Predictors of Sleep Quality in Children with Juvenile Polyarticular Arthritis

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A thesis submitted to the faculty of the University of North Carolina at Chapel Hill in partial fulfillment of the requirements for the degree of Master of Arts in the Department of Psychology (Clinical Psychology).

Chapel Hill
2009

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ABSTRACT

Maggie Hood Bromberg: Predictors of Sleep Quality in Children with Juvenile Polyarticular Arthritis
(Under the direction of Karen M. Gil, Ph.D.)

This study investigated the relationship between pain, coping efficacy, and sleep in 51 children and adolescents with polyarticular arthritis over a two month period. Consistent with expectations, pain significantly predicted sleep quality after controlling for disease and demographic variables. When the sample was restricted to days in which children experienced pain, this relationship was moderated by coping efficacy. The daily coping efficacy rating was found to be a valid measure of coping efficacy based on correlations to a validated self report measure of coping efficacy. This study also examined the coping of children in the sample. The use of specific coping strategies by parents and children was largely unrelated. These findings suggest that children who experience pain or have poor pain management may be at risk for sleep problems. As such, sleep quality should be assessed clinically and pain coping interventions should be developed to target sleep disturbances.
This work is dedicated to my family; my parents for their perpetual support and the value they have always placed on my education; my sister for the meals shared during the writing of this document, and my husband for his love and encouragement.
ACKNOWLEDGEMENTS

I would like to thank my Masters Committee for their guidance and support. I would also like to express my gratitude to my mentor, Dr. Karen Gil, for her mentorship and her outstanding example as a clinical researcher.
# TABLE OF CONTENTS

LIST OF TABLES ............................................................................................................................... vii

LIST OF FIGURES ........................................................................................................................... vii

ABBREVIATIONS .............................................................................................................................. ix

PREDICTORS OF SLEEP QUALITY IN CHILDREN WITH

JUVENILE POLYARTICULAR ARTHRITIS ........................................................................................................ 1

- *Sleep in Children with Recurrent Pain and Chronic Illnesses* .................................................... 1
- *Coping* ............................................................................................................................................. 5
- *Current Study and Hypotheses* .......................................................................................................... 6
- *Daily Diary* ...................................................................................................................................... 8

Method .................................................................................................................................................. 9

- Participants ......................................................................................................................................... 9
- Procedure .......................................................................................................................................... 10
- Measures .......................................................................................................................................... 11

Results ................................................................................................................................................ 14

- Descriptive Statistics .......................................................................................................................... 15
- *Pain and Coping Efficacy as Predictors of Sleep Quality* ............................................................... 15
- *Baseline Coping Efficacy and Longitudinal Coping Efficacy* ......................................................... 18
- Parent and Child Coping Strategies ..................................................................................................... 18

Discussion ........................................................................................................................................... 19

REFERENCES ........................................................................................................................................ 34
LIST OF TABLES

Table

1. Means and Standard Deviations of Aggregated Daily Variables ...................27
2. Summary of Hierarchical Regression Analysis Predicting Sleep Quality.........28
3. Summary of Hierarchical Regression Analysis Predicting Pain Rating............29
4. Summary of Hierarchical Regression Analysis Predicting Sleep Quality on Pain Days..........................30
5. Means and Standard Deviations of Coping Strategies for Children Subscales ....31
LIST OF FIGURES

Figure

1. Moderation Model for the Effect of Pain Coping Efficacy on the Relationship between Pain and Sleep Quality .................................................................32

2. Interaction between Pain and Coping Efficacy on Pain Days ........................................33
### ABBREVIATIONS

<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>DMARDs</td>
<td>Disease Modifying Antirheumatic Drugs</td>
</tr>
<tr>
<td>NSAIDs</td>
<td>Nonsteroidal Anti-inflammatory Drugs</td>
</tr>
<tr>
<td>PGA</td>
<td>Physician Global Assessment</td>
</tr>
<tr>
<td>SCD</td>
<td>Sickle Cell Disease</td>
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<td>VAS</td>
<td>Visual Analog Scale</td>
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PREDICTORS OF SLEEP QUALITY IN CHILDREN WITH JUVENILE POLYARTICULAR ARTHRITIS

Sleep has been identified as a critical element of health, physical development, and wellbeing in healthy children. In children with chronic illnesses, sleep disruptions from pain as well as physiological and psychological distress may effect psychosocial adjustment (Mindell et al., 1999). However, little research to date has explored the complex relationship between pain and sleep in children with arthritis.

Sleep is a developmentally important physiological function, characterized by different patterns across childhood and adolescence. Preadolescent, school-aged children generally sleep between 9-10 hours, experience little daytime sleepiness (Wolfson, 1996), and enjoy largely uninterrupted deep sleep (Anders, Sadeh, & Appareddy, 1995). During puberty the characteristics of sleep are affected by physiological and psychosocial changes. Adolescent sleep is characterized by increased need for sleep, shorter nightly sleep duration, and more daytime sleepiness (Carskadon, 2002). Health problems, anxiety, and depression can result in sleep disruptions in both children and adolescents. Disruptions in sleep can have adverse effects on physical growth, cognitive functioning, and emotional functioning (Wolfson, 1996). Moreover, poor sleep may lead to increased daytime sleepiness and disruptions in attention, learning, mood, and behavior. Given the physiological and emotional stress associated with physical illnesses, children with illnesses are at-risk for sleep problems (Mindell et al., 1999).
Sleep in Children with Recurrent Pain and Chronic Illnesses

In children and adolescents with recurrent pain, such as functional abdominal pain, research has shown that children experience difficulty initiating and maintaining sleep even when adjusting for symptoms of depression and anxiety, which may contribute to sleep difficulties independent of medical status (Huntley, Campo, Dahl, & Lewin, 2007). Furthermore, delays in sleep onset are a common complaint in children with abdominal pain and children with headache (Bruni, Russo, Violani, & Guidetti, 2004; Gilman, Palermo, Kabbouche, Hershey, & Powers, 2007; Haim et al., 2004). Recently, a longitudinal study of sleep and pain in children ages 8-12 years with sickle cell disease (SCD) detected a bi-directional or cyclical relationship between sleep and pain. Increased daily pain correlated with decreased sleep that night, which in turn predicted pain the next day (Valrie et al., 2007a).

