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Sleep Patterns and Parents' Experience in Caring for Young Children Newly Diagnosed with
Juvenile Idiopathic Arthritis

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Abstract

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Background: Juvenile idiopathic arthritis (JIA) is a common pediatric autoimmune condition with no cure. The cause of JIA is not completely understood. Children experience unpredictable and fluctuating episodes of active and inactive disease and symptoms of pain, fatigue, and poor sleep quality. JIA not only affects the child, but also affects the family. Parents must learn to manage the illness, diagnostic and treatment demands, and disruptions in daytime and nighttime routines, as well as balance family and other caregiving demands.

Purpose: The overall purpose of the dissertation was to gain new knowledge about the interrelations among sleep patterns, sleep quality, symptoms, and family functioning in parent-child dyads of young children newly diagnosed with JIA compared to their peers without this health condition, and to describe parents' experience in caring for their young children newly diagnosed with JIA.

Methods: The first part of the dissertation included two secondary analysis studies of data from an existing cross-sectional observational study that examined the impact of a new JIA diagnosis on sleep, disease-related symptoms and family functioning in 2-5-year-old children and their parents. The sample included 30 parent-child dyads (14 JIA and 16 control dyads). Participants wore actigraphs for 10 days and completed daily sleep and symptoms diaries. Parents completed one-time questionnaires about demographic information and family functioning survey. The second part of the dissertation involved a single-occasion semi-structured interview study with 9 of the 14 parents who participated in the previous observational study.

Results: Young children with JIA had decreased nighttime sleep duration and poorer sleep quality compared to children without JIA. Poor sleep quality was related to increased symptoms of pain and fatigue. Parents of children with JIA had poorer sleep quality compared to parents in the control group, and poor sleep quality was inversely correlated with daytime fatigue and nighttime worry in parents. Parent-child sleep quality was interrelated in the JIA dyads, but not in the control parent-child dyads. Family functioning was similar in families of children with and without JIA, and did not correlate with children's sleep quality. However, the parent interview data found the core construct "Struggling in the Dark to Help My Child" described parents' experience in caring for a young child with JIA. Parents struggled to understand this illness without a known cause, the different treatment options, and what the future might hold for their child. Parents reported feeling completely alone and in the dark, and tried to reach out for resources, only to find not much available. Parents struggled seeing their child in pain, but knew there was not much they could do to help. Parents blamed themselves, and some felt they were bad parents who had betrayed their children. Parents tried everything they could to work out the kinks and stay on top to manage JIA, even when they felt drained physically and emotionally.

JIA not only consumed their lives, but also affected the entire household, including siblings and spouse, and the relationships among family members.

Conclusions: Inadequate amounts of sleep and poor sleep quality were common in young children with JIA and their parents. Poor sleep quality was related to symptoms of pain and fatigue in children, and worry and fatigue in parents. Poor sleep quality is a modifiable health behavior and early interventions to improve sleep may alleviate future sleep problems and disease-related symptoms in children with JIA and their parents. Despite the advanced medical care for young children with JIA, the interview study findings are the first to highlight the day-to-day challenges parents encounter when caring for their young child during the initial JIA diagnosis period. Parents struggled with the unknown, searched for resources, witnessed their child's suffering without knowing how to help, and tried every possible way to stay on top of the child's illness and treatment. The prolonged struggle substantially impacted the emotional and physical health of the parents as well as the entire household. Healthcare providers need to assess the particular needs of an ill child and parents and the impact of the illness on the physical and psychosocial health in the entire household, so that proper resources can be provided. These findings will inform the development of family-centered intervention trials to assist families and improve child, parent, and family outcomes in the future.

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DEDICATION

To Grandpa Wang

Chapter 1. Introduction

Juvenile Idiopathic Arthritis (JIA) is one of the most common pediatric autoimmune conditions with prevalence between 1 and 8.6 per 10,000 children, affecting approximately 300,000 children in the United States (Espinosa & Gottlieb, 2012; Harrold et al., 2013). Children are diagnosed before the age of 16 and the cause is unknown. There are various categories of JIA including oligoarthritis, polyarthritis, systemic arthritis, enthesitis-related arthritis, psoriatic arthritis, and undifferentiated arthritis. JIA tends to occur more frequently in girls of European ancestry (Espinosa & Gottlieb, 2012). JIA is characterized by unpredictable episodes of active and inactive disease. During active disease, children may experience joint swelling, pain, tenderness, limited range of motion, fever, rash, and inflammation of the eyes, which can impact children's growth and development (Nordal et al., 2011; Prakken, Albani, & Martini, 2011). Fatigue, poor sleep quality (e.g., frequent night awakenings, inadequate amount of sleep), and daytime sleepiness are also common symptoms in JIA (Bromberg, Connelly, Anthony, Gil, & Schanberg, 2014; Butbul Aviel et al., 2011; Shyen et al., 2014; Ward et al., 2010).

Early childhood, 2 to 5 years, is a critical time for child growth and development. Young children begin to develop emerging skills and lifelong sleep habits are forming. When a young child is diagnosed with a chronic condition such as JIA, joint pain and limited range of motion may limit child's ability to develop gross and fine motor skills. Increased fatigue and disrupted sleep may interfere with the newly established daytime and nighttime routine. Early childhood might be a more opportune time to examine these children when the chronic condition is new so that potential health problems can be identified early and prompt interventions can be delivered to optimize health outcomes in these young children.

When a young child is newly diagnosed with a chronic condition, there is substantial increase in caregiving burden involved in treatment regimens, clinic visits, and symptoms

assessment and management in addition to the already demanding care for the young child. Parents' extensive involvement in the management of their child's chronic condition results in family and work life disruptions including less time with other family members, less personal time, and some parents decide to work part-time or stop working to manage their child's JIA (Bornstein, 2012). Although, caring for a young child newly diagnosed with a chronic condition is challenging, less is known about what particular challenges parents encounter, or how parents incorporate a new diagnosis within the family. A new diagnosis of JIA not only affects parents' daytime work and life routine, but also their nighttime routine. Parents tend to their child's nighttime needs and the repetitive nighttime awakenings lead to accumulated sleep deficits, fatigue, worry, and work absenteeism (Matthews, Neu, Cook, & King, 2014; Mawani et al., 2013). In addition, a child's new chronic illness diagnosis in the family may also affect how the entire family functions, including family cohesion and adaptation (Olson, Russell, & Sprenkle, 1983).

The overall purpose of the dissertation was to gain new knowledge about the interrelations among sleep patterns, sleep quality, symptoms, and family functioning in parent-child dyads of young children newly diagnosed with JIA compared to their peers without this health condition, and to describe parents' experience in caring for their young child newly diagnosed with JIA. The second chapter described daily sleep patterns, sleep quality, and sleep hygiene in 2-5-year-old children newly diagnosed with JIA and their parents in comparison with control children and their parents. The third chapter examined the temporal relationships between sleep quality and symptoms of pain, fatigue, and worry in these young children with JIA and their parents compared to control young children and their parents, and whether sleep quality was associated with family functioning in JIA and control children. The fourth chapter described

parent's perceived stress, illness-related challenges, coping behaviors, and adaptation to the stress and challenges when their 2-5-year-old child was newly diagnosed with JIA. Each of the three chapters form related but independent manuscripts, with separate introduction, methods, results, and discussion sections respectively.

Conceptual Framework

The integration of the Family Systems Theory and the Ecological Model for Human Development formed the conceptual framework for the dissertation (Broderick & Smith, 1979; Bronfenbrenner, 1979). The Family Systems Theory is a theory of human behavior that views the family as a unit and uses systems thinking to describe the complex interactions in the family unit. The Family Systems Theory provides a foundation to understand how JIA affects the young child, other family members, the relationships between family members, and the family system. In Chapter 4, the Family Systems Theory was discussed in detail and used to guide the interview study.

The Ecological Model for Human Development (Bronfenbrenner, 1979) was used to conceptualize interactions among sleep, symptoms, and the family system in JIA. The Ecological Model for Human Development describes health as a product of interdependence between the individual and subsystems (e.g., family) nested within an ecosystem. The model emphasizes the equal importance of the person and the environment (i.e., each subsystem), with a special attention to the child, parent and environmental interactions (Bronfenbrenner, 1979; Green, Richard, & Potvin, 1996). The Ecological Model for Human Development provides a framework to examine the relations among the person factor (young children with JIA), the health factors (sleep patterns, sleep quality, symptoms), and the environment factors (parents, family system, and family functioning).

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Chapter 2. Daily Sleep Patterns, Sleep Quality, and Sleep Hygiene Among Parent–Child Dyads
of Young Children Newly Diagnosed With Juvenile Idiopathic Arthritis and Typically
Developing Children

Abstract

Purpose: Describe daily sleep patterns, sleep quality, and sleep hygiene in 2-5-year-old children newly diagnosed with juvenile idiopathic arthritis (JIA) and their parents in comparison to typically developing (TD) children and parents.

Methods: Participants (13 JIA, 16 TD parent-child dyads) wore actigraphs for 10 days. Parents completed sleep diaries and sleep hygiene survey.

Results: Children with JIA had significantly less total sleep time, lower sleep efficiency (SE), and longer naps than TD children. Parents of children with JIA had significantly earlier bedtimes, more wake after sleep onset (WASO) and lower SE than TD parents. Parent-child SE and WASO were interrelated in JIA dyads. Sleep hygiene practices were inconsistent in both groups of children.

Conclusions: Inadequate amounts of sleep and poor sleep quality were common in parent-child dyads. Early interventions to improve sleep duration and promote sleep hygiene practices may alleviate future sleep problems and improve parent and child wellbeing.

Introduction

Juvenile idiopathic arthritis (JIA) is a chronic inflammatory disease with an unknown etiology. It is one of the common pediatric rheumatologic diseases, characterized with unpredictable episodes of active and inactive disease that impact children's growth and development (Nordal et al., 2011). An estimated 20% of children with JIA experience lifelong pain and significant disability (Gowdie & Tse, 2012). Children experience joint swelling, stiffness, tenderness and pain, limited range of motion, poor sleep quality (e.g., frequent night awakenings, inadequate amount of sleep), obstructive sleep apnea, and daytime sleepiness (Bromberg et al., 2014; Butbul Aviel et al., 2011; Shyen et al., 2014; Ward et al., 2010). While poor sleep hygiene practices including inconsistent pre-bedtime activities (e.g., screen time, roughhousing), lack of a regular bedtime routine and a regular bedtime contribute to inadequate amounts of sleep and impair sleep quality in typically developing children (Mindell, Li, Sadeh, Kwon, & Goh, 2015; Sadeh, Mindell, Luedtke, & Wiegand, 2009; Staples, Bates, & Petersen, 2015; Thompson & Christakis, 2005), less is known about young children with JIA. In typically developing children, poor sleep quality and inconsistent sleep hygiene practices not only predispose children to later development of behavioral and emotional impairments, but also have a negative impact on physical health and family stress (El-Sheikh, Kelly, Bagley, & Wetter, 2012; Gregory & Sadeh, 2012; Meltzer & Mindell, 2007; Sivertsen et al., 2015; Touchette et al., 2007). These effects may be even more pronounced in children with an underlying chronic condition like JIA, particularly in those with early childhood diagnoses when sleep habits are established.

Although the majority of sleep research in JIA examined school-age children and adolescents, early childhood may be a more critical period in JIA because lifelong sleep habits

are forming and rapid growth and development are occurring. Early childhood may provide a more opportune time for early identification and intervention to optimize health outcomes in young children with JIA. Irregular bedtime routines and bedtimes, disruptive nighttime awakenings, and inadequate amounts of sleep in JIA may play a major role in behavioral and emotional development and manifestations of disease-related symptoms. Given the importance of adequate sleep and healthy sleep hygiene, and the paucity of sleep research in young children, this study focused on 2-5 year-olds newly diagnosed with JIA and a comparison group of typically developing children.

Poor sleep quality and inconsistent sleep hygiene not only affect young children, but also their parents (Gallagher, Phillips, & Carroll, 2010; Matthews et al., 2014; Meltzer, Sanchez-Ortuno, Edinger, & Avis, 2015). Joint inflammation and pain can interfere with sleep quality and result in frequent nighttime awakenings for both the child and parent. Young children seek parents for comfort during the night, and irregular bedtime routines and bedtimes, and repetitive night awakenings can result in parental fatigue, daytime dysfunction (e.g., daytime sleepiness, anxiety), and work absenteeism (Matthews et al., 2014; Mawani et al., 2013). Several studies report poor sleep quality in children with a chronic condition (Lewandowski, Ward, & Palermo, 2011; Valrie, Bromberg, Palermo, & Schanberg, 2013), but less is known about the impact of a new chronic diagnosis on parents' sleep.

The purpose of this study was to describe daily sleep patterns, sleep quality, and sleep hygiene in 2-5-year-old children newly diagnosed with juvenile idiopathic arthritis (JIA) and their parents in comparison to typically developing (TD) children and parents. Sleep patterns were operationalized as actigraphy measured sleep onset and offset times, total sleep time (TST), and sleep diary reported bedtimes, wake times, and sleep duration. Sleep quality was

operationalized as actigraphy measured wake-after-sleep onset (WASO) and sleep efficiency (SE), as well as parent-report of child's sleep quality (SQ) and parent self-report diary SQ. The aims of the study were to: (1) describe parent-report sleep hygiene practices (regular bedtime routine and bedtime) in children with JIA and TD children; (2) describe daily sleep patterns (bedtime, wake time, TST) and sleep quality (WASO, SE, sleep diary SQ) in young children newly diagnosed with JIA in comparison to TD children; (3) describe daily sleep patterns (bedtime, wake time, TST) and sleep quality (WASO, SE, sleep diary SQ) in parents of young children newly diagnosed with JIA in comparison to parents of TD children; and (4) examine the relationships between parent and child sleep patterns and sleep quality and determine whether these relationships differ by group (JIA vs. TD). We hypothesized that: (1) in comparison to TD children, parent-reported bedtime routines and bedtimes would be inconsistent in children with JIA; (2) in comparison to TD children, children with JIA would have decreased TST, increased WASO, decreased SE, increased daytime naps, and decreased sleep diary SQ; (3) in comparison to parents of TD children, parents of children with JIA would have decreased TST, increased WASO, decreased SE, and decreased sleep diary SQ; and (4) parent and child TST, WASO, SE and sleep diary SQ would be positively associated in both groups, with the magnitude of this association being greater in the JIA dyads.

Methods

Participants

From January 2013 through September 2014, 30 children (14 newly diagnosed with JIA and 16 TD children), and 30 respective parents enrolled in the study. Eligibility for children included 2-5 years, and a JIA diagnosis within the past 10 months. Children in both groups were excluded if they had a developmental delay, a comorbid illness (diabetes, asthma, cancer), a

diagnosed sleep disorder reported by a parent and medical record (obstructive sleep apnea), or a family history of narcolepsy in a first-degree relative. Parent eligibility criteria included at least 18 years of age and able to speak and read English.

Of the 24 families of children with JIA approached, 14 were enrolled and completed the study protocol (58% response rate). One JIA parent-child dyad data was not included in the analysis due to loss of actigraphy data (i.e., swimming). Of the 29 families of TD children approached, 16 were enrolled and completed the study protocol (55% response rate). The final sample included 29 parent-child dyads (13 JIA dyads, 16 TD dyads).

Procedures

Approval for this study was obtained from the Institutional Review Board at a large urban children's hospital in the Pacific Northwest. Children newly diagnosed with JIA were recruited from a pediatric rheumatology clinic. During a clinic visit, children with JIA and their parents were informed about the study; if they expressed interest, the research coordinator explained the purpose of the study and scheduled a home visit.

TD children were recruited through flyers and media advertisements in the communities. Interested parents contacted a member of the team who informed parents about the study, and if interested a home visit was scheduled. The home visit was scheduled for all families to explain the study protocol and the instructions to complete sleep diaries and questionnaires, and to obtain parent consent and child assent. Actigraphy watches were placed on the child's ankle and the parents' non-dominant wrist. Parents and children wore the watch and completed sleep diaries for 10 consecutive days. Families were contacted each week to answer questions.

