An Electronic Daily Diary Study of Sleep Quality, Pain, and Emotion Regulation in Children with Juvenile Idiopathic Arthritis

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ABSTRACT

MAGGIE HOOD BROMBERG: An Electronic Daily Diary Study of Sleep Quality,
Pain, and Emotion Regulation in Children with Juvenile Idiopathic Arthritis
(Under the direction of Karen Gil)

Children with juvenile idiopathic arthritis (JIA) experience frequent, fluctuating

pain, not fully explained by medical factors. Previous research identifies sleep as an

important predictor of pain and daytime functioning. This study extended inquiry on the

role of sleep in daily pain by employing electronic daily diaries (e-diaries) that allowed

children to report symptoms and behavior in the natural environment 3 times each day.

In addition to replicating previous findings that daily and typical sleep quality predict

pain intensity ($t(2456) = -3.20, p = .001$;  $t(51) = -3.64, p < .001$), this study built on

research on emotions in the pain-sleep association by testing the role of emotion

recovery. The momentary association between sleep quality and pain intensity was

moderated by positive emotion recovery. Consistent with current pediatric pain models,

sleep and pain were also examined as predictors of functional outcomes (i.e., across

aspects of physical, social, and school functioning). Pain intensity mediated the

association between daily and typical sleep quality and functional limitations. Findings

support ongoing, mechanistic research on pain and sleep in children with JIA and a

clinical need to assess sleep and incorporate psychosocial interventions targeting sleep

and promoting positive emotions into the care of children with JIA.

# ACKNOWLEDGEMENTS

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# ABBREVIATIONS

e-diary        Electronic diary

ER        Emotion regulation

JIA        Juvenile idiopathic arthritis

VAS        Visual analog scale

**CHAPTER I**

An Electronic Daily Diary Study of Sleep Quality, Pain, and Emotion Regulation in Children

with Juvenile Idiopathic Arthritis

Juvenile idiopathic arthritis (JIA) is a common childhood illness, affecting

approximately 300,000 children in the United States (Sacks, Helmick, Luo, Ilowite, &

Bowyer, 2007). Children with JIA experience frequent pain, stiffness, and joint

inflammation. JIA is diagnosed before the age of 16, with arthritis in the same joint for at

least 6 weeks, and encompasses heterogeneous disease types distinguished by active arthritis

on physical exam at onset, biological disease markers, and a constellation of accompanying

symptoms (Petty et al., 2004). Over time, JIA can result in joint contractures, growth

abnormalities, and osteoporosis, in addition to daily incidences of recurrent pain, functional

impairment, and stress. There is no known cure for juvenile arthritis; however,

approximately 50% of children achieve remission and do not require ongoing treatments as

adults (Minden et al., 2000). JIA treatments are typically multimodal, incorporating

physical, occupational, and psychosocial interventions along with the use of disease

modifying and anti-inflammatory medications (Anthony & Schanberg, 2005). Recent studies

of JIA treatment indicate more aggressive treatments including a combination of nonsteridal

anti-inflammatory drugs (NSAIDs), disease-modifying drugs, biologics, and corticosteroid

joint injections promote better illness outcomes (Beukelman et al., 2011; Dewitt et al., 2012;

Hashkes & Laxer, 2005). In addition to disease management and together with improved

quality of life, physical functioning, and psychosocial functioning, pain reduction is

recognized as an important JIA treatment outcome (Giannini et al., 1997).

**Juvenile Idiopathic Arthritis**

**Pain in JIA.** Children with arthritis experience frequent pain (Sherry, Bohnsack,

Salmonson, Wallace, & Mellins, 1990) often reaching severe levels (Schanberg, Anthony,

Gil, & Maurin, 2003; Schanberg, Gil, Anthony, Yow, & Rochon, 2005). Indeed, children

with JIA reported pain on 70% of days in a paper diary study (Schanberg et al., 2003) with a

significant number of children (31%) reporting severe pain during the two-month study

period. Despite a better understanding of the fluctuating nature of JIA pain, factors that

predict the onset and maintenance of pain are still not well understood and biomedical factors

do not entirely explain variation in JIA pain. Schanberg and colleagues (1997) found that

medical variables accounted for only 30% of the variance in pain. Malleson et al. (2004)

found that disease activity accounted for only 6.5% of pain variance and a combination of

demographic (e.g., age, gender, race) and disease-related variables (e.g., disease duration,

joint count, morning stiffness) accounted for only 22% of pain intensity variance. Children

with minimal disease activity reported pain on the majority of days (58%) on a paper diary,

suggesting that nonmedical factors may drive day-to-day variability in JIA pain (Schanberg

et al., 2003).

**Functioning in JIA.** In addition to pain, children with JIA experience functional

limitations and emotional distress, despite improved medical treatments. In recent studies,

children with arthritis typically reported mild to moderate functional limitations, such as

difficulty completing physical and self-care tasks, schoolwork, and socializing with peers

(Ding, Hall, Jacobs, & David, 2008; Hyrich et al., 2010). Children with higher functional

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limitations appear to have persistent functional difficulties (Hyrich et al., 2010) and functional limitations fluctuate over time (Magni-Manzoni et al., 2008), similar to fluctuations in JIA pain (Schanberg et al, 2003, 2005). Based on results of questionnaires administered at a clinic visit, Sawyer and colleagues (2004) found that higher pain intensity was significantly associated with more difficulties in self-care, completing physical tasks (e.g., walking up stairs), and participating in activities in children with arthritis aged 8-18 years. Fluctuations in JIA pain also predict changes in day-to-day functioning. In a previous paper diary study (Schanberg et al., 2003), daily stress and mood predicted higher pain, which was associated with more reductions in activities. However, these findings are somewhat limited by the use of end-of day reporting practices, which may be affected by retrospective bias and could be influenced by current symptom levels.

In order to address this limitation, our research group has moved to using in-the-moment reports of pain, symptoms, and functioning. In our pilot study of electronic diary (e-diary) assessment of daily symptoms and functioning of children with arthritis (Connelly et al., 2010), average pain intensity was highly correlated with overall reductions in school, social, and physical activities ($r = .78$), suggesting that pain and functioning are highly associated and that this relationship warrants ongoing prospective research. A better understanding of the ways in which pain and functioning are related has implications for pain management and psychosocial interventions. For example, if a third variable is identified which relates to both pain and functioning, this would suggest new approaches for interventions to promote decreased pain and improved functioning.

**Emotional adjustment in JIA**. In addition to physical and social functioning, emotional adjustment has been associated with pain and JIA. LeBovidge and colleagues

(2003) conducted a meta-analysis of 21 studies of emotional adjustment in children with JIA, which employed retrospective psychological measures that resulted in overall psychological adjustment scores, internalizing symptoms scores, and externalizing symptoms scores. The majority of the studies used the Achenbach Child Behavior Checklist (Achenbach & Edelbrock, 1983); specific measures of depression or anxiety were not included in the analysis. When reports from parents of children with JIA were compared to those from parents of healthy children, children with JIA were at increased risk for internalizing symptoms (LeBovidge et al., 2003).

Building on this finding, LeBovidge and colleagues (2005) examined correlates of anxiety and depressive symptoms in children with JIA aged 8-17 years. Results indicated that higher illness-related stress reported in semi-structured interviews (e.g., problems with school, peers, or family) was associated with higher symptom scores on the Beck Depression Inventory for Youth (Beck, Beck, & Jolly, 2001) and the Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1985). Additionally, a more positive attitude towards illness, as measured by the Child Attitude Toward Illness Scale (Austin & Huberty, 1993) buffered the effects of higher stress on depressive symptoms. Based on these studies, children with arthritis face pain and accompanying physical and psychosocial challenges. As such, there is an ongoing need to identify predictors of pain in JIA beyond medical indices.

**Biobehavioral Model of Pediatric Pain**

The Biobehavioral Model of Pediatric Pain (Varni et al., 1996) provides a conceptual structure for testing the proposed pattern of associations between pain, sleep, emotion regulation, and functional limitations (e.g., difficulties in physical, school, and social tasks) in this study. The model also identifies the broader need to consider factors beyond medical

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indices affecting pain and functional outcomes. Consistent with a biopsychosocial approach, the Biobehavioral Model of Pediatric Pain is a dynamic model that encompasses reciprocal influences between pain precipitants. Aspects of the model have been tested in a variety of pediatric populations, including JIA (e.g., Sawyer et al., 2004). The Biobehavioral model suggests that precipitants, such as disease and stress, predict pain and functioning. These associations may be mediated or moderated by intervening variables, which include cognitive appraisals and coping strategies. Functional outcomes in the model include activities of daily living, school attendance, and behavioral and adjustment problems. Of importance, pain and functioning are conceptualized as sharing bidirectional influences. Broadly speaking, the variables in this study mapped on to the Biobehavioral model. Poor sleep quality can be conceptualized as a daily stressor, thus serving as a pain antecedent. Emotion regulation is similar to emotion-focused coping, which is included in the model as an intervening variable. Finally, consistent with the model, the key outcomes of the study are pain and functioning. An overall functional limitations score was used in this study, which included items related to physical, school, and social functioning.

**Evidence supporting the Biobehavioral Model of Pediatric Pain.** Several empirical studies have tested aspects of the Biobehavioral Model of Pediatric Pain. In a study of 5- to 18-year-old children with cerebral palsy (Berrin et al., 2007) results of path analyses showed that pain and fatigue were intermediaries between disease severity and school functioning. White and colleagues (2006) found that the Biobehavioral Model also applied to healthy children; in a community sample, the association between life stress (e.g., witnessing violence) and pain (e.g., headache, abdominal pain) was largely mediated by anxiety.

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Aspects of the Biobehavioral Model have been supported in pediatric chronic pain populations. A series of studies (Gil et al., 2003; Gil et al., 2000) examined predictors of pain and functioning in children and adolescents with sickle cell disease (SCD) using paper-and-pencil diaries. Parents of children aged 6-17 years completed daily diaries of child symptoms and behaviors for 14 days. Results indicated that children reduced participation in activities when experiencing pain, but disease severity did not predict pain or activity reductions (Gil et al., 2000). In the second study, in which adolescents completed daily diaries for up to 6 months (Gil et al., 2003), stress and positive mood predicted same-day pain, but did not predict pain on the next day. In contrast, pain predicted stress and mood in subsequent days. Increased pain, stress, and negative mood predicted more cutbacks in activities and increased positive mood was associated with fewer limitations in functioning. Findings from these studies suggest a complex temporal association between pain, functioning, stress, and mood, indicating that a biobehavioral approach is necessary for understanding pediatric chronic pain.

Using the Biobehavioral Model of Pediatric Pain as a conceptual framework, Sawyer and colleagues (2004) examined pain and coping strategies as predictors of functional outcomes in children with arthritis aged 8-18 years. Based on child reports, controlling for pain, coping strategies predicted aspects of functioning with medium to large effect sizes; seeking social support as a coping strategy predicted improved emotional ($r = -.56$, $p < .001$) and social functioning ($r = -.38$, $p < .01$). These studies suggest that the Biobehavioral Model of Pediatric Pain (Varni et al., 1996) is a valid and useful conceptual framework for understanding the pattern of associations between variables related to chronic pain in children.

An important feature of the Biobehavioral Model is the focus on functional outcomes; many studies highlight the importance of functioning in children with chronic illnesses and pain. Children with chronic pain consistently endorse high levels of physical disability, with estimates ranging from 75-91% of children reporting moderate to high physical impairment across studies using the Functional Disability Index (Gauntlett-Gilbert & Eccleston, 2007; Susmita Kashikar-Zuck et al., 2008; Walker & Greene, 1991). Similarly, children with chronic pain reported moderate difficulties in social functioning and activity participation (L. L. Cohen, Vowles, & Eccleston, 2010; Palermo, Witherspoon, Valenzuela, & Drotar, 2004). Based on academic records, Logan and colleagues (2008) found that 44% of adolescents with chronic pain missed greater than a quarter of school days, with similar rates of late arrivals and early dismissals. Via daily diaries, children with chronic pain reported more difficulty completing activities (e.g., walking, socializing, or participating in afterschool activities) on days they experienced higher pain intensity (Lewandowski, Palermo, Kirchner, & Drotar, 2009). These studies provide ample evidence that children with chronic pain experience significant functional limitations; improved understanding of mechanisms contributing to poor functioning is still needed.

In summary, the Biobehavioral Model of Pediatric Pain posits that complex associations between biological, psychological, and social factors contribute to pain and functioning. Research supports the utility of this conceptual approach in understanding pediatric pain. Moreover, the model emphasizes the importance of looking beyond medical indices in order to understand the etiology and consequences of pain. Stress, coping, and psychological adjustment have been examined as predictors of pediatric chronic pain in many past studies, but there remains unexplained variance in these models.

Recently, sleep has been identified as a significant problem for children with chronic pain (Chambers, Corkum, & Rusak, 2008). There is evidence that sleep may relate to both pain and functioning (Dahl, 1996; Dahl & Lewin, 2002; Lewin & Dahl, 1999; Sadeh, Gruber, & Raviv, 2002). As such, sleep quality served as the primary predictor of pain and functioning in this study.

**Sleep**

**Evidence for studying sleep in pediatric chronic pain.** Sleep problems have been identified across pediatric chronic pain groups.  In a study using parent-reports of sleep problems, children with headache had more difficulty initiating sleep, shorter sleep duration, and poorer sleep quality in comparison to healthy peers (Bruni, 1997).  Similarly, based on parent- and self-reports of sleep problems, children aged 8-15 years with functional abdominal pain (FAP) experienced more difficulty initiating and maintaining sleep (Huntley, Campo, Dahl, & Lewin, 2007) and significantly worse overall sleep quality than healthy controls.  In another study only 25% of children with FAP rated their sleep quality as good on daily sleep logs (versus 87% of the control group), despite no significant between group differences on sleep actigraphy (Haim et al., 2004).  In a mixed sample of adolescents with chronic pain conditions including sickle cell disease, headache, and JIA, Palermo and Kiska (2005) found a small, positive correlation between pain duration and bedtimes; depression and functioning had moderate, positive correlations with daytime sleepiness and sleep/wake problems on the School Sleep Habits Survey (Wolfson & Carskadon, 1998).  Taken together, there is support for studying sleep in children with chronic pain; research suggests that subjective aspects of sleep (e.g., sleep quality) may be important to assess even in the absence of objective sleep disturbances.  Additionally, more research is needed to progress

from describing sleep disturbances to understanding the influence of sleep problems on outcomes such as pain and daytime functioning in children with chronic pain.

**Sleep and JIA.** In one of the first studies of sleep in JIA, Zamir and colleagues (1998) found children with JIA experienced more sleep problems than healthy controls on polysomnography (PSG), thus laying a foundation for additional sleep research in this population. A few studies have examined the association between sleep, pain, and functioning in children with JIA, but the results are somewhat inconsistent across studies, due to methodological differences. Using a standardized sleep questionnaire, Bloom et al. (2002) found that parents of children with JIA reported higher total sleep problems scores and endorsed more night awakenings, parasomnias, sleep anxiety, sleep disordered breathing, and early morning waking/daytime sleepiness than parents of healthy controls. Children's pain ratings and overall sleep reports were strongly correlated (Bloom et al., 2002), but indices of disease severity and functioning were not associated with subjective sleep reports. Using a 2-night PSG protocol in 9- to 17-year-olds with and without JIA, Passarelli et al. (2006) found significant between-group differences in sleep efficiency, arousals, and leg movements, but parent-reported sleep problems were not different. Within the JIA group, those with higher functional limitations experienced more frequent arousals and decreased sleep efficiency on PSG during the night. Higher pain was correlated with more alpha-delta activity, which they interpreted to indicate decreased arousal thresholds.

Recently, Ward and colleagues conducted a series of studies examining sleep disturbances and aspects of daytime functioning in children with JIA. Controlling for age and medications, neither anxiety or evening pain significantly predicted sleep arousals on PSG at night (Ward et al., 2008). Children with active JIA experienced longer sleep latency than

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those with inactive disease, but did not differ on motor speed, reaction time, and sustained

visual attention (Ward et al., 2010). Children with JIA did not perform differently from

controls on neurobehavioral tasks, despite higher self-reported sleep disruption scores (Ward

et al., 2011). Taken together, children with JIA appear to have abnormal PSG findings, but

these sleep architectural differences do not predict impairment in neurocognitive functions

typically affected by sleep deprivation or disruption.

