Abstract

Objective: To quantify the effects of parent- and family-based psychological therapies for youth with common chronic medical conditions on parent and family outcomes (primary aim) and child outcomes (secondary aim).

Methods: MEDLINE, EMBASE, and PsycINFO were searched from inception to April 2013. Thirty-seven randomized controlled trials were included. Quality of the evidence was evaluated using GRADE criteria. Data were extracted on parent, family, and child outcomes.

Results: Pooled psychological therapies had a positive effect on parent behavior at post-treatment and follow-up; no significant improvement was observed for other outcome domains. Problem solving therapy (PST) improved parent mental health and parent behavior at post-treatment and follow-up. There was insufficient evidence to evaluate cognitive-behavioral and systems therapies for many outcome domains.

Conclusions: Parent and family-based psychological therapies can improve parent outcomes, with PST emerging as particularly promising. Future research should incorporate consensus statements for outcomes assessment, multi-site recruitment, and active comparator conditions.

Keywords: Psychological therapies, randomized controlled trials, meta-analysis, systematic review, parent, family, children, chronic illness, asthma, cystic fibrosis, diabetes mellitus, chronic pain, cancer, traumatic brain injury, epilepsy, spina bifida, cardiovascular disease, solid organ transplant.

Systematic Review and Meta-analysis: Parent and Family-Based Interventions for Children and Adolescents with Chronic Medical Conditions

Introduction

Medical advances in the past two decades have resulted in an increase in the prevalence of pediatric chronic medical illness as many children in developed nations are surviving or living longer with conditions such as cancer, cystic fibrosis, and sickle cell disease (Perrin, Bloom, & Gortmaker, 2007). Pediatric chronic illness has a negative impact on child, parent and family functioning. Parents of children with chronic medical conditions commonly report increased parenting stress, anxiety and depressive symptoms, financial strain, and family conflict (Cousino & Hazen, 2013; Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004; Logan & Scharff, 2005; Palermo, Putnam, Armstrong, & Daily, 2007; Quittner et al., 1998). Parents play a critical role in their child's ability to adapt to living with a chronic illness, both in terms of their child's emotional functioning as well as their child's ability to participate in activities of daily life. In particular, parent psychological distress is recognized as a risk factor for poorer outcomes in youth with a variety of chronic medical conditions such as cystic fibrosis (Cappelli, McGrath, MacDonald, Katsanis, & Lascelles, 1989), cancer (Robinson, Gerhardt, Vannatta, & Noll, 2007), spina bifida (Friedman, et al., 2004), and chronic pain (Logan & Scharff, 2005; Palermo, et al., 2007). Parents have significant potential to positively or negatively impact their child's adjustment to chronic illness.

Theoretical Model

The behavioral family systems theoretical model provides an over-arching framework for family-based psychological interventions that integrates cognitive-behavioral, problem solving, and systems approaches (Robin & Foster, 1998). Based on this theoretical model, child, parent and family adjustment to pediatric chronic illness may be influenced by several factors, including: family members' maladaptive thoughts, feelings, and behaviors; family members' ability to solve problems and communicate effectively; and patterns of interactions between family members, as well as between children, parents and broader community systems such as school and the hospital.

Existing Psychological Interventions

Existing interventions for parents and families of youth with chronic illness that fall under the behavioral family systems theoretical model include cognitive-behavioral (e.g., Palermo, Wilson, Peters, Lewandowski, & Somhegyi, 2009), problem solving (e.g., Sahler et al., 2002), and systemic treatments (e.g., Ellis et al., 2005; Wysocki et al., 2007). Cognitive-behavioral therapy (CBT) includes a range of strategies with the goals of modifying social/environmental and behavioral factors that may exacerbate or cause symptoms, and modifying maladaptive thoughts, feelings, and behaviors to reduce symptoms and prevent relapse (see Beck, 2011; Kendall, 2011). Problem solving therapy (PST) includes didactic instruction in the cognitions and behaviors required to effectively solve problems (i.e., problem solving skills), followed by modeling, behavioral rehearsal, and performance feedback (see D'Zurilla & Goldfried, 1971; Nezu (2005). Systemic therapies (ST) emphasize the role of the family and broader social context in an individual's emotional functioning and adjustment, and interventions focus on altering patterns of interactions between family members and collaborating with broader systems such as the patient's school, work, or medical team. Thus,

psychological interventions that fall under the behavioral family systems theoretical model include behavioral family systems therapy in addition to cognitive-behavioral therapy, problem-solving therapy and systems therapies such as behavioral family systems therapy, family therapy and multisystemic therapy (see Cotrell & Boston, 2002; Kazak, Simms, & Rourke, 2002).

