Interdisciplinary Behavioral Rehabilitation of Pediatric Pain-Associated Disability: Retrospective Review of an Inpatient Treatment Protocol

Cynthia S. Maynard, PsyD, Adrianna Amari, PhD, Beth Wieczorek, RN CRNP, James R. Christensen, MD, and Keith J. Slifer, PhD
The Kennedy Krieger Institute and Johns Hopkins University School of Medicine

Objective A biopsychosocial model was used to treat pain-associated disability in children and adolescents. We assessed the clinical outcomes of children and adolescents (8–21 years of age) with pain-associated disability who were treated in an interdisciplinary inpatient rehabilitation program which included physical, occupational, and recreational therapy, medicine, nursing, pediatric psychology, neuropsychology, psychiatry, social work, and education. Psychological treatment emphasized cognitive-behavioral intervention for pain and anxiety management, and behavioral shaping to increase functioning. **Methods** We conducted a retrospective chart review of 41 consecutive patients. School attendance, sleep, and medication usage were assessed at admission and discharge; functional disability and physical mobility were assessed at admission, discharge, and 3-month follow-up. **Results** As a group, significant improvements were observed in school status, sleep, functional ability, physical mobility, and medication usage. **Conclusion** Findings support the efficacy of an inpatient interdisciplinary behavioral rehabilitation approach to the treatment of pain-associated disability in pediatric patients.

Key words pediatric; chronic pain; functional disability; interdisciplinary rehabilitation, pain-associated disability.

Introduction

Epidemiological studies have determined that more than 30% of children and adolescents experience chronic or recurrent pain significant enough to cause suffering, school avoidance, limited participation in physical and social activities, and disruption of sleep and appetite (Palermo, 2000; Perquin et al., 2000). Some of these individuals are very difficult to treat and develop chronic functional disability. In many medical settings, chronic or recurrent pain and disability in the absence of an identified physical cause is attributed to psychological factors. Interventions that are based on psychological conceptualizations are often poorly received by families and typically do not lead to significant improvement. Families often seek the opinions of numerous healthcare providers in search of medical etiology and treatment of pain. As a result, specialists in the field of chronic pain treatment and research

have adopted a biopsychosocial conceptualization of chronic pain. This has called for a conceptual shift away from attempting to differentiate physical from mental pain and acknowledges the multidimensional nature of pain in which biological, psychological, social, and environmental variables interact in the development, maintenance, and subjective experience of pain and disability (Bursch, Joseph & Zeltzer, 2003; Zeltzer, Bursch, & Walco, 1998; Zeltzer, Tsao, Bursch, & Myers, 2006). The biopsychosocial model has been recommended with patients who meet the criteria for Pain-Associated Disability Syndrome (PADS). Severe disability that continues for two months or greater has been adopted as one of the criteria used to define PADS (Bursch et al., 2003). PADS has been used to describe a chronic pain condition with frequent and severe difficulties in functioning, regardless of the location or cause of the pain (Bursch, Walco, & Zeltzer, 1998;

Zeltzer et al., 1998). PADS is based on the observation that the actual tissue damage sustained by a patient, the perceived severity of the condition, and the degree of disability exhibited are often widely discrepant. PADS occurs with many variations in etiology and source of the original pain. It may begin with illness, injury, viral infection, or developmental or psychological challenge. Oftentimes the original trigger may not be identified. There also is wide variation in the time course of the development of the disability. Bursch and colleagues hypothesized that PADS develops when a physical trauma, illness, or other life circumstance becomes overwhelming to a vulnerable child who has poor coping skills and cannot regulate his/her own distress. Furthermore, the physical symptoms may allow the child to avoid or escape the stressor, associated discomfort, and negative emotions. Consequently, the child may fail to develop positive coping strategies and a behavior pattern in which avoidance of stressful or painful stimulation occurs along with increasing dependence on others, social withdrawal, avoidance of school and physical activity, and progression of functional disability.

Given the multiplicity of possible contributing factors, Bursch and colleagues (Bursch et al., 1998, 2003; Hyman et al., 2002) have advocated for a multidisciplinary rehabilitation intervention that addresses underlying pain mechanisms (to the extent possible), treats specific symptoms (anxiety, depression, avoidance behavior, diminished muscle strength/range of motion, academic deficits), and teaches active coping skills to the child and family. Emphasis is placed on reinforcing more independent child functioning in activities of daily living, academics, social interaction and physical exercise. Psychological interventions are provided in the context of physical rehabilitation in order to strengthen child and family coping skills and to help modify interaction patterns that may be maintaining pain and illness behavior.

The child and family are encouraged to move their focus away from total elimination of pain as a pre-condition for recovery. Adaptive functioning instead of pain severity is used to measure progress.