Ward and colleagues' studies (2010, 2011) examined the association between sleep

and specific aspects of daytime neurocognitive functioning that sleep is known to affect (e.g.,

response speed), but it remains to be seen how sleep affects functioning across the domains

most salient to children with JIA (e.g., social, physical, emotional functioning). Ward's

methodologies also may not capture the full range of sleep and pain problems children with

JIA experience from day-to-day. Indeed, the average evening pain rating for Ward and

colleagues' (2008) study was quite low (approximately 1 on a scale of 0-10) and children

experienced good sleep efficiency, on average. It is important to implement study designs

that can examine the temporal associations of sleep and pain under naturalistic settings in

order to better understand the role of sleep problems in the lives of children with JIA.

Consistent with broader research on children with chronic pain, research on sleep in

JIA provides convergent evidence that children with arthritis experience behavioral sleep

problems and abnormalities in underlying sleep architecture. Due to employed

methodologies, these studies have not addressed the ways sleep and pain may be directly

related to one another, or to other important aspects of the disease experience (e.g., emotional

functioning and functional limitations), over time. In their influential review of sleep in

pediatric pain, Lewin and Dahl (1999) suggested that pain may directly contribute to sleep

disruptions;  sleep problems in turn may result in emotional or functional difficulties that affect daytime pain.  Therefore, it is important to understand both pathways of influence by applying prospective methodologies and advanced statistical techniques.

**Temporal associations between sleep and pain.** Daily diary studies have tested the cyclical association between pain and sleep, based on Lewin and Dahl's (1999) conceptual framework.  Lewandowski et al. (2010) examined sleep in adolescents with chronic pain.  On actigraphy, sleep duration and time spent awake after sleep onset predicted next-day pain; e-diary reports of sleep quality did not predict pain.  Daytime pain did not predict sleep at night as measured objectively or subjectively.  In contrast to these findings, a cyclical relationship between pain and sleep was detected using paper diaries in Valrie et al.'s (2007a, 2007b, 2008) studies of children with SCD.  Daily mood also played an important role in the relationship between pain and sleep in children with SCD (Valrie et al., 2008).  Negative mood partially mediated both the relationship between high pain and poor sleep quality and between poor sleep quality and high pain the next day. Thus, increased negative mood underlay both directions of association between poor sleep and high pain. In contrast, positive mood buffered the effects of poor sleep quality on pain the next day, but did not buffer sleep quality at night from high pain during the day.

The previous Bromberg et al (2011) study prospectively examined sleep and pain in children with JIA between the ages of 8-16 years using paper daily diaries.  Self-reported sleep quality predicted pain intensity the next day, but daytime pain did not significantly predict sleep quality at night, suggesting that the cyclical relationship may not hold in the children with JIA.  Daily mood was assessed on the same scale used by Valrie et al. (2008), which prompted children to select the face that best reflected their mood for the day on a

scale ranging from a very happy face to a very sad face. Daily mood moderated the association between sleep and pain; the effects of poor sleep quality on pain were buffered as mood became more positive. This finding provides preliminary evidence that emotional functioning is important in the context of sleep and pain.

Surprisingly, the majority of the variance in pain lay at the between-children level, indicating that stable factors predicted much of the variance in pain. Sleep quality accounted for nearly half (49%) of the between-child variance in pain, whereas daily sleep quality only accounted for 10% of the total within-child (daily) variance in pain. These findings indicate that individual differences in overall sleep quality, rather than daily variations in each child's sleep experiences, better explain the relationship between sleep quality and pain. In other words, children who consistently experienced poor sleep quality also experienced higher pain across the study period.

Despite these findings, it remains important to more closely examine the temporal relationship of sleep and pain in JIA for several reasons. First, to attempt to replicate findings regarding the structure of the daily relationship between sleep and pain in JIA. Ongoing failure to detect a cyclical relationship would suggest a unique structure to the pain-sleep relationship in JIA and that findings may not generalize across pediatric pain groups. Second, additional prospective research is warranted because prospective data remain more ecologically valid than retrospective reports. Variables that fluctuate over time are best measured prospectively in children's typical environment, even if the association between pain and sleep continues to be explained at the between-child level.

An important limitation of the previous study was the use of end-of-day reports, completed each evening before bed. End-of-day reports are prone to backfilling and

retrospective bias (Takarangi, Garry, & Loftus, 2006).  The sleep quality item may be especially vulnerable to biases, because it asked the child to report on sleep quality for the previous night.  Intervening events or daytime pain may have affected the child's memory of sleep quality.  Therefore, it was important to attempt to replicate these findings using daily e-diaries, which have the added benefits of cued reporting, assessment of current symptoms, and limited potential for backfilling (Shiffman, Stone, & Hufford, 2008).

Additionally, the Facial Affective Scale (McGrath, de Veber, & Hearn, 1985; McGrath et al., 1996) used as a mood measure in previous studies limits the interpretation of findings (Bromberg et al, 2011; Valrie et al., 2007a, 2007b, 2008).  Using a univariate mood measure limits the potential to detect the unique contributions of negative and positive mood, which predict different health outcomes.   Moving to frequent, in-the-moment data capture allows examination of within-day variation in emotions, rather than relying upon the more temporally stable construct of daily mood.  Emotion researchers call for examining emotion process variables (Silk, Steinberg, & Morris, 2003) to capture emotional processes as they occur.  In this study, emotion regulation was measured as a dynamic variable in order to extend our understanding of how affective processes contribute to the pain-sleep relationship and to establish additional support for the need to develop emotion-focused components of broader psychosocial pain interventions.

**Emotion Regulation**

As previously discussed, daily affective experiences may contribute to the pain and sleep of children with chronic illness.  There is evidence that daily mood affects the relationship between sleep at night and pain during the day in children with chronic pain (Bromberg et al., 2011; Valrie et al., 2008).  However, these findings are limited by the use

of a single daily mood assessment, which may not capture the complexities of affective experiences over time; therefore, this study examined the role of emotion regulation in the context of sleep and pain in children with JIA using e-diaries collected across the course of each day.

**Defining emotion regulation.** Emotion regulation (ER) is a dynamic process encompassing both effortful and automatic processes aimed at the modulation of emotions. Emotion regulation is part of a broader constellation of regulatory processes, including behavioral, cognitive, and physiological regulation (Calkins & Fox, 2002; Calkins & Marcovitch, 2009), which may be considered a subset of coping when emotions are modified specifically in response to stress (Compas, 2009). Despite a lack of consensus, many researchers employ Thompson's (1994) definition of ER as "the extrinsic and intrinsic processes responsible for monitoring, evaluating, and modifying emotional reactions, especially their intensive and temporal features, to accomplish one's goals" (p. 27-28). In Thompson's definition, ER broadly encompasses many processes related to emotional reactions that follow a developmental trajectory. Changes in regulatory strategies and resources occur in conjunction with neurophysiological, social, and cognitive development (Zeman, Cassano, Perry-Parrish, & Stegall, 2006), thus it is important to consider age in the context of ER.

A major criticism of Thompson's model of ER is the potential difficulty distinguishing emotion activation (i.e., the onset of an emotion) and regulation (Campos, Frankel, & Camras, 2004). Hoeksma et al.'s (2004) conceptual and methodological framework identifies ER as a dynamic process; changes in emotion intensities across reports can be used to measure ER distinctly from emotion activation. Recent prospective research

(Connelly et al., 2012; Silk et al., 2011) used multiple assessments to capture changes in

emotion intensity over time as aspects of ER (emotional maintenance and recovery). These

temporal ER constructs do not capture other broader aspects of the ER process such as

emotion activation prevention, unsuccessful effortful attempts at regulating emotions, or the

use of specific ER strategies.  Consistent with these studies and nested within the broader

array of ER processes, the present study specifically examined the ER construct of *emotion

recovery*, broadly defined as the process of modifying emotion, over time, in the adaptive

direction (i.e., increasing positive emotions and decreasing negative emotions).  Thus, the

distinction between emotion activation and regulation is fundamental to the construct of

emotion recovery used in this study.

**ER and psychological adjustment in children.** ER is considered an important factor

in the development of childhood psychopathology (Cicchetti, Ackerman, & Izard, 1995;

Cicchetti & Toth, 1995); failure to adaptively respond to, and modulate emotions occurs in

depression, anxiety, and oppositional defiant disorder (Cole, Michel, & Teti, 1994).  Despite

an implicit aim to improve ER (e.g., decrease feelings of sadness and increase positive

emotions in depression interventions) many cognitive behavioral therapies (CBT) promote

change in emotions and ER through cognitive or behavioral techniques, rarely focusing upon

teaching specific ER skills as a key treatment component.  For example, in depression

treatments, children are taught to schedule pleasant activities as a form of behavioral

activation, leading to improved mood without directly teaching the child to respond to

changes in emotion as they occur.  Recent reviews emphasize the need to incorporate

emotion-focused components in child therapy (Hannesdottir & Ollendick, 2007; Suveg,

Southam-Gerow, Goodman, & Kendall, 2007; Trosper, Buzzella, Bennett, & Ehrenreich,

2009) and results of the few treatment studies directly employing ER skills (Kovacs et al., 2006; Trosper et al., 2009) support their use for children and adolescents with internalizing disorders.  In Kovacs and colleagues' (2006) CBT study, ER strategies of self-talk and seeking social support were applied in response to increased negative emotions.   Post treatment, 53% of children had full remission; 92% of children met remission criteria at 12-months (Kovacs et al., 2006).  Comorbid anxiety and major depression were greatly decreased in children who completed the intervention, suggesting that improved ER may generalize across internalizing symptoms.

Although learning to successfully identify and regulate emotions is a developmental task faced by all children, children with arthritis experience the daily challenges of managing pain, stiffness, and fatigue (Schanberg et al., 2003, 2005) in addition to typical childhood experiences of negotiating family, peer, school, and developmental demands.  These unique stressors may result in emotional burdens above what is typical for same-aged peers and children with JIA have been shown to be at somewhat increased risk for internalizing problems (e.g., depressive symptoms and anxiety; LeBovidge et al., 2003).  Based on the daily burdens of living with JIA and the potential for internalizing symptoms, the ability to regulate emotions may be particularly important in the context of JIA.  Research in juvenile arthritis has found associations between daily mood, sleep, and pain (Schanberg et al., 2005; Bromberg et al., 2011), but the mechanisms associating emotions and health-related outcomes remain unclear.

Findings from intervention studies provide support for promoting ER in children with psychopathology, but the role of ER in children facing ongoing significant stress from chronic illness remains to be clarified.  Furthermore, although the developmental

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psychopathology framework broadly encompasses normal and abnormal behavior and seeks to identify risk and protective factors, little research has explored the protective potential of ER in the absence of psychopathology. In other words, it remains unknown whether ER helps children adapt more successfully even when they do not meet criteria for a disorder such as depression or anxiety.

Despite accruing evidence of the importance of ER during childhood, confusion remains regarding basic definitions and models of emotion-related constructs. Thus, it is important to define terms and review conceptual models from the broader affective literature in the context of the current study. The construct of the emotion recovery ER variable in this study was based on conceptual affective models.

**Emotions, mood, and affect.** Although "emotion" and "mood" are sometimes used interchangeably, each term describes a set of related, yet distinct constructs with biological, behavioral, and cognitive components. Emotions are intense, discrete experiences with identifiable onsets, peaks, and durations. Emotions are experienced in direct response to environmental stimuli (Parkinson, Totterdell, Briner, & Reynolds 1996), are highly malleable (Gross & Thompson, 2007; Zalewski, Lengua, Wilson, Trancik, & Bazinet, 2011), and ideally, adapt in order to enhance performance or to obtain desirable outcomes (Thompson, 1994). For example, a child may feel sad after breaking a toy, but the sadness does not persist after the child begins coloring. In contrast to the discrete, fluctuating nature of emotions, mood is a more stable state without a distinct onset, which Larsen (2000) likens to the "backdrop against which the rest of our psychology gets played out in daily life" (p. 130). Unlike emotions, mood is not necessarily contingent upon the immediate situation. For

example, a depressed child may experience long periods of sadness, without an identifiable triggering event.

Emotions and mood can be understood within the broader construct of affect, the experienced *tone* of emotions and mood. Affect consists of both varying intensities of negatively and positively valenced emotions; affect is how strongly negative or positive a person feels. Affective measures, such as the Positive and Negative Affect Schedule (Watson, Clark, & Tellegen, 1988) assesses the intensity of discrete negative emotions (e.g., sadness, anger) and positive emotions (e.g., happiness, excitement) to obtain composite affective valence scores. Affective measures can be used to assess both mood and emotions, depending on wording of the instructions. Ratings of current, in-the-moment affect measure emotions and broader retrospective reports of affect (e.g., global ratings for a day) constitute mood. As such, the assessed duration and intensity of emotional experiences primarily distinguishes mood and emotions.

**Affective paradigms.** Although is it generally agreed that positive affect is comprised of emotions characterized by pleasant engagement (e.g., happiness, excitement) and negative affect is characterized by unpleasant and distressing emotions (e.g., anger, sadness), the relationship between negative and positive affect has long been debated (Barrett & Russell, 1998; Russell & Carroll, 1999; Watson et al., 1988; Watson & Tellegan, 1999). The disagreement centers around the independence of negative and positive affect; bivariate models identify negative and positive affect as separate, independent constructs and bipolar models position negative and positive emotions as opposites ends of a continuous spectrum. As Reich and colleagues (2003) summarize in their review of affective models, bivariate and bipolar models have both been supported by studies using factor analytic techniques (Green,

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Salovey, & Truax, 1999; Tuccitto, Giacobbi, & Leite, 2010), and in the context of negative and positive life events (Gable, Reis, & Elliott, 2000; van Eck, Nicolson, Berkhof, & Sulon, 1998). Furthermore, findings from studies using techniques to physiologically capture emotional arousal also support each affective paradigm (Lane et al., 1997; C. Wang et al., 1996). Based on the wealth of evidence for both models, it is possible that the structure of affect may be more complex than either model and may vary on other conditions.

Recently, theorists have proposed models that reconcile the two predominant approaches. Cacioppo and Berntson's (1994) Evaluative Space Model suggests that affective experiences often function in a bipolar manner; however, mixed emotional states are possible because negative and positive emotions are independently activated. The Dynamic Model of Affect (DMA, Reich et al., 2003) also suggests that the bivariate and bipolar structures may apply in different circumstances, with emotions becoming more bipolar in times of stress or pain. Both models have been supported in applied research (J. T. Larsen & McGraw, 2011; J. T. Larsen, McGraw, & Cacioppo, 2001; J. T. Larsen, To, & Fireman, 2007), including studies of adults with chronic pain (Davis, Zautra, & Smith, 2004; Potter, Zautra, & Reich, 2000).

In order to best understand the relationship between negative and positive affect in a given context, Reich et al. (2003) recommend using a 2-factor affective assessment method, such as the PANAS (Watson et al., 1988). This approach allows researchers to determine the affective structure in their own data, without imposing bipolar affective constructs a priori. Based on this recommendation, items from the child version of the PANAS (Laurent et al., 1999) were used in data collection in order to measure emotion intensity, to identify times

when emotions were activated (i.e., when emotions were elevated above the child's typical level), and to construct the ER variable.