Previous Meta-Analytic Reviews

Despite increasing appreciation for the importance of involving parents and family members in treatment, existing meta-analytic reviews of psychological interventions for children with chronic medical conditions often fail to report parent and family functioning as treatment outcomes (e.g., Astin, Beckner, Soeken, Hochberg, & Berman, 2002; Beale, 2006; Kahana, Drotar, & Frazier, 2008; Kibby, Tyc, & Mulhern, 1998; Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010); We are aware of only two meta-analyses that have considered this issue. In a meta-analysis evaluating the efficacy of psychological interventions for children with cancer and their parents, Pai, Drotar, Zebracki, Moore, & Youngstrom (2006) found that psychological interventions significantly reduced parental distress and improved parental adjustment but had no effects for child outcomes. Recently, we conducted a meta-analysis for the Cochrane Collaboration evaluating psychological interventions for parents and families of youth with asthma, cancer, chronic pain, diabetes, gynaecological disorders, inflammatory bowel diseases, skin diseases, and traumatic brain injury (Eccleston, Palermo, Fisher & Law, 2012). Due to lack of available studies, data analyses were carried out on a subset of these illnesses (i.e., asthma, cancer, chronic pain, diabetes, skin diseases, and traumatic brain injury). Results indicated that across illness groups, only problem-solving therapy demonstrated a positive effect on parental mental health and behavior; no such effects were found for cognitive-behavioral therapy, family therapy, or multisystemic therapy.

Together, these findings suggest that not all psychological interventions impact parental mental health and behavior; however, it is unclear why this might be the case. The meta-analytic reviews conducted by Pai et al. (2006) and Eccleston et al. (2012) included studies of psychological interventions that had a wide range of parent involvement, from interventions that primarily targeted children with only minimal parent participation to interventions that primarily targeted parents and had no child participation. It is possible that the efficacy of these interventions may depend upon whether the parent (rather than the child) is the primary treatment target. Research is also needed to evaluate the efficacy of psychological interventions for parents and families of youth with other chronic medical conditions that are commonly encountered by pediatric psychologists (e.g., cystic fibrosis, epilepsy, spina bifida, solid organ transplant).

Unique Contributions of the Current Review

The current systematic review and meta-analysis is similar to our previous Cochrane review on this topic (Eccleston et al., 2012), but differs in the following ways:

- First, we have broadened the scope of illnesses that were previously considered by searching for all of the chronic medical conditions that are reviewed in the Handbook of Pediatric Psychology, 4th edition (Roberts & Steele, 2010). These include asthma, cancer (patients in active treatment and survivors), cardiovascular diseases, cystic fibrosis, diabetes mellitus, epilepsy, painful conditions (i.e., sickle cell disease, chronic pain, fibromyalgia, juvenile rheumatoid arthritis, irritable bowel syndrome, irritable bowel disease), spina bifida, solid organ transplant, and traumatic brain injury. In particular, cardiovascular diseases, epilepsy, spina bifida, and solid organ

transplant have not been included in previous meta-analytic reviews of parent and family-based interventions for youth with chronic illness.

- Second, we have standardized the amount of treatment delivered to parents across included trials. Specifically, to be included in this review, parents had to be identified by the authors as a primary intervention target and treatment delivered to parents had to equal at least 50% of the child's treatment duration.

Aims

The primary aim of this review is to evaluate the efficacy of parent and family-based psychological interventions in improving parent mental health, behavior and family functioning among parents and families of children with chronic medical illness. A secondary aim of this review is to evaluate the efficacy of parent and family-based psychological interventions in improving mental health, behavior/disability and medical symptoms of children with chronic medical illness. An exploratory aim of this review is to examine the efficacy of parent and family-based psychological interventions based on therapy type (i.e., CBT, PST, or ST).

Method

Study Design

Only randomized controlled trials published in peer-reviewed journals were included in this systematic review. All included trials had a primary aim to evaluate a psychological intervention that directly targeted parents and families of youth with a chronic medical condition. A minimum sample size of 10 in the treatment and control arms at each data extraction point was also required to meet the inclusion criteria. Studies not written in English were excluded.

Types of Participants

Participants were parents of children and adolescents (ages 0 to 18) with one of the following chronic medical conditions: asthma, cancer (patients in active treatment and survivors), cardiovascular diseases, cystic fibrosis, diabetes mellitus, epilepsy, painful conditions (i.e., sickle cell disease, chronic pain, fibromyalgia, juvenile rheumatoid arthritis, irritable bowel syndrome, irritable bowel disease), spina bifida, solid organ transplant, and traumatic brain injury. Trials with more than one illness group that reported aggregated data were only included if all of the illness groups were on the above list.

Since most randomized controlled trials of behavioral interventions in pediatric psychology do not report specific details on family structure, we chose not to operationally define the term "family" or "parent" and instead relied on inclusion of the following terms in the description of the target population: parent, mother, father, caregiver, family (see Appendix A in the online supplementary materials for more specific details on the search terms used).

Types of Interventions

Only studies that included a psychological therapy delivered as an intervention were included in this review. A psychological intervention was defined as an intervention that was a) designed to change thoughts and/or behaviors of parents and/or family members, with the goal of improving parent and/or child outcomes, and b) incorporated psychological methods subsumed under the behavioral family systems theoretical model, including cognitive behavioral, problem solving, and/or systems approaches. Included interventions met the following criteria: 1) A primary aim of the intervention was to change thoughts, behaviors or psychological well-being of parents or families, and 2) Treatment duration (e.g., number of sessions) for parents equalled

at least 50% of the child's treatment duration. Comparator conditions included treatment as usual, attention control, or wait-list control.

