Efficacy of Psychosocial Group Intervention for Children With Chronic Illness and Their Parents

WHAT’S KNOWN ON THIS SUBJECT: Children with chronic illnesses are at risk for emotional and behavioral problems. Therefore, interventions that focus on coping with the negative consequences of the disease are needed. Evidence-based interventions are limited and often focus on a single diagnosis.

WHAT THIS STUDY ADDS: This study demonstrates the efficacy of a cognitive-behavioral group intervention for children with various chronic illnesses. The findings indicate that the involvement of parents is important to achieve long-term results.

abstract

OBJECTIVE: To investigate the efficacy of a cognitive-behavioral group intervention for children with chronic illnesses and to test the effect of an added parent component.

METHODS: Children (n = 194) and their parents participated in a multicenter randomized clinical trial comparing a child-only intervention and a parent-child intervention to a wait-list 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). Assessments took place at baseline and at 6- and 12-month follow-ups. Intention-to-treat mixed-model analyses were performed to test the difference in change in outcomes.

RESULTS: The intervention had a positive effect on changes in 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. Illness severity and illness type did not moderate the effects. There were no intervention effects on child-reported internalizing problems, parent-reported externalizing problems, relaxation, or medical compliance. Of the families in the wait-list control group, 74% sought alternative psychological support during the intervention period.

CONCLUSIONS: This RCT supports the efficacy of a protocol-based group intervention for children with chronic illnesses and their parents. Adding a parental component to the intervention contributed to the persistence of the effects. Future research should focus on moderating and mediating effects of the intervention. Pediatrics 2013;131:e1196–e1203

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ABBREVIATIONS: CI—chronic illness PRF—Parent Report Form YSR—Youth Self-Report

Ms Scholten conceptualized and designed the study, collected the data from all of the sites, carried out the analyses, and drafted the initial manuscript; Dr Willemen conceptualized and designed the study, carried out the initial analyses, and reviewed and revised the manuscript; Dr Last conceptualized and designed the study and reviewed and revised the manuscript; Dr Maurice-Stam conceptualized and designed the study, critically reviewed the analyses, and reviewed and revised the manuscript; Mrs van Dijk, Ms Ensink, Ms Zandbelt, and Mrs van der Hoop-Mooij coordinated and supervised data collection at 1 of the participating sites and critically reviewed the manuscript; Dr Schuengel conceptualized and designed the study and reviewed and revised the manuscript; Dr Grootenhuis conceptualized and designed the study and reviewed and revised the manuscript; and all authors approved the final manuscript as submitted.

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(Continued on last page)
Children growing up with a chronic illness (CI), such as asthma, diabetes, or arthritis, are at heightened risk for hospitalizations, painful medical procedures, and restriction of activities. Discomfort, pain, and limitations may lead to secondary psychosocial problems and adverse effects on academic functioning and social competence.\textsuperscript{1,2} Interventions have been developed\textsuperscript{3–5} with a strong focus on coping skills, which are important moderators of chronic illness effects.\textsuperscript{4,6} However, few interventions have been stringently evaluated, and the medium- to long-term effects are unknown.\textsuperscript{3–5,7} Furthermore, existing intervention programs have often been targeted at a single diagnosis, even though psychosocial challenges in CI (e.g., fear, sadness) usually overlap to a considerable extent.\textsuperscript{4,8} Generic interventions would also give perspective for children with rare diseases.

A standardized group intervention called Op Koers (“On Track”) was developed for heterogeneous groups of children with CI and was evaluated in a pilot study, with promising results.\textsuperscript{9} This intervention program teaches cognitive-behavioral skills, using methods common in clinical work with children with somatic complaints\textsuperscript{10} and children with behavior and/or anxiety disorders.\textsuperscript{11} There is substantial evidence linking parental support to decreased distress and increased psychosocial adjustment in children with CI;\textsuperscript{12–15} therefore, a parallel parent intervention was developed to enhance the effect of the child-only intervention.\textsuperscript{16} The objective of the current study was to test the efficacy of Op Koers, and the additional effect of the parent component, compared with a wait-list control group. Primary outcomes were parent- and self-reported child psychosocial problems; secondary outcomes were child disease-related coping skills.

