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Longitudinal change in parent and child functioning after internet-delivered cognitive-behavioral therapy for chronic pain

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Introduction

Chronic pain is common in childhood and results in high levels of disability and distress [13, 42]. The impact of pain extends to parents who experience their own significant levels of anxiety, depression, and parenting role strain [26]. Theoretical models have been proposed which emphasize longitudinal relationships between children's pain experiences and parent's emotional and behavioral functioning [25, 31]. Many cross-sectional studies support these models by demonstrating positive correlations between children's pain-related disability and parental distress and behavior [e.g., 7, 9, 17, 22, 38], where higher disability is associated with greater parental distress and more frequent maladaptive parenting behaviors.

Limited longitudinal data are available to test temporal relationships between child and parent functioning. Recently, Chow, Otis and Simons [6] examined parental distress and behavior at the start of multidisciplinary pediatric pain clinic treatment as a predictor of child disability at four-month follow-up. While there were no direct associations identified between baseline parental functioning and children's disability at follow-up, mediational analyses revealed that children's disability at follow-up was predicted by the interaction between baseline measures of parent and child functioning. To our knowledge, this is the only study to date that has examined longitudinal associations between parent and child functioning among families of youth with chronic pain. However, the interpretation and reproducibility of these findings may be limited due to the lack of a standardized treatment approach and the use of only two assessment time points.

Recognizing the important role of parenting behaviors, cognitive-behavioral interventions (CBT) for pediatric chronic pain have been developed that include a focus on modifying parent responses to support child wellness behaviors while decreasing reinforcement for illness behaviors. In two randomized controlled trials, efficacy of parent CBT strategies was demonstrated in reducing maladaptive parent behaviors (i.e., protective or solicitous behaviors) over time [19, 29]. However, these studies were designed only to examine parent behaviors as treatment outcomes. They did not examine the interrelationship of parent and child functioning over time in the context of treatment nor did these studies examine the potential impact of parent distress on child functioning. Therefore, there are a number of gaps in understanding of the relationship between child and parent functioning, which may have direct implications for how best to involve and intervene with parents in the clinical management of youth with chronic pain.

The aim of the present study was to examine longitudinal relationships between parent and child functioning among families of youth with chronic pain receiving a standardized family-based cognitive-behavioral therapy program for pain management delivered via the Internet (I-CBT). In our primary outcome analysis from this randomized controlled trial (RCT), we demonstrated beneficial effects of I-CBT, which significantly reduced child disability and maladaptive parent behaviors at the 6-month follow-up compared to the control condition [29]. In this secondary data analysis, we sought to extend our limited understanding of longitudinal associations between parent and child functioning by focusing on families who received the standardized CBT treatment protocol and examining outcomes over 12 months. As such, only families randomized to the I-CBT arm of the RCT were included in analyses.

For this study, we hypothesized that child disability, maladaptive parenting behaviors, and parent distress would improve over 12 months. Based on associations found in prior cross-sectional studies, we hypothesized a bi-directional relationship where: a) greater parental distress and maladaptive parenting behaviors at pre-treatment would be associated with less improvement in child disability over 12 months, and b) greater child disability at pre-treatment would be associated with less improvement in parent distress and maladaptive parenting behaviors over 12 months.

Methods

Study Design

Participants in this study were randomized to the active treatment arm in a RCT evaluating the efficacy of Internet-delivered family CBT for pediatric chronic pain. We have previously published three manuscripts using data from this trial. The first manuscript evaluated trajectories of change in children's pain intensity and activity limitations during the 8–10-week treatment period [30]. The second manuscript reported primary treatment efficacy outcomes at 6-month follow-up [29]. The third manuscript examined concordance between parent and child treatment goals [11]. The present manuscript is unique because it focuses solely on testing longitudinal relationships between parent and child functioning outcomes over 12 months among families randomized to the active treatment arm. We have not

previously reported data evaluating longitudinal trajectories of change in parent and child functioning.