Types of Outcomes

Parent and family outcomes were the primary target of this review paper; child outcomes were a secondary target. Outcome domains included: parent mental health, parent behavior, family functioning, child mental health, child behavior/disability, and child medical symptoms. When multiple measures were used to assess the same outcome domain, we extracted the measure that was indicated as primary by the authors. If the authors did not indicate a primary outcome measure, we selected the most generic, reliable, and frequently used measure within the field. We consulted the *Journal of Pediatric Psychology* evidence-based assessment special issue to aide in this decision making (Drotar, 2008). Where both parents and children reported on an outcome domain, we extracted the self-report item. For family functioning measures, we extracted the parent-report item. Multiple manuscripts reporting outcomes from the same sample were combined and treated as one trial. Qualitative outcome measures were excluded. Data were extracted at post-treatment (immediately following completion of intervention) and follow-up. Follow-up was defined as between three and 12 months following post-treatment. If there were two time points or more within this year, the longer of the two was extracted.

Search Methods for Identification of Studies

Three databases were searched for this review: MEDLINE, EMBASE, and PsycINFO. The search strategy was conducted from the conception of these databases through April 2013. For the exact search strategies used, please see Appendix A in the online supplementary materials. We also searched other resources including reference lists of included studies,

reference lists of relevant book chapters, and relevant reviews that were found in our initial search. We contacted authors of included studies, experts in the field, and authors of relevant abstracts from conference proceedings to identify any further studies that were not found in the initial search.

Data Extraction and Management

One review author performed the searches of each database and collated the results. Four review authors sorted abstracts, identified those eligible to be included, and read the manuscripts of eligible abstracts in full. A fifth author adjudicated any disagreements. Four authors carried out data extraction for studies that were identified as appropriate for inclusion. Disagreements regarding extracted data were arbitrated by a fifth author. An adapted data extraction sheet from Eccleston et al. (2012) was used, and included sample demographics, characteristics of the intervention and comparator(s), outcome measures, and outcome data. Following data extraction, authors of studies with incomplete data reporting were contacted to obtain the missing data.

Assessment of Risk of Bias in Included Studies

Risk of bias was assessed by four authors using the Cochrane risk of bias tool (Higgins et al., 2011), which evaluates selection bias, detection bias, attrition bias, and reporting bias. We eliminated the item assessing blinding of participants and personnel as it is not possible to blind therapists or participants receiving therapy, and is therefore redundant in psychological trials included in this review.

Quality of Evidence

Quality of evidence was assessed using the GRADE criteria (Guyatt et al., 2013). Each analysis was judged on risk of bias, inconsistency of evidence, indirectness of results, imprecision of evidence, and publication bias. Per the guidelines in Balshem et al. (2011), a four-tiered quality rating is given, ranging from 'high' to 'very low'. High quality ratings indicate that further research is very unlikely to change our confidence in the estimate of effect. Moderate quality ratings indicate that further research is likely to have an impact on our confidence in the estimate of effect. Low quality ratings indicate that further research is very likely to have an impact on our confidence in the estimate of effect. Finally, very low quality ratings indicate that we are very uncertain about the estimate of effect.

Data Analytic Approach

Data analyses were conducted in RevMan 5.1. For the purpose of this review, all extracted outcome data were continuous. Random effects models were used for all meta-analyses. This approach allows for weighting of each trial, and provides a mean difference score (treatment vs. comparator) and confidence interval (CI) that represents all of the trials included in a given analysis. Standardized mean difference (SMD) scores (rather than raw mean scores) were used in all meta-analyses to account for heterogeneity among extracted measures.

Results

Characteristics of Included Studies

Our search produced a total of 1,312 papers, of which 181 were read in full and 37 met inclusion criteria (see PRISMA Flow Diagram in Figure 1 for details). Of the 37 included studies, 18 used CBT, nine used PST, and 10 used ST. Eleven of the 37 studies are new to this review and were not included our previous Cochrane review on this topic (Eccleston, et al.,

2012). Six studies enrolled children with asthma, seven studies enrolled children with cancer, one study enrolled children with congenital heart disease, two studies enrolled children with cystic fibrosis, 11 studies enrolled children with diabetes, seven studies enrolled children with painful conditions, three studies enrolled children with traumatic brain injury, and. There were no studies that investigated children with epilepsy, spina bifida, or solid organ transplant. The comparison groups also varied. Eighteen studies used a "treatment as usual" comparison, six studies used a waitlist control comparison, nine studies used an active comparison group, three studies used both an active comparison group and a treatment as usual control group (three arm studies), and one study did not identify what type of comparison was used.

Insert Figure 1 here.

The mean number of parents entering treatment was 132 per study, (*M* age = 37.02 years, *SD* = 6.55). More mothers entered into treatment compared to fathers (Average $N_{Mothers}$ = 141/study, Average $N_{Fathers}$ = 13/study). The average number of children entering treatment was 120 per study (*M* age = 9.44, *SD* = 2.45; *Range* = 0-18 years). A similar number of boys and girls entered into treatment (M_{Boys} = 57, M_{Girls} = 55). A variety of settings were used to carry out the interventions. Of the 37 studies, 23 described the treatment setting; eight were conducted in office-based settings, 11 were conducted in patients' homes, and four used both office and home settings to conduct the intervention. Table 1 provides a brief summary of study characteristics. Appendix B in the online supplementary material provides detailed study characteristics including participant demographics, intervention characteristics, and outcome measures.