Increasingly, the nature of sleep disruptions in children with juvenile arthritis has emerged as a concern (Labyak, Bourguignon, & Docherty, 2003). Juvenile arthritis is a childhood rheumatologic disease affecting the joints. It is characterized by recurrent pain, joint contractures and inflammation, and growth abnormalities. Prevalence estimates in American populations have ranged from around 100,000 (Towner, Michet, O’Fallen, & Nelson, 1983) to 300,000 children under the age of 16 years (Cassidy, & Nelson, 1988; Gewanter, Roghmann, & Baum, 1983). Juvenile arthritis encompasses a heterogeneous group of childhood arthritis diagnoses, each distinguished by the number of affected joints, accompanying symptoms (e.g., fever, rash), and the presence or absence of antibodies such as rheumatoid factor. Polyarticular involvement is considered a more severe disease presentation and is diagnosed by the presence of active arthritis in five or more joints (Petty
et al., 2004). Recurrent musculoskeletal pain is a primary symptom across disease subtypes and may affect functioning across psychosocial domains. Due to the burdens associated with their illness, children with juvenile arthritis may be at risk for adjustment problems, including internalizing symptoms (LeBovidge, Lavigne, Donenberg, & Miller, 2003). Treatment of pediatric arthritis is targeted at reduction in pain and disease symptoms, reduction in joint inflammation, improvement in joint range of motion, and improvement in daily physical functioning and activities (Giannini, Ruperto, Ravelli, Lovell, Fellson, & Martini, 1997). Nonsteroidal anti-inflammatory drugs (NSAIDs), along with intra-articular steroids, are often the first-line treatment. Disease modifying antirheumatic drugs (DMARDs) such as methotrexate are used in many cases and new biologic agents may be part of treatment as well (Ilowite, 2002).

The nature of sleep disturbance in pediatric arthritis is beginning to be explored via both direct, observational measures (i.e., laboratory sleep studies) and subjective, retrospective measures (i.e., sleep questionnaires). Adults with rheumatoid arthritis experience reductions in sleep and poor sleep quality due to pain (Drewes et al., 1998). Previous research on sleep in children with arthritis consistently indicates that these children experience more sleep disturbances than healthy controls (Zamir, Press, Asher, & Tarasiuk, 1998; Amos, Curry, Drutz, Frost & Warren, 1997), but the differences in specific sleep disruptions and sleep behaviors and their relationship to disease variables and medication use continue to be explored and debated (Passarelli, et al., 2006; Labyak et al., 2003).

Recent studies have focused on identifying specific aspects of sleep disruptions in pediatric arthritis patients. Bloom et al. (2003) found that parents of 6-12 year old children with arthritis rated the children as having significantly higher levels of overall sleep
disturbance, night awakenings, parasomnia, sleep anxiety, sleep disordered breathing, and early morning waking/daytime sleepiness than age and sex matched healthy controls. Child ratings of overall sleep disturbance and average pain were correlated. A number of factors limit the interpretations of these data, including the small sample size, the variations in disease characteristics across subtypes, and the possible effects of pain medications on sleep, which were not controlled for. In a recent study using objective sleep measures, Passarelli and colleagues (2006) found that sleep disturbances were significantly higher in children and adolescents with arthritis than in matched controls. Sleep difficulties in the arthritis sample were characterized by lower sleep efficiency, higher arousal index, and frequent leg movements and a relationship between sleep and pain was detected. Passarelli et al. (2006) controlled for the effects of medication on sleep by having subjects discontinue all NSAIDs, DMARDs, steroids, and other immunosuppressors two days prior to the sleep study; however, it is unclear whether this brief medication hiatus truly removed the possible drug influences on sleep and the authors did not analyze sleep data by medication category. The relationship between sleep and medications commonly used in the management and care of arthritis is not well defined, although NSAIDs have been shown to have a deleterious affect on sleep in healthy samples (Labyak et al., 2003) and have been linked to reported decreased sleep disturbances in adults with rheumatoid arthritis (Lavie, Nahir, Lorber, & Scharf, 1991). It is possible that there is a similar relationship between medication use and sleep in children with arthritis, but further research is necessary to determine the effects of specific medications on nightly sleep.

In summary, research to date has focused on describing the nature of sleep disturbances in children with arthritis and in establishing the relationship between pain and
sleep in this recurrent pain population. However, other psychosocial variables that may affect this relationship, such as mood, stress, and coping, have not yet been examined. Furthermore, the influence of poor sleep on pain has not been described in this population and the relationship between pain and sleep has not been investigated longitudinally to determine the relationship between these variables over time.

*Coping*

Children with juvenile arthritis face ongoing, recurrent pain and its associated health consequences as well as functional and psychosocial burdens. As such, how the child and parent cope with pain is important. Coping can lessen the potential psychological distress resulting from recurrent pain, which can in turn promote better adjustment in a variety of domains.

Most conceptual models of adjustment in children with chronic illnesses consider the interaction between stressors and coping resources. Developmentally appropriate models for children have been proposed (Compas, Connor-Smith, Saltzman, Thomsen, & Wadsworth, 2001), largely based on the coping model developed by Lazarus and Folkman (1984). The most influential model of child coping is the Transactional Stress and Coping Model (Thompson, Gil, Burbach, Keith, & Kinney, 1993a, 1993b; Thompson, Gustafson, Hamlett, & Spock, 1992a, 1992b) which presents adaptational processes such as cognitive appraisals of stress, locus of control, and efficacy; coping strategies; and family processes as mediators between child and disease demographics and overall adjustment outcomes. Maternal and child coping and adjustment are interrelated in this dynamic process, with each influencing the other’s adjustment and subsequent adaptive processes. The utility of this model has been supported in children and adolescents cystic fibrosis and SCD (Thompson et al., 1993a,
1993b). The Transactional Stress and Coping Model and other child stress and coping models take into consideration the unique cognitive, social, and emotional development of children and the influence of development on the coping process. For example, in pediatric populations children’s concept and understanding of illness evolves over time (Berry, Hayford, Ross, Pachman, & Lavigne, 1993; Thompson & Gustafson, 1996). It is likely that increasingly sophisticated concepts of disease will result in different selections and applications of coping strategies.

