Empirically Supported Treatments in Pediatric Psychology: Disease-Related Pain

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Objective: To evaluate psychological literature addressing interventions for disease-related pain in children.
Methods: We conducted a literature review of all studies using psychological interventions for pain stemming directly from disease process as well as pain secondary to disease treatment.
Results: Few empirically validated psychological approaches to the treatment of disease pain were found. Although existing intervention studies do not meet Chambless criteria, some promising strategies were identified.
Conclusions: Clinical evidence suggests that cognitive-behavioral strategies for the management of disease pain in children are promising and manualized, controlled intervention studies are needed.

Key words: pain management; chronic illness; children; empirically validated interventions.

The focus of this article is pain secondary to disease in children, including pain arising from disease process itself and secondary to treatments for the pathophysiological condition. Interventions for pain related to medical procedures, including surgery, burn management, phlebotomy, lumbar punctures, and bone marrow aspirations, will be addressed elsewhere, as will the management of conditions in which pain is the primary presenting problem, such as recurrent abdominal pain and headaches. As will become clear, there are very few systematically controlled studies assessing the efficacy of psychological interventions for disease-related pain in children. The depth of the present discussion of various studies and case reports depends on the breadth of material originally published. A variety of syndromes will be considered, and thus overviews of incidence and treatment strategies applied to adult populations (for whom psychological interventions have been more extensively studied) will be provided for each major group of disorders. The disease entities to be highlighted include pain associated with neoplasms and their treatment, vaso-occlusive pain in sickle cell disease, and musculoskeletal pain associated with hemophilia and rheumatologic conditions. Table 1 summarizes the studies we found.

Cancer

Cancer is relatively rare in children age 15 and under, occurring in 129 per million per year among whites and 98 per million per year among African Americans in the United States (Li, 1993). The predominant types of pediatric cancer are leukemia...
Table 1. Summary of Intervention Studies of Pediatric Disease-Related Pain

<table>
<thead>
<tr>
<th>Citation</th>
<th>N</th>
<th>Baseline Design</th>
<th>Measures</th>
<th>Protocol</th>
<th>Outcome</th>
<th>Follow-up</th>
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<tr>
<td><strong>Cancer</strong></td>
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<tr>
<td>Olness, 1981</td>
<td>1 boy, age 13</td>
<td>None stated</td>
<td>Case study Imagery interview</td>
<td>Imagery sessions</td>
<td>Reported meds, pain decrease, thermal control</td>
<td>30 months</td>
</tr>
<tr>
<td>Hilgard &amp; LeBaron, 1984</td>
<td>1 man, age 20</td>
<td>None stated</td>
<td>Case study ACSHR</td>
<td>Hypnosis sessions</td>
<td>Reported meds, pain decrease</td>
<td>None</td>
</tr>
<tr>
<td>LaClave &amp; Blix, 1989</td>
<td>1 woman, age 20</td>
<td>Not defined</td>
<td>Case study Not defined</td>
<td>Hypnosis sessions</td>
<td>Reported pain decrease</td>
<td>None</td>
</tr>
<tr>
<td><strong>Rheumatology</strong></td>
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<tr>
<td>Lavigne et al., 1992</td>
<td>7 girls, 1 boy ages 9-17</td>
<td>28 days</td>
<td>Multiple Pain diaries, VAS</td>
<td>6 sessions, PMR, EMG biofeedback, thermal biofeedback</td>
<td>Reported pain decrease</td>
<td>6 months</td>
</tr>
<tr>
<td>Walco et al., 1992</td>
<td>8 girls, 5 boys ages 4-16</td>
<td>4 weeks</td>
<td>Longitudinal within subjects</td>
<td>PPQ, VAS, CAQ</td>
<td>Lowered VAS, improved activities' daily living</td>
<td>6 months</td>
</tr>
<tr>
<td>Walco &amp; Ilowite, 1992</td>
<td>7 girls, ages 8-17</td>
<td>Not defined</td>
<td>Within subjects PPQ, VAS</td>
<td>4-9 sessions, PMR, guided imagery</td>
<td>Lowered VAS</td>
<td>4-24 months</td>
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<td><strong>Hematology</strong></td>
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<tr>
<td>Varni &amp; Gilbert, 1982</td>
<td>1 man, age 31</td>
<td>5 weeks</td>
<td>Case study Self-monitoring</td>
<td>25-step PMR, meditative breathing, guided imagery</td>
<td>Symptom, meds, decrease</td>
<td>Not specified</td>
</tr>
<tr>
<td>Varni et al., 1981</td>
<td>1 boy, age 9</td>
<td>2.5 weeks</td>
<td>Case study Not defined</td>
<td>25-step PMR, meditative breathing, guided imagery</td>
<td>Less meds, pain, hospitalizations</td>
<td>1 year</td>
</tr>
<tr>
<td>Varni, 1981</td>
<td>2 men, ages 19, 27</td>
<td>1-3 weeks</td>
<td>Case study Self-monitoring</td>
<td>25-step PMR, meditative breathing, guided imagery</td>
<td>Symptom, meds, decrease</td>
<td>8 months</td>
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<td>Pt. 2, increased thermal control</td>
</tr>
<tr>
<td>Zeltzer et al., 1979</td>
<td>2 men, age 20</td>
<td>None stated</td>
<td>Case study Self-report, chart review</td>
<td>Eye fixation, PMR</td>
<td>Better socialization, decreased meds, clinic, impatient, ER visits</td>
<td>8 months</td>
</tr>
<tr>
<td>Thomas et al., 1984</td>
<td>7 women, 8 men, ages 22-35</td>
<td>6 months</td>
<td>T test for paired distributions</td>
<td>Self-report, chart review</td>
<td>15 sessions, PMR, biofeedback, self-hypnosis, cognitive strategies</td>
<td>Decreased ER, inpatient days, rep. improved mood, QOL</td>
</tr>
<tr>
<td>Cozzi et al., 1987</td>
<td>5 girls, 3 boys, ages 10-20</td>
<td>6 months</td>
<td>Repeated measures STAI, self-concept scales, drawings, pain, diaries</td>
<td>12 sessions, EMG, thermal biofeedback, practice tapes</td>
<td>Decreased state anxiety, headaches, symptoms, meds</td>
<td>6 months</td>
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Independent medical diagnoses were made by pediatric subspecialists and study participants met diagnostic criteria for oncologic, rheumatologic, and hematologic disorders.