Measures

Sleep hygiene. Two items from the General Sleep Information survey were used to assess parent reports of sleep hygiene during the previous one month. Parent reported “yes” or “no” to the questions “does your child have a regular bedtime routine?” and “does your child have a regular bedtime?”

Actigraphy. Actigraphy is a non-intrusive watch-like device to measure sleep-wake patterns in the natural home environment. Each child and parent wore an actigraph watch for 10 consecutive days (Actiwatch 64TM, Mini-Mitter Philips Respironics, Bend, OR). Actigraphy has an accelerometer that senses the occurrence and degree of motion in all directions. The motion is converted into an electric signal and digitally integrated to derive an activity count. Activity counts were recorded and stored in the actigraph at 1-minute intervals. Parents were instructed to press the event marker for their child and themselves to indicate the time they attempted to fall asleep, the time they woke up in the morning, the beginning and end of naps, and when the watch was removed (swimming, bathing). Sleep onset and offset were scored based on the sleep diary and event marker data. After study completion, actigraphy data were downloaded and stored in computer files using the Actiware software for scoring (Mini-Mitter Philips Respironics, Inc.). Activity counts were scored using a weighting algorithm with medium threshold, defined as 40 activity counts per epoch, for both parents and children. This algorithm has high sensitivity to detect sleep and low specificity to detect wake when compared to polysomnography or videosomnography in children with and without chronic conditions (Belanger, Simard, Bernier, & Carrier, 2014; Meltzer, Walsh, Traylor, & Westin, 2012; Sitnick, Goodlin-Jones, & Anders, 2008; Ward, Lentz, Kieckhefer, & Landis, 2011) and in adults (Morgenthaler et al., 2007). Interrater reliability was assessed with two members who individually scored actigraphy data, and the intraclass correlation coefficients (ICC) were 0.97 for sleep onset and 0.95 for sleep offset.

Actigraphy-derived sleep variables included: (a) sleep onset, defined as the time of the initial 3 or more consecutive minutes of sleep; (b) sleep offset, defined as the time of the last 5 or more consecutive minutes of sleep; (c) total sleep time (TST), defined as the total amount of time in minutes during the sleep period scored as sleep; (d) wake after sleep onset (WASO), defined as the number of minutes spent awake after sleep onset occurred; (e) sleep efficiency (SE), defined as the ratio of the amount of time scored as sleep to total sleep period time which yields the percentage of sleep within the sleep period; (f) daytime nap, defined as the total amount of time in minutes during the nap period scored as sleep per day; and (g) 24-hour sleep, defined as the total amount of nocturnal TST and daytime naps in minutes.

Sleep diary. Daily sleep diaries were used in conjunction with actigraphy. Parents completed the sleep diaries for themselves and their child every morning and at bedtime. Parents recorded the time they and their child went to bed, how long it took to fall asleep, number and duration of night awakenings, the time they woke up, sleep quality, duration of naps, and time and duration of actigraph removal. In addition, evening activities, medications, illnesses, and any unusual events that may have affected child or parental sleep (such as parent out of town) were recorded. Variables derived from sleep diary for both children and parents included: (a) bedtime, defined as the time parent recorded in the sleep diary as “falling asleep”; (b) wake time, defined as the time parent recorded in the sleep diary as “waking up”; (c) sleep duration, defined as the total amount of time spent asleep between bedtime and wake time; and (d) sleep quality, defined as 1 = “terrible” to 9 = “great” (higher numbers indicate better sleep quality).

Demographic characteristics. A demographic questionnaire was used to obtain information regarding parent demographics (parent’s age, sex, employment, ethnicity, education, and marital status), child demographics (age, sex), and childcare.

JIA disease duration was measured from the date the child was first diagnosed, and obtained from chart review. During a routine clinic visit, a pediatric rheumatologist assessed each child and determined active/inactive disease. Inactive disease was defined as a physician global assessment of disease activity of 0 on a 10-point numerical rating scale; no joints with active arthritis; no active uveitis; no fever, rash, serositis, splenomegaly, or lymphadenopathy attributable to JIA; normal erythrocyte sedimentation rate (ESR); and morning stiffness ≤ 15 minutes (Wallace, Giannini, Huang, Itert, & Ruperto, 2011). Children who did not meet criteria for inactive disease were categorized as having active disease.

Statistical Analysis

All analyses were conducted in the statistical software *R* (R Development Core Team, 2013). Descriptive statistics and graphs were computed for the demographic, actigraphy, and sleep diary data. Actigraphy sleep data with matching diary data were included in the analyses ($n=272$ number of days). Two-group independent *t*-test showed group differences in children's age ($p<0.01$; see Table 2.1); therefore, children's age was included as a covariate in the subsequent analyses. To address aim 1 – describe parent-report sleep hygiene practices (regular bedtime routine and regular bedtime), descriptive statistics were used to describe the two questions between the two groups.

Multilevel modeling (also known as mixed-effects modeling or hierarchical linear modeling) with restricted maximum likelihood estimation was used to analyze the data in order to account for the nesting effect of daily actigraphy and sleep diary assessed variables within each participant. The multilevel modeling approach allows for different numbers of observations (days) from each participant, providing a better mechanism to handle missing values compared to deleting all data from participants with missing data (Snijders & Bosker, 2012). Each

participant's sleep data from a particular day were treated as a single data point. Packet "lme4" within *R* was used to conduct multilevel analyses (Bates, Maechler, Bolker, & Walker, 2015). There were two levels of analysis in the data: "level 1" referred to the days across the study period within each subject, while "level 2" was the subject. The subject effect was entered into the models as a random effect. To address aim 2 – describe daily sleep patterns (bedtime, wake time, TST) and sleep quality (WASO, SE, sleep diary SQ) in children with JIA in comparison to TD children, and aim 3 - describe daily sleep patterns and sleep quality in parents of children with JIA in comparison to parents of TD children, the group effect (TD coded as 0, JIA coded as 1) and child age were entered as fixed effects, and the outcome variables were the sleep variables of the children and parents, respectively.

In multilevel modeling, it is important to separate the within-subject effect from the across-subject effect. Ignoring this distinction may lead to difficulty in interpreting the results and incorrect conclusions (Mancl, Leroux, & DeRouen, 2000). For aim 4 – examine the relationships between parent and child sleep patterns and sleep quality in the two groups and whether these relationships differ by group (TD vs. JIA), the across-subject term was calculated by averaging each child's sleep variables across the study period (e.g., TST_{mean}). The within-subject term was calculated as the deviation of the daily value of the child's sleep variables from the within-subject mean (e.g., $TST_{\text{diff}} = TST_{\text{day}} - TST_{\text{mean}}$). First, child sleep variables (within- and across-subject effects), age, and group (TD coded as 0, JIA coded as 1) were entered into the model as fixed effects, and subject as a random effect, to examine the relationships between parent and child sleep variables. For example, a significant child TST_{mean} predictor of parent TST would indicate that when the child has an increased sleep duration on average, the parent would have an increased sleep duration on average. A significant child TST_{diff} predictor of parent TST

would indicate that within a dyad, when the child has an increased sleep duration in one night, the parent would have an increased sleep duration that night. Interaction terms with the interaction of within-subject term \times group and across-subject term \times group were then added to the model. The coefficient of the interaction term would be interpreted as whether there was a group difference in parent-child relationships in the sleep variable examined. For example, a significant group \times TST_{diff} effect would indicate group differences in the relationships between parent-child TST. If there was a significant interaction, further analysis was performed to calculate the coefficients of the relationships for each of the two groups (TD and JIA).

Results

Sample Characteristics

Table 2.1 shows the demographic and clinical characteristics of the participants. The average age for children was 3.4 years. In children newly diagnosed with JIA, the average time between diagnosis and study was 6.9 months. The majority of children with JIA were girls with active disease.

[Table 2.1 about here]

Sleep Hygiene

Of the 16 parents of TD children, 100% ($n = 16$) reported that their child had a regular bedtime routine, and 88% ($n = 14$) reported that their child had a regular bedtime. Of the 13 parents of children with JIA, 85% ($n = 11$) reported that their child had a regular bedtime routine, and 92% ($n=12$) reported that their child had a regular bedtime.

Sleep Patterns and Sleep Quality

The average number of days of sleep examined with actigraphy was 9.4. Table 2.2 shows actigraphy and sleep diary data for parent-child dyads. In children, across the 10-day period, on

average, young children with JIA had an estimated 31 minutes less total sleep time ($p < 0.03$), 3% lower sleep efficiency ($p < 0.03$), and 24 minutes more of daytime naps ($p < 0.01$), as measured by actigraphy, than TD children, controlling for child age. The reports from the sleep diary did not show any difference in bedtime, wake time, sleep duration, and sleep quality between the two groups of children.

In parents, across the 10-day period, on average, the parents of children with JIA went to sleep 52 minutes earlier in the evening ($p < 0.02$), had 20 minutes more WASO ($p < 0.02$), and 4% lower sleep efficiency ($p < 0.02$), as measured by actigraphy, than the parents of TD children. Consistent with actigraphy, the sleep diary reports also showed that the parents of children with JIA went to sleep 40 minutes earlier in the evening ($p < 0.02$) than the parents of TD children. The reports from the sleep diary did not show any difference in wake time, sleep duration, and sleep quality between the two groups of parents.

[Table 2.2 about here]

Relationships between Parent and Child Sleep

To determine whether there was a relationship between parent and child actigraphy and sleep diary variables, regardless of group (TD vs. JIA), child sleep variables (actigraphy measured TST, WASO, SE, and child sleep diary SQ), both within- and across-subject effects, were entered into the model as fixed effects, and parent sleep variables (actigraphy measured TST, WASO, SE, and sleep diary SQ) as outcomes, respectively. Table 2.3 shows the relationships between parent and child sleep: for the across-subject effects, parent and child TST and SQ were positively related, suggesting that when a child slept one hour more on average, the parent was estimated to sleep 34 minutes more on average ($p < 0.002$). When a parent rated their child's sleep quality 1 unit higher on average, the parent rated their own sleep quality 0.71 unit

higher on average ($p < 0.001$). For the within-subject effects, parent and child TST, WASO, SE, and SQ were positively related (p 's < 0.01). Thus within a dyad, when a child slept one hour more during a night, the parent was estimated to sleep 19 minutes more that night. When a child had one hour more WASO, the parent was estimated to have 10 more minutes WASO that night. When a child had a 10% increase in SE during a night, the parent was estimated to have a 2% increase in SE that night. When a parent rated their child's sleep 1 unit higher in one night, the parent rated their own sleep 0.52 unit higher that night (See Table 2.3).

[Table 2.3 about here]

To examine whether the above relationships differed by group (TD vs. JIA dyad), interaction terms (child within-subject term \times group and child across-subject term \times group), were added to the model. No significant child sleep \times group interaction was found for TST ($p = 0.35$), but significant within-subject child sleep \times group interaction was found for actigraphy-measured WASO ($p < 0.03$), SE ($p < 0.003$) and sleep diary-measured SQ ($p < 0.02$) (See Table 2.4). In order to understand the relationships among parent-child WASO, SE, SQ and group (TD vs. JIA dyad), coefficients of the relationships in the two groups were calculated separately. In JIA dyads, for every one-hour increase in child WASO, there was an estimated 12-minute increase in parent WASO ($p < 0.001$) (See Figure 2.1). For every 10% decrease in child SE, there was an estimated 3% decrease in parent SE ($p < 0.001$). For every 1-unit increase in parental report of child SQ, there was 0.51-unit increase in parent SQ. In TD dyads, for every 1-unit increase in parental report of child SQ, there was 0.51-unit increase in parent self-report SQ; the coefficients for the relationships between parent-child WASO and SE were close to zero and not significant.

[Table 2.4 about here]

[Figure 2.1 about here]

Discussion

This is one of the first studies that assessed daily sleep patterns and sleep quality with both actigraphy and diary using multilevel modeling in 2-5-year-old children newly diagnosed with JIA and their parents in comparison to TD children and their parents. Regardless of JIA, our sample of young children obtained inadequate amounts of sleep, poor sleep quality, and had poor sleep hygiene. Although, parents of children with JIA had earlier bedtimes, their sleep quality was worse than the parents of TD children. Actigraphy-measured wake after sleep onset and sleep efficiency were interrelated in JIA parent-child dyads, but not in TD parent-child dyads. Sleep quality as measured by sleep diaries was interrelated in TD and JIA dyads.

Sleep Patterns and Sleep Quality

Average 24-hour sleep was approximately 9 hours in our sample of young children which is below the recommended 10 to 13 hours by the National Sleep Foundation (2014) and considerably less than prior reports in young children using actigraphy (Acebo et al., 2005; Staples et al., 2015; Ward, Gay, Anders, Alkon, & Lee, 2008). Previous actigraphy-based studies show that longer daytime naps were associated with shorter nighttime sleep, earlier wake times (Acebo et al., 2005; Jones & Ball, 2014; Ward, Gay, Alkon, Anders, & Lee, 2008), and later sleep onset (Tikotzky & Sadeh, 2001). Contrary to prior research our study did not support this. Rather our findings were consistent with El-Sheikh and colleagues (2013) and Lam and colleagues (2011), who also found inadequate amounts of 24-hour sleep (average 9 hours and 9.4 hours, respectively) as measured by actigraphy in young children (El-Sheikh, Arsiwalla, Staton, Dyer, & Vaughn, 2013; Lam, Mahone, Mason, & Scharf, 2011). We anticipated that nighttime sleep would be less in JIA owing to increased night awakenings, and that daytime naps would compensate for the shorter nighttime sleep durations, but this was not the case. JIA children had

longer daytime naps, but the daytime naps did not make up for the inadequate amounts of nighttime sleep. This finding was not explained by JIA disease activity as sleep patterns and sleep quality did not differ by active versus inactive disease or active joint count (data not shown). The inadequate amounts of sleep in our sample may be owing to poor sleep hygiene practices (discussed below) and/or child temperament (e.g., soothability), family and socio-cultural factors (family stress, socioeconomic status) previously reported in other studies report (El-Sheikh et al., 2013; Jones & Ball, 2014; Molfese et al., 2015; Ward, Gay, Alkon, et al., 2008), but were not included in this pilot study.

Sleep Hygiene

Sleep hygiene practices were inconsistent in both TD and JIA children that may explain the inadequate amounts of sleep observed in our sample. Interestingly, the majority of parents reported that their child had a regular bedtime and a regular bedtime routine, yet there was considerable variability in the actigraphy sleep onset times between days (50 to 68 minutes as shown in Table 2.2). This finding may be attributed to the differences in the duration of bedtime routines across the families that can contribute to inadequate amounts of sleep in children and/or parents' interpretation of "regular" bedtime routine and bedtime. Caring for young children is exhausting and parents may not realize the amount of sleep young children need, the variability in bedtime routines, bedtimes, or the amount of sleep their child actually obtained. For example, upon study completion, we had the opportunity to discuss the sleep findings with parents and review the sleep data from the actigram. Many parents were unaware of the insufficient amounts of sleep their child received and the irregularity in bedtimes. Attending to young children while managing work and family life is demanding for parents, making it plausible that parents may lose sight of the importance of a consistent bedtime routine and sleep needs for their child.

Additionally, some parents may need advice in how to implement sleep hygiene practices and manage behaviors associated with sleep (e.g., bedtime resistance) versus the underlying chronic condition (e.g., night awakenings). Although our sample was small, more than 70% of the parents had a college degree, and our finding highlights the need to routinely ask parents about their child's sleep habits including prebedtime activities, consistency in implementing regular bedtime routines and bedtimes, sleep consumption (e.g., nighttime and daytime sleep duration), and location of sleep (home vs. childcare). We did not obtain information on parental sleep hygiene practices, which can contribute poor sleep hygiene in children. Future studies should obtain information on sleep hygiene practices in both the child and parent, which would provide new knowledge in the development of interventions to promote healthy sleep hygiene practices.

