Furthermore, in addition to participation in the current study, participating adolescents were involved in a patient participation program initiated by Fonds NutsOhra (FNO; sponsor of the studies), focused on how psychosocial care for adolescents with a CI should be organized.

Another strength is that the thesis included an intervention for adolescents as well as an intervention directed at the wellbeing of parents, and thereby contributes to the family-focused care in families who deal with a (chronic) illness. A intervention directed at parental needs is innovative because parents are often uninvolved in interventions, indirectly involved or only for a short period (34). Psychosocial care becomes more family-focused, since there is better understanding of the influence of a CI on the whole family. Also, the intervention is illness-generic, which makes it possible for patients with rare illnesses and their parents to participate.

It is a strength that a pilot study on *Op Koers Online* for adolescents preceded the RCT, which gave directions for the RCT. Strengths of the RCT's in particular included recruitment of participants across the whole country from nine centers and via patient associations and using a standardized intervention protocol which is essential for conducting intervention research and for implementation in clinical practice.

There were also some limitations, among others, the unknown recruitment rate and the lack of information about non-respondents, because of partially online open recruitment. Furthermore, some outcome measures had moderate internal consistency. On the one hand, the use of scales with moderate internal consistency is acceptable for group comparisons because the internal consistency is an indication of random error and has nothing to do with systematic error (bias). On the other hand, Cronbach's alphas should preferably be 0.7 or higher because the lower the internal consistency, the larger the random measurement error, and so, the more difficult to detect differences between groups. It is possible that due to this, some intervention effects were not proven in the current studies. Concerning the RCT for parents, outcomes were based on mothers, which limits the representativeness for fathers.

Regarding the choice to use a waitlist-control group with 'care as usual' in the RCT's in the current thesis, it is important to realize that the added value of the intervention may be smaller than compared to a control condition with no care at all. On the positive side, studying *Op Koers Online* alongside regular care is actually more informative as it matches with the real-world situation and how it would be implemented. Also, for ethical reasons, the control condition in psychosocial research mostly receives 'care as usual'. In the current sample, care as usual entailed mostly no treatment and treatment in primary psychosocial care.

DIRECTIONS FOR FUTURE RESEARCH

Research focusing on working mechanisms of interventions is important but rare. However, the study of Scholten et al (2013) (2) on *Op Koers* face-to-face focused on moderating and mediating effects and showed that children who were more 'at risk' for developing psychosocial problems (children with a more disengaged coping style and lower self-worth) appeared to gain more form participating in the intervention, especially when parents were involved as well. The study of Willemen et al (2022) (18) showed that the working mechanisms of *Op Koers* are mediated by the disease-related coping skills taught during the intervention and parenting stress. For future research, it would be interesting to investigate whether the same moderating and mediating factors can be found for the new module *Op Koers Online* for parents.

RCT's are considered to be the gold standard in intervention efficacy research (35). Some researchers state RCT's are the only design allowing for conclusions on causality (36). A recent meta-analysis into group interventions for parents of children with cancer encourages further RCT's (37). However, attention for the downsides of RCT's is growing (38-40), which is in line with the experiences from the current thesis: since an RCT needs a control group, more participants are needed, which makes RCT's time- and money consuming. Also, such robust but laborious research design makes it difficult to evaluate continuous improvement and adaptions to the intervention, which impacts the validity of research results. Furthermore, according to Kwakkenbos et al (2018) (39), follow-up can be labor intensive and many RCT's have limited real-world generalizability. Research shows that that valid casual inferences can be drawn from single-case designs because these are also controlled experiments (41). To date, single-case designs are generally considered to be less valuable and therefore seemingly harder to publish, which is likely one of the reasons for many researchers to conduct an RCT. However, the options for single-case designs or other types of intervention evaluation and implementation research should be considered.

Concerning study designs, the current RCT studies used a waitlist-control design. Since peer support is a key element of *Op Koers Online*, it would be interesting to study the additional effect of CBT and ACT elements on peer support only by adding a control condition consisting of peer support without CBT and ACT elements. However, this would require more participants.

The use of appropriate outcome measures is essential to capture intervention effects (42, 43). In this thesis, no changes were found in the primary outcomes for adolescents (internalizing and externalizing problems). This is a common problem among studies on interventions with a preventative as well as curative character (20) such as *Op Koers Online*. Clinical levels of internalizing/externalizing problems were not an inclusion criterion. If the participants' coping skills were sufficient and participants were not experiencing high levels of psychosocial problems from the start of an intervention, there is little room for improvement and ceiling effects on measures could occur. In the future, primary outcomes should be reconsidered, in particular those for the adolescent intervention.

CLINICAL IMPLICATIONS AND FUTURE PLANS

With the results of the RCT's, *Op Koers Online* for adolescents with a CI and for parents of children with a CI can be implemented in clinical practice. There are some recommendations for clinical practice.

A point of attention is recruitment and inclusion. Including enough patients is important to be able to run the intervention. As stated before, it is helpful to recruit via health-care professionals. A personal approximation with extra explanation and/or demonstration of the intervention could be useful for some patients, parents in particular. Furthermore, it would be helpful when hospitals have an *Op Koers Online* 'ambassador' in the clinical department (44), who is responsible for carrying out the intervention program and can keep track of the planning of the courses, invitations of the patients, recruitment and inclusion.

Concerning the intervention format: the intervention was intentionally set up as a chat-only, without a webcam to ensure anonymity and keep the threshold for participation low. However, in an evolving field like e-health and e-intervention development, the material may became somewhat outdated quickly. At the time of developing *Op Koers Online*, video calls were not standard practice in the Netherlands. Over the past years, especially during the COVID-19 pandemic, people became more accustomed to online activities, and *Op Koers Online* participants seemed to be increasingly interested in an online course that is provided through video calls instead of only a chatroom. Even though there are advantages to typing instead of speaking (e.g. having time for reflection and ensure anonymity), it is important to consider new up-to-date options. Using videoconferencing could make it easier to further implement and disseminate the intervention internationally, since the chat platform would not have to be translated.

Future efforts should also focus on maintaining the effects on coping skills at one-year follow-up for both adolescents and parents. For adolescents, parental encouragement on use of coping skills should be evaluated and explored. Possibly, a telephonic appointment with parents to explain the intervention and help them support their child could be helpful. However, this should be handled with care since the anonymity and the possibility to participate separately from their parents is an advantage for some adolescents. For both groups, the option of an extra booster session at one-year follow-up in order to maintain long-term effects should be explored. Moreover, an online e-health self-study module with a summary of the coping skills taught during the intervention could be helpful to refresh participant's minds, for example once a year.

Op Koers Online for adolescents and parents is ready for implementation. Implementation is an extensive process and should be done thoroughly. Preferably, implementation should be evaluated structurally and guided by an implementation plan, which is grounded on theory and based on a conceptual framework (45). This requires time and effort (i.e. financial investment), which is a barrier for using such an approach.

Nevertheless, the *Op Koers* team aims to keep working on refining the intervention where needed and continues to make efforts for implementation of the interventions. An ambassador, as mentioned above, can promote the implementation process.

CONCLUSION

With this thesis, we contribute to the field of pediatric psychology with a protocol- and evidence based group intervention for adolescents and parents.

Concluding key messages

- *Op Koers Online* for adolescents and parents are innovative interventions given the online and illness-generic character and the focus on adolescents/parents themselves. The interventions can now be implemented in clinical practice.

Adolescents

- Participation in *Op Koers Online* for adolescents has a positive effect on the use of adaptive coping skills and health-related quality of life;
- Adolescents who participated are satisfied about the intervention content and format.

Parents

- Parents of children with a chronic illness have a need for psychosocial support focusing on themselves focusing on different themes: 1) the illness of the child, 2) relationships within the family, 3) taking care of yourself besides caregiving tasks, 4) relationships with others and 5) practical support. According to parental support needs, *Op Koers Online* for parents was developed;
- Participation in *Op Koers Online* for parents had a positive effect on decreasing feelings of anxiety and depression and the use of adaptive coping skills.

The Op Koers program

- Within the *Op Koers* program, multiple interventions were developed, evaluated and implemented over the past 25 years;

Implementation and future directions

- Implementation of *Op Koers Online* should be evaluated structurally, preferably guided by an extensive implementation plan and an ambassador to guide implementation.
- Future efforts should focus on maintaining the long-term effects of *Op Koers Online*;
- Future researchers are encouraged to carefully evaluate research designs and outcome measures.

