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## Coping with Pediatric Chronic Illness

Scholten, L.

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# Chapter 8

## Summary and General Discussion



The research for this thesis was conducted to extend understanding on how to promote positive adjustment to pediatric chronic illness by investigating a group intervention ('Op Koers') for children with various chronic illnesses and their parents. The 'Op Koers' intervention aims to teach children to use skills that help them cope with the consequences of their disease, and in an additional parent component parents are taught to encourage their children in using the learned coping skills in everyday life. As described in Chapter 1, specific aims for the five studies described in this thesis were: (1) to give insight in the level of behavior problems in children who applied for the intervention, and in associated risk- and protective factors, (2) to investigate whether 'Op Koers' is effective in reducing adjustment problems in children with a chronic illness, and to examine the extent to which the parent component enhances the effect, (3) to examine moderation of intervention effects by pre-intervention risk- and protective factors, (4) to explore whether changes in disease related coping skills and in parent-child interactions are the mechanisms through which the 'Op Koers' intervention induces changes in adjustment problems, and (5) to evaluate feasibility in order to provide directions for future implementation. In this concluding chapter, the findings of the previous chapters are summarized and integrated, and recommendations for future research and clinical implications are given. Objectives, characteristics, and main findings of the different chapters are also presented in Table 8.1.

## MAIN FINDINGS

### Level of adjustment problems and putative risk- and protective factors: Why intervene?

Our first aim was to determine the levels of adjustment problems (internalizing and externalizing behavior problems) in children who applied for the intervention. Furthermore, in line with existing transactional models of child adaptation to chronic illness (Wallander & Varni, 1998), we investigated associations between psychosocial adjustment (internalizing and externalizing behavior problems), and several illness (type, duration, severity), stress-processing (engaged- and disengaged coping), intrapersonal (self-worth), and parent factors (parenting stress and the security of the parent-child relationship). For this cross-sectional study, described in **Chapter 2**, we used the baseline data of the efficacy trial described in the later chapters ( $N = 194$  children aged 8-18 and their primary caregiver).

Findings revealed that both parents and, if they were old enough to self-report ( $> 11$  years of age), children reported higher levels of internalizing problems than samples from the general population ( $d = 1.04$ , and  $.62$ ). When the 'somatic complains' items were disregarded from the internalizing problem scale, parent-reported anxious/withdrawn behavior was also higher than the norm ( $d = .51$ ), while levels of self-reported



internalizing problems were comparable to the norm ( $d = -.06$ ). Parents reported more externalizing problems ( $d = .25$ ), and children reported less externalizing problems than the norm ( $d = -.25$ ). Percentages of internalizing problems within the subclinical or clinical range were 48% according to parents, 15% according to children. Percentages of externalizing problems within subclinical or clinical range were 19% based on parental report, and 4% based on self-report. Overall, levels of behavior problems were similar to those from previous studies in children with chronic illness (Pinquart & Shen, 2011), and underlined the value of multi-informant assessments, and caution in interpreting internalizing problems scales with somatic items (Perrin, Stein & Drotar, 1991). Furthermore, it should be noted that the sample of children with chronic illness was not recruited to be representative of the whole population of children with chronic illnesses, but represented children and their parents who responded to the offer of a psychosocial support intervention.

Results on risk- and protective factors showed that more use of child disengaged coping, lower self-worth, less security of the parent-child relationship, and more parenting stress were uniquely associated with more behavior problems. However, associations depended on outcome and informant. Disengaged coping contributed uniquely to explaining higher levels of self-reported internalizing and externalizing problems, while engaged coping explained no unique variance. Lower self-worth was uniquely associated with higher levels of self- and parent-reported internalizing problems. Lower security of the parent-child relationship contributed uniquely to explaining higher levels of self-reported externalizing problems, and higher parenting stress was uniquely associated with higher levels of parent-reported behavior problems. We found no links between illness characteristics (type, duration, course) or socio-demographic characteristics and behavior problems, with the exception of a weak association between illness severity and internalizing problems.

In sum, findings of this study indicated that behavior problems of children with chronic illness should be considered as an outcome of a dynamic process in which interrelated risk and protective factors contribute. Moreover, findings support the idea that interventions for children should be family-based and should target coping and positive self-perceptions. Finally, the lack of associations between adjustment problems and illness characteristics underscores the case for an illness-generic approach to pediatric psychosocial intervention program.