Insert Table 1 here.

Risk of Bias

Risk of bias was assessed according to the Cochrane Handbook risk of bias tool (Higgins, et al., 2011), including: 1) Random sequence generation (selection bias); 2) Allocation concealment (selection bias); 3) Blinding of outcome assessment (detection bias), 4) Incomplete outcome data (attrition bias); and 5) Selective reporting (reporting bias).

For random sequence generation, authors had to report a satisfactory method of randomization to be judged as low risk of bias; 15 studies had a low risk of bias, 22 studies were judged to be unclear, and no studies had high risk of bias.

For allocation concealment, authors had to report that allocation to study group was carried out by a third party to be judged as low risk of bias; 12 studies had a low risk of bias, 22 studies were judged to be unclear, and three studies had high risk of bias.

For blinding of outcome assessment, authors had to report that asssements were conducted by a third party who was blind to treatment allocation to be judged as low risk of bias; 13 studies had low risk of bias, 20 studies were unclear, and in four studies the authors stated that the individual who took assessments knew of the allocation to treatment group and were therefore judged as having a high risk of bias.

For incomplete outcome data, authors had to report attrition and specify that there were no significant differences on pre-treatment variables between completers and non-completers; 13 studies had low risk of bias, 16 studies were judged to be unclear, and eight studies were judged to have high risk of bias because the authors either reported attrition but did not assess differences between completers and non-completers or reported there were significant differences between completers and non-completers.

Selective reporting bias was judged to be low if authors fully reported all outcome data (mean, standard deviation, *N*), unclear if authors did not report outcome data in the published manuscript but responded to our request for these data, and high if authors did not report outcome data in the published manuscript and did not respond to our request for these data; 15 studies had low risk of bias, 10 studies were judged to be unclear, and 12 studies were judged to have a high risk of bias.

For a summary of risk of bias ratings by study, see Figure 2. The Characteristics of Included Studies table in Appendix B of the online supplementary materials provides more detailed information on risk of bias ratings.

Insert Figure 2 here.

Meta-Analysis Results

Data were analysed twice. First, data were pooled across treatment types to determine the effect of all parent- and family-based psychological interventions for youth with a chronic illness at post-treatment and at follow-up. Second, data were analyzed within each treatment type (CBT, PST, or ST) to determine the effect of each treatment type at post-treatment and follow-up. Outcomes included parent mental health, parent behavior, family functioning, child mental health, child behavior/disability, and child medical symptoms.

Missing data. Of those studies which assessed relevant outcome domains, complete outcome data (i.e., sample size, means, standard deviations) were available from the published manuscript in 15 trials (Ellis, et al., 2005; Ellis et al., 2004; Hoekstra-Weebers, Heuvel, Jaspers, Kamps, & Klip, 1998; Laffel et al., 2003; McCusker et al., 2012; Murphy, Wadham, Hassler-Hurst, Rayman, & Skinner, 2012; Nelson et al., 2011; Ng et al., 2008; Palermo, et al., 2009;

Sassmann, de Hair, Danne, & Lange, 2012; Seid, Varni, Gidwani, Gelhard, & Slymen, 2010; Stehl et al., 2009; Wade, Wolfe, Brown & Pestian, 2006; Wade, Carey, & Wolfe, 2006a; Walders et al., 2006). We wrote an average of two emails to 29 authors. Ten authors provided data in response to our requests (Ahari, Younesi, Borjali, & Damavandi, 2012; Ambrosino et al., 2008; Barakat, Schwartz, Salamon, & Radcliffe, 2010; Barry & von Baeyer, 1997; Celano, Holsey, & Kobrynski, 2012; Lehmkuhl et al., 2010; Levy et al., 2010; Sahler et al., 2013; Sahler et al., 2005; Sahler, et al., 2002). Other authors were unable or unwilling to provide additional data or did not respond. Authors who were unwilling to provide additional data stated that the data were available to them but they were too busy to provide it for this review.

Adverse events. Only two trials explicitly stated that no adverse events occured (Nansel, Iannotti, & Liu, 2012; Stark et al., 2005). The presence or absence of adverse events was not described in the remaining 35 trials.

Meta-analysis for pooled psychological interventions. Table 2 provides a summary of the results of the overall meta-analysis for each of the outcomes at two assessment points (post-treatment and follow-up). Appendix C in the online supplementary materials provides forest plots for each of the analyses described below. Appendix D in the online supplementary materials provides ratings on quality of evidence for each analysis using GRADE criteria.

Parent outcomes. Twelve studies including 1079 participants were entered into an analysis to determine the effect on parent mental health at post-treatment, and follow-up data were available from eight studies including 1047 participants. Parent- and family-based psychological interventions did not significantly improve parent mental health post-treatment

(SMD = -0.19, CI -0.43 to 0.04, $z = 1.63$, $p = 0.10$) or at follow-up (SMD = -0.03, CI -0.22 to 0.17, $z = 0.27$, $p = 0.78$)

Five studies including 769 participants were entered into an analysis to determine the effect on parent behavior at post-treatment, and follow-up data were available from three studies including 625 participants. Parent- and family-based psychological interventions had a small but significant effect on parent behavior post-treatment (SMD = -0.25, CI -0.39 to -0.11, $z = 3.44$, $p < .01$; Figure 3) and at follow-up (SMD = -0.21, CI -0.37 to -0.05, $z = 2.64$, $p < .01$; Figure 4).

