Use of Smartphones to Prospectively Evaluate Predictors and Outcomes of Caregiver Responses to Pain in Youth with Chronic Disease

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Abstract

This study examined outcomes and predictors of different types of responses to child pain used by caregivers of youth with chronic disease. Sixty-six children and adolescents (ages 7–18) with juvenile idiopathic arthritis answered questions about pain, pain interference in activities, and mood on a smartphone three times per day for one month, while a caregiver contemporaneously answered questions about their own mood and use of protecting, monitoring, minimizing, or distracting responses to their child's pain. Multilevel models were used to evaluate (a) how a child's pain and pain interference changes after a caregiver uses different types of pain responses; (b) the extent to which caregiver responses to pain vary across days; and (c) whether variability in caregiver responses to pain is predicted by changes in child pain characteristics, child mood, and/or caregiver mood. Results showed that children's pain intensity and pain interference increased following moments when caregivers used more protective responses, whereas children's pain interference decreased following times when caregivers responded with minimizing responses. Caregiver pain responses varied considerably across days, with caregivers responding with more protecting and monitoring responses and fewer minimizing responses at moments when their child reported high levels of pain unpleasantness and pain interference. Caregivers also were found to respond with fewer protective responses at moments when they themselves were in a more positive mood. Implications for clinical recommendations and future studies are discussed.

Keywords

pain; children; adolescents; arthritis; parent response

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1.0 Introduction

Children with certain chronic illnesses such as Juvenile Idiopathic Arthritis (JIA) experience pain that can disrupt participation in typical activities. How parents respond to their child's pain may mitigate or exacerbate the child's perception of pain and the impact of pain on functioning [38]. Parent monitoring and protecting responses to pain tend to be associated with greater pain expression and disability in children due to maintaining a focus on pain and/or helping facilitate escape from perceived pain-provoking settings [1,13,28,39,47,51]. Conversely, parent distracting responses that redirect a child's attention from pain tend to be associated with reduced pain and distress [12,49]. Clinicians therefore often advise parents of youth experiencing pain to limit discussion about pain and to help focus attention elsewhere. However, insights about parent pain responses have been generated from studies using experimentally-induced pain or from samples of youth with idiopathic/benign chronic pain [12,38]. Comparatively less is known about predictors and outcomes of parent responses to pain in youth with a painful chronic disease. Pain expression in a child with chronic disease may be likely to elicit extra vigilance and protective responses from parents due to a heightened perception of the child's vulnerability to a medical issue [24]. Which responses lead to better or worse pain or functioning outcomes in youth with a painful chronic disease remains speculative.

Most studies on parent pain responses also have inferred parent response style from a single observation or subscale score and correlated this with a health outcome. This approach implicitly assumes that parent responses to pain are relatively consistent over time. Yet variables known to change dynamically, such as emotional distress, have been shown to be associated with parents' use of certain pain responses [10,11,13]. By extension, parent responses to pain may also shift from one moment to the next depending on context. The extent to which and why parent responses to their child's pain vary has been overlooked.

We sought to address these knowledge gaps by using smartphone-based ecological momentary assessment methodology to investigate variability, predictors, and outcomes of different types of caregiver responses to pain in youth with chronic disease (JIA). This study represents the parent-focused portion of several related studies evaluating child pain and functioning in JIA [6,7,14]. Smartphone-based ecological momentary assessment increasingly has been used to understand contextual predictors of a wide variety of behavior such as substance use and physical activity [e.g., 5,31]. In the current study, we used ecological momentary assessment in dyads consisting of a youth with JIA and a parent/ caregiver to examine the hypothesis (based on operant learning theory [20]) that children's pain intensity and interference will increase after caregivers use more protective and monitoring responses and decrease after parents use more distracting responses. We also sought to determine the extent to which caregiver responses to child pain vary more between or within caregivers, and the extent to which baseline covariates (demographics, disease severity, and perceived child vulnerability) and time-varying contextual variables (child pain characteristics and child and caregiver mood) predict caregivers use of pain responses.

2.0 Method

2.1 Participants

Participants were recruited over the course of one year from a pediatric rheumatology clinic associated with an academic medical center. Eligibility criteria for the study included ages between 7–18 years (inclusive), a diagnosis of polyarticular JIA from a board-certified pediatric rheumatologist, joint pain within the past 6 months, and the presence of a primary caregiver at time of enrollment. Children were excluded if they had another chronic pain condition (e.g., fibromyalgia), a poorly controlled psychiatric disorder, significant cognitive impairment or illiteracy. Children also were excluded if they were not currently attending school (due to the larger study requiring school attendance to be able to assess functioning at school) or were non-English speaking. Of the 93 families approached for participation in the study, 3 were ineligible and 19 declined to participate. There were no significant differences in child age or sex between those that declined participation and those that agreed. An additional 5 families were excluded from the final sample due to early study withdrawal; excluded families were comparable on child age, sex, and disease severity, but were significantly younger in parent age ($\underline{M}$ = 27 years old vs $\underline{M}$ = 40 years old).

The final analyzed sample comprised 66 dyads. Child participants ranged in age from 7 to 18 years (M = 12.8, SD = 2.8), with 73% female. The child race distribution was as follows: 79% White/Caucasian, 19% Black/African-American, and 2% Native Hawaiian or other Pacific Islander. These demographics are largely representative of the general population of youth with JIA [42]. Baseline disease activity, based on a global rating provided by the attending pediatric rheumatologist, was rated as minimal for 15%, mild for 45%, moderate for 32%, and severe for 8% of the sample. Children continued on their usual medications during the study (disease-modifying anti-rheumatic drugs, biologics, and/or acetaminophen or non-steroidal anti-inflammatory drugs). Caregiver participants ranged in age from 29 to 64 years, with most being the biological mother of the child (86% = biological mother; 9% biological father; 5% other). Of the caregiver sample, 73% were married (14% single, 8% divorced, 4% separated, and 1% widowed). Most primary caregivers were working full-time (80%), and 78% had completed at least some schooling beyond high school.