### **Efficacy of the 'Op Koers' intervention: Does it work?**

The second and main objective of this thesis was to investigate the efficacy of the 'Op Koers' intervention for children with chronic illnesses, and to test the effect of an added parent component. Previous studies have suggested that adjustment in children with chronic illness may be promoted by teaching active coping skills using cognitive behavioral therapeutic techniques (Barlow & Ellard, 2004; Beale, 2006; Plante et al.,

2001; Thompson et al., 2011), and that multi-component interventions that include parents might enhance efficacy (e.g., Beale, 2006; Eccleston et al., 2012; Thompson et al., 2011). However, few parent-child interventions have been stringently evaluated, and interventions have been developed with specific diseases in mind, while evidence fails to support illness-specific risk factors for behavioral maladjustment.

To study the efficacy of 'Op Koers', a research protocol was developed following Consort Statements (Schulz et al., 2010) (**Chapter 3**). A total of 194 children and their parents participated in a multicenter randomized controlled trial (RCT) which compared the 'Op Koers' intervention for children only (child-only intervention), and the 'Op Koers' intervention with a parent component (parent-child intervention), to a waitlist control group. Primary outcomes were parent- and self-reported internalizing and externalizing problems; secondary outcomes were child disease-related coping skills (information seeking, relaxation, social competence, medical compliance, and positive thinking). Questionnaires were assessed online at baseline, at 6-month, and at 12-month follow-ups.

Results showed that the 'Op Koers' intervention had a positive effect on changes in parent-reported internalizing problems, child-reported externalizing problems, information seeking, social competence, and positive thinking, compared to the waitlist group (**Chapter 4**). The additional effect of a parent component was observed on parent-reported internalizing problems, child-reported externalizing problems, information seeking, and social competence. For internalizing problems parent involvement appeared to be particularly important for the long term effect (6 to 12 month follow-up). Although intervention effects were comparable to effects of similar interventions (Hampson et al., 2000; Nguyen et al., 2011), they were small in magnitude ( $d = .22 - .42$ ). In addition, there were no intervention effects on child-reported internalizing problems, parent-reported externalizing problems, relaxation, or medical compliance. Small effect size and lack of intervention effects on some outcomes may be due to the fact that 74% of the families in the waitlist control group sought alternative psychological support during the intervention period. While it is unclear to what extent this additional support may have attenuated effects, it does suggest that the intervention addresses a need for psychosocial support in children with chronic illness.

Concluding, the RCT supported the efficacy of the 'Op Koers' intervention. Adding a parental component to the intervention contributed to the (persistence of the) effects.

### **Moderators of intervention effects: For whom does it work best?**

Our third aim was to investigate whether illness, child and family risk- and protective factors, act as moderators of the effect of a the 'Op Koers' intervention. It is unrealistic to think that all children benefit equally from an intervention (La Greca, Silverman, & Lochman, 2009). Therefore, to investigate for whom it works best, and with which protocol (child-only versus parent-child), we studied pre-treatment factors as moderators (Hinshaw, 2007; Simon & Perlis, 2010).



Generally, moderators in intervention studies are based on routinely obtained information such as age, gender or initial level of problems (Kazdin, 2007). However, moderators that are theoretically grounded may be even more important to consider. Little is known about the intervention effects for children with different types and levels of risk and resilience (La Greca, Silverman, & Lochman, 2009), limiting the development of practices that tailor interventions to individual needs and strengths (Scott & Dadds, 2009; Weisz et al., 2012).

In **Chapter 4** we tested moderation of illness severity and illness type, and found no evidence that the intervention works better or less for children with different diagnoses or different levels of severity. This is important information, because it provides support for the disease generic approach of the 'Op Koers' intervention.

In **Chapter 5** we investigated whether age, gender, baseline levels of behavior problems, and the risk- and protective factors that were identified in Chapter 2 (coping style, self-worth, the security of the parent-child relationship, and parenting stress) moderated intervention effects on behavior problems. Findings indicated that for children with a more disengaged coping style and for children with lower self-worth, the parent-child intervention was more effective than the child-only intervention in reducing behavior problems. In addition, we found that intervention effects were the same for both boys and girls, for children of all ages, for children with all levels of baseline problems, and for children which parents experienced different levels of parenting stress. Our moderation results were in line with previous studies that describe that greatest improvements can be expected for children who have the most to learn from the intervention (e.g., Hautmann et al., 2009). However, although previous research has found that inclusion of parents in intervention was especially effective for the most distressed parents (Crawford & Manassis, 2001), our findings pointed in a different direction. The parent-child intervention modality was especially effective if the parent-child relationship was experienced as more secure. This indicates that the intervention itself did not have an influence on security of the relationship, but that parents and children were able to apply more effectively the skills learned if there was already good communication and trust between parents and children.

It was concluded that children who are more 'at risk' on factors that are targeted in the 'Op Koers' intervention have the greatest scope for positive behavior change, and for these children involving parents in the intervention is even more important. However, parental encouragement of child coping skills appeared to be more successful in emotionally secure, good quality relationships. This suggests that for children with insecure parent-child relationships, alternative interventions that focus more thoroughly on parent-child interactions are needed.