Families in this study were enrolled from 15 multidisciplinary pediatric pain management clinics across the United States and Canada. In the analyses reported here, only families randomized to the active intervention group are included ($n = 138$). All families received the I-CBT program in addition to usual care at their pain clinic. Further details on the participants, design, and procedures of this study can be found in our prior publication reporting primary outcome analyses [29].

The Institutional Review Boards at the primary academic medical center and the referring pain clinics approved the study procedures. Parents gave informed consent and children gave assent prior to initiating study procedures. The trial was registered at clinicaltrials.gov prior to enrolling participants (NCT00853138).

Procedures

Internet-delivered Cognitive Behavioral Therapy (I-CBT)—Treatment content is based on social learning and cognitive-behavioral theories of pain management. The program has a travel theme and each treatment module represents a different destination around the world. There are eight treatment modules designed to be completed within 30 minutes each over 8–10 weeks. Children and their parents were given access to separate versions of the I-CBT program. Children completed modules on pain education, recognizing stress and negative emotions, relaxation methods, coping with pain at school, cognitive skills, sleep hygiene and lifestyle habits, and maintenance and relapse prevention. Parents completed modules on pain education, recognizing stress and negative emotions, operant strategies (using praise and reward systems), modeling, sleep hygiene and lifestyle habits, communication skills, and maintenance and relapse prevention. Homework was assigned at the end of six of the eight modules to facilitate skills practice. An online coach (PhD level post-doctoral fellows with expertise in CBT for pediatric pain management) provided feedback on the behavioral assignments and encouraged continued treatment engagement and skills practice through an email message center in the web program.

Assessment Protocol—All assessments were completed online through the secure study website. There were four assessment time points: pre-treatment (prior to randomization), immediate post-treatment (following the 8–10-week treatment period), 6-month follow-up, and 12-month follow-up. Children and parents were instructed to complete their assessments privately and independently. All assessments were completed in participants' homes.

Measures

Demographics—Parents reported on parent and child age, sex, race, parent marital status, parental education, parent employment, and family income at pre-treatment.

Child disability—To assess children's disability due to pain, the Child Activity Limitations Interview (CALI-21) [32] was administered via a prospective 7-day online diary. At study enrollment, children responded to an item-selection list of 21 activities and chose

the eight activities that were most difficult due to pain and important in their daily lives (e.g. going to school, sports, playing with friends). At each assessment period, children rated the difficulty of these activities once daily for 7 days on a 5-point scale (0=no difficulty; 4=extremely difficult). Total scores ranged from 0–32, with higher scores indicating greater disability due to pain. Mean scores across the 7 days of diary ratings at each time point were used in analyses.

Parent protective behavior—Parents completed the Protect subscale from the Adult Responses to Children's Symptoms questionnaire (ARCS) [40]. The 13-item Protect subscale assesses maladaptive, protective parental behaviors that reinforce children's pain complaints through increased attention and permission to avoid unpleasant activities. Parents rated how likely they were to engage in each parenting behavior on a 5-point scale (0=never; 4=always). Responses were averaged to provide the subscale score, which ranged from 0 – 523.5. Higher scores indicated more frequent engagement in protective parental behaviors. Scale reliability for each parent distress subscale was calculated using McDonald's coefficient omega with a 95% bias-corrected bootstrap confidence interval [24, 34, 35, 36]. Reliability was .86, 95% CI [.83, .88] at pre-treatment, .88, 95% CI [.86, .90] at immediate post-treatment, .90, 95% CI [.87, .92] at 6-month follow-up, and .90, 95% CI [.88, .92] at 12-month follow-up.

Parent distress—Parent distress related to caring for a child with chronic pain was assessed using the Bath Adolescent Pain – Parental Impact Questionnaire (BAP-PIQ) [15]. Parents reported on their own depressive symptoms (nine items), anxiety symptoms (six items), and catastrophizing about their child's pain (five items) over the past two weeks. Response options ranged from 0 = never to 4 = always. Items were summed to create total scores for each subscale, with higher scores representing greater parental distress.

