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Introduction

The diagnosis of pediatric cancer is a traumatic stressor; both parents and the child have to face the impact of the disease itself and its associated sequelae. Most caregivers are shocked by the unexpected diagnosis of their children's disease (Kazak et al., 2001; Kazak et al., 2002). With the rapid advances in medical treatment and research, the survival rate has improved substantially; however, the chronic nature of cancer prolongs the sufferings in caregivers (Cancer Research UK, 2016; Census and Statistics Department, Hong Kong, 2015; So et al., 2013; Ward et al., 2014). Studies have documented well the devastating impacts associated with caring for pediatric cancer patients. Parents may experience psychological stress and posttraumatic stress at any phase in the cancer trajectory and treatment period. More than half of the parents whose children were undergoing cancer treatment experience moderate to severe levels of posttraumatic stress symptoms (PTSS),which refer to psychological and physiological symptom response after exposure to disease or treatment-related traumatic experience (Graziano et al., 2016; Kazak et al., 2005a). It has been reported to correlate negatively with quality of life (QoL) and correlate positively with parental distress, including negative mood, depression, and anxiety (Morrill et al., 2008; Norberg and Noman, 2008). Parents report increased levels of anxiety and depression, and they frequently experience dysfunctional coping and impaired problem solving (Griffiths et al., 2006; Vrijmoet-Wiersma et al., 2008). However, the quality of care parents provide and their ability to care for the child are associated with how well they cope with the cancer diagnosis (Norberg and Noman, 2008). Hence, interventions for parents are warranted.

Psychoeducational interventions

Many interventions have been introduced in oncology. Psychoeducational intervention (PEI) is one of the frequently implemented interventions that target family caregivers. PEIs are "therapeutic approaches that involve information giving and receiving, discussion of concerns, problem solving, coping-skills training, expression of emotions, and social support" (Barsevick, Sweeney, Haney, & Chung, 2002, p. 75).

PEI can be delivered by different modalities, including through health education, problem-solving skill training, cognitive behavioral therapy, social support, or counseling (Barsevick et al., 2002). PEI is a promising intervention by increasing caregivers' knowledge of the disease the patient is suffering, strengthening their stress coping skills, and improving psychological outcomes (Sörensen et al., 2002). Thus, PEI can empower the caregivers of children who have cancer throughout the cancer trajectory.

However, individual studies have drawn different conclusions about the effects of PEI on caregiver outcomes, and there is a need to summarize the studies on the effects of PEIs and critically analyze the interventions for caregivers of children with cancer.

Although a meta-analysis was conducted to examine the effects of psychological interventions for parents of children with chronic illness, researchers have clustered the outcomes into 2 broad categories, namely psychological distress and adjustment (Pai et al., 2006). Thus, the efficacy of PEIs on individual psychological outcomes, such as PTSS, anxiety, and depression, were not evaluated. Besides, the included studies investigated the effects of the intervention on caregivers of cancer survivors; hence, the pooled effects of PEI on caregivers of children with cancer who are undergoing active treatment remain unknown (Pai et al., 2006).

In addition, 2 other recent meta-analyses summarized the effects of psychological interventions on parents of children with chronic illness (Eccleston et al., 2012; Law et al., 2014). Similar to the study by Pai et al. (2006), the Eccleston et al. study (2012) also categorized the outcomes into two classes: parent behavior and parent mental health. The other meta-analysis did not perform subgroup analysis for each individual disease, and the inclusion of patients with various diseases in the analysis could hamper accurate determination of the true effects of interventions on pediatric cancer patients (Law et al., 2014). In particular, parents of children with cancer have significantly higher stress than do parents of children with other chronic illnesses (Masa'Deh, 2015). Hence, a meta-analysis evaluating the effects of PEIs among parents of children with cancer is still warranted.

Aims

This paper aimed to review the best available evidence from randomized controlled trials (RCTs) to understand the effects of the PEIs on caregivers of children with cancer. Only RCTs were included in this review because this ensures that only high-quality studies are reviewed (Burns et al., 2011). The data regarding the effects of the PEIs from different studies was pooled to provide a more precise estimate. In addition, characteristics of the interventions, including content and type of interventions, were also analyzed.