Another central issue in coping is coping efficacy, that is, the self-perceived effectiveness of the individuals’ coping responses. As coping responses can buffer the effects of stress (Compas, Worsham, & Eys, 1992), this self-evaluation of successful coping can serve as an estimate of the impact of stress on the individual. Barlow et al. (2001) defined coping efficacy as “children’s perceived ability to control, or manage, salient aspects of life with juvenile rheumatoid arthritis” (p.160). The majority of coping measures for children are designed to assess specific coping strategies and their individual success in responding to the stressor (Blount et al., 2008) and often do not include a measure of overall coping efficacy.

Coping is a critical aspect of the adjustment process with outcomes affecting psychological functioning, social functioning, and health. A number of models have been proposed to describe the relationship between stress, coping, and adjustment. Although the relationship between coping and psychological adjustment has been described in children with arthritis and other pediatric pain populations, researchers are only beginning to address the relationship between coping and health related behaviors, such as sleep.

*The Current Study and Hypotheses*
Based upon conceptual models of childhood pain and coping, the current study examined the relationship between pain, coping efficacy, and sleep in children and adolescents with polyarticular juvenile arthritis over a two-month period. For this study, coping efficacy was used as a post hoc, global measure of the feelings of control associated with coping attempts made by the child. The second purpose of this study was to examine aspects of coping in children with arthritis in more detail, given the important role of coping in adjustment to chronic illness and pain. Specifically, I investigated the correlation between baseline reports, and aggregate daily reports of coping efficacy of children with arthritis. This relationship was be analyzed to determine if these reports are consistent, valid, and reliable. I also examined the relationship between parent and child coping at baseline, given previous research that suggests that maternal coping and adjustment are related to the child’s coping and adjustment in pediatric chronic illness populations.

**Specific Aim 1:** To examine the impact of pain on sleep quality in children with arthritis.

**Hypothesis 1:** Higher pain ratings will predict poorer sleep quality in children with juvenile polyarticular arthritis.

**Hypothesis 2:** The relationship between high pain and poor quality of sleep will be weakened at increasing levels of coping efficacy (see Figure 1).

**Specific Aim 2:** To examine the measure of coping efficacy in more detail by investigating the consistency in reports of child coping efficacy between child baseline retrospective reports and child longitudinal daily reports, as well as the consistency between parent and child reports of coping efficacy.
Hypothesis 3: Baseline child ratings of coping efficacy will predict the aggregate coping efficacy ratings from the daily diary.

Hypothesis 4: Baseline parent self-reports of coping strategies and coping efficacy will predict child baseline self-reports of coping strategies and coping efficacy.

Daily Diary

In order to accomplish the goals of this study, a prospective daily diary was implemented. Recently, in order to understand variations in health symptoms such as pain and sleep patterns, researchers studying chronic illnesses have begun to utilize strategies such as daily diaries. Prospective longitudinal studies that use daily diaries are preferable to retrospective summary methods for determining the frequency, characteristics, and effects of chronic and recurrent pain and other symptoms. Retrospective pain assessments conducted at a single time point may introduce increased recall bias (Butz, 2004; van der Brink, Bandell-Hoekstra, & Abu-Saad, 2001). Conversely, daily diary methodology reduces recall bias and allows for naturalistic data collection and captures day-to-day variations in symptoms. Diaries offer many advantages when examining individual differences in the pain experience, fluctuations in pain levels, and the ongoing process of adjustment to pain. The final advantage of daily data collection is the application of statistical analysis methods that allow researchers to investigate the temporal relationships amongst variables, thus capturing the unique richness of longitudinal data.

The use of daily diaries has been validated in pediatric SCD patients (Gil et al., 2000; 2003) and has resulted in important knowledge regarding the pain experience in children with arthritis and pain’s relationship with other variables such as daily mood, coping, and functional impairment (Schanberg et al., 2000, 2003, 2005). Findings indicated that daily
mood, stress, and pain are predictive of daily functioning and that children with juvenile polyarticular arthritis experience daily fluctuations in symptoms and more frequent pain than previously indicated based on single time point data collection (Schanberg et al., 2003, 2005). Arthritis pain and adjustment are ongoing experiences best understood by longitudinal studies.

Method

Child and parent dyads were recruited as part of a larger, longitudinal study investigating the daily pain, mood, stress, and disease activity of children with polyarticular arthritis (Schanberg et al., 2003, 2005). The complete dataset contained daily dairies of pain, mood, stress, coping and disease symptoms completed by 51 children ages 8-16 years over a two month period. Additionally, in the complete dataset, there are self- and parent-report questionnaires (e.g. mood, coping, stress, physical function) and biological measures (e.g., erythrocyte sedimentation rate, hemoglobin, prolactin, IL-2 receptor) obtained at baseline and every two weeks. Potential participants in the overall study were excluded if they were currently using psychotropic medications or systemic steroids, due to the collection of hormonal measures as part of the larger study.

To date, two publications regarding the correlates of pain in children with arthritis have resulted from this line of inquiry (Schanberg et al., 2003, 2005). The current study uniquely focused on variations in sleep and coping by analyzing data obtained from the child-completed daily diary. These data have not been examined previously.

Participants

Patients were recruited from the patient populations of Duke University Medical Center. All participants experienced arthritis onset prior to the age of 16 years, had arthritis
in 5 or more joints (i.e., polyarticular arthritis), and had experienced arthritis for at least 6 weeks. The study sample consisted of 51 patients (33 girls, 18 boys) with the mean age 12.4 years (SD = 2.8 years), and one of their parents. The majority of the sample was Caucasian (88%), reflecting the population of the Pediatric Rheumatology Clinic at Duke University Medical Center. In regard to disease characteristics, the average time since diagnosis or disease duration was 4 years (range = 3 months to 12 years) and the mean physician global assessment score was 31.5 (SD = 25.6) on a 100 mm visual analog scale (VAS), indicating that children in the sample experienced mild to moderate arthritis on average. Most were taking medications, with the majority of participants taking a NSAID (86.3%) alone or in addition to methotrexate (72.5%). Children reported pain on 76% (SD = 34.8%) of days during the study period with the average pain rating on days in which pain occurred of 29.9 (SD = 19.2) on a 100 mm VAS.

