

# Introduction to the Special Issue on Pain: From Pain to Pain-Associated Disability Syndrome

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Pediatric pain research has grown exponentially in the past decade, reflecting the increasing awareness of chronic pain as a significant health problem in pediatrics. Various epidemiologic studies across countries indicate that more than 30% of children suffer from chronic or recurrent pain significant enough to create suffering and impact function, such as school attendance, sleep, and physical and social activities (Goodman & McGrath, 1991; Kristjansdottir, 1997; Palermo, 2000; Perquin et al., 2000; Stang & Osterhaus, 1993). The most comprehensive recent epidemiologic study of pediatric pain was reported by Roth-Isigkeit, Thyen, Stoven, Schwarzenberger, and Schmucker (2005) in Germany with almost 750 school-recruited children and adolescents. In this nonclinical sample, more than 80% of the children reported having pain during the previous 3 months, with a third reporting pain for more than half a year. One third reported pain that occurred more than once a week. Pain types, in order of frequency ranging from 60 to 30%, were headache, abdominal pain, limb pain, and back pain. Half of the children with pain reported pain-related disruption of sleep, personal pleasurable activities, eating, school absence, and social activities. Pain-related functional disability increased with age. Over half of the children with pain regularly took analgesics. Gender differences emerged during adolescence, with girls reporting the use of pain medications and more pain-related functional disability than did boys. Also, adolescent girls were more likely to attribute stress and emotions as triggers for their pain than were adolescent boys, who were more likely to attribute physical causes for their pain. The above study indicates that pain is prevalent during childhood, with

prevalence increasing with age, and is associated with significant functional disability. The study also supports the sex-related differences in pain condition and functional disability that has been reported in studies of pain in adults and provides support for this sex-related difference in pain prevalence emerging during adolescence.

Pediatric psychology can play an important role in the study of pediatric pain, as this special issue demonstrates. For example, systematic assessment of pain and pain-related disruption in function is a critical first step, so that meta-analyses of studies can be carried out for the broadest and most generalizable understanding of the prevalence of pain and its consequences. The spiral of increasing pain-related disruption of function has been labeled “pain-associated disability syndrome” or “PADS” for short by Bursch, Walco, and Zeltzer (1998). Although this eponym provides a short cut to describing a phenomenon that clinicians treating children with pain disorders all recognize, more specific definitions and assessment methods that cut across existing and future studies are needed. Common assessments also allow specifications of populations and outcomes in studies of interventions, key to the prevention and reduction of pain and pain-related disability. For example, a consensus working group of investigators in adult pain developed pain-specific domains and recommended measures for each domain (Dworkin et al., 2005; Turk et al., 2003). Such trans-study consistency allows for meta-analyses across studies with better generalizability across populations.

Finally, as the study by Roth-Isigkeit et al. (2005) illustrates, pathways to pain vulnerability, including mediators and moderators of chronic pediatric pain,

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need to be identified to develop targeted interventions based on causal models of pain. The relationship between pain and both biological sex and sociologic gender role is ripe for targets to understanding some key factors in pain and pain-related disability.

The topics covered by the articles selected for inclusion in this special issue are grouped into three broad categories: conceptual articles examining cognitive and functional aspects of chronic pediatric pain, assessment of chronic pediatric pain, and interventions for pediatric chronic pain. Each of these articles addresses important aspects of the biopsychosocial model of pain and expands the scope of current research to include relatively neglected areas, including family factors, laboratory pain, and health-related quality of life (HRQOL) in children with chronic pain. Two of the intervention articles break new ground by evaluating new computer-based treatments. It is hoped that these studies will stimulate future research that will advance our understanding of the etiology and treatment of chronic pain syndromes in younger populations.

### **Conceptual Papers on Cognitive and Functional Aspects of Chronic Pain**

The overarching theoretical framework for the conceptual papers included in this issue of the journal is the biopsychosocial model (Engel, 1977), which acknowledges interdependent and mutually influencing relationships among biological, psychological, individual, family, and community factors in the onset, maintenance, and experience of illness and its impact. Within the biopsychosocial model, pain is conceptualized as a multidimensional construct, including sensory and affective dimensions as well as functional domains, that are influenced by internal states as well as interpersonal interactions (Drossman, 1996; Hyams & Hyman, 1998; Melzack, 1999; Rainville, Duncan, Price, Carrier, & Bushnell, 1997; Zeltzer, Bursch, & Walco, 1997). For example, a partial list of factors related to pediatric functional abdominal pain includes modulation of sensory transmissions in the nervous system (Hyams & Hyman, 1998), increased focus on pain-related stimuli, emotional responses to pain, and efforts to cope with pain (Zeltzer et al., 1997), and family history of pain and parental responses to a child's pain (Walker, 1999). Three conceptual articles in this collection examine psychosocial and environmental aspects of pediatric chronic pain and its impact.

