

Parent outcomes and the role of parent psychological flexibility following intensive interdisciplinary pain treatment based on Acceptance and Commitment Therapy for adolescents with chronic pain: A non-randomized trial

Mike Kemani, Karolinska University Hospital

Marie Kanstrup, Karolinska University Hospital

Abbie Jordan, University of Bath

Line Caes, University of Stirling

Jeremy Gauntlett-Gilbert, Bath Centre for Pain Services

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Abstract

Objective

Parental factors are central in the development and maintenance of chronic pain in youths. Only a handful of studies have investigated the impact of psychological treatments for pediatric chronic pain on parental factors, and the relationships between changes in parental and adolescent factors. In the current study, we evaluated the effects of an intensive interdisciplinary pain treatment (IIPT) program based on Acceptance and Commitment Therapy (ACT) for adolescents with chronic pain, on adolescent and parental variables, and the relationship between parental psychological flexibility and adolescent pain acceptance.

Methods

Adolescents ($N = 164$) with chronic pain were included, with a mean age of 15.5 years, and completed the 3-week treatment with an accompanying parent ($N = 164$). Linear mixed-effects models were used to analyze change over time (from pre-treatment to 3-month follow-up) on parent (depression, health related quality of life and parent psychological flexibility) and adolescent (physical, social and emotional functioning, and adolescent pain acceptance) variables. Additionally, linear mixed-effects models were used to analyze the relationship between parent psychological flexibility and adolescent pain acceptance.

Results

Results illustrated significant improvements over time in depressive symptoms and levels of psychological flexibility in parents. Excluding social development, adolescents improved significantly in all assessed aspects of functioning, and pain acceptance. Additionally, changes in parent psychological flexibility were significantly associated with changes in adolescent pain acceptance.

Conclusions

Results indicated that treatment had positive effects for parents and adolescents, and a significant positive relationship between changes in parent psychological flexibility and adolescent pain acceptance was found.

Introduction

Background

Pediatric chronic pain is common and often associated with pain-related disability, emotional distress (Hoftun, Romundstad, Zwart, & Rygg, 2011; Vinall, Pavlova, Asmundson, Rasic, & Noel, 2016) and disrupted family functioning (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010). The impact of parenting a youth with chronic pain is illustrated in studies reporting significant levels of anxiety, depression and parenting stress in parents, in addition to financial burden and relationship difficulties (Eccleston, Malleon, Clinch, Connell, & Sourbut, 2003; Groenewald, Essner, Wright, Fesinmeyer, & Palermo, 2014; Jordan, Eccleston, & Crombez, 2008). These studies also highlighted the important role of certain parent factors (e.g. parent mental health and parent's own pain) in the development and maintenance of pediatric chronic pain (Chow, Otis, & Simons, 2016; McKillop & Banez, 2016). A growing body of research supports Cognitive Behavior Therapy (CBT) (Fisher et al., 2014), Acceptance and Commitment Therapy (ACT) (Gauntlett-Gilbert, Connell, Clinch, & McCracken, 2013; Wicksell, Melin, Lekander, & Olsson, 2009b) and intensive interdisciplinary pain treatment programs (Hechler et al., 2015) in treating pediatric chronic pain. A recent Cochrane review on children with chronic illnesses identified limited evidence that parent-only interventions produce positive outcomes, either for the parent or indirectly for the child (Eccleston, Fisher, Law, Bartlett, & Palermo, 2015). Examples of studies that

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have reported parent outcomes include an early study by Eccleston and colleagues (2003) in which results illustrated improvements in parent anxiety, depression and parental stress following an interdisciplinary CBT-program for pediatric chronic pain. Further confirming these benefits, a study by Law et al. (2017a), using a mixed methods approach to evaluate the feasibility of a parent problem solving skills training in the context of an intensive pediatric pain rehabilitation program, indicated that the intervention holds promise in improving parent problem solving skills and reducing parent distress.

For ACT in particular, results from two recent pilot studies suggested the utility of this approach for parents of adolescents with chronic pain, and illustrated positive changes in parent psychological flexibility, protective parenting responses and pain reactivity (Kanstrup et al., 2016; Wallace, Woodford, & Connelly, 2016). Relatedly, studies evaluating the Parent Psychological Flexibility Questionnaire (PPFQ), assessing parents' values based actions and acceptance of their own distress concerning their adolescent's pain, have highlighted the potential importance of targeting parent psychological flexibility as a means to enhance treatment effects for the children and adolescents (McCracken & Gauntlett-Gilbert, 2011; Wallace, McCracken, Weiss, & Harbeck-Weber, 2015; Wiwe Lipsker, Kanstrup, Holmström, Kemani, & Wicksell, 2016). In several studies evaluating ACT for chronic pain in adults, pain acceptance and psychological flexibility, i.e. the ability to in an accepting manner act in line with values also in presence of distress, have been shown to mediate improvements in pain related disability (Kemani, Hesser, Olsson, Lekander, & Wicksell, 2016; Vowles, Witkiewitz, Sowden, & Ashworth, 2014; Wicksell et al., 2013). Results from a few studies evaluating ACT in the field of pediatric chronic pain have indicated a similar role for pain acceptance as a predictor and mediator of improvements in pain related disability (Gauntlett-Gilbert et al., 2013; Wicksell, Olsson, & Hayes, 2011). However, more studies are needed to investigate the impact of ACT, e.g. provided in an intensive interdisciplinary format, on parental

psychological flexibility and parental acceptance, but also how these parental factors influence treatment targets for pediatric chronic pain.