Procedure

The study protocol was approved by Institutional Review Board at Duke University Medical Center. Eligible patients from this population were contacted regarding the study. Upon completing informed consent, children and parents completed baseline measures of coping and were trained on a daily recording measure by a research assistant. Children were instructed to complete daily reports of symptoms including pain, mood, fatigue, stress, coping efficacy, and sleep without the assistance of a parent. Children completed daily diaries each evening and returned them in provided envelopes by mail the following morning. Diaries were completed for approximately two months. A research assistant contacted each participant twice during the initial week and once a week thereafter throughout the recording period to confirm ongoing study participation and to reinforce the
independent completion of the daily report. Parents received $25 upon entry to the study to reimburse for travel related expenses. Children were reimbursed $0.50 for each diary completed and an extra $1.25 for each week in which all 7 daily recordings were returned.

**Measures**

**Daily Diary.** The daily dairy measure used for this study was based on a previous version of the diary (Gil et al., 2000, 2003; Schanberg et al., 2000) that was developed for children with chronic illnesses such as sickle cell disease or chronic arthritis. Specific variables from the diary analyzed for the present study will include pain, sleep quality, and coping efficacy.

**Arthritis-related pain.** Children were asked to rate their average level of pain for the day on a 100 mm VAS taken from the Pediatric Pain Questionnaire (Varni, Thompson, & Hanson, 1987), anchored by “No hurting, No discomfort, No pain” and “Hurting a whole lot, Very uncomfortable, Severe pain.” Previous research by the Varni and colleagues (1987) has established the reliability and validity of this commonly used measure of musculoskeletal pain.

**Quality of sleep.** Each day, children reported quality of sleep during the previous night on a 100 mm VAS anchored by “Did not sleep well” and “Slept very well.” This scale was the primary rating of sleep quality for the present study. The use of daily sleep measures has been found to be reliable and valid in comparison to physiological sleep measures (Gaina, Sekine, Chen, Hamanishi, & Kagamimori, 2004; Sadeh, Raviv, & Gruber, 2000) and is considered optimal for assessing sleep patterns (Knab & Engel-Sittenfeld, 1983; Wolfson et al., 2003). Recent research with pediatric SCD patients further supports the reliability and validity of daily sleep diaries (Valrie et al., 2007a, 2007b, 2007c). Overall, results of these
studies found adequate consistency between parent and child baseline reports and daily reports, further supporting the use of daily diaries in measuring sleep and disease-related variables in a pediatric population. However, consistent with previous research, Valrie and colleagues found the average daily report of sleep quality to be significantly higher than baseline reports of sleep quality, suggesting that daily reports are less influenced by recall bias than baseline measures (Valrie et al., 2007a).

Coping efficacy. Each day, children rated their perceived ability to control pain on a question taken from the Coping Strategies Questionnaire for Children (CSQ-C) (Gil et al., 1991, 1993; Gil, Wilson, & Edens 1997; Schanberg et al., 1996, 1997). This coping efficacy item asked the child to circle one response to the question “Based on all the things you did to cope, or deal with your pain today, how much control do you feel you had over the pain?” Answers were presented on a 4-point Likert-type scale ranging from “No control” to “Complete control.” This item served as the primary measure of daily coping efficacy in the data analysis.

Baseline measures

Parent coping strategies. Each parent completed the Coping Strategies Questionnaire (CSQ; Rosenstiel & Keefe, 1983), a self-report measure consisting of 42 items regarding strategies often used by individuals experiencing pain. Since the original measure was developed for adults with recurrent or chronic pain, the instructions were modified for use with parents, given that they may not experience recurrent pain. Parents were asked to consider any time they have experienced pain such as a toothache, a headache, or a minor injury such as a cut or bruise. Parents were instructed to rate how frequently they use each strategy in response to pain on a 7-point scale ranging from “never do that” to “always do
that.” Items include strategies such as “I try to think of something pleasant,” “I tell myself I can overcome the pain,” and “I pretend it’s not there.” The specific coping strategies are grouped into 7 coping response categories: Diverting Attention, Reinterpreting Pain Sensations, Coping Self-Statements, Ignoring Sensations, Praying/Hoping, Catastrophizing, Increase Behavioral Activities. This measure also has a global rating of the individual’s overall ability to control pain. This question asks “Based on all the things you did to cope, or deal with your pain, on an average day, how much control do you feel you have over it?” Parents rated their sense of control on a 0-6 scale anchored by “not control” and “complete control.” The “Control” item will be used as the primary parent coping efficacy variable in data analysis. The CSQ is a valid and reliable measure of pain coping strategies (Keefe et al., 1987, 1990, 1997) that has been widely used as a measure of pain coping strategies with adult pain patients (Jenson, Turner, Romano, & Karoly, 1991) and was adapted for use with pediatric pain patients and their parents.

Child coping strategies. Each child completed the Coping Strategies Questionnaire for Children (CSQ-C; Rosenstiel & Keefe, 1983; Schanberg et al., 1996), a self-report child version of the Coping Strategies Questionnaire. Children were instructed to rate how much they used each coping strategy in response to pain on the same scale as adults ranging from “never” to “always.” The individual items and scales are identical to the adult version, with developmentally appropriate modifications to individual items. For example, the second item on the adult version is “I leave the house and do something, such as going to the movies or shopping.” On the child version, shopping was replaced with “going for a walk.” The CSQ-C items compose 7 coping strategy scales that are identical to the adult measure. Children also rated their coping efficacy on an item that was identical to the item on the adult
version, with the instructions “Based on all the things you do to cope, or deal with your pain on an average day, how much control do you feel you have over it?”

This measure has been widely used with adult pain patients and was adapted for use with pediatric pain patients. Reliability and validity of the CSQ-C has been shown for children with SCD pain, fibromyalgia, and arthritis (Gil, Abrams, Phillips, & Keefe, 1989; 1992; Gil et al, 1991; 1993; Schanberg et al., 1996, 1997).