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Addendum

SUMMARY

An increasing number of children (age 0-18 years) in the Netherlands is growing up with a chronic illness, such as asthma, diabetes, rheumatoid arthritis, Crohn's disease and more rare diseases such as cystic fibrosis and diverse metabolic diseases. Due to increased medical knowledge and improved treatments, more of these children grow up into adulthood. An important consequence of this positive development is that the psychosocial consequences of pediatric chronic illness need attention. Children and adolescents with a chronic illness have to face challenges such as insecurity about the future, frequent hospital visits, medical treatments, feeling tired, limits in social and/or sportive activities and school absenteeism. These factors are of influence on the daily functioning of children and adolescents. Interventions that help support families in coping with psychosocial aspects of a pediatric chronic illness are essential to avoid negative consequences as social problems or the development of psychopathology.

In the current thesis, two separate online psychosocial group interventions *Op Koers Online* for adolescents (12 to 18 years) with a chronic illness and parents of a child (0 to 18 years) with a chronic illness are described. *Op Koers Online* focusses on teaching how to use adaptive coping skills. Coping is known as the way someone reacts in stressful situations. The intervention consists of eight (adolescents) or six (parents) weekly online sessions in a chatroom. Participants learn how to use adaptive coping skills which help them to think positive, relax during medical treatments and stressful situations and how to communicate about their illness/the illness of their child. besides, the intervention focusses on improving quality of life and decreasing emotional- and behavioral problems. Support from others in a similar situation is an important part of *Op Koers Online*.

This thesis describes the development and efficacy of *Op Koers Online* for adolescents and parents and the development, evaluation and implementation of the *Op Koers* program over the past 25 years. Specific research questions were: 1) is *Op Koers Online* for adolescents effective in enhancing engaged coping and in preventing/reducing emotional- and behavioral problems?, 2) what are parental support needs?, 3) is *Op Koers Online* for parents effective in enhancing engaged coping skills and in preventing and/or reducing emotional problems?, and 4) what are the lessons learned after 25 years of intervention development?

1. Is *Op Koers Online* for adolescents effective in enhancing engaged coping and in preventing/reducing emotional- and behavioral problems?

Adolescents growing up with a chronic illness have to face several challenges concerning their illness, and are therefore at risk for developing psychosocial problems such as feeling down or isolating oneself (1, 2). Especially during puberty, with the formation of identity, self-image and self-esteem, a chronic illness constitutes a major challenge (3-5). Interventions that help adolescents to support how to cope with their illness are essential. Interventions are available, however, these interventions are mostly focused on a specific illness (5, 6). Research shows that psychosocial consequences are comparable across illnesses (7). An illness-generic intervention has the benefit of offering adolescents with rare diseases the possibility to participate.

After proven positive effects of *Op Koers* face-to-face for children and adolescents with chronic illness (8), the wish to lower the threshold to participate in the intervention for adolescents arose. *Op Koers Online* for adolescents was developed, at first for adolescent survivors of childhood cancer (9) and hereafter for adolescents with chronic illness.

The first research question is answered with two studies, first of which the pilot study (**Chapter 2**). The pilot study was conducted in order to assess the feasibility and effectiveness of *Op Koers Online* for adolescents, before a larger effect study was carried out. In total, 33 adolescents participated in the pilot study. Eventually, 23 adolescents filled out the pre- and post measurement questionnaire. Participants were satisfied about the intervention; the online format appeared to be feasible. After following the intervention, use of coping skills (information seeking and social competence) of participants improved, withdrawn/depressed behavior decreased and quality of life (emotional and psychosocial) improved.

After the promising results of the pilot study, a randomized controlled trial (RCT) was conducted (**Chapter 5**) in order to assess the efficacy of *Op Koers Online* for adolescents. The design of the RCT was described in a research protocol developed according to the CONSORT Statement (10, 11) (**Chapter 4**).

Nine hospitals across the Netherlands participated in the RCT, of which 22 trained psychologists carried out the intervention. In total, 59 adolescents participated in the RCT (intervention group N = 35, control group N = 24). Adolescents filled out questionnaires at four time-points, before the intervention (baseline), after following the intervention/after 8 weeks, six and twelve months after baseline. Parents were asked to fill out one questionnaire concerning emotional functioning of their child at each time-point.

The intervention had a positive effect on adaptive coping skills (relaxation, social competence and information seeking) and on quality of life. No intervention effects were found for internalizing and externalizing problems. After one year, intervention effects diminished. Future efforts should focus on maintaining the effects in the long term.

2. What are parental support needs?

In order to study parental support needs, a support needs study for parents of children with a chronic illness was conducted (**Chapter 3**). Previous studies show that parents of children with a chronic illness have to deal with additional (caregiving) tasks and stressors, and therefore are at risk for psychosocial problems (12-15). Offering support to those parents is important, since parents who experience less stress are more able to take care of their chronically ill child (16-18). Most existing interventions focus on the child, parents are not or limited involved, mostly focused on how to support their child (8, 19). The current support needs study was designed to assess parental support needs focusing on themselves.

The support needs study was both quantitative (questionnaire) as qualitative (focus groups and telephonic interviews). Recruitment was done via patient associations and social media, using a link to an online questionnaire. The questionnaire included questions about themes which could be important to address in an intervention, which type of psychosocial intervention parents would like and practical preferences for an online group intervention. At the end of the questionnaire, parents could give up the wish to participate in a focus group. focus groups were held to acquire more in-depth information, in addition to the questionnaire.

In total, 272 parents filled out the questionnaire. Three focus groups were held with 15 parents, telephonic interviews were held with seven parents. According to the support needs study, parents appeared to have a need for an intervention focusing on themselves. An illness-generic design was appealing to parents, since it would offer parents of children with rare illnesses the possibility to participate. Parents indicated five important themes to address in an intervention: 1) the illness of the child, 2) the family, 3) taking care of yourself, 4) relationships with others and 5) practical support. Concerning practical preferences, parents preferred groups composed based on the ages of the child. At first, parents were somewhat hesitant about an online format and had a preference for face-to-face. However, after explanation and demonstration of what an online format would look like, the preference changed. An online format was appealing for parents, given the possibility to participate from home and the option for anonymity.

According to parental support needs, *Op Koers Online* for parents for children with a chronic illness was developed. Parental wishes were integrated in the intervention where possible.

3. Is *Op Koers Online* for parents effective in enhancing engaged coping skills and in preventing and/or reducing emotional problems?

In order to assess the efficacy of *Op Koers Online* for parents, an RCT was conducted (**Chapter 6**). The study protocol is presented in **Chapter 4** and is designed in accordance with the CONSORT Statement (10, 11). Nine hospitals across the Netherlands participated, of which 22 trained psychologists carried out the intervention.

In total, 73 parents participated in the study (intervention group N = 34, control group N = 33). Parents filled out questionnaires before the intervention (baseline), after following the intervention/after six weeks, six- and twelve months after baseline.

The intervention had a positive effect on symptoms of anxiety and depression and on use of adaptive coping skills (open communication, relaxation, social support, acceptance of the disease, predictive control, helplessness and positive thinking). Parents were satisfied about the intervention, especially because the intervention is focused on themselves and that participation could be done from home, which offered practical preferences and lowered the threshold for participation. Effects on anxiety and depression maintained at one-year follow-up, effects on some of the coping skills diminished.

In conclusion, *Op Koers Online* for parents is an innovative and unique intervention, given the illness-generic character (and therefore the possibility for parents of children with rare illnesses to participate), the focus on parents themselves instead of their child and the possibility to participate from home. The intervention was proven to be effective in decreasing symptoms of anxiety and depression and the use of adaptive coping skills. Next step is to implement *Op Koers Online* for parents in clinical practice.

4. What are the lessons learned after 25 years of intervention development?

In the past 25 years, several psychosocial group interventions for children with an illness and their parents within the *Op Koers* program were developed, evaluated and implemented in clinical practice. **Chapter 7** of the current thesis presents the lessons learned over the past 25 years. Using the National Institutes of Health Stage Model for Behavioral Intervention Development, the activities in the different stages were critically appraised.

Within the *Op Koers* program, intervention modules for different populations are available: children, adolescents, siblings and parents who have to deal with chronic illness or cancer. Several pilot studies and larger effect studies (RCT's), in which *Op Koers* appeared to be effective in teaching adaptive coping skills and improving emotional functioning.