Despite a number of studies pointing to the importance of including parental programs in pediatric chronic pain treatments and the role of parental factors in pain-related disability in youth, most interventions primarily address the symptoms and related disability of the child. There is a continued need for additional research examining the effects of pediatric chronic pain treatments that include parental interventions. Importantly, there is a general lack of, and need for studies that investigate how potential changes in parental variables relate to changes in child process and outcome variables (Palermo & Chambers, 2005). Specifically, for ACT-based approaches for pediatric pain, there is a need for larger studies that investigate if parental interventions influence general factors such as parental mental health, but also treatment specific targets such as parental psychological flexibility. As mentioned, previous studies evaluating ACT-based approaches for pediatric chronic pain illustrate improvements in child and adolescent pain related disability as well as in acceptance and psychological flexibility, but also that improvements in pain acceptance and psychological flexibility are related to improvements in pain related disability. However, there is a need for larger treatment studies that more closely investigate the relations between potential improvements in parent psychological flexibility and improvements in pain acceptance and functioning in children and adolescents.

Aims and hypotheses

In the current study, we aimed to evaluate the effects of an intensive interdisciplinary pain treatment program based on ACT for adolescents with chronic pain and their parents on:

- (1) parent and child outcomes (parent health related quality of life and depression, and

adolescent physical, social and emotional functioning); and (2) the relationship between changes in parent psychological flexibility and adolescent pain acceptance.

Based on previous findings (Gauntlett-Gilbert et al., 2013) we hypothesized that results would show significant improvements on all adolescent measures following treatment. With respect to parents, our more exploratory hypotheses were that (1) parents would improve on the included measurements following treatment; and (2) a positive relationship between improvements in parent psychological flexibility and adolescent pain acceptance would be observed.

Methods

Study setting and design

The residential treatment was carried out at a tertiary national specialist center in the south west of England. A non-randomized repeated measures design was used to evaluate treatment. The reporting was done in line with Transparent Reporting of Evaluations with Nonrandomized Designs (TREND) (Des Jarlais, Lyles, Crepaz, & TREND-group, 2004), but as the study was based on data collected as part of routine clinical data-collection it was not registered in a clinical trials registry.

Participants

Participants eligible for study inclusion were adolescents aged 11-18 years who experienced chronic pain and who consecutively attended a residential adolescent pain management program together with an accompanying parent. The adolescents were referred to the service from secondary or tertiary centers all across the U.K., and referral to this unit indicated that the adolescents had particularly severe or complex problems that had not responded to treatment in their local area. Adolescents were assessed for eligibility in a two-

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hour joint assessment with a Medical Consultant and a Clinical Psychologist, both of whom were experts in pediatric chronic pain. Inclusion criteria for adolescents included: (1) presence of pain-related disability; (2) expressed motivation to participate in the rehabilitation program; (3) an absence of medical pathology that would either require treatment or prevent safe participation in physical rehabilitation and (4) an absence of other effective medical options for pain control. No specific inclusion criteria were applied to parents, other than being the caregiver present during the residential treatment program. The data in the current study were derived from the same clinical service used in the study by Gauntlett-Gilbert et al. (2013), however the data used in the current study are from an entirely separate set of patients.

Results from power analyses using GPower (Faul, Erdfelder, Lang, & Buchner, 2007) illustrated that a total of 161 dyads were needed to detect a small effect ($d = 0.2$) with 80% power using an F-test for repeated measures (three assessment points) and an alpha value of 0.05. In total, 187 adolescents and their parents met the inclusion criteria, provided informed consent and were included in the study between May 2011 and March 2016. Figure 1. Illustrates the participant flow through each stage of this study. Recruitment rate could not be calculated as data for non-eligible or non-consenting patients were not collected. Analyses report on all eligible participants who provided data on at least one assessment point across all measures ($N = 164$ adolescents, $N = 164$ parents). Parental ages ranged from 29.7 to 61.7 years ($M = 46.3$; $SD = 5.6$) whilst adolescent ages ranged from 11.3 to 18.9 years ($M = 15.5$; $SD = 1.8$). Pain duration for the adolescents ranged from 8 to 199 months ($Mdn = 46.5$). Further demographic and background information regarding parents and adolescents can be found in Table 1 and Table 2.

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Please insert Table 1 about here.

Please insert Table 2 about here

Treatment content

Treatment comprised IIPT based on ACT. According to Hechler and colleagues (2015), IIPT includes three or more disciplines at the same facility (e.g. pain specialist, psychologist, and physiotherapist) working in an integrated manner to provide treatment in a day hospital or an inpatient setting for an average of eight hours per day over a 1-3-week period. The treatment (Gauntlett-Gilbert et al., 2013) included three main components: (1) physical conditioning; (2) activity management; and (3) psychological interventions. The ACT framework was consistently applied throughout the program by all professions in the clinical team (medics, psychologists, physiotherapists, occupational therapists and nurses). Emphasis was placed on the short- and long-term ends behaviors served, i.e. their function, in contrast to their form. Exposure in line with valued, but previously avoided activities, was promoted, using interventions based on acceptance, mindfulness, and valued living. Increased functioning was a primary target, and distressing experiences that could not be directly reduced, such as pain and related thoughts and emotions, were managed by means of acceptance and present moment focus rather than by strategies oriented towards changing these experiences, such as thought-challenging or similar techniques. All six ACT processes (Hayes, Luoma, Bond, Masuda, & Lillis, 2006) were targeted in the psychological sessions (acceptance, defusion, present moment contact, values, committed action, and self as context), and the content of the sessions and the exercises were delivered with consideration to developmental aspects.

Approximately 90 hours of treatment was provided during a 3-week period. More specifically, treatment comprised 34 hours of physical conditioning sessions, 22 hours of

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psychology, 15 hours of activity management (delivered by occupational therapist or nurse), and 15 hours of mixed sessions including medical talks, skills sessions (e.g. communication role play), individual sessions, and entry and exit assessments. Adolescents received, on average, three hours of individual input (i.e. the majority of the treatment was provided in a group setting).

Parents participated in all sessions, with the exception of a 4-day period in the middle of treatment during which adolescents worked independently of their parents. Parent involvement in treatment took three forms: (1) active participation in group sessions and modeling performance of treatment skills; (2) parent-only group sessions; and (3) some individual sessions with a keyworker, either with or without the child. In group sessions where their children (approximately six children per group) were present, parents (approximately six parents per group) were expected to support their child's treatment by modeling the acquisition and practice of skills themselves. Thus, parents exercised, and practiced psychological skills (e.g. mindfulness) and practical techniques (e.g. communication role play) in all sessions where they were alongside their children. They also received three hours of parent-only group input during the four-day phase where their children were working independently. These focused on the direct application of the skills (e.g. mindfulness, values focused behaviors) that they had learned in the previous week to challenging parenting situations.