Depressive symptoms. Children completed the Children’s Depression Inventory (CDI; Kovacs, 1992) a 27-item self-report scale assessing depressive symptoms including sleep disturbance, appetite loss, interpersonal problems, negative self-esteem, and dysphoria. The measure possesses adequate reliability and validity and is the most widely used and researched measure of childhood depressive symptoms in children 8 to 17 years of age (Kovacs, 1992).

Disease severity. As part of the larger study, during a clinic visit at the start of the study, the pediatric rheumatologist completed a physical examination. Results of this examination served to inform her rating on a global assessment of disease severity for each child. A 100 mm VAS anchored by the points “asymptomatic” and “very severe” was used to measure disease severity. This measure has been widely used across rheumatology research and is designated as part of the core set of outcome variables in juvenile arthritis clinical research (Giannini et al., 1997).

Results

Results regarding the relationship between pain severity and sleep quality will be presented first, including post hoc analyses, followed by results for models investigating the characteristics of coping in this sample. Prior to analysis, the daily reports for pain, sleep,
and coping were aggregated across each participant to obtain an average rating of each variable over the course of the 2 months or 60 days of the study. Continuous variables (i.e., pain, sleep quality, disease severity, coping efficacy) were centered such that the mean is equal to 0. Disease severity as assessed by physician global assessment (PGA) at baseline and age of participant were controlled for in the linear regression models predicting sleep quality.

Descriptive Statistics

On average, children in the sample completed diaries on 84% of days during the 60 day response period. Completion rate was not significantly correlated with baseline pain rating, age, or disease severity. Similarly, results of independent sample t-tests indicate that the number of completed diaries was not significantly different between Caucasian and African American participants and male and female participants.

The means of the aggregate values for age, PGA, pain rating, coping efficacy rating, and sleep quality rating are presented in Table 1. Overall, children in the sample experienced mild to moderate pain ($M = 26.79, SD = 21.25$) and moderate to high sleep quality ($M = 75.25, SD = 18.53$). In regards to coping efficacy, children endorsed an average score of 1.80 ($SD = 0.82$) indicating moderate perceived control over pain as a result of all coping efforts.

Pain and Coping Efficacy as Predictors of Sleep Quality

Hypothesis 1, that high pain severity ratings would predict low sleep quality ratings was tested using hierarchical linear regression (see Table 2). Covariates to control for age and disease severity were entered at Step 1 and neither age nor disease severity predicted significant variance in aggregate sleep quality. Pain was entered into the model at Step 2 and resulted in a significant full model ($R^2 = .39, p < .0001$), indicating that pain predicted a
significant amount of variance in sleep quality beyond the effects of age and disease severity ($\Delta R^2 = .34, p < .0001$).

Additional predictors were added to the regression model at Steps 3 and 4 to test the hypothesis that the relationship between high pain ratings and lower sleep quality would be weakened at increasing levels of coping efficacy. Coping efficacy was entered into the model at Step 3 and the main effect of coping efficacy was not significant ($\Delta R^2 = .01, p > .05$). Similarly, the interaction between pain and coping efficacy was not significant ($\Delta R^2 = .03, p > .05$). The full regression model uniquely accounted for 43% of the variance in sleep quality.

Due to the potentially reciprocal relationship between pain and sleep, the alternate hypothesis was also tested to determine if coping efficacy and sleep predicted significant variance in reported pain when controlling for age and disease severity (see Table 3). Covariates were entered into the model at the first step and the main predictors and interaction term were added at later steps. Results indicate that age and disease severity predicted 12% of the variance in reported pain ($p < .05$). When controlling for age and disease severity, sleep quality significantly predicted pain ratings ($\Delta R^2 = .32, p < .05$). Coping efficacy accounted for an additional 12% of the variance in pain ($p < .05$), but the interaction between sleep quality and coping efficacy was not significant ($\Delta R^2 = 0, p > .05$). The full model accounted for 56% of the variance in pain ratings.

Finally, post hoc analyses were conducted to determine if coping efficacy moderated the relationship between pain severity and sleep quality on days when pain was reported. Children reported no pain on 18% of diary days. On days when no pain was reported, 81% of children reported complete control over their pain, which may have artificially inflated the
average coping efficacy score. Descriptive statistics for average pain, coping efficacy, and sleep on pain days are reported in Table 1. A hierarchical regression analysis was conducted to test hypotheses regarding pain and the interaction between pain and coping efficacy as predictors of sleep quality, limiting the sample to days when pain was reported, as shown in Table 4. Again, covariates were entered into the model at Step 1 and did not account for a significant amount of variance in sleep quality ($R^2 = .06, p > .05$). Consistent with hypothesis 1, pain rating predicted a significant amount of the variance in sleep quality ($\Delta R^2 = .37, p < .0001$), when limited to days when pain was reported. Again, coping efficacy did not account for a significant portion of the variance in sleep quality in this analysis ($\Delta R^2 = 0, p > .05$). The results of Step 4 diverged from the results of analyses using the aggregate scores from all diary days; in the sample restricted to pain days, the relationship between pain rating and sleep quality was moderated by coping efficacy ($\Delta R^2 = .06, p < .05$). These results indicate that coping efficacy played an important role in explaining the relationship between pain and sleep; however, the direction of this interaction was unclear.

In order to further investigate how coping efficacy interacted with pain in relation to sleep quality, a series of regression models, as outlined by Aiken and West (1991), were conducted. The relationship between pain severity and quality of sleep was analyzed at each of three levels of pain coping efficacy: low (1 standard deviation below the mean), medium (at the mean), and high (1 standard deviation above the mean). This method allows results to be immediately comparable with standard ANOVA procedures and outcomes (Aiken & West, 1991). Simple slopes for each level of coping efficacy were calculated and plotted using the interactive calculation tool developed by Preacher and colleagues (2006). As seen in Figure 2, pain and sleep quality were negatively related at each level of coping efficacy,
indicating that as pain increased sleep quality decreased correspondingly. This relationship is modified at increasing levels of coping efficacy, such that the higher pain severity is associated with less sleep quality disruption for children with high coping efficacy.