**ER and affective valence.** Consistent with current affective models, both emotional valence and intensity are important to consider in the context of ER. Indeed, ER may be distinguished from the related construct of coping in part via the emphasis on modifying both negative and positive emotions (Gross & Thompson, 2007), as well as the different role of stress in coping and ER. Coping behaviors occur following appraisals of stimuli as harmful, threatening or challenging (i.e., as stressful; (Lazarus & Folkman, 1984); thus, *stress* must be present in order to elicit a coping response aimed at decreasing accompanying *negative* emotions. Compas (2009) proposes that ER functions as a subset of coping when emotions are regulated specifically in response to stress. More broadly, ER may occur in response to changes in emotions, with or without an identified stressor, and incorporates the adaptive modification of positive emotion. For example, if a boy and girl compete for an award and he loses, he may benefit from the ability to experience genuine happiness for her success, which in turn may help to strengthen the friendship. Despite the potentially adaptive function of increasing negative emotions (e.g., arousing anger during a confrontation) or decreasing positive emotions (e.g. dampening joy upon hearing sad news), ER is typically studied in the context of decreasing negative emotional intensity and increasing positive emotional intensity (R. J. Larsen & Prizmic, 2004), consistent with the construct of emotion recovery used in this study.

Much of the current applied research on ER in children and adolescents has focused on adaptively decreasing negative emotions. Most standardized measures of child ER assess anger or sadness (Suveg & Zeman, 2004; Zeman, Shipman, & Penza-Clyve, 2001) and

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clinical research has largely focused on maladaptive or unsuccessful negative ER as a vulnerability to psychopathology. The use of maladaptive ER strategies (e.g., emotional suppression, experiential avoidance) has been associated with internalizing disorders in adults and difficulties regulating anger and sadness have been linked to internalizing and externalizing symptoms in children (Eisenberg et al., 2001; Suveg, Hoffman, Zeman, & Thomassin, 2009). Kovacs et al.'s (2006) ER-focused intervention taught children with mood disorders to use ER strategies to quickly decrease high intensity negative emotions as they occur. As a hypothetical example, if a child began to experience sadness, she could talk to a friend or play a game to try to decrease her sadness. She successfully regulated her emotions if she felt less sad after trying these strategies. Kovacs and colleagues' (2006) ER-focused intervention greatly decreased children's symptoms across comorbid internalizing disorders, suggesting that improved regulation of negative emotions contributed to changes in overall adjustment. The focus on regulating negative affect in previous research is not surprising, given evidence that negative experiences and emotions are stronger than positive experiences or emotions (Baumeister, Bratslavsky, Finkenauer, & Vohs, 2001).

**Positive emotions, resiliency, and health.** Positive affect, however, may be uniquely important for promoting adaptation and improved health-related outcomes in children. Positive psychology departs from the historic emphasis on psychopathology in applied psychology and fundamentally seeks to identify factors that promote well-being, adjustment, and optimal health (Seligman & Csikszentmihalyi, 2000). In Fredrickson's broaden-and-build theory (Fredrickson, 1998b, 2001), positive emotions broaden attention, allowing exploration and creativity. This in turn helps to build social, cognitive, psychological, and physical resources, which may provide more resilience to stressors or

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improve coping in response to stress. As an illustration of this theoretical model, Tugade and Fredrickson (2004) found that individuals with higher positive emotions also had higher trait resiliency. Additionally, following a stress induction task, more resilient individuals experienced shorter cardiovascular recovery durations and benefitted more from the use of positive emotions in the physiological stress recovery process. The relation between positive emotions and resiliency has also been demonstrated longitudinally. Over a 1-month period, higher daily positive affect predicted increased resilience in college students, suggesting that positive emotions helped to build new coping resources over time. Increased life satisfaction was related to higher daily positive affect, but was not associated with lower daily negative affect, and the unique effects of positive affect remained despite accompanying high negative affect (Cohn, Fredrickson, Brown, Mikels, & Conway, 2009). Fredrickson (1998a) also specifically identified the need to promote positive emotions in children, especially through parental cultivation, which could increase coping skills and social competencies.

There is evidence of unique, direct effects of positive emotions on a wide variety of health-related outcomes (S. Cohen & Pressman, 2006). For example, in a prospective study of adults with RA, Strand and colleagues (2006) found that positive affect buffered the relationship between pain and negative affect, thus promoting resiliency during times of increased pain. The role of positive affect may also vary by pain population. In one study (Zautra et al., 2005), women with fibromyalgia experienced negative affect comparable to women with RA, but the role of positive affect varied by pain diagnosis. Compared to women with RA, women with fibromyalgia experienced lower levels of positive affect overall and experienced greater difficulty sustaining positive affect during weeks with higher

interpersonal stress, suggesting that they may have fewer opportunities to benefit from the protective benefits of positive affect.

Findings from previous daily diary studies with children with SCD and JIA highlight the benefits of positive affect in children with chronic pain. Gil et al. (2003) found that positive mood was related to a variety of desirable outcomes including increased activity participation and decreased pain and health care utilization for adolescents with SCD. Building upon this research, Valrie and colleagues (2008) found that increasingly positive mood buffered the effects of poor sleep at night on pain the next day in children with SCD. This finding was replicated in our research with children with arthritis (Bromberg et al., 2011), suggesting that the protective potential of positive affect is seen across pediatric pain populations and may be especially important in the context of sleep difficulties. However, these studies were limited by the use of once-daily mood measures that do not capture the fluctuating nature of emotions as they occur over time, which may be important in the context of pain and functioning. Therefore, it is important to extend this research by examining emotion and ER as they occur, in relation to aspects of JIA that have been associated with the more temporally stable construct of mood.

**Emotion regulation research using ecological momentary assessment.** This study built upon previous findings regarding the importance of affect in the daily pain (Schanberg et al., 2005) and sleep (Bromberg et al., 2011) of children with arthritis by incorporating ER as an emotional process variable. A few prospective studies of ER established precedence for the construction of the ER variable in this study.

In adults with RA, Connelly and colleagues (2007) collected daily diary reports of negative and positive affect scores from the PANAS (Watson et al., 1988). Based on

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procedures by Paquet et al. (2005), positive and negative ER scores were constructed by calculating daily changes in emotion intensities and identifying instances of maintaining or recovering emotion intensities.  Adaptive regulation of negative emotions predicted decreases in pain intensity.  Moreover, results from multilevel models showed that both negative *and* positive ER predicted pain intensity reductions in adults with arthritis (Connelly et al., 2007).

The few studies to prospectively examine ER in children or adolescents support ongoing longitudinal studies of ER.  Silk and colleagues (2003) examined the difference between current and peak negative emotion on each diary as a measure of ER.  For example, a change from high peak sadness to low current sadness indicated adaptive ER.  Adolescents completed 88% of diaries despite multiple reporting periods each day.  Additionally, Suveg et al. (2010) found that e-diaries are a feasible method of capturing affective data from 7- to 12- year-old children.  These studies suggest that experience sampling is a useful way to examine emotional experiences of children and adolescents, allowing researchers to move beyond trait-based emotion variables to those that are dynamic and temporally constructed.

**Emotion regulation conclusion.** In conclusion, the ability to adaptively regulate emotions is an important developmental competency that has implications for adjustment and health outcomes.  A prospective approach to assessing ER is well suited to capturing  discrete positive and negative emotional states as they occur and change. The construction of emotion recovery as an aspect of ER followed strategies outlined by Connelly et al. (2007; 2012), using prospective diary data collection.  Using these methods, this study furthered understanding of the role of ER in children who face significant stressors in the absence of psychopathology.

**Ecological Momentary Assessment and Electronic Daily Diaries**

Prospective methodologies are frequently used to assess health behaviors and symptoms. Ecological momentary assessment (EMA) is a form of experience sampling that allows data to be recorded in real-time (Shiffman et al., 2008) in order to capture processes as they occur in the person's environment. Electronic devices such as smartphones (i.e., cellular phones with computer operating systems) allow participants to provide cued, mobile reports of current experiences as they occur in the natural environment, instead of a laboratory or clinical setting.

E-diaries overcome many of the limitations of paper and pencil diaries (memory biases, diary hoarding, backfilling), can use electronic tools (alarms) to cue reports, and allow the measures to be easily accessible at all times. In an innovative study using technology to assess when paper diaries were opened, adults with chronic pain reported completing paper pain diaries within a 90-minute period on 94.9% of entries, but actual compliance was only 19.6% (Stone, Shiffman, Schwartz, Broderick, & Hufford, 2003). In contrast, participants completed 94% of e-diaries and all e-diary entries were completed within the appropriate timeframe. These findings provide strong evidence for the use of e-diaries to promote accurate reporting in real-time and suggest that these methods are suited to frequent pain assessments.

**Using e-diaries with children.** Moving to using e-diaries is particularly suitable to research with children, who are quite familiar and comfortable with new technology. Recent United States Census (2009) findings indicated that majority (68.7%) of households with children and adolescents have home internet access and access to mobile phones, which are predominantly smartphones (Lenhart, Purcell, Smith, & Zickuhr, 2010; A. Smith, 2010,

2012). In one study, children reported using computers and cellular phones a few times each week, on average (Jackson et al., 2008), but given the significant increase in access to technology occurring over the past few years, it is likely that children use technology even more frequently at present. Children also appear to enjoy using these new technologies. In both our pilot study exit survey and that of Stinson and colleagues (2006), children with JIA reported high satisfaction with the e-diaries and electronic devices, often commenting that the methodology was novel and enjoyable.

Children and adolescents' comfort and familiarity with new technology affords new strategies for tracking behaviors, ranging from assessing health behaviors to daily stress and symptoms of psychopathology. For example, e-diaries have been widely used to track physical activity over days and weeks (Dunton, Whalen, Jamner, & Floro, 2007; Gorely, Marshall, Biddle, & Cameron, 2007) and to identify the settings in which children engage in physical activities or sedentary behavior. E-diaries have also been used to better understand daily symptoms of psychopathology in children, including ADHD symptoms (Whalen et al., 2006), disordered eating (Hilbert, Rief, Tuschen-Caffier, de Zwaan, & Czaja, 2009), and self-injurious behavior (Nock, Prinstein, & Sterba, 2009). These studies provide support for the ongoing use of e-diaries in children. Furthermore, by allowing researchers to understand the antecedents, occurrences, and outcomes of behaviors, e-diary studies may lead to better diagnostic conceptualizations, more ecologically valid treatment approaches, and improved intervention delivery methods (Boschen & Casey, 2008).

**E-diaries in pain research.** In-the-moment reports on e-diaries are especially appropriate for capturing symptoms and behaviors that fluctuate over time and show great promise for assessing pain (Keogh, Rosser, & Eccleston, 2010) and emotions (Wenze & Miller, 2010). Research using e-diaries in pain populations has shown low reactivity (i.e.,

change in the variable of interest due to measurement) and high completion rates. Stone and colleagues (2004) compared reporting patterns of adults with pain prompted to completed e-diaries 3, 6, or 12 times per day and found high completion rates (> 94%) regardless of number of prompts. Participants also showed little change in pain reports over time, indicating low reactivity to electronic pain monitoring; low reactivity in pain and emotion ratings has also been demonstrated in other e-diary studies (Aaron, Manci, Turner, Sawchuk, & Klein, 2004; Peters et al., 2000). Litcher-Kelly and colleagues (2007) prompted adults with inflammatory bowel syndrome to complete an e-diary during each hour of the day. Despite the high reporting demand, participants completed 88% of all possible entries within the 3-minute window. Similarly, a systematic review found high completion rates across e-diary studies; short surveys and incentives promoted compliance (Morren, van Dulmen, Ouwerkerk, & Bensing, 2009). These studies support the feasibility of, and improved reporting from e-diaries.

Although Morren and colleagues' (2009) review of e-diaries found that older age predicted higher completion rates in adults with pain, children also appear to benefit from the use of e-diaries. Palermo and colleagues (2004) found that children with JIA or headache completed significantly more entries on an e-diary than on a paper diary, with an average of 6.6/7 e-diaries completed. In a study of the feasibility and validity of e-diaries in JIA, completion rates were between 70.5% and 72.9% when children were required to complete entries 3 times per day (Stinson et al., 2008). All participants in the reviewed studies were able to complete electronic measures on mobile devices, suggesting that this format is appropriate for use with children.

In our pilot study using e-diaries with 10 children with JIA, children completed 2 out of 3 reports on most days over the course of 2 weeks and had an overall completion rate of 87.5% (Connelly et al., 2010). Upon completion of each e-diary entry, data were wirelessly uploaded to a central server. No participants in the pilot study experienced technical problems when completing the e-diaries; however, all data for one participant were lost due to smartphone malfunction. Some children in the current study experienced occasional difficulty with accessing or uploading surveys, mostly due to limited wireless or cellular access. However, some data were obtained from all participants, unlike previous studies that stored data on the electronic devices, which lost a few complete sets of diaries. For example, Palermo and colleagues (2004) were unable to use data from 5 participants due to technical malfunction. Although a potential for lost data remains, e-diaries clearly offer many advantages, allowing researchers to better understand the nature of chronic pain as it occurs. Children and adults have demonstrated high completion rates and low reactivity, demonstrating that recurrent prompts for self-reports are tolerable, and thus were appropriate for use in this study.

**Specific Aims and Hypotheses**

The primary aim of this study was to understand the role of ER in the sleep and pain of children with arthritis. A unique aspect of this study was the focus on the regulation of both negative and positive emotions. Based on research identifying sleep and pain as predictors of functional limitations in children with persistent pain, I intended to understand the underlying mechanisms associating these variables through this prospective longitudinal study. Our previous study using paper daily diaries detected an association between sleep and pain, and identified sleep quality as a strong predictor of pain (Bromberg et al., 2011).

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However, use of end-of-day sleep reports introduced limitations to interpreting those findings, including the potential influence of intervening experiences on retrospective reports of the day.  As such, it was important to confirm those results using e-diaries, which eliminate backfilling and minimize retrospective bias. The following hypotheses were tested in this study.

*Hypothesis 1:  Sleep quality predicts pain intensity in children with JIA and this association is moderated by emotion regulation (Figure 1).*

*Hypothesis 1a: Daily sleep quality predicts daily pain ratings using e-diaries.* Previous findings using paper-and-pencil diaries were expected to be replicated using e-diaries, such that poor sleep quality at night will predict high pain intensity ratings on the following day.  Also, I expected to continue to find that children who experience lower sleep quality, overall, also experience higher pain intensity, on average.

*Hypothesis 1b:  Adaptive regulation of emotions buffers the effects of lower sleep quality at night on pain intensity the next day.* Adaptive regulation of negative emotions buffers the effects of poorer sleep quality at night on pain the next day.  Moreover, I expected to find that the regulation of positive emotions (i.e., recovering from instances of low positive emotions) also weakens the association between sleep quality and pain.  As such, regulation of both negative and positive emotions were expected to protect against the deleterious effects of a poor night's sleep on subsequent pain.

In addition to confirming previous findings and examining the role of ER in the pain-sleep relationship, this study further developed understanding of how sleep and pain contribute to overall functioning across physical, social, and school-related tasks.  This

second aim was consistent with the importance of understanding functional outcomes, as highlighted in the Biobehavioral Model of Pediatric Pain (Varni et al., 1996). Furthermore, in addition to disease and pain management, improvements in physical, social, and academic functioning are additional JIA treatment goals (Giannini et al., 1997). Therefore, it was important to understand how factors contribute to both pain and overall functioning across these important domains.

*Hypothesis 2: Daily pain intensity ratings mediate the relationship between sleep quality and overall functional limitations.* Lower sleep quality at night predicts higher pain intensity ratings during the day, which in turn predicts higher same-day functional limitations across physical, social, and academic domains. I also expected to detect a direct relation between lower sleep quality at night and higher overall functional limitations during the day.

An important feature of this study was the use of e-diaries to track and report sleep, pain, and functional limitations under naturalistic settings. This innovative data collection tool is best suited to capturing fluctuating symptoms and behaviors by obtaining real-time data at home. E-diaries have been validated for use with children with arthritis (Stinson et al., 2006, 2008; Connelly et al., 2010), but have not previously been used to examine emotional processes or the daily relationships between sleep, pain, and functional limitations in children with arthritis.

# CHAPTER II

## Method

### Overview

Data for the current study were collected as part of a larger study (NIH/NIAMS 1R01AR53845-1A2) led by Laura E. Schanberg, M.D., examining daily functioning in 8- to 18-year-old patients with polyarticular juvenile idiopathic arthritis. Using an e-diary on a smartphone, children and their caregivers reported on child pain, symptoms, behaviors, and emotions. Data collection for the larger study began in September 2009 and was completed in June 2011. Seventy-four caregiver-child dyads were recruited for the study and 59 children completed full study procedures. Daily items related to sleep were specifically designed for the current study and were not intended for use in the data analysis for the larger study.