Reliability for depressive symptoms was .88, 95% CI [.84, .90] at pre-treatment, .88, 95% CI [.85, .90] at immediate post-treatment, .87, 95% CI [.83, .90] at 6-month follow-up, and .89, 95% CI [.86, .91] at 12-month follow-up. Reliability for anxiety symptoms was .91, 95% CI [.89, .93] at pre-treatment, and .92, 95% CI [.90, .93], .90, 95% CI [.88, .92], and .93, 95% CI [.91, .94] for the remaining assessment time points through 12-month follow-up (respectively). Reliability for catastrophizing was .84, 95% CI [.80, .87] at pre-treatment, and .84, 95% CI [.80, .87], .87, 95% CI [.83, .89], and .90, 95% CI [.86, .91] for the remaining assessment time points (respectively).

Data Analysis Plan

The first aim of this study was to examine the shape of the mean trend of child disability, parent protective behavior, and parent distress across four measurement occasions from pre-treatment to 12-month follow-up and the degree to which significant between-person variability existed in the shape of these trajectories. Because parent distress was measured using multiple sub-scales, a Confirmatory Factor Analysis (CFA) was first used to test longitudinal invariance over time [3]. Invariance was essential because inferential statistics (e.g., means and regression parameters) require that measurements are consistent across time

[3, 20]. We then specified a multivariate curve-of-factors LGM (second-order LGM) to estimate the shape of the mean trend of parent distress [3, 20].

To evaluate change in child disability, parent protective behavior, and parent distress over time, a series of latent growth models (LGMs) were specified within the structural equation modeling framework [20, 33]. We hypothesized that child disability, parent protective behavior, and parent distress scores would generally decline (show improvement) over the 12-month period, however, we did not have a strong theory to suggest a specific functional form for change. Therefore, we identified the shape of the mean trend over time through the following sequence of nested model comparisons with a less restricted model compared to a more restricted model: (1) assess a level and shape model to allow the basis weights to optimally estimate the shape of the mean trend, (2) test a linear mean trend, and then, (3) evaluate a quadratic mean trend.

Specifically, we first specified a freely estimated level and shape model where time scores were anchored at 0 for Wave 1 at the pre-treatment assessment (to set the initial status) and 1 for Wave 4 at the 12-month follow-up assessment. The loadings for assessments at immediate post-treatment (Wave 2) and 6-month follow-up (Wave 3) were freely estimated [33]. Next, we imposed linearity on the change process (time scores: 0, 1, 2, and 3). Then, we tested a quadratic model (time scores: 1, -1, -1, 1). Fixing some of the time scores of the freely estimated model to a non-zero value allowed for direct comparisons of linear, quadratic and freely estimated models using a series of Satorra-Bentler scaled chi-square difference tests [33, 37].

The second aim of this study was to examine relationships between growth parameters of child disability and parent protective behavior and distress. Unlike multilevel modeling (random coefficient models, mixed-effects models), an extension of LGM for parallel processes allowed for a simultaneous examination of change in child disability, parent protective behavior, and parent distress over time and tested the influence of one change process on the other. This model combined the best fitting LGM for child disability, parent protective behavior, and parent distress into a single model that contained three sets of intercepts and slopes, one for each repeated-measures variable [33]. See Figure 1 for an illustration of the parallel process model.

All models were estimated using direct ML in the Mplus 7.3 software program to handle missing data with the robust standard error option to correct for non-normality [41]. For this study, goodness of fit was evaluated using multiple indices including the root mean square error of approximation (RMSEA) and its 90% confidence interval [39], the Tucker-Lewis Index (TLI) [1], and the Comparative Fit Index (CFI) [2]. Acceptable model fit criteria was RMSEA .08, TLI .90, and CFI .90 [4, 14].

Power Analysis

A primary goal of this paper was to examine change processes across four measurement occasions. Therefore, we were interested in determining the statistical power to detect a non-zero slope parameter. A series of Monte Carlo simulations were conducted a priori using Mplus 7.32. The intercept and slope growth factor variance ratio was 1 to 2 with a growth

curve reliability of .50, at pre-treatment (Wave 1) and .60 for assessments at post-treatment through 12-month follow-up (Waves 2–4) and a growth rate reliability of .60. For the simplest LGM with 6 free parameters and 8 df, results indicated a sample size as low as 66 would provide power of .80 to reject the null hypothesis that a slope parameter was equal to zero. Similarly, for the higher-order LGM with 40 free parameters and 50 df, a sample size of 75 would provide 80% power.