An inpatient interdisciplinary rehabilitation hospital setting, in which patients are seen daily by professionals from several disciplines across multiple situations, is particularly well suited for implementing a systematic behavioral intervention to shape functional behavior and reinforce its generalization (Amari, Slifer, Sevier, Spezio & Tucker, 1998; Palermo & Scher, 2001). To date, there are limited published data on inpatient interdisciplinary rehabilitation programs that have used interventions comparable to those described in this study. In one case

example, Palermo and Scher (2001) describe the improvements in physical and psychosocial functioning of an adolescent with a severe pain disorder following an inpatient rehabilitation admission. Eccleston, Malleson, Clinch, Connell and Sourbut (2003) investigated 57 adolescents with chronic pain and their accompanying caregivers who were enrolled in a 3-week interdisciplinary residential program that emphasized cognitive-behavioral and physical therapy interventions. Results of their study demonstrated that the intervention was effective in reducing distress and improving functioning and school attendance.

There are a few programs located in children's hospitals and other centers nationally and internationally (e.g., Cleveland Clinic; http://my.clevelandclinic.org/childrens_hospital/departments/rehabilitation_services/programs/pediatric_pain_rehab.aspx) that use the interdisciplinary rehabilitation model to treat patients with chronic pain (Dobe, Damschen, Reiffer-Wiesel, Sauer, & Zemikow, 2006; Eccleston, Cornell, & Carmichael, 2006; Lee et al., 2002; Sherry, Wallace, Kelley, Kidder & Sapp, 1999). These programs appear to vary based upon type of setting, length of stay, and various disciplines involved, and it appears that programs may differ regarding the type and level of psychological interventions used. To our knowledge the outcomes of patients in these programs have not been described.

The goal of this study was to assess the outcomes of implementing an inpatient interdisciplinary behavioral rehabilitation protocol to treat children and adolescents with pain-associated disability and distress. The conceptualization and the intervention approach are based, in large part, on the prior work of Zeltzer, Walco, Bursch, Tsao and colleagues as reviewed above (Zeltzer et al., 2006). We conducted a retrospective chart review to obtain clinical outcome data on a sample of children and adolescents hospitalized for treatment of PADS. We hypothesized that the group would exhibit significant improvement across measures of functioning after completing the inpatient protocol as compared to pre-admission functioning. The clinical sample and the intervention protocol are described in detail, and the clinical results are presented in order to contribute empirical support for the interdisciplinary behavioral rehabilitation approach to treating PADS.

Method
Rehabilitation Setting

The inpatient setting is a 33-bed neurorehabilitation unit that specializes in acute medical and intensive

interdisciplinary rehabilitative therapies. Patients can be admitted to the inpatient program in several different ways. Patients can come through the Multidisciplinary Outpatient Pain Management Clinic and be screened for inpatient candidacy. Patients can also be referred from medical specialists (both locally and out-of-state) or can also be transferred directly from local medical institutions. The comprehensive treatment team for the pain program includes physical medicine and rehabilitation, occupational and physical therapy, child life/therapeutic recreation, nursing, education, nutrition, social work, pediatric psychology, neuropsychology, and psychiatry. At times, diagnostic consultations with other medical departments (i.e., neurology, anesthesiology, rheumatology, etc.) were warranted for some patients in order to assess, confirm or rule-out various medical etiologies (i.e., neurally mediated hypotension, Celiac Disease, Lyme's Disease, etc.).

Procedures and Participants

Patients were determined to be appropriate for the inpatient pain program if they were 6–21 years old, had pain and functional disability, and had an adult caregiver willing to participate in treatment. Formal functional disability measures were not used during clinical screening for inpatient admission; rather, patients had to demonstrate significant pain complaints with functional disability in the above domains (i.e., social, educational, and physical) to qualify for admission. Inpatient admission was considered based on whether the patient had participated in outpatient services (e.g., physical therapy or counseling) without significant improvement, per patient/family report and review of preadmission records. Additionally, individuals who had significant psychiatric needs thought to require more intensive psychiatric services (e.g., those with active suicidal ideation) were not admitted to the inpatient pain program and referrals were provided as appropriate.

This study consisted of a retrospective chart review. Forty-nine patients were admitted for inpatient treatment of PADS in the preceding 7 years. With approval from the institutional review board, we conducted a retrospective chart review to determine which patients met criteria for PADS (and thus, inclusion in the current report) and also for the data presented in the report. We reviewed preadmission summaries, notes from therapist interviews with child and family members, scores from standardized assessments of functional disability, clinical questionnaires regarding coping strategies, inpatient evaluation and discharge summary reports, written behavioral protocols (for staff and/or family), and data sheets from direct behavioral observations. After reviewing these records,

41 patients met the criteria for PADS. Inclusion criteria for this study included chronic or recurrent pain and functional disability without relief from medications or outpatient therapy. Functional disability was defined by severe impairment in school attendance, participation in social activities, activities of daily living, and/or physical mobility. An adult caregiver had to be willing to participate in rehabilitation treatment. Exclusion criteria for this study consisted of individuals who had significant psychiatric needs which required more intensive psychiatric services, in addition to individuals and parents who were not in agreement with and who were unwilling to participate in the rehabilitation approach because they were seeking purely medical rather than rehabilitative services in order to ''cure'' the pain. Data obtained from reviews were de-identified and stored separately from the original clinical files.