To address the research problem pertinently, the following research question was formulated:

> What are the effects of the PEIs on the psychological and coping outcomes of caregivers of children with cancer, compared to a control group?

In this meta-analysis, psychological outcomes refer to negative emotions, including

PTSS, emotions, or moods experienced in response to stress, whereas coping outcomes refer to behaviors, skills, or cognitive efforts developed in managing stress. The primary outcome is PTSS, whereas other psychological and coping outcomes are secondary outcomes.

Methods

Search strategies

Studies were searched from inception to June 2017 in 11 English-language databases, including Embase, MEDLINE, PsycINFO, CINAHL, Scopus, AMED, Joanna Briggs Institute EBP Database, EBM reviews, British Nursing Index, Nursing & Allied Health Database, and ERIC. Moreover, a search of gray literature, including unpublished studies, reports, and dissertations, was performed. Furthermore, a manual search of the reference list of all the identified articles was performed to identify additional studies. The keywords and the medical subject headings used for searching, included “caregiver$”, “carer$”, “parent$”, “child*”, “p?ediatric*”, “adolescen*”, “oncology”, “cancer”, “ neoplasms”, “ carcinoma”, “psychoeducation”, “psychological”, “supportive”, “ educat*”, “ social support”, “psychosocial”, “ quality of life”, “depression”, “anxiety”, “stress”, “paediatric,” “child,” “parent,” “intervention,” and “education.”

Inclusion and exclusion criteria

Studies were included if they were (1) written in English; (2) conducted on caregivers of children ranging in age from birth to 18 years old diagnosed with any type of cancer and undergoing treatment, and (3) RCTs. Studies were excluded if they reported (1) outcomes on patients only and (2) did not focus on caregivers of pediatric cancer patients receiving active treatment.

Search results

In total, 17,777 citations were yielded from the database search. After removing 8,191 duplicates, titles, and abstracts of the remaining 9,586, citations were screened manually; 74 potential studies were identified and the full texts were reviewed. Two additional citations were identified from the reference lists of the potential studies, and 65 studies were excluded after full-text screening. Reasons for exclusion included irrelevance (n = 50), non-RCT studies (n = 13), subjects not meeting the inclusion criteria (n = 1), focus on patient outcomes only (n = 1). Eleven studies met the inclusion criteria and were included in the review. Figure 1 illustrates the search and selection procedures.

[Figure 1]

Figure 1 Flowchart illustrating the searching and selection procedures

Data extraction

Details of included studies, including study methods, settings, study participants, contents of the interventions, outcome measures, and their study results were extracted by 2 independent reviewers. Disagreements between the reviewers were resolved by discussion. The study authors were contacted to retrieve any missing data in the included studies.

Quality assessment

The Cochrane Collaboration's tool for assessing risk of bias was used to evaluate the risk of bias in the included studies (Higgins et al., 2011). The tool covered 7 domains, namely sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data, selective reporting, and other sources of bias. These domains aimed at detecting selection bias, performance bias, detection bias, attrition bias, and reporting bias.

Statistical analysis

Data were synthesized and statistically pooled using Review Manager 5.2. For outcomes measured by different instruments across the studies, the standardized mean difference (SMD), which is the effect size of Hedges' (adjusted) g, was calculated and reported. The mean difference and Cohen's d were computed if the outcome measurements used the same scale. Cohen's d was computed manually based on mean difference, standard error or standard deviation. The inverse-variance statistical analysis method was used in the meta-analysis; thus, weight was given to each study by the inverse of the variance of the effect measure~~, which can minimize the inaccuracy of the pooled effect estimate~~. The differences in the direction of instruments were corrected by subtracting the reported mean from the maximum possible value for the instrument. The heterogeneity of the studies was assessed by ~~the evolution of heterogeneity (~~I^2~~)~~, which measures the proportion of variability of intervention effect estimates in a meta-analysis that is attributed to differences between the included trials rather than to sampling error (Kitsiou et al., 2013; Thorlund et al., 2012). Random-effects models were used to calculate pooled mean differences to incorporate heterogeneity related to ~~different settings, interventions, and measurement tools across studies~~treatment effects (Riley et al., 2011). Findings of studies that were not comparable and could not be included in statistical pooling are presented in a narrative format.