### Participants

All participants recruited from the Duke Pediatric Rheumatology clinic met International League of Associations for Rheumatology (ILAR) diagnostic criteria for juvenile idiopathic arthritis with polyarticular presentation (Petty et al., 2004) and were excluded if they had another medical or psychiatric disorder that could affect pain, displayed a significant cognitive impairment, were non-English speaking, were not currently enrolled in an academic curriculum, or if children or caregivers had low computer literacy.

Consistent with the demographics of the population served by the Duke Pediatric Rheumatology Clinic, participants were predominantly Caucasian girls. The majority of participants had mild (43.1%) or moderate (39.7%) disease activity. The average age of participants was 13.3 years (SD = 2.8).

**Procedure**

Eligible patients from the Duke Pediatric Rheumatology clinic were prescreened by a research assistant using clinic schedules. Caregivers of eligible children were then contacted via letter from the Duke Pediatric Rheumatology faculty in order to introduce the study and describe study procedures. Research personnel approached families at their next regularly scheduled clinic visit. All participants were recruited and participated during the academic year in order to obtain data in the context of a typical school year. The study involved 3 phases – a baseline clinic visit, a 4-week e-diary reporting phase, and a follow-up evaluation at the end of the e-diary reporting period.

At the baseline visit, patients were given a physical examination as part of their routine appointment and study personnel explained study procedures and obtained assent and consent. Participants completed baseline questionnaires of psychological adjustment, sleep disruptions, emotion regulation, and physical, academic and social functioning on a secure website via wireless access on a laptop computer after selecting a personal login and password that was used for all electronic data entry. Following completion of questionnaires, a research staff member trained the parent and child on the use of the smartphone and e-diary. Finally, the family selected a starting date for daily monitoring. The 4-week response period was marked on a calendar, which was provided to the family along with the smartphones, outlet charger, study manuals, and prepaid mailers.

During the reporting phase of the study parents and children each completed e-diary entries for 4 weeks.  The research coordinator contacted the family via email or telephone during the first week of reporting.  Participants were encouraged to contact study personnel to report technical problems or other barriers to study completion during the e-diary reporting period.

For the study follow-up, participants completed questionnaires similar to baseline assessments at home via the secure website or with a study staff member via telephone.  All participants were reimbursed $10 for completing baseline procedures and $10 for follow-up questionnaires.  Consistent with approaches in other daily diary studies (e.g., Palermo et al., 2004; Whalen et al., 2006; Schanberg et al., 2003, 2005) diary completion was promoted via incentives.  Caregivers and children earned $0.25 for each e-dairy entry during the first two weeks of the study.  In order to decrease participant dropoff, participants earned $0.50 for each e-diary entry completed during the last two weeks.  As an additional incentive, participants earned a bonus of $0.50 for completing 3 reports in one day and $1.25 for completing all entries for one week.  The total amount earned was applied towards a gift card at the end of the study.

**Equipment**

E-diary reports were collected via T-Mobile ® Dash smartphones (see Figure 2) and the keypad was used to enter all responses.  Each smartphone had both cellular and wireless connectivity; however, the telephone features were largely disabled (i.e., calls could only be made to 911 and text messaging was disabled).  Participants were able to use the Internet, but a webguard was placed on all smartphones in order to ensure safety for children's Internet use.

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**E-Diary Programming, Pilot Testing, and Data Security**

In order to program the e-diary mobile application, we contracted with U-Inc. (Overland Park, Kansas) a software development firm. Our research group provided U-Inc. all content and formatting for the e-diary smartphone application and the study website. The e-diary mobile application development occurred over the course of several months and various changes to the formatting and flow were implemented based on pilot testing conducted with a sample of 15 children aged 8-16 years with rheumatic diseases other than JIA. U-Inc. also managed the secure website that housed the baseline electronic questionnaires and stored all study-related data, including e-diary reports. The website could only be accessed through a study login and password. Separate administrative areas of the website housed all baseline and e-diary data and could only be accessed by study personnel. At the time of each e-diary entry, information was uploaded to the secure web-based database via cellular or wireless technology built into the smartphones. Each e-diary entry was automatically dated and time-stamped and participants did not have access to their responses after uploading.

**Measures**

**Electronic diary.** E-diary content was developed based on previous paper diaries (Gil et al., 2000, 2003; Schanberg et al., 2003, 2005) containing pain, mood, and sleep items, in addition to a review of published measures of child activity limitations and emotion regulation. E-diaries are validated for use with children with arthritis and are increasingly used to assess pediatric pain prospectively (Connelly, Miller, Gerry, & Bickel, 2009; Stinson et al., 2008).

E-diaries were accessed on the smartphone by launching the e-diary mobile application. Three distinct e-diaries were developed for morning, afternoon, and evening, and include different items depending on activities typical during each timepoint (i.e., the morning diary included items about getting dressed and the afternoon diary included school-related items). The reporting periods are similar to those of Stinson and colleagues' previous JIA pain e-diary (2006). E-diary items were presented in three formats: electronic visual analog scales (e-VAS; Figure 3), body map images, and multiple-choice items (see Figure 4). Only e-VAS and multiple-choice items were used in the current study.

Symptom ratings on 50mm e-VAS were based on a 100mm VAS from paper diaries and items related to emotions and functioning were based upon standardized questionnaires such as the CALI (Palermo, Lewandowski, Long, & Burant, 2008) that used multiple-choice formats. Upon uploading the data, e-VAS scores were automatically converted to a 100-point scale in order to be comparable to previous VAS. Multiple-choice items included a question followed by a radio dial for each response option (Figure 5). At the end of the e-diary, participants were instructed to submit their e-diary and a confirmation screen appeared to indicate that the e-diary successfully uploaded.

***E-diary instructions.*** Participants were provided with detailed manuals identifying the use of each button on the smartphone and instructions for completing each type of diary question. A research team member modeled the use of the smartphone and completion of diary items for the family before assisting the family with completing sample diary questions. We encouraged participants to practice using the smartphone and e-diary until they were comfortable with the technology.

As part of the study orientation, a research team member worked with each participant to select response times for morning, afternoon, and evening diary entries based on their unique schedules. Morning response times were selected close to the individual's daily waking time. Children were instructed to select afternoon times after they typically arrived home from school and caregivers were instructed to select afternoon times after they expected to have contact with their child after school or work. Evening times for caregivers and children were scheduled to be close to bedtime. At the scheduled times, participants received an email alert that triggered an auditory alarm on the smartphone. The appropriate e-diaries were available only during a set window (e.g., afternoon e-diaries could be accessed and completed between 12pm-6pm.)

*E-diary completion.* Beginning on the designated start date, families completed e-diary entries for 4 weeks. The study coordinator monitored daily completion and contacted all families during the first week of the reporting period to assess for challenges and to encourage ongoing participation. Research personnel worked with the family to promote ongoing participation and address difficulties that emerged. For example, a new smartphone could be shipped overnight if a participant's device malfunctioned. Participants were encouraged to call, page, or email study personnel regarding any difficulties. At the end of the reporting period the smartphones were returned to the Duke Pediatric Rheumatology headquarters in prepaid mailers.

*Morning sleep e-diary item.* Sleep was measured via a global rating of sleep quality on an e-VAS anchored by "did not sleep well" and "slept very well." A similar sleep quality VAS has been used on previous paper diaries (Schanberg et al., 2003; Schanberg et al., 2005; Valrie, Gil, Redding-Lallinger, & Daeschner, 2007a, 2007b, 2008). Daily sleep logs are a

valid and reliable method for assessing sleep (Gaina, Sekine, Chen, Hamanishi, & Kagamimori, 2004; Sadeh, Raviv, & Gruber, 2000). Morning sleep quality reports capture the subjective sleep experience in the home setting. Although other data on sleep were collected as part of the overall study (e.g., number of sleep disruptions during the night), sleep quality served as the sleep variable in hypothesis testing.

*Pain e-diary items.* At the start of each diary, children were asked to rate their current pain intensity on a 50mm e-VAS anchored by "no pain" and "a lot of pain." Children were prompted to "Move the blue bar in either direction along the line to show how much pain you are having right now." The pain e-VAS was similar to a validated pain VAS from the Pediatric Pain Questionnaire (Varni, Thompson, & Hanson, 1987) and was based upon those in paper diaries (Schanberg et al., 2003, 2005.) VAS are a well validated method of assessing subjective pain intensity (L. L. Cohen et al., 2008) and have been recommended for use in children ages 8 and older (Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006).

*Emotion e-diary items*. Children completed items from an abbreviated version of the Positive and Negative Affect Schedule for Children (PANAS-C; Laurent et al., 1999) on each e-diary. The PANAS-C is based on a bivariate affective model and thus contains discrete negative and positive emotion scales with varying intensity levels. The PANAS-C has good validity and reliability (Lonigan, Hooe, David, & Kistner, 1999; Wilson, Gullone, & Moss, 1998). Five items with varying levels of arousal were selected from both the negative affect scale (e.g., "sad," "lonely") and the positive affect scale (e.g., "happy," "excited"). Children were instructed to rate their current intensity for each emotion on a 4-point scale ranging from "not at all or slightly" to "extremely." Items were averaged to obtain

negative and positive affect scores, which were used to calculate the ER variable for this study.

*Emotion regulation*. The method for constructing the ER variable was based upon Connelly and colleagues' (2007) methodology. Similar to Silk and colleagues' (2003) ER construct, Connelly et al. labeled ER as occurring if an emotion intensity rating returned to a typical level, following an increase in negative emotions or a decrease in positive emotions.

Changes in emotions from one to the next completed entry that day (e.g., from afternoon to evening) were used as the basis for detecting ER via a series of steps similar to those employed by Connelly et al. (2007). As outlined in Table 1, I first examined PANAS-C (Laurent et al., 1999) negative and positive affect ratings to identify instances of emotion activation. Negative emotion activation occurred when a child's negative emotion score was greater than .5SD above the child's mean negative emotion intensity. Similarly, positive emotion activation occurred when positive emotion intensity was greater than .5SD below the child's mean positive emotion intensity. The child's overall mean emotion intensity was used as the point of comparison based on preliminary findings that there was little difference in emotion intensity scores across the morning, afternoon, and evening e-diaries, indicating diurnal patterns do not need to be addressed when constructing the ER variable.

The next e-diary entry after times of emotion activation was examined to determine if the child adaptively regulated the emotion. Adaptive emotion recovery occurred if a child's negative or positive affect returned to within .5SD of his or her mean emotion intensity on the next e-diary entry. Consistent with bivariate affective models, separate variables were constructed to code for positive emotion recovery and negative emotion recovery and were

coded as follows: 0 = no emotion activation, 1 = adaptive emotion recovery, 2 = failure to adaptively recover emotions.

This approach to constructing ER addressed many of the methodological issues raised by Cole and colleagues (2004). First, ER was a well-defined variable constructed based on current theoretical model of ER (Gross & Thompson, 2007). Second, prospective data collection allowed me to examine change in emotions over time, thus avoiding confusing ER with emotion activation. Also, a temporally constructed ER variable directly measured ER and avoided the pitfall of interpreting the association between emotion valence and outcomes as evidence of ER.

*Functional limitations e-diary items*. E-diary items assessing physical, social, and school-related functional limitations were taken from two standardized and validated measures. Items assessing physical functioning and activities of daily living were derived from the Activity Scale for Kids (Young, Williams, Yoshida, & Wright, 2000) and items related to school and social functioning were taken from the Child Activity Limitations Questionnaire (Palermo et al., 2008), which have been shown to have adequate psychometric properties. Each item assessed the difficulty of completing the activity (e.g., "How difficult was it to put your clothes on this morning?), and all responses were rated on a 4-point Likert scale (ranging from 1-not very difficult to 4- extremely difficult). An average limitation score was calculated for each diary entry, with higher scores indicating greater difficulties in functioning, independent of the number of items endorsed.

**Baseline Measures.** Children and caregivers completed a variety of measures assessing baseline psychosocial functioning and adjustment. Although most questionnaires were not used in hypothesis testing, results of the questionnaires provide important

descriptive information regarding the study sample and were used to validate the e-diary measures. Disease activity and age were used in data analysis in order to control for the effects of a global medical variable and the potential developmental differences in sleep, pain, and ER.

*Disease activity.* Physicians completed a disease activity rating based on a routine physical exam conducted during the baseline visit. Disease activity ratings were completed on a 4-point scale ranging from 0 (inactive) to 3 (severe). Disease activity ratings are routinely used in rheumatology clinics.

*Demographics.* Caregivers completed a demographics form at baseline. Caregivers reported family demographics (e.g., caregiver education level and age) as well as information on the child (e.g., race/ethnicity, grade, age). Caregivers also provided a list of the child's current medications.

*Depressive symptoms.* Children completed the Children's Depression Inventory (Kovacs, 1983). The CDI is a 27-item, self-report scale assessing a variety of depressive symptoms including anhedonia, changes in appetite, and depressed mood. It provides a total score of depressive symptoms; higher scores indicate increased depressive symptoms. The CDI is a valid, widely used, and well-researched measure of child depressive symptoms (Kovacs, 1983, 1992). Children's scores on the CDI were used to test discriminant validity for the ER variable.

*Anxiety.* Children completed the Multidimensional Anxiety Scale for Children (March, Parker, Sullivan, Stallings, & Conners, 1997) at baseline. Children responded to items on four scales regarding physical symptoms, harm avoidance, social anxiety, and

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separation/panic on a 4-point Likert-type scale ranging from 0 ("Never true about me") to 3 ("Often true about me.")  Subscale and Total Anxiety scores were converted to T-scores. The MASC is an empirically derived scale with a clear construct of anxiety, which includes physical symptoms and approach/avoidance symptoms (Myers & Winters, 2002) and has been shown to have better construct and discriminant validity than other common child anxiety measures  (Dierker et al., 2001).  Similar to the CDI, MASC scores were used to discern the validity of the constructed ER variable.

# CHAPTER III

## Results

### Descriptive Statistics and Completion Rates

Data from participants who completed < 25 e-diary entries were not used in data analysis, which resulted in a final sample of n=59. Of the original 74 children recruited, 3 participants were excluded from analysis due to completing < 25 entries and the remainder of excluded participants did not meet eligibility criteria upon enrollment (n=3) or withdrew from the study before completing 25 e-diary entries (n=9). In the final sample, individual completion rates ranged from 26-91 entries (m=56.17, SD=18.45). A total of 3258 entries were logged. The 66% completion rate was similar to that of our initial e-diary pilot study (Connelly et al., 2010) in which we found that participants typically completed 2 out of 3 entries per day. Neither gender ($F$ (1, 56) = 5.94, $p$ > .05) or disease activity ($F$ (3, 51) =.10, $p$ > .05) significantly predicted the number of e-diary entries completed. Similarly, age was not correlated with number of e-diary entries ($r$ = .06, $p$ > .05).

The average pain intensity rating was 26.29 (SD = 27.51; Table 3), which can be interpreted as being in the mild to moderate pain range (Bulloch & Tenenbein, 2002; Collins, Moore, & McQuay, 1997). Children reported experiencing pain (> 0) on 68.6% of all entries and reported pain in the high intensity range (> 40) on 31.3% of all diary reports. The average sleep quality across all children and all moments on a 100-point VAS was 67.94 (SD = 27.38). Children reported taking at naps on 17.4% of e-diary days and most of these naps

(76%) lasted between 1-2 hours.  Children typically experienced low levels of functional

limitations, with an average total functional limitations score of 3.86 (SD = 7.26), out of a

possible range of 0-32.  Participant's scores on the CDI and MASC were generally below the

clinical range (Table 2).