### **Mediators of intervention effects: How does it work?**

The fourth objective of this thesis was to explore whether changes in disease related coping skills and in parent-child interactions are the mechanisms through which the

'Op Koers' intervention induces changes in child problem behaviors. Although many scholars stress the importance of identifying underlying mechanisms of interventions, studies on mediators of change are scarce in pediatric literature (e.g., Kraemer et al., 2008; La Greca et al., 2009). In **Chapter 6** results of mediation analysis are described. Mediator variables included self-reported use of information seeking, positive thinking, and social competence, parent-reported child use of general coping skills, and observations of parent-child interactions (measured with a semi-structured observation task).

Findings revealed that the effects of the intervention on decreases in behavioral outcomes were mediated by improvements in disease related coping skills. In families where children and parents both participated in the intervention, social competence skills improved more, which in turn was related to stronger decreases in internalizing and externalizing problems. Indirect effects were also found via information seeking and positive thinking, but no significant mediation was found. Positive parent-child interactions were related to decreases in problems, but did not explain the additional effect of parental involvement in the intervention. Effects of coping skills on decreases in problems were stronger when parents were also involved in the intervention, indicating that this may promote the use of coping skills in daily life.

Results emphasize that improvement in disease related coping skills are important working mechanisms of a group-intervention for children with chronic illness. This result provides important experimental evidence that these coping skills are causally related to behavioral adjustment in children with chronic illness. Parental involvement in the intervention appears important to promote the use of coping skills in daily life, resulting in child adjustment on the long term.

### **Feasibility: Can it be implemented?**

Our fifth and final aim was to learn about ways in which implementation of the 'Op Koers' intervention in clinical pediatric practice may be promoted, by studying feasibility and practical issues. Previous research indicated that even when interventions are proven to be effective, the translation of these interventions into clinical practice remains a complex and haphazard process (Drotar, 2010; Spirito & Kazak, 2006; Stark, 2010). This is problematic, because it is recognized that poorly implemented and executed interventions are significantly less effective (Berwick, 2003).

In **Chapter 7** we investigated several outcomes that provide avenues for future implementation. First, we discussed recruitment, attrition and logistical issues of conducting the 'Op Koers' intervention during the RCT (in three academic hospitals, four non-academic hospitals, and two primary schools for chronically ill children). Second, we assessed whether the intervention goals matched the support needs of the families. Finally, we evaluated satisfaction with the intervention reported by children, parents and psychologists directly after the interventions.





**TABLE 8.1** Summary of Main Findings

CHAPTER	TITLE	OBJECTIVES	PARTICIPANTS/PROCEDURES
2	Behavior problems in children with chronic illness: Risk- and protective factors	To describe behavior problems in children with various chronic illnesses who seek psychosocial support, and to examine associated risk factors and protective factors.	<i>N</i> = 194 children (8-18) with various chronic illnesses, and their primary caregiver  Baseline data of the RCT
4	Efficacy of psychosocial intervention in pediatric chronic illness	To investigate the efficacy of the 'Op Koers' intervention, and to test the effect of an added parent component	<i>N</i> = 194 children with various chronic illnesses, and their primary caregiver  Assessments were at baseline, 6-month and 12-month follow-up
5	Moderators of intervention effects on behavior problems in children with chronic illness	To investigate psychosocial characteristics of children and parents as well as parent-child relationships as moderators of the effect of the 'Op Koers' intervention	<i>N</i> = 194 children with various chronic illnesses, and their primary caregiver  Assessments of outcome were at baseline, 6-month and 12-month follow-up. Assessments of moderators took place at baseline
6	Coping and Parental Involvement as Working Mechanisms of Intervention Effects on Behavior Problems in Children with Chronic Illness	To explore whether changes in disease related coping skills and in parent-child interactions are the mechanisms through which the 'Op Koers' intervention induces changes in child problem behaviors.	<i>N</i> = 194 children with various chronic illnesses, and their primary caregiver  Assessments of outcome were at baseline, 6-month and 12-month follow-up. Assessments of mediators were at baseline, directly after intervention, and 6-month follow-up
7	Feasibility and directions for future implementation	To provide leads for future implementation in clinical pediatric practice by investigating feasibility of the 'Op Koers' intervention.	<i>N</i> = 95 children <i>N</i> = 42 parents <i>N</i> = 35 psychologists  Assessments were directly after the intervention