Insert Figures 3 and 4 here.

Family functioning. Eight studies including 433 participants were entered into an analysis to determine the effect on family functioning post-treatment, and at follow-up data were available from three studies including 170 participants. Parent- and family-based psychological interventions did not significantly improve family functioning post-treatment (SMD = -0.05, CI -0.24 to 0.14, $z = 0.56$, $p = 0.57$) or at follow-up (SMD = -0.22, CI -0.53 to 0.09, $z = 1.42$, $p = 0.16$).

Child outcomes. Five studies including 439 participants were entered into an analysis to determine the effect on child mental health post-treatment. Parent- and family-based psychological interventions did not significantly improve child mental health post-treatment (SMD = 0.00, CI -0.27 to 0.28, $z = 0.02$, $p = 0.98$). Only two studies reported on child mental health at follow-up, therefore this effect was not estimated.

Seven studies including 422 participants were entered into an analysis to determine the effect on child behavior/disability post-treatment and at follow-up data were available from three studies including 244 participants. Parent- and family-based psychological interventions did not

significantly improve child behavior/disability post-treatment (SMD = -0.32, CI -0.74 to 0.10, z = 1.50, p = 0.13) or at follow-up (SMD = -0.20, CI -0.45 to 0.05, z = 1.55, p = 0.12).

Eighteen studies including 1599 participants were entered into an analysis to determine the effect on child medical symptoms post-treatment, and follow-up data were available from nine studies including 1031 participants. Parent- and family-based psychological interventions did not significantly improve child medical symptoms post-treatment (SMD = -0.08, CI -0.19 to 0.04, z = 1.29, p = 0.20) or at follow-up (SMD = -0.03, CI -0.26 to 0.20, z = 0.24, p = 0.81).

Analysis by intervention type. Appendix C in the online supplementary materials provides forest plots for each of the analyses described below. Appendix D in the online supplementary materials provides ratings on quality of evidence for each analysis using GRADE criteria.

Cognitive-behavioral therapy.

Parent outcomes. Five studies including 268 participants were entered into an analysis to determine the effect of CBT on parent mental health post-treatment, and results were not significant (SMD = -0.14, CI -0.71 to 0.44, z = 0.47, p = 0.44). Because fewer than three studies presented data on parent mental health at follow-up and parent-behavior (post-treatment and follow-up), these effects were not estimated.

Family functioning. Three studies including 133 participants were entered into an analysis to determine the effects of CBT on family functioning post-treatment, and results were not significant (SMD = -0.09, CI -0.44 to 0.25, z = 0.53, p = 0.60). Because fewer than three studies presented data on family functioning at follow-up, this effect was not estimated.

Child outcomes. Three studies including 287 participants were entered into an analysis to determine the effect of CBT on child mental health post-treatment, and results were not significant (SMD = 0.18, CI -0.05 to 0.42, $z = 1.52$, $p = 0.13$). Three studies including 243 participants were entered into an analysis to determine the effect of CBT on child behavior/disability post-treatment, and similarly results were not significant (SMD = -0.25, CI -0.73 to 0.24, $z = 1.00$, $p = 0.32$). Fewer than three studies presented data on child mental health and child behavior/disability at follow-up, therefore these effects were not estimated.

Eight studies including 645 participants were entered into an analysis to determine the effect of CBT on child medical symptoms post-treatment, and at follow-up data were available from four studies including 379 participants; however results were not significant post-treatment (SMD = -0.03, CI -0.19 to 0.12, $z = 0.42$, $p = 0.67$) or at follow-up (SMD = 0.07, CI -0.13 to 0.28, $z = 0.70$, $p = 0.48$).

***Problem solving therapy*.**

Parent outcomes. Five studies including 737 participants were entered into an analysis to determine the effectiveness of PST interventions on parent mental health post-treatment, and follow-up data were available from four studies including 690 participants. PST had a small but significant effect on parent mental health post-treatment (SMD = -0.29, CI -0.48 to -0.10, $z = 2.95$, $p = < .01$) and at follow-up (SMD = -0.21, CI -0.36 to -0.06, $z = 2.75$, $p < .01$). Three studies were entered into an analysis to determine the effect on parent behavior post-treatment ($N = 664$) and at follow-up ($N = 625$). PST had a small but significant effect on parent behavior post-treatment (SMD = -0.28, CI -0.43 to -0.13, $z = 3.61$, $p < 0.01$), and at follow-up (SMD = -0.21, CI -0.37 to -0.05, $z = 2.64$, $p < 0.01$).

Family functioning and child outcomes. Fewer than three PST studies presented data on family functioning, child mental health, child behavior/disability, or child medical symptoms at post-treatment or follow-up; therefore, these effects were not estimated.

Systems therapy.

Parent outcomes. Fewer than three ST studies presented data on parent mental health and parent behavior post-treatment and at follow-up; therefore, these effects were not estimated.

Family functioning. Three studies including 233 participants were entered into an analysis to determine the effect on family functioning post-treatment, but results were not significant (SMD = -0.01, CI -.0.27 to 0.25, $z = 0.06$, $p = 0.95$). Fewer than three ST studies presented data on family functioning at follow-up, therefore these effects were not estimated.