**METHODS**

**Study Design**

A multicenter randomized controlled trial was designed in accordance with the Consolidated Standards of Reporting Trials statement. Full details of the study protocol and the content of the intervention were reported by Scholten et al.\textsuperscript{16} Participants within each of the 9 centers were block randomized into the following study groups: (1) the Op Koers child-only intervention, (2) Op Koers with a parallel parent program, and (3) a wait-list control group.

**Participants, Procedure, and Randomization**

Participants were recruited between May 2009 and August 2010 from outpatient clinics of 3 academic hospitals, 4 nonacademic hospitals, and 2 primary schools for children with CI in the Netherlands. Children and parents received an information letter and a reply form from their pediatrician. After a positive reply, parents were phoned to assess eligibility and to obtain informed consent. Eligibility criteria included age between 8 and 18 years and having a CI according to the definition of van der Lee et al.\textsuperscript{17} Children were excluded if severe learning difficulties or language problems limited their ability to independently fill out the questionnaires. Participants were computer allocated by an independent researcher, in blocks of 12 to 24 children within each center.\textsuperscript{18} In 2 of the 9 centers, it was not possible to randomize into all 3 study-groups because of a shortage of psychologists carrying out the intervention. Therefore, in those centers, participants were randomized into child-only and wait-list group. Variation between centers in study-groups to which randomization took place might introduce potential confounders. However, we controlled for possible center effects in our data-analyses. Assessments occurred online at baseline (T0), 6 months (T1), and 12 months (T2). Interventions were organized at 3 time points and started within 1 month after baseline. Participants in the wait-list control group were invited to take part in the intervention after the final follow-up assessment at 12 months.

**Interventions**

Children in both intervention groups received the same group course consisting of 6 weekly 90-minute sessions, and a booster session after 6 months, with 4 to 8 participants per group (M = 5.04, SD = 0.89). Two qualified psychologists carried out the intervention, based on a detailed manual. All psychologists (n = 35) were extensively trained in the protocol. Five coping strategies were taught: (1) seeking and giving information about the disease, (2) using relaxation techniques during stressful situations, (3) increasing knowledge of self-management and medical compliance, (4) improving social competence, and (5) positive thinking.\textsuperscript{9,10} Two slightly different versions of the protocol were developed, 1 for children at primary school age (8–12 years old) and another for adolescents (12–18 years old).

The parent component consisted of six 90-minute sessions parallel to the child sessions, also led by 2 psychologists and based on a detailed protocol. Parents were reinforced in sensitively attending to their child’s needs and in encouraging their child to use the taught coping strategies.\textsuperscript{16} All sessions took place in the hospital or school where the child was recruited.

**Measures**

Psychosocial functioning was assessed with the Child Behavior Checklist (Dutch version; Parent Report Form [PRF] 4–18 years and Youth Self-Report [YSR] 11–18 years).\textsuperscript{19,20} Questionnaires consisted of 120 (PRF) and 119 (YSR) problem items, and a 5-point Likert scale (from 0 indicating “not true” to 2 indicating “very true or often true,” considering behavior}
during the past 6 months). We used the 2 broadband scales: Internalizing Problems and Externalizing Problems. The Internalizing Problems scale (range: 0–42) included the subscales “Anxious/Depressed” and “Withdrawn/Depressed.” Items from the subscale “Somatic Complaints” were disregarded. The Externalizing Problems scale (PRF range: 0–70, YSR range: 0–64) included the subscales “Rule-breaking Behavior” and “Aggressive Behavior.” Higher scores indicated more problems. Cronbach’s alphas for the PRF and YSR internalizing and externalizing scales ranged from 0.84–0.91, which were satisfactory and comparable with those reported for the full version. Dutch community norm scores are available. To indicate the percentages of children scoring within the subclinical and clinical range, T scores were computed from the raw scale score. A T score of 63 (90th percentile in the norm population) demarcates the clinical range, which is an indication that a child has clinically relevant symptoms and needs professional treatment.