In this meta-analysis, the intervention effect at immediate post-intervention and 3 months post-intervention were evaluated and reported.

Results

Study characteristics

Eleven studies were included and they were published between 1998 and 2015; of these, 7 were conducted in the United States (n = 7) (Askins et al., 2009; Kazak et al., 2005b; Marsland et al., 2013; Mullins et al., 2012; Sahler et al., 2013; Stehl et al., 2009; Streisand et al., 2000), 2 across the United States and Israel (n = 2) (Sahler et al., 2005; Sahler et al., 2002), 1 in Sweden (n = 1) (Cernvall et al., 2015), and 1 in the Netherlands (n = 1) (Hoekstra-Weebers et al., 1998). Among the 11 studies, only 2 employed theoretical frameworks: the Marsland et al. study (2013) was guided by the Transaction Model of Stress and Coping, whereas Mullins et al. (2012) adopted Mishel's model of illness uncertainty.

In addition to baseline and immediately after intervention data collection time points, 5 studies also conducted follow-up at 3 months post-intervention (Askins et al., 2009; Mullins et al., 2012; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002), and 1 study performed follow-up at 6 months (Hoekstra-Weebers et al., 1998).

All included studies focused on psychosocial outcomes: PTSS (n = 8), depression (n = 5), mood (n = 4), anxiety (n = 4), and stress (n = 2). A few of them investigated coping outcomes: problem-solving (n = 4) and social support (n = 2). The characteristics and findings of the included studies are shown in Table 1.

Four studies reported high attrition rates (34%–48%) (Askins et al., 2009; Hoekstra-Weebers et al., 1998; Kazak et al., 2005b; Sahler et al., 2013).

Quality assessment

Overall, a low risk of selection bias was detected among all included studies. Among the 11 studies, only 4 studies were judged as unclear risk (Marsland et al., 2013; Sahler et al., 2013; Sahler et al., 2005; Streisand et al., 2000), and the remaining 7 had low risk. For allocation concealment, 3 were at low risk (Cernvall et al., 2015; Kazak et al., 2005b; Stehl et al., 2009) and the remaining 8 studies were at unclear risk (Figure 2).

For the detection bias, only 1 study (Streisand et al., 2000) stated clearly in the methods that the outcome assessor was blinded and was therefore judged as low risk in blinding of outcome assessment. The other 10 studies did not provide information on this issue and were considered to have unclear risk.

Regarding the attrition bias, 2 studies (Sahler et al., 2002; Streisand et al., 2000) had unclear risk, and the other 9 studies were judged as low risk because they reported reasons for dropout and compared the difference between the participants who completed the study and those who withdrew from the study.

[Figure 2]

+ low risk of bias   ? unclear risk of bias

Figure 2 Risk-of-bias summary of each included study with the Cochrane Collaboration tool

Publication bias

Publication bias of each outcome variable was assessed by the funnel plot. It revealed asymmetry in PTSS, mood, depression, and anxiety outcomes, which might be due to the variation in the sample size or the inconsistent study results. For outcomes including PTSS, mood, and anxiety, the asymmetry of the funnel plot was due to outlier data from one study. With regard to depression, despite the clinical homogeneity, by which all 4 studies examining depression adopted the same intervention protocol and had the same inclusion criteria for subjects, the insignificant finding of the study by Askins et al. (2009) and Sahler et al. (2013) resulted in statistical heterogeneity with high I^2. This phenomenon might contribute to the asymmetry of the funnel plot. It should be cautious when interpreting the funnel plot as SMDs are correlated with standard errors, leading to the asymmetry.

Participants' characteristics

All included studies were RCTs, involving 1,515 participants in total. The sample size ranged from 22 to 430 with a mean of 138. In terms of subject recruitment from each family, 6 studies included only mothers as the participants (Askins et al., 2009; Mullins et al., 2012; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002; Streisand et al., 2000), 2 studies recruited 2 caregivers, including parents, grandparents, siblings, or extended family (Kazak et al., 2005b; Mullins et al., 2012; Stehl et al., 2009), whereas 1 study included both parents (Hoekstra-Weebers et al., 1998). Two studies, conducted by Marsland et al. (2013) and Cernvall et al. (2015), recruited 1 primary caregiver, the majority mothers. The mean age of the participants was comparable among the studies, ranging from 35 to 41.4 years.