The lessons learned can be categorized in three domains. First, **participants and health-care providers**: health-care providers are important for including participants for *Op Koers*. Especially when the intervention is carried out in the context of an RCT study, recruitment was difficult. It is important that health-care providers motivate patients for participation in the study and intervention. Having an *Op Koers* ambassador in the hospital who is responsible for education and motivation of health-care providers and planning and organization of the intervention is recommended. The ambassador should monitor and adjust the planning and organization where needed. Besides, patient participation is important, to adjust the intervention content according to participant's preferences.

Second, the **research methodology**. To assess the efficacy of *Op Koers*, multiple RCT's were performed. RCT's are considered to be the gold standard in intervention efficacy research (20). However, attention for the downsides of RCT's is growing: since an RCT needs a control group, more participants are needed, which makes RCT's time- and money consuming. This is in line with the experiences in the current thesis, the recruitment and inclusion was extended multiple times. Some researchers advocate for single-case designs, which should be more feasible and generalizable to real-world situations (21). For future studies, it is important to carefully select a research design. Besides, in order to be able to find intervention effects, it is important to critically evaluate outcome measures.

Finally, concerning **technology**, the world of e-health is developing fast. *Op Koers Online* was designed a few years ago. Over the years and especially during the COVID-19 pandemic, people became more accustomed to online activities and for example video calls. The chatroom format of *Op Koers Online* became somewhat outdated. Even though the use of chatroom with only chatting (typing) was deliberately chosen to ensure anonymity and keep the threshold for participation as low as possible, it is worth the effort to experiment with adding video call to the intervention format. Depending on the experiences, the intervention format can be adjusted eventually.

In conclusion, *Op Koers* is an unique and widely developed intervention program for children and adolescents with a chronic illness or (previous) cancer diagnosis, their siblings and parents. The face-to-face modules are part of standard care in different hospitals in the Netherlands and one hospital in Sweden. In total, fifteen articles are published in peer-reviewed journals. *Op Koers* is one of the few psychosocial interventions in pediatric psychology that has been studied and implemented this intensively. The main factor for this achievement is the close collaboration between, and the perseverance of the clinical care and research departments.

Main findings

- Participation in *Op Koers Online* for adolescents had a positive effect on use of adaptive coping skills and health-related quality of life;
- Adolescents who participated are satisfied about the intervention content and format;
- Parents of a child with a chronic illness indicate a need for psychosocial support focusing on themselves, focused on different themes: 1) the illness of the child, 2) the family, 3) taking care of yourself, 4) relationships with others and 5) practical support. According to parental support needs, *Op Koers Online* for parents was developed;
- Participation in *Op Koers Online* for parents had a positive effect on decreasing feelings of anxiety and depression and the use of adaptive coping skills;
- Some effects diminished at long-term. Future efforts should focus on how to maintain long-term effects of the interventions;
- Within the *Op Koers* program, multiple interventions were developed, evaluated and implemented over the past 25 years;
- *Op Koers Online* for adolescents and parents can now be implemented in clinical practice. It is importance that the process of implementation is done structured and thoroughly.

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NEDERLANDSE SAMENVATTING / SUMMARY IN DUTCH

In Nederland groeien steeds meer kinderen op met een chronische ziekte. Voorbeelden van chronische ziektes zijn astma, diabetes, reuma, de ziekte van Crohn, maar ook meer zeldzame aandoeningen zoals cystic fibrosis en verschillende metabole ziekten. Door de vooruitgang in de medische behandelmethoden kunnen steeds meer van deze kinderen opgroeien tot volwassenen. Een belangrijk gevolg van deze positieve ontwikkeling is dat de consequenties van ziekte op de kinderleeftijd voor de psychosociale ontwikkeling van kinderen, jongeren en hun families aandacht behoeven. Kinderen en jongeren met een chronische ziekte krijgen veelal te maken met onzekerheid over de toekomst, frequent ziekenhuisbezoek, medicamenteuze behandeling, vermoeidheid, beperkingen in het meedoen aan sociale en/of sportieve activiteiten en schoolverzuim. Deze aspecten hebben een grote invloed op het functioneren van kinderen en jongeren in het dagelijks leven. Om gezinnen te ondersteunen in het omgaan met de psychologische en sociale aspecten van een chronische ziekte kan hulpverlening gewenst zijn. Interventies gericht op vaardigheden die kunnen helpen bij het omgaan met een chronische ziekte zijn van cruciaal belang om de veerkracht van kinderen, jongeren en ouders te vergroten en mogelijke negatieve gevolgen, zoals sociale problemen, of de ontwikkeling van psychopathologie te voorkomen.

In dit proefschrift worden twee aparte online psychosociale groepsinterventies *Op Koers Online* voor jongeren (12 tot 18 jaar) met een chronische ziekte en ouders van een kind (0 tot 18 jaar) met een chronische ziekte beschreven. *Op Koers Online* richt zich op het aanleren van adaptieve coping vaardigheden. Onder coping wordt de manier waarop je omgaat met stressvolle omstandigheden en tegenslagen verstaan. De interventie bestaat uit acht (jongeren) of zes (ouders) wekelijkse online sessies in een chatbox. Deelnemers leren adaptieve coping vaardigheden die hen helpen positief te denken, te ontspannen bij medische ingrepen en stressvolle situaties en open te communiceren over hun ziekte/ de ziekte van hun kind. Daarnaast is de interventie gericht op het verbeteren van kwaliteit van leven en het verminderen van emotionele- en gedragsproblemen. Lotgenotencontact is tevens een belangrijk onderdeel van *Op Koers Online*.

Dit proefschrift beschrijft de ontwikkeling en effectiviteit van *Op Koers Online* voor jongeren en ouders en de ontwikkeling, toetsing en implementatie van het hele *Op Koers* programma over de afgelopen 25 jaar. Specifieke onderzoeksvragen waren: 1) is *Op Koers Online* voor jongeren effectief in het aanleren van adaptieve coping vaardigheden en in het voorkomen en/of verminderen van emotionele- en gedragsproblemen?, 2) wat zijn de behoeftes van ouders aan psychosociale steun?, 3) is Op Koers Online voor ouders effectief in het aanleren van adaptieve coping vaardigheden en in het voorkomen en/ of verminderen van emotionele problemen? En 4) Wat hebben wij geleerd van 25 jaar interventie ontwikkeling, onderzoek en implementatie?

1. Is Op Koers Online voor jongeren effectief in het aanleren van adaptieve coping vaardigheden en in het voorkomen en/of verminderen van emotionele- en gedragsproblemen?

Jongeren die opgroeien met een chronische aandoening krijgen te maken met verschillende stressoren rondom hun ziekte en lopen daarmee risico op psychosociale problemen zoals somberheid en zichzelf afzonderen (1, 2). Zeker tijdens de puberteit, een belangrijke periode voor het ontwikkelen van een eigen identiteit, vormt een chronische aandoening een uitdaging (3-5). Interventies om jongeren te ondersteunen bij het omgaan met hun aandoening zijn essentieel. Er bestaan verschillende interventies voor deze doelgroep, echter zijn deze interventies meestal gericht op een specifieke aandoening (5, 6). Uit onderzoek is gebleken dat de psychosociale uitdagingen die horen bij het hebben van een chronische aandoening vergelijkbaar zijn voor verschillende aandoeningen (7). Een ziekte-generieke interventie geeft jongeren met een zeldzame aandoening de mogelijkheid om ook deel te nemen.

Na de positieve resultaten van het onderzoek naar de *Op Koers* face-to-face interventie voor kinderen en jongeren met een chronische aandoening (8), ontstond de wens om de drempel voor deelname aan de interventie voor adolescenten te verlagen. *Op Koers Online* werd ontwikkeld, in eerste instantie voor jongeren die behandeld zijn voor kanker (9), en vervolgens voor jongeren met een chronische aandoening.

De eerste onderzoeksvraag wordt beantwoord met twee studies, te beginnen met een pilotstudie (**Hoofdstuk 2**). De pilotstudie werd uitgevoerd om een indicatie te krijgen van de haalbaarheid en effectiviteit van *Op Koers Online* voor jongeren, voordat een grote effectstudie werd ondernomen. Er deden 33 jongeren mee aan het pilot onderzoek, uiteindelijk vulden 23 jongeren de vragenlijsten van de voor- en nameting in. Deelnemers waren tevreden over de cursus; het online format bleek haalbaar. Na de interventie waren de coping vaardigheden (informatie geven en zoeken en sociale competenties) van de deelnemers verbeterd, teruggetrokken/somber gedrag verminderd en kwaliteit van leven (emotioneel en psychosociaal) verbeterd.