Ethical considerations

The relevant health ethics committee, and hospital research committees approved the study; adolescent and parent data form part of a research database that includes, with consent, all treatment outcome data from the host clinical unit (RB502 and 17/SW/0002). All adolescents aged over 16 gave informed, written consent for themselves; participants under

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16 gave their assent, with formal consent from their parents. In addition, parents separately gave informed, written consent for the use of their own data.

Assessments

Assessments were conducted pre- and post-treatment and at a 3-month follow-up. Data, including demographic and medical information, was collected by means of self-report questionnaires that were administered by a psychology assistant not otherwise involved in later data preparation or statistical analyses.

Adolescent measures

Pain intensity. Average pain experienced over the past week was assessed using a numerical rating scale (NRS) with the endpoints 0 (“No pain at all”) and 10 (“Worst pain possible”) (Jordan et al., 2008).

Bath Adolescent Pain Questionnaire (BAPQ). The seven scales comprising the BAPQ were used to assess seven different domains related to longstanding pain in children and adolescents (Eccleston et al., 2005). Higher scores indicate more severe impact of pain in the respective domains, which include: (1) *Social functioning* (BAPQ-SF; nine items); (2) *physical functioning* (BAPQ-PF; nine items); (3) *depression* (BAPQ-DP, six items); (4) *general anxiety* (BAPQ-GA; seven items); (5) *pain-specific anxiety* (BAPQ-PSA; seven items); (6) *family functioning* (BAPQ-FF; 12 items); and (7) *development* (BAPQ-DE; 11 items). Item scales range from 0 (“never”) to 4 (“always”), with the exception of BAPQ-DE. In this domain respondents are asked to compare their own perceived progress regarding a specific developmental task with healthy same age peers using a 0 (“very behind”) to 4 (“very ahead”) response scale. The original psychometric evaluation paper for the BAPQ identified

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that α -values ranged from 0.79 to 0.89 (Eccleston et al., 2005), indicating adequate reliability of the seven BAPQ scales, and that concurrent validity was satisfactory when tested against other relevant measures (Eccleston et al., 2005). In the current sample, Cronbach's alphas for the respective subscales were: 0.64 (BAPQ-SF); 0.66 (BAPQ-PF); 0.84 (BAPQ-DP); 0.82 (BAPQ-GA); 0.87 (BAPQ-PSA); 0.79 (BAPQ-FF); and 0.85 (BAPQ-DE).

Chronic Pain Acceptance Questionnaire Adolescent (CPAQ-A). The adolescent's acceptance of pain was measured using the Chronic Pain Acceptance Questionnaire Adolescent (CPAQ-A) (McCracken, Gauntlett-Gilbert, & Eccleston, 2010). The questionnaire comprises 20 items, rated from 0 ("never true") to 4 ("always true") and uses a 0 to 4 scale with the verbal labels: "Never True," "Rarely True," "Sometimes True," "Often True," and "Always True. The measure includes two components of acceptance of chronic pain: activity engagement and pain willingness. Activity engagement assesses the degree of participation in regular daily activities in the presence of pain and pain willingness reflects non-attempts to avoid or control pain. Larger scores indicate higher levels of acceptance. The CPAQ-A has been validated for children and adolescents and demonstrates adequate reliability as illustrated by a Cronbach's alpha value of 0.87 (McCracken et al., 2010) for the total scale. In the current study, we used the total scale and the Cronbach's alpha for the total scale in this sample was 0.82.

Parent measures

Short Form Health Questionnaire 36 (SF-36). The Short Form-36 Health Survey (SF-36) is a 36-item measure assessing health-related quality of life in the following eight health domains (Ware & Sherbourne, 1992): Physical functioning (PF); role limitations due to physical problems (RP); bodily pain (BP); general health perception (GH); vitality (VT);

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social functioning (SF); role limitations due to emotional problems (RE); and mental health (MH). These domains are part of a physical component (PF, RP, BP, and GH) and a mental component (RE, VT, MH, and SF). Scores are commonly calculated for Physical and Mental Component Summary Scores (PCS and MCS) and are transformed to a range from 0 to 100 for the 8 subscales. The two components are normed using z -scores ($Mean = 50.0$; $SD = 10.0$). Higher component scores indicate better health status on each dimension. In the current study we used norms for the UK to calculate the sum scores for the two components (Jenkinson, Stewart-Brown, Petersen, & Paice, 1999). Cronbach's alpha in the current sample was 0.93 for the PCS and 0.91 for the MCS.

Perceived Health Questionnaire 9 item (PHQ-9). The 9-item Patient Health Questionnaire (PHQ-9) was used to assess severity of depressive symptoms in parents (Kroenke, Spitzer, & Williams, 2001). The PHQ-9 is a self-administered questionnaire that scores each of the 9 DSM-IV criteria as "0" (not at all) to "3" (nearly every day), and the total score can range from 0 to 27. Based on suggestions from a study by Manea and colleagues (2012) we considered a score of 11 to be an adequate cut-off value to classify participants as depressed. In a large study ($n = 6,000$) the questionnaire demonstrated adequate reliability and construct and criterion validity (Kroenke et al., 2001). Cronbach's alpha in the current sample was 0.85.

Acceptance and Action Questionnaire version 2 (AAQ-II). The Acceptance and Action Questionnaire (AAQ-II) assesses the construct of psychological flexibility. The AAQ-II is a 7-item unidimensional measure rated on a likert-type scale ranging from 1 ("Never true") to 7 ("Always true"), with higher scores indicating greater levels of psychological inflexibility. The questionnaire shows satisfactory structure, reliability, and validity (Bond et

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al., 2011). In a study by Bond and collaborators (Bond et al., 2011) the mean alpha coefficient was 0.84 (0.78-0.88), and the 3- and 12-month test–retest reliability was .81 and .79, respectively. In addition, the AAQ-II demonstrates appropriate discriminant validity. Cronbach’s alpha in the current sample was 0.90.