MEASURES	MAIN FINDINGS
<p><i>Outcomes:</i> Internalizing and externalizing behavior problems (CBCL/YSR)</p> <p><i>Risk- and protective factors:</i> Illness severity, Coping strategies (CSI), Self-worth (SPPC/A), Security of the parent-child relationship (CSS), Parenting stress (PSI)</p>	<ul style="list-style-type: none"> <li>● Parents and children reported elevated levels of internalizing problems compared to the Dutch norm population, and children with chronic illness in general</li> <li>● More use of child disengaged coping, lower self-worth, less security of the parent-child relationship and more parenting stress were uniquely associated with more behavior problems</li> </ul>
<p><i>Primary outcomes:</i> Internalizing and externalizing behavior problems (CBCL/YSR)</p> <p><i>Secondary outcomes:</i> Disease related coping skills (QOK: information seeking, relaxation, social competence, medical compliance and positive thinking)</p>	<ul style="list-style-type: none"> <li>● Positive intervention effects were found on parent-reported internalizing problems, child-reported externalizing problems, information seeking, social competence, and positive thinking</li> <li>● Adding a parental component to the intervention contributed to the (persistence) of the effects on parent-reported internalizing problems, child-reported externalizing problems, information seeking, and social competence</li> </ul>
<p><i>Outcomes:</i> Internalizing and externalizing behavior problems (CBCL/YSR)</p> <p><i>Moderators:</i> Disengaged coping scale (CSI), Self-worth (SPPC/A), The security of the parent-child relationship (CSS), Parenting stress (PSI), age, gender, Baseline problems (CBCL/YSR)</p>	<ul style="list-style-type: none"> <li>● Children who are more ‘at risk’ on factors targeted in the intervention appear to gain more from participating in intervention, especially if their parents are involved as well</li> <li>● Benefit of parents’ involvement may depend on the quality of the parent-child relationship</li> </ul>
<p><i>Outcomes:</i> Internalizing and externalizing behavior problems (CBCL/YSR)</p> <p><i>Mediators:</i> Disease related coping skills (QOK), Parent-child interactions (FIT)</p>	<ul style="list-style-type: none"> <li>● Child coping skills and social competence mediated respectively the effect of the child-only intervention and the parent-child intervention compared to the waitlist, indicating that these skills were working mechanisms of the intervention.</li> <li>● Involvement of parents in the intervention strengthened the effect of coping skills on behavioral outcomes</li> <li>● Positive parent-child interactions were related to decreases in problems, but did not explain the additional effect of parental involvement in the intervention.</li> </ul>
<p><i>Outcomes of feasibility:</i> Recruitment rates, logistical issues reported by psychologists, support needs reported by parents, and satisfaction with the intervention reported by children, parents and psychologists</p>	<p>Findings indicate that the ‘Op Koers’ intervention is feasible for implementation in clinical practice of pediatric settings. Possible directions for future implementation are:</p> <ul style="list-style-type: none"> <li>● Improve recruitment methods and risk screening</li> <li>● be innovative to overcome logistical problems</li> <li>● tailor interventions to individual needs</li> <li>● secure treatment integrity</li> <li>● monitor implementation with research</li> </ul>



Results of this evaluation study showed that overall recruitment resulted in 19% who applied for the intervention. Attrition rates were low (7.0% attrition in the child-intervention, and 4.1% in the parent-child intervention). In addition, there was considerable overlap of reported support needs and intervention goals. Finally, children, parents and psychologists were overall satisfied with the content of the program. Improvements that were reported were renewal of the relaxation exercise, and more time for exchange of personal experiences in the parent protocol.

In sum, implementation of the 'Op Koers' intervention in clinical pediatric practice appears to be possible. Although the majority of children and their parents might not perceive a need for this intervention, participation from the population of children with chronic illness was still considerable (about 1 out of 5). Possible directions for future implementation are; (1) improve recruitment methods (e.g., systematic screening), (2) be innovative to overcome logistical problems (e.g. offer interventions online), (3) beware of the match between support needs and intervention goals (e.g., tailor interventions to individual needs or offer stepped care), and (4) improve treatment integrity (e.g., extensive training).