All patients with sickle cell disease were African American. No other racial information was provided.

ACSHR = Adult Clinical Scale of Hypnotic Responsiveness.

PMR = progressive muscle relaxation.

VAS = visual analog scale.

PPQ = Pediatric Pain Questionnaire.

EMG = electromyogram.

CBCL = Child Behavior Checklist.

CAQ = Children's Activity Questionnaire.

QOL = quality of life.

STAI = State Trait Anxiety Inventory.
(30.5%), brain tumors (18.8%), lymphoma (11.9%), neuroblastoma (8.0%), and soft tissue sarcomas (6.4%). These are quite different than prevalence figures in adults, in whom solid tumors, such as carcinomas, are much more typical (Stevens, Pozza, Cavalleto, Cooper, & Kilham, 1994). The differential epidemiology of cancer types between children and adults implies a variation in the prevalence and nature of disease-related pain as well. In all age groups, pain may stem from tumor invasion or infiltration of bone, nerve, or other organ systems, as well as soft tissue invasion, capsular distention (e.g., splenic metastases), hollow viscous obstruction, and invasion or compression of central or peripheral nervous systems (Miser & Miser, 1989). Cancer pain may range in severity from mild to excruciating, depending on the type, location, and progression of the tumor (Bonica, 1980).

An effort to determine the prevalence and nature of pain in the pediatric and young adult cancer population was conducted by Miser, Dothage, Wesley, and Miser (1987), who assessed 139 patients at the National Cancer Institute (NCI) over a 6-month period. It should be noted that this represented a high-risk group undergoing aggressive treatment protocols. During that period, pain was appreciated 54% of the time among inpatients and 27% of the time among outpatients. Of note, at study entry only 29.3% and 12.9% of inpatients and outpatients, respectively, experienced pain due to neoplastic process per se. This was most common within 2 weeks of diagnosis or in later terminal stages. Most often pain was found to be secondary to treatment, including pain from mouth sores (mucositis or stomatitis) and neuropathic pain secondary to chemotherapy. In a related study conducted at nine community treatment centers during a 1-week period, Elliott et al. (1991) found that 17.5% of pediatric cancer patients experienced pain, 57.8% of which was secondary to disease treatment, while 21.1% was disease-related pain. Thus, a comprehensive approach to pediatric cancer pain treatment must encompass difficulties related both to neoplastic process and its treatment (Miser & Miser, 1989).

The mainstay for cancer pain relief is analgesics (World Health Organization [WHO], 1990). An “analgesic ladder” is invoked with nonsteroidal anti-inflammatory drugs or acetaminophen prescribed for mild pain, narcotics (e.g., codeine) for moderate pain, and opioids (e.g., morphine) for severe pain. Tricyclic antidepressants or anticonvulsants are indicated for neuropathic pain. With the exception of procedural pain (see article, by Scott Powers, pp. 131–145 in this issue), psychological treatments for cancer pain in children have been uncommon, likely because there is deemed to be such a strong organic component to this pain (Jay, Elliott, & Varni, 1986).

There are also very few studies addressing psychological interventions for cancer pain in adults. Spiegel and Bloom (1983) included self-hypnosis as part of a group program for women with metastatic breast cancer. Lowest pain ratings were found in the condition that combined group support strategies and these techniques. Syrjala, Cummings, and Donaldson (1992) found that mucositis pain associated with bone marrow transplant was best managed by those who had received hypnosis training in comparison with other psychological interventions (cognitive-behavioral strategies that included relaxation, but not hypnosis or imagery, attention control, or treatment as usual). Syrjala, Donaldson, Davis, Kippes, and Carr (1995) reported on a similar controlled study with 94 patients undergoing bone marrow transplantation in the same institution and demonstrated the efficacy of hypnosis for various aspects of controlling nausea and vomiting. Besides the value of demonstrated treatment outcomes, this study is significant for the present discussion as it highlights the disparity between studies of adult and pediatric cancer pain. Because of the relative low frequency of bone marrow transplants in children, securing 94 pediatric bone marrow transplant patients in a single setting is nearly impossible, and thus multicenter studies are imperative.