Voortbordurend op de pilotstudie is er gerandomiseerd onderzoek (Randomized Controlled Trial, RCT) uitgevoerd (**Hoofdstuk 5)** om de effectiviteit van *Op Koers Online* voor jongeren verder te onderzoeken en beter te kunnen onderbouwen. De opzet wordt beschreven in een onderzoeksprotocol ontwikkeld conform het CONSORT Statement (10, 11) (**Hoofdstuk 4**).

Aan het onderzoek deden in totaal negen ziekenhuis verspreid over Nederland mee, waarvan in totaal 22 getrainde psychologen de cursus gaven. Er deden 59 jongeren mee aan de RCT (interventiegroep N = 35, controlegroep N = 24). Jongeren vulden vragenlijsten in voor de start van het onderzoek, na het volgen van de interventie/na acht weken, en een half jaar en een jaar na de start. Ouders vulden per meetmoment ook één vragenlijst in over het emotioneel functioneren van hun kind.

De interventie had een positief effect op ziekte gerelateerde coping vaardigheden (ontspanning, sociale competentie en informatie zoeken) en op kwaliteit van leven. Er werden geen effecten van de interventie gevonden op internaliserende en externaliserende problemen. Een jaar na de interventie waren de effecten afgenomen. Een belangrijke vervolgstap is nagaan hoe de effecten op lange termijn behouden kunnen blijven.

Concluderend kan gesteld worden dat *Op Koers Online* voor jongeren van meerwaarde is voor het gebruik van adaptieve coping vaardigheden en het verbeteren van kwaliteit van leven. De vervolgstap is het implementeren van *Op Koers Online* voor jongeren in de klinische praktijk.

2. Welke behoeftes aan psychosociale steun hebben ouders van kinderen met een chronische ziekte?

Om deze vraag te kunnen beantwoorden is een behoeftepeiling gedaan onder ouders van een kind met een chronische aandoening (**Hoofdstuk 3**). Uit eerder onderzoek is gebleken dat ouders van kinderen met een chronische aandoening te maken krijgen met extra (zorg-) taken en stress, en daarmee risico lopen op het ontwikkelen van psychosociale problemen (12-15). Ondersteuning hierbij is belangrijk, omdat ouders met minder stress beter in staat zijn om voor hun chronisch zieke kind te zorgen (16-18). De meeste bestaande interventies zijn vooral gericht op het kind, waarbij ouders niet of zijdelings worden betrokken ter ondersteuning van het kind (8, 19). De behoeftepeiling was gericht op het onderzoeken of ouders behoefte hebben een hulp voor zichzelf en welke voorkeuren ze hierbij hadden.

In de behoeftepeiling werd gebruik gemaakt van kwantitatief (vragenlijst) en kwalitatief (focus groepen en telefonische interviews) onderzoek. De werving verliep via patiëntenverenigingen en sociale media, waarop een link naar de vragenlijst gepresenteerd werd. De vragenlijst bevatte vragen over de thema's die belangrijk zijn in een interventie, welk type psychosociale interventie ouders zouden willen en er werd gevraagd naar praktische voorkeuren voor een online groepsinterventie. Aan het eind van de vragenlijst konden ouders aangeven of ze mee wilden werken aan een focusgroep of telefonisch interview. Het doel van de focusgroepen en interviews was het verkrijgen van meer diepgaande informatie in aanvulling op de vragenlijst.

In totaal vulden 272 ouders de vragenlijst in. Er werden drie focusgroepen gehouden met in totaal 15 ouders, en met zeven ouders werd een telefonisch interview gehouden. Uit de behoeftepeiling bleek dat ouders inderdaad behoefte hebben aan een interventie gericht op zichzelf. Voor ouders was het prettig dat er een ziekte-generiek hulpaanbod zou komen, om ook ouders van kinderen met een zeldzame aandoening de kans te geven om mee te doen aan een groepscursus. Hulp zou volgens ouders moeten gaan over verschillende thema's: 1) de chronische aandoening van het kind, 2) het gezin, 3) zorgen voor jezelf, 4) relaties met anderen en 5) praktische steun. Uit de inventarisatie van praktische voorkeuren bleek onder andere dat ouders voorkeur hadden voor groepen samengesteld op basis van de leeftijden van de kinderen. Ouders waren aanvankelijk wat terughoudend over een online format, en gaven voorkeur aan voor face-to-face. Echter, na uitleg en demonstratie van een online interventie veranderde de voorkeur. Een online interventie bleek aantrekkelijk, met als belangrijkste redenen de mogelijk om vanuit huis deel te nemen en de optie om anoniem te blijven.

Op basis van de resultaten van de behoeftepeiling werd *Op Koers Online* voor ouders van een kind met een chronische aandoening ontwikkeld. De wensen van ouders werden waar mogelijk verwerkt in de interventie.

3. Is Op Koers Online voor ouders effectief in het aanleren van adaptieve coping vaardigheden en in het voorkomen en/of verminderen van emotionele problemen?

Om de effectiviteit van *Op Koers Online* voor ouders te onderzoeken is een RCT uitgevoerd (**Hoofdstuk 6**). Het onderzoeksprotocol wordt gepresenteerd in **Hoofdstuk 4** en is ontwikkeld conform het CONSORT Statement (10, 11). Er deden negen ziekenhuizen verspreid over Nederland mee aan het onderzoek, waarvan in totaal 22 getrainde psychologen de cursus gaven.

Aan het onderzoek deden 73 ouders mee (interventiegroep N = 34, controlegroep N = 33). Ouders vulden vragenlijsten in voor de start van het onderzoek, na het volgen van de interventie/na zes weken, een half jaar en een jaar na de start.

De interventie had een positief effect op symptomen van angst en depressie, en op het gebruik van coping vaardigheden (open communicatie, ontspanning, sociale steun, acceptatie van de ziekte, predictieve controle, hulpeloosheid en positief denken). Ouders waren tevreden over de interventie, met name over het feit dat de interventie op henzelf gericht is en dat deelname vanuit huis mogelijk was wat voor ouders de drempel om mee te doen verlaagde. De effecten op angst en depressie bleven bestaan een jaar na de interventie, de effecten op een aantal coping vaardigheden waren een jaar na de interventie afgenomen.

Samengevat kan gesteld worden dat *Op Koers Online* voor ouders een innovatie en unieke interventie is, gezien het ziekte-generieke aspect (en daarmee de mogelijkheid voor ouders van kinderen met zeldzame ziekten om deel te nemen), de focus op ouders zelf in plaats van hun kind en de mogelijkheid om vanuit huis deel te nemen. De interventie is effectief gebleken in het verminderen van symptomen van angst en depressie en het gebruik van adaptieve coping vaardigheden. De volgende stap is het implementeren van *Op Koers Online* voor ouders in de klinische praktijk.

4. Wat hebben wij geleerd van 25 jaar interventie ontwikkeling?

In de afgelopen 25 jaar zijn er binnen het *Op Koers* programma verschillende psychosociale groepsinterventies voor zieke kinderen en hun familieleden ontwikkeld, geëvalueerd en geïmplementeerd in de klinische praktijk. In dit proefschrift worden in **Hoofdstuk 7** de geleerde lessen uit de afgelopen 25 jaar gepresenteerd aan de hand van het National Institutes of Health Stage Model for Behavioral Intervention Development, waarbij de gedane activiteiten kritisch beoordeeld worden aan de hand van een aantal stappen.

Binnen het *Op Koers* programma bestaan interventiemodules voor verschillende doelgroepen: kinderen, jongeren, broers en zussen en ouders die te maken hebben met een chronische aandoening of kanker. Er zijn verschillende pilotstudies en grotere effectstudies (RCT's) gedaan waarbij *Op Koers* effectief bleek in het aanleren van adaptieve coping vaardigheden en het verbeteren van emotioneel functioneren.