Furthermore, we specified a series of Monte Carlo simulations to determine the statistical power for predictive effects within the parallel process LGM. The focus of this power investigation was the proportion of replications for which the null hypothesis that a parameter equal to zero was rejected at the .05 level, given bias did not exceed 10% for any parameter and 5% in the key parameter of interest. Simulations were based on a sample size of 138. Results indicated 80% power to detect correlations among intercept parameters of .18 or larger, slope parameters of .07 or larger and predictive effects of slope regressed on intercept of .24 or larger.

Results

Participant Characteristics

Participants included 138 children with chronic pain and their parents, between the ages of 11 and 17 years ($M = 14.7$, $SD = 1.6$). Table 1 shows demographic characteristics of the children and parents in the sample. Children and their parents were primarily female, Anglo-American, and middle class with most parents having completed college or higher education. Children had musculoskeletal pain (42%), abdominal pain (12.3%), headache pain (8.0%), and multisite pain (42.0%), that was, on average, moderate to severe in intensity ($M = 6.2$, $SD = 1.7$). Unadjusted means and standard deviations for child disability, parent protective behavior, and parent distress from pre-treatment to 12-month follow-up are presented in Table 2.

The overall rate of missing data was low. Attrition was responsible for an average of 8.8% missing data (range: 1.0%–9.5%). In addition, due to a data collection error, 11.4% of participants ($n = 31$) were missing parent data at pre-treatment. Little's Missing Completely at Random (MCAR) test indicated that missing data were not likely to introduce bias, $\chi^2(554) = 605.889$, $p = .063$.

Parent Distress Construct

A confirmatory factor analysis (CFA) with repeatedly measured indicators was used to test longitudinal invariance of the parent distress construct [3]. While traditional methods (e.g., OLS approaches) assume all variables are free of measurement error and would not allow for a determination of whether time-related change is due to true change or changes in measurement of the parent distress subscales, the CFA framework corrects for measurement error and allowed for a straightforward examination of parent distress measurement equality across the four time points [3, 20].

A single factor model was specified in which indicators for depressive symptoms, anxiety symptoms, and catastrophizing loaded onto the latent variable for parent distress at each

time point. The indicators were sum scores with a range from 0 to 36 (depression), 0 to 24 (anxiety), and 0 to 20 (catastrophizing), with higher scores reflecting higher levels of the parental distress dimension. The effect-coding method was used to set the parent distress scale [21]. The initial configural CFA contained 12 indicators (3 indicators at each of 4 waves) and 4 constructs (parent distress at each of 4 waves) with 30 df.

Longitudinal measurement invariance was tested through the following sequence of steps: (1) test of equal patterns of indicator-construct loadings (configural invariance) over time, (2) test of equal factor loadings over time (loading invariance), and (3) equality test of the magnitude of the relationships between the indicators and constructs (intercept invariance) [3]. Beginning with the least restricted configural model, subsequent nested model constraints for the loading and intercept invariance models were tested using the RMSEA model test [20, 39] and CFI model test [5].

Each of the overall goodness-of-fit indices suggested that the configural model fit the data well and that the parent distress construct was measured equivalently across all four time points (see Table 3). Factor loading estimates suggested depressive symptoms, anxiety symptoms, and catastrophizing sum scores were reliable indicators of parent distress (R^2 range .50, .82). Moreover, scale reliability for parent distress was .85, 95% CI [.78, .90] at pre-treatment and .85, 95% CI [.79, .90], .87, 95% CI [.81, .91], and .89, 95% CI [.84, .92] for assessments at immediate post-treatment through 12-month follow-up, indicating strong internal consistency over time.