Parent Psychological Flexibility Questionnaire (PPFQ). The Parental Psychological Flexibility Questionnaire (PPFQ) aims to assess parents’ ability to in an accepting manner handle their own distress concerning their adolescent’s pain, while keeping actions directed towards goals and values (McCracken & Gauntlett-Gilbert, 2011). The version used in the current study comprised 17 items and four subscales (Wallace et al., 2015): *Values based action* (VBA); *Emotional acceptance* (EA); *Pain acceptance* (PA); and *Pain willingness* (PW). Parents respond to each item using a 7-point scale ranging from 0 = never true to 6 = always true. Higher scores indicate more psychological flexibility. In a previous study (Wallace et al., 2015) the 17-item version (including the three subscales VBA, EA and PW) results indicated good internal consistency ($\alpha = 0.88$) for the total scale. In the current study, we used the total score for the 17-item scale with a Cronbach’s alpha of 0.82.

Data analytical approach

We specified and estimated linear mixed-effects models of longitudinal change, including all time points (pre, post and 3-months follow-up). The models incorporated person-specific intercepts and slopes (Bolger & Laurenceau, 2013). Random effects and their associated covariances were retained based on their model contribution, as determined by a significant log likelihood ratio test. Residual maximum likelihood estimation (REML) was used to model parameters and standard errors, based on all participants who provided at least one valid assessment for the dependent variables (i.e. intention-to-treat analysis). REML

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produces unbiased estimates in the presence of missing data, under the assumption that data are missing for ignorable non-random reasons (Enders, 2011). Time was scaled such that 0 represented pre-treatment assessment, 1 post-treatment assessment, and 5 represented 3-month follow-up assessment (15 weeks from treatment start). When analyzing change over time in adolescent (BAPQ, Average pain past week and CPAQ) and parent variables (MCS, PCS, PHQ-9, PPFQ and AAQ) we also controlled for the influence of adolescent and caregiver demographics (gender and age) and baseline clinical variables (pain duration, pain intensity past week and parental chronic pain) by adding these variables as additional covariates.

We evaluated changes in adolescent pain acceptance over time, and the extent to which change was associated with improvements in parent psychological flexibility, by specifying a mixed-effects model with adolescent pain acceptance as the dependent variable and that in addition to time included parental psychological flexibility as a covariate. The model also included all BAPQ-scales as covariates in order to ensure that potential relationships between changes in adolescent and parent variables were not better explained by generic treatment improvements for the adolescents. First, we specified simple models including time as the only covariate. If a significant effect ($p < .05$) of time on the dependent variable was found we used a backwards selection approach to identify the most parsimonious model, which means that we included all variables in the model, according to descriptions above, and iteratively removed the variables with the largest p -value until the p -values for the remaining variables were below the 0.05 significance level. As a last step interactions between time and potential remaining significant covariates were tested, and retained in the model if significant.

In line with recommendations by Bolger and Laurenceau (2013) we included time as predictor and grand mean centered the other predictors (parent psychological flexibility and adolescent functioning) included in the models. All data was analyzed using IBM SPSS

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Statistics version 24.0 (IBM, 2016.). Using formulas in articles by Feingold (2009, 2015) and Morris and DeShon (2002), we calculated effect sizes comparable to Cohen's d and their corresponding confidence intervals, based on parameters from the simple outcome analyses, including time as the only covariate. In line with Cohen's suggestion we categorized these effect sizes as small ($d \approx 0.20 - d < 0.50$); medium ($d \geq 0.50 - d < 0.80$) and large ($d \geq 0.80$) (Cohen, 1992).

Results

Missing data

Missing questionnaire items were imputed via person mean score imputation for adolescents and parents for each subscale, in cases where $\leq 25\%$ of the items were missing (resulting in imputation for 0.3% and 0.5% of total items, respectively) (Peyre, Lepage, & Coste, 2011). In addition, 0.6% single-item scales in the SF-36 were missing and were imputed using group-mean imputation. In total, the completion quota for all included self-report questionnaires across all individuals and the three time points was 77.6%. Little's Missing Completely At Random (MCAR) test ($\chi^2(827) = 9.157$ and $p = 1.000$) indicated that this data was missing completely at random (Chen & Little, 1999). Data on serious adverse events were not collected systematically but were carefully monitored as a part of the clinical routines, and in doing this we did not detect any serious adverse events in relation to the study. Means and standards deviations for all self-report questionnaires, as well as the specific number of assessments for the different time points for both adolescents and parents are presented in Table 3.

Please insert Table 3 about here

Effects of treatment on parent and adolescent outcomes

Parents. There were significant main linear effects of time, from treatment onset to 3-month follow up, illustrating improvements on depression ($B = -0.191$, $SE = 0.090$, $p < 0.05$); acceptance of parents own discomfort (AAQ-II: $B = -0.273$, $SE = 0.108$, $p < 0.05$) and parent psychological flexibility ($B = 2.370$, $SE = 0.246$, $p < 0.001$). There were no significant main linear effects of time on mental and physical health-related quality of life ($B = 0.218$, $SE = 0.279$, $p > 0.05$ and $B = 0.245$, $SE = 0.170$, $p > 0.05$, respectively).