2.2 Procedure

Study staff mailed information letters to the caregivers of pre-screened patients with JIA approximately one week prior to a routine pediatric rheumatology clinic visit. A research assistant met with interested families following the routine clinic visit and obtained written informed consent and assent on a form approved by the hospital's Institutional Review Board. Enrolled children and caregivers then independently completed computerized baseline self-report measures and were trained by a research assistant in the completion of electronic (e-) diaries on a T-Mobile® Dash smartphone. The caregiver and child both were provided a T-Mobile® Dash phone (with data service only) to use for the duration of the study. Phones were programmed to prompt children and caregivers to complete the e-diaries three times per day (morning, afternoon, and evening) at times that the child and caregiver approved at the initial study visit. Participants were automatically sent a reminder prompt 2

minutes after the initial prompt if they had not yet began the survey; if responses were still not entered, data were considered missing for that time interval.

After completing the e-diary training, both caregivers and child participants took home a smartphone with a printed instruction manual. The participating dyads were asked to complete e-diaries for 30 days. Data were automatically uploaded from the phones to a password-secured internet server using the phones' wireless data plan. At the end of the data collection period, smartphones were returned to the study team by mail in a prepaid mailer. Participants were reimbursed up to a maximum of $10 for participation based on the number of completed weeks of e-diary entries.

2.3 Measures

2.3.1 Caregiver electronic diary measures

Caregiver responses to child pain: Caregiver pain responses were assessed on the caregiver e-diary using a 20-item subset of questions taken from the Adult Responses to Children's Symptoms Questionnaire (ARCS), which is a well-validated measure of caregiver responses to child pain [44, 48]. Caregivers were asked to check boxes next to any pain response they had used since last completing the electronic diary. The 20 items used from the ARCS were classified into 4 subscales (protecting – 9 items, monitoring – 4 items, minimizing – 4 items, and distracting – 3 items) based on results of a recently redone factor analysis of ARCS items with caregivers of children with pain-related conditions [35]. Table 1 provides definitions and sample items each of these four types of pain responses. Responses to items within each category were summed together for each moment of assessment, such that values indicated the number of pain responses within each category that caregivers reported using at the given time. Cronbach alpha internal consistency values for the subscales were .83 (Protect), .80 (Monitoring), .57 (Minimizing), and .78 (Distracting).

Mood: Caregivers were asked to rate their own current mood using the 20-item Positive and Negative Affect Schedule [50]. Caregiver participants indicated the degree to which they were currently experiencing a given affective descriptor using a response scale of 1 ("very slightly or not at all") to 4 ("extremely"). Raw scores for negative and positive affect items were averaged separately to comprise indices of negative and positive affect per instructions for the original scale, with higher scores indicating greater negative or positive affect. The scale has excellent psychometrics and previously has been used for state assessment of mood in daily diary studies [15]. Internal consistency (Cronbach alpha aggregated across measurement occasions) for this scale for the caregiver sample in this study was .95 for positive affect and .87 for negative affect.

Interaction with child: At each e-diary assessment, caregivers were asked whether or not they were presently or recently with their child; this information was used for descriptive information on the number of times each day that parents had the opportunity to influence their child through their responses to pain.

2.3.2 Child electronic diary measures

Pain characteristics: Children were asked to rate pain intensity on a 50mm electronic visual analog scale (converted to a 0 to 100 scale for analyses) with anchors "no pain" to "worst pain possible." Children also were asked to rate the unpleasantness of their pain using a similar visual analog scale with anchors "not bothering me at all" to "bothering me a lot." Electronic visual analog pain scales have been validated in pediatric populations and are comparable to their paper-based counterparts [37,43]. Additionally, children were asked to identify the location(s) of their pain using a body map that had 7 main areas represented (head, upper body, left hand, right hand, hip and upper legs, right foot, left foot) along with branching options to select more specific locations within each main area; the number of larger pain locations identified was summed for use in analyses.

Pain interference: Pain interference was measured by items taken from the Child Activity Limitations Questionnaire [23,36], which is a validated pediatric measure of level of interference in doing regular activities due to pain. Children were asked to report their extent of difficulty with social activities (e.g., activities with friends and family, group or club activities), physical activities (e.g., sports, walking), and academic activities (e.g., writing, doing schoolwork) due to pain using a 4-point response scale ranging from "Not very difficult" to "Extremely difficult." The total score on the 8 items asked at each assessment period was used for analyses, with higher scores indicating greater difficulty in carrying out typical activities due to pain. Internal consistency (Cronbach alpha aggregated across measurement occasions) for this scale was .84 for the study sample.

Mood: Children were asked to rate the extent to which they were currently feeling each of 10 affective descriptors (5 positive and 5 negative) taken from the Positive and Negative Affect Schedule for Children [26]. A 4-point rating scale ranging from "very slightly or not at all" to "extremely" was provided for each item. Separate scores were computed for the positively and negatively valenced affective descriptors, per the recommended scoring for the PANAS-C. Higher scores indicate higher levels of negative or positive affect. The original PANAS-C and abbreviated (10-item) version have been well-validated for measurement of mood in children [16,26], and the PANAS-C has been used for ecological momentary assessment of mood in prior studies [17]. Internal consistency based on data from the current study was .82 for positive affect and .76 for negative affect.

2.3.3 Baseline measures—Information on child age, child sex (0=female, 1=male), and caregiver education level (6-point scale from "some high school" to "postgraduate degree") was obtained using a questionnaire for the purpose of describing the sample and for evaluating baseline covariates associated with caregiver pain responses. Physician-rated disease activity (using a 0–3 scale from "none" to "severe") was obtained as another potential covariate. Additionally, individual differences in caregiver perception of child vulnerability was measured as a potential covariate associated with caregiver pain response by using the Child Vulnerability Scale [18]. On this questionnaire, caregivers are asked to rate to what extent each of 8 statements is congruent with their perceptions of their child using a 4-point scale ranging from "definitely false" to "definitely true." Sample items include "I sometimes get concerned that my child doesn't look as healthy as s/he should"

and "When there is something going around, my child usually catches it." Studies have supported the internal consistency, concurrent and predictive validity, and convergent validity of the measure in children with arthritis [3, 4], and prospective studies have found scores to be relatively stable over time [46]. Higher scores indicate greater perceived child vulnerability. Internal consistency on this scale for the present sample was found to be adequate (α=.84).

2.4 Analyses

Multilevel models using HLM software [41] were used for primary analyses. This type of analysis accounts for repeated measurements ("Level 1 units") nested within each child-caregiver dyad ("Level 2 units") and is considered the most accurate analysis for the type of data furnished by ecological momentary assessment [2,33]. With 66 dyads each completing up to 90 repeated measurements of the study variables of interest, we had above the minimum sample sizes recommended from simulation studies for adequate power and valid parameter estimation [30]. Missing entries were handled within the models by using maximum likelihood estimation from all available data from each dyad.