Disease-related coping skills were assessed with the 26-item Questionnaire Op Koers for children. On a 4-point scale, children were asked if they agreed with statements concerning the 5 learning goals (eg, “I know how to get answers to questions concerning my disease” or “When I am nervous about going to the hospital, I am able to relax myself”). Higher scores reflected better use of skills. Cronbach’s alphas of the scales were moderate to good, ranging from 0.55 (medical compliance) to 0.88.

Illness characteristics (illness type, duration and severity) were collected at baseline. Parents rated illness severity using a proxy measure based on the occurrence of the following 13 possible consequences of chronic illness in the past year (0 = no, 1 = yes, scale 0–13): doctor visits, hospitalization, surgery, use of medication, dietary consequences, visible malformations, use of appliances, diet limitations, exercise, hearing, vision and speech, and course of the disease (0 = improving, 0 = stable, 1 = deteriorating, 1 = unstable). Background and control variables were also obtained from parents. Family composition and socioeconomic status (income), and family members’ ages, genders, and ethnicities were recorded. The setting for the intervention was also recorded (academic hospital, non-academic hospital, or school), as well as stressful life events and the use of psychological care besides Op Koers.

Treatment integrity was observed and rated by trained psychology students (n = 6), using videotapes of randomly selected sessions. For each separate group exercise within the session, scores were assigned for using the course materials (scale: 0–1) and for using the explanations from the protocol (scale: 1–5). Both aspects were summed and averaged across all parts of the session (range: 1–6). For each group and session, psychologists reported on whether they had followed the manual (yes/no) and on attendance of participants. Scores were summarized across all sessions (range: 0–7).

Statistical Analyses

Power calculations indicated that to detect differences of medium effect size (d = 0.5) between study groups over time at a significance level of .05, a minimum of 42 participants had to be enrolled per study group to achieve adequate power (0.80). Preliminary analyses examined participant flow and baseline differences between groups on all study variables. The longitudinal design of this study, together with the nested data structure of children within intervention groups and centers (hospital/school), required multilevel analysis. The multilevel model accounted for dependency among assessment occasions (level 1), intervention groups (level 2), and centers (level 3). To indicate the dependency within each level, intraclass correlations were calculated separately for each dependent variable. Levels were rejected from further analyses if the intraclass correlation coefficient was not significant (or < .05). Intention-to-treat analyses were performed based on the random allocation, using the mixed-model procedure in SPSS (17.0) with full maximum likelihood estimation. Dependent variables were psychosocial problems (PRF and YSR Internalizing and Externalizing scales) and disease-related coping skills. In Step 1, an empty model with the appropriate number of levels and an unrestricted within-subjects (covariance) structure were fitted to the data. In Step 2, “measurement occasion” (time) was added as a random effect. We expected a stronger decrease in psychosocial problems during the intervention period than during the follow-up period; therefore, a quadratic effect of time was added to the linear effect of time for primary outcomes. In Step 3, all background and control variables (see Measures) that differed between study groups were added as fixed effects and were subsequently removed if not found to be significantly related to the level or a change in the dependent variables. In Step 4, study group and the interaction between group and time were entered into the model. This procedure was carried out twice, to test whether change in the outcome variable was different for (1) the child-only and the parent-child intervention groups compared with the wait list as a reference category and (2) the child-only intervention compared with the parent-child intervention as a reference category. In Step 5, the interaction between study group, time, and illness severity was added to test a possible moderation effect of illness severity. The same was done for illness type (7 diagnosis groups, see Table 1). After each step, a deviance
statistic was computed. If the model fit improved, the multivariate statistics of the predictors were interpreted. An $\alpha$ of .05 was used to test the statistical significance of the effects.