**Hypothesis Testing using Multilevel Hierarchical Model Building**

Hypothesis testing was completed through a series of multilevel models consistent

with recommendations made by Raudenbush and Bryk (2002).  Multilevel modeling is an

advanced statistical approach that allows for the nesting of observations within each

participant and has been recommended for use in daily diary studies (Schwartz & Stone,

1998).  Multilevel modeling is able to accommodate varying numbers of observations across

individuals and can accommodate missing data, without estimating these values. Multilevel

modeling partitions variance based on the nesting structure; in this case, e-diary reports are

nested within a child.  Thus variance is partitioned into within-child, momentary sources

(Level 1) and between-child, average sources (Level 2) of variance.  Given the expected

correlations between observations in e-diary data, a serial autocorrelation residual variance

structure (spatial power) was applied to all models to allow for observations taken closer

temporally to be more similar than those taken further apart in time.  A series of hierarchical

multilevel model building was conducted in order to determine the additional variance in the

dependent variable accounted for by the inclusion of new predictors.  All baseline predictors

were centered around the grandmean (i.e., the mean for all children) and all momentary

predictors from the e-diaries were centered around each child's mean score, with the child

means entered into the model in order to decompose effects by level of variance. All

hypothesis testing using multilevel modeling was conducted in SAS using PROC MIXED (SAS, 2008).  Results of hierarchical multilevel model building are displayed in Table 4.

**Random Effects ANOVA.** The first model, a test of the random effects of analysis of variance (REANOVA) contained no predictors and was used to calculate the interclass correlation (ICC) to decompose sources of variance in pain intensity. The ICC indicates that of the total observed variability in pain intensity scores, approximately 59% is due to between-child differences.  In other words, the correlation between pain intensity scores of the same child is $r = .59$.  More variance in pain intensity resides at the between-child level than at the within-child level, indicating that trait factors influence momentary pain intensity ratings more than state factors.

**Model 2- Adding Time Invariant Covariates as Predictors of Pain Intensity.** Grandmean centered age and disease activity were entered into level two at the second step of multilevel model building.  Neither age, $t(52)= -.85, p >.05$, or disease activity $t(52)= 1.36, p >.05$ significantly predicted pain intensity.  However, consistent with the proposed method of hierarchical model building, these covariates were retained in all subsequent steps of model building.

**Model 3- Adding Within- and Between-Child Sleep Quality as Predictors of Pain Intensity.**

 Childmean centered sleep quality significantly predicted daily pain intensity, $t(2456) = -3.20, p =.001$, indicating that higher sleep quality at night predicted lower pain intensity the following day.  Similarly, children who experienced higher average sleep quality during the study period experienced lower pain intensity, on average, $t(51) = -3.64, p < .001$. These findings support Hypothesis 1a;sleep quality significantly predicted both within and between-child variance in pain intensity.

As previously noted, the majority of variance in pain intensity is due to between-child factors. Mean sleep quality accounted for 21% of between-child variance in pain intensity, whereas daily sleep quality accounted for only 7% of the total within-child variance in pain intensity. It appears that how well a child sleeps on any given night is not as important as how well a child typically sleeps, when predicting pain intensity.

**Model 4- Negative and Positive ER Indices and Interaction Terms as Predictors of Pain Intensity.**

The third step of model construction was initially tested using the proposed ER constructions, based on changes in PANAS-C scores using each child's mean and .5SD to determine cut points and coding for instances of no emotion activation, adaptive recovery, and failure to adaptively recover emotions. Separate categorical positive and negative ER indices were added to the model at this step. Results indicate that there was no significant overall effect of negative ER on pain intensity ($F(2, 82) = .42, p > .05$), indicating that pain intensity did not significantly differ at times when children had no emotion activation, compared to times when children adaptively recovered negative emotions, and to times when children failed to adaptively recover negative emotions. There was a significant effect of positive ER, $F(2, 103) = 12.45, p < .0001$. Compared to instances when children failed to recover from drops in positive emotion, pain intensity was significantly lower at times when children adaptively recovered from drops in positive emotions $t(103) = -4.56, p < .0001$, as well as at times when children experienced no positive emotion activation, $t(103) = -4.68, p < .0001$. The interactions between negative ER and sleep quality, $F(2, 2411) = .42, p > .05$ and positive ER and sleep quality, $F(2, 2411) = .22, p > .05$, were not significant.

Taken together, contrary to Hypothesis 1b, adaptive ER (i.e., adaptive recovery from instances of emotion activation) did not buffer the effect of sleep quality on pain intensity

using the proposed ER construct.  Although ER did not moderate the association between sleep quality and pain intensity, the significant main effect of positive ER suggests that more adaptive positive emotional functioning protected against increased pain intensity.

   ***Model 4 with Alternate ER construct.***  A different pattern of findings emerged when employing an alternate construction of ER.  Based on statistical concerns related to the use of means and standard deviations in the construction of a temporally unstable ER construct (e.g., possibility of regression to the mean, issue with the instability of standard deviations), alternate negative and positive ER indices were constructed using extreme, outlying observations as the criteria for cut points.   Jahng et al. (2008) discuss different options for determining cut points in their test of different indices for constructing measures of temporal instability of affect.  In situations where there is no clear theoretical or statistical cut point, they suggest using a location relative to the distribution, such as the 95$^{th}$ percentile of the distribution.  The alternate measure of negative ER was constructed using a cut point at the 95$^{th}$ percentile of each child's distribution of negative PANAS-C score and the 5$^{th}$ percentile for positive ER.  Emotion activation occurred if a child's score was outside the cut point in the respective distribution (i.e., negative emotion scores above the 95$^{th}$ percentile were coded as instances of negative emotion activation).  Similar to the initial method for constructing the ER variables, adaptive emotion recovery was defined as instances in which a child successfully returns to a level below the cut point for negative emotions or above the cut point for positive emotions following an instance of emotion activation.

   Results of the model testing the effects of the alternate ER constructs indicated that the overall effect of positive ER trended towards significance, $F(2, 73) = 2.54$, $p = .09$. There was also a significant interaction between sleep quality and positive ER, $F(2, 2411) =$

3.78, $p < .05$. However, upon probing this interaction, the graphical representation of the interaction is not consistent with the slopes of the graphed raw data (Figures 6-8). As such, the findings displayed in Figure 5 cannot be interpreted with confidence. Visual inspection of Figures 6-8 suggest that higher sleep quality was associated with lower pain intensity, but this association was stronger at times of no positive emotion activation and positive emotion recovery. Across levels of positive emotions, pain intensity is in the severe range (>40) when children experience the lowest sleep quality.

In sum, employing an alternate ER construct to address statistical concerns of the proposed ER construct resulted in a pattern of findings more consistent with study hypotheses. The proposed construct detected a main effect of positive ER, such that pain intensity was highest when children failed to adaptively recovery from drops in positive emotion. The alternate ER construct, which is believed to be more statistically sound, resulted in a more robust pattern of findings and detected an interaction effect. However, the nature of the interaction effect is unclear and should be interpreted with caution given the above described discrepancies between the graphed interaction and the graphs if the raw data at each level of positive ER.

**Exploratory Model Testing Random Slopes for Sleep Quality**

An exploratory model including random slopes for sleep quality was tested in order to determine whether the association between sleep quality and pain intensity should be allowed to statistically vary by child. Results of this model were similar to the final model (model 4 with alternate ER construct). The Akaike's Information Criteria (AIC) was compared between the two models as a measure of model fit; smaller AIC values indicate better model fit (Bozdogan, 1987). The difference between the AIC for the original model with fixed

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slopes (20921.0) and the exploratory model with random slopes (20916.2) was minimal.

Based on this finding, together with limited theoretical guidance for model selection, the

original fixed slopes model holding the relationship between sleep quality and pain intensity

constant for all participants was interpreted in this study.

**Exploratory Multilevel Model Building Controlling for Previous Pain**

Two fixed-slope models were constructed to explore the effects of pain intensity from

the previous night on pain intensity the following day.  The influence of evening pain on

current pain intensity was tested simultaneously with age and disease activity.   Evening pain

significantly predicted pain during the next day $t(764) = 13.39$, $p <.0001$.  When childmean

centered sleep quality and the means for child sleep quality were entered into the model

containing age, disease activity, and evening pain, the effects of daily sleep quality were not

significant, $t(763) = -1.23$, $p >.05$, but children's typical sleep quality remained a significant

predictor of overall pain intensity, $t(51) = -4.19$, $p = .0001$. Due to the large loss of degrees of

freedom associated with limiting the dataset to observations with a previous evening pain

report, a second set of models was tested using average pain from the prior day and a similar

pattern of findings was detected with a similar degrees of freedom ($df = 955$).  Results of

these exploratory models suggested that the strongest predictor of daytime pain was previous

pain intensity, such that sleep quality did not predict pain intensity during the following day

when evening pain was included as a covariate.  However, these results must be interpreted

with caution, given the reduced number of observations included in the exploratory analyses

and the corresponding reduction in degrees of freedom, which limits the ability to detect

weaker associations.

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**Alternative Hierarchical Multilevel Models Testing the Effects of Pain Intensity on Sleep Quality**

Based on Lewin and Dahl's (1999) theoretical model of the bidirectional association between sleep and pain, alternative models were constructed to test the effects of pain intensity on sleep quality.  A series of hierarchical multilevel models similar to those employed in hypothesis testing were constructed to test this pattern of association.  In the first step of model building, age and disease activity were entered as predictors of sleep quality.  Only disease activity significantly predicted sleep quality, $t(52) = -2.22$, $p < .05$; higher disease activity predicted lower sleep quality.  The average pain intensity from the previous day was used to test the influence of pain intensity on sleep quality. Within-child, pain intensity from the previous day did not predict sleep quality at night, $t(955) = -1.53$, $p > .05$, but children's average pain intensity did significantly predict typical sleep quality $t(51) = -3.74$, $p < .001$.  Of note, fewer observations were retained for these models than for the models predicting pain intensity from sleep quality and these results should be interpreted with some caution.  An exploratory model using evening pain intensity ratings was also tested; based on a similar pattern of results and a greater loss of degrees of freedom for that model, only results from models including average daytime pain intensity are presented and interpreted here.

**Exploratory Model Probing the Effects of Age**

A final exploratory model was tested to determine whether the combined effects of ER and sleep quality on pain intensity varied by age, given that both sleep and ER are expected to vary across development.  A simultaneous model was constructed including the covariates, sleep quality, the proposed measures of negative and positive ER, and two 3-way interaction terms. The interaction between age, sleep quality, and negative ER was not

significant, $F(2, 2410) = 0.57$, $p > .05$; nor was the interaction term constructed with positive ER, $F(2, 2410) = 0.58$, $p > .05$.

**Multilevel Mediation Models Testing the Effects of Sleep Quality and Pain Intensity on Functional Limitations**

Hypothesis 2, that daily pain intensity ratings mediate the relationship between sleep quality and overall functional limitations was tested using Zhang et al.'s (2009) recommendations for testing 1-1-1 multilevel mediation using the centered within context with reintroduction of the subtracted means (CWC(M)) approach, which consists of a series of model testing to obtain parameter estimates. Per these guidelines, significance testing was conducted via a Sobel test (Sobel, 1982), using an online calculator (Preacher & Leonardelli, 2001). This approach allows mediation testing at both the within- and between-child levels; although the hypothesized effect lies at the within-child level, both sets of results will be reported and interpreted in order to avoid confounding effects.

**Within-child (level 1) mediation analysis**. At any given moment, lower sleep quality was related to higher pain intensity ($\beta = -.07$, $p < .001$), higher pain intensity was related to higher total functional limitations ($\beta = .11$, $p < .0001$), and lower sleep quality was related to higher total functional limitations ($\beta = -.03$, $p < .0001$; Figure 9). When the influence of momentary pain intensity was taken into account, the influence of sleep quality on total functional limitations dropped, but remains significant ($\beta = -.02$, $p < .0001$). Results of the Sobel test for level 1 mediation were significant ($z = -3.27$, $p = .001$), which supports the hypothesis that daily pain intensity ratings mediate the association between sleep quality and total functional limitations. Given the small $p$ value detected for the Sobel test, no additional hypothesis testing (e.g., parametric bootstrapping) was conducted.

**Between-child (level 2) mediation analysis**. On average, lower typical sleep quality was related to higher pain intensity ($\beta = -.50$, $p < .001$), higher typical pain intensity was related to higher total functional limitations ($\beta = .13$, $p < .0001$), and lower typical sleep quality was related to higher total functional limitations ($\beta = -.09$, $p < .001$) (see Figure 10). When the effect of pain intensity was taken into account, the relationship between typical sleep quality and total functional limitations was no longer significant ($\beta = -.03$, $p > .05$). Together with the results of the Sobel test for significant mediation ($z = -3.19$, $p = .001$), this pattern of findings indicated that the influence of typical sleep quality on total functional limitations was accounted for the by influence of the child's typical pain intensity.

**ER Construct Validation**

In order to test the validity of the ER construct, correlations between t-scores on the MASC, CDI, and the frequencies of adaptive emotional functioning (i.e., combined frequencies of times of no emotion activation and times when adaptive recovery of positive or negative emotions occurred), and the frequencies of instances in which children failed to recovery from positive or negative emotion were calculated as a measure of divergent validity. This was done using both the proposed and alternate ER constructs. Given that difficulties with ER are believed to underlie clinical presentations of anxiety and depression, I expected to find that higher frequencies of adaptive emotion functioning would be negatively correlated with CDI and MASC t-scores and that higher frequencies of failure to recover from emotion activation would be positively correlated with t-scores. However, there were no significant correlations using the proposed or alternate ER constructs. Overall, these analyses failed to establish convergent validity with the CDI and MASC, validated measures of internalizing symptoms.

**Final Model Diagnostics**

   Diagnostics were conducted to examine the residuals of level 1 variables, level 2 variables, and pain intensity scores using the final full model (model 4) with the alternate ER construct.  Results show that both the level 1 (Figure 11) and level 2 (Figure 12) residuals were relatively close to, but not normally distributed, indicating that the assumption of normality of the residuals was not met.  Given that the distributions were not extremely skewed, no corrections were implemented.  Additionally, based on plots of residuals by predictors, linearity assumptions of the model were approximately met.  Although model assumptions are not extremely violated in this case, in the future, daily pain process researchers may consider employing statistical modeling techniques that are not based upon assumptions of normal distributions.

**CHAPTER IV**

**Discussion**

The primary aim of this study was to understand the role of ER in the association between sleep quality and pain intensity of children with arthritis, focusing on the function of adaptive recovery of both negative and positive emotions. Expanding on previous paper diary research (Bromberg et al., 2011), I sought to understand underlying mechanisms associating these variables through this prospective longitudinal study. Using e-diaries allowed for more frequent assessments and increased ecological validity compared to data obtained from paper diaries. Thus, it was possible to go beyond examining daily associations between sleep quality and pain intensity to examine the influence of sleep quality and emotion recovery on pain intensity in any given moment.

Findings from this study largely supported the proposed hypotheses. The significant association between poor sleep quality at night and higher pain the following day detected in our previous paper diary study was replicated using e-diary entries; poor sleep quality at night predicted higher pain intensity on any given observation during the next day. Similarly, poor typical sleep quality continues to be an important risk factor for higher typical pain. Typical sleep quality accounted for 21% of the overall variance in pain intensity, indicating that sleep quality accounts for a similar proportion of JIA pain intensity variance as medical indices tested in previous studies (Malleson et al., 2004; Schanberg et al., 1997, 2003). However, the primary medical variable included in this study (disease activity)

did not predict pain intensity; it will continue to be important to determine the unique and combined influences of sleep and medical factors on pain in JIA. The overall patterns of findings regarding the pain-sleep relationship indicate that while a poor night of sleep is associated with higher pain the following day, children who typically experience poor sleep quality are at greater risk of experiencing more pain overall. In studies with healthy adults, sleep deprivation (Onen, Alloui, Gross, Eschallier, & Dubray, 2000), lack of REM sleep (M. T. Smith, Edwards, Stonerock, & McCann, 2005), and daytime sleepiness (Chhangani et al., 2009) contributed to hyperalgesia; supporting a causal pathway between insufficient or disrupted sleep and heightened pain sensitivity. Alternately, children with this symptom profile may experience more global physiological arousal, contributing to both disrupted sleep and increased pain sensitivity (Palermo, 2012).