De geleerde lessen zijn te categoriseren in drie domeinen. Ten eerste: **deelnemers en zorgverleners**: zorgverleners spelen een belangrijke rol bij het includeren van deelnemers voor *Op Koers*. Zeker wanneer de cursus in het kader van een onderzoek gegeven wordt, is de werving lastig gebleken. Het is belangrijk dat zorgverleners patiënten zorgvuldig informeren en enthousiasmeren voor onderzoek. Wat helpt is om een *Op Koers* ambassadeur op de afdeling te hebben, die verantwoordelijk is voor de planning en organisatie rondom *Op Koers* en dit ook monitort en bijstuurt. Een ambassadeur is ook verantwoordelijk voor het voorlichten en motiveren van zorgverleners, die vervolgens hun patiënten motiveren. Daarnaast is gebleken dat patiëntparticipatie bij *Op Koers* van grote meerwaarde was, om het groepsaanbod af te stemmen op de behoeftes van deelnemers.

Ten tweede het domein **onderzoeksmethodologie**. Er zijn meerdere effectstudies (RCT's) gedaan om de effectiviteit van *Op Koers* vast te stellen. RCT's worden gezien als de gouden standaard in effectonderzoek (20), echter neemt de kritiek op de onderzoeksmethode toe. RCT's zouden gezien de controlegroep waarvoor meer deelnemers nodig zijn tijd- en geld rovend zijn. Deze kritiek sluit aan bij de ervaringen opgedaan met het onderzoek in dit proefschrift, waar de werving en inclusie meer tijd heeft gekost dan van tevoren gedacht. Er zijn onderzoekers die pleiten voor bijvoorbeeld single-case studies (21), die beter haalbaar en generaliseerbaar zouden zijn. Voor toekomstig onderzoek is het belangrijk het onderzoeksdesign zorgvuldig af te wegen alvorens een keuze te maken. Daarnaast is het, om een interventie effect goed te kunnen onderzoeken, van belang om een weloverwogen keuze te maken voor uitkomstmaten.

Tot slot, aangaande **technologie**, is de wereld van e-health zich snel aan het ontwikkelen. *Op Koers Online* is een aantal jaren geleden ontwikkeld. Sindsdien is men, in het bijzonder tijdens de COVID-19 pandemie, meer gewend aan online zijn en bijvoorbeeld videobellen. Dat maakt het concept van *Op Koers Online* waarbij alleen een chatbox gebruikt wordt enigszins achterhaald. Hoewel het gebruik van een chatbox waarin alleen tekst gebruikt wordt een bewuste keuze is geweest om anonimiteit te kunnen waarborgen en deelname zo laagdrempelig mogelijk te houden, is het de moeite waard te experimenteren met het gebruik van videoverbinding. Op basis van de ervaringen kan wellicht het format van de interventie worden aangepast.

Al met al staat er met *Op Koers* een uniek breed ontwikkeld interventieprogramma voor kinderen en jongeren met een chronische aandoening of kanker, hun broers en zussen en hun ouders. De face-to-face cursussen worden gegeven door verschillende ziekenhuizen in Nederland, in een ziekenhuis in Zweden en er zijn momenteel vijftien artikelen verschenen in internationale peer-reviewed tijdschriften. Dit maakt *Op Koers* een van de weinige psychosociale interventies in de pediatrische psychologie die intensief onderzocht en geïmplementeerd is. Een belangrijke factor die hiertoe heeft bijgedragen is de nauwe samenwerking tussen de klinische zorg en de onderzoeksafdeling.

Belangrijkste bevindingen

- Deelname aan *Op Koers Online* voor jongeren had over het algemeen een positief effect op het toepassen van adaptieve coping vaardigheden en kwaliteit van leven;
- Jongeren zijn tevreden over deelname aan een online psychosociale groepscursus;
- Ouders van een kind met een chronische aandoening hebben behoefte aan psychosociale hulp voor zichzelf, gericht op verschillende thema's zoals 1) de chronische aandoening van het kind, 2) het gezin, 3) zorgen voor jezelf, 4) de omgeving en 5) praktische steun. *Op Koers Online* voor ouders is ontwikkeld aan de hand van de behoeftes van ouders;
- Deelname aan *Op Koers Online* voor ouders had een positief effect op het verminderen van gevoelens van angst en depressie en het toepassen van ziekte gerelateerde coping vaardigheden;
- Sommige effecten namen af op de lange termijn. Vervolgstappen moeten zich richten op het behouden van de lange termijn effecten van de interventies;
- Binnen het *Op Koers* programma zijn de afgelopen 25 jaar verschillende interventies ontwikkeld, geëvalueerd en geïmplementeerd in de klinische praktijk;
- *Op Koers Online* voor jongeren en ouders is klaar om geïmplementeerd te worden in de klinische praktijk. Het is belangrijk dat implementatie gedegen en gestructureerd uitgevoerd wordt.

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LIST OF PUBLICATIONS

International peer-reviewed publications

Douma, M., Joosten, M. M. H., Scholten, L., Maurice-Stam, H., & Grootenhuis, M. A. (2019). Online cognitive-behavioral group intervention for adolescents with chronic illness: A pilot study. *Clinical Practice in Pediatric Psychology, 7*(1), 79–92. https://doi.org/10.1037/cpp0000274

Douma, M., Bouman, C. P., van Oers, H. A., Maurice-Stam, H., Haverman, L., Grootenhuis, M. A., & Scholten, L. (2020). Matching psychosocial support needs of parents of a child with a chronic illness to a feasible intervention. *Maternal and Child Health Journal*, *24*, 1238-1247. https://doi.org/10.1007/s10995-020-02925-3

Douma, M., Scholten, L., Maurice-Stam, H., & Grootenhuis, M. A. (2018). Online cognitive-behavioral based group interventions for adolescents with chronic illness and parents: study protocol of two multicenter randomized controlled trials. *BMC Pediatrics*, *18*(1), 1-10. https://doi.org/10.1186/s12887-018-1216-6

Douma, M., Maurice-Stam, H., Gorter, B., Houtzager, B. A., Vreugdenhil, H. J., Waaldijk, M., Wiltink, L., Grootenhuis, M. A. & Scholten, L. (2021). Online psychosocial group intervention for adolescents with a chronic illness: A randomized controlled trial. *Internet Interventions*, *26*, 100447. https://doi.org/10.1016/j.invent.2021.100447

Douma, M., Maurice-Stam, H., Gorter, B., Krol, Y., Verkleij, M., Wiltink, L., Scholten, L., & Grootenhuis, M. A. (2021). Online psychosocial group intervention for parents: positive effects on anxiety and depression. *Journal of Pediatric Psychology*, *46*(2), 123-134. https://doi.org/10.1093/jpepsy/jsaa102

Joosten, M., van Gorp, M., Maurice-Stam, H., Douma, M., Scholten, L., & Grootenhuis, M. A. Development, research and implementation of the Psychosocial Group intervention Op Koers: Lessons learned. *Submitted*

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Chapter 2: Online cognitive-behavioral group intervention for adolescents with chronic illness: A pilot study

Miriam Douma: Conception and design of the study; data acquisition; data preparation and analysis; data interpretation; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Mala M. H. Joosten: Conception and design of the study; data acquisition; data preparation and analysis; data interpretation; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Linde Scholten: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version. Heleen Maurice-Stam: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version. Martha A. Grootenhuis: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version.

Chapter 3: Matching psychosocial support needs of parents of a child with a chronic illness to a feasible intervention

Miriam Douma: Conception and design of the study; data acquisition; data preparation and analysis; data interpretation; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Charlotte P. Bouman: Conception and design of the study; data acquisition; data preparation and analysis; data interpretation; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Hedy A. van Oers: Supervision of conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Lotte Haverman: Supervision of conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Martha A. Grootenhuis: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version. Linde Scholten: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version.

Chapter 4: Online cognitive-behavioral based group interventions for adolescents with chronic illness and parents: Study protocol of two multicenter randomized controlled trials

Miriam Douma: Conception and design of the study; writing first draft of manuscript;

critical revision of manuscript for intellectual content; approved final version. Linde Scholten: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Heleen Maurice-Stam: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Martha A. Grootenhuis: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version.