**RESULTS**

**Sample Characteristics**

Figure 1 shows participant flow from recruitment to follow-up. Of those given the opportunity to participate, 19% applied. This percentage was expected, considering reported mental health care use of children with CI, combined with the obligations of participating in a RCT. Of the applicants, 98% provided informed consent, and 89% completed baseline assessments. Subject retention to study completion was 82%. There were no significant differences in age, gender, ethnicity, setting, illness severity, or baseline outcome scores between the families retained and those who withdrew; however, withdrawal was significantly lower in the parent-child intervention group (8%; $\chi^2[2] = 6.55$, $P = .038$) than in the child-only intervention (21%; Exp $B = 0.24$; 95% confidence interval 0.07–0.89) and wait-list (20%; Exp $B = 0.26$; 95% confidence interval 0.07–0.94) groups. No differences in attendance were observed between the 2 intervention groups, and no study-related adverse events were reported. Seventy-six participants were assigned to sessions for primary school age children (M age = 10.34, SD = 1.31 range: 7.98–13.03), and 44 participants were assigned to the sessions for adolescents (M age = 15.17, SD = 1.59, range: 11.96–18.07).

During the intervention period, 74% of the children in the wait-list group received alternative psychosocial care, mostly individual psychological treatment. During the follow-up period, 29% in the child-intervention, 28% in the parent-child intervention, and 18% in the wait-list control group received additional alternative psychological care; these percentages did not differ significantly ($P = .33$).

Despite randomization, baseline characteristics were not completely balanced between study groups (Table 1). Children in the parent-child intervention were significantly older, were more frequently of Dutch origin, and had higher illness severity and higher scores on internalizing problems (PRF, $P < .05$) compared with the wait-list control group. Parent-child intervention was more frequently performed in academic hospitals than in the other settings.

The average treatment integrity did not differ between child-only (psychologist: M = 0.89, SD = 0.14; range: 0.57–1.00; observed: M = 5.44, SD = 0.50; range: 4.04–6.00) and parent-child (psychologist: M = 0.83, SD = 0.17; range: 0.50–1.00; observed: M = 5.47, SD = 0.59; range: 4.27–6.00) interventions, and intraclass correlations indicated no significant group or center effects. Therefore, we only accounted for dependency within children in the multilevel analyses.

**Primary Outcomes**

The mean outcome scores and SDs by study group at each 6-month interval are presented in Table 2 and graphically shown in Fig 2. To give an indication of the severity of the problems, percentages of internalizing and externalizing problems within subclinical or clinical range were also reported in Table 2. In addition, Dutch community mean norm scores are included in Fig 2 as a straight line.

**Internalizing Problems (PRF)**

Regardless of the study-group, parent-reported internalizing problems (Fig 2A) decreased linearly over time ($t_{1,320} = 7.92$, $P = .005$, $B = −3.67$, $P = .005$, $d = 0.31$). The quadratic term was significant and positive, indicating that the rate of decrease diminished over time.
The intervention had a significant positive effect ($\chi^2 [6] = 13, P = .043$) and was significantly related to the change in internalizing problems over time ($F = 4.09, P = .018$). In the child-only intervention group, internalizing problems decreased more than in the wait-list group ($B = -8.13, P = .008, d = .30$). However, in the child-only intervention, internalizing problems increased again from T1 to T2 ($B = 1.75, P = .034, d = .24$), whereas in the parent-child and wait-list groups internalizing problems continued to decrease, showing a significant interaction of time (quadratic) with study-group ($F = 3.75, P = .025$). Academic setting (vs nonacademic hospital, school) and illness severity were significantly positively related to the level of parent-reported internalizing problems. The moderation effects of illness severity ($\chi^2 [3] = 2.28, P = .44$) and illness type ($\chi^2 [60] = 60.72, P = .45$) did not significantly add to the model fit.

**Externalizing Problems (PRF)**
Regardless of the study-group, parent-reported externalizing problems (Fig 2B) decreased significantly over time ($B = -0.92, P = .000, d = .61$). The intervention had no significant effect on the change in parent-reported externalizing problems ($\chi^2 [3] = 2.28, P = .44$). Illness severity was positively associated with the level of parent-reported externalizing problems.

**Internalizing Problems (YSR)**
Regardless of the study-group, child-reported externalizing problems (Fig 2C) decreased significantly over time ($B = -1.35, P < .001, d = .95$). The intervention had no significant effect on the change of child-reported internalizing problems.
problems ($x^2 [3] = 1.23, P = .75$). Age and illness severity were significantly positively associated with the level of child-reported internalizing problems.