We found a handful of case studies describing the application of hypnosis and imagery for disease-related pain in cancer. Olness (1981) reported several case studies in which self-hypnosis was used to reduce pain and nausea related to illness and treatment. Of the 25 cases discussed, procedural pain was the focus of treatment in all but one, a 13-year-old child with Ewing’s sarcoma who was referred for pain control and assistance with sleeping problems. Using imagery techniques that invoked numbness in the areas of discomfort, the child demonstrated improvement in pain control and a decrease in requirements for pain medication while in the hospital. After discharge, he was seen for three additional sessions, as well as group practice sessions. Olness made the point that children exposed to self-hypnosis early in their treatment may have greater utility in using the techniques for symptom management later on.

Hilgard and LeBaron (1984) discussed the use of
hypnotherapy as a method of relieving pain and anxiety in children with cancer. Once again, the vast majority of cases focused on procedural pain. They described an intervention with a 20-year-old suffering from chronic pain secondary to an abdominal tumor. The patient reported pain in his back, abdomen, and hips, rating his pain at 6 on a 10-point scale, with ratings as high as 9. Two days after being taught relaxation and imagery, he reported pain to be at an average level of 2. Additionally, morphine requirements were reduced from 3 to 1 mg per hour. The authors highlighted self-report and reduction in morphine as key outcome measures, but provided little detail on the nature and length of intervention.

LaClave and Blix (1989) used hypnosis to manage cancer-related symptoms in a 6.5-year-old girl with malignant astrocytoma. The patient had undergone seven sessions of hypnosis to address vomiting secondary to chemotherapy, which was successful. Two months later she began to experience severe headaches, which were reportedly alleviated by associating pain with colors and visual imagery. Although they reported that hypnosis was beneficial in decreasing pain, the authors did not quantify the pain in any fashion or state the method of pain assessment. No other studies that evaluated the efficacy of psychological intervention for pediatric cancer pain were found.

**Chambless Criteria**

Based on the Chambless criteria for empirically validated treatments, it is difficult to draw any conclusions about the value of psychological interventions for cancer-related pain in children. Although case reports and clinical anecdotes indicate that self-regulatory strategies, such as hypnosis, have been beneficial, no controlled studies were found in this area. Extrapolating from the adult oncology literature, it seems plausible that these interventions would have merit for younger patients. Furthermore, such strategies may be helpful in pediatric oncology for other reasons. First, various aspects of mood, such as anxiety and depression, affect the pain experience, and cognitive-behavioral strategies focused on pain may be generalized to these affective components. Second, cognitive-behavioral techniques may provide some semblance of control in a situation that otherwise fosters passivity. Third, because patients experience pain while away from the health care setting, cognitive-behavioral techniques acquired at the hospital may be generalized to other situations. Finally, the potent analgesics used to control pain are not without side effects. Techniques such as hypnosis and distraction may be helpful as an adjunct to reduce the amount of medication needed for adequate pain control.

**Musculoskeletal Disorders**

Over the past 15 years, multiple studies have documented the efficacy of cognitive-behavioral interventions for pain associated with arthritis in adults. As discussed in a recent review by Keefe and Caldwell (1997), three patient populations have been the focus: rheumatoid arthritis, osteoarthritis, and fibromyalgia.

Among patients with rheumatoid arthritis, pain is typically persistent and there are acute flare-ups to be addressed. As a result, protocols that enhance patients’ abilities to control or decrease pain are important, as demonstrated in a number of controlled studies with large samples (greater than 50 patients). Similar results have been shown in affecting pain experience among patients with osteoarthritis, but outcomes reflecting improvement in physical functioning have been less forthcoming. In contrast to the number of well-designed studies on pain in adults with arthritis, the number of quality studies on fibromyalgia syndrome are far fewer. While some studies with smaller sample size or poorer controls demonstrate promise for cognitive-behavioral interventions for pain and disability in this group, the benefit of psychological strategies over physical interventions has not been shown (Keefe & Caldwell, 1997).

**Juvenile Rheumatoid Arthritis**

Juvenile rheumatoid arthritis (JRA) is an autoimmune disorder characterized by inflammation of organs, joints, and other parts of the body that occur before or at age 16 years (Jaworski, 1993). The prevalence rates for chronic JRA is 0.6 to 1.1 per 1,000 children (Gewanter, Roghmann, & Baum, 1986), and girls are generally affected about four times more often than boys (Leak, 1994). Pain is one of the most prominent features of JRA (Konkol et al., 1989), and the degree to which it is managed successfully is a powerful predictor of children’s adaptation to their illness (Lovell & Walco, 1989).
Pharmacologic interventions are a mainstay in managing JRA. Nonsteroidal anti-inflammatory drugs (including aspirin and ibuprofen) serve to reduce inflammation and alleviate pain (Leak, 1994). In more severe cases, powerful chemotherapeutic agents (e.g., methotrexate) may be used to combat the disease process. Unfortunately, even with recent pharmacological advances, coping with chronic pain remains a major challenge for many of these children. To date, very few studies have systematically explored the efficacy of psychological techniques for chronic joint pain in children with JRA.