Chapter 5: Online psychosocial group intervention for adolescents with a chronic illness: A randomized controlled trial

Miriam Douma: Conception and design of the study; data acquisition; data preparation and analysis; data interpretation; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Heleen Maurice-Stam: Conception and design of the study; (supervision of) data acquisition; (supervision of) data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version. Bianca Gorter: Critical revision of manuscript for intellectual content; approved final version. Bregje A. Houtzager: Critical revision of manuscript for intellectual content; approved final version. Hestien J. I. Vreugdenhil: Critical revision of manuscript for intellectual content; approved final version. Maaike Waaldijk: Critical revision of manuscript for intellectual content; approved final version. Lianne Wiltink: Critical revision of manuscript for intellectual content; approved final version. Martha A. Grootenhuis: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version. Linde Scholten: Conception and design of the study; supervision of data acquisition; supervision of data preparation and analysis; data interpretation; critical revision of manuscript for intellectual content; approved final version.

Chapter 6: Online psychosocial group intervention for parents: Positive effects on anxiety and depression

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Chapter 7: Development, research and implementation of the psychosocial group intervention Op Koers: Lessons learned

Mala M. H. Joosten: Conception and design of the study; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Marloes van Gorp: Conception and design of the study; writing first draft of manuscript; critical revision of manuscript for intellectual content; approved final version. Heleen Maurice-Stam: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Miriam Douma: Critical revision of manuscript for intellectual content; approved final version. Linde Scholten: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version. Martha A. Grootenhuis: Conception and design of the study; critical revision of manuscript for intellectual content; approved final version.

FINANCIAL SUPPORT

The research in this thesis was supported by Fonds Nuts Ohra (FNO) under grant number 100.977).

PhD PORTFOLIO

Name: Miriam Douma
PhD period: March 2016 – June 2023
Promotor: Prof. dr. Martha A. Grootenhuis
Co-promotores: Dr. Linde Scholten
Dr. Heleen Maurice-Stam
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Emma Children’s Hospital, Amsterdam UMC

1. PhD training	**Year**	**Workload (ECTS)**
Courses		
Scientific writing in English for publication	2016	1.5
Basic course legislation and organization for clinical researchers (BROK)	2017	1.0
Practical Biostatistics	2017	0.8
Research data management	2018	0.7
Medical literature: EndNote	2018	0.1
Medical literature: Searching Literature	2018	0.1
Oral Presentation in English	2018	0.8
Project management	2019	0.6
Writing patient information forms	2019	0.3
Seminars, workshops and master classes		
Two-weekly research meeting *Psychosocial department, Emma Children’s Hospital*	2016 - 2020	0.2
TULIPS 2-years PhD-curriculum (Training Upcoming Leaders in Pediatric Science)	2018 - 2020	2.8
TULIPS Young Investigators Day (Training Upcoming Leaders in Pediatric Sciences)	2017 - 2020	1.0
Oral presentations		
Op Koers Online: psychosociale groepscursussen voor jongeren met een chronische aandoening, broers, zussen en ouders *Symposium “Zorg aan tieners”*	2019	0.8
Positive effects of an online psychosocial group intervention for parents of children with a chronic illness: A randomized controlled trial *Amsterdam Kindersymposium AKS*	2020	0.5
Presentations about *Op Koers Online* in the participating hospitals for health-care professionals (concerning recruitment and inclusion) *The Netherlands*	2016 - 2018	0.5
Presentations about *Op Koers Online* at several patient organizations *The Netherlands*	2016 - 2020	
Poster presentations		
Op Koers Online: Effectiveness and feasibility of a psycho-educational group intervention for adolescents with a chronic illness: A pilot study *Amsterdam Kindersymposium AKS*	2017	0.3
Development, content and first evaluation of an online group intervention for parents of children with chronic illness: Op Koers Online *European Pediatric Psychology Conference, Gent, Belgium*	2018	0.5

1. PhD training	**Year**	**Workload (ECTS)**
Development, content and first evaluation of an online psychosocial group intervention for parents of children with chronic illness *Amsterdam Public Health Research Institute Annual Meeting*	2018	1.0
Matching psychosocial support needs of parents of a child with a chronic illness to a feasible intervention *Amsterdam Kindersymposium AKS*	2019	0.3
Positive effects of an online psychosocial group intervention for parents of children with a chronic illness: A randomized controlled trial *Amsterdam Kindersymposium AKS*	2020	0.3
(Inter)national conferences		
Medical Psychology Research Meeting *Amsterdam, the Netherlands*	2016 - 2019	1.2
Amsterdam Kinder Symposium (AKS) *Amsterdam, the Netherlands*	2017 - 2020	1.2
Pediatric Psychology Network Netherlands *Utrecht, the Netherlands*	2016 - 2019	1.2
European Pediatric Psychology Conference *Gent, Belgium*	2018	0.5
Amsterdam Public Health Research Institute 3rd Annual Meeting *Amsterdam, the Netherlands*	2018	0.3
Child Health symposium, Training Upcoming Leaders in Pediatric Sciences (TULIPS) *Noordwijk, the Netherlands*	2018	0.5
Symposium "Zorg aan Tieners" (Princess Maxima Center for Pediatric Oncology) *Utrecht, the Netherlands*	2019	0.5
Child Health symposium, Training Upcoming Leaders in Pediatric Sciences (TULIPS) *Rotterdam, the Netherlands*	2019	0.5
ISOQOL-NL symposium *Amsterdam, the Netherlands*	2019	0.3
Child Health symposium *Utrecht, the Netherlands*	2018, 2019	0.6
Child Health symposium, Training Upcoming Leaders in Pediatric Sciences (TULIPS) *Egmond aan Zee, the Netherlands*	2021	0.5

2. Teaching	**Year**	**Workload (ECTS)**
Lecturing		
Lecturing medical students	2016 - 2020	1.2
Supervising		
Co-supervising applied science student	2019	0.3

3. Parameters of esteem	**Year**	**Workload (ECTS)**
Grants		
ZonMw Grant E7500,- for organizing the Young Investigator's Day (Training Upcoming Leaders in Pediatric Sciences (TULIPS)) "Van lab tot ledikant: De verbinding tussen basaal onderzoek en de kliniek." *Number 446001008*	2018	
Awards and prizes		
Best Poster Award (Amsterdam Kindersymposium AKS)	2020	

3. Parameters of esteem	Year	Workload (ECTS)
Selected for the two-year PhD curriculum Training Upcoming Leaders in Pediatric Science (TULIPS)	2018 -2020	
Other		
Member of the organizing committee of the PhD Day	2016	0.3
Member of the organizing committee of the Pediatric Psychology Network Netherlands	2017, 2018	0.6
Member of the organizing committee of the Young Investigator's Day (Training Upcoming Leaders in Pediatric Sciences (TULIPS))	2018, 2019	0.6

ABOUT THE AUTHOR

Miriam Douma was born on the 21st of May 1989 in Zwolle, where she grew up and finished high school at the Thorbecke Scholengemeenschap in 2007.

Miriam moved to Utrecht to start studying at Utrecht University. After finishing her Bachelor's degree and travelling in Asia, she moved to Amsterdam where she finished her Master's degree Forensic Psychology at the University of Amsterdam in 2014. The master included two research internships and a clinical internship. While studying, Miriam had several part-time jobs as a research assistant on different projects at the University of Amsterdam.

After graduation, Miriam started working as a research assistant at the University Medical Center Utrecht, Department of Psychiatry. She conducted a pilot study and was involved in a large European study, both in the field of neuropsychology and developmental disorders. In addition, Miriam worked part time as a psychologist at the National Psychotrauma Center at Wilhelmina Children's Hospital in Utrecht.

During her time at the UMC Utrecht, Miriam's research interests and passion for medical psychology blossomed. She started working as a PhD-student at the Emma Children's Hospital in 2016, under supervision of Prof. dr. Martha A. Grootenhuis, dr. Heleen Maurice-Stam and dr. Linde Scholten. The research was focused on the development and (pilot) feasibility and efficacy studies concerning an online psychosocial group interventions for adolescents and parents (called *Op Koers Online*), resulting in this thesis. During her time as a PhD student, she was selected for the 2-year TULIPS PhD curriculum (Training Upcoming Leaders in Pediatric Science). She was also involved in the organizing committees of several symposia.

After her time in the Emma Children's Hospital, Miriam started working as a child psychologist at the Medical Psychology of Medical Spectrum Twente. Miriam completed her PhD in her own time, simultaneously with the postmaster training to become a healthcare psychologist for children, adolescents and their parents, which she currently almost finished.

Miriam lives happily together with Martin, their children Klaas (2020) and Anna (2022) and dog Froning in Enschede.