**Externalizing Problems (YSR)**

Regardless of the study-group, child-reported externalizing problems (Fig 2D) decreased significantly over time ($B = 20.765, P = .001, d = 0.56$). The intervention had a significant positive effect ($x^2 [4] = 10, P = .04$). Decrease of externalizing problems was stronger after participation in the parent-child intervention than in the child-only intervention ($B = 1.21, P = .027, d = 0.42$). No control variables were significantly associated with the level of externalizing problems. The interactions with illness severity ($x^2 [2] = 0.27, P = .874$) and illness type ($x^2 [12] = 9.65, P = .62$) did not significantly add to the model fit.

**Secondary Outcomes**

Regardless of the study group, all disease-related coping skills improved significantly over time: information seeking ($B = 0.80, P < .001, d = 0.88$), relaxation
(B = 0.68, P = .001, d = 0.34), social competences (B = 0.42, P = .001, d = 0.52), positive thinking (B = 0.88, P < .000, d = 0.50), and medical compliance (B = 0.67, P < .000, d = 0.74). The intervention had a significant positive effect on the changes of coping skills for information seeking, social competence, and positive thinking. The increases in information seeking (B = 0.69, P = .046, d = 0.30) and in social competence (B = 0.62, P = .048, d = 0.31) were stronger in the parent-child intervention compared with the wait-list group. The increase in positive thinking was stronger in the child-only intervention compared with the wait-list group (B = 0.90, P = .045, d = 0.22). No control variables were significantly associated with the skill levels.

**DISCUSSION**

The current study found evidence for the efficacy of Op Koers (a group intervention for children with various chronic diseases) and for the added value of involving their parents in this intervention. The interventions had a positive effect on parent-reported internalizing problems, child-reported externalizing problems, and disease-related coping skills (information seeking, social competence, and positive thinking). The additional effect of parental involvement was observed on parent-reported internalizing problems, child-reported externalizing problems, and disease-related coping skills (information seeking and social competence). These findings were expected, because parental support is considered necessary to successfully apply adaptive coping skills in everyday life. Effects sizes were small to moderate, which is in line with earlier studies on efficacy of psychosocial interventions for children with CI. Small effect size and lack of intervention effects on some outcomes may be because 74% of the families in the wait-list control group sought alternative psychological support during the intervention period. Although 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 CI.

Unexpectedly, we found no effect of the child-only intervention on self-reported externalizing problems, despite substantial improvement in the parent-child intervention. One possible explanation is that externalizing behavior is to a large extent determined by parenting practices and parental support. However, it is important to consider that in our sample mean self-reported externalizing problem levels were below the community norm (Fig 2D), thus the lack of change in these particular problems may not necessarily be worrisome. Although all disease-related coping skills improved and Op Koers showed positive effects on information seeking, social competence, and positive thinking, we found no effect of the interventions on the use of relaxation and knowledge of medical compliance. For medical compliance, this may be explained by the moderate internal consistency of the scale. For relaxation, it is possible that children already used adequate relaxation methods (eg, techniques taught by their pediatrician or nurse practitioner) to cope with stressful or painful medical situations. Future efforts should elaborate more extensively on these coping skills.

The current study is the first multisite randomized controlled trial of a cognitive-behavioral group intervention for children with various medical diagnoses. Strengths include recruitment from 9 centers across the country, use of a protocol-based intervention that involved parents, and an intervention protocol well suited for implementation in clinical practice. Limitations include reliance on nonblind self-report outcome measurements and compensatory help seeking by the control group. In addition, 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. Illness parameters did not appear to influence the effect; however, it is unknown which elements of the intervention were effective and whether the intervention was equally effective for all types of children with CI. Future empirical efforts should focus on moderating and mediating effects of the intervention to determine what works for whom (eg, based on age, gender, baseline levels of risk and protective factors), and on studying effectiveness and implementation outside the scope of a controlled trial.

**CONCLUSIONS**

This study supports the efficacy of a protocol-based, group intervention for children with CI and their parents. Effects persisted over time, which can mainly be attributed to the involvement of parents in the intervention. Providing access to Op Koers multidisciplinary care for children with CI will likely prevent the development of emotional and behavioral problems secondary to the CI.

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REFERENCES


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