Patient characteristics

Among all the patients (n = 1,228), 52.6% (n = 646) of the patients were male, with their mean age ranging from 4.5 to 13.9 years. The most common cancer type among the patients was hematologic cancer, (leukemia or lymphoma; n = 650), followed by brain tumor (n = 167) and non–central nervous system (CNS) solid tumor (sarcomas, Wilms tumor, neuroblastoma) (n = 137). All patients were newly diagnosed (24 hours–16 weeks) except those in Streisand et al. (2000).

Intervention characteristics

The interventions were in different modalities in the included studies. Four studies used a problem-solving skills training program as the intervention (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002). Such interventions included periodic discussion and revision of individual concerns and the promotion of problem-based and emotion-based coping strategies. Two studies used cognitive behavioral and family therapy (Kazak et al., 2005b; Stehl et al., 2009) and 2 studies used cognitive behavioral therapy only (Cernvall et al., 2015; Hoekstra-Weebers et al., 1998). Another 2 studies used stress management and coping enhancement therapy (Marsland et al., 2013; Streisand et al., 2000). One study combined different modules, including cognitive coping, problem solving, and social support (Mullins et al., 2012).

The dose of intervention was defined as the total number of hours and total number of sessions, with the total number of hours ranging from 1.5 hours to 12 hours (mean: 6 hours) and total number of sessions ranging from 1 session to 8 sessions (mean: 5.9 sessions). Two studies did not report the duration of the intervention (Kazak et al., 2005b; Stehl et al., 2009). For those reporting the duration of intervention, 4 lasted for 8 weeks (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002), 3 studies lasted for 12–18 weeks (Cernvall et al., 2015; Marsland et al., 2013; Mullins et al., 2012), one study lasted for 24 weeks (Hoekstra-Weebers et al., 1998) and 1 study had only 1 session (Streisand et al., 2000).

Format of intervention

The target recipients of interventions were solely the caregivers. Regarding delivery mode, all interventions were delivered in face-to-face sessions. Four studies added an extra component: telephone contacts (Marsland et al., 2013; Mullins et al., 2012), access to a study website (Marsland et al., 2013), and access to a personal digital assistant (Askins et al., 2009).

Interventions in all the studies were manual- or protocol-guided except Streisand et al., (2000), which did not provide information. All studies, except Marsland et al. (2013), were delivered by an individual with a psychology background. Among them, 2 studies involved a nurse and a psychologist in the delivery of the intervention (Mullins et al., 2012; Stehl et al., 2009).

Effects of psychoeducational interventions on psychological outcomes

Post-traumatic stress symptoms (PTSS)

Nine studies assessed the immediate post-intervention effects on PTSS. All studies, except Marsland et al. (2013) and Stehl et al. (2009), reported a significant improvement in PTSS. Of these 9 studies, the results of 5 studies can be pooled. Four studies were excluded from the analyses because mean scores and the standard deviations of the outcome variables were not provided (Kazak et al., 2005b; Mullins et al., 2012; Stehl et al., 2009; Streisand et al., 2000). Among these 4 studies, 2 reported significant improvement in PTSS in the intervention group (Kazak et al., 2005b; Mullins et al., 2012).

The overall effect showed a significant improvement in PTSS in the intervention group compared to the control group immediately after the intervention (SMD: –0.27, 95% CI: –0.50, –0.05, p = 0.02, Heterogeneity: Chi^2:: 10.43, df: 4 (p = 0.03), I^2 = 62%). Based on the results from 3 studies, the significant effect persisted for 3 months post-intervention (SMD: –0.29, 95% CI: –0.45, –0.12, p <0.001, Heterogeneity: Chi^2: 3.09, df: 2 (p = 0.21), I^2 = 35%) (Figure 3). According to Cohen (1988), the effect size of PEI on PTSS was considered small.