Lavigne, Ross, Berry, Hayford, and Pachman (1992) evaluated a psychological treatment package for children experiencing moderate to high levels of pain associated with JRA. Seventy-eight patients between the ages of 6 and 17 years were approached for the study. Of those, 10 refused and 12 failed to return diaries. Of the 56 eligible subjects, 21 met study criteria for severity of pain. Ultimately, eight subjects participated in the treatment phase of the study.

This study employed a multiple baseline across-subjects design. Half of the subjects were assigned to an immediate treatment group, while the other half were assigned to a delayed treatment group (which served as a control). Children’s self-reported pain diaries and maternal pain diaries were collected at pretreatment, posttreatment, and follow-up. Measures included in the diary were 10-cm visual analog scales for worst pain rated at three times during the day by both subjects and their mothers. Scores were recorded, as well as the number of episodes in which pain was greater than or equal to +5. Mothers also rated pain behavior, as did a physical therapist, and data on functional status were obtained.

The intervention consisted of six individual sessions, conducted biweekly, to be completed in a 3-month period. Both parent and child were seen at each session and specific homework assignments were given. A treatment manual was developed for this study. Sessions 1 and 2 focused on the teaching and implementation of progressive muscle relaxation exercises. Sessions 3 and 4 built on relaxation and included electromyogram biofeedback. In addition, operant pain management strategies were taught to the mothers and discussed during all subsequent sessions. Sessions 5 and 6 involved training in thermal biofeedback.

Because the number of participants was relatively small, the authors discussed treatment outcomes for individual subjects. Overall, there was a 25% reduction in +5 pain periods for five of the eight subjects from baseline to posttreatment and a 25% reduction in mean pain scores from baseline to long-term follow-up in seven of the eight. Repeated measures (baseline, posttreatment, follow-up) analyses of variance indicated significant improvement from baseline to follow-up for both mean pain scores and the number of +5 pain periods. Maternal ratings yielded similar results. Although the authors reported large and statistically significant differences between the treatment and delayed treatment group (assessing immediate impact of intervention), there were only four subjects per cell. A comparison of pre- and posttreatment pain ratings indicated no differences between the immediate and delayed treatment groups.

The weakness of this study lies in its small sample size. The large number of eligible children who declined to participate in the study illustrates the difficulty associated with research of this nature. Perhaps those children and mothers who agreed to participate are significantly more motivated than nonparticipants and therefore are not representative of the population at large. Furthermore, it is difficult to truly ascertain the impact of the treatment program as virtually all children with significant disease were receiving concurrent pharmacological interventions aimed at reducing inflammation and pain.

A study conducted by Walco, Varni, and Ilowite (1992) also examined cognitive-behavioral pain management in children with JRA. Similar to the Lavigne et al. (1992) study, 58 children were approached for the study, 26 of whom agreed to participate. Only half of that group, (13 subjects between the ages of 4.5 and 16.9 years) actually completed the intervention program, which consisted of cognitive-behavioral techniques aimed at reducing pain intensity and enhancing adaptive functioning. The authors systematically explored reasons for nonenrollment or discontinued participation and found that a range of demographic and disease-related variables were not predictive. Most family variables were also not significant, with the exception of maternal ratings of marital satisfaction.

Joint pain was assessed at intake using the Pediatric Pain Questionnaire (PPQ; Varni & Thompson, 1985) and subsequent visual analog scales (VAS) used twice per day over a 4-week baseline period. A questionnaire focused on adaptive functioning was
also used at intake. Follow-up data were gathered at 6 and 12 months after the final treatment session.

A standardized treatment package was followed for eight weekly sessions that involved three sequential phases: (1) progressive muscle relaxation, (2) meditative breathing, and (3) guided imagery aimed at facilitating distraction or pain reduction through the use of metaphor. Minor modifications were made to tailor the intervention to each child’s developmental level, pain experience, interests, and abilities. Children were instructed to practice the techniques on a regular basis at home and were provided with audiotapes as a means of facilitating generalization. Parents were seen on two additional occasions to discuss aspects of chronic pain management and to be taught basic elements of behavioral strategies to maximize adaptive functioning.

The results indicated short-term benefits of the intervention as mean VAS scores decreased from baseline to immediate posttreatment. Indices of both statistical and clinical significance were reported for interventions conducted in the clinic as well as those completed by the subjects at home. In addition, long-term effects were found at the 6-month follow-up, at which time children rated their pain as significantly lower both in the morning and afternoon/evening periods, and were maintained at the 12-month follow-up. It is noteworthy that data from parents closely paralleled those of their children. Along with the reduced pain experience following intervention, children showed improvement in activities of daily living at the 6- and 12-month follow-up assessment periods relative to the pretreatment baseline. Subject attrition was so high, however, that no statistical tests were conducted on long-term functional status data.

Similar to the previous study, this project also suffered from a small number of subjects. As a result, there was no control group and all assessments of outcome were within-subjects designs. Inclusion of appropriate control groups would demonstrate the superiority of the intervention. Second, in this study there was a 50% dropout rate. Thus, there may be a selection bias as those subjects who completed the program were likely to be the most motivated. Third, although characteristics of the patient sample were delineated and information about age range, breakdown of gender, medical diagnosis and medication use were all provided, demographic data on socioeconomic status and race were not given, thereby potentially limiting external validity.