Relationships between Parent and Child Sleep

Our findings provide preliminary evidence for the interrelations between parent and child sleep in JIA dyads but not in TD dyads, which supports previous studies in parent-child dyads with autism and cancer (Goldman, Wang, & Fawkes, 2014; Matthews et al., 2014). We attribute this finding to parents of children with JIA may be more sensitive to their child's nighttime awakenings due to stress, worry, and/or more attentive to their child's nighttime needs, thus wake up more often than TD parents. Goldman and colleagues (2014) examined actigraphy-based sleep patterns and sleep quality and parent report of sleep (e.g., "good vs. poor sleeper") in mother-child dyads with and without autism. Sleep efficiency, sleep fragmentation, and wake after sleep onset were positively correlated in mother-child dyads with autism who were "poor sleepers," and total sleep time was positively correlated in TD dyads and mother-child dyads with autism who were "good sleepers." Another study by Matthews and colleagues also examined sleep in mother-child dyads with and without acute lymphoblastic leukemia (ALL),

and found that total sleep time was positively correlated in the mother-child dyads with ALL, but not in control dyads. Although we have few studies for comparison, findings from the above studies suggest that parents caring for children recently diagnosed with a chronic condition are vulnerable to inadequate amounts of sleep and poor sleep quality. Early interventions tailored to meet the specific sleep health needs of the child and parent caring for a child with a chronic condition are needed to promote parent and child well being. Interventions to improve sleep early in a child's disease course may alter later sleep-related comorbidities.

Limitations

Results from this study should be considered in light of its limitations. First, this was a small pilot study, and our findings need replication with a larger sample of parent-child dyads. Multiple comparisons were not adjusted for, with significant p -values set at < 0.05 . Additional studies are needed to provide new knowledge to develop interventions to improve sleep quantity and/or sleep hygiene in parent-child dyads. Second, there may be selection bias in our sample because families in this study may have been concerned about their sleep and/or their child's sleep prior to being approached, and thus more likely to participate in the study. Third, this study had a fairly homogeneous sample from two-parent, middle class families, limiting the generalizability of our findings. Fourth, age was included as a covariate in the analyses; however, the group difference in age may contribute to the difference in children's sleep (e.g., older children less likely to nap). Lastly, we did not measure light in the bedroom, which may have influenced study findings.

Conclusion

Routinely asking parents about their child's sleep needs, prebedtime activities, and consistency and duration of bedtime routines during well-child visits is needed to increase

parental awareness about the importance of sleep. This information would provide clinicians with new knowledge about how to intervene and work with parents caring for a child with a chronic illness during early versus later development. Parents caring for children with a chronic condition may also need practical advice about how to implement consistent sleep hygiene practices and manage behaviors associated with sleep versus the chronic illness. Disrupted sleep and poor sleep hygiene in parents and children are treatable, and early interventions to improve sleep hygiene practices and promote healthy sleep may alleviate future sleep problems and improve parent and child well-being and daytime function.

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Table 2.1 *Demographic Characteristics of the Sample*

	Child Characteristics		Parent Characteristics	
	TD (n = 16)	JIA (n =13)	TD (n = 16)	JIA (n =13)
Age, years *	4.0 (1.14)	2.73 (0.73)	Age, years	34.87 (4.19) 32.75 (4.43)
Sex, n (%)			Sex, n (%)	
Male	4 (25%)	3 (23%)	Male	2 (13%) 0
Female	12 (75%)	10 (77%)	Female	14 (88%) 13 (100%)
JIA Category, n (%)			Education, n (%)	
Oligoarticular	N/A	6 (46%)	Some college	2 (13%) 6 (46%)
Extended oligoarticular		1 (8%)	College degree	10 (63%) 5 (39%)
Polyarticular		6 (46%)	Master's degree	4 (25%) 2 (15%)
Disease Condition, n(%)			Marital Status, n (%)	
Active JIA	N/A	9 (69%)	Married	16 (100%) 11 (85%)
Inactive JIA		4 (31%)	Divorced	0 2 (15%)
JIA Duration, months	N/A	6.92 (2.22)	Ethnicity, n (%)	
JIA Active Joint, (#)	N/A	2.31 (2.14)	Caucasian	13 (81%) 10 (77%)
PGA Disease Activity	N/A	1.68 (1.87)	Hispanic	0 1 (8%)
Attend Childcare, n (%)	8 (50%)	6 (46%)	Asian	3 (19%) 1 (8%)
Attend Kindergarten	3 (19%)	0	Other	0 1 (8%)

JIA – juvenile idiopathic arthritis; TD –typically developing; PGA – physician global assessment. Data are mean \pm SD or n (%).

* denote significant group differences.

Table 2.2 *Actigraphy Sleep and Sleep Diary in Parent-Child Dyads*

Variables	Mean (SD)		Group Fixed Effects				
	TD	JIA	<i>b</i>	s.e.	<i>t</i>	<i>p</i>	<i>Cohen's d</i>
<i>Actigraphy Child Sleep</i>							
Sleep Onset	21:17 (0:50)	21:14 (1:08)	-0.02	0.26	-0.07	0.94	0.02
Sleep Offset	7:24 (0:46)	7:06 (1:03)	-0.34	0.24	-1.41	0.16	0.54 +
TST (min)	509 (42)	483 (72)	-30.65	13.89	-2.21	0.03 *	0.86 ++
WASO (min)	82 (24)	93 (36)	10.97	5.91	1.86	0.06	0.69 +
SE (%)	84.84 (3.75)	82.53 (6.05)	-2.51	1.14	-2.2	0.03 *	0.84 ++
Daytime Nap (min)	19 (37)	63 (49)	23.94	9.51	2.52	0.01 **	1.72 ++
24-Hour Sleep (min)	528 (52)	546 (76)	-2.04	19.85	-0.1	0.92	0.35
<i>Diary Child Sleep</i>							
Bedtime	21:01 (0:47)	21:01 (1:04)	0.09	0.25	0.34	0.74	0.22
Wake Time	7:24 (0:36)	7:11 (0:59)	-0.24	0.21	-1.15	0.25	0.36
Sleep Duration (min)	623 (47)	610 (62)	-20.66	15.75	-1.31	0.19	0.62 +
SQ	7.80 (1.54)	6.98 (2.23)	-0.63	0.61	-1.04	0.3	0.46
<i>Actigraphy Parent Sleep</i>							
Sleep Onset	23:43 (1:06)	22:56 (1:34)	-0.86	0.37	-2.35	0.02 *	0.89 ++
Sleep Offset	7:27 (1:05)	7:01 (1:10)	-0.33	0.3	-1.13	0.26	0.44
TST (min)	397 (65)	404 (77)	4	18.04	0.22	0.82	0.16
WASO (min)	48 (19)	67 (41)	20.02	8.33	2.4	0.02 *	0.92 ++
SE (%)	87.81 (4.47)	84.28 (6.93)	-3.76	1.63	-2.3	0.02 *	0.88 ++
<i>Diary Parent Sleep</i>							
Bedtime	23:30 (1:05)	22:52 (1:20)	-0.67	0.29	-2.31	0.02 *	0.88 ++
Wake Time	6:55 (0:57)	6:46 (1:01)	-0.21	0.25	-0.84	0.40	0.31
Sleep Duration (min)	444 (76)	475 (86)	31.89	18.11	1.76	0.08	0.72 +
SQ	6.54 (1.47)	5.74 (2.08)	-0.29	0.53	-0.54	0.59	0.56 +

TD –typically developing; JIA – juvenile idiopathic arthritis; TST– total sleep time, WASO – wake after sleep onset, SE - sleep efficiency, min – minutes, SQ – sleep quality reported in sleep diary; bedtimes and wake times are presented as 24-hour clock.

Group fixed effect was entered as TD-0, JIA-1; child age was entered as a fixed effect.

Cohen's d was calculated based on the individual sleep data averaged across the study period;

+ medium effect size, ++ large effect size.

* $p < .05$, ** $p < .01$

Table 2.3 *Fixed Effects of Child Sleep as Predictors of Parent Sleep*

Outcome	Parent TST			
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
Child TST Mean (Across)	0.56	0.18	3.08	0.002 **
Child TST Diff (Within)	0.31	0.08	3.81	<0.001 ***
Group (JIA)	0.30	0.27	1.11	0.27
Age	-0.06	0.12	-0.56	0.58
Outcome	Parent WASO (minutes)			
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
Child WASO Mean (Across)	0.09	0.28	0.33	0.74
Child WASO Diff (Within)	0.17	0.05	3.32	0.001 **
Group (JIA)	16.62	10.63	1.57	0.12
Age	-1.88	4.57	-0.41	0.68
Outcome	Parent SE (%)			
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
Child SE Mean (Across)	0.19	0.29	0.67	0.51
Child SE Diff (Within)	0.17	0.06	3.03	0.002 **
Group (JIA)	-3.15	2.01	-1.51	0.13
Age	0.10	0.91	0.11	0.91
Outcome	Parent SQ (%)			
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
Child SQ Mean (Across)	0.71	0.10	6.87	<0.001 ***
Child SQ Diff (Within)	0.52	0.05	9.83	<0.001 ***
Group (JIA)	0.16	0.33	0.49	0.63
Age	0.31	0.14	2.18	0.03 *

TST – total sleep time, SE – sleep efficiency, WASO – wake after sleep onset; SQ – sleep quality reported in sleep diary; group fixed effect was entered as TD-0, JIA-1.

* $p < .05$, ** $p < .01$, *** $p < .001$

Table 2.4 *Fixed Effects and Interaction Terms (Child Sleep x Group) as Predictors of Parent Sleep*

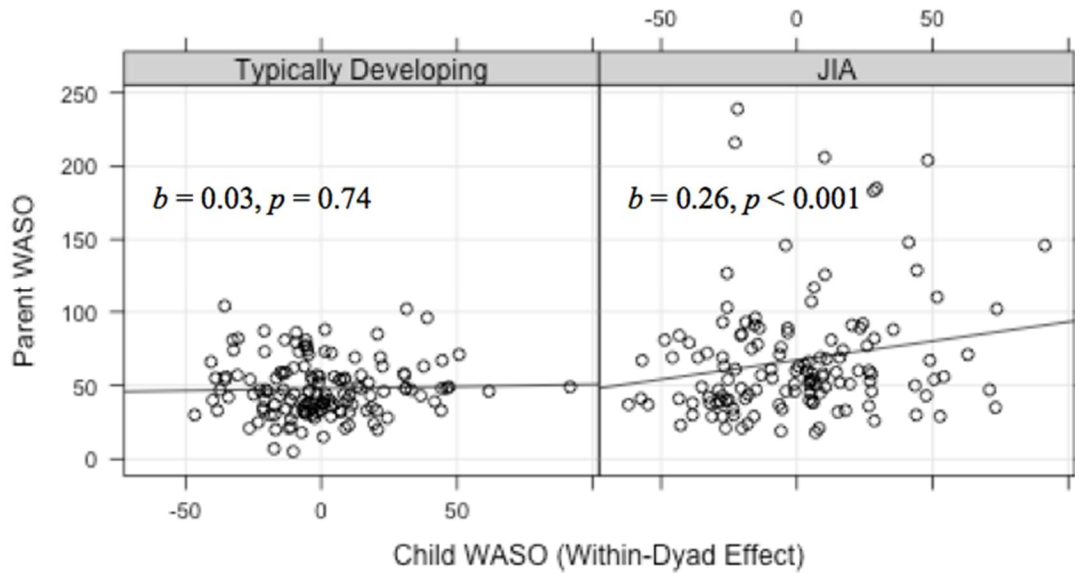
Outcome	Parent WASO (minutes)				
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>	
Child WASO Mean (Across)	0.21	0.68	0.30	0.76	
Child WASO Diff (Within)	0.03	0.08	0.33	0.74	
Group (JIA)	30.64	64.70	0.47	0.64	
Child WASO Mean x Group	-0.16	0.75	-0.22	0.83	
Child WASO Diff x Group	0.24	0.11	2.24	0.03	*
Age	-1.78	4.70	-0.38	0.70	

Outcome	Parent SE (%)				
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>	
Child SE Mean (Across)	0.74	0.76	0.98	0.33	
Child SE Diff (Within)	-0.05	0.09	-0.50	0.62	
Group (JIA)	52.51	68.30	0.77	0.44	
Child SE Mean x Group	-0.66	0.81	-0.82	0.41	
Child SE Diff x Group	0.35	0.12	2.94	0.003	**
Age	-0.17	0.97	-0.18	0.86	

Outcome	Parent SQ (%)				
Fixed Effect	<i>b</i>	s.e.	<i>t</i>	<i>p</i>	
Child SQ Mean (Across)	0.40	0.16	2.55	0.01	*
Child SQ Diff (Within)	0.33	0.10	3.45	0.001	**
Group (JIA)	-3.36	1.48	-2.28	0.02	*
Child SQ Mean x Group	0.49	0.20	2.44	0.02	*
Child SQ Diff x Group	0.28	0.11	2.43	0.02	*
Age	0.37	0.13	2.78	0.006	**

SE - sleep efficiency, WASO – wake after sleep onset, SQ –sleep quality reported in sleep diary; group fixed effect was entered as TD-0, JIA-1.

* $p < .05$, ** $p < .01$

Figure 2.1 *Interaction Between Parent-Child WASO by Group*

WASO – wake after sleep onset (minutes); Child WASO for each night was centered at child mean WASO across the 10 days; each circle represents a night, the corresponding x-value is the centered child WASO that night, and the corresponding y-value is the parent WASO that night; Slopes and p -values were calculated using multilevel modeling and controlled for child age.

Chapter 3. Family Functioning and Temporal Daily Relationships between Sleep Quality and Symptoms in Young Children with Juvenile Idiopathic Arthritis and Their Parents

Abstract

Purpose: Examine the temporal daily relationships between sleep quality and symptoms of pain, fatigue, and worry in young children recently diagnosed with JIA and their parents compared to young children without JIA and their parents (control), and to examine whether sleep quality was associated with family function in JIA and control children.

Methods: Participants (14 JIA, 16 control parent–child dyads) wore actigraphy watches for 10 days. Parents completed daily sleep and symptoms diaries for their child and themselves, and the Family Adaptability and Cohesion Scale (FACES) survey.

Results: In young children with JIA, both daily and average daytime pain predicted poor sleep quality, which subsequently predicted increased next-day pain; average fatigue severity, not daily fatigue predicted poor sleep quality, and poor sleep quality predicted increased next-day fatigue. In parents of young children with JIA, both daily and average nighttime worry predicted poor sleep quality, which subsequently predicted next-day fatigue. Associations between child's sleep quality and family functioning were not found.

Conclusions: Findings from this study show the complex temporal relationships between sleep quality and symptoms in young children newly diagnosed with a chronic condition and their parents. Poor sleep quality is a modifiable behavior, and improving sleep quality may alleviate symptoms in young children with a new chronic condition and their parents. Routine sleep assessments in young children and their parents are needed during clinical care to identify families at high risk for poor sleep quality.

Introduction

An estimated 10-40% of young children (2-to-5 years) in the United States have sleep disturbances, including inadequate sleep or poor quality sleep (e.g., nighttime awakenings) (Byars, Yolton, Rausch, Lanphear, & Beebe, 2012; Honaker & Meltzer, 2014). Sleep disturbances in early childhood often persist into later years, and are associated with subsequent development of emotional and behavioral problems (Sivertsen et al., 2015; Touchette et al., 2007). Further, poor quality sleep negatively affects parents' sleep as well as daytime function (e.g., fatigue, worry), and family function (e.g., marital conflict, family adaptability and cohesion) (El-Sheikh et al., 2012; Meltzer & Montgomery-Downs, 2011). Despite the prevalence of sleep disturbances and their negative impact, little is known about disturbed sleep in young children living with Juvenile Idiopathic Arthritis (JIA). To address this gap, we studied the interrelations among sleep quality (better quality indicates less disturbances), daytime function (e.g., fatigue, worry) and family functioning (family adaptability and cohesion) in young children with JIA and their parents in comparison to a control group of parent-child dyads.