Adolescents. For the adolescents, analyses with linear mixed-effects models illustrated that there was a significant positive effect of time, from treatment onset to 3-month follow up, on social and physical functioning (BAPQ-SF; $B = -0.605$, $SE = 0.137$, $p < 0.001$ and BAPQ-PF; $B = -0.325$, $SE = 0.118$, $p < 0.01$); depression (BAPQ-DP; $B = -0.264$, $SE = 0.066$, $p < 0.001$); general and pain specific anxiety (BAPQ-GA $B = -0.468$, $SE = 0.126$, $p < 0.001$ and BAPQ-PSA $B = -0.733$, $SE = 0.080$, $p < 0.001$); as well as on family functioning (BAPQ-FF $B = -0.650$, $SE = 0.142$, $p < 0.001$). Additionally, there were significant positive effects of time on pain acceptance (CPAQ; $B = 1.867$, $SE = 0.227$, $p < 0.001$) and pain intensity ($B = -0.085$, $SE = 0.036$, $p < 0.05$). There was no significant main linear effect of time, on social development (BAPQ-DE; $B = 0.111$, $SE = 0.194$, $p > 0.05$). See Table 4 for model specific estimates, standard errors, significance values, confidence intervals and effect sizes for all parent and adolescent outcomes.

Please insert Table 4 about here

Associations between parent psychological flexibility and adolescent pain acceptance

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We specified and estimated a mixed-effects models to analyze if changes over time (pre, post, and follow up) in parental psychological flexibility were related to changes in adolescent pain acceptance over time (pre, post, and follow-up), while controlling for changes in different dimensions of adolescent functioning during these time periods, as measured with the BAPQ (see Table 4). The results from these analyses illustrated that changes in parent psychological flexibility was significantly associated with changes in adolescent pain acceptance while controlling for changes in all included domains of functioning ($B = 0.069$, $SE = 0.026$, $p = < 0.01$). See Table 5 for model specific estimates, significance values and confidence intervals, for the included parameters.

Please insert Table 5 about here

Discussion

In the current study, we evaluated the effects of an interdisciplinary IIPT program based on ACT for adolescents with chronic pain and their parents. For the adolescents, there were significant small to medium-sized improvements in physical, social and emotional functioning, and pain intensity, as well as a medium effect on pain acceptance. Importantly, these results are in consonance with previous studies showing that child and adolescent functioning and pain acceptance improves following ACT for adolescents with chronic pain (Gauntlett-Gilbert et al., 2013; Kanstrup et al., 2016; Wicksell, Melin, Lekander, & Olsson, 2009a). No improvements were identified in social development (as assessed by the BAPQ) for the adolescents, which somewhat aligns with the results from a previous study which found significant improvements at post-assessment in this domain, but not at follow-up (Gauntlett-Gilbert et al., 2013). Future qualitative studies could provide an important avenue to gain more in-depth understanding of how adolescents themselves perceive and understand

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completion of normative adolescent tasks; such as gaining social skills, in the context of living with chronic pain. Broadly, the persons included in the current study appear to be representative when compared to the adolescents in studies included in a systematic review of intensive interdisciplinary pain treatments for children (Hechler et al., 2015). In our sample approximately 78% of the participants were females, compared to 74% in the studies included in the systematic review. In the current study the average age of participants was 15.5 years ($SD=1.8$) compared to 13.9 years ($SD=1.5$) in the studies included in the review. As regards pain duration, mean pain duration was 4.9 years ($SD=3.6$) in the current sample, compared to 2.95 years ($SD=2.8$) in the studies included in the review.

For parents, there were small but significant improvements over time on depressive symptoms and acceptance of parents' own distress, as well as a significant medium effect size on parental psychological flexibility. The improvements in parent depression are in line with results from previous studies: an intensive residential treatment (Eccleston et al., 2003); an internet-delivered CBT study (Palermo et al., 2016b) for adolescents with chronic pain and their parents; and in two studies focusing specifically on parent problem solving skills (Palermo et al., 2016a; Palermo, Law, Essner, Jessen-Fiddick, & Eccleston, 2014). Health-related quality of life in parents did not improve following treatment and to our knowledge; previous studies evaluating similar treatments were also unsuccessful in identifying any longitudinal changes in health-related quality of life following parent-interventions for adolescents with chronic pain. It could be, in order to achieve long-term changes in parental health-related quality of life, that parents would require a higher treatment dose and/or more specific and targeted parental interventions. Possibly, other questionnaires, for example the PedsQol Family Impact Module (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004), may be better suited to assess change in this type of intervention.

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Prior studies have shown that parental psychological flexibility has improved following ACT (Kanstrup et al., 2016; Wicksell et al., 2011). However thus far, this is the largest study to demonstrate that parental psychological flexibility in relation to the child's pain – that is, the ability to deploy acceptance, mindfulness and values-based action – can improve in relation to treatment. Furthermore, these findings suggest that the Parent Psychological Flexibility Questionnaire can adequately capture treatment related change in ACT-based parental interventions and both supports and further adds knowledge to findings from previous studies.

Additionally, we found a significant relationship between improvements in parental psychological flexibility and the adolescents' improvement in pain acceptance. This relationship was specific in that it was not solely accounted for by generic improvements in adolescent functioning. Adolescent pain acceptance is associated with the most important facets of adolescent mood and functioning in the context of pain (McCracken et al., 2010). Additionally, studies have repeatedly shown that treatment-related improvements in pain acceptance are an important predictor and mediator of treatment-based improvements in functioning in ACT for adolescents (Gauntlett-Gilbert et al., 2013; Wicksell et al., 2011) and adults (Kemani et al., 2016; Vowles et al., 2014) with chronic pain. Thus, we were interested in how this important variable might be impacted by parent changes. It should be stated that these analyses were exploratory and need to be replicated in future studies. This being said, findings from the current study extend existing evidence by highlighting for the first time, a significant relationship in concurrent change in parental psychological flexibility and adolescent acceptance. It could reasonably be argued that this relationship was an epiphenomenon of treatment – as adolescents become less distressed and more functional, then their pain acceptance can improve, as it can for their parents. However, the results from this study indicates this not to be the case as changes in parent and adolescent acceptance

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were linked, independently of improvements to the child's overall mood or functioning. This underscores the interplay between adolescents and parents, and further implies the need to address parent's ability to deal with their child's symptoms and related distress in treatment. Important findings in this context have been reported by other research groups, such as in a recent study by Chow and colleagues (2016), where parent avoidance and protective behaviors were found to predict child functioning over time. Furthermore, in a long-term follow up of internet-delivered CBT for adolescents with chronic pain and their parents, parent distress predicted child disability over a 12-month period (Law et al., 2017b).