2.4.1 Outcomes of caregiver responses to pain—The four caregiver pain response types (protecting, monitoring, minimizing, distracting) were specified in multilevel models as lagged time-varying predictors of current child pain intensity and pain interference (while controlling for prior pain intensity and pain interference scores, respectively, and the baseline covariates of child age and sex, disease severity, caregiver education level, and perceived child vulnerability). Only consecutive moments were evaluated in lagged models. The resulting model coefficients of focus indicated the shift in a child's current pain intensity or pain interference directly following a caregiver's use of one more than their usual number of protecting, monitoring, minimizing, or distracting response to the child's pain.

2.4.2 Variability in caregiver responses to pain—Unconditional mean multilevel models (one-way ANOVAs with random effects) were specified for each of the four caregiver response types in order to derive the variance estimates needed to calculate an intraclass correlation coefficient (ICC). The ICC in this context represents an estimate of the relative proportion of total variance in the given type of caregiver pain response that is static/ trait-like (varying between caregivers only). ICC values below .5 (i.e., < 50%) indicate that most of the total variation is due to *within*-caregiver variance.

2.4.3 Predictors of caregiver responses to pain—Multilevel models were specified with frequency of use of the different types of caregiver responses to pain (averaged over all assessments) as the outcome and forced entry of the baseline covariates (child age, child sex, caregiver education level, physician-rated disease activity, and Child Vulnerability Scale scores). Estimated model coefficients of focus indicated the shift in caregivers' average use of the different responses types for each unit change in the covariate. Significant covariates were retained in models as applicable.

Multilevel models then were specified with simultaneous forced entry of child pain characteristics (intensity, unpleasantness, number of pain locations, and pain impact on activities) as time-varying predictors of the extent to which each caregiver response type was used at a given moment or at a subsequent moment (using lagging of the predictor variables). Similar models also were used with forced entry of child and caregiver negative and positive affect as predictor variables. Coefficients of focus from these models indicated the expected change in how much a caregiver would use a pain response type at or after times when the given predictor variable is one unit higher or lower than the respondent's average level.

3.0 RESULTS

3.1 Descriptive statistics

Caregivers and children on average had complete diaries on 75% of moments (83% of days) and 81% of moments (87% of days), respectively. Caregivers reported some contact with their child on almost all measurement occasions ($\underline{M}$ = 90%; $\underline{SD}$ = 12%). Table 2 presents descriptive statistics for the child and caregiver variables measured by the electronic diary. Child scores for pain intensity, unpleasantness, and interference on average were in the lower range of their respective scales, though standard deviations were fairly high relative to mean values. Both children and caregivers generally reported low levels of negative affect and comparatively higher levels of positive affect.

Figure 1 shows box plots for the percentage of times caregivers reported using each of the four pain response types. Monitoring responses were used most often by caregivers, followed by protecting responses, distracting responses, and minimizing responses. Although in general caregivers rarely reported using minimizing responses, three caregivers reported using them at least one-third of the time.

3.2 Primary analyses

3.2.1 Outcomes of caregiver responses to pain—Table 3 shows the results of multilevel analyses evaluating lagged (prior) caregiver pain response variables as predictors of a child's current pain intensity and pain interference (while controlling for prior pain intensity and pain interference scores, respectively, and the baseline covariates). After moments when caregivers used more protecting responses, child pain intensity and pain interference significantly increased. Following moments when caregivers used more minimizing responses, child pain interference significantly decreased. Caregiver use of distracting and monitoring responses was not significantly associated with subsequent changes in child pain intensity or pain interference.

3.2.2 Variability in caregiver responses to pain—Figure 2 graphically depicts the total variance accounted for in caregiver response types that is between- and within-caregivers, based on ICCs computed from multilevel models. ICCs for the caregiver pain response types ranged from .22 (minimizing) to .48 (monitoring), indicating that only between 22–48% of the total variation in caregiver pain responses was due to relatively fixed

individual differences. Thus, all caregiver pain response types showed evidence of varying more within caregivers (varying from moment to moment) than between caregivers.

3.2.3 Baseline predictors of caregivers' overall use of pain response types—

Table 4 shows the results of multilevel analyses evaluating predictors of caregiver responses to pain. Different baseline covariates were related to different caregiver response types. Specifically, caregiver education was significantly inversely associated with the frequency of using protective responses. Caregivers of male children more frequently used distracting responses, and caregivers of children with higher physician-rated disease activity more frequently used monitoring responses. Greater perceived child vulnerability was found to be significantly positively associated with use of all caregiver response types other than minimizing responses. There was no association between any baseline covariate and frequency of using minimizing responses to pain.

3.2.4 Time-varying predictors of when caregivers' use more or less of the pain response types—With regard to time-varying predictors of caregivers' use of pain responses, caregivers reported significantly more protecting and monitoring responses at and after times when their child reported more pain unpleasantness, more pain interference, and more pain locations. Caregivers used significantly fewer minimizing responses at and after times when their child reported more pain interference with activities. Additionally, caregivers reported using *fewer* protecting responses at and after times when they had higher levels of positive affect; neither caregiver or child affect significantly predicted use of any of the other caregiver pain responses.

4.0 DISCUSSION

The current study extends the literature on caregiver responses to child pain by employing an intensive sampling methodology in a chronic disease population. Caregivers used smartphones to monitor their mood and responses to child pain while their child with JIA contemporaneously reported on their pain and symptoms. By using this methodology, we were able to examine the temporal relevance of caregiver responses to child pain. A key finding was that caregiver use of more protective responses at a given moment predicted a subsequent increase in child pain intensity and pain interference, whereas caregiver use of more minimizing responses at a given moment predicted a subsequent reduction in pain interference for the child.

How caregiver responses to pain translate into changes in child pain outcomes is a clinically important question that has been minimally examined to date in youth with chronic disease. In the case of persistent pain without an underlying disease, protective and monitoring pain responses have been considered problematic by virtue of being inconsistent with the primary treatment goal of improved functioning. Specifically, in youth with idiopathic pain, caregiver protective responses are associated with relatively lower participation of children in academic and physical activities and greater somatic complaints [1,13,28,51]. Results of the current study in youth with JIA demonstrated that when caregivers respond to pain with protective behavior, child pain intensity and pain interference subsequently increases. Thus, our results likewise suggest that caregiver protective responses generally should be

minimized in this population. However, protective responses may still be helpful for other outcomes (e.g., protecting inflamed joints) at moments when increased pain indicates a disease flare. Whether moments of pain in youth with a chronic disease reflects disease activity or some other occurrence, such as normal childhood aches or disordered pain processing, can be difficult to decipher for both caregivers and clinicians [8]. As such, conclusions regarding at what moments caregivers should limit protective responses in youth with a chronic disease requires further investigation.