Dependent Measures

School Attendance. School status was assessed by the pediatric psychology therapist, neuropsychologist, and/or the special educator who interviewed the parents and/or school staff. School attendance was categorized in the following manner: No school (not attending regular school or receiving any formal educational services), home and hospital teaching (receiving formal educational services in the home or hospital), part-time (attending regular school for less than the full scheduled school day), and full-time (attending regular school for the entire scheduled school day). If patients were admitted during the summer time when school is typically not in session, an anecdotal history was taken regarding school attendance and functioning during the previous school year.

Sleep. Presence of sleep problems was defined as any disruption to a developmentally appropriate sleep/wake cycle, including late onset of sleep, excessive day sleep, and frequent night-awakenings. Mindell and Owens (2003) published age-specific norms for sleep pattern and duration, which were used to evaluate the developmental appropriateness of the patient's sleep. Information on sleep was obtained by parent and child report and behavioral observations. Data on sleep disruption were classified categorically as present or not, which is consistent with the method used by Hyman et al. (2002).

Functional Status. Functional status was assessed using the Functional Independence Measure for Children, or WeeFIM® (Guide for the Uniform Data Set for Medical Rehabilitation for Children, 1998), a measure commonly utilized in pediatric rehabilitation settings nationwide, and with growing application internationally

(Wong, Wong, & Wong, 2002). The WeeFIM is a comprehensive 18-item, seven-level ordinal scale instrument designed to measure a child's needs and changes in performance in the main functional domains of mobility, self-care, and cognition. Total scores range from 18 (dependent in all areas) to 126 (complete independence across all items). The WeeFIM is well validated and highly reliable, as studied in children with and without disabilities (Msall et al., 1994; Ottenbacher, Taylor, Msall, Braun, & Granger, 1996; Ottenbacher et al., 1997; Sperle, Ottenbacher, Braun, Lane, & Nochajski, 1997).

Clinical significance was determined based upon baseline and discharge WeeFIM scores. For instance, a patient with a WeeFIM score of 18 would have significant difficulties with self-care (i.e., assistance with grooming, eating, bathing, toileting, and dressing), cognition (i.e., problem-solving, memory, comprehension, expression, and social interaction) and mobility (i.e., transfers with shower/tub, toilet, and chair/wheelchair, walking, and use of stairs). At discharge, a patient with a WeeFIM total score of 126 would require less supervision and would be more independent in these areas.

The WeeFIM can be scored by interview with a familiar caretaker or direct observation of a child's performance of a task to criterion standards. WeeFIM data presented here were systematically collected throughout each patient's inpatient admission by trained rehabilitation staff (e.g., nursing, occupational and physical therapists) based on direct observation, and recorded in a weekly interdisciplinary rounds format by trained staff.

Physical Mobility

The WeeFIM mobility subscale was examined separately. Subscale scores range from 5 (dependent in all areas) to 35 (complete independence). For instance, a patient who receives a score of 5 would require total assistance for transfers (both in and out of a wheelchair, toilet and tub/shower). They would also require total assistance for stairs and walking. In comparison, a patient who receives a score of 35 would demonstrate autonomy with these tasks. It is routine clinical and research practice to examine and report WeeFIM total scores as well as relevant separate domain subscores (Grilli et al., 2006; Majnemer et al., 2008; Prosser, 2007). Because the majority of patients were not ambulating or using limbs normally at admission, this subscale was thought to be particularly relevant.

Medication Usage. The total number of medications prescribed and taken prior to admission and at discharge was calculated.

Utilization of Coping Skills. Patient's use of cognitive behavioral coping skills was tracked throughout admission on a daily schedule data sheet. A daily schedule was utilized by all therapists and staff throughout the day to record observed functional gains in scheduled therapies, in generalized therapeutic activities with child life and therapeutic recreational specialists, in various settings and on the inpatient unit. Staff was instructed to provide the participants with verbal praise as they observed and recorded target positive behaviors. The daily schedule consisted of the following domains: time of day broken down into 30-min increments, therapy activity, participation and use of coping strategies (coded using a three-point Likert scale), tasks demands performed in each therapy session, and therapist's comments on patient's progress.

Inter-Coder Agreement

Except for the WeeFIM, which has published reliability studies, all of the outcome measures were coded from the medical charts and a second coder independently coded 20% of the cases selected at random from the total 41 cases. For sleep disturbances and school status exact agreement percentages were 100 and 87.5, respectively. For number of medications prescribed/taken and number of coping strategies employed throughout the whole admission, an agreement was scored if the primary and reliability coders were within plus or minus 1 point. Using this method, the agreement percentage was 87.5 for both medication and coping scores.

Inpatient Interdisciplinary Rehabilitation

Baseline. Detailed information about pre-admission history was obtained from review of previous outpatient and inpatient medical reports, consultation with medical providers, and from family and child/adolescent interviews. Once admitted to the inpatient program, patients received several days of baseline evaluations of functioning by the interdisciplinary staff.