Based on theoretical models of pediatric pain and sleep (Lewin & Dahl, 1999), it was also important to test the alternate pathway of influence; pain intensity predicting sleep quality. Although typical pain intensity predicted typical sleep quality, pain intensity on a given day did not predict sleep quality that night. Together with results from the models predicting pain intensity, a bidirectional association is supported, but consistent with recent research in both pediatric and adult populations (Lewandowski et al., 2010; Tang, 2012) findings from this study contribute to accumulating evidence that the pathway from sleep to pain may be stronger. However, there were fewer observations available for examining this directional pathway, resulting in decreased power and fewer degrees of freedom and these results are interpreted with some caution. As such, it remains important to study both directions of influence and to continue to examine mechanisms associating sleep with pain, such as emotional processes.

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Both the proposed and an alternate construct of emotion recovery were tested in this study; each revealed an important role of positive ER in children with JIA. A main effect of positive ER was detected using the proposed construct and an interaction between positive ER and sleep quality was detected using the alternate construct. Although the alternate construct used theoretical methods for identifying extreme observations (Jahng et al., 2008) and detected a more complex pattern of findings, these results must be interpreted with caution due to discrepancies between the graphed interaction and the graphs of the raw data. There may be issues with the statistical ER construct that continue to affect interpretability of findings. Suggestions for additional alternative temporally constructing ER variables are discussed further below. Additional research may be necessary to further delineate and explain the pattern of findings detected in this study regarding the role of ER in the daily pain of children with JIA.

Consistent with positive psychology theory (Fredrickson, 1998b, 2001; Seligman & Csikszentmihalyi, 2000) and previous research on the role of emotions in pediatric chronic pain (Valrie et al., 2008; Gil et al.; 2003, Bromberg et al., 2011), adaptive positive emotions were important to consider in the context of JIA pain. A significant interaction between positive ER and sleep quality was detected, but regulation of positive emotions alone did not predict pain intensity when using the alternate ER construct. Whether a child maintained, adaptively recovered, or failed to recover negative emotion intensity had no effect on the pain-sleep association; intervening upon negative emotional states may not have as great of an impact upon the relationship between sleep and pain in JIA. Although the interpretation of findings regarding the interaction is somewhat limited by statistical concerns, the general pattern of findings suggestions that positive emotions and their regulation warrant additional

research. These findings may align with evidence supporting the protective nature of positive emotions and adaptive ER and their role in the development of resilience (Alvord & Grados, 2005) in children facing psychosocial adversity (Curtis & Cicchetti, 2007) or chronic illness (Haase, 2004; Phipps, 2007). It is likely that the associations between emotions, sleep quality, and pain intensity are more complex than those captured in this study. Poor sleep quality may actually contribute to emotion activation the following day; it will be important to continue to test mechanistic processes underlying the associations detected in this study.

In addition to further delineating patterns of association between sleep and pain, this study aimed to determine the ways in which sleep and pain contribute to daily functioning. Based upon Varni and colleagues' (1996) Biobehavioral Model of Pediatric Pain, I hypothesized that daily pain intensity ratings would mediate the relationship between sleep quality and overall functional limitations. Using the CWC(M) (Zhang et al., 2009) approach revealed that pain intensity mediated the relationship between sleep quality and functional limitations at any given moment and the association between typical sleep quality and functional limitations was also partially accounted for by children's typical pain intensity. It appears that the overall effect of typically experiencing poor sleep quality on functional limitations is accounted for by high pain intensity, but on any given day sleep quality affects daily functioning above and beyond the effects of pain intensity, further supporting the need to intervene to improve sleep quality in children with JIA. Disrupted sleep affects daytime functioning across multiple domains in all children (Dahl, 1996; Sadeh et al., 2002) and these findings identify pain intensity as a mechanism linking poor sleep quality at night to higher functional limitations the following day in children with JIA. Impact on daily functioning is a marked clinical concern for the families of children with JIA and other chronic pain

conditions. The use of e-diaries offered the opportunity to delineate the temporal mechanisms associating sleep, pain, and functioning and results imply that interventions promoting both improved sleep quality and decreased pain intensity may lead to improved functional outcomes.

**Study Strengths**

There are a number of methodological strengths in this study and the findings build upon previous research in several important ways. First, the use of e-diaries offered the opportunity to capture behavior and symptom reports as they occurred, with multiple momentary assessments in a single day. The prospective nature of this methodology allows for the detection of mechanisms of change over time, as well as identification of related processes in a given moment using MLM. In the future, e-diary tools may be expanded to track and intervene upon mechanistic processes as they occur. For example, findings from this study could be applied to develop mobile intervention tools that continue to track sleep quality, pain intensity and functional limitations. Results from the mediation models suggest that it may be particularly important to prevent or abort increased pain intensity on days following nights of poor sleep quality. As such, mobile tools could be used to identify moments when children are at heightened risk for increased pain or functional limitations and to provide in-vivo suggestions for pain coping skills and ER strategies to practice at these times. Results of this study also indicate that children with JIA who typically experience poor sleep quality are also at risk for high pain intensity. Mobile programs could be used to help children identify factors contributing to disrupted sleep and to promote behavioral changes aimed at increasing sleep quality (e.g., establishing a developmentally-appropriate bedtime, limiting evening caffeine intake, modifying maladaptive thoughts about sleep).

The use of e-diaries overcomes some of the limitations of traditional paper diaries, which often ask participants to provide aggregate reports of symptoms during the day (e.g., typical pain level) and are subject to memory biases, such as the availability heuristic (Cone, 1999).  Paper diaries also burden participants with remembering to complete the reports.  E-diaries can take advantage of electronic tools to cue reports (via alarms) and allow the measures to be mobile and easily accessible at all times.  The potential for falsified reports (e.g., diary hoarding or backfilling) is an important limitation of paper diaries.  Although results from this study continue to indicate that much of the relationship between sleep quality and pain intensity lays at the between-child level, it remains important to employ e-diaries in future research on sleep in children with JIA.  For example, it will be important to use this technology to identify specific sleep disruptions that occur over time, which may also help to further validate existing retrospective sleep questionnaires for use with this population and assist in developing sleep-focused interventions.  Perhaps of greater importance, as research on daily pain processes progresses, e-diaries will play an important role in implementing in-vivo pain interventions and self-management programs and may be used to prospectively track treatment outcomes.

The use of a prospectively constructed ER variable (emotion recovery) was a notable strength of this study.  This approach addressed many of the ER construct issues raised by Cole and colleagues (2004).  First, emotion recovery was a well defined variable constructed based on a current theoretical model of ER (Gross & Thompson, 2007).  Second, prospective data collection allowed this study to capture change in emotions over time, and the steps for constructing emotion recovery explicitly identified instances of emotion activation as a step in a process potentially resulting in emotion recovery, thus avoiding confusing ER with

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emotion activation. Also, the temporally constructed ER variable directly measured a specific aspect of ER, rather than assessing the use of ER strategies as a proxy for adaptive ER. This temporal construct also avoided interpreting the association between emotion valence and behavioral outcomes as evidence of ER (i.e., successful achievement of behavioral outcomes being interpreted as adaptive ER), which has been identified as a potential pitfall of measuring ER (Cole et al., 2004). Beyond the theoretical strengths of a temporally constructed emotion recovery variable, the use of this construct as a momentary state measure of an aspect of emotion functioning extended previous paper pain diary research. In past studies (Valrie et al., 2008; Bromberg et al., 2011) positive mood was shown to influence the association between pain and sleep. However, these studies were limited by the use of end-of-day measures that required participants to provide retrospective mood ratings. The methods employed in the presented study offered the opportunity to determine the influence of momentary changes in emotional functioning on the pain-sleep association, thus further capturing temporal processes occurring in children's daily lives and moving the understanding of the role of emotion beyond the more temporally stable construct of mood. Previous studies also used bipolar affective constructs, with positive and negative valences situated on a continuous spectrum. A strength of this study was the use of a bivariate affective measure that allowed positive and negative emotion recovery to be examined separately. Finding that positive emotion recovery is more important than negative emotion recovery in the pain-sleep association bolsters previous findings on the importance of positive emotions from studies using bipolar affective constructs.

**Weaknesses and Limitations**

Despite the many strengths of this study there are a number of weaknesses and limitations related to the sample, constructs, and statistical methods employed. Similar to previous research on children with JIA (Schanberg et al., 2003; 2005), convenience sampling resulted in a sample predominantly composed of Caucasian females. Although these demographics are generally consistent with the JIA patient population at Duke University Medical Center and with the majority of pediatric chronic pain patients (Lynch, Kashikar-Zuck, Goldschneider, & Jones, 2006; Scharff et al., 2004), the small number of minority or male participants limits the ability to examine demographic differences in behaviors or JIA symptoms. Additionally, few children in this sample had severe disease activity, which may also limit the generalizability of these findings to all children with JIA. It may be that disease activity homogeneity contributed to null findings when predicting pain intensity from disease activity. It will be important to continue to determine best practices regarding methods for identifying and involving children most severely affected by JIA in research, particularly in order to better elucidate the role of disease factors in the finding that typically poor sleep quality results in higher overall pain intensity. It is possible that an underlying disease factor contributes to both heightened sleep problems and pain intensity. There was also a large age range of children in this this study, which was selected due to the low base rate of JIA and in order to represent much of the age range of children affected by JIA. However, there are developmental differences in both pain and sleep expected with the onset of adolescence. Sleep problems and pain prevalence increase during adolescence (Carskadon, 2002; Perquin et al., 2000; Stanford, Chambers, Biesanz, & Chen, 2008), likely related to pubertal and psychosocial changes (Dahl & Lewin, 2002). The exact influences of

age, development, and gender on the pain-sleep association largely remain unclear. In the future, applying the methods used in this study to multisite investigations with larger sample sizes may allow researchers to determine the presence of demographic and developmental differences in associations between sleep quality and pain intensity.

Sleep is a complex, biobehavioral process and poor sleep quality may be due to sleep disruptions caused by physiological (e.g., restless leg syndrome, sleep disordered breathing), behavioral (e.g., bedtime refusal, daytime napping, difficulty finding a comfortable position), and environmental factors (e.g., bedroom environment, neighborhood noise) (Mindell & Owens, 2010). A global construct of subjective sleep quality as used in this study is an important first step in understanding sleep in JIA, but does not capture the complex nature of sleep problems, making it difficult to know what specific sleep factors contributed to participants' e-diary sleep quality ratings. For example, it is difficult to know how heavily sleep disruptions versus sleep duration contributed to global sleep quality ratings. It may be that a child experienced no difficulty initiating or maintaining sleep, but only attempted and obtained a few hours of sleep, and thus rated sleep quality lower for that night. Although this study included a mixed sample of school-aged children and adolescents, who often obtain insufficient hours of sleep due to interactions between physical sleep regulatory processes and psychosocial factors (e.g., school schedules; Dahl & Lewin, 2002), the average sleep quality rating for the sample was relatively high. This finding may be due in part to the subjective nature of the global sleep variable, which does not protect against the possibility of children acclimating to ongoing sleep difficulties.

In the future, it will be important to include e-diary items assessing specific sleep disruptions (e.g., bedtime resistance, difficulty falling asleep, nightmares), sleep duration,

and frequency and severity of nighttime awakenings. Additional sleep monitoring via actigraphy would supplement these reports with objective data on delayed sleep onset, sleep maintenance, and total sleep duration (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). Collecting these data will offer the opportunity to prospectively identify specific sleep problems children with JIA experience. This will help to clarify findings from previous cross sectional research, which has produced equivocal results regarding the type and role of sleep problems in JIA (Bloom et al., 2002; Passarelli et al., 2006; Ward et al., 2008), partially due to differences in measures. Collecting more detailed sleep information will also assist in determining if children who typically experience poor sleep quality and high pain meet diagnostic criteria for specific sleep disorders (e.g., insomnia, SDB), consistent with other recent prospective research focusing on insomnia (characterized by difficulty with sleep onset, frequent night awakenings, and early morning waking) in adolescents with chronic pain (Palermo, Wilson, Lewandowski, Toliver-Sokol, & Murray, 2011). Finally, obtaining more detailed sleep information will lead to a better understanding of the aspects of sleep in JIA that contribute to daily health outcomes. However, given that a variety of sleep problems may contribute to heightened pain sensitivity (Chhangani, et al., 2009; Onen, et al., 2000; M. T. Smith, et al., 2005), it was important to use a global subjective measure as a preliminary step in developing this area of research in children with chronic pain. It would also be helpful to assess daily sleep hygiene and medication use to better understand factors affecting sleep at night in children with JIA, particularly in the context of attempting to draw causal conclusions regarding pain as a sleep disruptor.

There were also measurement issues related to the original and alternate emotion recovery constructs. The alternate construct was used and interpreted in this study in order to

address concerns about the originally proposed construct, related to the instability of standard deviations and potential regression to the mean, but does not take into account each child's degree of variability in emotions. In the future, additional alternate statistical methods for measuring temporally constructed ER variables should be considered, such as Wang et al.'s (2012) method for modeling between child differences in within child variability considering both fluctuation amplitude and temporal dependency in the data.

However, statistically constructed indices of ER are purely objective measures of emotion recovery as constructed by the researcher. This approach did not account for children's subjective appraisals of emotion recovery, nor is the context in which the regulatory process occurred clear. A similar prospective study of ER in adolescents with (Silk, et al., 2011) assessed emotion recovery following participants' self-reported highest and lowest emotions in the reporting period and assessed the context of these emotions (i.e., stressful events). Silk et al.'s (2011) method of contextualizing emotion recovery in relation to specific stressors avoided some of the issues discussed above, but introduced more subjectivity through the participants' self-identification of emotion activation. Stress was not assessed on the e-diary, but may be uniquely important in the context of pediatric sleep and pain (Gil et al., 2003; Valrie et al., 2007a) and may be important to examine in the future. Additionally, examining the role of negative versus positive stressors may be important in contextualizing ER. For example, negative stressors may elicit a drop in positive emotions and an increase in negative emotions, whereas in the context of stressful positive life events, negative emotions may increase while positive emotions may be maintained or even increase. It will be important to employ measures of perceived stress intensity and stressful daily events in future research in order to both contextualize ER and to identify true instances of regulation, thus avoiding over interpreting natural fluctuations in emotion intensities. The

methods employed by Matthews and colleagues (Matthews, Owens, Allen, & Stoney, 1992; Matthews, Salomon, Kenyon, & Zhou, 2005; Raikkonen, Matthews, Flory, Owens, & Gump, 1999) may be modified for future EMA research on ER in pediatric pain. Primarily investigating the effects of stress on ambulatory blood pressure (ABP), Matthews and colleagues used a similar measure of momentary affect to that used in this study, with the addition of questions regarding location, social interaction, and valence of social interactions (conflict/disagreement vs. pleasant interactions) prior to the diary recording in order to contextualize ABP and affective intensity scores.

Similar to Matthews et al.'s studies (1992, 2005), it may be informative to incorporate measure of physiological reactivity in future research on the associations between emotional functioning, pain, and sleep in children with JIA or chronic pain. A variety of psychophysical ambulatory monitoring methods have been validated as measures of physiological affect and stress responses in laboratory-based emotion research (Wilhelm & Grossman, 2010) and have been used as proxies for affect dysregulation in adults (Ebner-Priemer et al., 2007). However, psychophysical reactivity varies with development and measurement techniques should be developmentally tailored for use in pediatric populations (Obradovic & Boyce, 2012; Spear, 2009).  Incorporating ambulatory psychophysiological monitoring would supplement e-diary reports in order to further contextualize emotional functioning and ER in daily life, which in turn may offer another option for validating a temporally constructed ER variable.