The current study also found that after caregivers use more *minimizing* responses, pain interference in a child's activities decreases as well. Minimizing responses (i.e., responses involving discounting of the significance of pain) also have been conceptualized as problematic, with some support for this from cross-sectional studies in idiopathic pain showing an association between minimizing responses and greater child disability [e.g., 13]. Overall, caregivers in our sample reported infrequent use of minimizing responses. However, on the occasions that a minimizing response *was* used by a caregiver, it appeared to actually promote a child's subsequent maintenance of regular activity despite pain. This finding may still be interpreted as consistent with operant learning theory, and the fear-avoidance model of pain [21], in that a caregiver's minimizing response may fail to reinforce the child's avoidance of activities due to pain. Perhaps an occasional minimizing response by a parent is sufficiently compelling to alleviate a child's concern about the significance of pain, such that the child carries on with usual activity. Although it is premature to make clinical recommendations on this finding without replication, the finding suggests that in a chronic disease sample there may be a potential benefit of judicious use of minimizing responses.

Another unexpected result of the current study was that distracting and monitoring responses to pain did not change the child's pain intensity or pain interference. This finding contrasts with existing studies that have shown that a caregiver's increased attention to pain increases a child's symptom complaints, whereas distracting responses reduce them [13,49]. In these studies, however, pain is induced through experimental manipulation. It may be that the association of distracting and monitoring parent responses with child pain outcomes are inconsistent and not very robust for the more persistent pain of JIA, or that these responses predict other variables associated with pain such as fear or catastrophizing. Future studies might consider including multiple groups (e.g., well-children with acute pain, otherwise healthy children with idiopathic pain, and children with chronic disease) to more directly evaluate the moderating role of disease presence/absence on which caregiver responses improve a child's pain and other health outcomes.

An additional contribution of the current study was the finding that caregiver responses to pain were variable over time. This finding suggests that how a caregiver responds to pain in a child with chronic disease is partly context-specific. Relevant child factors that predicted how caregivers responded to pain at any given moment were found to be the severity of the child's pain unpleasantness and pain interference. In particular, at or after times when children reported being more bothered by pain and/or more pain interference, caregivers responded with more than their usual amount of protecting and monitoring responses and with less minimizing responses. It may be that child's pain unpleasantness and pain interference are at least in part the contextual clues caregivers use to determine whether to

respond with increased protective responses and attention to pain. These variables are in part behaviorally observable [22,29,40] and thus when at high levels may incite a more urgent response from caregivers. These findings add to our existing work in understanding how child variables improve or exacerbate daily pain and functioning in JIA [6,7,14] by determining that caregiver responses to pain are important to consider as well.

Interestingly, our study also found that caregivers' emotional state predicted their response to their child's pain. In particular, caregivers responded with fewer than usual protective responses at or after times when they reported more positive mood. Prior cross-sectional studies in samples of youth with painful chronic disease have suggested that the most distressed caregivers and those most likely to catastrophize about their child's pain are the caregivers that are most likely to report high levels of protective and monitoring responses [9,25,27]. Results of the present study add to these findings by suggesting that *when* caregivers are experiencing higher levels of positive affect, they may be less likely to respond to their child's pain in a way that subsequently increases pain intensity or interference. This may be due to caregivers feeling less compelled by fear and emotional distress to exclusively respond with increased protectiveness in these instances. Instead, as suggested by theories of positive emotions [19,32], when caregivers experience more positive emotions they may be better able to consider a broad range of response options. We have previously shown that in both children and adults with rheumatologic conditions, how emotions are regulated predicts pain and pain-related disability for that child or adult [14,15]. The current study adds to these data by suggesting that how caregivers regulate their positive emotions may partly determine how they respond to their child's pain, and thereby may predict pain outcomes for the child. By extension, pain outcomes in youth with a painful chronic disease may improve by teaching caregivers strategies that can enhance their positive mood in a given moment. Future interventional research is required to test this hypothesis directly. However, recent work has shown that a cognitive-behaviorally based intervention for caregivers of youth with chronic pain that improved caregiver distress led to a reduction in caregiver protective responses and reduced child pain intensity [34].

Limitations of the present study should be considered when interpreting results. Data were generated from caregiver/child self-report, which may not be an accurate representation of the child or caregiver behavior. For example, some caregivers may have been hesitant to report minimizing responses even if they actually used these, and this may have influenced results. Subsequent studies might consider augmenting self-report data with behavioral observations to increase confidence in the validity of our findings. It also is important to note that the internal consistency of the subscale used to measure minimizing responses to pain was relatively low, as has been reported elsewhere [1,44]. This was likely due in part to only a few items comprising this scale but also can suggest broader issues in how items on this scale are interpreted or relate to one another for this population. One consequence of relatively low internal consistency is a reduction of power to detect associations with other variables. An additional limitation of our study is that we only collected data from one caregiver (typically mothers); different results may emerge from including a different caregiver given that all caregivers may not be consistent with one another in how they respond to child pain [45]. Similarly, we collected data on a small subset of all the variables that potentially could explain the context in which caregivers use more or less of a given

pain response type; future studies might consider expanding the focus to include variables such as parent catastrophizing [10, 25], child catastrophizing [45, 51], fear of pain [21], and accompanying symptoms that may co-occur with the pain. Additionally, although the design of the study enabled us to establish temporal precedence of relationships among caregiver responses and child outcomes, this is insufficient for inferring causal relationships. Indeed, child pain intensity and pain interference did not universally change or always change in the same direction following caregiver protective or minimizing responses.

In conclusion, the use of smartphone-based prospective sampling methodology enabled us to start understanding the context in which caregiver responses to child pain may change and the effect these changes may have on important outcomes for youth with chronic disease. Taking the study findings together, strategies aimed at increasing times of caregiver positive affect and decreasing times of high pain unpleasantness for a child may decrease the number of moments caregivers use protective pain responses; in turn this may help decrease the child's subsequent pain intensity and interference in daily activities. Future studies should consider caregiver responses to pain as at least partly context-specific, and therefore consider employing novel methodologies to examine context in order to better guide caregiver recommendations for youth with painful chronic disease.