Comprehensive Mental Health Assessment. Patient and family psychosocial stressors, strengths, coping styles, patient personality traits, and cognitive abilities were assessed by pediatric psychology, neuropsychology and social work, as well as psychiatry as needed. Assessments consisted of structured and semi-structured interviews used to obtain psychosocial and family history.

Cognitive-Behavioral Pain Management and Coping Skills Training. All patients met with a pediatric psychology therapist and received individualized cognitive-behavioral therapy approximately two to three times a week to develop active coping and pain management strategies such as distraction, focused breathing, progressive

muscle relaxation, visualization, guided imagery, journal writing and biofeedback. Preferred coping and pain management strategies were identified for each patient and strengthened through verbal instruction, modeling, behavioral rehearsal, and positive reinforcement. Additionally, patients were provided individual therapy to identify and address psychosocial stressors, and to learn to differentiate psychological distress (e.g., school anxiety) from pain or other somatic symptoms.

Differential Reinforcement/Shaping. In conjunction with the medical team and rehabilitation therapists, pediatric psychology therapists developed and implemented a structured behavioral program to differentially reinforce and shape functional behavior for each patient. Behavioral shaping strategies were implemented individually based upon the patients' own pace and tolerance of demands. A daily schedule data sheet was utilized by therapists and staff to record observed functional gains and use of coping strategies in various settings throughout the day (scheduled therapies and in generalized therapeutic activities with child life/therapeutic recreation and on the inpatient nursing unit). The staff provided the patients with verbal praise as they observed and recorded target positive behaviors. Aside from structured assessment of pain by nursing and medical staff for medication assessment, and prompting of coping strategies in response to pain complaints, the staff was instructed to provide minimal social attention to pain behavior. Staff closely monitored each patient's tolerance for therapy demands, and demands and goals were progressed/modified accordingly on a weekly, daily, and within-session basis. At the end of each therapy day, a pediatric psychology therapist reviewed each patient's daily schedule data sheet, and provided verbal and tangible reinforcement for participation in daily therapies and particularly for any functional gains and coping skills used.

Systematic Desensitization. Within-session goals were broken down as needed into the smallest steps necessary to enhance the patient's tolerance of and success with increasing physical/environmental demands. Pediatric psychology therapists worked approximately two to three times a week with physical and occupational therapists to identify appropriate steps and to coach the patient in relaxation or distraction strategies as they progressed through a desensitization hierarchy.

Additional Therapeutic Intervention. Throughout the inpatient stay, each patient received appropriate pain medication and/or underwent medication weans based on routine pain assessment by nursing staff, physical medicine, and pediatric anesthesiology consultation. When they became available in this setting, complementary and alternative medicine methods (i.e., acupuncture, massage, energy therapy) were offered to four patients and families, and utilized by four patients. Finally, sleep hygiene was addressed via application of relaxation strategies at bedtime as planned on the schedule.

Parent Training. Pediatric psychology staff provided ongoing support and education for family members. Family members were taught how to differentially reinforce their child's coping and participation while refraining from attending to pain and illness behavior. The interdisciplinary staff also modeled and reinforced positive interactions. Social workers monitored family caregivers' coping, and reinforced behavioral recommendations that allowed children to experience developmentally appropriate independence. Some caregivers were encouraged to view their child's participation and positive coping behavior during therapies and activities from behind a one-way mirror prior to direct integration into sessions. This intervention was employed based upon the clinical judgment of the pediatric psychology therapist and social worker who had daily interactions with the caregivers' progress and role in providing appropriate reinforcement of healthy behavior.

Generalization/Discharge Planning. Families were assisted in developing a plan for a structured but gradual reintegration of the child into more developmentally typical home and school routines. The social worker and pediatric psychology staff met with the child/adolescent and caregivers to plan a daily schedule to be implemented upon discharge and return to home and school based upon interdisciplinary team recommendations. The daily schedule included activities of daily self-care, school attendance, homework, physical exercise, and social activities with family and peers. Discharge planning also included arrangement for continued outpatient services as needed with psychology and psychiatry (for medication management), as well as physical and/or occupational therapy. Members of the interdisciplinary team, including special educators, assisted in planning for school re-entry and school accommodations as needed (e.g., via school contact, development of individualized education plans). Therapeutic recreation specialists conducted community outings to support generalization of coping strategies to developmentally appropriate leisure and social activities.

Follow-Up. After discharge, follow-up WeeFIM scores were obtained for a portion of patients 19 (51%) who were available by telephone interviews which were conducted as part of the program's continuum of care. Patients that were local attended a follow-up outpatient clinic more easily.

Design and Analysis

This study employed a single group, repeated measures (admission, discharge) design. Data obtained during the retrospective chart review were taken from information and measures completed upon the patient's presentation at admission to the rehabilitation unit and again at discharge. For available cases ($n = 19$), functional data were obtained at three months post-discharge. Pre-treatment to post-treatment and pre-treatment to follow-up comparisons were analyzed using t-tests for two related samples for ratio scaled measures, and the nonparametric equivalent, Wilcoxon signed-rank test for related samples, for categorical measures. An alpha value of $p < .05$ was set as the a priori criterion for statistical significance. All statistical analyses for this study were conducted using SPSS version 15.0.