## A CHILD'S PERSPECTIVE

*My pediatrician recommended me to participate in the 'Op Koers' group. She thought it would be nice for me to talk to other children who are also ill, because I actually never talk about it. When I first met the psychologist, I heard that my mother was also going to participate. At first, I didn't like this idea, because my mother already lectures me enough about my medication. But when I heard that the groups would be separate, it was fine with me. There were 6 children in our group, and I was the youngest. The first session was a bit awkward, because we didn't know each other and nobody spoke much. But the second time we started to know each other better. I really liked two older girls in my group. We still have contact with each other, but only via MSN chat, because they live far away. What I liked about the group is that you do all the exercises together and you can really talk with each other about all the things that other children, who are not ill, don't understand. What I learned in the group is that I can still do a lot of nice things in my life, regardless of my illness, and thinking this helps me feel better!*

## A PARENT'S PERSPECTIVE

*As a mother, I first was reluctant to participate in the 'Op Koers' intervention, because I have two younger children and finding a sitter is difficult after school time. But because I believed it would be a good idea to engage in something with my adolescent daughter, and to share experiences with other parents I participated anyway. In the intervention, I found myself surrounded by a group of very nice parents (mostly mothers), and two competent and understanding psychologists. I really gained a lot by exchanging experiences and learning from other parents who sometimes were further along in the process of accepting the consequences of their child's illness for their families. I learned to help my daughter to think more positive about her life in general, and to assist her in making choices about the treatment regime. It also made me stand less alone and feel less incompetent when I recognized certain feelings of anxiety ('Am I a good enough parent for all my children?'), worrying about the future ('Will my daughter make it on her own as an adult?'), and the difficulty of letting my child grow up and do things without my constant guiding.*

## REFLECTION ON MAIN FINDINGS

Throughout this thesis, several efforts were made to expand existing understanding on the role of coping in adjustment to pediatric chronic illness. Stress and coping theory suggests that the stressors faced by children and parents dealing with pediatric chronic illness are multifaceted (e.g. uncertainty, restriction of activities, responsibility of treatment) and that the links between illness related stressors and child adjustment are moderated or mediated by children's coping strategies (Wallander & Varni, 1998; Compas, 2012). Considerable evidence across chronic childhood illnesses suggests that the use of disengaged coping, including cognitive and behavioral avoidance, is generally related to poor adjustment, and that engaged coping, including reappraisal, positive thinking, acceptance, or distraction, is associated to better adjustment (Compas et al., 2006; Spirito et al., 1994; Grootenhuis, Maurice-Stam, Derkx, & Last, 2009). In addition, when confronted with a chronic illness, the whole family faces significant stressors, and coping with these stressors is demands mutual efforts of both children and parents. Therefore, distress in parents can pose risk to the way children cope with chronic illness, and, in turn, parental support and guidance can help children to cope better with illness related stressors (Compas et al., 2010). This theory and research formed the foundation for the 'Op Koers' intervention aimed to improve adjustment



through teaching children active disease related coping skills, and through teaching parents how to support their child to apply the coping skills in everyday life.

Results from our cross-sectional study confirmed that the use of disengaged coping strategies is associated with more adjustment problems. Child engaged coping was also negatively associated with internalizing problems, but had weaker links. This pattern suggests that it is more important to reduce the use of disengaged coping than to increase the use of engaged coping. In a recent critical review, Compas, Jaser, Dunn, and Rodriquez (2012) discussed evidence pointing to which coping strategies are effective in promoting adjustment to chronic illness. They found that more use of secondary control coping<sup>1</sup> (or accommodative coping) and less use of disengagement coping were associated to better adjustment, but that primary control coping, such as problem solving, showed mixed associations with adjustment. The scale we used to measure engaged coping consisted of items that represented both primary (problem solving) and secondary control coping (cognitive restructuring). Compas and colleagues (2012) emphasized that primary control strategies may be effective for the more controllable sources of stress, such as problems related to missing school or changes in treatment regimen, but may be ineffective for others, such as feeling different from peers. In our scale that measured engaged coping both primary and secondary control strategies were represented. However, to answer questions on the use of these strategies children had to imagine a specific stressor (not specifically illness related). It is possible that particular strategies are not effective for each particular stressor that can be imagined. For example feeling sad about a negative consequence of the illness (stressor) requires cognitive restructuring (secondary control), while problems with treatment adherence requires problem solving (primary control). With that in mind, one could argue that the effectiveness of coping strategies depends on the match between characteristics of the stressor, and the child's coping responses. This also underscores the importance of measuring specific coping strategies opposed to a combined scale.

Understanding effects of coping strategies was further expanded by the results regarding the main objective of this thesis, which was to investigate the effect of a group intervention that teaches disease related coping skills, 'Op Koers'. In the 'Op Koers' intervention a set of adaptive coping strategies were translated into five intervention goals, and taught using cognitive behavioral techniques. Intervention goals were: information seeking and giving (problem solving, expressing emotions and seeking social support), relaxation during stressful situations (relaxation and distraction), increasing knowledge on self-management and medical compliance (problem solving), enhancement of social competence (emotional expression and seeking social support), and positive thinking (cognitive restructuring). Positive thinking actually touches all previously described intervention goals to a certain extent. Positive thinking refers to teaching children to replace inaccurate or unhelpful thoughts about the illness or about

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<sup>1</sup> Primary control refers to coping efforts that are intended to actively influence stressful situations, and secondary control involves coping aimed adaptation (or accommodation) to the stressor (Rudolph, Dennig, & Weisz, 1995).

themselves. This cognitive restructuring ability enables children to adapt to the negative consequences of the illness on their every day functioning, for example thoughts like ‘my life is useless, because I cannot compete in sports’ are replaced by ‘despite my illness I am able to go to school and have lots of friends’. All of the coping strategies together were proposed to enhance adjustment.