Acknowledgments

References

1. Achiam-Montal M, Lipsitz JD. Does parental response to children's pain moderate the association between pain severity and functional disability? An examination of noncardiac chest pain. J Pediatr Psychol. 2014; 39(1):35–44. [PubMed: 24023364]

2. Affleck G, Zautra A, Tennen H, Armeli S. Multilevel daily process designs for consulting and clinical psychology: a preface for the perplexed. J Consult Clin Psychol. 1999; 67(5):746–754. [PubMed: 10535241]

3. Anthony KK, Bromberg MH, Gil KM, Schanberg LE. Parental perceptions of child vulnerability and parent stress as predictors of pain and adjustment in children with chronic arthritis. Child Health Care. 2011; 40(1):53–69.

4. Anthony KK, Gil KM, Schanberg LE. Brief report: Parental perceptions of child vulnerability in children with chronic illness. J Pediatr Psychol. 2003; 28(3):185–190. [PubMed: 12654943]

5. Benarous X, Edel Y, Consoli A, Brunelle J, Etter JF, Cohen D, Khazaal Y. Ecological momentary assessment and smartphone application intervention in adolescents with substance use and comorbid severe psychiatric disorders: study protocol. Front Psychiatry. 2016; 20:157.

6. Bromberg MH, Connelly M, Anthony KK, Gil KM, Schanberg LE. Prospective mediation models of sleep, pain, and daily function in children with arthritis using ecological momentary assessment. Clin J Pain. 2016; 32(6):471–477. [PubMed: 26340651]

7. Bromberg MH, Connelly M, Anthony KK, Gil KM, Schanberg LE. Self-reported pain and disease symptoms persist in Juvenile Idiopathic Arthritis despite treatment advances: an electronic diary study. Arthritis Rheumatol. 2014; 66(2):462–469. [PubMed: 24504820]

8. Bromberg MH, Schechter NL, Nurko S, Zempsky WT, Schanberg LE. Persistent pain in chronically ill children without detectable disease activity. Pain Manag. 2014; 4(3):211–219. [PubMed: 24953073]

9. Caes L, Vervoort T, Devos P, Verlooy J, Benoit Y, Goubert L. Parental distress and catastrophic thoughts about child pain: implications for parental protective behavior in the context of child leukemia-related medical procedures. Clin J Pain. 2014; 30(9):787–799. [PubMed: 24042348]

10. Caes L, Vervoort T, Eccleston C, Vandenhende M, Goubert L. Parental catastrophizing about child's pain and its relationship with activity restriction: the mediating role of parental distress. Pain. 2011; 152(1):212–222. [PubMed: 21126822]

11. Caes L, Vervoort T, Trost Z, Goubert L. Impact of parental catastrophizing and contextual threat on parents' emotional and behavioral responses to their child's pain. Pain. 2012; 153(3):687–695. [PubMed: 22273548]

12. Chambers CT, Craig KD, Bennett SM. The impact of maternal behavior on children's pain experiences: an experimental analysis. J Pediatr Psychol. 2002; 27(3):293–301. [PubMed: 11909936]

13. Claar RL, Simons LE, Logan DE. Parental response to children's pain: the moderating impact of children's emotional distress on symptoms and disability. Pain. 2008; 138(1):172–179. [PubMed: 18221837]

14. Connelly M, Bromberg MH, Anthony KK, Gil KM, Franks L, Schanberg LE. Emotion regulation predicts pain and functioning in children with juvenile idiopathic arthritis: an electronic diary study. J Pediatr Psychol. 2012; 37(1):43–52. [PubMed: 22037006]

15. Connelly M, Keefe F, Affleck G, Lumley M, Anderson T, Waters S. Effects of day-to-day affect regulation on the pain experience of patients with rheumatoid arthritis. Pain. 2007; 131(1–2):162–170. [PubMed: 17321049]

16. Ebesutani C, Regan J, Smith A, Reise S, Higa-McMillan C, Chorpita BF. The 10-Item Positive and Negative Affect Schedule for Children, child and parent shortened versions: application of Item Response Theory for more efficient assessment. J Psychopathol Behav Assess. 2012; 34(2):191–203.

17. Forbes EE, Stepp SD, Dahl RE, Ryan ND, Whalen D, Axelson DA, Birmaher B, Silk JS. Real-world affect and social context as predictors of treatment response in child and adolescent depression and anxiety: an ecological momentary assessment study. J Child Adolesc Psychopharmacol. 2012; 22(1):37–47. [PubMed: 22339611]

18. Forsyth BW, Horwitz SM, Leventhal JM, Burger J, Leaf PJ. The child vulnerability scale: an instrument to measure parental perceptions of child vulnerability. J Pediatr Psychol. 1996; 21(1):89–101. [PubMed: 8820075]

19. Fredrickson BL, Branigan C. Positive emotions broaden the scope of attention and thought-action repertoires. Cogn Emot. 2005; 19(3):313–332. [PubMed: 21852891]

20. Gatzounis R, Schrooten MG, Crombez G, Vlaeyen JW. Operant learning theory in pain and chronic pain rehabilitation. Curr Pain Headache Rep. 2012; 16(2):117–126. [PubMed: 22261987]

21. Gordon JGA, Noel M, Petter M, Packerson HA. Pediatric fear-avoidance model of chronic pain: foundation, application, and future directions. Pain Res Mgmt. 2012; 17(6):397–405.

22. Hadjistavropoulos T, Craig KD. A theoretical framework for understanding self-report and observational measures of pain: a communications model. Behav Res Therapy. 2002; 40(5):551–570.

23. Hainsworth K, Davies W, Khan K, Weisman S. Development and preliminary validation of the child activity limitations questionnaire: flexible and efficient assessment of pain-related functional disability. J Pain. 2007; 8(9):746–752. [PubMed: 17631055]

24. Haverman L, van Oers HA, Maurice-Stam H, Kuijpers TW, Grootenhuis MA, van Rossum MA. Health related quality of life and parental perceptions of child vulnerability among parents of a child with juvenile idiopathic arthritis: results from a web-based survey. Pediatr Rheumatol Online J. 2014; 12:34. [PubMed: 25120411]

25. Langer SL, Romano JM, Mancl L, Levy RL. Parental catastrophizing partially mediates the association between parent-reported child pain behavior and parental protective responses. Pain Res Treat. 2014; 2014:751097. [PubMed: 24579047]

26. Laurent J, Catanzaro SJ, Joiner TE, Rudolph KD, Potter KI, Lambert S, Osborne L, Gathright T. A measure of positive and negative affect for children: Scale development and preliminary validation. Psychol Assess. 1999; 11(3):326–338.