Results

Demographic and Clinical Characteristics

Forty-nine patients were admitted to the inpatient rehabilitation unit for the pain management program and data from 41 patients were included from the initial record reviews. Four patients did not meet inclusion criteria for this chart review (i.e., did not have PADS), and four others met exclusion criteria (i.e., presented with severe psychiatric disorders requiring intensive psychiatric intervention, or were unwilling/unable to work within the rehabilitation model). These eight patients were originally identified as having been admitted for the comprehensive pain management program; however, their clinical courses were modified and did not follow the program described above. Four out of the eight patients had severe psychiatric symptoms (e.g., active suicidal ideation) and the other four patients were admitted for acute rehabilitation and intensive pain management secondary to their medical conditions (i.e., acute burns and juvenile rheumatoid arthritis with acute medical complications).

For the 41 included here, a wide range of primary medical and mental health diagnoses were represented. Diagnoses included complex regional pain syndrome in 19 (46%) patients, headaches in nine (22%) patients, abdominal pain in eight (20%) patients, functional gait disorder and traumatic brain injuries in four (10%) patients, post-concussive syndrome, developmental delays, and Ehlers-Danlos syndrome in three (7.3%) patients. Twenty-five (61%) patients also had other primary and/or co-morbid medical diagnoses. Mental health diagnoses included anxiety in 17 (41%) patients, depression in 14 (34%) patients, somatoform disorders in nine (22%) patients, and cluster B traits in three (7.3%) patients.

Table I. Participant Demographic and Clinical Characteristics

Characteristics	n (%)	Range	Mean (SD)
Age (years)		8–21	13.8 (2.79)
Gender			
Male	11 (26.8)		
Female	30 (73.2)		
Ethnicity			
White	35 (85.4)		
Black	5 (12.2)		
Hispanic	1 (2.4)		
Number of visits to healthcare specialists prior to admission			10.25 (9.94)
Number of previous interventions prior to admission			8 (8.50)
Number of medications prior to admission			9 (9.16)
Length of days from onset to admission		13–2096	613 (597.6)
Length of staying days		4–78	27.1 (16.6)

Overall, the group encompassed a diverse array and combination of medical conditions and pain symptoms.

Twenty-five (60%) patients had been seen by at least four or more healthcare specialists prior to admission; 33 (80%) had undergone four or more outpatient diagnostic procedures, medical interventions or surgeries, and 20 (49%) had been prescribed four or more medications (Table I). Despite receiving these healthcare services, these patients continued to exhibit pain, distress, and decreased functioning in the months since injury or onset of symptoms and prior to admission to the rehabilitation unit. The mean length of stay was 27.1 days with a range of 4–78 days.

A comparison of the 19 patients for whom follow-up data were obtained to those without follow-up data using a Pearson's chi-square test showed no statistically significant difference for gender [$\chi^2(1) = .41$, $p > .52$] or ethnicity [$\chi^2(2) = 1.70$, $p > .43$]. An independent samples t-test was used to compare the mean age of the group with follow-up data to the group without data and indicated no statistically significant difference [$t(39) = -.31$, $p > .76$].

School Attendance

Review of the data revealed that 36 (88%) patients had disrupted school attendance prior to intervention. Specifically, 15 (37%) did not receive any formal school services, 19 (46%) received home and hospital services, and 2 (5%) attended school on a part-time basis. Only 5 (12%) attended school full-time. A comparison of the 19 patients with follow-up data to those without follow-up data on their school status at admission using

the Pearson Chi-Square revealed no statistically significant difference [$\chi^2(3) = 4.3$, $p > .24$]. At discharge, 35 (86%) patients were reintegrated into a full day school program, while 3 (7%) continued with part-time status, 2 (5%) received home and hospital services, and 1 (2%) did not return to school (had previously dropped out of college coursework). School status rank sums between admission and discharge were significantly different [Wilcoxon signed-rank test $N = 34$, $T = 7$, $p < .001$]. A comparison of the 19 patients with follow-up data to those without follow-up data on their school status at discharge using the Pearson Chi-Square test did not reveal statistical significance [$\chi^2(3) = 4.5$, $p > .21$].

Sleep

Twenty-nine patients (71%) reported or exhibited disrupted sleep at admission. At discharge, this number was significantly reduced to 3 (7%) patients (Wilcoxon signed-rank test $N = 26$, $T = 15$, $p < .001$). Comparison of the 19 patients with follow-up data to those without follow-up data on sleep status showed no statistically significant difference at either admission [$\chi^2(1) = 3.1$, $p > .08$] or discharge [$\chi^2(1) = 3.7$, $p > .06$].

Functional Status

The mean WeeFIM total score increased significantly from 106.5 ($SD = 14.8$) at admission to 115.4 ($SD = 9.5$) at discharge [related samples $t(35) = -7.16$, $p < .001$]. For the 19 (46%) patients having 3-month follow-up data, the mean WeeFIM total score increased significantly from admission (108.9; $SD = 12.7$) to follow-up [121.5; $SD = 5.2$], $t(18) = -3.86$, $p < .01$]. Independent samples t-tests found that when comparing the 19 patients with follow-up data to those without follow-up data on the total WeeFIM score indicated a statistically significant difference at both admission [$t(39) = -2.6$, $p < .01$] and discharge [$t(39) = -2.5$, $p < .02$].