In the efficacy trial we found that the ‘Op Koers’ intervention was effective in reducing adjustment problems. Parental involvement appeared to be particularly important to achieve effects on the long term, and to achieve effects for children at higher risk for the factors that were targeted in the intervention (disengaged coping and lower self-worth). These effects could partly be explained by changes that the intervention effected in three of the five taught coping skills. The other two skills (‘relaxation’ and ‘medical compliance’) could not be investigated because the scales that were used to measure these goals were unreliable. It is therefore not known whether full mediation would have been obtained by including the full set of skills, or that other factors are at work as well.

To conclude, overall findings from this thesis indicate that teaching disease related coping skills promotes adjustment in children with chronic illness. Furthermore, involving parents in interventions aimed at child coping was consistently shown to bolster long term effects. Working elements that were found in this thesis were the enhancement of social competence, positive thinking, and information seeking and giving. However, it is likely that there are other mechanisms of change that were not under investigation (e.g., relaxation, enhancement of medical compliance, group support). Future efforts should continue to search for effective ways to promote adjustment in children with chronic illness.

## STRENGTHS, LIMITATIONS AND DIRECTIONS FOR FUTURE RESEARCH

To our knowledge, this is the first experimentally controlled study that investigated efficacy, moderators and mediators of a group intervention for children with various chronic illnesses and their parents. Strengths of the ‘Op Koers’ intervention include that it is based on well-established theory on stress and coping with chronic illness, that it uses evidence based cognitive behavior techniques to improve coping, and that it can be expanded by including parents. In addition, ‘Op Koers’ is designed for children with all chronic medical illnesses, which makes recruitment easier and participation of children with rare diseases possible. Furthermore, the intervention is described in a detailed manual, and was successfully implemented in nine centers during the RCT, which makes it well suited for implementation in clinical practice.

The randomized controlled trial design strengthened the validity of the findings, in particular because we 1) followed CONSORT guidelines (Schulz et al., 2010), 2)



achieved a relatively large sample size due to multicenter recruitment by pediatric psychologists working in clinical practice, 3) included a relatively long follow-up (one year), 3) used online assessments, 4) used measures of treatment integrity, 5) used multiple methods (questionnaires and observations) and informants, 6) investigated potential mediating and moderating factors in order to investigate the underlying mechanisms of the intervention, and for whom it works best.

Above described findings should be interpreted in the light of several limitations. The first noteworthy limitation concerns the waitlist control group. The effect analyses revealed that behavior problems decreased in all study-groups, including the waitlist control group. Although this can be attributed to spontaneous recovery (Cook & Campbell, 1979), a more likely explanation is that problem behaviors decreased because the majority of the families in our sample (74%) sought alternative psychosocial support. Families that were assigned to the control group were placed on a one year waitlist, however, because of obvious ethical reasons, were not prevented to seek support for their children or themselves from other sources. At each assessment, we documented if the families received other mental health care, and asked what kind of care (open question). From this open questions it was extremely difficult to determine the content and intensity of the received support. Future studies should operationalize this threat to validity of the findings in more detail. Because the sample consisted of non-referred children, and because the preventive nature of the intervention, we had not anticipated such a high need for immediate support. We therefore expected that children could wait a longer period than, for example, referred children with serious psychopathology. However, with hindsight, the waitlist period of one year was probably too long. Future studies that use a waitlist control group should consider this carefully, and should document compensatory help seeking comprehensively.

Second, despite randomization, baseline characteristics were not completely balanced between the three study-groups. Children in the parent-child intervention were significantly older, had higher illness severity, and higher scores on parent-reported internalizing problems compared to the waitlist control group. These baseline differences might be explained by the fact that the parent-child intervention was more frequently performed in academic hospitals than in the other settings, and children in the academic settings had higher baseline problem scores, and higher illness severity. Parent-child interventions were performed less in non-academic hospitals and schools, because in two of the nine centers it was not possible to randomize into all three study-groups due to a shortage of psychologists carrying out the intervention. Although we statistically controlled for baseline levels of problems, a possible floor effect might explain the lower decline of behavior problems in the control group. These baseline differences highlight the importance of a precisely conducted randomization process, with similar randomization procedures in all participating centers.