Child outcomes. Eight studies including 738 participants were entered into an analysis to determine the effect on child medical symptoms post-treatment, and follow-up data were available from three studies including 391 participants; however, results were not significant post-treatment (SMD = -0.11, CI - 0.30 to 0.07, $z = 1.18$, $p = 0.24$) or at follow-up (SMD = -0.12, CI -0.31 to 0.08, $z = 1.14$, $p = 0.25$). Fewer than three ST studies presented data on child mental health or child behavior/disability post-treatment and at follow-up; therefore these effects were not estimated.

Quality of evidence. GRADE criteria were used to assess quality of evidence for each meta-analysis. Appendix D in the online supplementary materials includes tables with GRADE ratings for each of the following eight analyses: combined therapies (post-treatment, follow-up), CBT (post-treatment, follow-up), PST (post-treatment, follow-up), and ST (post-treatment, follow-up). Of the 48 possible GRADE ratings, only 41 judgements could be made due to lack of

necessary data for some analyses. Of the 41 judgements, two were rated as high quality, 13 were rated as moderate quality, seven were rated as low quality, and 19 were rated as very low quality. Ratings of in the very low quality category were given primarily due to the small number of participants available for inclusion in the analysis.

Meta-analysis evaluating combined psychological therapies received low to moderate GRADE ratings at post-treatment and follow-up (see Table 3 and Table 4). This means that we are somewhat confident about the estimates of these effects but that further research could influence these findings.

Insert Table 3 and Table 4 here.

For CBT, analyses of parent outcome domains and the family functioning domain were rated as very low quality, meaning that we are very uncertain about the estimates of these effects and future research would influence these findings. In contrast, analyses of child outcome domains for CBT were rated as low to moderate quality, meaning that we have more confidence in the estimates of these effects but further research is still likely to have an important impact on these findings. Low quality ratings for analyses of outcomes from CBT trials were primarily due to the small number of studies contributing to those estimates. In general, authors of CBT trials were more likely to report child outcome domains and less likely to report parent outcome and family functioning domains.

For ST, analyses of all available outcome domains (parent, family, and child) at post-treatment and follow-up were rated as low to very low quality, meaning that our confidence in the estimates of these effects is low and further research is very likely to have an important

impact on these findings. Low quality ratings for analyses of outcomes from ST trials were primarily due to the small number of studies contributing to those estimates.

For PST, analyses of parent mental health at post-treatment and follow-up were rated as high quality, meaning that further research is very unlikely to change our confidence in the estimate of these effects. Analyses of parent behavior at post-treatment and follow-up were rated as moderate quality, meaning that further research may have an important impact on these findings. Analyses of child and family functioning outcome domains for PST were rated as very low quality at post-treatment and follow-up, meaning that we are very uncertain about the estimates of these effects and further research is likely to have an important impact on these findings. Very low quality ratings for analyses of child and family outcomes from PST trials were primarily due to the small number of studies contributing to those estimates.

Discussion

Summary of Findings

Results from this systematic review and meta-analysis indicate that parent- and family-based psychological interventions can significantly impact parent behavior at post-treatment and follow-up for children and adolescents with chronic medical conditions. Across all psychological therapies, no effects were found for parent mental health, family functioning, child behavior/disability, child mental health, and child medical symptoms at post-treatment or follow-up. These findings are based on RCTs comparing psychological treatments to wait-list control and active comparators. PST emerged as an efficacious intervention for improving parent behavior and parent mental health at post-treatment and follow-up. There was insufficient evidence ($n \leq 2$ trials per analysis) to determine the effect of PST on other outcomes. CBT

showed no effect on extracted outcome domains at post-treatment. At follow-up, there was no effect of CBT on child medical symptoms. It was not possible to determine the effect of CBT on the other outcome domains at follow-up due to lack of studies reporting follow-up data. ST showed no effect on family functioning at post-treatment or on child symptoms at post-treatment or follow-up. It was not possible to determine the effect of ST on the other outcome domains at post-treatment or follow-up due to lack of studies reporting on those domains. More work is needed to evaluate the effect of PST on child and family outcome domains. Further work is also needed to determine the effect of CBT on child behavior/disability and mental health as well as parent and family outcome domains. Similarly, work is needed to evaluate the effect of ST on child behavior/disability and mental health as well as parent outcome domains. This lack of data limits our understanding of the efficacy of CBT and ST treatments for parents and children.

Findings from this study are consistent with our previous meta-analysis regarding the effectiveness of parent- and family-based interventions for youth with chronic medical conditions (Eccleston, et al., 2012), which also showed positive effects for PST on parent behavior and parent mental health. These results are also consistent with a meta-analysis of psychological interventions for pediatric oncology patients and their families, which showed no effects on child behavior or child mental health but positive effects for parent mental health and parent behavior (Pai, et al., 2006).