Despite employing the alternate cut point for identifying instances of emotion activation, results of the convergent validity assessment were not significant.  This may be due in part to the low overall levels of clinically significant anxiety and depressive symptoms

endorsed on the CDI and MASC.  These measures may not be sensitive enough to the

variability in emotional functioning across a generally well-adjusted sample to be optimal

measures for discriminant validity testing.   However, the pattern of null validity findings are

consistent with other studies examining emotion change scores via EMA, which found poor

correspondence between momentary mood assessment and questionnaires (Connelly et al.,

2012; Solhan, 2009) and highlight a need for further consideration of potential validation

methods.  In a study of emotion lability in adults with borderline personality disorder and

healthy controls, Solhan and colleagues (2009) found that scores on cross sectional mood

measures were generally not correlated with prospective indices of emotion lability, which

they attributed to differences in trait versus state assessments. Solhan and colleagues

suggested examining external correlates of emotion change indices (i.e., examining

predictive validation) in addition to using retrospective measures for convergent validation.

Using this approach in a previous study with a subset of this dataset (Connelly et al., 2012),

we found that the construct of momentary ER (constructed as originally proposed) was better

able to predict daily JIA symptoms than baseline measures of negative ER.  Of note, this

evidence of better predictive validity of the prospective ER construct is somewhat limited;

cross sectional measures of ER primarily assess behavioral responses to negative emotions

(e.g., ER strategies). It will need to be determined whether cross-sectional assessments of

positive emotional experiences have similarly poor predictive validity of important

symptoms and behaviors in children with JIA.  Although emotion recovery is an important

adaptive component of ER, it will be necessary to incorporate aspects of the broader subset

of processes composing ER (e.g., regulatory strategies, the role of parents in emotion

socialization, behaviors intended to prevent emotion activation) into future prospective

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research on ER in children with JIA and other chronic illnesses in order to better understand how ER functions in populations without psychopathology.

Finally, there are a few general statistical limitations that may be addressed in future research by applying an alternate modeling technique. I attempted to capture potential reciprocal or cyclical processes between sleep and pain in two ways: by testing the alternate model, and by controlling for evening pain intensity in an exploratory model. Some limitations regarding interpreting reciprocal influences between pain and sleep are described above, but it is also important to note that constructing two separate models to test both pathways of influence may not optimally capture cyclical temporal processes. Also, there was a large loss of degrees of freedom in the exploratory model controlling for evening pain, due to a larger number of missing observations; there were only 14 consecutive evening pain and morning sleep quality reports per child, on average. It is possible that there were systematic reason for missing reports, such as not completing diaries during severe pain flares, which could bias findings from this limited subsample of observations. In the future, it may be helpful to apply multivariate methods (e.g., multivariate MLM or multivariate latent curve models) that allow researchers to identify concurrent patterns of changes in multiple variables over time (MacCallum, 1997), while continuing to similarly accommodate missing data and different intervals between observations as offered by the current MLM analyses.

**Clinical Implications and Future Directions**

In conclusion, this study resulted in a number of important implications for future research and clinical practice. Future research should expand upon this study to identify specific aspects of sleep underlying global sleep quality scores that affect daytime pain intensity and functional limitations. Ongoing research on the mechanistic associations between sleep quality, regulation of positive emotions, and pain intensity will continue to help refine future intervention approaches. This study also has important implications for the ongoing development and use of electronic (e-health) tools to monitor and treat children with chronic illness and pain.

Clinically, children with JIA would benefit from learning additional techniques to promote maintenance of positive emotions. Findings from this study suggest that solely addressing negative emotions in children with JIA may not be as clinically useful as promoting positive emotions. Future interventions promoting ER in children with JIA should incorporate more techniques from positive psychology (e.g., mindfulness meditation, benefit finding) rather than solely relying on intervention techniques developed to address regulation of negative emotions in the context of internalizing disorders. Extending upon previous stress and coping interventions for children with chronic illness (Gil et al., 1996; Gil et al., 1997; Sansom-Daly, Peate, Wakefield, Bryant, & Cohn, 2012; Scholten et al., 2011), these findings suggest that there is a need to develop ER-focused interventions for children with chronic illness and pain, who often experience significant psychosocial challenges, but do not necessarily develop psychopathology.

Findings from this study emphasize the importance of clinically identifying children with JIA who typically experience both poor sleep quality and high pain intensity.  This subset of children may be at particular risk for poor functional outcomes and may need more comprehensive assessment and intervention services.  Findings suggest that intervening on pain intensity alone will not fully improve functional impairments; sleep interventions should also be incorporated into clinical care. In the clinical setting, in addition to routinely monitoring pain intensity via brief screening (e.g., pain VAS), all patients should be also screened for specific sleep problems, potentially using a standardized questionnaire, such as the Pediatric Sleep Questionnaire (Chervin, Dillon, Bassetti, Ganoczy, & Pituch, 1997; Chervin, Hedger, Dillon, & Pituch, 2000) or a brief, multidimensional sleep interview such as the 'BEARS' (B= Bedtime Issues, E=  Excessive Daytime Sleepiness, A = Night Awakenings, R = Regularity and Duration of Sleep, S = Snoring) (Owens, 2005) aimed at obtaining more detailed sleep information to inform treatment planning.  Children identified as experiencing sleep problems should receive education about developmental sleep needs and sleep hygiene strategies; referrals to behavioral or sleep medicine specialists should be considered for children who experience significant sleep disruptions.  In addition to monitoring sleep outcomes, pain and functional outcomes should be examined following sleep interventions in children with JIA.  There is a strong evidence base for cognitive-behavioral therapy for insomnia (CBT-I) in adults (M. T. Smith, Huang, & Manber, 2005), but in children, there has been less treatment outcomes research on CBT for sleep and only a few pain-focused CBT interventions include a specific sleep module (Degotardi et al., 2006; S. Kashikar-Zuck, Swain, Jones, & Grant, 2005).  Comparative trials of CBT for pain versus combined approaches have only recently been conducted in adults with chronic pain (Pigeon

et al., 2012; Von Korff et al., 2012).  Results of a pain intervention meta-analysis (Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010) demonstrated the need to supplement pain-focused treatments with other interventions in order to improve functional and emotional outcomes.

Findings from this study clearly support the association between sleep and pain in children with JIA and identify mechanisms affecting this relationship.  As combined interventions are developed for pediatric pain populations, it will be important to determine optimal combinations of pain and sleep treatment components; treatments may equally incorporate pain and sleep intervention strategies (similar to recent adults studies), or unique sleep treatment modules may supplement existing pediatric CBT pain interventions (similar to recommendations per Palermo, 2012).  Consistent with adult (Gatchel, Peng, Peters, Fuchs, & Turk, 2007) and child models of chronic pain (Varni, et al., 1996) and findings from this study, intervention techniques targeting positive emotions should also be incorporated into these treatments and functional limitations should be measured as a primary treatment outcome.
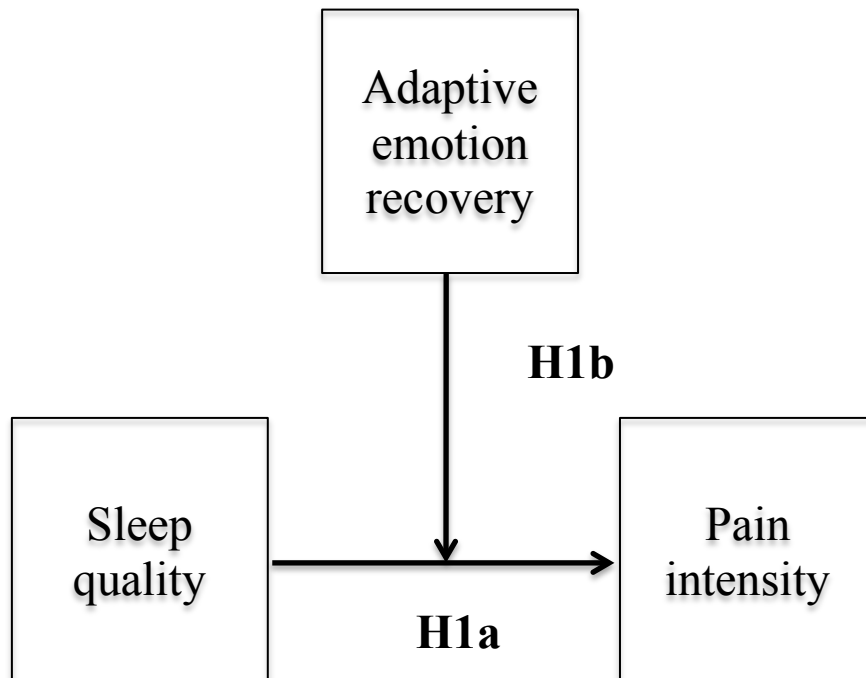
*Figure 1*. Model for Hypothesis 1. This diagram illustrates the significant pathways expected

between sleep quality and pain (H1a) and the role of adaptive emotion recovery as a

moderator of this relationship (H1b).

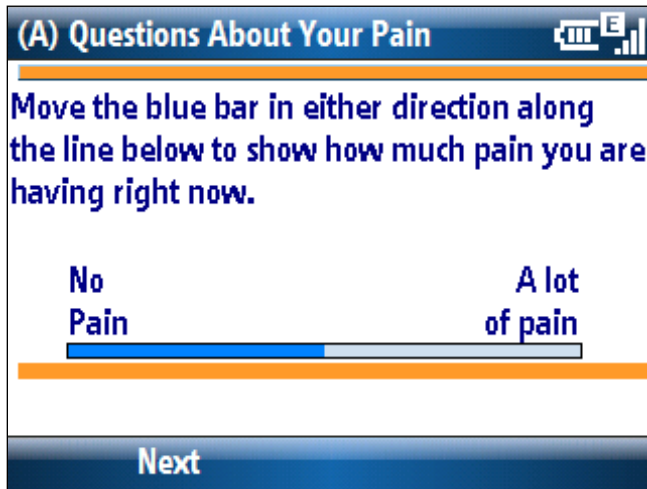*Figure 2*. T-Mobile ® Dash Smartphone used for e-diaries. All e-diary entries were made using the keypad.

*Figure 3.* E-diary electronic visual analog scale (e-VAS). The blue line appeared at the

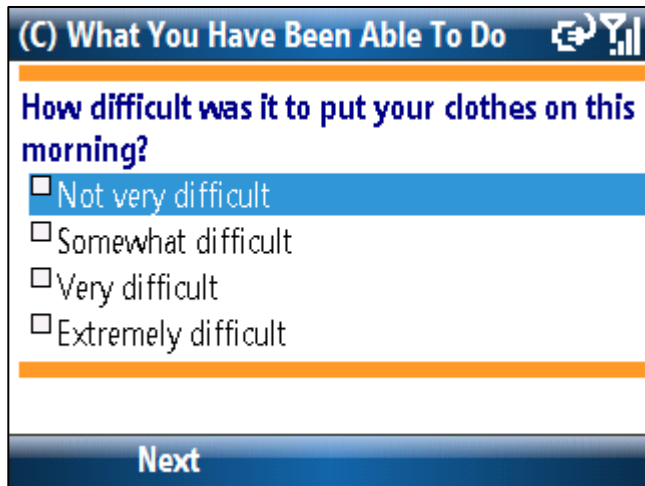midpoint and children used arrow keys to move the e-VAS to the appropriate spot.

*Figure 4.* E-diary multiple-choice item.  Children used keys to navigate to and select a response on multiple-choice items.

Table 1

*Quantifying ER: Steps for Identifying Instances of Emotion Activation and Coding Instances of Adaptive Emotion Recovery*

| Step | Variable | Task | Examine changes in PE and NE | | Code |
|------|----------|------|---------|---------|------|
| 1 | Emotion Activation | Identify times of emotion activation. | Is PE >.5SD below the child' mean PE score? | Is NE > .5SD above the child's mean NE score? | 0 = No emotion activation<br><br>1 = Emotion activation |
| 2 | Emotion Regulation | Examine next diary entry to determine if ER occurred. | Did PE return to >.5SD below the child's mean PE score? | Did NE return to <.5SD above the child's mean NE score? | 0= No emotion activation<br><br>1= Adaptive emotion recovery<br><br>2= Failure to recover to a typical emotion intensity |

*Note.* Separate variables were constructed to code for positive and negative emotion recovery.

PE= Positive emotion score averaged from PANAS-C e-diary items.

NE= Negative emotion score averaged from PANAS-C e-diary items

Table 2

*Sample Demographic Descriptive Information*

|  | m (SD) | Frequency |
|---|---|---|
| Age | 13.38 (2.78) | |
| Gender (% female) | | 74.1% |
| Race (% Caucasian) | | 73% |
| CDI t-score | 46.25 (9.37) | |
|    Clinical range | | 5.4% |
| MASC t-score | 50.80 (12.01) | |
|    Clinical range | | 14.3% |
| Disease activity | Minimal | 11% |
| | Mild | 42% |
| | Moderate | 43% |
| | Severe | 4% |

Table 3

*Electronic Diary Reports Descriptive Information*

|  | m (SD) | range |
| --- | --- | --- |
| Pain intensity | 26.29 (27.51) | 0-100 |
| PE score | 2.12 (.75) | 1-4 |
| NE score | 1.19 (.41) | 1-4 |
| Functional Limitations | 3.86 (7.26) | 0-32 |
| Sleep Quality | 67.94 (27.38) | 0-100 |

PE= Positive emotion score averaged from PANAS-C e-diary items.

NE= Negative emotion score averaged from PANAS-C e-diary items

Table 4

*Results of Hierarchical Multilevel Random Effects Analysis Predicting Pain Intensity and Sleep Quality*

| | Models predicting *pain intensity* | | | Alternative Models predicting *sleep quality* | | |
|---|---|---|---|---|---|---|
| Step | | **B** | **t** | | **B** | **t** |
| 2 | Age | -.90 | -.85 | Age | -.38 | -.42 |
| | Disease activity | 5.32 | 1.36 | Disease activity | -7.49 | -2.22* |
| 3 | Within-child sleep quality | -.07 | -3.20** | Within-child pain intensity | -.07 | -1.53 |
| | Between-child sleep quality | -.52 | -3.64** | Between-child pain intensity | -.40 | -3.74** |

| | Model 4 predicting *pain intensity* with the proposed ER construct | Model 4 predicting *pain intensity* with the alternate ER construct |
|---|---|---|
| | **F** | **F** |
| 4 | PE recovery | 12.45*** | 2.54 |
| | NE recovery | .42 | .14 |
| | Sleep quality x PE recovery | .22 | 3.78* |
| | Sleep quality x NE recovery | .42 | 1.84 |