**Hemophilar Arthropathy**

Hemophilia is a congenital hereditary disorder of blood coagulation, which affects approximately 1 in 10,000 males (Robinson & Linden, 1993). It is a lifelong chronic disorder characterized by recurrent, unpredictable bleeding episodes that can be triggered by physical trauma or emotional factors in any part of the body, but typically in the joints and extremities. Prior to the consistent prophylactic administration of factor products, repeated bleeding into the joints produced a chronic condition similar to osteoarthritis, characterized by destruction of articular cartilage, pathologic bone formation, and impaired function (Sokoloff, 1975).

Although the use of specific factor replacement products has reduced the morbidity of conditions secondary to hemophilia, in the past approximately 75% of adolescents and adults with hemophilia were affected by chronic arthritis pain (Dietrich, 1976). The child with hemophilia needed to differentiate acute pain due to hemorrhage from the chronic nature of arthritic pain (Varni & Gilbert, 1982). The former provides a functional signal, indicating the necessity of intravenous infusion of factor replacement, which temporarily replaces the missing clotting factor, converts the clotting status to normal, and allows a functional blood clot to form. Historically, approximately 10% of children with hemophilia developed an inhibitor to factor VIII in antihemophilic concentrate. Pain in these children could be severe, often requiring narcotic analgesics. A goal of psychological intervention was to provide a complement to medications for pain relief.

In a series of controlled single subject studies, Varni and associates (Varni, 1981a, 1981b; Varni & Gilbert, 1982; Varni, Gilbert, & Dietrich, 1981) described the use of biobehavioral methods in helping children and adults with chronic arthritic pain in hemophilia. The investigators used progressive muscle relaxation, meditative breathing exercises, and guided imagery to demonstrate that these techniques could reduce chronic pain without obscuring acute bleeding pain perception.

Varni, Gilbert, and Dietrich (1981) reported a case study involving a 9-year-old boy with hemophilia. At 4 years of age the child developed factor inhibitor and factor replacement became impossible. The child then required narcotics to tolerate pain associated with hemorrhage. Pain was so se-
vere that analgesic doses kept increasing, even with continued joint immobilization for bleeding episodes. The child eventually was confined to a wheelchair 50% of the time due to bleeding and arthritic pain, was hospitalized 16 times in the 4.5 years prior to the study period, and for a total of 80 days following the development of the inhibitor. Immediately prior to the study the child was brought to the emergency room due to painful left knee hemorrhage that had not responded to home therapy. The child also had no pain relief after meperidine and intravenous diazepam.

Training in self-regulation of pain perception consisted of three sequential phases (which served as the basis for the Walco et al. [1992] study described above): (1) a 25-step progressive muscle relaxation sequence involving the tensing and releasing of major muscle groups, (2) meditative breathing exercises, and (3) guided imagery techniques. Prior to self-regulation training, the patient was taking adult doses of meperidine for pain relief, as well as daily doses of acetaminophen with codeine. Following self-regulation training, there were no further requests for meperidine, and acetaminophen and codeine were decreased. There were also improvements in mobility, as evidenced by physical therapy measures of the left knee in comparison to the right, increased school attendance, and decreased hospitalizations. Parents reported an elevation in the child’s mood, noticing less depression during pain episodes since the child had developed skills to reduce pain perception. Additional benefits included improved self-control, self-esteem, and peer relationships.

Additionally, Varni (1981a, 1981b) and Varni and Gilbert (1982) reported other well-controlled case studies with adults in which the aforementioned self-regulation sequence was taught with similarly effective results. Varni and Gilbert reported that the use of self-regulation training with a 31-year-old man with severe hemophilia showed clinically significant decreases in chronic arthritic pain intensity and an adjustment of the analgesic regimen to within recommended dosage levels after self-regulation training maintained over a follow-up period.

Varni (1981a) reported on the use of training in progressive muscle relaxation exercise, meditative breathing, and imagery with two adult men with hemophilia. The patients were 19 and 27 years old, both with severe arthritis. Imagery training resulted in clinically significant reductions in arthritic pain perception for both patients, maintained over an 8-month follow-up period. Thermal biofeedback assessment of the arthritic joint in the second patient provided a physiological measure of learned temperature control through the imagery techniques. In a third paper (Varni, 1981b), results were discussed in the context of a multiple baseline design with three adult subjects, but outcomes were the same.

**Juvenile Primary Fibromyalgia Syndrome**

Juvenile primary fibromyalgia syndrome (JPFS) is of unknown etiology and is characterized by widespread and persistent pain, multiple tender or trigger points on physical examination, difficulty initiating or maintaining sleep, and disturbance of mood, such as anxiety or depression (Bossevain & McCain, 1991; Wolfe et al., 1990). Pharmacological therapy thus far has been limited and unsuccessful. Nonsteroidal anti-inflammatory drugs and tricyclic antidepressants have proven to be of minimal short- or long-term benefit in adult populations (Goldenberg, Felson, & Dinerman, 1986) and have shown limited utility in adolescents (Yunus & Masi, 1985).