However, results from the current study are not consistent with our previous meta-analysis, which found support for the effects of CBT on child medical symptoms across a range of chronic medical conditions (Eccleston, et al., 2012) and specifically within chronic pain (Palermo, et al., 2010). Our findings are also not consistent with narrative reviews of systems interventions for youth with diabetes which have shown positive effects on child medical

symptoms and family functioning (Armour, Norris, Jack, Zhang, & Fisher, 2005; Grey, 2000; Harris, Freeman, & Duke, 2010; McBroom & Enriquez, 2009. There appears to be increasing interest in the field of pediatric psychology on the indirect impact of parent interventions on child mental health, behavior, and medical symptoms (e.g., Fedele et al., 2013), and publication of additional high quality RCTs in this area could increase our confidence about the estimate of effect for outcomes in this area.

The lack of effects for CBT and ST may be surprising to some, particularly because this review only included trials where parents were a primary treatment target. In contrast, our previous review identified positive effects for CBT on child medical symptoms but included numerous trials where parents were not a primary treatment target (Eccleston et al., 2012). This discrepancy may be due to the fact that the current review was more expansive in the types of patients that were included (i.e., a broader range of medical conditions) compared to our previous work. As a result, there was high variability in the outcome measures that were extracted which may have diluted the effects of the interventions included in the meta-analysis. In addition, many of the analyses planned for CBT and ST were not conducted due to a lack of studies reporting on the necessary outcome domain at post-treatment or follow-up. Some studies did not assess a given outcome domain, while others did not provide complete outcome data to allow for inclusion in the analysis. In general, these findings reflect that this is a young and developing area of research.

Taken together, results of this meta-analysis indicate that the evidence base for parent- and family-based psychological interventions for youth with chronic medical conditions is still in its infancy. The significant effects identified were small, and should be interpreted with caution. These findings are based on RCTs of psychological therapies compared to active (n = 14) and

no-treatment or wait-list control conditions (n = 22). Average sample size of included studies was moderate ($M_{parents}$ = 132/study; $M_{children}$ = 120/study), however the sample size of most studies (n = 23; 62%) was under 100. Only two analyses in the current review were rated as high quality (PST on parent mental health at post-treatment and follow-up), which suggests that other significant and non-significant findings presented here could be altered by future research.

This review has several strengths. First, we searched for RCTs of behavioral interventions for a broad range of pediatric populations commonly encountered by pediatric psychologists in clinical practice. Second, the amount of parenting content was standardized across included trials such that parents had to be identified by the authors as a primary intervention target and treatment delivered to parents had to equal at least 50% of the child's treatment duration. This represents an extension of our previous work (Eccleston et al., 2012), which had a more restricted range of illness groups and pooled studies with varying amounts of parent treatment content.

Findings from this review should be interpreted in light of several limitations. First, significant effects were small and emerged when there was greater homogeneity in outcome assessment and illness condition. For example, the same measure was used across studies for the analysis of PST on parent behavior (i.e., the Social Problem Solving Skills Inventory) and cancer was the only medical condition included in that analysis. In contrast, there was large variability in the outcome measures and illness conditions for many of the other analyses both within and across therapy types.

Second, several trials included multiple measurement tools to evaluate a single outcome domain without a-priori identification of the primary measure. While we attempted to select the

most generic, reliable, and frequently used measure within the field when this occurred, this may have influenced effect size estimates.

Third, this review is limited to RCT designs and does not include uncontrolled trials, case studies, or observational studies. The focus on RCTs allowed us to increase the precision of our estimates of effect size, however it does not allow us to make conclusions about the effectiveness of these interventions in clinical practice.

Fourth, our ability to summarize data for the meta-analyses of CBT, PST, and ST was limited due to the low quality and small number of trials reporting on the outcome domains assessed in this review. There is a need for randomized controlled trials that are high quality and low bias to evaluate the efficacy of parent- and family-based interventions for youth with chronic medical conditions. In addition, the CBT, PST, and ST interventions included in this review differed on several factors other than treatment type, including whether the intervention targeted the entire family system vs. parents only, as well as the number and length of sessions. Although beyond the scope of this review, future meta-analyses on this topic should consider evaluating these factors as potential moderators of treatment effectiveness.

Clinical Implications

In clinical practice, little guidance is available to determine whether and how to involve parents in psychological treatment for youth with chronic medical conditions. Results from this meta-analysis suggest that psychological interventions that specifically target parents can lead to improvements in parent behavior. In particular, PST appears to be a promising intervention for improving parent behavior and parent mental health in pediatric populations. Specifically, PST was found to improve parents' ability to solve problems as well as parents' anxiety and

depressive symptoms. This meta-analysis included trials of PST targeting parents of youth with newly diagnosed cancer (*n*=3; Sahler et al., 2005; Sahler et al., 2002; Sahler et al., 2013), traumatic brain injury (*n*=3; Wade et al., 2011; Wade, Wolfe et al., 2006; Wade et al., 2006a), asthma (*n*=1; Seid et al., 2010), congenital heart defects (*n*=1; McCusker et al., 2012), and diabetes (*n*=1; Nansel et al., 2012). Clinicians can consider PST for parents of youth with these medical conditions as well as others.

Although results from the present study did not show an effect of parent- and family-based psychological interventions on child outcomes, there are numerous descriptive studies which suggest that improvements in parent and family functioning could have indirect effects on child mental health, behavior and medical symptoms (Cappelli, et al., 1989; Friedman, et al., 2004; Logan & Scharff, 2005; Palermo, et al., 2007; Robinson, et al., 2007). Given these findings, pediatric psychologists in clinical practice should consider screening for concerns about parent mental health and behavior as part of routine intake procedures. This assessment can then inform clinical decision making regarding whether to deliver treatment only to the child, only to the parent, or jointly to the child and parent.