JIA is the most common chronic rheumatologic disease in children. It is not only a disease of the individual child, but must be viewed in the context of the child's family, including the parents and the family as a unit (Barlow, Harrison, & Shaw, 1998; Knafl et al., 2013; Vandvik, Hoyeraal, & Fagertun, 1989). Parents must learn about the child's chronic condition, manage the treatment regimens, monitor the symptoms, and adapt to new roles and responsibilities, all while balancing other family demands such as attending to spouses and any non-ill children (Barlow et al., 1998; Frank et al., 1998; Knafl et al., 2013). At the same time, young children with JIA experience daytime pain and joint inflammation that can interfere with nighttime sleep quality. Likewise, poor sleep quality has been shown to exacerbate next-day pain

(Bromberg, Gil, & Schanberg, 2012; Lewandowski, Palermo, De la Motte, & Fu, 2010; Palermo & Kiska, 2005). Nighttime awakenings not only disrupt the sleep of the child but also of their parent and can result in parental fatigue, worry, and poor family functioning. In the few studies that examined sleep in parents caring for a child with a chronic condition, increased rates of worry, fatigue, and disturbed sleep were reported in comparison to parents of children without a chronic condition (Meltzer & Mindell, 2006; Meltzer et al., 2015; Neu, Matthews, & King, 2014; Stinson et al., 2012; Ward, Rankin, & Lee, 2007).

The purpose of this study was to examine the temporal relationships between sleep quality and symptoms of pain, fatigue, and worry in young children recently diagnosed with JIA and their parents compared to young children without JIA and their parents (control), and to examine whether sleep quality was associated with family function in JIA and control children. Family function was operationalized as family cohesion (e.g., emotional bonding among family members) and adaptability (e.g., relationship roles and rules in response to stress) (Olson et al., 1983). Specific study questions were: (1) What are the relationships between daily sleep quality and symptoms of fatigue and nighttime worry in parents of young children with JIA and control parents? (2) What are the relationships between daily sleep quality and symptoms of fatigue and pain in young children with and without JIA? (3) On average, is children's sleep quality associated with family functioning in families of young children with and without JIA?

Methods

Participants

Approval for this study was obtained from the Institutional Review Board in a University setting. Written informed consent was obtained from parents; assent was obtained from children. From September 2013 through December 2014, 30 children (14 JIA), 2-5 years, and their parent

were enrolled in the study. Inclusion criteria included children 2-5 years, and a new JIA diagnosis within the last 10 months. Exclusion criteria included a developmental delay, a comorbid illness (e.g., asthma), a diagnosed sleep disorder reported by a parent and the medical record (e.g., obstructive sleep apnea), prior tonsillectomy and/or adenoidectomy, or a family history of narcolepsy in a first-degree relative. Parent inclusion criteria included at least 18 years of age and able to speak and read English.

Screening Procedures

Young children with JIA and one parent were recruited from a large urban hospital in the Pacific Northwest. A clinical research associate (CRA) screened the medical clinic records for potential participants. During a routine clinic visit, the CRA met with potential participants to confirm eligibility, discuss the study, and invite participation. Control children and their parents were recruited using flyers and through media advertisements in the community. Interested parents contacted a member of the research team who discussed the study and screened for eligibility. Eligible parent-child dyads in both groups were scheduled for a home visit to obtain parent consent, child assent, explain daily sleep and symptom diary, and application of the actigraphy watch.

Measures

Sleep Quality

Actigraphy sleep efficiency (SE). Each child wore an actigraph watch around the ankle and a parent wore the actigraph watch on the non-dominant wrist (Actiwatch 64TM, Mini-Mitter Philips Respironics, Bend, OR) for 10 consecutive days. Parents kept a sleep diary and were instructed to press the event marker at bedtime when they and their child attempted to fall asleep, and at wake time (final wake time to start the day). Data were recorded in 1-minute

epochs and downloaded to the actigraphy scoring software for analysis (Mimi-Mitter Philips Respironics, Inc.). Sleep variables measured included: 1) sleep onset, defined as the initial three or more consecutive minutes of sleep; 2) sleep offset, defined as the last five or more consecutive minutes of sleep; 3) total sleep time (TST), defined as minutes scored as sleep between sleep onset and offset; and 4) sleep efficiency (SE), defined as TST divided by the time from sleep onset to offset, reported as a percent. SE was used as the objective measure of sleep quality, and values closer to 100% were indicative of better sleep quality. Approximately 10.6% of nights for children and 10.9% of nights for parents were not scored due to artifact, participants not wearing the device, or actigraphy data without corresponding sleep diary data.

Diary sleep quality (diary SQ). Parents completed daily sleep diaries each night at bedtime and each morning upon awakening for 10 consecutive days. Each morning upon waking up, parents rated their own sleep quality and their child's sleep quality on a Likert Scale (1 = terrible, 9 = great).

Symptoms

Parent symptoms (daytime fatigue and nighttime worry). Parents completed daily symptom diaries for themselves each night at bedtime and each morning upon awakening for 10 consecutive days. The daily diaries included measures of daytime fatigue and nighttime worry. Parents rated the severity of daytime fatigue at bedtime and nighttime worry upon awakening in the morning. Item responses for symptom severity ranged from 0 to 3 (0 = not at all; 1 = slight; 2 = moderate; 3 = extreme).

Child symptoms (daytime fatigue and pain). Parents completed daily symptom diaries for their child for 10 consecutive days. Parents rated the severity of their child's daytime fatigue and pain at bedtime each night. Item responses for symptom severity ranged from 0 to 3 (0 = not

at all; 1 = slight; 2 = moderate; 3 = extreme).

Family Functioning. A parent completed the Family Adaptability and Cohesion Evaluation Scale II (FACES II), a 30-item report of family adaptability and cohesion. Parents rate the frequency of each behavior (e.g., “family members are supportive of each other during difficult times”) on a 5-point Likert scale (“1= almost never” to “5=almost always”). Responses were summed to yield adaptability and a cohesion scores. Higher scores indicate better family adaptation and cohesion. The reliability of the FACES is well established in children with and without chronic conditions (Huygen, Kuis, & Sinnema, 2000; Kouneski, 2000; Olson, 1986). In this study the Cronbach’s α was 0.78 for adaptability and 0.59 for cohesion.

Demographic and JIA Clinical Characteristics

Parents completed demographic survey (e.g., child age, sex, parent education, marital status, ethnicity). JIA clinical characteristics included disease duration and disease activity. JIA disease duration was defined as the time between diagnosis and data collection, and obtained from the clinic record. A pediatric rheumatologist assessed disease activity during a clinic visit, and included: 1) physician global assessment (PGA) of overall JIA disease activity rated on a scale of 0 to 10 (0 “no disease” to 10 “very severe disease”), and 2) number of active joints, defined as number of joints with active synovitis during the examination. Active disease was defined as PGA disease activity > 0; joints with active arthritis; active uveitis; fever, rash, serositis, splenomegaly, or lymphadenopathy attributable to JIA; elevated erythrocyte sedimentation rate (ESR); or morning stiffness >15 minutes (Wallace et al., 2011).

Statistical Analysis

The data were analyzed using the statistical software *R* (R Development Core Team, 2013). Descriptive statistics and graphs were used to describe the demographic and clinical

characteristics, sleep quality (actigraphy sleep efficiency [SE], diary sleep quality [diary SQ]), child symptoms (daytime fatigue and pain) parent symptoms (daytime fatigue and nighttime worry), and FACES (adaptability score, cohesion score). Differences between the two groups (JIA and control) in the demographic characteristics and FACES scores were examined using *t*-tests for continuous variables and chi-square analyses for categorical variables.

For sleep quality and symptom variables, only days with both actigraphy and matching sleep diary data were included in the analyses. All subjects had matched actigraphy and diary data for at least 5 nights. Fourteen children in the JIA group and 16 in the control group were included in the analysis for a total of 262 days of data; thirteen parents in the JIA group and 16 in the control group were included in the analysis for a total of 261 days of data. One parent's actigraphy and symptom diary data were not included in the analysis due to data loss.

For question 1 - whether daily sleep quality was related to symptoms in *parents* (daytime fatigue and nighttime worry), two types of models were used: (1) four models were tested to examine whether parent's daytime fatigue and nighttime worry predicted parent's sleep quality that night (SE and diary SQ); and (2) two additional models with temporal reverse analysis were tested to assess whether parent's sleep quality (SE and diary SQ) predicted next-day fatigue. For question 2 – whether daily sleep quality was related to symptoms in *children* (daytime fatigue and pain), similar models were used to examine whether child's daytime symptoms predicted sleep quality that night and whether child's sleep quality predicted next-day symptoms. The effects of age and group status (controls coded as 0, JIA coded as 1) were tested in each of the models to determine how these factors impacted the relationships between sleep and symptoms. *P*-values less than 0.05 were considered significant.

The analyses of the relationships between sleep quality and symptoms were conducted

using generalized estimating equations (GEE) models with packet “gee” within *R* (Carey, 2015). GEE is a method for fitting regression models to hierarchical data while accounting for the correlations between daily measures (sleep quality and symptoms) within each cluster (subject). Working independence matrix and robust variance estimates were used in the analyses. The robust estimates were considered valid even if the assumptions about the working covariance are not accurate (Hanley, Negassa, Edwardes, & Forrester, 2003). Similar to simple regression models, the GEE models produce an unstandardized β coefficient that indicates the magnitude and direction of association in changes in the predictor variable with changes in the outcome variable. To facilitate comparison of the β coefficients across models, the *z* scores (over all days) of the predictor and outcome variables were used in the analyses. When analyzing clustered data, it is important to separate the within-subject effect (daily effect) from the across-subject effect (average effect) (Mancl et al., 2000). For example, the statistical model below shows the child daytime pain as a predictor of child SE. The across-subject term PainMean_i was calculated as the mean pain level in each child across the 10 days. The coefficient of PainMean_i (β_1) would be interpreted as an estimate about for each child, how much SE would change on average when pain ratings changed on average. The within-subject term $(\text{PainDay}_{ij} - \text{PainMean}_i)$ was calculated as the deviation of the daily pain level from the child mean pain level. The coefficient of $(\text{PainDay}_{ij} - \text{PainMean}_i)$ (β_2) would be interpreted as an estimate about within a child in one day, how much SE would change when pain ratings changed that day. The multiple correlation coefficient squared (R^2) was also computed to decompose the predictor’s effects on the outcome variable into within- and across-subject sources of variance.

$$SE_{ij} = \beta_0 + \beta_1 \text{PainMean}_i + \beta_2 (\text{PainDay}_{ij} - \text{PainMean}_i) + \varepsilon_{ij}$$

To address question 3 – whether family functioning is associated with sleep quality in children, Pearson's r correlations were computed between FACES scores (adaptability and cohesion scores) and average sleep quality (SE and diary SQ) across days for each child.

Results

Participants

Table 3.1 shows the sample demographic and clinical characteristics. Significant age group differences were found between young JIA and control children ($p < 0.05$). Control children were 16 months older than JIA children.

[Table 3.1 about here]

Question 1: What Are the Relationships between Daily Sleep Quality and Symptoms of Fatigue and Nighttime Worry in Parents of Young Children with JIA and Control Parents?

Table 3.2 shows the symptoms of fatigue, worry, and sleep quality for parents of children with JIA and control parents. Parents of children with JIA had significantly more nighttime worry and lower SE compared to parents of control children (p 's < 0.05), but significant differences in fatigue were not found.

[Table 3.2 about here]

Table 3.3 shows the daily symptoms (daytime fatigue, nighttime worry), and sleep quality for parents of children with JIA in comparison to control parents. In the entire sample, on average, daytime fatigue and nighttime worry negatively predicted diary SQ (p 's < 0.05); and diary SQ negatively predicted next-day fatigue ($p < 0.05$) (See Table 3.3). Figure 3.1.a shows the within-subject daily relationships between fatigue, worry, and sleep quality. Daily nighttime worry negatively predicted diary sleep quality ($p < 0.001$), and both daily SE and diary SQ

negatively predicted next-day fatigue (p 's <0.05). Daily daytime fatigue did not predict SE or diary SQ. These relationships did not differ between the two groups of parents.

[Table 3.3 about here]

[Figure 3.1 about here]

Question 2: What Are the Relationships between Daily Sleep Quality and Symptoms of Fatigue and Pain in Young Children with and without JIA?

Table 3.4 shows the daily symptoms (pain, fatigue) and sleep quality for children with JIA in comparison to control children. Young children with JIA had significantly more daytime fatigue and pain, and lower SE compared to control children (p 's <0.05), but not diary SQ ($p >0.05$; see Table 4). In the entire sample, on average, daytime fatigue negatively predicted SE, and both daytime fatigue and pain negatively predicted diary SQ (p 's <0.05). On temporal reverse analyses, SE did not predict next-day fatigue or pain; rather diary SQ negatively predicted next-day fatigue and pain (p 's <0.05) (See Table 4).

Figure 3.1.b shows the within-subject daily relationships between symptoms and sleep quality. Daily daytime pain negatively predicted SE and diary SQ within a child. Diary SQ negatively predicted next-day pain within a child (p 's <0.05). The within-subject relationships between daytime fatigue and sleep quality were not significant, and these relations did not differ between the two groups of young children. The addition of age as a covariate in the models did not change the relationships.

[Table 3.4 about here]

When examining the effects of fatigue and pain on sleep quality (SE and diary SQ), we decomposed the effects into within- and across-subject sources of variance using R^2 (see table 4). As shown in table 4, the across-subject term had larger R^2 than the within-subject term,

suggesting that subject-level variables (e.g., demographic or trait variables) accounted for more variability in sleep quality versus the variability in daily symptoms. The within-subject model using daytime pain to predict SE had a larger R^2 , which suggests a stronger daily effect of pain on sleep efficiency (e.g., physiological mechanisms) within a child. Similar results were found in the models using sleep quality to predict next-day symptoms.

Question 3: On Average, Is Children's Sleep Quality Associated with Family Functioning in Families of Young Children with and without JIA?

Table 5 shows the mean parent-reported FACES scores between JIA and control groups (adaptability and cohesion). The cohesion and adaptability scores were not significantly different between JIA and controls (p 's > 0.05). Neither cohesion nor adaptability scores were correlated with children's average sleep quality scores (p 's > 0.05).

[Table 5 about here]

Discussion

In parents of young children with JIA, both daily and average nighttime worry predicted poor sleep quality, which subsequently predicted next-day fatigue. In young children with JIA, both daily and average daytime pain predicted poor sleep quality, which subsequently predicted increased next-day pain; average fatigue severity, not daily fatigue predicted poor sleep quality, and poor sleep quality predicted increased next-day fatigue. Our findings extend the results from existing literature to young children recently diagnosed with JIA and their parents, and suggest that sleep is an important variable associated with disease-related symptoms. Young children and their parents may benefit from interventions to promote better sleep quality.

Parent Sleep Quality and Symptoms

Parents of young children with JIA reported more nighttime worry that predicted worse sleep quality. This finding may be attributed to parents' concern about their child's recent diagnosis and/or caregiving responsibilities (e.g., managing disease-related symptoms, medications) that could negatively impact parents' sleep quality. Our finding replicates previous studies that also report inverse associations between parental nighttime worry and poor sleep quality (e.g., prolonged sleep onset, symptoms of insomnia) (Herbert, Monaghan, Cogen, & Streisand, 2015; Matthews et al., 2014; Neu et al., 2014). For example, Herbert and colleagues (2015) found positive associations between increased parental worry and worse sleep quality in parents of children with diabetes. Neu and colleagues (2014) also found positive associations between increased parental worry related to child treatment regimes, financial problems, and employment and poor sleep quality in mothers caring for children with ALL. Parental worry in the above studies was related to child's treatment regimens, financial and employment challenges. Additional research is needed to further explore parents' concerns and worry in caring for a young child newly diagnosed with JIA. This information would be helpful to pediatric rheumatologists and nurse practitioners who care for newly diagnosed children and their families. Parental adjustment to the chronic condition, family strain, and the impact of a new chronic health condition on the daily home and work schedule may also contribute to our findings, but these variables were not measured in this pilot study.