Longitudinal Trajectories of Change

Child disability—As shown in Table 4, the freely estimated level and shape model demonstrated marginal fit to the data. While the model chi square was non-significant and the CFI and TLI demonstrated good fit, the RMSEA point estimate was .09 with a relatively wide confidence interval (.000, .161). Because the RMSEA has been shown to be positively biased given lower degrees of freedom models [16], such as the current child disability LGM, given some support for .10 as the RMSEA cut off for poor fitting models [23], and considering the other fit indices [3], we determined this model sufficient. Neither the linear nor the quadratic model for child disability showed acceptable fit (see Table 4). Results indicated the child disability mean trend was best represented by the freely estimated level and shape model.

Based on the level and shape model, the average child disability score at pre-treatment (Wave 1) was 7.43 ($p < .001$). The total change from Wave 1 (pre-treatment) to Wave 4 (12-month follow-up) was -2.6 ($p < .001$). Of the total change, 73% occurred between the first two waves and 26% between the last two waves. These estimates suggest a rapid decline from pre-treatment to post-treatment followed by a leveling off from post-treatment to the 6-month follow-up before a final decline between the 6- and 12-month follow-up (see Figure 2). Both the intercept (14.02, $p < .001$) and slope (8.74, $p < .01$) variances were significant, indicating children significantly vary around the average intercept and slope. The growth curve reliability estimates ranged from $R^2 = .67$ to $.72$ over time, indicating the level and shape LGM model explained a meaningful amount of variation among children. The covariance between the intercept and slope was -5.32 ($p = .02$), signifying that on average

children with a higher disability due to pain at pre-treatment had a sharper decrease in disability due to pain over time.

Parent protective behavior—The initial level and shape model for parent protective behavior demonstrated excellent fit to the data (see Table 4). An imposed linear trend significantly degraded model fit, $\chi^2(2) = 35.36, p < .001$ suggesting that a linear model does not adequately capture the parent protective behavior trajectory. As shown in Table 4, the quadratic model indicated excellent model fit and did not significantly degrade model fit relative to the level and shape model.

Based on the quadratic model, the mean parent protective behavior score at Wave 1 was 1.33 ($p < .001$). There was a significant linear decline in parent protective behavior of $-.12$ ($p < .001$) over the four waves, suggesting a decrease of .12 points every measurement occasion. However, a significant quadratic effect of $.11$ ($p < .001$) indicated that parent protective behavior had an accelerating aspect such that the large decrease early in the trajectory began to diminish over time (see Figure 3). Results indicated a significant degree of variability among parent protective behavior scores at pre-treatment (Wave 1) ($.19, p < .001$); however, the linear slope and curvature did not vary across parents, suggesting a similar rate of linear decline and acceleration. There was no association among the intercept and linear slope or curvature.

Parent distress—Given the establishment of longitudinal factorial invariance for the parent distress construct, a second-order LGM was specified to allow for the separation of variance related to departures from the mean trend of parent distress over time and variance related to measurement error [3; 20]. As shown in Table 4, the level and shape second-order LGM for parent distress (i.e., latent parent distress construct) demonstrated excellent fit to the data. A chi square difference test comparing the level and shape model to a linear model resulted in a significant difference, $\chi^2(2) = 35.36, p < .001$, indicating that a linear trend was not adequate to model change in parent distress. The quadratic model resulted in excellent model fit and resulted in a nonsignificant difference relative to the freely estimated level and shape model, demonstrating the quadratic model was appropriate for the data (see Table 4).

The quadratic model for parent distress resulted in a mean score at pre-treatment (Wave 1) of 9.16 ($p < .001$) and a significant linear decrease of $.62$ ($p < .001$) points each wave. There was also a significant quadratic effect of $.63$ ($p < .001$), suggesting that the large decrease between pre-treatment (Wave 1) and immediate post-treatment (Wave 2) began to diminish over time (see Figure 4). While parents significantly varied around the mean distress score at pre-treatment (Wave 1) ($11.32, p < .001$), parents were essentially the same in how they changed over time. Results indicated no relationships among the intercept and linear slope or curvature.