Self-regulatory techniques such as progressive muscle relaxation, meditative breathing, and guided imagery have been used to manage pain associated with fibromyalgia in adults (Bradley, 1989). Haanen et al. (1991) conducted a controlled study with adult patients with fibromyalgia, using hypnotherapy to successfully reduce pain experience. In order to test these techniques with children and adolescents, Walco and Ilowite (1992) implemented cognitive-behavioral interventions to treat seven girls between the ages of 8.6 and 17.7 years satisfying diagnostic criteria for JPFS.

The PPQ (Varni & Thompson, 1985) was used to assess various aspects of patients’ pain experiences. Visual analog scales were included to assess the intensity of present, average, and worst pain in the past week. Subjects were also screened for significant psychiatric dysfunction through diagnostic interview and psychometric assessments. Based on these data, a psychiatric diagnosis was assigned if relevant DSM-III-R criteria were met.

The treatment paradigm was modeled after a program used successfully with children with JRA (Walco, Varni, & Ilowite, 1992, described above). In-
struction was given on self-regulatory techniques aimed at reducing pain, which were generalized to sleep and mood problems. Cognitive-behavioral self-regulation of musculoskeletal pain consisted of the following: (1) patients were taught progressive muscle relaxation, (2) guided imagery techniques were taught that (a) utilized distraction and then (b) invoked metaphors for pain. This final step involved review of the nervous system and images of “pain switches” used to block transmission of pain messages.

Subjects were instructed to practice daily and were provided with audiotapes to facilitate generalization. Finally, self-efficacy and mastery over the pain were discussed to augment the intended effect of pain control strategies and to reduce anxiety and depressive thoughts. Treatment was terminated when patients and parents reported that the techniques had improved pain, sleep, and mood difficulties.

Data gathered at intake from the PPQ indicated high levels of average pain intensity (VAS: mean = 8.0 cm, SD = 1.02, range = 6.8–9.5), which reportedly interfered with school attendance and maintenance of daily routine. Two patients dropped out before completing treatment. Patients who completed treatment were seen for four to nine sessions, after which time pain was reported to be absent or negligible. Patient and parent reports in response to questions about activities of daily living, school attendance, participation in family activities, and interaction with peers revealed that patients had returned to premorbid levels of functioning.

Parents were recontacted by telephone 4 to 24 months after the conclusion of treatment and asked about their child’s pain experience, difficulty initiating or maintaining sleep, and general mood. Of the five subjects who completed the treatment, four reported no subsequent pain, and the final one reported low-level pain that could be controlled through use of self-regulatory techniques.

**Chambless Criteria**

Although these studies have demonstrated that cognitive-behavioral interventions hold promise for treating disease-related musculoskeletal pain in juvenile rheumatoid arthritis, hemophilia, and juvenile primary fibromyalgia syndrome, they do not meet the Chambless criteria for well-established treatments. With regard to hemophilic arthropathy, which seems promising, the pediatric case study by Varni and associates focused on a single child. The other cases (Varni, 1981a, 1981b; Varni & Gilbert, 1982) were adult subjects and thus results may not be generalizable to children and adolescents. It should also be noted that with the increased use of prophylactic factor replacement therapy, the occurrence of joint disease among patients with hemophilia is extremely rare.

For JRA, the study by Walco, Varni, and Ilowite (1992) had a replicable treatment, and documented effects, showing positive results with both clinical and statistical significance. However, the sample size was quite small and there was no control group. The Lavigne et al. (1992) study had a standardized methodology and a comparison group, but the sample was very small and there was no assessment of clinical significance. Although both studies demonstrated statistical significance related to treatment, samples failed to represent the heterogeneous nature of JRA, thereby limiting the replicability and reliability of results. Both studies also suffered from high rates of nonparticipation or subject attrition, which may bias outcomes.

Finally, for JPFS, Walco and Ilowite (1992) used sound methodology but provided only a clinical series. There were no control groups and no statistical analyses (not even a within-subjects design) to document the significance of observed improvements. The reader was also not provided with information about the two cases that failed to complete treatment, which may affect the clinical significance of the reported findings.

Based on the criteria generated by the Society of Pediatric Psychology, it is clear that psychological strategies for reducing musculoskeletal pain are promising interventions. The available data suggest that cognitive-behavioral interventions can potentially be effective as an adjunct to standardized pharmacotherapy for musculoskeletal pain disorders. Future studies with larger sample sizes and specified comparison groups, including those that control for effects of pharmacotherapy, will provide more definitive results as to the efficacy of psychological strategies in this arena.

**Sickle Cell Disease**

Sickle cell disease consists of a group of inherited hematological disorders involving defects in normal hemoglobin production. In the United States,
incidence of sickle cell anemia is estimated at 1 per 500 births in the African American population, while thalassemia has an incidence of approximately 1 per 1,000, occurring also in individuals of Greek and Italian descent (Robinson & Linden, 1993). Sickle cell disease is typically accompanied by painful vaso-occlusive episodes, which occur intermittently and may result in hospitalization. Management of painful crises principally has been conducted via the pharmacologic “pain ladder,” described earlier.