In particular, clinicians should consider parent- and family-based psychological therapies when parent behavior and parent mental health are identified as particular areas of concern. It is possible that child-only treatment may be sufficient for families with low parent distress and good family functioning. Parent-only or parent + child treatment may be indicated for families with high parental distress and poor family functioning. PST in particular may be a useful primary or adjunctive treatment for families with highly distressed parents.

Research Implications

There are several avenues for research to improve the quality of evidence for parent- and family-based psychological therapies. First, no RCTs of parent- and family-based psychological interventions were found for several medical conditions that are commonly encountered by pediatric psychologists (i.e., epilepsy, spina bifida, solid organ transplant). Replication studies conducted by independent research teams are needed, both within illness groups and across treatment types. For example, PST for families of children with newly diagnosed cancer has not been evaluated by any research team outside of Sahler and colleagues (2002; 2005; 2013).

Second, improvement in measurement and a-priori identification of the primary outcomes targeted by parent- and family-based psychological interventions for pediatric populations is necessary. Of the intervention types evaluated in this review, PST was the only treatment with high homogeneity in measurement of treatment outcomes particularly for the parent behavior and parent mental health domains. This is likely a reflection of strong leadership in the field of PST regarding the development and dissemination of guidelines for outcome assessment in both adult and pediatric populations (D'Zurilla & Nezu, 1999, 2007). This may also be a function of the relatively small number of research groups that have evaluated PST interventions in pediatric populations. Although consensus statements on outcome assessment are beginning to emerge for some pediatric medical conditions (e.g., McGrath et al., 2008), these guidelines do not yet exist for the majority of the medical conditions included in this review. In addition to guidelines on measurement for specific illness conditions, researchers should consider the theoretical underpinnings and purported targets of the treatment when designing a measurement plan.

Third, the sample size of most included studies was small. Researchers will need to consider multi-site recruitment methods to facilitate larger trials that will allow for appropriately powered tests of treatment efficacy and evaluation of treatment mechanisms. Little is known

about how parent- and family-based psychological intervention components lead to changes in parent, child and family outcomes. Furthermore, as mentioned above, no information is available to guide clinicians in determining whether and how to involve parents and families in treatment. To address these gaps, researchers should consider measurement of potential predictors, mediators and moderators early in the process of intervention development and trial design.

Fourth, reporting of age range of youth in the included trials was variable. For example, many of the trials evaluating youth with cancer did not report on the age range of youth, and those that did reported very wide ranges (e.g., 0-17; 11-18; Hoekstra-Weebers et al., 1998; Kazak et al., 2004; Stehl et al., 2009). In contrast, some medical conditions focused on only one age group. For example, the majority of trials targeting parents of youth with diabetes focused on adolescent populations. Increased standardization of reporting is needed so that all published trials of parent- and family-based interventions report on the age range of youth included in the study. Research is also needed to determine whether and how adaptations could be made to existing interventions for parents of youth at varying ages and developmental levels.

Finally, there is a need to set a standard in the field of parent- and family-based psychological interventions for pediatric populations to make treatment manuals and data publicly available to facilitate replication of intervention trials and re-analysis of results. Reluctance to share unpublished data for reanalysis is a pervasive problem in psychological research (Wicherts, Borsboom, Kats, & Molenaar, 2006). There are many reasons researchers may be unable to share unpublished data, such as loss or destruction of data, technological advances that make data stored on older devices no longer accessible, and lack of personal time/resources to respond to data requests.

Regardless of the reason, reluctance to share unpublished data has been associated with weaker evidence and a higher prevalence of errors in the reporting of statistical results (Wicherts, Bakker, & Molenaar, 2011). There is also a need to improve reporting standards within journals that publish RCTs of parent- and family-based psychological interventions. Only three studies included in this review were rated as having low risk of bias across all domains (Palermo, et al., 2009; Seid, et al., 2010; Stehl, et al., 2009). Editorial polices are needed to inform authors about reporting standards for RCTs that address concerns about risk of bias (e.g., requiring detailed descriptions of randomization and assessment procedures as well as reporting sample size, means and standard deviations for all analyses).

Conclusions

Findings from this meta-analysis suggest that parent- and family-based psychological therapies produce an improvement in parent behavior at post-treatment and follow-up, and PST in particular is promising for improving parent behavior and parent mental health. However, important issues remain to be addressed in this field. First, clinicians should routinely assess parent distress and determine whether and how to incorporate parents into treatment. Second, RCTs of parent- and family-based psychological therapies for youth with epilepsy, spina bifida, and solid organ transplant are needed. Third, important improvements (e.g. larger sample size, active comparator conditions, consensus statements for outcome assessment, and registration of trials) will improve the quality of RCTs investigating the effectiveness of parent- and family-based psychological interventions in this field and allow for more accurate meta-analyses.

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Note: Included studies are marked with an asterisk (*). Studies marked with the same letter after the asterisk (e.g., *[a]) are from the same trial. In the text, manuscripts from the same trial are cited using the author and year of the first published manuscript from that trial.

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