Physical Mobility

The mean WeeFIM mobility subscale score increased significantly from 25.9 ($SD = 8.0$) at admission to 29.8 ($SD = 5.2$) at discharge [Related samples $t(35) = -4.66$, $p < .001$]. The mean physical mobility subscale score for the 19 patients with 3-month follow-up increased significantly from 27.0 ($SD = 8.0$) at admission to 33.4 [$SD = 2.1$; $t(18) = -3.39$, $p < .01$] at discharge. Independent samples t-tests comparing the 19 patients with follow-up data to those without follow-up data on the physical mobility subscale found statistically significant differences at both admission [$t(39) = -2.5$, $p < .016$] and discharge [$t(39) = -2.3$, $p < .026$].

Medication Usage

Patient's mean number of prescribed medications (anti-depressant, anxiolytic, analgesic, anticonvulsant, etc.) decreased from 6.8 ($SD = 4.0$) at admission to 4.6 ($SD = 3.1$) at discharge [$t(40) = 4.66$. $p < .001$]. The number of patients taking 7 or more medications decreased from 19 (46%) at admission to 8 (20%) at discharge and 17 (41%) were taking 3 or fewer. Comparison of the 19 patients with follow-up data to those without follow-up data on the number of medications taken using an independent samples t-test showed no statistically significant difference at either admission [$t(39) = -.44$, $p > .66$] or discharge [$t(39) = -.24$, $p > .81$].

Utilization of Coping Skills

During intervention, data were collected on patient's use of coping strategies via the written daily schedule data sheet. Members of the interdisciplinary team recorded what strategies were used, if any, in each of their own scheduled therapy activities. Patients were observed to engage in a variety of coping strategies throughout their admission: 95% utilized distraction, 83% used focused breathing, 61% engaged in guided imagery, 56% practiced positive self-talk, 44% engaged in progressive muscle relaxation, and 29% used biofeedback. In addition, 17% engaged in journal writing, while one patient manipulated a stress ball during physical therapy sessions. Patients used an average repertoire of four different coping strategies during their structured day (range 1–6).

Discussion

The patients described in this study were admitted to the inpatient rehabilitation program for treatment of PADS. We hypothesized that, as a group, the patients in this sample would show significant improvement across measures of functioning after completing the inpatient protocol as compared to their pre-admission functioning. As a group, patients showed statistically and clinically significant improvements in the areas of functional ability and physical mobility after participation in the rehabilitation program. Clinical significance was determined by comparing patient's admission and discharge WeeFIM total and subscale mobility scores. For example, a patient at admission may have had significant difficulties with self-care, cognition and mobility. At discharge, a patient would likely exhibit more autonomy and require less supervision in these areas of functioning. Patients also showed improvements in school status, sleep, and medication use. Patients appeared responsive to the treatment

protocol consisting of differential positive reinforcement of coping behavior and compliance with demands of gradually increasing difficulty within the rehabilitation model. For those with available follow-up information, it appeared that physical functioning was maintained or continued to improve in this group after discharge. Given the diversity of medical conditions and wide age range of patients, we were encouraged to find that this integrated approach was useful and generalized across ages, diagnoses, and presenting problems. We believe that the protocol was feasible for such a wide developmental age range because of individualized shaping procedures and cognitive-behavioral interventions (e.g., distraction, imagery, etc.) that could be modified for different ages.

This study contributes to the literature on PADS by presenting the clinical outcomes of an interdisciplinary inpatient program for the treatment of PADS. As previous literature has suggested (Bursch et al., 1998; Eccleston, et al., 2003, 2006; Hyman, 2002; Palermo, 2000) the single most important consideration for the treatment of PADS may be the need for a biopsychosocial rehabilitation model, rather than the dichotomizing of etiologic factors (i.e. physiological and psychological). In this study the integrated approach included medical, educational, family, and environmental interventions. It illustrates the benefits of individualized shaping and cognitive-behavioral interventions. Use of shaping may allow children to experience their first successes in small steps, which may then help promote increased coping and motivation to recover. The rehabilitation hospital setting provides a unique environment, in which multiple staff members across settings and situations can prompt and differentially reinforce functional gains and coping behavior while not reinforcing pain or illness behavior.

The results presented in this study are based on a retrospective chart review of the outcomes for a clinical intervention protocol. Thus there are obvious limitations, such as lack of a treatment control group to account for any non-specific effects of inpatient treatment. We also could not fully evaluate variability in response to treatment because of the large number of potential sources of variability, patient characteristics and the relatively small sample size studied. Therefore, we did not have enough statistical power to examine which specific types of patients responded best to the treatment protocol. We also did not employ blinded raters or systematically assess the fidelity with which the protocol was implemented across patients.