[Figure 3]

SD: standard deviation; Std. mean difference: standardized mean difference; CI: confidence interval

Figure 3. Overall effect of PEI on PTSS (immediately post-intervention and 3 months post-intervention)

Mood

Four studies evaluated the effects of PEI on mood. The PEI demonstrated significant reduction in the caregivers' mood immediately after the intervention (MD: –8.71, 95% CI: –13.26, –4.16, p <0.001, Heterogeneity: Chi^2: 8.00, df: 3 (p = 0.05), I^2 = 63%). The inconsistent findings between the studies contributed to the statistical heterogeneity. The significant effect persisted for 3 months post-intervention (MD: –7.37, 95% CI: –12.13, –2.61, p = 0.002, Heterogeneity: Chi^2: 3.61, df: 3 (p = 0.31), I^2 = 17%) (Figure 4). Small effect size was detected at both immediate post-intervention (Cohen's *d*: –0.2338, 95% CI: –0.3564, –0.1112) and 3 months post-intervention (Cohen's *d*: –0.1889, 95% CI: –0.3114, –0.0664).

[Figure 4]

SE: standard error of mean difference; CI: confidence interval

Figure 4. Overall effect of PEI on mood (immediately post-intervention and 3 months post-intervention)

Depression

The immediate post-intervention effects of PEI on depression were evaluated in 5 studies, and no significant effect was found (MD: –1.40, 95% CI: –3.67, 0.86, p > 0.05, Heterogeneity: Chi^2: 28.57, df: 3 (p <0.001), I^2 = 86%). Three studies examined the effect of the intervention 3 months post-interventions, where again no significant effect was detected (MD: –1.15, 95% CI: –3.10, 0.80, p > 0.05, Heterogeneity: Chi^2: 14.03, df: 2 (p <0.001), I^2 = 86%). (Figure 5). Similar to mood, a small effect size was detected at both immediate post-intervention (Cohen's *d*: –0.075, 95% CI: –0.1966, 0.1466) and 3 months post-intervention (Cohen's *d*: –0.0758, 95% CI: –0.2039, 0.0523).

[Figure 5]

SE: standard error of mean difference; CI: confidence interval

Figure 5. Overall effect of PEI on depression (immediately post-intervention and 3 months post-intervention)

Anxiety

PEIs were not successful in reducing the level of anxiety among caregivers. Five studies addressed the immediate post intervention effects of the intervention, and no significant effect was found (SMD: –0.15, 95% CI: –0.44, 0.13, p = 0.22, Heterogeneity: Chi^2:5.68, df: 4 (p = 0.29), I^2 = 30%) (Figure 6). The only study that assessed the 3-month post-intervention effects reported an insignificant result (Hoekstra-Weebers et al., 1998).

[Figure 6]

SD: standard deviation; Std. mean difference: standardized mean difference; CI: confidence interval

Figure 6. Overall effect of PEI on anxiety (immediate post-intervention and 3 months post-intervention)

Stress

The effects of PEI on stress could not be summarized because mean scores and the standard deviations were lacking in one study (Streisand et al., 2000). Both studies reported a significant time effect on stress, but there was no significant difference between groups with regard to caregivers' level of stress (Marsland et al., 2013; Streisand et al., 2000).

Effects of psychoeducational interventions on coping outcomes

Problem solving

Four studies assessed the caregivers' problem-solving ability. The PEIs were found to be successful at enhancing the caregivers' problem-solving skills immediately post-intervention, with small effect size (SMD: 0.26, 95% CI: 0.1, 0.43, p = 0.002, Heterogeneity: Chi^2: 5.06, df: 3 (p = 0.17), $I^2 = 41\%$); however, the effect did not last for 3 months (SMD: 0.16, 95% CI: –0.06, 0.38, p = 0.16, Heterogeneity: Chi^2: 5.68, df: 3 (p = 0.04), $I^2 = 64\%$) (Figure 7).

[Figure 7]

Std. mean difference: standardized mean difference; SE: standard error of standardized mean difference; CI: confidence interval

Figure 7. Overall effect of PEI on problem solving (immediately post-intervention and 3 months post-intervention)

Social support

Two studies assessed the effect of PEI on social support and no significant result was detected (SMD: 0.10, 95% CI: –0.34, 0.54, p = 0.65, Heterogeneity: Chi^2: 0.03, df: 1 (p = 0.87), $I^2 = 0\%$) (Figure 8).