**Baseline Coping Efficacy and Longitudinal Coping Efficacy**

A hierarchical regression analysis was conducted to test the hypothesis that baseline coping efficacy scores on the SCQ-C would predict aggregate daily coping efficacy scores on the daily diary, even with the difference in response format. CDI scores were entered as a covariate at Step 1 and predicted a significant portion of the variance in coping efficacy on daily reports ($R^2 = .13, p < .05$). Baseline coping efficacy score on the CSQ-C was added at Step 2 and an interaction between child depression and baseline coping efficacy was tested at Step 3. As predicted, baseline coping efficacy score significantly predicted daily coping efficacy score when controlling for the effects of child depressive symptoms ($\Delta R^2 = .16, p < .01$), but results did not suggest that this relationship is moderated by depressive symptoms ($\Delta R^2 = .01, p > .05$).

**Parent and Child Coping Strategies**

Parent and child ratings of coping strategy use were analyzed to test the hypothesis that parent coping strategies predict child coping strategies. Descriptive statistics for child coping strategies are reported in Table 5. Pearson product correlations were calculated for child coping strategies, parent coping strategies, child age, child depressive symptoms on the CDI, and parent pain history in order to identify covariates for the regression analyses for each coping domain. Of the 7 coping strategies, parent and child scores were significantly correlated on only the Distracting Attention subscale ($r = .46, p < .001$) and no significant covariates were identified in the correlation analyses in regards to Distracting Attention.
coping strategies. As such, no regression models were constructed to test hypothesis 4. However, some significant correlations between some child coping strategies and child depressive symptoms and age were identified. Specifically, Reinterpreting Pain Sensations subscale scores were significantly correlated with CDI scores \( (r = .34, p < .05) \) meaning that higher scores on Reinterpreting Pain Sensations was related to higher scores of depressive symptoms. In order to further investigate this relationship, the Reinterpreting Pain Sensations subscale was disaggregated and a correlation analysis between individual items and the CDI scores was run. This analysis resulted in significant correlations between CDI scores and two items: “I imagine that the pain is outside of my body” and “I pretend it’s not a part of me,” suggesting that children who endorse more depressive symptoms also reconceptualize pain as being external. The relationship between Catastrophizing and CDI scores approached significance \( (r = .27, p = .06) \), such that higher depressive scores was related to more use of Catastrophizing coping strategies. Taken together, these results indicate that children’s depressive symptoms were related to the use of some coping strategies. In terms of demographic variables, age was positively correlated with Coping Self Statements \( (r = .37, p < .01) \), indicating that older children endorsed greater use of Coping Self Statements as a coping strategy.

Discussion

The present study applied prospective daily diary methodology in order to provide additional support for the pain-sleep relationship and is one of the first studies to incorporate psychological aspects of the disease experience, namely, coping efficacy. Consistent with previous research (Passarelli et al., 2006; Valrie, 2007a) and the study hypotheses, aggregate pain rating significantly predicted aggregate sleep quality rating, such that higher pain was
associated with decreased sleep quality. Importantly, this finding suggests that children with high pain or poor pain management may also be at risk of sleep problems. Given that good sleep is linked to daily functioning in a variety of domains, including academic performance and emotional functioning, assessment and treatment of sleep difficulties may be needed as part of comprehensive medical care.

Neither of the hypothesized covariates significantly predicted variance in sleep quality, suggesting that variables other than disease and demographic factors likely account for sleep quality in children with arthritis. This is similar to findings regarding pain in children with arthritis (Malleson et al., 2004; Schanberg et al., 1997), which have consistently found that the relationship between disease variables and pain is weak and psychosocial factors are more strongly associated with children’s arthritis pain experiences. However, given the expected developmental differences in the sleep of children and adolescents (Iglowstein, Jenni, Molinari, & Largo, 2003), it is somewhat surprising that age did not predict sleep quality and that sleep quality was relatively high overall for the sample. The lack of difference in sleep quality due to age may be an artifact of using a self report measure, which was a global assessment of sleep quality on the previous evening and did not assess specific aspects of the sleep experience such as number of night awakenings, difficulty with sleep onset, or early morning awakenings. Although adolescents as a group may experience increased need for sleep coupled with decreased nightly sleep duration (Carskadon, 2002), their individual subjective experiences of nightly sleep may still be positive overall (Anderson, Storfer-Isser, Taylor, Rosen, & Redline, 2009). Taken together, the children and adolescents in the sample reported relatively high average sleep quality. The limited variability in overall sleep quality may be related to using a global rating rather
than separate assessments of specific sleep disruptions. For example, previous research has identified night awakening and sleep anxiety as sleep disruptions in children with arthritis (Bloom et al., 2003), which may not be captured in a global rating. These disruptions might have become part of their nightly sleep experience and are thus discounted in their overall report of sleep quality. Also, subjective reports may not capture physiological sleep disruptions in children with arthritis that have been identified via polysonography (PSG). Including more detailed questions regarding specific aspects of nightly sleep quality may be important in describing the characteristics of sleep quality in children who experience recurrent pain.

The inclusion of coping efficacy as a moderating psychological variable was a unique strength of the current study. When restricting the data to days when pain was reported, the relationship between pain and sleep quality was significantly moderated by coping efficacy. Children with arthritis who felt more control over their pain experienced better sleep quality overall than children who perceived themselves as having low to moderate control over their pain. Coping efficacy buffers the effects of pain on sleep quality, but the specific mechanism remains to be identified. This finding demonstrates the connection between coping and sleep in the context of recurrent pain and highlights the need for further research on other psychological aspects of the disease experience that may affect sleep in children who experience arthritis pain.

The alternate hypothesis was also analyzed in order to test for a reciprocal relationship between pain and sleep. In this analysis, age was a significant predictor of pain rating, but physician global assessment rating remained insignificant. Sleep and coping efficacy both significantly predicted pain ratings, suggesting that a more complex
relationship between these variables may exist, but the interaction between sleep quality and coping did not predict significant variation in pain. The interpretation of these results is somewhat limited as it is not possible to determine a statistically significant difference in the direction of the relationship between sleep and pain in the current study, but these findings do indicate the need for longitudinal studies which can capture the temporal relationship between variables and help to further distinguish the direction of the relationship over time. No studies addressing this question have been conducted with children with arthritis, but a recent study in children with sickle cell disease (Valrie et al., 2007a) lends support for the cyclical nature of daily pain and sleep and the need for research with other pediatric recurrent pain populations.