DANKWOORD

Het boek is af! Mijn baby van 2023. Misschien wel de zwaarste bevalling tot nu toe, maar bovenal een ontzettend leuk en leerzaam traject. Er zijn een heleboel mensen die direct en indirect hebben bijgedragen aan de totstandkoming van dit proefschrift, die ik graag hier wil bedanken.

Allereerst wil ik de **jongeren** en **ouders** die hebben deelgenomen aan het Op Koers Online onderzoek hartelijk bedanken voor hun inzet, tijd en openheid. Luisteren naar en praten met jullie is het mooiste aan mijn vak en ik vind het heel bijzonder om in Op Koers Online te zien hoe jullie van elkaar leren. Alle respect voor jullie, hoe jullie in het dagelijks leven omgaan met ziekte en ziekenhuis.

Prof. dr. M. A. Grootenhuis, lieve **Martha**, veel dank voor je enthousiasme, hulp en betrokkenheid. Ik wist dat ik altijd bij jou terecht kon wanneer het nodig was, en dat was ontzettend prettig. Het is inspirerend om te zien hoe jij je inzet voor de psychosociale zorg voor het zieke kind en daarbij altijd bezig bent met de brug tussen wetenschap en kliniek. Wat een eer om met mijn promotie jouw AMC tijd officieel af te sluiten. The end of an era, waar we zeker op moeten proosten op 1 december.

Dr. H. Maurice-Stam, lieve **Heleen**, zonder jou was dit boekje er niet geweest. Veel dank voor al je hulp, het samen analyseren van de resultaten, je begeleiding bij het schrijven van de artikelen en je geduld en steun tijdens de laatste (lange) loodjes. Bij jou kon ik altijd terecht voor advies en een eerlijke visie. Ik zie ons nog zo zitten; in een klein belhokje op G8 vol spanning naar het computerscherm aan het kijken toen de SPSS output met de RCT resultaten aan het laden was. Als het te zenuwslopend werd kwam jij op het juiste moment met de conclusie dat het écht even tijd was voor koffie. Ik had dat niet zonder jou willen meemaken, dankjewel.

Dr. L. Scholten, lieve **Linde**, heel erg bedankt voor je hulp en begeleiding. Ik heb zowel vakinhoudelijk en op het gebied van onderzoek als privé veel van je geleerd. Jouw bron aan informatie lijkt soms onuitputtelijk. Wanneer ik in overleg een zin zei kwam jij altijd met honderd-en-één ideeën die ik dan driftig probeerde mee te pennen. Onze samenwerking bestond naast alle leerzame en gezellige overleggen uit zwangerschapsverloven van ons beiden. Daaruit volgden uiteraard de liefste ‘Op Koers baby’s’, maar ook wat uitdagingen. Jij hebt echter altijd en zonder enig oordeel het volste vertrouwen gehad dat het goed zou komen en dat ik dit boekje uiteindelijk af zou krijgen. Dat was ontzettend fijn. Mede dankzij jou is het er nu echt.

Geachte **promotiecommissie**, **prof. dr. J. B. van Goudoever**, **prof. dr. A. W. M. Evers**, **prof. dr. A. M. Bosch**, **prof. dr. E. M. W. J. Utens, dr. A. M. Willemen**, en **dr. C. M. Verhaak**, dank voor jullie bereidheid plaats te nemen in mijn promotiecommissie en de tijd die jullie genomen hebben voor het kritisch beoordelen van mijn proefschrift.

Charlotte, jouw keus voor klinisch werk maakte dat de vacature voor promovendus op het Op Koers Online project vrij kwam. Dank voor al je gedane (voor-)werk en de fijne inwerkperiode.

Om in die fase te blijven bedank ik graag **Dr. P. de Zeeuw** en **Dr. B. van Hulst**, Patrick en Branko, mijn begeleider en collega's uit mijn UMC Utrecht tijd. Dank voor jullie enthousiasme die me heeft doen beseffen dat onderzoek doen echt leuk kan zijn, de borrels en natuurlijk de referentie die jullie gaven aan het Emma Kinderziekenhuis.

Graag wil ik alle cursusleiders bedanken die Op Koers Online hebben gegeven in de deelnemende centra, te weten het Antonius Ziekenhuis, Canisius Wilhelmina Ziekenhuis, DeKinderKliniek, Deventer Ziekenhuis, Jeroen Bosch Ziekenhuis, Ziekenhuis St. Jansdal, Amsterdam UMC, locatie VUmc en mijn eigen locatie: het Emma Kinderziekenhuis. Dank aan alle kinderartsen, verpleegkundigen, medisch maatschappelijk werkers, pedagogisch medewerkers en alle anderen die altijd open stonden voor een presentatie tijdens overdrachtsmomenten en me enorm hebben geholpen bij het werven van deelnemers voor het onderzoek.

Speciale dank gaat uit naar de lokale coördinatoren van de deelnemende centra: **Inge Visser**, **Lianne Wiltink**, **Bianca Gorter**, **Yvette Krol** (tevens dank voor het huidige supervisietraject!), **Sandra Jansen-Kamphorst**, **Maaike Waaldijk** en **Alice van Dijk**. Dank voor jullie inzet en de altijd zeer prettige samenwerking. Zonder jullie was het zeker niet gelukt!

Veel dank gaat uit naar alle (oud)medewerkers van de **Psychosociale Afdeling (tegenwoordig VKC Psyche)** van het Emma Kinderziekenhuis, Amsterdam UMC, locatie AMC. Dank voor jullie hulp bij de werving en het meedenken. Speciale dank gaat uit naar het secretariaat dat er werkte in 'mijn tijd'. **Marga**, dank voor je hulp, altijd een praatje en het lachen. **Michal**, in liefdevolle herinnering, we hadden ons geen betere Op Koers ambassadeur op de afdeling kunnen wensen.

Pieter en **Ronald** van IPPO, zonder website geen online groepscursus, dank voor de prettige samenwerking en jullie eeuwige geduld met uitleggen en blijven aanpassen.

Lieve collega's van H7, ofwel **Teven van H7**, het leven van een onderzoeker wordt leuk gemaakt door collega's. Dank voor de fijne samenwerkingen, het spuien als het bij een van ons even tegenzat, de koffies op het Voetenplein, het planken en de push-ups. **JP**, het voelt al als een eeuwigheid geleden dat we samen op H7 zaten (is het ook), dank voor het lachen. **Florrie**, **Perrine** en **Mala**, Teven van H7 back in the days, ook wel de Harde Kern. Er bestaat geen appgroep die zo levendig is terwijl ik de mensen zo zelden zie. Bedankt voor het klagen, lachen en *sparren*. **Anouk** en **Madelief**, inhoudelijk hadden we op H7 niet veel met elkaar te maken, maar des te meer hebben we met elkaar gedeeld (en gelachen). **Hedy**, bij jou kon ik altijd terecht voor een luisterend oor en een oprecht antwoord. **Lorynn**, bijna samen gepromoveerd, maar nu is het over twee weken gewoon nog een keer tijd voor een feestje. **Maud**, het toonbeeld van een harde werker die toch altijd in is voor koffie en gezelligheid. **Michiel**, altijd geïnteresseerd in de ander en super behulpzaam, je altijd geduldige adviezen en het meedenken bij statistische uitdagingen (voor mij dan) hebben me enorm geholpen. **Thirsa**, zoveel hebben we niet tegelijk op H7 gezeten, maar wij gaan van CrossFit in het hoge noorden tot eindeloze SPSS app sessies. **Sophie**, de aanhouder wint, we zouden een lotgenotengroep moeten oprichten. Nu al zin in jouw promotie!

Last but not least, **Lotte**, moedergans van alle onderzoekers. Hoewel ik als een van de weinigen van onze groep niet officieel onder jouw vleugels viel, was je altijd bereid tot meedenken en een goed advies. Dankjewel daarvoor.

De volgende mensen hebben niet zozeer bijgedragen aan de totstandkoming van dit proefschrift, maar zijn wel onmisbaar in mijn leven en verdienen daarom een bedankje.