Another limitation was that follow-up data were only available for about one-half of the patients and this limits the generalizability of the results. Additionally, we found a significant difference between the patients with follow-up data compared to those without data in physical mobility and total functional status scores at admission and discharge, which limits our ability to make conclusions about the long-term outcomes of these patients. Also, apart from the interdisciplinary scoring of the WeeFIM, different therapists and clinicians utilized a variety of formal and informal methods and measures to assess social functioning, coping, psychosocial stress, etc. Given that this was a retrospective study conducted over the course of 7 years, it is limited by heterogeneity in the patient cohort and chart documentation.

It is also important to note that, although the nursing staff and medical staff obtained subjective ratings of pain, these were not emphasized with the patients or included as an outcome measure in this article. The emphasis on functional measures rather than subjective pain ratings is an important part of the behavioral rehabilitation approach. Furthermore, we have noted that patients show little variability in their pain ratings during admission, often giving continued high ratings of pain (9 or 10 out of 10) despite observed positive affect during participation in therapeutic and leisure activities. Given that this was not a controlled experiment, we cannot claim a causal relationship, nor did we attempt to dismantle the relative contributions of the many therapeutic components included in the protocol.

Future research should examine prospective randomized controlled clinical trials involving multiple centers utilizing the same protocol. Future studies should also incorporate additional standardized outcome measures commonly used with this population (e.g., the Functional Disability Inventory, Walker & Greene, 1991). Pediatric psychology staff on the unit now administers objective measures to assess coping, pain, functional disability, mood, and anxiety. All patients fill out these standardized measures at time of admission, discharge, and follow-up in the outpatient pain management clinic. Future prospective studies should focus on long-term maintenance of gains, use of consistent standardized, reliable, and valid measures to comprehensively assess sleep, emotional functioning, functional disability, coping, and pain at admission, discharge, and follow-up. Future research also should examine when children begin to report decreased subjective pain, as they exhibit significant and sustained improvements in function across domains. However, this may not occur until later during follow-up care.

Future research should examine the cost-benefit ratio for an interdisciplinary inpatient rehabilitation treatment approach that targets PADS. This requires close consideration given the great cost of an inpatient admission and

likelihood that insurance companies may deny inpatient treatment for this reason. However, the burden of pain and resultant impairment frequently leads to extensive health-seeking behavior. Health-seeking patterns can result in frustration and be costly for patients, their families, and medical professionals. Of note, Palermo (2000) published a case report that examined the cost-effectiveness of an inpatient rehabilitation treatment approach. Based on the cost analysis, Palermo hypothesized that the expenses of an effective and relatively short (i.e., 22 days) inpatient admission may be far less compared to the cost of several years of outpatient medical interventions. In the above case study, the length of the inpatient admission and estimated baseline are comparable to the mean length of stay and baseline found in the present study. Overuse of healthcare resources can have significant economic costs, but with appropriate treatment, potential lifelong patterns of unnecessary health care use can be avoided. Future studies are needed to examine individual differences in response to treatment because it is likely that variability exists. Studies aimed at predicting responsiveness to treatment (e.g., who benefits most and why) are necessary in the future to better understand the contributing factors for treatment outcome variability (i.e. age, diagnosis, length of baseline, location of pain, etc.) with a larger sample size.

Conclusions

Children and adolescents with PADS are a subset of chronic pain patients with common difficulties in daily functioning, regardless of the location and etiology of the pain. Once a downward spiral of pain and disability begins, use of a biopsychosocial model that addresses contextual factors is highly recommended to ensure that a comprehensive and appropriate assessment and treatment plan is adopted. The interdisciplinary behavioral rehabilitation approach described in this study appears promising for increasing functioning, as defined by physical performance, increasing school attendance, improving sleep, use of active coping strategies, reducing medication usage, and possibly reducing future over-utilization of healthcare resources.

Acknowledgments

References

Amari, A., Slifer, K. J., Sevier, R. C., Spezio, J., & Tucker, C. L. (1998). Using differential reinforcement to treat functional hypophonia in a pediatric rehabilitation patient. Pediatric Rehabilitation, 2, 89–94.

Bursch, B., Joseph, M. H., & Zeltzer, L. K. (2003). Pain-associated disability syndrome. In N. L. Schechter, C. B. Berde, & M. Yaster (Eds.), Pain in infants, children, and adolescents (pp. 841–848). Philadelphia, P.A.: Lippincott, Williams, & Williams.

Bursch, B., Walco, G. A., & Zeltzer, L. (1998). Clinical assessment and management of chronic pain and pain-associated disability syndrome. Journal of Developmental and Behavioral Pediatrics, 19, 45–53.

Dobe, M., Damschen, U., Reiffer-Wiesel, B., Sauer, C., & Zernikow, B. (2006). [Three-week multimodal inpatient treatment of children with chronic pain. First results of the long-term follow-up], Schmerz, 20, 51–60.

Eccleston, C., Connell, H., & Carmichael, N. (2006). Residential treatment settings for adolescent chronic pain management: Rationale, development and evidence. In G. A. Finley, P. J. McGrath, & C. T. Chambers (Eds.), Bringing Pain Relief to Children: Treatment Approaches. New Jersey: Humana Press.