We found three studies that address cognitive or behavioral approaches to the management of sickle cell disease pain. Zeltzer, Dash, and Holland (1979) reported on case studies utilizing hypnosis with two 20-year-old black men to treat sickle cell disease pain. Both participants were referred for self-hypnosis training following extensive histories of painful episodes, hospitalizations, reliance on multiple analgesics, as well as disruption in daily life and psychosocial functioning. Medical record review provided historical information and follow-up data.

Hypnosis sessions involved eye fixation and progressive relaxation techniques, although session number and specifics of the intervention were not reported. At 4 and 8 months posthypnosis training, both participants were said to have marked reduction in painful crises, outpatient visits, emergency visits, and hospitalizations. Additionally, narcotic use was reduced and both patients were described as better able to pursue adaptive life goals.

Another study (Thomas, Koshy, Patterson, Dorn, & Thomas, 1984) addressed a combination of cognitive and behavioral strategies for pain management in 15 adult patients, ages 22 to 35, with sickle cell disease. Posttreatment data were compared to archival records of emergency room visits and hospitalizations prior to psychological intervention. Individual treatment spanned 15 sessions, but the actual protocols varied across patients. All patients were trained in a variation of progressive muscle relaxation, biofeedback, and several cognitive strategies. Participants were permitted to select or decline self-hypnosis training. Attendance and completion of measures were described to be erratic and consequently data are unclear. Results suggest that, compared to pretreatment, utilization of medical services was less frequent for participants in the 6 months following treatment. Long-term follow-up showed continued reduction of medication use, but these data were recorded for only three patients. This study and the case studies described previously suffer from inadequate treatment description and lack of a standardized approach.

The third study devoted to the evaluation of psychological approaches to pain management in sickle cell disease (Cozzi, Tryon, & Sedlacek, 1987) involved eight patients, ages 10 to 20 years. Treatment was more formalized with 13 sessions of biofeedback-assisted relaxation training, all of which were prerecorded and administered by tape. As in the previous studies, archival data served to provide baseline information about clinic visits and hospitalization and no other control group was included. An attempt at generalization was made during sessions, as patients were encouraged to engage in conversation about stressors both related and unrelated to their disease. Self-report data were collected at the first and last visits on a variety of psychological variables.

In contrast to the Zeltzer et al. (1979) and Thomas et al. (1984) studies, the Cozzi et al. (1987) study did not result in fewer emergency room visits or hospitalizations following treatment. However, Cozzi et al. (1987) described posttraining reductions in self-reported headaches, frequency of and pain associated with self-treated episodes, analgesic use, and state anxiety. These data were all obtained from subjective patient report and are vulnerable to biases inherent to self-report data.

Chambless Criteria

These studies do not meet Chambless criteria due to small sample sizes, absence of control groups, and the lack of manualized or even clearly replicable treatments. Participants were included who were over 18 years old in all of the studies, which limits demonstrated validity for children and adolescents. One study included subjects who were under 18; however, only eight patients participated and their ages ranged from 10 to 20 years old. Additionally, this study of biofeedback efficacy did not support objective criteria for reduced pain crises, resulting in unchanged hospitalization and emergency room visit frequencies following treatment.

Based on the results described by Zeltzer et al. (1979) and Thomas et al. (1984), as well as anecdotal evidence, hypnosis and progressive relaxation may warrant further examination to determine their adequacy as interventions for sickle cell dis-
ease pain. Studies designed to explore the efficacy of hypnosis and progressive relaxation skills should adhere to a defined research protocol and compare experimental subjects to an appropriate control group.

With sickle cell disease, in particular, the intermittent nature of pain crises may be appropriate for behavioral intervention designed to encourage the avoidance of crises. As children with sickle cell disease require large quantities of fluid to increase blood volume and, subsequently, decrease sickling episodes, behavioral programs designed to increase fluid intake or avoid extreme temperatures may be conducted to examine the potential to avert or minimize painful episodes.

**Future Directions in Psychological Interventions for Pediatric Pain**

The majority of the studies described here focus on reducing pain intensity as an initial treatment outcome and then measure various indices of adaptive behavior reflecting either decreased pain or enhanced coping. In future studies, researchers should carefully delineate their outcomes and the manner in which treatment strategies will address goals.

Varni (1983) offered a simple delineation of biobehavioral pain treatment modalities. Pain perception regulation includes cognitive-behavioral strategies aimed at reducing subjective experiences of discomfort or at increasing tolerance to that discomfort. Techniques such as progressive muscle relaxation, meditative breathing, guided imagery, and self-hypnosis are commonly used as part of a larger cognitive-behavioral framework designed to increase mastery over the pain. Pain behavior regulation involves strategies based on operant paradigms designed to decrease “pain” or “sick” behaviors, while maximizing “well” behaviors. Thus, pain behaviors are carefully defined and measured in terms of their frequency, duration, or intensity. Attention is then turned to antecedent stimuli and specific outcomes that increase or decrease the probability of those behaviors occurring.

Intervention research in pediatric psychology focused on disease-related pain is quite limited. One area of future research should be devoted to standardizing current treatment approaches aimed at modifying both pain behavior and pain perceptions. Anecdotal reports, case studies, and uncontrolled studies with small sample sizes have provided initial evidence of the success of such techniques. It is clear, however, that several steps must be taken before the psychological literature on the management of pediatric disease pain meets Chambless criteria for experimental rigor.