A third limitation is that moderation and mediation effect sizes were small and, although we had a relatively large sample size and used multilevel modeling, several

models lacked sufficient power. These problems are reported in many moderation and mediation studies (MacKinnon, 2008). In addition, with the many analyses performed, false positives are a possibility. However, given the early stage of research in this field, taking precautions for multiple testing was not a priority. An important task for future research will be to replicate our findings, using larger samples. This would also allow to investigate multiple moderators or mediators at the same time, and sequential models to examine whether mediators affect changes in other, more proximal, outcomes, such as parental encouragement in daily life and self-perception.

Fourth, several instruments that were used had their own specific limitations. Most importantly, the primary outcomes of this study (CBCL and YSR) were non-blind self-reported measures. Although we used multiple informants (parents and children), child-report was only available from adolescents (>11 years), and parent-report could be biased by parents own levels of distress. In addition, because bias can occur in interpreting data concerning physical symptoms (Perrin et al., 1991), we excluded the 'somatic complaints' from the internalizing scale. However, this could also result in biased interpretations of this scale, because in some cases somatic complaints that are not related to the illness could also pose as internalizing symptoms. Although we still believe that the CBCL is the most suitable outcome measure for assessment of behavior problems, future studies should take these limitations into account. Furthermore, the Coping Strategies Inventory (CSI), and the Questionnaire Op Koers (to measure disease related coping skills) both showed low internal consistency on some of the scales, respectively 'engaged coping', and 'relaxation and medical compliance'. Therefore, making assumptions with regard to engaged coping as a risk factor, and relaxation and medical compliance as mediators was not possible. Future efforts should focus on the development and validation of good coping measures, and to provide sufficient norm data. This will allow us to assess, screen and monitor coping with pediatric illness in research and in clinical practice.

Finally, although we believed that we recruited a representative group of children with chronic illness, we had few ethnic or cultural diversity in our sample. In addition, mainly mothers participated in the study, leaving understanding of father involvement untouched. Future research should strive to include more fathers, and more culturally diverse samples. In addition, although this is outside the scope of this thesis, it is known that chronic illness also poses risk for siblings (Barlow & Ellard, 2005). Future efforts should also include intervening on sibling adjustment problems.

To conclude, our study had several strengths that contributed to successfully conducting a RCT in clinical practice. However, conducting an RCT in pediatric practice comes with limitations. Future research should be aware of, and strive to limit bias due to randomization, practical issues, small sample sizes, and non-reliable instruments.





## CLINICAL IMPLICATIONS

The integration of research and practice is an important but elusive goal (Drotar, 2010). Published research in pediatric psychology often fails to focus on clinical significance, or implementation of evidence based interventions (Brown, 2007; Drotar, 2012). Furthermore, most practicing pediatric psychologists are not engaged in research that evaluates the effectiveness of their clinical interventions, which results in a gap between research and practice (Drotar, 2010). In order to contribute to ‘bridging’ this gap, this thesis provides several implications for clinical pediatric practice. The most important implications are listed below.

### Screening

The results of this thesis highlight the importance of screening for family risk factors and adjustment problems in clinical practice. These factors and problems were shown to be relevant as predictors of benefit from intervention, and relevant to matching families to the optimal intervention modality. To systematically assess psychosocial functioning and to monitor children over time Patient Reported Outcomes (PROs) can be used by professionals in pediatric health care. In the past decades there has been a growing interest in these PROs as a tool for the pediatricians to discuss psychosocial issues during medical consultation (Haverman, Engelen, van Rossum, Heymans, & Grootenhuis, 2011). For example, a recent study investigating the effectiveness of the use of Health Related Quality of life (HRQoL) assessments for children with Juvenile Idiopathic Arthritis in clinical practice, showed that providing information to pediatricians on a child’s HRQoL, leads to significantly more discussion of emotional and social functioning during consultation and improves the child’s, parent’s, and pediatrician’s satisfaction with the provided care (Haverman et al., 2012). When pediatric psychologists and pediatricians work together, PROs can be used to closely monitor children with chronic illnesses in a multidisciplinary context and referral to psychosocial interventions can be better facilitated (Haverman et al., 2012). Next to generally monitoring psychosocial functioning of the child, PROs can also be used to screen for risk within the family. Such family screening tools are scarce in pediatric psychology. Recently, Patel and colleagues (2011) and Kazak and colleagues (2011) developed the Psychosocial Assessment Tool (PAT), which is a brief parent-report screening tool of psychosocial risk (e.g. financial resources, social support, stress reactions, child problems, and sibling problems) in families that cope with pediatric illness (Patel et al., 2011; Kazak et al., 2011). The PAT is described as a promising screening tool in families of children with cancer (Kazak et al., 2012), kidney transplantation (Pai et al., 2008), and sickle cell disease (Karlson et al., 2012). The use of psychosocial screening instruments in clinical practice can help to identify families at risk, and to match the level of risk to appropriate interventions.