[Figure 8]

SD: standard deviation; Std. mean difference: standardized mean difference; CI: confidence interval

Figure 8. Overall effect of PEI on social support (immediately post-intervention and 3 months post-intervention)

Discussion

This is the first meta-analysis that has collected evidence from existing RCTs to examine the efficacy of the effects of PEIs on caregivers of children with cancer undergoing active treatment. These PEIs were found to be effective in reducing PTSS (Askins et al., 2009; Marsland et al., 2013; Sahler et al., 2013; Sahler et al., 2005) and improving the mood of caregivers immediately after intervention (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002) and 3 months post-intervention. PEI was also found to improve problem-solving skills immediately post-intervention. Nevertheless, the effect of the intervention was not sustained after 3 months post-intervention (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002).

The evidence established might be explained by the mediating mechanism of the intervention. Two studies examined the mediating effects underpinning the intervention and hypothesized that negative affectivity, such as distress and depression, would be reduced through the enhancement of problem-solving skills (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002). They also reported that a change in mood was partially mediated by the change in problem-solving skills. Constructive problem-solving skills were enhanced by improving caregivers' rational problem-solving skills (Sahler et al., 2005; Sahler et al., 2002). In addition, the researchers explained that the intervention facilitated a reduction in adoption of dysfunctional coping strategies, including negative problem orientation and avoidance. Because these dysfunctional coping strategies accounted for the change in mood, decrease in their usage helps improve psychological well-being. This implies that enhancing caregivers' coping skills is beneficial to emotional outcomes. In fact, numerous studies provided evidence that maladaptive coping, such as avoidance, denial, or distraction coping, are associated with maladjustment and negative health outcomes, such as increase in levels of depression and anxiety (Blalock and Joiner, 2000; Carver et al., 1993; Miller et al., 1992; Pakenham and Bursnall, 2006).

The current findings were comparable to the results generated from previous meta-analyses, which studied interventions for caregivers of adults with cancer and parents of children with cancer (Northouse et al., 2010; Pai et al., 2006). Both meta-analyses demonstrated that PEIs provided to caregivers could improve their adjustment and reduce distress. However, it should be noted that the evidence established in the current meta-analysis was mainly generated from problem-solving skills training interventions, which derived from the same protocol (Askins et al., 2009; Sahler et al., 2013; Sahler et al., 2005; Sahler et al., 2002), so this finding may not apply to other problem-solving training or PEI.

In this meta-analysis, the beneficial effects of PEIs were not found in 3 outcomes:

anxiety, depression, and social support, and only small effect sizes were detected for PTSS and problem-solving outcomes in this meta-analysis. This might be due to several factors. First, 4 studies were pilot studies with relatively small sample sizes (Kazak et al., 2005b; Mullins et al., 2012; Sahler et al., 2002; Streisand et al., 2000), so they might have been underpowered for detecting significant intervention effects. Second, the effect size of PEIs for caregivers of cancer patients tends to be small (Pai et al., 2007). The included studies might have had insufficient power to detect a genuine effect because the 9 included studies either did not report the statistical power or aimed at detecting a medium effect size. Third, the dosage of the intervention varied substantially across the included studies. The number of sessions varied from 1 to 8 sessions, constituting a total contact time of 1.5 to 12 hours, and the duration of the intervention period varied from 1 day to 6 months. The large variation in dosage precludes the researchers from examining the related effect. The non-significant findings might also be due to the methodological flaws of the included studies.

In addition, 4 studies reported a high attrition rate (ranging from 34% to 48%) (Askins et al., 2009; Hoekstra-Weebers et al., 1998; Kazak et al., 2005b; Sahler et al., 2013). Studies reported the common reasons for subject withdrawal, as reported by the studies, were that caregivers felt overwhelmed and found it difficult to spare the time to attend the sessions. This might suggest that caregivers were unable to afford the time for a large number of sessions in the intervention or endure a lengthy intervention. In particular, the family-based interventions in some of the included studies jointly involved both parents or 2 caregivers; therefore, this might not be practical because the caregivers might not have sufficient time to schedule the participation in the intervention. Such participation might increase the burden on the families and hence increase the attrition rate of the intervention. Besides, most of the included studies recruited caregivers of children newly diagnosed with cancer, and these caregivers were experiencing a critical time in their lives; they might have felt overwhelmed about participating in research studies and seen such participation as a further burden. The aforementioned factors might contribute to the high attrition and reduce the effect of the intervention. Thus, further research studies should develop interventions with a format and intensity that are most appropriate for the caregivers and likely to increase adherence.