In order to further investigate the coping experiences of children in the sample, baseline reports of pain coping strategies were analyzed to determine if a social learning process occurs between parent and children and to identify covariates of coping strategy use. Findings indicate that children’s pain coping strategies were not significantly related to those that their parents use, suggesting that children’s selection of coping strategies is influenced by variables other than parent modeling. In regard to demographics, age was significantly correlated with use of Coping Self Statements, such that older children use more of these statements. Developmentally, this is consistent with expectations that children will increasingly implement cognitive strategies as they age (Skinner & Zimmer-Gembeck, 2007a). Some aspects of psychological functioning did correlate with coping strategies. Scores on depressive symptoms were significantly correlated with increased use of Reinterpreting Pain Sensations. At the individual item level, it appears that children with higher depressive symptoms scores endorsed using cognitive strategies to externalize the
pain more frequently. Children with higher depressive symptoms scores also endorsed increased use of catastrophizing as a coping strategy. No psychological or demographic variables correlated with children’s baseline reports of coping efficacy, indicating that depressive symptoms likely do not contribute to the children’s sense of control over pain. This finding is important in light of previous research on the relationships between pain and mood, and mood and sleep. It suggests that depressive symptoms do not confound coping efficacy, such that the two may be treated as individual psychological variables in future research on pain and sleep.

This study builds upon previous research in several important ways. First, it provides additional support for findings in sleep research with a variety of pediatric chronic illness populations showing that higher pain is related to poorer sleep quality (Haim et al., 2004; Miller, Palermo, Powers, Scher, & Hershey, 2003; ) and highlights the need to for better sleep management interventions in children with recurrent pain. Second, coping efficacy was identified as an important moderator of the pain-sleep relationship, suggesting that future interventions may be focused on improving pain coping in order to promote sleep quality. This finding also lends support for the inclusion of psychological variables in future sleep research in children with recurrent pain. Finally, the prospective nature of data collection was a unique strength. Previous research with this population was cross sectional and prone to biases in retrospective reports or limited generalizability due to methodologies that captured only a very limited sample of sleep behavior. Given that this study implemented a prospective daily diary, tracking day-to-day patterns in sleep, coping, and pain, it eliminated some of the errors occurring in retrospective studies. Daily data collection is preferable to current or retrospective pain assessments conducted at a single time point, which may fail to
present a comprehensive view of the pain experience or which may introduce increased recall bias (Butz, 2004; van der Brink, Bandell-Hoekstra, & Abu-Saad, 2001).

Despite the many positive features of this study, there are a number of limitations to address. The recruitment method resulted in a sample of convenience, which may not reflect the full range of disease severity in children with arthritis. Indeed, the average disease severity rating was mild to moderate, suggesting that the pain and sleep experiences of the most impaired children may not be reflected in this study. However, a significant relationship between pain and sleep was detected in this sample, which indicates that even children with less severe disease status still experience difficulties in sleep related to pain and that sleep disruptions may be even more of a concern in children with more severe arthritis. Furthermore, although some aspects of the disease experience were incorporated in the present study, there may be other important aspects to include such as disease duration and medication use. Controlling for medications may be important as research in adult healthy populations has suggested that NSAIDs have a negative effect on sleep (Horne, 1989; Murphy, Badia, Myers, Boecker, & Wright; 1994), but research in adult arthritis has suggested that NSAIDs may improve the subjective sleep experience of patients (Baumgartner, Hohmeister, & Blumenberg-Novoselec, 1988; Lavie, Nahir, Lorber, & Scharf, 1991). The direct relationship between sleep and medications commonly used in treating pediatric arthritis remains to be determined.

Beyond issues pertaining to disease related variables, there are some issues regarding sleep measurement that should be noted in interpreting these findings. First, the present study relied on self reports of global sleep quality and did not include objective measures of sleep characteristics. This assessment technique may be more biased than
physiological measures such as PSG and actigraphy, which can capture specific sleep difficulties as they occur. Measuring the influence of specific sleep disturbances and overall sleep quality on other outcome measures will help identify the most critical sleep domains. It may be the case that specific sleep disturbances that cannot be captured via self report (such as leg movements, less time spent in deep sleep) adversely affect daytime functioning even when the individual perceives his or her overall sleep quality as high. As Bourguignon and colleagues (2003) point out in their review of sleep in adults with rheumatoid arthritis, individuals with chronic, specific sleep disruptions, as captured by PSG, may acclimate to a certain amount of sleep disturbances, thus influencing their perceptions of adequate sleep quality. Additional research on the effects of individual sleep disturbances on a variety of functional outcomes, including academic performance, activity reductions, and daytime mood is needed in order to identify the most critical aspects of sleep in pediatric arthritis patients.

Finally, a few issues regarding data collection and analysis should be noted. First, all measures included in this study were collected via self report and no concurrent parent reports were available for validation purposes. However, in Valrie et al.’s (2007a) recent daily diary study of sleep in SCD patients, parent and child reports of aspects of sleep were fairly consistent, lending support for the use of child reports alone. Furthermore, although the prospective nature of these daily self reports was a significant strength of the current study, pencil and paper daily diaries allow for some potential backfilling of information. Although participants in this study were provided stamped envelopes in order to return reports each day, some reporting errors may have occurred. This technique has not completely eliminated the issue of retrospective error in similar studies (Gendreau, Hufford,
& Stone, 2003), given the potential effects of inattention and mood at the time of completion on ratings (Anthony & Schanberg, 2007). Moving to electronic ecological momentary assessments of pain and sleep will eliminate some of these reporting issues in future research by capture data in real time. Future research should also employ more advanced statistical techniques that allow the examiner to study an individual’s unique pattern of change over time and to compare it to the other participants’ patterns of change.