27. Link CJ, Fortier MA. The relationship between trait anxiety and parent-reported pain, solicitous behavior, and quality of life impairment in children with cancer. J Pediatr Hematol Oncol. 2016; 38(1):58–62. [PubMed: 26056789]

28. Logan DE, Simons LE, Carpino EA. Too sick for school? Parent influences on school functioning among children with chronic pain. Pain. 2012; 153(2):437–443. [PubMed: 22169177]

29. Lynch-Jordan AM, Kashikar-Zuck S, Goldschneider KR. Parent perceptions of adolescent pain expression: the adolescent pain behavior questionnaire. Pain. 2010; 151(3):834–842. [PubMed: 20961688]

30. Maas CJM, Hox JJ. Sufficient sample sizes for multilevel modeling. Methodology. 2005; 1(3):86–92.

31. Marszalek J, Morgulec-Adamowicz N, Rutkowska I, Kosmol A. Using ecological momentary assessment to evaluate current physical activity. Biomed Res Int. 2014; 2014:915172. [PubMed: 25126580]

32. Moskowitz JT, Shmueli-Blumberg D, Acree M, Folkman S. Positive affect in the midst of distress: implications for role functioning. J Community Appl Soc Psychol. 2012; 22(6):502–518. [PubMed: 23175617]

33. Nezlek J. Multilevel random coefficient analyses of event- and interval-contingent data in social and personality psychology research. Pers Soc Psychol Bull. 2001; 27:771–785.

34. Noel M, Alberts N, Langer SL, Levy RL, Walker LS, Palermo TM. Sensitivity to change and responsiveness of the Adult Responses to Children's Symptoms in children and adolescents with chronic pain. J Pediatr Psychol. 2016; 41(3):350–362. [PubMed: 26493601]

35. Noel M, Palermo TM, Essner B, Zhou C, Levy RL, Langer SL, Sherman AL, Walker LS. A developmental analysis of the factorial validity of the parent-report version of the Adult Responses to Children's Symptoms in children versus adolescents with chronic pain or pain-related chronic illness. J Pain. 2015; 16(1):31–41. [PubMed: 25451623]

36. Palermo T, Lewandowski A, Long A, Burant C. Validation of a self-report questionnaire version of the Child Activity Limitations Interview (CALI): The CALI-21. Pain. 2008; 139(3):644–652. [PubMed: 18692316]

37. Palermo T, Valenzuela D, Stork P. A randomized trial of electronic versus paper pain diaries in children: impact on compliance, accuracy, and acceptability. Pain. 2004; 107(3):213–219. [PubMed: 14736583]

38. Palermo TM, Valrie CR, Karlson CW. Family and parent influences on pediatric chronic pain: a developmental perspective. Am Psychol. 2014; 69(2):142–152. [PubMed: 24547800]

39. Peterson C, Palermo T. Parental reinforcement of recurrent pain: the moderating impact of child depression and anxiety on functional disability. J Pediatr Psychol. 2004; 29(5):331–341. [PubMed: 15187172]

40. Prkachin KM. Assessing pain by facial expression: Facial expression as nexus. Pain Res Manag. 2009; 14(1):53–58. [PubMed: 19262917]

41. Raudenbush SW, Bryk AS, Congdon R. HLM 7.01 for Windows [Computer software]. Skokie, IL: Scientific Software International, Inc; 2013.

42. Ringold S, Beukelman T, Nigrovic PA, Kimura Y. CARRA Registry Site Principal Investigators. Race, ethnicity, and disease outcomes in juvenile idiopathic arthritis: a cross-sectional analysis of the Childhood Arthritis and Rheumatology Research Alliance (CARRA) Registry. J Rheumatol. 2013; 40(6):936–942. [PubMed: 23588937]

43. Stinson J, Stevens B, Feldman B, Streiner D, McGrath PJ, Dupuis A, Gill N, Petroz GC. Construct validity of a multidimensional electronic pain diary for adolescents with arthritis. Pain. 2008; 163(3):281–292.

44. Van Slyke D, Walker L. Mothers' responses to children's pain. Clin J Pain. 2006; 22(4):387–391. [PubMed: 16691093]

45. Vervoort T, Huguet A, Verhoeven K, Goubert L. Mothers' and fathers' responses to their child's pain moderate the relationship between the child's pain catastrophizing and disability. Pain. 2011; 152(4):786–793. [PubMed: 21272996]

46. Vrijmoet-Wiersma CMJ, Egeler RM, Koopman HM, Bresters D, Norberg AL, Grootenhuis MA. Parental stress and perceived vulnerability at 5 and 10 years after pediatric SCT. Bone Marrow Transplant. 2010; 45:1102–1108. [PubMed: 19881554]

47. Walker L, Claar R, Garber J. Social consequences of children's pain: when do they encourage symptom maintenance? J Pediatr Psychol. 2002; 27(8):689–698. [PubMed: 12403859]

48. Walker L, Levy R, Whitehead W. Validation of a measure of protective parent responses to children's pain. Clin J Pain. 2006; 22(8):712–716. [PubMed: 16988567]

49. Walker LS, Williams SE, Smith CA, Garber J, Van Slyke DA, Lipani TA. Parent attention versus distraction: impact on symptom complaints by children with and without chronic functional abdominal pain. Pain. 2006; 122(1–2):43–52. [PubMed: 16495006]

50. Watson D, Clark L, Tellegen A. Development and validation of brief measures of positive and negative affect: the PANAS scales. J Pers Soc Psychol. 1988; 54(6):1063–1070. [PubMed: 3397865]

51. Welkom JS, Hwang WT, Guite JW. Adolescent pain catastrophizing mediates the relationship between protective parental responses to pain and disability over time. J Pediatr Psychol. 2013; 38(5):541–550. [PubMed: 23471361]