Both groups of parents had similar fatigue severity. We anticipated that JIA parents would report more fatigue severity but this was not the case. This finding may be related to the fact that caring for a young child is demanding regardless of a chronic condition and/or the fatigue measure used in this study was not sensitive enough to detect differences. Despite the lack of group differences in parental fatigue, our finding is similar to Meltzer and colleagues who

also found similar fatigue levels in parents caring for children with cystic fibrosis in comparison to parents of control children (Meltzer & Mindell, 2006). Future studies should include fatigue measures that assess different domains of fatigue including general fatigue, sleep/rest, and cognitive fatigue that would provide important information for pediatric providers in caring for these families.

As expected, poorer sleep quality predicted an increase in next-day fatigue. Our finding replicates previous studies that also show positive correlations between sleep disturbances and fatigue in parents caring for children with leukemia, cystic fibrosis, and ventilator dependency (Meltzer & Mindell, 2006; Zupanec, Jones, & Stremmler, 2010). Our findings added to the existing literature that show the relationship between fatigue and sleep quality in parents maybe directional with poor sleep quality affecting next-day fatigue rather than fatigue affecting sleep quality. The development of interventions to promote better sleep quality is needed to not only improve sleep but may also reduce fatigue levels in parents caring for young children.

Child Sleep Quality and Symptoms

In both JIA and control children, average daytime fatigue predicted poorer sleep quality as measured by the sleep diary, but not actigraphy. This finding may be explained by parent's subjective perception of fatigue that was related to their subjective perception of sleep quality. It is unclear whether the association between sleep quality and fatigue is primarily due to the underlying sleep disturbances and/or JIA disease mechanisms that may contribute to the development of fatigue over time. Similar to our findings, a previous study of adolescents with chronic pain including JIA also report positive associations between average fatigue and poor sleep quality (Tham, Holley, Zhou, Clarke, & Palermo, 2013). Tham found that poorer baseline self-report sleep quality, not actigraphy sleep quality, predicted increased fatigue severity 12

months later. The combined findings provide evidence for the need to develop behavioral interventions to improve sleep quality, which may subsequently decrease fatigue in children with JIA.

Daily and average daytime pain predicted poorer diary sleep quality in young children with and without JIA. Similarly, both daily and average poor diary sleep quality predicted next-day pain. These findings support the theoretical framework about the bidirectional relationships between sleep and pain (Lewin & Dahl, 1999). Previous studies in school-age children and adolescents report similar findings of the temporal relationships between daily pain and sleep quality (Bromberg, Connelly, Anthony, Gil, & Schanberg, 2015; Bromberg et al., 2012). Bromberg and colleagues (2012) found that average pain intensity, not daily pain intensity, negatively predicted sleep quality in adolescents with JIA, but both average and daily sleep quality negatively predicted next-day pain intensity. The difference between the current study and Bromberg and colleagues (2012) is the time of the day symptoms were reported. In the current study, daytime pain was reported at bedtime, and sleep quality was reported in the morning, whereas Bromberg and colleagues reported pain and sleep quality in the evening. The differences in the time of day may have influenced the recall of previous night's sleep quality.

Daily variations in pain severity predicted actigraphy sleep quality (SE). Our finding may suggest an underlying biological relationship between pain and sleep quality such that prolonged and recurrent pain may cause elevated vigilance to signal pain onset, which in turn can contribute to poor sleep quality (Lewin & Dahl, 1999). Lewandowski and colleagues examined the temporal daily associations between self-report pain and sleep quality measured by both actigraphy and self-report in adolescents with and without chronic pain (Lewandowski et al., 2010), and show that pain did not predict sleep quality by self-report or actigraphy; rather

increased wake after sleep onset predicted increased next-day pain. Our sample of young children with JIA were also taking medications to treat JIA, as well as pain medications as needed, which may alter the relations between pain and sleep quality. Other factors may also influence the relationship between pain and sleep. For example, mood and daily functional limitations were found to mediate or moderate the relationship between sleep and pain (Bromberg et al., 2015; Bromberg et al., 2012). Future studies examining the temporal relationships between daily sleep and pain may consider the mediating or moderating effect of other variables such as mood and functional limitations.

Family Functioning and Children's Sleep Quality

Family functioning did not differ between families of young children with and without JIA, and it was not correlated with sleep quality. No visible patterns were detected via scatterplots and Lowess curve fitting. This finding was unexpected and may be explained by the measure we used to examine family function or the small sample size. Family adaptability and cohesion were the two constructs measured, and family functioning is a multidimensional construct (Rolland, 1993). Important dimensions, such as family stress, family communication, organization and structure were not examined in this study. Prior studies that examined family function in JIA are inconsistent (Gerhardt et al., 2003). For example, Gerhardt and colleagues examined family functioning in four dimensions (family relationship, support, conflict, and control), and found no differences in school-age youth with and without JIA (Gerhardt et al., 2003). Other studies show significant differences in some dimensions of family functioning, including family conflict, family cohesion, and family adaptation (Conte, Walco, & Kimura, 2003; Huygen et al., 2000). Further research is needed to examine the multidimensional constructs of family functioning in young child with chronic conditions, and the extent to which

a chronic condition, like JIA may affect the family so that tailored programs could be developed to assist these families.

Notable strengths of this study include objective and self/proxy-report measures of sleep in parent-child dyads with and without a new chronic condition, and the temporal relations of sleep and symptoms. There are also a number of limitations in this study. First, multiple comparisons were not controlled for given the small sample size and the exploratory nature of the pilot study. Second, symptom frequency in fatigue, pain, and worry was not assessed and may explain the low variability in some of these measures. Third, we did not control for medication use in JIA, which can impact sleep and symptoms of pain and fatigue. Lastly, parental mood (stress) was not measured and may contribute to our findings.

Conclusion

Symptoms of pain, fatigue, and poor sleep quality were interrelated in children newly diagnosed with JIA. A new diagnosis of JIA was associated with parent's sleep quality and health and wellbeing. Poor sleep quality is a modifiable health behavior, and improving sleep quality may alleviate or reduce disease-related symptoms in young children with a new chronic condition and their parents. Routine sleep assessments in both children with JIA and their parents are needed during clinical care to identify families at high risk for poor sleep quality.

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Table 3.1 *Demographic and Disease Characteristics of the Sample*

Parent Characteristics	Control (n = 16)	JIA (n =13)
Age, years	34.9 ± 4.2	32.8 ± 4.4
Female, n (%)	14 (88%)	13 (100%)
Education, n (%)		
Some college	2 (13%)	6 (46%)
College degree	10 (63%)	5 (39%)
Master's degree	4 (25%)	2 (15%)
Married, n (%)	16 (100%)	11 (85%)
Ethnicity, n (%)		
Caucasian	13 (81%)	10 (77%)
Hispanic	0	1 (8%)
Asian	3 (19%)	1 (8%)
Other	0	1 (8%)
Child Characteristics	Control (n = 16)	JIA (n =14)
Age, years *	4.0 ± 1.1	2.8 ± .7
Female, n (%)	12 (75%)	11 (79%)
Ethnicity, n (%)		
Caucasian	10 (63%)	9 (65%)
Hispanic	0	2 (14%)
Asian	2 (12%)	1 (7%)
Other	4 (25%)	2 (14%)
JIA Characteristics		
JIA Category, n (%)		
Oligoarticular	-	7 (50%)
Extended oligoarticular		1 (7%)
Polyarticular		6 (43%)
Active JIA, n(%)	-	10 (71%)
JIA Duration, months	-	7.1 ± 2.3
JIA Active Joint, (#)	-	2.2 ± 2.1
PGA Disease Activity	-	1.6 ± 1.8

JIA – juvenile idiopathic arthritis; PGA – physician global assessment.

Data are mean ± SD or n (%).

* $p < .05$, ** $p < .01$, *** $p < .001$ (significant group differences).

Table 3.2 Mean and Standard Deviations of Child and Parent Sleep Quality and Symptoms

Parent	Mean (SD)		Group Differences (Control vs. JIA)			
	Control	JIA	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
DT Fatigue	.81 (.88)	1.04 (.83)	.23	.21	1.10	.27
NT Worry	.16 (.46)	.47 (.77)	.31	.16	1.95	.05 *
SE (%)	87.76 (4.49)	84.08 (6.98)	-3.67	1.84	-1.99	.04 *
Diary SQ	6.45 (1.56)	5.65 (2.06)	-.79	.45	-1.78	.08
Child	Mean (SD)		Group Differences (Control vs. JIA)			
	Control	JIA	<i>b</i>	s.e.	<i>t</i>	<i>p</i>
DT Fatigue	.18 (.51)	.64 (.82)	.47	.14	3.24	.001 **
DT Pain	.11 (.45)	.70 (.89)	.59	.15	3.91	<.001 ***
SE (%)	84.81 (3.77)	82.21 (6.10)	-2.60	1.10	-2.39	.02 *
Diary SQ	7.77 (1.54)	6.97 (2.25)	-.80	.46	-1.73	.08

JIA – juvenile idiopathic arthritis, DT – daytime, NT – nighttime, SQ – sleep quality.

* $p < .05$, ** $p < .01$, *** $p < .001$; Control coded as 0, JIA coded as 1.

Table 3.3 Relationships between Parent Sleep Quality and Symptoms

Symptoms as Predictors of Sleep Quality									
Symptoms → SE		Across-subject (Average effect)				Within-subject (Daily effect)			
Predictors	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²	
DT Fatigue	-.02	.19	.91	<.1	-.08	.08	.30		.4
NT Worry	-.14	.23	.55	.8	-.08	.04	.07		.3
Symptoms → Diary SQ									
Predictors	<i>b</i>	s.e.	<i>p</i>		<i>R</i>²	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²
DT Fatigue	-.35	.15	.02	*	5.0	-.4	.05	.49	.1
NT Worry	-.73	.16	<.001	***	20.9	-.31	.10	<.001	*** 5.3
Sleep Quality as Predictors of Next-Day Symptoms									
SE → Symptoms		Across-subject (Average effect)				Within-subject (Daily effect)			
Outcome	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²	
Next-D Fatigue	.001	.14	.96	<.1	-.14	.07	.047	*	.6
Diary SQ → Symptoms									
Outcome	<i>b</i>	s.e.	<i>p</i>		<i>R</i>²	<i>b</i>	s.e.	<i>p</i>	<i>R</i>²
Next-D Fatigue	-.32	.14	.02	*	4.2	-.20	.08	.01	* 2.1

SE – sleep efficiency, SQ – sleep quality, DT – daytime, NT – nighttime, Next-D – Next day, *R*² – percent variance of the outcome explained by the predictor.

* *p* < .05, *** *p* < .001; Group (control-0, JIA-1) was entered as a covariate in all models.

Table 3.4 Relationships between Child Sleep Quality and Symptoms

Daytime Symptoms as Predictors of Sleep Quality									
Symptoms→SE		Across-subject (Average effect)				Within-subject (Daily effect)			
Predictors	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	
DT Fatigue	-.10	.22	.65	.3	.07	.10	.46	.3	
DT Pain	-.001	.28	.99	<.1	-.22	.06	<.001	***	2.7
Symptoms→ Diary SQ									
Predictors	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	
DT Fatigue	-.68	.18	<.001	***	13.3	-.14	.08	.09	1.2
DT Pain	-.51	.21	.01	*	7.1	-.14	.05	.002	**
Sleep Quality as Predictors of Next-Day Symptoms									
SE→Symptoms		Across-subject (Average effect)				Within-subject (Daily effect)			
Outcomes	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	
Next-D Fatigue	-.13	.19	.52	.5	-.01	.04	.85	<.1	
Next-D Pain	-.01	.25	.96	<.1	-.05	.06	.42	.1	
Diary SQ →Symptoms									
Outcomes	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	<i>b</i>	<i>s.e.</i>	<i>p</i>	<i>R</i>²	
Next-D Fatigue	-.43	.15	<.001	***	8.5	-.06	.05	.25	.2
Next-D Pain	-.39	.12	.002	**	6.9	-.22	.10	.03	*

SE – sleep efficiency, SQ –sleep quality, DT- daytime, NT – nighttime, Next-D – Next-day, *R*²- percent variance of the outcome explained by the predictor.

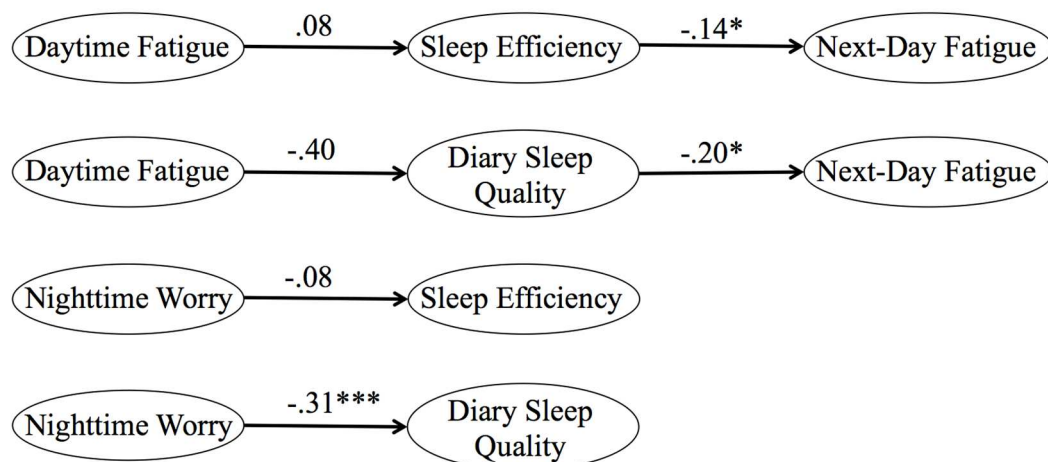
* *p* < .05, ** *p* < .01, *** *p* < .001; Group (Control-0, JIA-1) was entered as a covariate in all models.

Table 3.5 *Family Adaptability and Cohesion Scale (FACES) and Sleep Quality*

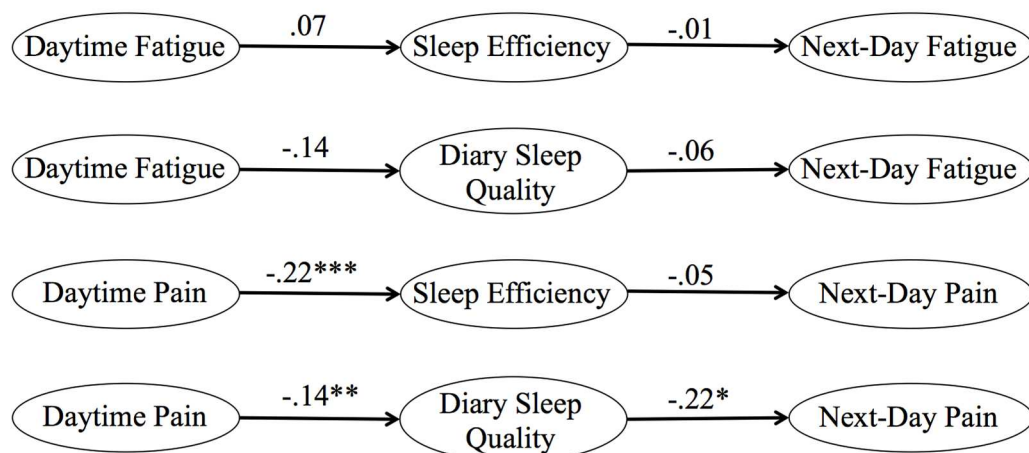
	Family Functioning			Pearson's <i>r</i> Correlations with Child Sleep Quality	
	Control	JIA	Group Differences	Sleep Efficiency	Diary Sleep Quality
Family Adaptation	50.2±5.6	52.5±6.2	-	-.25	-.23
Family Cohesion	70.9±5.6	70.8±6.5	-	.03	.04

Figure 3.1 *Temporal Daily Relationships between Sleep Quality and Symptoms in Parents and Children with and without JIA*

a. *Temporal Daily Relationships between Sleep Quality and Symptoms in Parents of Children with JIA and Control Parents*



b. *Temporal Daily Relationships between Sleep Quality and Symptoms in Children with and without JIA*



* $p < .05$, ** $p < .01$, *** $p < .001$

The parameter estimates (β 's) are the within-subject daily effects; Group (control = 0, JIA = 1) were entered as a covariate in all models.