Accurate assessment of pediatric pain with reliable and valid measures is a necessary prerequisite for the experimental application of psychological interventions. Multiple aspects of the subjective pain experience (e.g., intensity, quality, duration) and of pain behavior must be systematically assessed (see McGrath, 1990, for a review). Researchers must specify outcomes and the precise interventions aimed at achieving treatment goals. Of the two types of interventions available for the treatment of pediatric disease-related pain, there is some experimental evidence for the efficacy of self-regulatory techniques, such as hypnosis, imagery, and relaxation. There are far fewer reports on operant models aimed at reducing pain behavior.

Studies should include adequate sample sizes, homogeneous disease populations, manualized treatment approaches with valid assessment of pain intensity pre- and posttreatment, and extensive follow-up. Due to the limitations of large groups of children with any particular painful disease, multisite studies should be considered. When samples are limited, single case designs may be used with multiple baseline designs to compensate for small numbers. Finally, in light of health care economics, cost analyses should be included among outcome variables as psychological strategies may prove to be extremely cost-effective in treating chronic pain syndromes.

Finally, all of the studies reviewed focused on pain as a relatively isolated symptom, ignoring the broader context of coping with a chronic condition. Among the conditions discussed in this article, studies focused on the comprehensive assessment of pain and coping in JRA and sickle cell disease indicate that pain is one factor embedded in more complicated models of stress and psychological adjustment.

Psychological literature devoted to sickle cell disease pain has focused primarily on its moderators, assessment, coping, and adjustment to illness and pain. Social support and involvement in activities have been associated with more frequent home management of sickle cell disease pain and less hospitalization (Vichinsky, Johnson, & Lubin, 1982).
Quality of pain experience in adolescents with sickle cell disease has been shown to be quantifiable, variable in intensity, and associated with socioemotional factors (Walcott & Dampier, 1990). Comprehensive studies of pain and coping in children and adolescents with sickle cell disease have been completed by Gil and her colleagues. In assessing pain coping strategies, they found that negative thinking, passive adherence, and coping attempts were related to activity and use of health care services (Gil, Williams, Thompson, & Kinney, 1991). Longitudinal data showed the above relationships to persist over time and that coping attempts were related to school, household, and social activities during painful episodes (Gil et al., 1993). Later studies indicated that pain coping strategies change over time during childhood and adolescence (Gil, Wilson, & Edens, 1997), and that there is a relationship between pain coping and psychological adjustment in this population (Thompson et al., 1994). Clearly, coping strategies and thought patterns have been shown to predict report of pain, health care utilization, and psychological adjustment (e.g., Gil et al., 1997). While these data are convincing, no published treatment studies have integrated these hierarchical models into pain management.

Among children and adolescents with JRA, Varni and his colleagues have shown statistically predictive relationships among pain experience, psychological adjustment, and functional behaviors (Thompson, Varni, & Hanson, 1987; Varni, Wilcox, Hanson, & Brik, 1988). More recently, a biobehavioral model has been invoked (Varni et al., 1996a, 1996b), examining precipitants (e.g., disease, injury, stress), intervening variables (biological predispositions, family environment, coping strategies), pain perception and pain behavior, and functional status (activities of daily living, school attendance, mood and behavioral difficulties). In a similar vein, Schanberg and colleagues assessed pain coping strategies in children with JRA (Schanberg, Lefebvre, Keefe, Kredich, & Gil, 1997) and with JPFS (Schanberg, Keefe, Lefebvre, Kredich, & Gil, 1996). In both instances they advocated the use of behavioral and cognitive interventions aimed at increasing the perception of pain control and pain coping efficacy.

Recent data on the development of pain networks and spinal cord and central nervous system mechanisms related to pain system excitation and inhibition raise a number of questions about the relationship between what traditionally has been thought of as “physiological” and “psychological” factors (Zeltzer, Bursch, & Walco, 1997). Although still somewhat speculative, a developmental biopsychosocial model that truly defines these factors as flip sides of the same coin may invoke more focused psychological and medical interventions. While certain treatment strategies have shown promise in this regard (see Bursch, Walco, & Zeltzer, 1998), empirical studies are lacking.

Finally, if we wish to extrapolate from studies of adult patients to interventions with children and adolescents, the continuity (or discontinuity) of relevant developmental and environmental factors must be evaluated. Very few longitudinal studies examining these relationships have been conducted, although theoretical perspectives on this topic have been described (Walcott & Harkins, in press).

In sum, there have been very few well-controlled empirical studies of psychological interventions for disease-related pain in children. The available data highlight interventions aimed at treating pain as an isolated symptom. Studies focusing on pain in the context of the individual coping with a chronic medical condition have added to our insights of relevant variables to be included in a comprehensive treatment approach. The major tasks that lie before us include demonstrating the emergence of pain experiences and coping over time, identifying causal relationships among variables, and validating treatment approaches to address the range of relevant variables. These prospects certainly make this field fertile ground for future research.

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References


Treatments for Disease-Related Pain