Predictive Effects among Longitudinal Trajectories of Change

The second aim of this study focused on modeling change in child disability, parent protective behavior, and parent distress simultaneously to examine the predictive relations among initial pre-treatment levels of child disability, parent protective behavior, and parent

distress scores on change over time. To evaluate this question, we first specified a parallel process LGM by combining each best fitting LGM into a single model that contained three sets of growth parameters, one for each repeated-measures variable [33]. Then, predictive effects were added by regressing slopes onto intercepts.

The initial parallel process model demonstrated excellent fit, $\chi^2(135, n = 138) = 149.57, p > .05$, RMSEA = .028, 90% CI [.000, .052], TLI = .984, CFI = .989. All intercept and slope factors were significant, with no marked differences in parameter estimates from the prior individual LGMs. Results indicated no associations among initial pre-treatment levels of child disability, parent protective behavior, and parent distress. Also, there were no relationships among change parameters of slopes or curvature.

Next, linear and quadratic growth parameters were regressed on intercepts. The addition of predictive effects did not alter model fit. Results from the parallel process predictive model indicated that holding initial level of child disability and parent behavior constant, there was also a significant effect of parent distress at pre-treatment on total change in child disability. Specifically, the higher the initial level of parent distress the shallower the decline (standardized $\beta = .51, p < .05$) in total change in child disability. That is, on average, children whose parents had higher distress at pre-treatment had less improvement in disability than children whose parents had lower initial levels of parent distress.

Similarly, holding initial level of child disability, and parent distress constant, we found a significant effect of parent behavior at pre-treatment on linear change in parent distress. Specifically, on average parents with higher initial levels of protective behavior had a shallower linear decline in distress (standardized $\beta = 1.04, p < .05$) than parents with lower protective behavior levels at pre-treatment, indicating higher initial levels of parent protective behavior predicted higher levels of distress over time.

Furthermore, holding initial level of parent protective behavior and parent distress constant, there was a significant effect of child disability at pre-treatment (intercept) on the total change in child disability over time. Specifically, on average children with greater disability at pre-treatment had a sharper decline in disability over the course of the study (standardized $\beta = -.39, p < .05$) than children with lower initial levels of disability.

No other predictive effects were significant.

Discussion

In this study, we tested longitudinal relationships between parent and child functioning among families of youth with chronic pain receiving I-CBT for pain management. Findings from this secondary data analysis partially supported existing theoretical models of pediatric chronic pain, which propose longitudinal associations between children's pain experiences and parent's emotional and behavioral functioning [25, 31]. As expected, child disability, parent protective behavior, and parent distress improved over the 12-month study period. The greatest improvements occurred between the pre-treatment and immediate post-treatment assessment periods, with a leveling off or slight decline in improvement at 6-month and 12-month follow-up. Consistent with our hypothesis, we found that greater parent

distress at pre-treatment predicted less improvement in child disability over 12 months. These findings suggest that parents who are highly distressed may have more difficulty learning and implementing behavioral pain management strategies to support their child's adaptation to chronic pain. We also found that greater parent protective behavior at pre-treatment predicted less improvement in parent distress over the 12-month study period. Finally, we found that greater child disability at pre-treatment predicted more improvement in child disability over time. However, we did not find evidence of a bi-directional influence of child disability on parent functioning.

Understanding how parent and child functioning relate over time can help to identify treatment targets that may enhance the efficacy of I-CBT for youth with chronic pain. All families in this study received I-CBT that aimed to reduce children's pain-related disability, decrease maladaptive parenting behaviors, and increase parent's confidence in their ability to support their child. We have previously reported that our I-CBT intervention resulted in significant improvements in child disability and maladaptive parenting behaviors compared to a control condition [29]. However, these effect sizes were small, which indicates that I-CBT may benefit some but not all families of youth with chronic pain. Findings from the present study suggest that children in families of highly distressed parents may benefit less from the I-CBT intervention. In addition, findings from our study suggest that pre-treatment parent protective behavior may impact change in parent distress over time. Specifically, we found that parents with greater protective behaviors at pre-treatment demonstrated less change in parent distress over time.