Palermo and Chambers (2005) suggest an integrated model including operant-behavioral theories and family systems theories, that highlight a multi-level interplay between family (e.g. family functioning) and individual parent factors (e.g. parenting style) and child pain and functioning, while taking into consideration e.g. child gender, developmental factors and coping strategies. In this study, we specifically investigated the relation over time between an individual parent factor, parent psychological flexibility, and a potential adolescent moderator/mediator variable, i.e. adolescent pain acceptance, and we tentatively suggest that parental psychological flexibility may be a treatment target that holds promise for improving both family and adolescent factors.

There are a number of limitations that need to be considered when interpreting the results from the current study. First, the study was non-randomized which undermines the causal inferences that can be drawn. Second, relatively few assessments were made, which did not allow for adequate separate analyses of change during the treatment and follow-up periods, using linear mixed effects-models. Third, with the exception of parent psychological flexibility we did not assess other parental factors that may be relevant in explaining improvements in the adolescent behavior and functioning, for example parental solicitous responses to pain behaviors. Fourth, internal consistencies at pre-assessment for the physical

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and social functioning subscales of the BAPQ were relatively low ($\alpha = 0.64$ and $\alpha = 0.66$ respectively) as compared to the other BAPQ subscales and to the original questionnaire validation (Eccleston et al., 2005), indicating lower reliability of these subscales for this particular sample. This should be kept in mind when interpreting the results.

A feature that we believe stands out in comparison with other treatment programs, and needs to be taken into consideration when assessing the results, is that adolescents and parents participated in nearly all aspects of the program together. In fact, some IPPT-programs have quite the opposite approach, in which parents and children spend the majority of treatment time apart. We see benefits and drawbacks with both approaches, but have chosen the former in order to ensure that the same information, practical and experiential treatment aspects were provided to both the adolescent and the parent, with the aim to further facilitate and bolster potential treatments benefits. Furthermore, this approach as well as the residential setting, provides the advantages being able to observe and to intervene in on-going interactions between adolescents and parents, on a daily basis over an extended period of time (Dunford, Thompson, & Gauntlett-Gilbert, 2014).

As regards the residential setting specifically, the format facilitates close collaboration between professionals and a thoroughly integrated treatment approach. Furthermore, such settings may provide an excellent context to study processes of change (predictors, moderators and mediators) using daily assessments, observational and other objective data (e.g. actigraphy). A drawback with this highly specialized type of treatment is that it cannot be made available to all those who need it. Furthermore, even though there is some support for the cost-effectiveness of this mode of delivery (Evans, Benore, & Banez, 2016), but additional studies are needed to compare this approach with potential alternatives, e.g. treatments that provide a combination of residential, outpatient and Internet provided modes of delivery. Given the needed commitment in terms of time and costs (e.g. related to school

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and work) the approach may be better suited for, for example, patients reporting higher levels of pain related distress and disability. This however, is also an important question related to issues previously touched upon, i.e. what in these treatment approaches that are key to improvements in outcomes and for whom and under what circumstances this type of treatment is effective in the short and long term?

Future studies using adequate designs that take the complex interplay between family, parent and child factors into consideration, as illustrated by the model of Palermo and Chambers (2005), can advance our understanding of how to improve child functioning, and which potential treatment targets that are likely to bring about change. On a related note, approximately 25% of the parents in the current sample reported that they themselves experienced pain and about 19% scored above the cut-off for clinical depression, which also indicates a need to conduct further studies which directly address parental chronic pain and related distress in the parent intervention, as these factors can have detrimental effects on the family environment (Dueñas, Ojeda, Salazar, Mico, & Failde, 2016). Furthermore, more than 90% of the adolescents in the current study reported that pain disturbed their sleep, which is illustrative of the extent of this problem for adolescents with chronic pain. This also points to the need for thorough assessment of sleep problems, and the need to properly address and evaluate such difficulties in clinical trials, in order to understand the role of sleep for functioning outcomes in the context of pediatric chronic pain.

In regard to study design and methodology, future studies ought to incorporate more intensive longitudinal assessments on core outcome and process measures (e.g. functioning and parent psychological flexibility and adolescent pain acceptance). Whilst challenging within the constraints of clinical practice, randomization and the incorporation of additional control conditions will strengthen inferences of causality in regard to both outcome and process analyses. In addition, qualitative studies exploring processes of change would make

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an important contribution to the field. For example, studies could focus on factors that parents perceive as important for effective coaching of a child with chronic pain towards improved functioning, and how psychological flexibility relates to this.

Results from the current and previous studies highlight the utility of parental interventions in conjunction with treatment for adolescents suffering from chronic pain. In particular, findings illustrate the potential relevance of targeting parent behaviors related to acting in greater accordance with long-term values while being willing to experience discomfort related to their children's suffering (i.e. parental psychological flexibility). This approach, central to the ACT model, is quite at odds with an intuitive approach to parent support that emphasizes the reduction of parent distress. So far, the amount of parental support that should be provided remains unclear, but the current study and two previous pilot studies (Kanstrup et al., 2016; Wallace et al., 2016) incorporated relatively minimal direct parental support (only a few clinical sessions). Thus, there is reason to believe that relatively low-intensity interventions can help, and that research on more substantial parent interventions is warranted.

In the current study, we evaluated the effects of IIPT based on ACT for adolescents with chronic pain and their parents. Results illustrated significant improvements in depressive symptoms in parents and parental psychological flexibility and in adolescent functioning and pain acceptance. In addition, results showed that changes in parental psychological flexibility were linked to changes adolescent pain acceptance, independent of improvements in functioning. These results provide important knowledge concerning the efficacy of parental support for adolescents suffering from chronic pain and the relationship between changes in central parent and adolescent factors.