## Match risk to interventions

To match risk levels to interventions Kazak (2006) introduced the Pediatric Psychosocial Preventative Health Model. This pyramid-shaped model defines three health care levels. When families have limited risk factors and are resilient (Universal level), it is suggested to provide general support (e.g. give information). When some risk factors are present, along with moderate levels of distress (Targeted level), it is recommended to monitor distress and provide preventive (group) interventions. When more risk factors are present and distress becomes persistent, clinical treatment from a health specialist (e.g. pediatric psychologist) is necessary (Clinical level) (Kazak, 2006). In earlier research in pediatric oncology, the majority of families (65% to 75%) fall within the universal level of risk, 20% to 25% within the targeted range and 10% in the clinically distressed range (Kazak et al., 2011).

The 'Op Koers' intervention can be classified as an intervention in the targeted level (preventive group intervention). One could argue that only families in which risk factors are present or who report significant adjustment problems should be advised to participate in the 'Op Koers' intervention. However, matching families to interventions according to risk/distress could also disregard possible preventive or resilience-increasing effects in families on the universal level. When non-distressed children learn to cope with problems at an early stage, one could expect that when children do face significant stressors, they are better equipped to cope well. Therefore, a stepped care approach might be more suited. In stepped care models, all patients start with the same intervention, and only those patients who do not respond adequately to this intervention step up to an intervention of higher intensity (Bower & Gilbody, 2005). The assumption of stepped care is that many families benefit from low-intensity treatments and do not need further treatment. In theory, this means that stepped care is the most cost-efficient model of care delivery (van Straten, Seekles, Veer-Tazelaar, Beekman, & Cuijpers, 2010). However, in order to develop better recommendations on the dissemination of stepped care, research that investigates preventive effects and/or cost-effectiveness is needed.

## Implementation and future plans

Findings from this thesis indicated that the 'Op Koers' intervention is feasible for implementation in clinical practice. The practical implications from our findings and recommendations described in Chapter 7 will lead to future plans for implementation. Because there might be considerable interest from hospitals throughout the Netherlands, the next step will be to make 'Op Koers' available for these centers. However, in Chapter 7 it was concluded that implementation is a complex process, in which different and innovative strategies must be used to gain success. Several government agencies in the Netherlands have implementation on their agenda (NJI, ZonMW). They recommend to approximate implementation as a planned process with different phases (dissemination,



adoption, implementation and assurance), and to use strategies that consider the population and the context (Yperen & Veerman, 2008). Therefore, the implementation process of 'Op Koers' should be conducted in light of these recommendations.

In addition, because logistical issues are frequently described in intervention studies, including ours, future efforts should be made to overcome these problems and reach more families. Therefore, 'Op Koers' has to be translated into an online chat intervention for the adolescent (12-18) group. There is increasing interest in the development of online interventions (Cushing & Steele, 2010; van der Zanden, Kramer, Gerrits, & Cuijpers, 2012). A first effort of an online chat intervention for children who are treated for cancer is currently being evaluated in our hospital (Maurice-Stam et al., in preparation). However, research that investigates the effectiveness of online interventions for children is scarce, and therefore future efficacy trials are necessary.

Finally, because our and other studies point in the direction of significant stress in parents of children with chronic illnesses, parental distress should also be screened and monitored in clinical practice, and (online) interventions that focus on distress of parents themselves will need to be developed and studied. All of these and future efforts eventually lead to the ultimate goal of delivering all-round evidence-based care for children with chronic illness and their families.

## CONCLUDING KEY MESSAGES

- Children with chronic illness who apply for psychosocial care have more internalizing behavior problems than their healthy peers
- Use of disengaged coping, low self-worth, parenting stress and insecure parent-child relationships are important risk factors associated with adjustment problems
- Adjustment to chronic illness can be promoted by teaching disease related coping skills in a group intervention using cognitive behavioral techniques
- Disease related coping skills, and especially social competence, are important working elements of a group-intervention aimed to promote adjustment
- Parental involvement in the intervention promotes the benefit of learned coping skills, resulting in better child adjustment on the long term
- Parental involvement in the intervention is especially important for more 'at risk' children
- A secure parent-child relationship seems necessary to provide a secure base from which intervention goals can be achieved
- Illness characteristics are only minimally related to psychosocial adjustment, and not related to intervention outcome
- Future researchers are encouraged to investigate high quality screening instruments, and implementation of interventions in clinical practice