There are several limitations in this meta-analysis. First, the participants in this meta-analysis were mainly the caregivers of children newly diagnosed with cancer, and more than half of the included studies provided intervention to the mothers only (n = 6), so caution should be exercised in applying or interpreting the results of this meta-analysis. Furthermore, the included studies only investigated the intervention effect up to 3 months post-intervention; thus, the long-term effect of the PEI remains unclear. Future studies could follow up the participants for a longer period to examine the durability of the intervention effects. Despite these shortcomings, this meta-analysis highlighted the empirical evidence of the positive effects of PEI on PTSS, problem solving, and mood.

Implications for practice

The physical and psychological well-being of caregivers greatly affects the quality of care provided to patients. Hence, the importance of care of caregivers, particularly in the arena of chronic illness, has drawn researchers' attention. This is particularly true in pediatric oncology care, because caregivers are usually the parents, who suffer from both parenting and caregiving stress. The evidence generated from the current meta-analysis demonstrates that PEI facilitated coping enhancement. With the increased usage of positive coping or the reduction of dysfunctional coping, emotional outcomes were improved. Coping affects both physical and psychological health because it can mediate between stress and physical and psychological health. Therefore, health care professionals should consider coping a major element in PEI for the sake of boosting the caregiver's health.

Recommendations for future research

There are several recommendations for future research. First, this meta-analysis included small numbers of RCTs, reflecting a limited number of trials that examine the effects of PEIs provided to the caregivers of children with pediatric cancer. More RCTs are warranted to strengthen the evidence of the benefits of PEIs. Concerning the methodological quality of the 11 included studies, only 1 study reported adequate allocation concealment. Inadequate allocation concealment might introduce selection bias, and these studies tended to overestimate the intervention effect (Hewitt et al., 2005; Schulz, 2001). Blinding was also not explicitly reported in most of the studies. Whether the participants were blinded to the allocated treatment or the outcome assessors or investigators is unknown. The Hawthorne effect or reporting bias might occur. Thus, it is recommended that future research improve the methodological rigor.

In addition, although 2 studies examined the mediating mechanisms between the intervention and study outcomes (Sahler et al., 2005; Sahler et al., 2002), only 2 studies were guided by a theoretical or conceptual framework (Marsland et al., 2013, Mullins et al., 2012). Thus, the hypothesis identification, the selection of outcome measures, and the mediating mechanism of most studies were unknown, indicating a need for theory-driven studies to examine the mechanism–outcome effect.

Owing to the high attrition occurring in the study interventions, we recommend more studies to assess the appropriate duration and format of the intervention and thus deliver effective and feasible intervention. In this meta-analysis, all the included interventions were delivered in a face-to-face manner. Some of the participants reported that it was difficult to spare the time to attend the interventions. With the substantial increase in the use of the Internet and mobile phones, intervention delivery through these platforms might be feasible. We also recommend that future studies include molecular biomarkers as a physiological outcome. The use of molecular biomarkers as study outcomes is becoming increasingly popular in psychological research, and it can provide objective and reliable data to complement the evidence of the effects of the intervention (Piazza et al., 2010). Further studies are needed to examine the sustainability of treatment effects with a long-term follow-up. Enrolling a control group could be employed to strengthen the evidence of intervention effects and increase the validity of the intervention (Freedland et al., 2011; Latham et al., 2003).

Conclusion

The current meta-analysis examined the effects of PEIs on psychological outcomes for the caregivers of pediatric cancer patients. A moderate level of evidence has been established in PTSS, mood, and problem-solving outcomes. The PEIs play a pivotal role in improving problem-solving skills, reducing PTSS, and improving negative moods of caregivers; thus, it is important to incorporate caregivers' care into pediatric oncology care. To help caregivers cope more effectively through the cancer trajectory, more research is needed to explore further the most effective and feasible PEIs that suit their needs.

[Table 1]

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