References

- Bolger, N., & Laurenceau, J.-P. (2013). *Intensive longitudinal methods : an introduction to diary and experience sampling research*. New York: Guilford Press.
- Bond, F. W., Hayes, S. C., Baer, R. A., Carpenter, K. M., Guenole, N., Orcutt, H. K., . . . Zettle, R. D. (2011). Preliminary Psychometric Properties of the Acceptance and Action Questionnaire–II: A Revised Measure of Psychological Inflexibility and Experiential Avoidance. *Behavior Therapy*, 42, 676-688.
- Chen, H. Y., & Little, R. (1999). A test of missing completely at random for generalised estimating equations with missing data. *Biometrika*, 86(1), 1-13.
- Chow, E. T., Otis, J. D., & Simons, L. E. (2016). The Longitudinal Impact of Parent Distress and Behavior on Functional Outcomes Among Youth With Chronic Pain. *The Journal of Pain*, 17, 729-738.
- Cohen, J. (1992). A power primer. *Psychological Bulletin*, 112(1), 155-159.
- Des Jarlais, D. C., Lyles, C., Crepaz, N., & TREND-group. (2004). Improving the reporting quality of nonrandomized evaluations of behavioral and public health interventions: The TREND statement. *American Journal of Public Health*, 94, 361-366.
- Dueñas, M., Ojeda, B., Salazar, A., Mico, J. A., & Failde, I. (2016). A review of chronic pain impact on patients, their social environment and the health care system. *Journal of pain research*, 9, 457.
- Dunford, E., Thompson, M., & Gauntlett-Gilbert, J. (2014). Parental behaviour in paediatric chronic pain: a qualitative observational study. *Clinical Child Psychology and Psychiatry*, 19(4), 561-575.
- Eccleston, C., Fisher, E., Law, E., Bartlett, J., & Palermo, T. M. (2015). Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database Systematic Review*, 8.
- Eccleston, C., Jordan, A., McCracken, L. M., Sled, M., Connell, H., & Clinch, J. (2005). The Bath Adolescent Pain Questionnaire (BAPQ): Development and preliminary psychometric evaluation of an instrument to assess the impact of chronic pain on adolescents. *Pain*, 118, 263-270.
- Eccleston, C., Malleson, P. N., Clinch, J., Connell, H., & Sourbut, C. (2003). Chronic pain in adolescents: evaluation of a programme of interdisciplinary cognitive behaviour therapy. *Archives of disease in childhood*, 88, 881-885.
- Enders, C. K. (2011). Analyzing longitudinal data with missing values. *Rehabilitation Psychology*, 56, 267-288.
- Evans, J. R., Benore, E., & Banez, G. A. (2016). The Cost-Effectiveness of Intensive Interdisciplinary Pediatric Chronic Pain Rehabilitation. *Journal of Pediatric Psychology*, 41(8), 849-856.
- Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods*, 39(2), 175-191.
- Feingold, A. (2009). Effect sizes for growth-modeling analysis for controlled clinical trials in the same metric as for classical analysis. *Psychological Methods*, 14(1), 43-53.
- Feingold, A. (2015). Confidence interval estimation for standardized effect sizes in multilevel and latent growth modeling. *Journal of Consulting and Clinical Psychology*, 83(1), 157-168.
- Fisher, E., Heathcote, L., Palermo, T. M., de, C. W. A. C., Lau, J., & Eccleston, C. (2014). Systematic review and meta-analysis of psychological therapies for children with chronic pain. *Journal of Pediatric Psychology*, 39(8), 763-782.

- Gauntlett-Gilbert, J., Connell, H., Clinch, J., & McCracken, L. M. (2013). Acceptance and Values-Based Treatment of Adolescents With Chronic Pain: Outcomes and Their Relationship to Acceptance. *Journal of Pediatric Psychology*, 38, 72-81.
- Groenewald, C. B., Essner, B. S., Wright, D., Fesinmeyer, M. D., & Palermo, T. M. (2014). The Economic Costs of Chronic Pain Among a Cohort of Treatment-Seeking Adolescents in the United States. *The Journal of Pain*, 15, 925-933.
- Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and commitment therapy: model, processes and outcomes. *Behavior Research and Therapy*, 44(1), 1-25.
- Hechler, T., Kanstrup, M., Holley, A. L., Simons, L. E., Wicksell, R., Hirschfeld, G., & Zernikow, B. (2015). Systematic Review on Intensive Interdisciplinary Pain Treatment of Children With Chronic Pain. *Pediatrics*.
- Hoftun, G. B., Romundstad, P. R., Zwart, J.-A., & Rygg, M. (2011). Chronic idiopathic pain in adolescence – high prevalence and disability: The young HUNT study 2008. *Pain*, 152, 2259-2266.
- IBM. (2016.). IBM SPSS Statistics for Windows (Version 24.0). Armonk, NY: IBM Corp.
- Jenkinson, C., Stewart-Brown, S., Petersen, S., & Paice, C. (1999). Assessment of the SF-36 version 2 in the United Kingdom. *Journal of epidemiology and community health*, 53, 46-50.
- Jordan, A., Eccleston, C., & Crombez, G. (2008). Parental functioning in the context of adolescent chronic pain: a review of previously used measures. *Journal of Pediatric Psychology*, 33(6), 640-659.
- Kanstrup, M., Wicksell, R., Kemani, M., Wiwe Lipsker, C., Lekander, M., & Holmström, L. (2016). A Clinical Pilot Study of Individual and Group Treatment for Adolescents with Chronic Pain and Their Parents: Effects of Acceptance and Commitment Therapy on Functioning. *Children*, 3, 30.
- Kemani, M. K., Hesser, H., Olsson, G. L., Lekander, M., & Wicksell, R. K. (2016). Processes of change in Acceptance and Commitment Therapy and Applied Relaxation for long-standing pain. *European Journal of Pain*, 20(4), 521-531.
- Kroenke, K., Spitzer, R. L., & Williams, J. B. (2001). The PHQ-9: validity of a brief depression severity measure. *Journal of general internal medicine*, 16, 606-613.
- Law, E. F., Fales, J. L., Beals-Erickson, S. E., Failo, A., Logan, D., Randall, E., . . . Palermo, T. M. (2017a). A Single-Arm Feasibility Trial of Problem-Solving Skills Training for Parents of Children with Idiopathic Chronic Pain Conditions Receiving Intensive Pain Rehabilitation. *Journal of Pediatric Psychology*, 42(4), 422-433.
- Law, E. F., Fisher, E., Howard, W. J., Levy, R., Ritterband, L., & Palermo, T. M. (2017b). Longitudinal change in parent and child functioning after internet-delivered cognitive-behavioral therapy for chronic pain. *Pain*.
- Lewandowski, A. S., Palermo, T. M., Stinson, J., Handley, S., & Chambers, C. T. (2010). Systematic review of family functioning in families of children and adolescents with chronic pain. *The Journal of Pain*, 11(11), 1027-1038.
- Manea, L., Gilbody, S., & McMillan, D. (2012). Optimal cut-off score for diagnosing depression with the Patient Health Questionnaire (PHQ-9): a meta-analysis. *CMAJ*, 184(3), E191-196.
- McCracken, L. M., & Gauntlett-Gilbert, J. (2011). Role of psychological flexibility in parents of adolescents with chronic pain: Development of a measure and preliminary correlation analyses. *Pain*, 152, 780-785.
- McCracken, L. M., Gauntlett-Gilbert, J., & Eccleston, C. (2010). Acceptance of pain in adolescents with chronic pain: Validation of an adapted assessment instrument and preliminary correlation analyses. *European Journal of Pain*, 14, 316-320.