In conclusion, these findings have important implications for future research endeavors and clinical practice. Future research should continue to capture daily pain and sleep ratings, while incorporating new technological advances that enable us to obtain ratings frequently without the influence of retrospective bias, and employing statistical techniques that test the cyclical relationship between pain and sleep. Also, sleep should be examined as a predictor of functional daily outcomes such as academic performance and participation in activities to further investigate the role of sleep in children with recurrent pain. Objective measures of sleep should be used to capture the specific aspects of sleep disruptions in children with arthritis in order to guide the selection of sleep variables for further study and to tailor future clinical interventions. In regard to clinical practice, these results support the need for additional assessment and monitoring of sleep in pediatric arthritis patients. Also, interventions should be developed to improve children’s sense of control in coping with the challenges of recurrent pain, as targeting this moderating variable is an avenue to reducing the relationship between pain and sleep. Consistent with the biopsychosocial model, other aspects of the psychological functioning of children with recurrent pain should be considered for interventions to promote sleep improvement.
Table 1

*Means and Standard Deviations of Aggregated Daily Variables*

<table>
<thead>
<tr>
<th></th>
<th>All Days</th>
<th></th>
<th>Pain Days</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( M (SD) )</td>
<td>Range</td>
<td>( M (SD) )</td>
<td>Range</td>
</tr>
<tr>
<td>Pain Severity</td>
<td>26.79 (21.25)</td>
<td>0.3-74.94</td>
<td>27.18 (20.68)</td>
<td>1-73.97</td>
</tr>
<tr>
<td>Coping Efficacy</td>
<td>1.80 (0.82)</td>
<td>0.05-3.0</td>
<td>1.75 (0.83)</td>
<td>0-3</td>
</tr>
<tr>
<td>Sleep Quality</td>
<td>75.25 (18.53)</td>
<td>40.79-100</td>
<td>74.88 (18.65)</td>
<td>40.97-100</td>
</tr>
</tbody>
</table>
Table 2

Summary of Hierarchical Regression Analysis Predicting Sleep Quality

<table>
<thead>
<tr>
<th>Step</th>
<th>Variable</th>
<th>β</th>
<th>F</th>
<th>ΔR²</th>
<th>R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Age, PGA</td>
<td>-.07, -1.32</td>
<td>1.31</td>
<td>.05</td>
<td>.05</td>
</tr>
<tr>
<td>2</td>
<td>Pain</td>
<td>-.54</td>
<td>9.79</td>
<td>.34***</td>
<td>.39***</td>
</tr>
<tr>
<td>3</td>
<td>Coping Efficacy</td>
<td>2.77</td>
<td>7.48</td>
<td>.01</td>
<td>.40**</td>
</tr>
<tr>
<td>4</td>
<td>Pain x Coping Efficacy</td>
<td>-.18</td>
<td>6.53</td>
<td>.03</td>
<td>.43**</td>
</tr>
</tbody>
</table>

*p < .05, **p < .001, *** p < .0001
Table 3

Summary of Hierarchical Regression Analysis Predicting Pain Rating

<table>
<thead>
<tr>
<th>Step</th>
<th>Variable</th>
<th>β</th>
<th>F</th>
<th>ΔR²</th>
<th>R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Age, PGA</td>
<td>.25*,</td>
<td>1.01</td>
<td>3.21</td>
<td>.12*</td>
</tr>
<tr>
<td>2</td>
<td>Sleep Quality</td>
<td>-0.66</td>
<td>11.72</td>
<td>.32*</td>
<td>.44***</td>
</tr>
<tr>
<td>3</td>
<td>Coping Efficacy</td>
<td>-10.02</td>
<td>14.01</td>
<td>.12*</td>
<td>.56***</td>
</tr>
<tr>
<td>4</td>
<td>Sleep x Coping Efficacy</td>
<td>.08</td>
<td>11.05</td>
<td>0</td>
<td>.56***</td>
</tr>
</tbody>
</table>

*p < .05, **p < .001, ***p < .0001
Table 4

*Summary of Regression Analysis Predicting Sleep Quality on Pain Days*

<table>
<thead>
<tr>
<th>Step</th>
<th>Variable</th>
<th>β</th>
<th>F</th>
<th>ΔR²</th>
<th>R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Age, PGA</td>
<td>-.08, -1.30</td>
<td>1.35</td>
<td>.06</td>
<td>.06</td>
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<tr>
<td>2</td>
<td>Pain</td>
<td>-.59</td>
<td>11.16</td>
<td>.37***</td>
<td>.43***</td>
</tr>
<tr>
<td>3</td>
<td>Coping Efficacy</td>
<td>.75</td>
<td>8.21</td>
<td>0</td>
<td>.43***</td>
</tr>
<tr>
<td>4</td>
<td>Pain x Coping Efficacy</td>
<td>-.28</td>
<td>8.18</td>
<td>.06*</td>
<td>.49***</td>
</tr>
</tbody>
</table>

*p < .05, **p < .001, ***p < .0001
Table 5

*Means and Standard Deviations of CSQ-C Subscales*

<table>
<thead>
<tr>
<th>Subscale</th>
<th>Mean (SD)</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Diverting Attention</td>
<td>9.45 (6.62)</td>
<td>0-24</td>
</tr>
<tr>
<td>Reinterpreting Pain Sensations</td>
<td>5.94 (6.40)</td>
<td>0-22</td>
</tr>
<tr>
<td>Coping Self Statements</td>
<td>18.47 (9.56)</td>
<td>0-36</td>
</tr>
<tr>
<td>Ignoring Pain Sensations</td>
<td>13.55 (8.26)</td>
<td>0-31</td>
</tr>
<tr>
<td>Praying or Hoping</td>
<td>19.90 (9.39)</td>
<td>0-36</td>
</tr>
<tr>
<td>Catastrophizing</td>
<td>8.69 (6.95)</td>
<td>0-28</td>
</tr>
<tr>
<td>Increased Behavioral Activations</td>
<td>12.84 (6.89)</td>
<td>0-28</td>
</tr>
<tr>
<td>Coping Efficacy</td>
<td>3.70 (1.52)</td>
<td>0-6</td>
</tr>
<tr>
<td>Ability to Decrease Pain</td>
<td>3.18 (1.44)</td>
<td>0-6</td>
</tr>
</tbody>
</table>
Figure 1

*Moderation Model for the Effect of Pain Coping Efficacy on the Relationship between Pain and Sleep Quality*
Figure 2

*Interaction between Pain and Coping Efficacy on Pain Days*

![MLR 2-Way Interaction Plot](image)
References