Study synopsis

Caregiver protective responses were found through smartphone-based ecological momentary assessment to predict increases in pain intensity and interference for youth with Juvenile Idiopathic Arthritis.
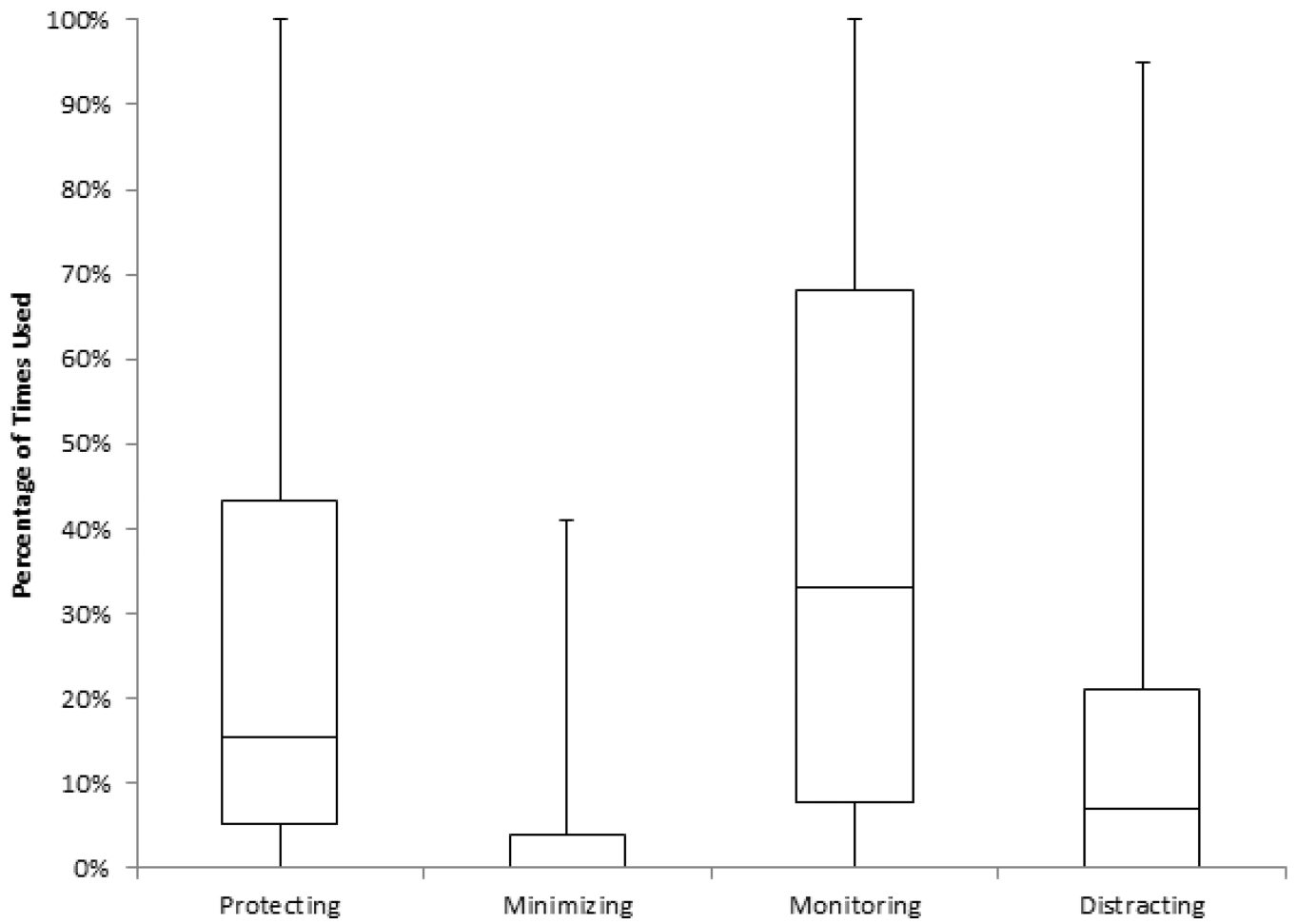
Figure 1.
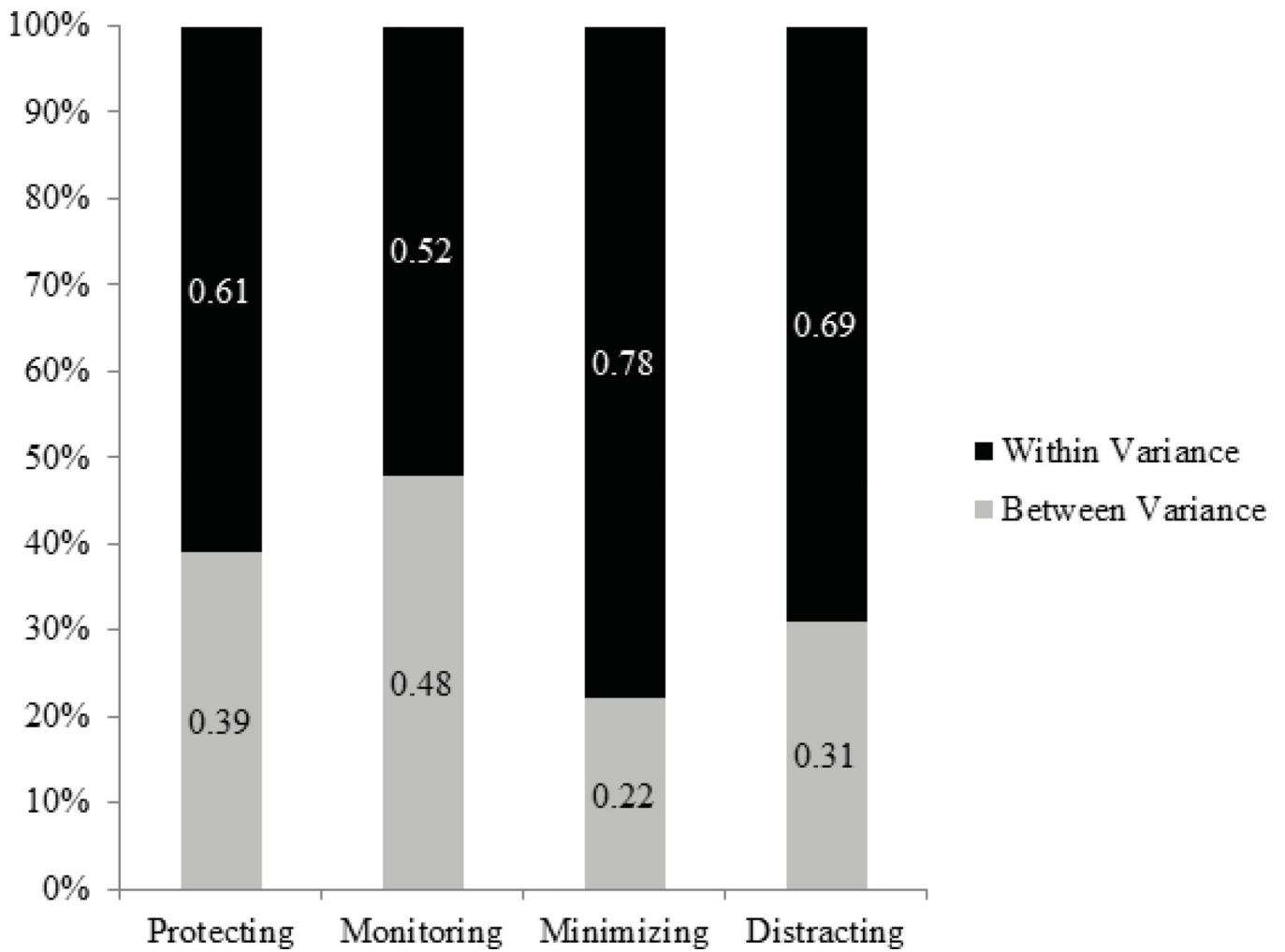Box and whisker plot for caregiver pain response types