Although high levels of parent stress, anxiety, and depressive symptoms have been reported in pediatric pain samples [7,9], surprisingly few interventions have been specifically directed at modifying parent distress. Indeed, there is variability in parent-focused interventions in psychological treatments for youth with chronic pain. Typically, parent intervention aims to change maladaptive parenting behaviors in the service of reducing children's disability [12].

The development of interventions that target distress among parents of youth with chronic pain is in its infancy. For example, one pilot RCT has evaluated the feasibility, acceptability and preliminary efficacy of problem-solving skills training (PSST) for parents of children with chronic pain [27, 28]. PSST is based on the social problem solving model [8] and teaches parents a structured approach to solving short- and long-term problems to reduce stress. Compared to standard care in a multidisciplinary pediatric pain clinic, parents who received PSST demonstrated greater decreases in distress and maladaptive behavioral responses to pain [27]. Positive downstream effects on children's mental health were also found for the PSST group, even though children did not receive the PSST intervention [27]. This pattern of findings is consistent with results from randomized controlled trials of PSST for parents of youth with other chronic medical conditions, including cancer, traumatic brain injury, and asthma [10, 18].

Our results suggest that parent distress may be an important factor to screen in evaluation of youth with chronic pain. Intervention targeting parent distress (e.g., PSST) may be beneficial for these families to help reduce parent stress, which ultimately may enhance children's outcomes. However, research is needed to identify the usefulness of screening and targeting

parent distress to help guide clinicians in the provision of comprehensive care for youth with chronic pain. We found that three subscales (anxiety, depression, and catastrophizing) from the BAPQ-PIQ [15] might be a useful composite measure of parent distress. Determining optimal screening measures for identifying parent distress, including the relative utility of global vs. construct-specific assessment tools, requires further investigation.

Contrary to our expectation, protective parent behaviors at pre-treatment did not predict change in child disability over time, and pre-treatment child disability did not predict longitudinal change in parent distress or behavior. Prior studies have reported cross-sectional associations between parent behavior and child disability [7, 17, 22, 38]. The only other study examining longitudinal associations between parent and child functioning among youth with chronic pain identified significant predictive associations between baseline parent behavior and child disability at follow-up [6]. Our study is the first to evaluate these relationships within families receiving a standardized CBT protocol and with long-term follow up. Further longitudinal research with larger sample sizes is needed to evaluate the natural course of these relationships over time, and to more stringently evaluate these relationships in the context of pain treatment.

We also found that children with greater disability at pre-treatment demonstrated more improvement in disability over the 12-month study period. This finding is difficult to interpret because it may reflect a regression toward the mean. For example, one might expect that youth with mild to moderate pre-treatment disability may be better suited for a low-intensity intervention such as I-CBT compared to youth with severe pre-treatment disability. Further research is needed to determine whether youth at varying levels of disability at pre-treatment benefit equally from I-CBT.

Generalizability of findings from this study are limited by the demographic characteristics of the sample which, like other published studies of youth with chronic pain, was primarily female, college-educated, Anglo-American and middle-to-upper middle class. Although our sample size was adequate to test the proposed models, research is needed to determine whether longitudinal relationships between parent and child functioning identified in the current study generalize to the broader population of youth with chronic pain, including families from more diverse socioeconomic backgrounds. The impact of cultural and socioeconomic factors on associations between parent distress, parent behavior, and children's pain experiences is poorly understood and should be considered an important area for future research.

In summary, findings from this study indicate that parent's emotional functioning can have a long-term impact on their children's outcomes from I-CBT intervention. Our findings suggest that families of children with chronic pain may benefit from additional intervention targeting parent distress. Research is needed to determine how to effectively screen for parent distress and what interventions may provide the most benefit for not only improving parent emotional functioning but also for enhancing pain outcomes in youth with chronic pain.

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Figure 1.

Parallel Process LGM. *Note.* Not all paths are displayed for clarity of presentation. Bolded predictive paths are significant at the .05 level.

Figure 2.
Model implied change in child disability.

Figure 3.
Model implied change in parent protective behavior.