Lieve vriendinnen, dank voor jullie vriendschap. **Soof**, mijn 'oudste' beste vriendinnetje, dat zul je altijd blijven. Vroeger waren we praktisch zusjes, hopelijk pakken we na de tropenjaren die lijn weer meer op. **Tessa**, we leven op ruim 10.000 km afstand van elkaar en toch hoor ik niemands stem vaker dan die van jou in ontelbaar veel minuten aan voicemessages. Met jou kan ik echt alles delen. **Kwijl** oftewel **Lijn** (even die bijnaam goed maken), wij hebben er samen al vele hardloopkilometers en vakanties op zitten. Jij bent een ontzettend relaxt persoon en altijd in voor een uitje. Als we niet inmiddels beiden een gezin hadden gesticht, hadden we misschien nog steeds samen op de Spaarn gewoond. Zo gek was dat eigenlijk nog niet geweest. **Fiek**, het begon allemaal tijdens onze scriptiestage. Vele uren met jou (en rijstwafels met pindakaas) in een hokje dossiers doorspitten en typen, en ons verschuilen voor die enge onderzoekers daar. Wat is het fijn om nog steeds lief en (werk-)leed met elkaar te kunnen delen. **Teun** en **Knabbel**, onze Schoolstraat tijd ligt lang achter ons maar altijd als we elkaar zien is het weer even fijn als toen. De goede oude **wijnclub**, helaas sinds Corona en alle veranderende levens niet meer in ere, maar tijdens mijn promotietraject nog wel en ik koester warme herinneringen aan avonden vol wijn en verhalen en zelfs heuse wijnclubvakanties.

Mijn vriendinnetjes van vroeger, **Madelon**, **Taban**, **Margot**, **Eslie** en **Ellemieke**, de Peperbus blijft altijd knus (en de IJsselbrug ook).

Vrienden die ik wil bedanken voor de gezelligste dubbeldate avonden met lekker eten en drinken en vooral veel spelletjes: **Floris en Hedi**, **Wytze en Christa** (extra dank voor de fijne weken in Portugal, wanneer gaan we weer?), **Michiel en Bente**, **Jelle en Vera** en onze nieuwe Twentse spelletjes slachtoffers: **Laura en Johan**.

Mijn **collega's van het MST**, die me een leerzame tijd en (zonder dat jullie het zelf misschien weten) een warm welkom hebben gegeven in Enschede. Dank voor de fijne samenwerking. Speciale dank aan mijn 'kindercollega's' **Anneke**, **Anouk** en **Ingrid**, het is inspirerend om zo nauw met jullie samen te werken.

Mijn paranimfen, wat ben ik blij en trots dat jullie letterlijk en figuurlijk naast mij staan.

Lieve **Ell**, van MSN'en na schooltijd op de middelbare school tot het nachtnet terug vanuit Delft naar Utrecht tijdens onze studententijd, tot elke woensdag meisjesavond in Amsterdam, tot elkaar nu veel te weinig zien maar weten dat het altijd goed zit. Jij bent de meest oprechte persoon die ik ken en waar ik altijd bij terecht kan. Dankjewel voor je onvoorwaardelijke vriendschap.

Lieve **Mala**, naast je eindeloze mentale steun heb jij ook in praktische zin veel betekent voor mijn onderzoek. Samen reisden we naar de deelnemende centra om honderden wervingsbrieven te versturen. Dat ging niet altijd even soepel en ik krijg nog steeds de slappe lach als ik aan Harderwijkewout denk. Ik ben blij dat ik via werk zo'n goede vriendin heb leren kennen, en wat is het daarbij bijzonder dat we tegelijk kindjes kregen en een tijdje bij elkaar om de hoek woonden. Dankjewel voor je altijd nuchtere kijk op de zaken en urenlange app sessies over alles (sorry Wessel!).

Lieve **Jona**, **Nikki**, **Sam** en **Lot**, **Thomas**, **Alisa**, **Willem (en de baby)**, **Madelief**, **Koen**, **Guus** en **Kate**, **Joost**, **Judith**, oftewel (samengestelde) broers en zussen, aanhang, neefjes, nichtjes en een bonusgezinslid. De tafel wordt steeds voller en daarmee ook steeds gezelliger. Jullie hebben misschien geen idee waar dit boekje over gaat, maar jullie zijn van grote waarde bij de verdediging. Hetzelfde geldt voor mijn schoonfamilie **Willem Jan**, **Maartje**, **Jan Willem**, **Floris** en **Myrthe**, **Pauline**, **Sander**, **Noek**, **Stach**, **Maes** en **Lauren**, **Jorine**, **Isaac**, **Samuel** en **Sophie**, dank voor alle chaotische gezelligheid van de afgelopen jaren met als hoogtepunt een jaarlijks weekendje Hof van Saksen.

Oma Jannie, helaas niet meer bij ons, bedankt dat u me leerde te genieten van de kleine dingen.

Lieve **Engelien (mama)**, **Johan**, **Mame** en **Alette**; ik kan jullie alle vier niet genoeg bedanken voor jullie hulp, interesse, onvoorwaardelijke steun en betrokkenheid.

Lieve **mama**, ik hoop dat ik later net zo'n band mag hebben met mijn kinderen als ik met jou heb. Ik geniet enorm van samen zijn met de kindjes, maar minstens even veel van onze moeder-dochter uitjes (die we gelukkig nu weer regelmatig plannen). Lieve **Johan**, ik ken niemand die zoveel weetjes kent als jij. Bedankt voor je altijd oprechte interesse in alles waaronder mijn werk en ook in dit boekje.

Lieve **Mame**, bedankt voor je vertrouwen en je altijd ontnuchterende visie. Van jou leerde ik dat je je best doet, en meer kun je niet doen. Iets wat ik nog regelmatig tegen mezelf zeg als ik iets spannends moet doen.

Lieve **Alette**, dat jij geen onderscheid maakt in 'koude kant' is echt te merken. Bedankt voor onze gesprekken over van alles. Ik had me geen betere schoonmoeder kunnen wensen.

Lieve **Froning**, Frootje, het is niet gebruikelijk om je hond te bedanken in je proefschrift, maar aangezien verschillende zinnen in dit boekje zijn bedacht tijdens een wandeling met jou kon je niet ontbreken. Meer dan eens moest ik tijdens het lopen stilstaan om snel een bepaalde formulering of idee in te typen op mijn telefoon. Dank dat je ons altijd naar buiten dwingt voor een verfrissende wandeling en 's avonds onze laptops van schoot duwt als we te lang werken en het écht tijd wordt om aandacht aan jou te besteden.

Dan mijn kinderen, wat ben ik dol en trots op jullie. Lieve **Klaas**, onze grappenmaker. Jij kletst de hele dag door. Je hebt fantasie voor tien en dat werkt erg relativerend, want je kunt altijd doen alsof. Je bent ontzettend zorgzaam en leeft zo mee met 'mama's boek'. Helaas is het niet zo interessant voor jou, maar ooit zal je in ieder geval deze alinea zelf lezen. Lieve **Eva**, je leefde maar een halve zwangerschap in mijn buik, maar bent voor altijd ergens bij me. Lieve **Anna**, onze knuffelkont. Al vanaf dat je een paar maanden oud was geef jij de lekkerste knuffels. Je weet heel goed wat je wil, bent het liefst omringd door anderen, dol op dieren en het liefst buiten op je loopfiets. Je wordt helemaal wild van elke tractor of brandweerauto die we zien, dat is je met de paplepel ingegoten door je broer. Lieve boefmuisjes, jullie doen me elke dag beseffen wat echt belangrijk is in het leven. Ik ben blij dat ik jullie moeder ben.

Liefste **Martin**, Piet, het laatste plekje is natuurlijk voor jou. Toen ik in het AMC werkte belde jij geregeld naar mijn vaste AMC telefoon die vaak door iemand anders werd opgenomen. Daarbij ontstond verwarring over wie Martin zou zijn, want ik was toch aan het daten met ene Piet? Daarom voor de duidelijk hier beide namen. Het is zover, nu hoef je nooit meer te vragen hoe het met mijn boekje gaat. Nou ja, dat mag wel, maar dan kan ik je eindelijk verwijzen naar de boekenkast. Dankjewel dat jij, hoe druk het ook is of misschien júist als het druk is, altijd zorgt voor een (flauwe) grap om me aan het lachen te krijgen, de nodige knuffels en voor tijd voor elkaar. Ik ben elke dag weer blij en trots dat jij bij mij hoort. Jouw ambitie is bewonderingswaardig en ik vind het ontzettend knap wat je allemaal bereikt. Ik hou van je en kijk uit naar de toekomst met jou (en naar meer laptopvrije avonden). Ik zeg altijd maar zo: als we maar samen zijn.