*Notes*. The initial step of each model contained no predictors per procedures as detailed by Raudenbush & Bryk (2002). Average pain intensity for the day was used as a predictor of nighttime sleep quality in the model predicting sleep quality.*p* < .05, ** *p* < .01, *** <.0001
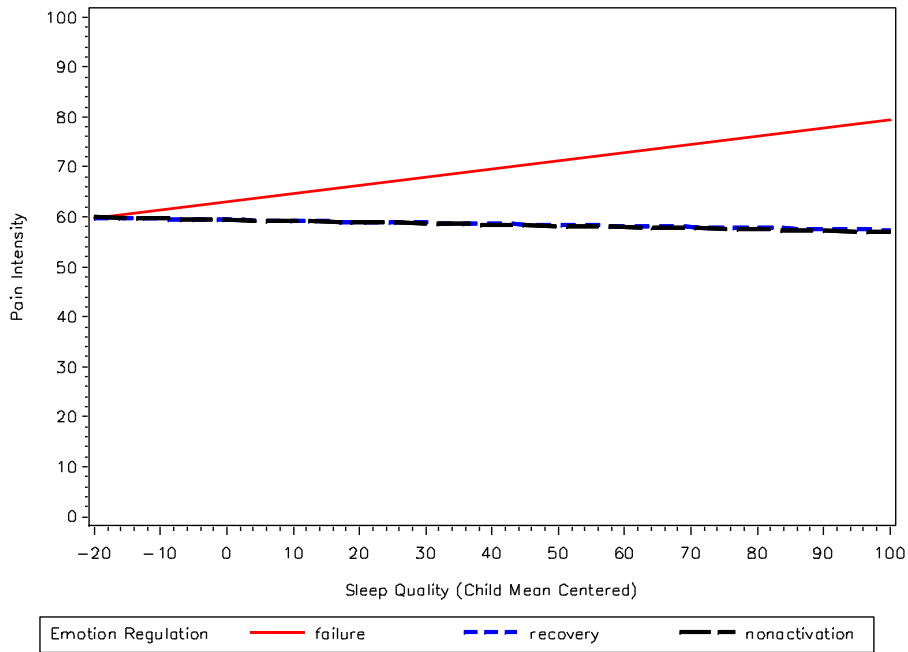
*Figure 5.* Interaction between sleep quality and alternate ER construct for positive affect regulation predicting pain intensity. At times of no positive emotion activation or when positive emotions were adaptively recovered to a typical level, pain intensity was slightly lower at higher levels of sleep quality, but failure to recover positive emotions in the presence of higher sleep quality resulted in higher pain intensity.
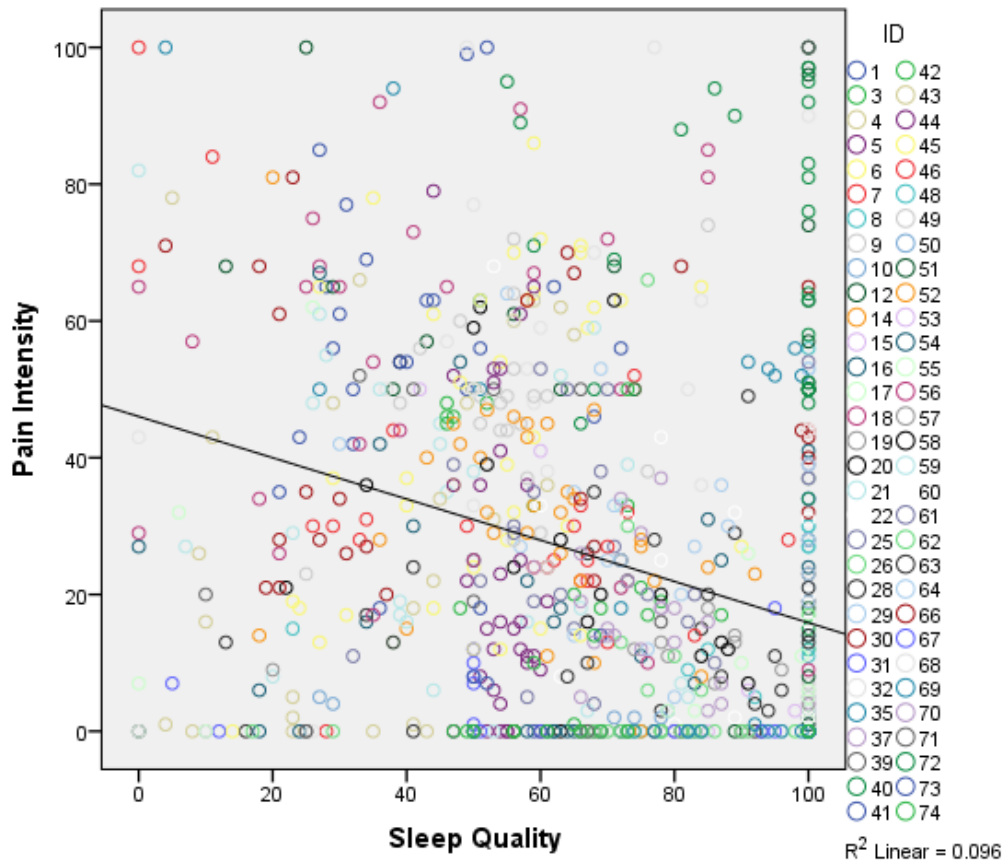
*Figure 6.* Graph of relationship between sleep quality and pain intensity at times when children experienced *no positive emotion activation.*
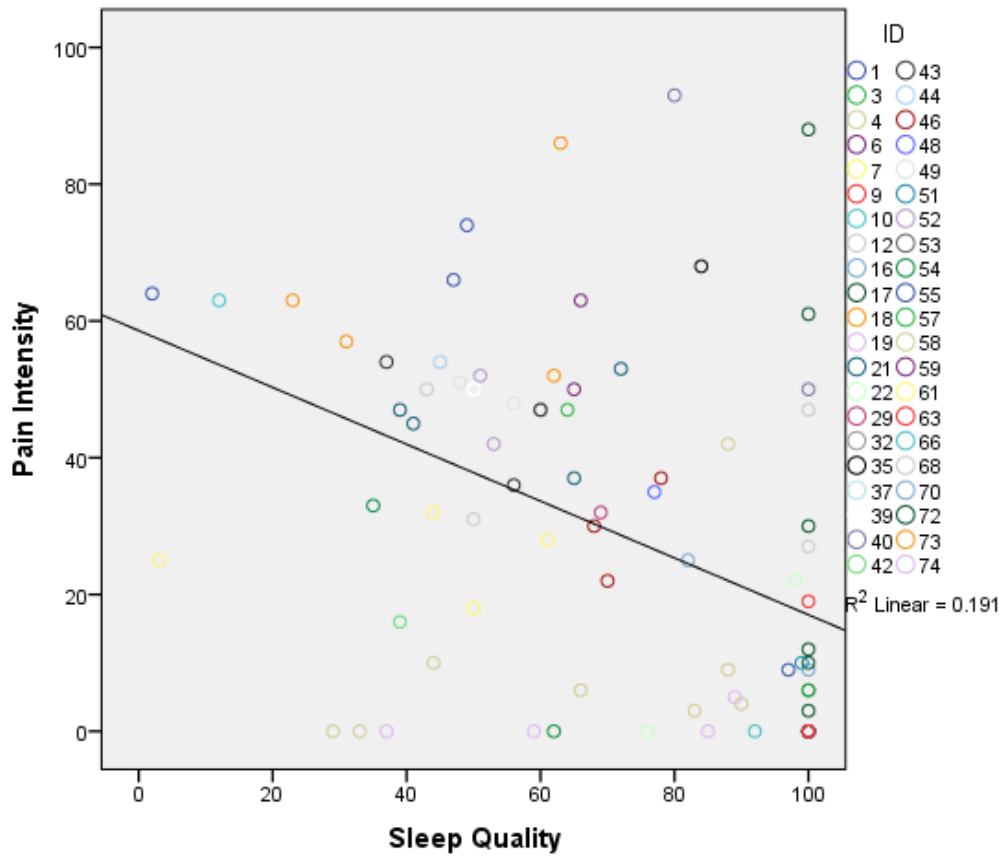
*Figure 7.* Graph of relationship between sleep quality and pain intensity at times when children experienced *recovery from drops in positive emotion.*
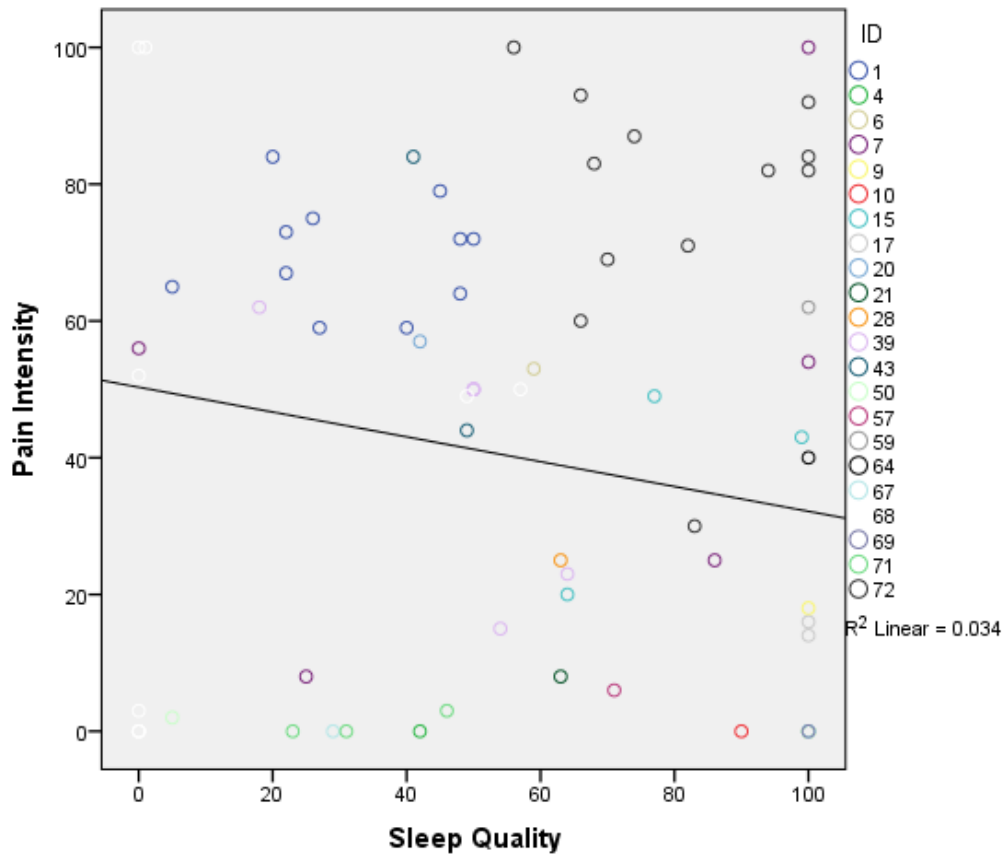
*Figure 8.* Graph of relationship between sleep quality and pain intensity at times when children *failed to recover from drops in positive emotion.*
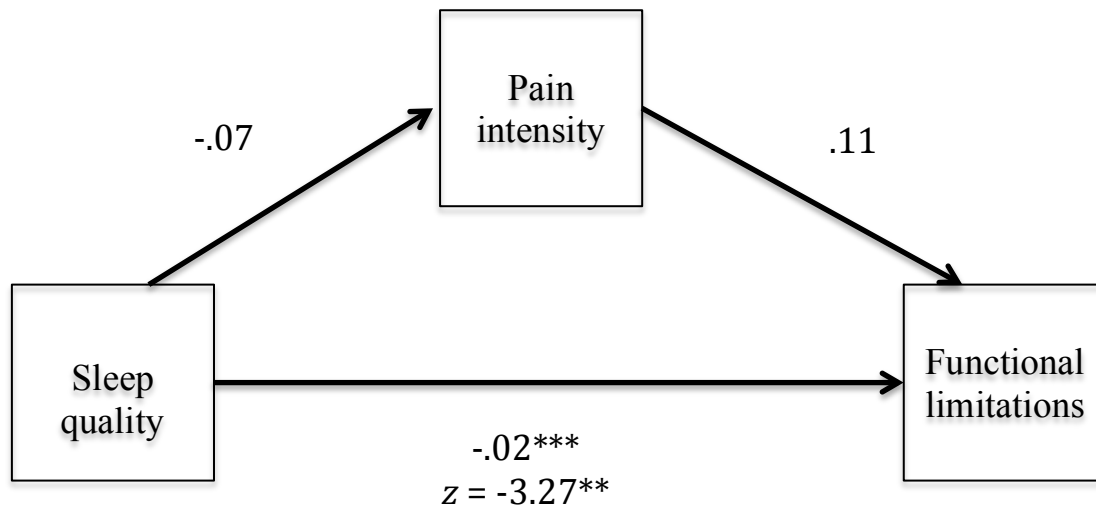
*Figure 9*. Level 1 within-child mediation model. *Momentary* pain intensity accounted for some of the relationship between sleep quality and functional limitations.

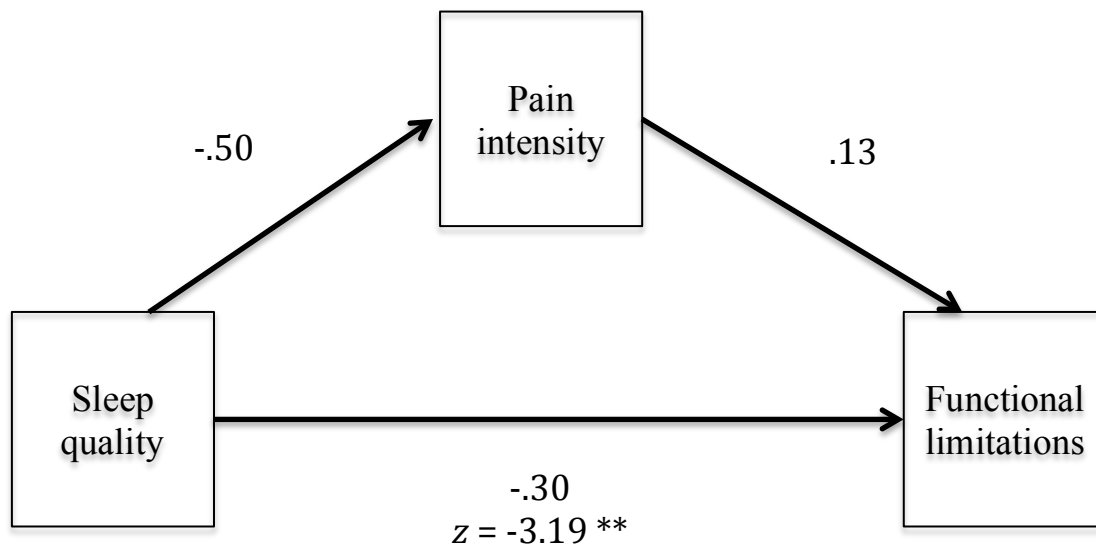** *p* < .001, *** *p* < .0001

*Figure 10.* Level 2 between-child mediation model. *Typical* pain intensity accounted for the relationship between typical sleep quality and functional limitations.
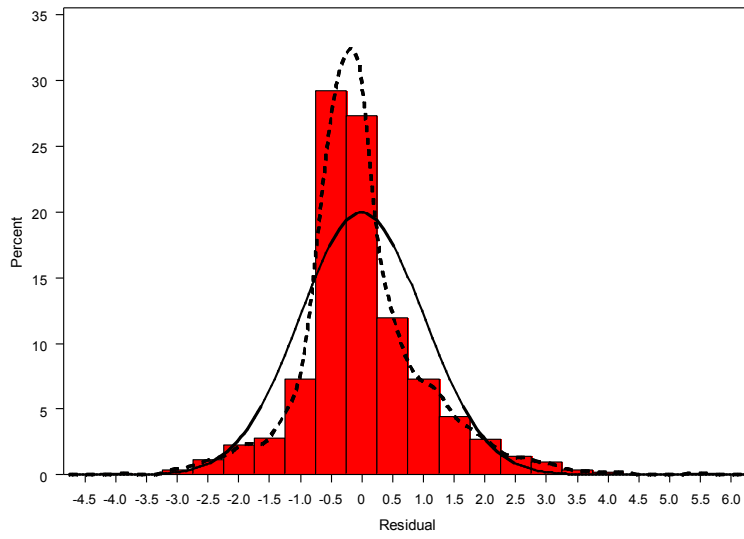
** *p* < .001, ****p* < .0001
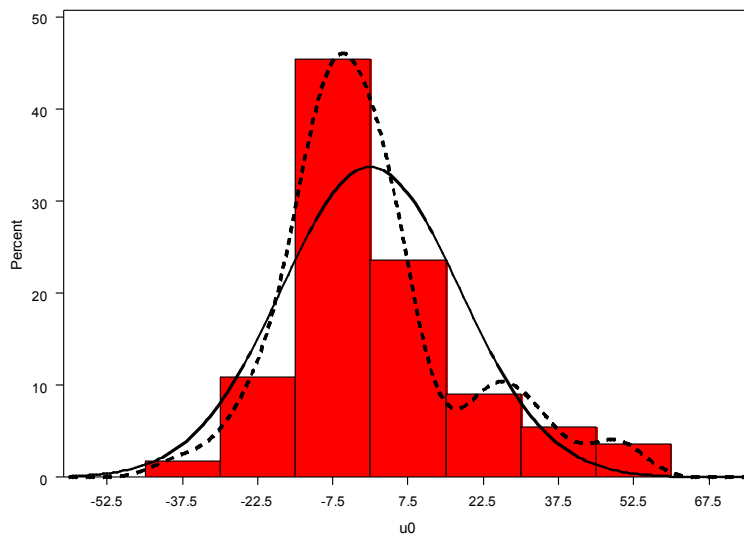
*Figure 11*.  Distribution of Level 1 Residuals

*Figure 12.* Distribution of Level 2 Residuals

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