Figure 4.
Model implied change in parent distress.

Table 1

Parent and child demographic characteristics at pre-treatment (n = 138 dyads).

Child demographic characteristics	
Gender (% female)	78.3%
Age in years (M, SD)	14.6 (1.6)
Race	
Anglo-American	92.0%
Black or African American	1.4%
Hispanic	1.4%
Other	4.5%
Missing	.7%
Primary pain location	
Head	8.0%
Abdomen	12.3%
Musculoskeletal	37.7%
Multiple	42.0%
Parent demographic characteristics	
Gender (% female)	92.8%
Race	
Anglo-American	93.5%
Black or African-American	.7%
Hispanic	2.9%
Other	2.2%
Missing	.7%
Marital status	
Married	75.4%
Not married	25.6%
Education	
High school or less	9.4%
Vocational school/some college	25.4%
College	38.4%
Graduate/professional school	25.4%
Missing	1.4%
Household annual income	
< \$10,000	2.3%
\$10,000–\$29,999	6.9%
\$30,000–\$49,000	15.4%
\$50,000–\$69,999	32.3%
\$70,000–\$100,000	13.8%
> \$100,000	29.2%
Employment status	

Full time	49.3%
Part time	24.6%
Not working	19.6%
Missing	6.5%

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Table 2

Sample descriptive statistics (M, SD) for parent and child functioning over time (n = 138).

Domain	Pre-treatment	Post-treatment	6-month follow-up	12-month follow-up
Child disability	7.43 (4.51)	5.58 (4.33)	5.30 (4.27)	4.92 (4.29)
Parent protective behavior	1.45 (0.55)	1.06 (0.57)	1.02 (0.59)	1.06 (0.67)
Parent distress ^I	9.81 (3.89)	7.79 (3.94)	7.37 (4.03)	7.89 (4.56)

^IEstimates from the intercept invariant CFA model.

Table 3

Fit Indices for the nested sequence in the longitudinal confirmatory factor analysis

Model	χ^2	df	p	χ^2_{sb}	df	p	RMSEA	RMSEA 90% CI	TFI	CFI
Null Model	1077.94	84	—	—	—	—	—	—	—	—
Configural Invariance	28.39	30	>.05	—	—	—	0.000	.000–.062	0.999	0.999
Loading Invariance <i>I</i>	33.36	36	>.05	—	—	—	0.000	.000–.056	0.999	0.999
Intercept Invariance <i>I</i>	38.95	42	>.05	—	—	—	0.000	.000–.053	0.999	0.999

I Evaluated with the RMSEA and CFI model test

Note. Each nested model contains its constraints, plus the constraints of all previous, tenable models.

Table 4

Fit Indices for level and shape, linear, and quadratic models for child disability, parent protective behavior, and parent distress.

LGM model	χ^2	df	p	χ^2	df	p	RMSEA	RMSEA 90% CI	TFI	CFI
Child Disability										
Free Slope	12.56	6	>.05	—	—	—	0.090	.000–.161	0.950	0.950
Linear Slope	24.58	8	<.01	12.02	2	<.01	0.124	.070–.182	0.905	0.873
Quadratic Slope	16.88	4	<.01	4.32	2	>.05	0.155	.084–.235	0.852	0.901
Parent Behavior										
Free Slope	6.63	6	>.05	—	—	—	0.028	.000–.118	0.996	0.996
Linear Slope	41.99	8	<.001	35.36	2	<.001	0.178	.127–.233	0.841	0.788
Quadratic Slope	6.00	4	>.05	0.63	2	>.05	0.061	.000–.155	0.981	0.987
Parent Distress										
Free Slope	48.91	48	>.05	—	—	—	0.012	.000–.161	0.998	0.999
Linear Slope	72.76	50	<.05	23.85	2	<.001	0.059	.000–.059	0.959	0.969
Quadratic Slope	44.88	46	>.05	4.03	2	>.05	0.000	.000–.056	0.999	0.999

Note. Each nested model contains its constraints, plus the constraints of all previous, acceptable models.