Eccleston, C., Malleson, P.M., Clinch, J., Connell, H., & Sourbut, C. (2003). Chronic pain in adolescents: Evaluation of a programme of inter-disciplinary cognitive behaviour therapy. Archives of Disease in Childhood, 88, 881–885.

Grilli, L., Feldman, D. E., Majnemer, A., Couture, M., Azoulay, L., & Swaine, B. (2006). Associations between functional independence measure (WeeFIM) and the pediatric quality of life inventory (PedsQL4.0) in young children with physical disabilities. Quality of Life Research, 15, 1023–1031.

Guide for the Uniform Data Set for Medical Rehabilitation for Children (WeeFIM). Version 5.0. Inpatient. Buffalo: Center for Functional Assessment Research, Uniform Data System for Medical Rehabilitation, State Univ New York; 1998.

Hyman, P. E., Bursch, B., Sood, M., Schwankovsky, L., Cocjin, J., & Zeltzer, L. K. (2002). Visceral pain-associated disability syndrome: A descriptive analysis. *Journal of Pediatric Gastroenterology and Nutrition, 35*, 663–668.

Lee, B.H., Scharff, L., Sethna, N.F., McCarthy, C.F., Scott-Sutherland, J, Shea, A.M., et al. (2002). Physical therapy and cognitive-behavioral treatment for complex regional pain syndromes. *Journal of Pediatrics, 141*, 135–140.

Majnemer, A., Limperopoulos, C., Shevell, M., Rohlicek, C., Rosenblatt, B., & Tchervenkov, C. (2008). Developmental and functional outcomes at school entry in children with congenital heart defects. *Journal of Pediatrics, 153*, 55–60.

Mindell, J. A., & Owens, J. A. (2003). *A clinical guide to pediatric sleep: Diagnosis and management of sleep problems*. (pp. 18). Philadelphia, PA: Lippincott, Williams & Wilkins.

Msall, M. E., DiGuadio, K. M., Duffy, L., LaForest, S., Braun, S., & Granger, C. V. (1994). WeeFIM: Normative sample of an instrument for tracking functional independence in children. *Clinical Pediatrics, 33*, 431–438.

Ottenbacher, K. J., Msall, M. E., Lyon, N. R., Duffy, L. C., Granger, C. V., & Braun, S. (1997). Interrater agreement and stability of the Functional Independence Measure for Children (WeeFIM): Use in children with developmental disabilities. *Archive of Physical Medicine and Rehabilitation, 78*, 1309–1315.

Ottenbacher, K. J., Taylor, E., Msall, M. E., Braun, S., & Granger, C. V. (1996). The stability and equivalence of the Functional Independence Measure for Children (WeeFIM). *Developmental Medicine and Child Neurology, 38*, 907–916.

Palermo, T. M. (2000). Impact of recurrent and chronic pain on child and family daily functioning: A critical review of the literature. *Journal of Developmental and Behavioral Pediatrics, 21*, 58–69.

Palermo, T. M., & Scher, M. S. (2001). Treatment of functional impairment in somatoform pain disorder: A case example. *Journal of Pediatric Psychology, 26*, 429–434.

Perquin, C. W., Hazebroek-Kampschreur, A. A., Hunfled, J. A., Bohnen, A. M., van Suijlekom-Smit, L. W., Passchier, J., & Van der Wouden, J. C. (2000). Pain in children and adolescents: A common experience. *Pain, 87*, 51–58.

Prosser, L. A. (2007). Locomotor training within an inpatient rehabilitation program after pediatric incomplete spinal cord injury. *Physical Therapy, 87*, 1224–1232.

Sherry, D.D., Wallace, C.A., Kelley, C., Kidder, M., & Sapp, L. (1999). Short- and long-term outcomes of children with complex regional pain syndrome type I treated with exercise therapy. *Clinical Journal of Pain, 15*, 218–223.

Sperle, P. A., Ottenbacher, K. J., Braun, S. L., Lane, S. J., & Nochajski, S. (1997). Equivalence reliability of the functional independence measure for children (WeeFIM) administration methods. *American Journal of Occupational Therapy, 51*, 35–41.

Walker, L.S., & Greene, J. W. (1991). The functional disability inventory: Measuring a neglected dimension of child health status. *Journal of Pediatric Psychology, 16*, 39–58.

Wong, V., Wong, S., & Wong, W. (2002). Functional independence measure (WeeFIM) for Chinese Children: Hong Kong cohort. *Pediatrics, 109*, 36–50.

Zeltzer, L., Bursch, B., & Walco, G. (1998). Pain responsiveness and chronic pain: A psychobiological perspective. *Journal of Developmental and Behavioral Pediatrics, 18*, 413–422.

Zeltzer, L. K., Tsao, J. C., Bursch, B., & Myers, C. D. (2006). Introduction to the special issue on pain: From pain to pain-associated disability syndrome. *Journal of Pediatric Psychology, 31*, 661–666.