Figure 2.
Relative contributions of within- and between-caregiver variance for caregiver pain responses

Table 1

Definitions of caregiver pain response types.

Caregiver Pain Response Type	Definition	Sample Items
Protecting	Provision of special attention or privileges, or assisted reduction of normal responsibilities, in response to the expression of pain or potential for pain	"Give your child special privileges" "Keep your child inside the house"
Monitoring	Use of inquiry and extra vigilance to check on the child's pain status	"Check on your child to see how he/she is doing" "Ask your child questions about how he/she feels"
Minimizing	Verbal discounting of the significance of pain or criticizing the expression of pain as excessive	"Express irritation or frustration with your child" "Tell your child not to make such a fuss about it"
Distracting	Efforts to engage the child's attention in other activities or sensory stimuli besides pain	"Talk to your child about something else to take your child's mind off it" "Try to involve your child in some activity"

Table 2

Descriptive statistics for the child and caregiver electronic diary variables

Variable	Possible Score Range	Observed Mean (SD)
Child pain intensity	0 to 100	26.0 (20.6)
Child pain unpleasantness	0 to 100	26.0 (20.0)
Child pain locations	0 to 7	1.0 (1.1)
Pain interference	0 to 32	6.9 (2.3)
Child positive affect	1 to 4	2.1 (.5)
Child negative affect	1 to 4	1.2 (.2)
Caregiver positive affect	1 to 4	2.7 (.8)
Caregiver negative affect	1 to 4	1.3 (.4)
Proportion of times monitoring responses were used	0 to 1.0	.40 (.33)
Proportion of times protective responses were used	0 to 1.0	.29 (.32)
Proportion of times minimizing responses were used	0 to 1.0	.04 (.09)
Proportion of times distracting responses were used	0 to 1.0	.14 (.19)

Table 3

Results of lagged multilevel analyses for predictors of child's current pain intensity and pain interference.

Lagged Predictor	Current Outcome			
	Pain Intensity		Pain Interference	
	b ± SE	t	b ± SE	t
Protecting	1.96 ± .58	3.34*	.51 ± .21	2.41*
Monitoring	−0.49 ± .52	−.93	.28 ± .19	1.47
Minimizing	−2.50 ± 1.72	−1.46	−1.96 ± .62	−3.16*
Distracting	.62 ± .81	.76	.16 ± .29	.54

Note: Values for b-coefficients represent the expected change in current pain intensity or pain interference associated with the use of one additional protecting, monitoring, minimizing, or distracting response at a prior moment (while controlling for child age and sex, disease severity, caregiver education level, and perceived child vulnerability).

*
$p < .05$.

Table 4

Results of multilevel analyses for predictors of caregiver responses to pain

Predictor	Parent Response Type							
	Protecting		Monitoring		Minimizing		Distracting	
	b ± SE	t	b ± SE	t	b ± SE	t	b ± SE	t
Baseline								
Child age	.01 ± .02	.70	.02 ± .03	.57	.01 ± .01	.90	.00 ± .01	.06
Child sex	.18 ± .13	1.40	−.02 ± .18	−.09	.02 ± .03	.74	.19 ± .07	2.47*
Caregiver education	−.10± .04	−2.45*	−.05 ± .06	−.84	.02 ± .01	1.70	−.05 ± .03	−1.94
Disease activity rating	.13 ± .09	1.45	.28 ± .13	3.00*	.03 ± .02	1.48	.09 ± .05	1.70
CVS Total Score	.03 ± .01	2.75*	.05 ± .02	2.89*	.00 ± .00	.32	.03 ± .01	3.52*
Time-Varying								
Pain intensity	.00 ± .00	.45	.00 ± .00	.93	−.00 ± .00	−1.82	−.00 ± .00	−.01
Pain unpleasantness	.01 ± .00	3.54*	.01 ± .00	3.28*	.00 ± .00	.65	.00 ± .00	1.07
# of pain locations	.03 ± .01	2.19*	.07 ± .01	4.55*	.01 ± .01	1.25	.01 ± .01	.93
Pain interference	.02 ± .00	9.71*	.01 ± .00	5.76*	−.01 ± .00	−3.72*	.00 ± .00	.58
Child Positive Affect	.02 ± .02	1.09	−.03 ± .02	−1.11	.00 ± .01	.23	.02 ± .02	1.10
Child Negative Affect	.04 ± .03	1.29	−.03 ± .04	−.80	−.00 ± .01	−.02	−.03 ± .02	−1.40
Caregiver Positive Affect	−.09 ± .03	−2.74*	.02 ± .04	.56	.00 ± .01	.03	−.00 ± .02	−.11
Caregiver Negative Affect	−.10 ± .07	−1.56	.03 ± .07	.42	−.04 ± .02	−1.78	.04 ± .05	.95

Note: CVS = Child Vulnerability Scale. Values for b-coefficients represent the expected increase in the number of the caregiver responses used on average (for the baseline predictors) or at a given moment (for time-varying predictors) for each unit increase in the predictor variable.

*
 p<.05.