Chapter 4. Struggling in the Dark to Help My Child: Parents' Experience in Caring for a Young
Child Newly Diagnosed with Juvenile Idiopathic Arthritis

Abstract

Purpose: The purpose of this study is to describe parents' experiences in caring for 2-5-year-old children newly diagnosed with JIA.

Methods: A qualitative study using single-occasion semi-structured interviews was conducted. Nine parents participated in the study (eight mothers and one father). Parents were interviewed in-person or via telephone. Interview data were analyzed using inductive content analysis. Methods were used to protect the trustworthiness of study results: maintenance of an audit trail, peer debriefing, and a formal member check.

Results: The core construct *Struggling in the Dark to Help My Child* explained parents' experience in six domains: *not knowing, trying to reach out in the dark, feeling my child's pain, working out the kinks to stay on top to manage, feeling drained from the whole process, and being hard on the entire household*. Parents struggled trying to understand the illness with unknown cause, the different options of treatments, and what the future might hold for their child. Parents felt totally alone and in the dark, and tried to reach out for resources, but found there was not much available. Parents struggled seeing their child in pain, but knew that there was not much they could do to help. Parents blamed themselves, and some even felt they were a bad parent who betrayed their child. Parents tried everything they could to work out the kinks and stay on top to manage JIA, even when they felt totally drained physically and emotionally. JIA not only consumed their lives, but also affected the entire household, including the siblings and spouse, and the relationships among family members.

Conclusions: Findings from this study highlight the day-to-day lived challenges parents face when caring for a young child newly diagnosed with JIA. Additional research with a larger sample is needed to inform future intervention trials.

Introduction

Juvenile idiopathic arthritis (JIA) is a common pediatric autoimmune disease that currently has no cure. Children experience unforeseeable episodes of active disease that are characterized by joint inflammation, pain, tenderness, and limited mobility. The management of JIA can be time-consuming and complex, and includes pharmacological interventions, physical therapy, routine laboratory and imaging tests, and regular appointments with specialists such as ophthalmologists and dentists (Prakken et al., 2011). The majority of the JIA care and management requires parental involvement particularly for young children 2-5 years old, yet few studies have examined the illness-related demands and challenges parents encounter when caring for a young child with JIA.

To date, five qualitative studies have been conducted to describe parents' experience in caring for children and adolescents with JIA (see Table 4.1). Common parents' experiences have included confusion and emotional turmoil; feelings of guilt, anxiety, anger, frustration, and fear; and a lack of social support (Barlow et al., 1998; Gomez-Ramirez et al., 2016; Jerrett, 1994; McNeill, 2004; Stinson et al., 2012). Barlow and colleagues found that parents' personal time with other family members was often restricted; this was particularly true in parents caring for younger children who required more supervision and assistance (Barlow et al., 1998). The purpose of this descriptive study was to describe parents' perceived stress, illness-related challenges, coping behaviors, and adaptation to the stress and challenges when caring for their 2-5-year-old child with a new diagnosis of JIA.

[Table 4.1 about here]

Conceptual Framework

An integration of the Family Systems Theory and the Resiliency Model of Family Stress, Adjustment, and Adaptation – hereafter called the Resiliency Model (Broderick & Smith, 1979; M. A. McCubbin & McCubbin, 1993; Olson et al., 1983) formed the conceptual framework for this study. The Family Systems Theory is a theory of human behavior that views the family as an emotional unit and uses systems thinking to describe the complex interactions in the family unit. The Family Systems Theory holds that families are systems of interconnected and interdependent individuals. Illness in a family member can impose different levels of burden and strain on parents, siblings, caregivers, and other members of the family (Broderick & Smith, 1979; Kerr & Bowen, 1988). In this study, the Family Systems Theory laid the foundation for understanding how a child's JIA affects the family system. However, this framework does not address the processes of stress, coping, and adaptation that occur in a family. Thus, the Resiliency Model was selected to complement the Family Systems Theory to inform the current study.

The Resiliency Model views illness as a stressor that affects family life. The family's appraisal of the situation influences their coping and problem-solving abilities as they deal with the stressor (M. A. McCubbin & McCubbin, 1993). The key concepts chosen from this model for the current study were stress, coping, and adaptation. A stressor is a demand placed on the family that produces, or has the potential to produce changes in the family system. An illness of a family member is considered a stressor in the family that can produce tension in the family. If the tension is not reduced, stress merges (Lewis, 2010). Coping refers to the family's behaviors designed to maintain or strengthen the family as a whole, maintain the emotional stability and well-being of its members, use resources to manage the situation, and initiate efforts to resolve family challenges created by the stressor. Family adaptation describes the outcome of family

efforts to bring a new level of balance, coherence, and functioning to a family stress. Family adaptation involves positive changes within the family and positive transactions between the family and the community (in this case, the healthcare system). Family struggles to achieve a balance between meeting the needs of the individual members of the family and the well-being of the family as a whole (Lewis, 1993). Families with an ill member also benefit from the support and professional care and guidance received from the healthcare professionals (M. A. McCubbin & McCubbin, 1993; Olson et al., 1983).

Methods

Participants

Fourteen primary caregiving parents (either mother or father) of a 2-5-year-old child with JIA were recruited from an existing observational study of sleep and health outcomes in young children with JIA and their parents. Parents were eligible if they were least 18 years of age, able to speak and read English, and had a 2-5-year-old child diagnosed with JIA within the past 10 months. Parents had given consent in the original form to be contacted for future studies. Of the fourteen eligible families, eight mothers and one father ($n = 9$) from eight families were enrolled and completed the study (57% response rate). In one family both parents identified themselves as the primary caregivers.

Procedure

Human Subjects approval was obtained from the University of Washington Institutional Review Board. Parents were mailed a passive letter informing them about the study and were instructed to contact the author within two weeks if they wish not to participate. None of the parents opted out within the two-week period. After two weeks, the author contacted the parents and asked if they were willing to participate. If the parents agreed to participate, the author

mailed the consent form, and explained the study purpose and procedure over the phone. Interested parents signed and mailed the consent form to the author, and then the author scheduled an in-person or phone interview. One parent chose to be interviewed in-person at the University of Washington and eight parents chose to have a phone interview.

The semi-structured interview consisted of open-ended questions listed in Table 4.2. Interviews lasted 22 to 74 minutes (median 45 minutes) and were audio-recorded with permission. The recordings were transcribed by a professional transcriptionist and verified 100% for accuracy by the author. Each participant received a \$75 gift card for participation.

[Table 4.2 about here]

Data Analysis

Transcribed interview data were imported into qualitative analysis software Atlas.ti Mac Version 1.0. Using a multi-phased process, data were analyzed using inductive content analysis. Inductive content analysis was developed by Glaser and Strauss (1967), Krippendorff (1980) and Strauss and Corbin (1990), and extended by Lewis and Deal (1995). This method was derived from grounded theory methodology and rooted in symbolic interactionism. Symbolic interactionism is the process of interaction in the formation of meanings. Meanings were formed by the experience of the individuals and the social processes from which individuals derive socially constructed meaning (Strauss & Corbin, 1990). Inductive content analysis is a method for making inferences from data to their context, with the purpose of providing knowledge and new insights. Data analysis occurred in five phases: 1) Unitizing data; 2) Open coding and identification of initial categories; 3) Naming and defining categories; 4) Identification of higher order domains, naming and defining domains; and 5) Identification of a core construct that

further organized and explained the domains and categories. Details of the data analysis steps are described in Table 4.3.

[Table 4.3 about here]

The trustworthiness of study results were protected by keeping an audit trail, peer debriefing and member check (Cho & Trent, 2015; Corbin & Strauss, 2008). Audit trails, chronological records of analysis that provide documentary evidence of the sequence of activities, began with the first analytic session and continued throughout the analytic process. Audit trails were also used for peer debriefing. Peer debriefing by a senior researcher with expertise in inductive content analysis occurred in five areas that examined: 1) the accuracy of the unitized data; 2) the initial open codes from the units; 3) the categories and category labels; 4) the organization of the categories within the domains; and 5) the identification of the core construct. Throughout the analysis process, the senior researcher used constant comparative analysis to protect the fit of the unit with the category and the uniqueness and distinctness of each category and domain, and worked with the senior researcher to constantly review and refine the categories, their definitions, and the domains and their definitions. Disagreement about any aspect of the analysis was discussed and resulted in a refinement in the definition of a category or a domain, a reassignment of a unit/category, or the development of a new category or a domain. Coding decisions were based on 100% consensus between the senior researcher and the author. After the initial analysis, a one-page summary was mailed to the participants. All participants agreed to receive the study summary and were approached for a member check with the author with a follow-up phone call. Four questions were asked during member check process: 1) What is your overall response of the summary? 2) What are the parts that fit your experience?

3) What are the parts that did not fit your experience? 4) What else about the summary that you may want to tell me?

Results

Parents ranged from 31 to 39 years (median age 34 years). Among the nine parents, 11% ($n=1$) were divorced at the time of JIA diagnosis, and 44% ($n=4$) were separated or divorced at the time of the interview; 67% ($n=6$) were employed full time. The median time since their child's JIA diagnosis was 18 months (range from 10 to 33 months). Median age for the child with JIA was 3.5 years.

Core Construct: *Struggling in the Dark to Help My Child*

The core construct *Struggling in the Dark to Help My Child* described parents' experiences in caring for their young child newly diagnosed with JIA. Parents struggled in trying to understand the illness, the cause, the various options for treatment, and what the future might hold for their child. Parents felt totally alone and in the dark, and tried to reach out for resources, but found few available. Parents also struggled seeing their child in pain, but knew that there was not much they could do to help. They blamed themselves, and some even felt they were a bad parent who betrayed their child. Parents tried everything they could to stay on top to manage the illness, even when they were physically and emotionally drained. JIA consumed their life, and also affected the entire household, including the siblings, spouse, and the relationships among family members. The core construct described the parents' experience in six domains: *not knowing, trying to reach out in the dark, feeling my child's pain, working out the kinks to stay on top to manage, feeling drained from the whole process, and being hard on the entire household*. The domains and their categories are summarized in Table 4. A description of the domains and the categories follows, including illustrative quotes from parents.

[Table 4.4 about here]

Domain 1: Not Knowing

Parents received an overwhelming amount of information about the new diagnosis. However, they did not fully understand the illness, had substantial uncertainty about their child's future, and had difficulty making decisions about treatments. Parents often felt they had to guess what their child was experiencing because it was challenging for young children to articulate their feelings. The uncertainties made them feel scared and unsettled. "Not Knowing" consisted of seven categories. The description of each category and sample emics are described below.

Not really understanding. Parents did not understand what was happening with their child's body with JIA, how bad the illness was, or the underlying damage. "The biggest [challenge] for us really...just emotionally dealing with the fact that we didn't really understand."

Not able to predict the future. Parents described the uncertainty of the treatment options and the prognosis of JIA. They were informed that the illness could progress in different directions, but this depended on how their child would respond to the treatment. Parents were told that they would just have to wait and see how the treatment progressed. Parents felt uncertainty about their child's future. "Is your child gonna end up you know, with being sick all the time and with long term effects and um, and if you like, if you don't do something quick enough, she has bone damage."

Not knowing what to do. Parents did not know what decisions to make, such as whether they should or should not start certain medications for their child. "Even when you make a choice to do methotrexate or whatever, you don't know if that's the right choice or the best choice... We don't know what, how to best treat [JIA] because the treatment that works for one

child doesn't necessarily work for the others.” Parents also did not know if they should vaccinate their child or wait on a type of immunosuppressing medication to treat JIA; they did not know whether to take their child with compromised immunity to school where some children were not vaccinated. As one parent said, “there’s just no good answers.”

Unsettling to have no closure on any aspect. Sometimes parents were told that a medication was supposed to help their child, but the child seemed worse. “They [doctors] don't have the explanation of why this is happening, we don't know why it's happening... it's hard to feel like you come to any sort of acceptable resolution on it. It's like this unresolved thing and it's very unsettling to have no, no closure on any aspect of it.” Parents felt frustrated that even the best sources available did not provide answers.

Hard to discern between arthritis-related and other causes. Parents had to guess whether the child was in pain because of JIA or other things such as growth pain. Young children were not able to express themselves when they felt hurt or needed something. “That's a huge challenge like from a parent perspective...like sometimes he'll wake up and say, I'm having pain in my bone. You know. And you're just like, oh is it a flare, or is it something we should worry about... You don't know uh, how to trust, like you don't even know if he knows what he's saying half the time, you know?”

Feeling blindsided and overwhelmed. Parents felt blindsided by the huge amount of information about the diagnosis and the treatment options. They viewed the JIA diagnosis as so overwhelming they could not process the information. Despite feeling blindsided and overwhelmed, they had to make decisions for their child. “What if I, as my child's mother, decide that he's gonna go on this medicine, and he's the one out of the 600 or whatever that ends up having something really bad happen, even develop lymphoma, down the road and die. I, I just

felt like, having to be responsible for making that decision is, is really difficult and really overwhelming... or if I decided that he's not gonna go on it and then his arthritis progresses, and then he's limping around his whole life, it destroys his joints, then I, I'm the one that made that decision too.”

Scared of not knowing. Parents felt scared that they did not know what was happening. “When he was first diagnosed, it took them actually three months to diagnose him and initially they thought it was just a fracture. And we didn't even know. His dad was putting his shirt on and his arm was um bent. And he couldn't straighten it. And that's how we, we you know initially found out about this. And so just that fear of, I don't wanna miss anything again.” Sometimes when their child was on an immunosuppressant and had subtle symptoms of infection, parents felt “extremely terrified” that they did not know what was going on, or whether the infection might trigger other disease activity.

Domain 2: Trying to Reach Out in the Dark

Parents felt totally alone and in the dark when their child was diagnosed. They felt helpless with so many unknowns and uncertainty about JIA. They tried to talk to people with similar experience or search online, but there was not much helpful resource available. “Trying to Reach Out in the Dark” consisted of four categories.

Feeling totally alone. Parents felt that nobody could understand them and they felt alone. “I felt really kind of alone and in the dark when [child] was diagnosed. And I had um, my mom talked to a woman that she knows, whose daughter, has a daughter with JIA. And my mom got the girl's phone number and everything... And so I tried calling this girl a number of times and she never returned my call.”

Hearing stories here and there. Parents tried to reach out to people who knew about JIA, but could only hear scattered stories here and there. They reached out to others to gain understanding even if the others knew nothing about JIA, or had another totally different diagnosis. “[I had] another friend who has a friend with an older daughter with arthritis just told me that story I guess.” And another parent shared, “I mean I have an aunt that has osteoarthritis... It's totally different from what I understand than what [child] has, but she's the only person I know with arthritis so if I have any questions, you know, I, I ask her.”

Having no resources to draw on. Parents felt they did not really have any resources other than the medical team. For example, some children experienced side effects from JIA treatments, such as “extreme fatigue,” disturbed sleep, and compromised immune system. Parents did not know who to ask for help when they had a question about caring for their child with these symptoms. Parents also had challenges in administering medications to their child. In addition, some parents didn't feel like there was support resource to turn for help with their own emotions. “I just felt, I, I had no resources to draw on.” One consequence of the few resources was that parents “just felt helpless and powerless.”