- McKillop, H., & Banez, G. (2016). A Broad Consideration of Risk Factors in Pediatric Chronic Pain: Where to Go from Here? *Children*, 3, 38.
- Morris, S. B., & DeShon, R. P. (2002). Combining effect size estimates in meta-analysis with repeated measures and independent-groups designs. *Psychological Methods*, 7, 105-125.
- Palermo, T. M., & Chambers, C. T. (2005). Parent and family factors in pediatric chronic pain and disability: an integrative approach. *Pain*, 119(1-3), 1-4.
- Palermo, T. M., Law, E. F., Bromberg, M., Fales, J., Eccleston, C., & Wilson, A. C. (2016a). Problem-solving skills training for parents of children with chronic pain: a pilot randomized controlled trial. *Pain*, 157(6), 1213-1223.
- Palermo, T. M., Law, E. F., Essner, B., Jessen-Fiddick, T., & Eccleston, C. (2014). Adaptation of problem-solving skills training (PSST) for parent caregivers of youth with chronic pain. *Clinical Practice in Pediatric Psychology*, 2, 212-223.
- Palermo, T. M., Law, E. F., Fales, J., Bromberg, M. H., Jessen-Fiddick, T., & Tai, G. (2016b). Internet-delivered cognitive-behavioral treatment for adolescents with chronic pain and their parents. *Pain*, 157, 174-185.
- Peyre, H., Leplege, A., & Coste, J. (2011). Missing data methods for dealing with missing items in quality of life questionnaires. A comparison by simulation of personal mean score, full information maximum likelihood, multiple imputation, and hot deck techniques applied to the SF-36 in the French 2003 decennial health survey. *Quality of Life Research*, 20(2), 287-300.
- Varni, J. W., Sherman, S. A., Burwinkle, T. M., Dickinson, P. E., & Dixon, P. (2004). The PedsQL Family Impact Module: preliminary reliability and validity. *Health and quality of life outcomes*, 2, 55.
- Vinall, J., Pavlova, M., Asmundson, G., Rasic, N., & Noel, M. (2016). Mental Health Comorbidities in Pediatric Chronic Pain: A Narrative Review of Epidemiology, Models, Neurobiological Mechanisms and Treatment. *Children*, 3, 40.
- Vowles, K. E., Witkiewitz, K., Sowden, G., & Ashworth, J. (2014). Acceptance and commitment therapy for chronic pain: evidence of mediation and clinically significant change following an abbreviated interdisciplinary program of rehabilitation. *Journal of Pain*, 15(1), 101-113.
- Wallace, D. P., McCracken, L. M., Weiss, K. E., & Harbeck-Weber, C. (2015). The Role of Parent Psychological Flexibility in Relation to Adolescent Chronic Pain: Further Instrument Development. *The Journal of Pain*, 16, 235-246.
- Wallace, D. P., Woodford, B., & Connelly, M. (2016). Promoting Psychological Flexibility in Parents of Adolescents With Chronic Pain: Pilot Study of an 8-Week Group Intervention. *Clinical Practice in Pediatric Psychology*, 4(4), 405-416.
- Ware, J. E., & Sherbourne, C. D. (1992). The MOS 36-item short-form health survey (SF-36). I. Conceptual framework and item selection. *Medical care*, 30, 473-483.
- Wicksell, R. K., Kemani, M., Jensen, K., Kosek, E., Kadetoff, D., Sorjonen, K., . . . Olsson, G. L. (2013). Acceptance and commitment therapy for fibromyalgia: a randomized controlled trial. *European Journal of Pain*, 17(4), 599-611.
- Wicksell, R. K., Melin, L., Lekander, M., & Olsson, G. L. (2009a). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – A randomized controlled trial. *Pain*, 141, 248-257.
- Wicksell, R. K., Melin, L., Lekander, M., & Olsson, G. L. (2009b). Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain--a randomized controlled trial. *Pain*, 141(3), 248-257.

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- Wicksell, R. K., Olsson, G. L., & Hayes, S. C. (2011). Mediators of change in Acceptance and Commitment Therapy for pediatric chronic pain. *Pain, 152*, 2792-2801.
- Wiwe Lipsker, C., Kanstrup, M., Holmström, L., Kemani, M., & Wicksell, R. (2016). The Parent Psychological Flexibility Questionnaire (PPFQ): Item Reduction and Validation in a Clinical Sample of Swedish Parents of Children with Chronic Pain. *Children, 3*, 32.