Getting scary stories from the Internet. When parents had questions about the care for their child, but could not obtain help from other resources, they turned to the Internet. However, they found the worse case scenarios. One parent described her experience after reading scary stories from the Internet, “I'm in my bedroom having a full-on meltdown while my daughter is eating dinner in the kitchen because I read that [scary story]... They [hospital] you know, told you before don't Google. Yes I know that. Um but what are you supposed to do? Who am I supposed to ask? You know, if [hospital] tells me the, the medication is supposed to be helping it and it's not. And she seems to be getting worse, then, of course you're gonna, inquiring minds

you'll wanna know.” Another parent said, “if you're doing like a surface level research, it's very scary.” Some parents paid fees to obtain access to online medical journals for information about JIA.

Domain 3: Working Out the Kinks to Stay on Top to Manage

As children experienced JIA-related symptoms and medication side effects, parents were extra vigilant in monitoring their children. At the same time, parents had to figure out how to best manage their child's care, including attending medical appointments, medication administration, and finding ways to make their child feel better. “Working out the Kinks to Stay on Top to Manage” consisted of eight categories.

Battling with JIA symptoms. Children experienced joint pain, swelling, and limited range of motion. For some children, symptoms were so severe that they would refuse to walk. For example, one parent shared, “She'd complain a lot that she couldn't color or she couldn't draw. Um, I know that a lot of the time she had a lot of, a lot, a lot of sleepless nights... So a lot of crying, especially at night, very uncomfortable, very restless, to the point where I'd wanna pick her up and she wouldn't let me touch her. She wouldn't let me pick her up because she'd say, ‘ow’.”

Seeing my child getting sick all the time from treatment. Some children experienced medication side effects from the JIA treatment. Parents felt that the medication that was supposed to fix JIA was making their child sick in other ways. Parents were told explicitly by the doctors to expect some “subtle” side effects, but the side effect their child experienced did not appear subtle at all to them. Some side effects were particularly challenging for parents to accept or manage. For example, one parent described her child experiencing severe fatigue from methotrexate, “When she was fatigued, we'd give her shot and uh, her shot of methotrexate. She

would be fatigued for almost the next four days and when I say fatigued, she's usually a, about an hour napper normally. Um, she would take like three two-hour naps, for, for almost four days. So, when I'm saying fatigued, it was extreme fatigue. Um, so she'd have four days where she just was almost lethargic. Um, and then she'd be active for two. And then we'd give her her next dose." Other symptoms included upset stomach, nausea, and diarrhea. Some medications such as methotrexate suppressed children's immune system, so children were more likely to get sick. "Last December she was sick three weeks out of the month, again in February, and, and with different colds. So...she still would catch every possible bug that was coming through. Um and so that's hard because your child um is already having all those other problems, but even when the arthritis goes away with the treatment, she's been sick all the time."

Being extra vigilant. Parents heard that children with joint injuries could subsequently trigger JIA flare-up, and so parents watched their children very carefully. "One of my bigger fears is that she'll injure something and then that will reactivate the whole process you know, whether it's the same knee or a different joint." Parents were extra vigilant about preventing their child from getting sick due to a compromised immune system or not being able to receive vaccination due to treatment. For example, one parent offered, "So you know, I, I would make a mental note of oh okay it looked like strep throat was going around in his class that time. So we would be extra vigilant about him washing his hands... he, he has this really bad habit of putting his hands in his mouth." Another parent also shared, "I think that's the most difficult part is, okay, are all his [blood] counts good or okay, he's not getting sick, someone at school has a cold, remind the teacher to make sure everybody's using hand sanitizer 'cause you know, it's more difficult for, on his body when he gets sick."

Feeling paranoid about my child getting sick. In addition to being extra vigilant, some parents expressed feeling paranoid about their child getting sick. “Once when he was first on methotrexate, I was really, really paranoid about him being sick all the time...and I'd get kind of frustrated and upset when we had friends that would bring a sick, their sick child, over and then [my child] would get sick and so for quite a while I was really paranoid about everything he was gonna be exposed to.” Parents were not sure whether they needed to wear a mask around the child when they were sick, whether they could take their child to the grocery store, or eat out as a family for fear that people were sick. As another parent stated, “I'd love to take her to Disneyland but after the measles outbreak I don't want to take her anywhere.”

Working out the kinks of medications. Parents described the issues with medication administration. Some parents found it challenging to keep the medication dosage straight. For some, medication dosages changed over time, while others were administered as needed. Some parents found it difficult to administer oral medications to their young child. “Hiding medicine, oh my gosh, such a struggle to even get her to take her medicine.” Another parent shared, “some of the medicines that were prescribed were um like in a liquid form. And it's the volume of the medicine was, that we needed to get in her, you couldn't like hide it in yogurt or hide it in apple sauce, like it was too much, you would taste that, or in milk or whatever. So um so I had to try different medicines and finally we found meloxicam which is less volume and easier to mask and thank goodness we did 'cause it, it was really hard, that we could finally get her to take.” Parents learned to use distraction or games for their child to play while giving medications. Parents who live outside of the city had difficulty obtaining their child’s medication. Keeping the medication at the required cool temperature while traveling was another challenge.

Staying on top to manage JIA. Parents wanted to be on top of their child's illness and health condition, but found it difficult to manage all of the JIA-related medical appointments, including doctors' appointments, physical therapy, and periodic check-ups with blood draws. "Keeping up with all of the things he needs to do, so like making sure he gets his eye appointment every three months and making sure that we see the dentist so that we can like make sure his jaw is all, you know, not affected and, and all that kind of stuff." Another parent said, "I think the biggest thing for us is... making sure that we have that conversation with his, with, with the you know, Dr. [doctor] and you know, where are things at, what do we need to do."

Trying everything I could to help. Parents tried many alternative ways to help their child, such as naturopathic and homeopathic remedies and interventions. "I did try to go gluten free for a very short period of time. I think I only made it, I was strict about it for like a week, um trying to, I don't know, you see so many different theories with this and so I was just trying to do whatever I could possibly do... at the time I was doing all the research and finding you know, theories about different you know, inflammatory foods and then there's casein allergy or sensitivity and gluten allergies and sensitivities and all these different things related." Some parents tried non-pharmacological interventions such as warm bath, heating pads, and blankets to keep the affected joints warm.

Traveling with a young child. Some families lived far away from a children's hospital with a rheumatology clinic. Parents found it challenging to travel with their young child, because of issues such as feeding, long-time sitting in the car, and preparing for a multiple-day trip. "The drive is about um two and a half to three hours and traffic and the pass that's, you know, and especially with the mountains. Sometimes the passes are closed and that can make the trip even way worse and when you're traveling with little kids, it's like ten times more challenging than it

is with just adults driving because there's feeding and using the bathroom and diapering and you know... while two and a half, three hours doesn't seem like a long time, it is for a little kid to sit still and hang out in one little position strapped into a car seat... besides just the average you know, well I mean, stress, whenever you travel and trying to pack and do all those things that you need to do for little kids, thinking ahead, you know, oh you gotta bring their change of clothes and their snacks and their toys and how am I gonna entertain them and, and all that kind of stuff.” Some children were being nursed. As one mother described, “we always nurse for comfort and, and he was going through traumatic thing having to be in his car seat for five and a half hours and then going to the hospital and him not being able to nurse and then um, by the time he was supposed to go into the joint injection, he had just lost it, he was just screaming and there was nothing that could console him because he couldn't nurse.”

Domain 4: Feeling My Child's Pain

Parents had a horrifying experience giving injections to their child and taking them in for painful procedures. They felt there was not much they could do to help their child and they felt like a bad parent who betrayed their child. “Feeling My Child's Pain” consisted of five categories.

Feeling traumatized by giving my child injections. Giving injections to a child was a huge challenge for parents. Sometimes parents had to have a friend or neighbor come to their home to help hold their child down and inject a needle. “Every time you're doing that injection and you're pulling out all the tricks in your hat and you know, your child is not responding to them. You still have to give the injection. So, to hold them down and, and like force it onto them, it's traumatizing.” Most parents had never given injections before, but even for parents who had professional experience giving injections, giving injections to their own children while the child

was screaming and yelling was a horrifying experience. Another parent offered, “giving her something that I actually have to inject her with, it was really scary and I have to say that I cried the first couple times. Because she would cry, I would cry, she'd scream, and it's hard.”

Feeling painful to watch. Children often had to have blood drawn, receive injections into their joints, and experience medication side effects. Parents hurt when their child was hurting, and some even felt they experienced the pain themselves. “[You’re] empathizing with them and feeling their pain. You almost feel it more than they do.” Some parents also described the feeling in situations where their child could not play with others due to JIA, “my heart just breaks watching her have to watch the other kids play and she can't play.”

Knowing there was not much to do for my child. Although parents felt their child’s pain, sometimes they did not know how to help their child. “I was just like, totally bawling at one night, I remember, because the one thing [a medication] that can help make her feel better I cannot even get her to take it down.” For some parents, it was even more difficult because they actually knew that they could not do anything to help in situations such as joint pain, disrupted sleep at night, or medication side effects.

Betraying my child. Parents had the feeling that they were a bad parent when doing things that hurt their child. “I just felt like I was, he looked at me as the bad parent because I was the one doing the shot, you know?” Parents also felt they betrayed their child by taking them for painful procedures. “I was supposed to be his comfort and the one there for him and, and then, them telling me I can't nurse and I'm kinda distracting him while they put this mask on his face and hold it on there while he's screaming... in his mind, um, I could see that he would feel like that, that I had betrayed him or abandoned him because he always turned to me to comfort him and I couldn't comfort him the way that he was used to.”

Blaming myself. Parents felt guilty and thought it was their fault that their child had JIA. “You feel guilt, too, as a parent, that you're not doing enough or even though the doctors are saying that like it's nothing you did to cause it or anything, you still wonder if it, if like, did I feed her too much dairy?” Some parents felt a child’s flare up was their fault. They also blamed themselves for the medication side effects children experienced. “I was angry at myself for allowing it [medication side effects] to go on.” Some parents blamed themselves for not disciplining their child with JIA. “To be super honest, that first year, I probably ruined her because, because I was really...lenient um, discipline wise...like when she's, when she would throw tantrums...Um, because I felt like any sort of jarring was adding to what she already had... So I let that go. Um, which did affect how, um, how the everyday went...which was hard on everybody.”

Domain 5: Feeling Drained from the Whole Process

Parents felt that since their child’s JIA diagnosis, challenging situations continued to happen one after another. Parents felt that JIA was consuming their life and they felt emotionally and physically drained. They took time off work to take care of their child with JIA, but at the same time had to make money to pay for all the medical bills. “Feeling Drained from the Whole Process” consisted of five categories.

Feeling kicked down like a dog. Parents described feeling like they were being kicked down like a dog. "It kind of started with [child] getting diagnosed with JIA and then a lot of things happened since then that have been hard and difficult and not necessarily related to him having JIA but just kind of like, feeling like, kicking a dog when it's down type of thing and then the first blow was, was finding out about [child]'s JIA. And then after that there's just been like, one kick after another, it feels like."

Taking an awful amount of time off work. Parents took time off for JIA-related appointments and caring for their ill child. “I’ve taken off a good deal of work between appointments and then her not feeling well.” In some families one parent was the primary caregiver that took time off work to care for the child while the other parent had to work to support the family; in other families, both parents took lots of time off work trying to be there for the appointments.

Escaping to work. Although parents took lots of time off to care for their ill child, some parents shared the feeling that work was a way for them to “get a little breather.” “The majority of his focus was his full time job you know, during the day time and so he has to kinda disengage from what was going on at home so he can do what he has to do at work. And um so I think in some ways it was a little easier for him than for me, you know. But, 'cause I was in the throws of a disorganized home and a child with you know, a big swollen knee and limping and, and I got to see that you know, pretty much every minute of the day versus he got to kinda escape to work.”

Financial stress. Parents reported the financial burden of JIA, including medical bills, medication costs, and travel expenses. Some parents took extra jobs to supplement their income. “I took on um some extra jobs like work uh from contracts and, and so I mean, it meant less time on the weekends with [child] but it, it helped get us through the hump of um paying the bills that I was getting.”

Consuming my life. Parents felt emotionally and physically exhausted. “It [JIA] kinda consumes your life... until it's in remission, it really consumes your life.” Parents had to take time off work to care for the child, and at the same time they had to work extra hours that cut into their leisure time. “I think I was probably a lot more tired than uh, I didn't have as much energy to, to play with him so we would do less energetic activities and more of uh, um

sedentary activities, if you will.” In some families, a child’s JIA would go into remission for a while, but then became active again. The new flare-up made the parents feel even more tired than the initial diagnosis. “Just last night he [the other parent] actually told me he's probably gonna seek out a therapist for it. Um because we have this new flare up and it's really affecting us again...we're having this emotional stuff happening from the new flare up three weeks ago so it's kinda bringing me back to where we were with the new diagnosis.”

Domain 6: Being Hard on the Entire Household

The JIA not only affected the child, it also impacted the parents, siblings, and the relationships between the family members. Parents felt that every aspect of their lives was affected by JIA and had to change daily routines to deal with it. “Being Hard on the Entire Household” consisted of six categories.

Limiting our life as a family. Because of the symptoms, the child’s activities were restricted. “It's very limiting and it, it affects, I guess it affects other, socially other ways because um we may not go to as many social activities or something... Or we would go to the 4th of July stuff and not stay as long. We would not go out trick or treating as long... She's had to miss out from a lot of stuff, not just, even when she's feeling good but her immune system is down.” JIA also restricted activities the family could do together. “There're things that we could normally do, before that [JIA diagnosis], we can't now... so just, I guess, the things that we used to be able to all do together, is more limited.” Some parents decided to go out less often in order to prevent the sad feelings child and themselves had watching what other children could do and they could not. “Like we held, held back a little bit during that time 'cause for her to go and watch other kids having, being able to have all this fun, um, it's like we would rather stay home, rather than go there and be sad about it, you know.”

Being hard on siblings. In families with more than one child, the JIA was hard on siblings. Siblings could not understand the illness and had a difficult time seeing their sister or brother in pain. “It really hurt his brothers because they felt helpless also because seeing their brother in pain is never fun.” Siblings had hurt feelings because parents paid more attention to or had different standards for their sister or brother with JIA, “I know that sometimes they [siblings] say to me even now, um, why would you let her [child with JIA] get away with that?” Some siblings were also frustrated about less time they spent together as a family. “I do know that they get exasperated cuz it’s a lot of the time that so we have to go to this appointment or we need to take her here, or we have to have another doctor visit, or, you know, those kinds of things.”

Being hard on the marriage. Some parents reported tension between the parents after the JIA diagnosis. Sometimes the two parents disagreed on JIA treatment plans. “During those times [when child had medication side effects], I think I was probably most angry with my husband 'cause then he would say, remember this is, this is what they said to expect. So...which to me wasn't right.” Some parents felt that the other parent did not share the burden to care for their child. “I know that there was resentment towards my husband because I was always the one that got up with [child] when he woke up during the night and always when I got up with him first thing in the morning when he woke up, and there was just building resentment that he didn't do more with, I was the one that was sleep deprived all the time instead of him taking a turn being sleep deprived sometimes taking turns with [child].” Overtime, parents felt that their tempers got shorter with each other, and they argued and/or fought a bit more than normal. These stressful times of elevated tension made the parents felt estranged from each other. Sex life between couples was also affected. “Sometimes it would be her sleeping in our bed and

[husband] would sleep in our guest room. So it definitely disrupts the fact that we can't even sleep together... so it affects the sex life, which affects your relationship.”

Feeling judged. Parents felt that their children were judged by other people because of the illness, “When we go places where we're gonna have to do a lot of walking, he goes in a stroller because that's just what's easiest for him. Um you know, we get a lot of questions, ‘Isn't he kinda big for that’, ‘Shouldn't he be walking.’ I think the most difficult part is kind of the judgment part... It was very frustrating how this goes because um I feel like there's a lot of judgment.” Parents were also judged by other people about how they care for their child. “The other thing [that was challenging] too is explaining it to your friends and family and stuff and then everybody has their own opinion of what you should do... people around you have different ideas and want me to use some hemp oils and all these different things... there's definitely medicinal values but like people around you judging you like or, or feeling like you should be doing this, or you should be doing that. And so that's hard because you have to deal with the, the social aspect of people thinking you should be doing something different.” Some parents decided not to disclose their child’s JIA diagnosis to avoid the judgment.

Losing sleep was hard on the entire family. Parents described that their child just could not seem to get comfortable at night especially after having a flare-up or being in pain during the day. The child’s disrupted sleep affected the sleep in the whole family. “The biggest thing was as far as affecting the whole family was the sleep stuff... the fact that she was being woken up you know, more than five times a night, um and that affects us. So basically almost you know, more than half of that first year, we had horrible sleep.” One parent reported how the sleep problem affected her, “It affected me hugely. It, it still continues to affect me. He has nights, two nights ago when pretty much all night long he's waking up, he's saying stuff in his sleep, he's moaning

in his sleep and it wakes me up all night. I am, it makes me really grumpy. And... I've totally come to understand why, why sleep deprivation is used as a torture tactic and it has been used as a torture tactic because it is just, you get to this, this state where you're so tired and you just wanna sleep and, and not have it disturbed. And so from the time [of the diagnosis], pretty much, I feel like I was constantly sleep deprived for over, for almost three years consecutive of my life.”

Member Check

Five parents participated in the member check phone interviews. The interview duration ranged from 7 to 19 minutes. Data were analyzed based on the four member check questions. Parents' overall responses to the summary were: it was very clear and accurate; it encompassed many daily things parents thought about; and it was nice to know that other parents had the same experience. For example, one parent shared, “You're kinda spot on to everything that we dealt with in the beginning... I think it's excellent and anything that I thought or would say is in here.” Parents highlighted the areas that they thought were especially good, “the part about not knowing...not understanding the illness um a lot of uncertainties about the child's future. I still feel that way. And not sure about what decisions to make about child's treatments. I still feel that way...” Parents mentioned some parts that did not fit their experience. For example, when parents did not have more than one child, the category about being hard on the siblings did not fit. Another parent mentioned the category of being hard on the marriage did not fit because she was not married.

When asked to share any additional things about this summary, parents said, “I'm just really glad that you did this and just reading it, for me, has helped a lot and, and knowing that um that it wasn't just me that went through this experience but knowing that other parents who have gone through the experience with their children pretty much sound like they went through the

exact same things as I did.” Parents thought that the summary of study results would also be helpful to other people, “I think it, it's just wonderful. Like I think that um it's, it's a lot of data that, that um you've collected to give a picture of the effects of, you know, the diagnosis and I would, I think even physicians that treat, treat JIA or arthritis generally would benefit from having this perspective, you know? Um and the nurses and, and the people who are involved in the care team...” Another parent said, “I really was pleased with it and I actually had friends read it just because I think I don't vocalize things that are in my head and my worries enough to people that are around us...I think it helps our family and my mother and his other caregivers really understand where I was coming from when I voiced those worries about what's going on with him. So it was a nice resource to have for other caregivers to read.”

Discussion

Despite the advanced medical care for young children with JIA, our findings show parents encountered multiple challenges in trying to help their young child during the new diagnosis period. Parents struggled with the unknown, searched for resources, witnessed their child's suffering without knowing how to help, and tried every possible way to stay on top of the child's illness and treatment. The prolonged struggle substantially impacted the emotional and physical health of the parents and the entire household. The findings from this study expand our understanding about the impact of a young child's new JIA diagnosis on that child's family. Specific areas included the lack of information parents received about the illness, support, and available resources; parents' struggles to help their child; the impact of JIA on the parents and the impact of JIA on the entire household. Each of the four areas of impact is discussed below.

Parents were overwhelmed by the amount of scattered and unclear information given to them about JIA. The lack of resources to meet the parents' needs led to parents searching outside of the healthcare teams, but these searches were not successful. This finding is consistent with Barlow and colleagues (1998) who also found a lack of information and inadequate support for parents to care for their school-age children or adolescents with JIA (Barlow et al., 1998). Findings show that the resources for parents of caring for a child with JIA are limited and inadequate. The lack of information and resources available for parents during the acute phase of the initial diagnosis period is evident. Information and resource that meets parents' specific needs are needed to assist parents in managing the care during the initial diagnosis period.

Unique to this study is that our sample focused on young children who are not always able to articulate and describe their feelings in comparison to school-age children and adolescents. At times, parents had to guess at their child's feelings in order to better manage their care. Parents did not have external measuring tools to judge whether what was happening to their child was part of the expected disease course, improvement or worsening of JIA, or side effects from medications. These findings contradict other studies reporting that parents caring for children with JIA considered themselves as well-informed, and as developing expertise in managing their child's illness over time (Jerrett, 1994; Thon & Ullrich, 2009). These differences may be attributed to the fact that parents in the prior studies had cared for their child with JIA for years after the initial diagnosis, whereas the current study focused on parents of children with a new JIA diagnosis. It is plausible that some of the struggles such as the difficulty in discerning between arthritis-related and other causes of child's behavior decreased over time.

Parents tried everything they could to manage the illness and its effects, but they encountered multiple challenges. Even when the disease-related symptoms were under control,

JIA treatments made their child sick in other ways. For example, administering methotrexate alleviated pain, but some children experienced severe fatigue, nausea and vomiting, as well as compromised immunity. This finding is consistent with previous studies that also report children with JIA experienced difficulty and side effects in taking methotrexate (Bechard et al., 2014; Mulligan et al., 2013; Mulligan, Wedderburn, & Newman, 2015). In addition to the side effects of methotrexate, our findings also show that parents were affected. Parents struggled when administering injectable medications due to the pain, discomfort, and side effects experienced by their children. Some parents blamed themselves for their child's suffering. Previous studies in JIA show that parents experienced emotional distress that was above the clinical cutoff score for psychological symptoms (Bruns, Hilario, Jennings, Silva, & Natour, 2008; Gerhardt et al., 2003). Specific programs that provide appropriate educational and psychological support need to be developed and evaluated to improve parents' and children's experience to reduce the psychological impacts of certain medication and alleviate the caregiving burden.

One of the underlying assumptions that guided this study was the stress, coping, and adaptation process when a stressor – the diagnosis of JIA in the child – occurred in the family. A linear occurrence of stress, coping, and subsequent adaptation to the new JIA diagnosis were rarely reported with the challenges parents shared. Rather most of the challenges persisted and still affected the family long after the first year of diagnosis. The concept of stress pile-up in the McCubbin and Patterson's framework of Family Stress Process is relevant to these results (H. I. McCubbin & Patterson, 1983). It argues that families rarely are dealing with a single stressor, but rather, experience a pile-up of stressors and strains. The stress pile-up particularly occurs after a major stressor - in this particular case, the diagnosis of a new chronic condition in the child. Parents described their feeling of “kicking a dog when it's down...and then the first blow

was...finding out about [child]'s JIA.” Many things happened after the diagnosis, and prior strains were exacerbated. Families became aware of these prior unresolved hardships as demands in and of themselves. McCubbin and Patterson argue that these strains contributed to the pile-up of demands families must contend with in an already stressful situation. These stressors feed into each other and become multiplicative, resulting in a family crisis (H. I. McCubbin & Patterson, 1983).

The adaptation phase described in the Resiliency Model was not observed in the current study, which may be attributed to the timing of the interview (during the initial acute phase of a new chronic condition onset). Parents described most challenges continuing after the first year of diagnosis. Previous studies of parents caring for children years after JIA diagnosis also reported substantial burden of care and sense of strain (Barlow et al., 1998; Gomez-Ramirez et al., 2016). However, Jerrett described that parents were able to eventually develop expertise in managing their child’s illness; however, the time between the interview and JIA diagnosis was not reported (Jerrett, 1994). Conflicting results were also found in previous quantitative studies of adaptation in families of children with JIA, with some reporting lower adaptation and others reporting similar levels of adaptation in comparison to families of children without JIA (Gerhardt et al., 2003; Huygen et al., 2000). Whether parents adapt to the child’s chronic condition may depend on many factors including previous stressors in the family, the resource and support available to the family, the severity of child’s illness, and the time since diagnosis.

The application of Family Systems Theory to examine the impact of JIA on the family and its members in the current study revealed substantial evidence that the new diagnosis of JIA profoundly impacted different aspects of the lives of the entire household. Parental feelings of being judged and the negative impact of JIA on siblings, the marriage, and relationships between

family members were unique findings in the current study. Prior studies of other chronic condition (e.g., cancer, diabetes) in one family member also report the profound impact of the illness on the family system (Lewis, 1993; Lewis, Woods, Hough, & Bensley, 1989; Zahlis & Lewis, 2010). Healthcare providers need to assess the impact of JIA not only in the ill child and the parents, but also the entire household. The family as whole and individual family members could benefit from tailored resource and support programs.

Limitations

There are several limitations in the study. First, the data are limited to parents' recall. Although parents were interviewed within a year after their child's new JIA diagnosis and they continued to struggle. Second, the cross-sectional design limits the ability to understand how parents cope and manage the illness-related challenges overtime. Third, the nature of the interview questions developed may have resulted in parents identifying challenges. Fourth, the parents reported difficulties in sleeping for both parents and children; this may be attributed to the prior study about sleep in these young children with JIA and their parents, although no direct questions were asked about sleep.

Implications and Conclusion

Three tentative clinical implications are suggested from the study results. First, healthcare providers need to assess the particular needs of an ill child and parents, and the impact of the illness on the physical and psychosocial health in the entire household so that proper resources can be provided. The second implication is that parents' connections with other families in similar situations needs to be facilitated. Parents in the current study expressed feelings of isolation and a strong desire to share their experience with other families of children with the same diagnosis at a similar young age. Third, parents encountered multiple ongoing challenges

after their child's JIA diagnosis. Not all challenges may be due to the child's illness, but may have been escalated with stress piling up in their lives. Often family is considered the context of care when a child is diagnosed with a chronic condition, rather than the unit of care. We may be more interested in helping the family members help the child than helping the family members themselves (Shands, Lewis, Sinsheimer, & Cochrane, 2006). Healthcare providers need to re-evaluate their commitment to family-centered care, and consider various challenges families encounter when providing care, not merely those that directly relate to the child's illness.

The current study findings are the first to highlight the day-to-day challenges parents encountered when caring for their young child during the initial JIA diagnosis period. The new knowledge generated from this study may help healthcare providers understand parents' stress and illness-related challenges when their young child is newly diagnosed with JIA. Future studies may benefit from studying a larger number of families, and at the initial diagnosis period and following up periodically for years after to examine the patterns of stress, coping, and adaptation that occur after the diagnosis of a chronic condition in the child. The results will inform the development of family-centered intervention trials to assist families and improve child, parent, and family outcomes in the future.

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Table 4.1 *Qualitative Studies Describing Parents' Experience In Caring for A Child with JIA*

Study	Sample	Child Age	Time Since JIA Diagnosis	Design	Interview Questions/Topics
Barlow et al. (1998)	5 groups: 6 health professionals, 5 with mild JIA, 6 parents, 5 with severe JIA, 7 parents	8-15 years	-	Focus group interviews	Symptoms of JIA, the day-to-day impact of JIA, strategies used to overcome problems associated with JIA, and the impact of JIA on other people.
Jerrett (1994)	19 parents from 10 JIA families	-	-	Phenomenological perspective with individual interviews	Parents' perspective of their experience in managing their child's illness.
McNeil l (2004)	22 fathers with JIA children	Average 8.7 years	Average 5.6 years Newly dx (<1 year) excluded	Grounded theory with semi-structured interviews	Experience of caring for their child, approach to parenting, parenting relationship with their partner, potential needs for emotional and social support, relationship with the health care team, advice for fathers of newly diagnosed children.
Gomez-Ramirez et al. (2016)	15 experienced parents (9 months - 14 years of care) and 8 novice parents (<6 months since diagnosis)	2-16 years	mixed	Focus groups and written reports of reciprocal interviews (parents interview each other)	(1) What are the predominant emotional experiences of parents caring for a child with JIA? (2) How are these parental emotions associated with different phases or events during the disease?
Stinson et al. (2012)	41 children with JIA and 48 parents	8-11 years, Average 9.7	Average 4.8 years	Descriptive exploratory interviews (dyad and focus groups)	(1) Family perspectives of how they managed JIA; (2) Information needs of children with JIA and their parents.

Table 4.2 *Interview Questions*

1. During the first year after your child was diagnosed with JIA, what were some challenging situations you and your family experienced?
2. How did the situation (or these situations) affect you/your partner/your child with JIA/your other child(ren)/the relationships between family members?
3. What are the things that you have been doing to manage this challenging situation (or these challenging situations)?
4. How is it working for you and your family?
5. What resources have you used to manage this situation (or these challenging situations)?

Table 4.3 *Inductive Content Analysis Steps*

Steps	Details
1. Unitizing data	Data were unitized based on complete ideas, not complete sentences. A complete idea was defined as a verbal expression that included both a verb and noun, and the idea could be explicit or implicit. Thus, a compound sentence with more than one idea was analyzed into its component parts and each part was coded as one data unit.
2. Open coding and identification of initial categories	All unitized data identified in Step 1 were open coded; none was discarded. Open coding required the analyst to put aside preconceived notions about what was expected to be found in the research, be open to all potentials and possibilities contained within the data, and let the data and its interpretation guide the analysis. Open coding involved analyzing, comparing, and categorizing the data units. First, open coding happened within each interview question. Data units were reviewed and organized into categories of units based on one or more common properties. Then, all data units across all interview questions were open coded together to develop the initial set of categories. No unit of data appeared under more than one category.
3. Naming and defining categories	Categories were defined with critical attributes according to the shared properties of the units in each category. Category names were chosen from the parents' own words. Constant comparisons were utilized to ensure mutual exclusiveness of the categories. Each unit was compared with each category to maximize the fit of the unit with the category. If the unit did not fit a category, a new category was created. All categories were compared with one another to ensure non-overlapping.
4. Identification of higher order domains, naming and defining domains	Categories were organized into higher order groups called "domains." Categories within a domain represent different dimensions of the domain. Domains were defined according to the shared properties of the categories within each domain. Domain names were chosen to best capture the manifest meaning of the categories, which were labeled with the study participants' own words. Constant comparisons were also utilized to ensure mutual exclusiveness of the domains. Defining and refining categories and domains were performed continuously during the analysis period.
5. Identification of a core construct	Domains and categories were reviewed and a core construct that organized the domains and categories was identified, with the goal that the core construct further organized the domains and described the social processes in which the parents engaged caring for their young child newly diagnosed with JIA.

Table 4.4 *Struggling in the Dark to Help My Child: Domains and Categories of Parents' Experiences*

Domain 1: Not Knowing
1.1 Not really understanding
1.2 Not able to predict the future
1.3 Not knowing what to do
1.4 Unsettling to have no closure on any aspect
1.5 Hard to discern between arthritis-related and other causes
1.6 Feeling blindsided and overwhelmed
1.7 Scared of not knowing

Domain 2: Trying to Reach Out in the Dark
2.1 Feeling totally alone
2.2 Hearing stories here and there
2.3 Having no resources to draw on
2.4 Getting scary stories from the Internet

Domain 3: Working Out the Kinks to Stay on Top to Manage
4.1 Battling with JIA symptoms
4.2 Seeing my child getting sick all the time from treatment
4.3 Being extra vigilant
4.4 Feeling paranoid about my child getting sick
4.5 Working out the kinks of medications
4.6 Staying on top to manage JIA
4.7 Trying everything I could to help
4.8 Traveling with a young child

Domain 4: Feeling My Child's Pain
3.1 Feeling traumatized by giving my child injections
3.2 Feeling painful to watch
3.3 Knowing there was not much to do for my child
3.4 Betraying my child
3.5 Blaming myself

Domain 5: Feeling Drained from the Whole Process
5.1 Feeling kicked down like a dog
5.2 Taking an awful amount of time off work
5.3 Escaping to work
5.4 Financial stress
5.5 Consuming my life

Domain 6: Being Hard on the Entire Household
6.1 Limiting our life as a family
6.2 Being hard on siblings
6.3 Being hard on the marriage
6.4 Feeling judged
6.5 Losing sleep was hard on the entire family

VITA

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