Lipani and Walker (this issue) used Lazarus and Folkman's (e.g., Folkman, Lazarus, Gruen, & DeLongis,

1986) formulation of stress appraisal and coping as a framework within which to assess pain appraisal in children with chronic or recurrent abdominal pain (RAP). Primary appraisal, secondary appraisal, problem-focused coping, and emotion-focused coping were examined within this model. The primary hypothesis was that family disruption would be a function of the extent to which children perceived their abdominal pain as threatening and perceived themselves as unable to cope with the pain. The results of this application of the stress appraisal and coping model extend the literature on families of pediatric pain patients. The findings highlight the need for health care providers to assess children's and parents' pain beliefs and correct misperceptions about the medical implications of pain, while aiding children to increase their efficacy in coping with pain and for parents to support their child's adaptive efforts.

Vervoort and colleagues (this issue) investigated variability in ratings of pain, disability, and somatic complaints associated with pain catastrophizing, beyond the variability caused by negative affect. Catastrophizing, which refers to focusing on and magnifying the threat value of pain while negatively evaluating one's ability to deal with pain, has been associated with increased pain report in both clinical and experimental studies with adults and children and has emerged as one of the most robust psychosocial predictors of pain responses (Crombez et al., 2003). Vervoort et al. (this issue) assessed pain catastrophizing, negative affectivity, pain, disability, and somatic complaints in school children as well as in a clinical sample of children with recurrent or chronic pain. Catastrophizing significantly accounted for pain outcomes beyond the effects of age, sex, and negative affectivity, and mediated relationships between negative affectivity, somatic complaints, and functional disability. Clearly, these results underscore the importance of assessing pain catastrophizing and the identification of interventions to decrease the use of this cognitive style by children with pain conditions.

Logan and Scharff (this issue) studied family characteristics associated with functioning by children with clinically significant recurrent pain syndromes. In their study, children completed pain diaries and a measure of functional disability. Mothers reported on the family environment, and mothers and fathers both reported on their own psychological distress. Results indicated that family factors played a central role in determining how children functioned. These results add to the critically important understanding of systems-level influences on functional outcomes among children with recurrent

pain. The findings also contribute to model development that can lead to testing interventions designed to maximize functional abilities by incorporating family systems-based techniques into psychological interventions for children and adolescents with pain problems.

### **Pain Assessment**

The accurate assessment of children's pain and functioning is a critical aspect of evidence-based approaches to pain management. Reliable and valid measurement of the child's pain and pain-related disruptions in functioning is necessary to achieve a comprehensive picture for diagnostic purposes as well as to guide researchers and clinicians in evaluating the effectiveness of interventions. One of the main limitations of existing research is the focus on pain as the sole dimension for assessment (see Eccleston et al., this issue). A more comprehensive approach to pain assessment may include measurement of functional limitations, perhaps across a variety of contexts (e.g., school, home) as well as the impact of pain on children's HRQOL (see Connelly & Rapoff, this issue). Moreover, the development of behavioral analogs that expand the assessment of pain beyond the domain of self-report is an important advancement in the field (see Walker et al., this issue).

As outlined by Eccleston and colleagues (this issue), a plethora of measures have been developed to evaluate pain in adolescents. Yet, a systematic review of the randomized controlled trials of behavioral treatments for chronic pain reveals that the vast majority of trials did not include instruments designed to assess domains other than pain experience. In particular, Eccleston et al. note a paucity of instruments to evaluate the impact of pain on cognitive functioning and social development during adolescence, as well as the economic impact of chronic pain. The challenge for researchers will be to develop instruments that capture the full range of pain-related experience in children across broad domains of functioning, while at the same time ensuring the psychometric quality and clinical utility of such instruments. As recommended by Eccleston et al. (this issue), the development and validation of such measures should optimally be guided by theories of adolescent chronic pain.

The Eccleston review does not include the construct of HRQOL which is addressed by Connelly and Rapoff (this issue) in their evaluation of the PedsQL in children with recurrent headache. HRQOL, or the impact of a chronic condition on one's physical and emotional functioning, is a useful construct that is well-established in

the fields of medicine and health service research. HRQOL may be assessed generally or specifically (i.e., in relation to specific conditions). Notably, scores on widely used HRQOL measures are generally considered among the most robust predictors across a variety of conditions and health outcomes (Ware, Kosinski, & Keller, 1994; Ware, 2005). However, the concept of HRQOL has not been extensively researched in relation to pain, particularly in pediatric populations. Thus, the Connelly and Rapoff study represents an important addition to the field by providing evidence to support the utility of a HRQOL measure in children with recurrent headache. It is hoped that other investigators will follow their lead by incorporating HRQOL measures in the assessment of pediatric chronic pain outcomes.

Although self-report of the pain experience is generally considered the "gold standard" for pain assessment (McGrath et al., 1990), at least for pediatric populations who can provide such verbal or written reports, Walker and colleagues (this issue) illustrate the utility of a laboratory-based behavioral assessment task in children with RAP. Their contribution highlights the importance of developing valid and reliable measures of children's pain experience that are based on both conceptually and clinically relevant models of pain. The systematic and rigorous methodology employed by the Walker group provides an excellent illustration of the type of research that will advance our understanding of the mechanisms that underpin recurrent pain in children. Laboratory pain analogs, such as that developed by the Walker group, allow researchers to test hypotheses regarding such aspects as individual differences and intervention effects without confounding variables (e.g., variations in duration/intensity) that are inherent to clinical pain episodes and "real world" medical procedures. Walker and colleagues have thus furnished us with a valuable tool that will promote further research dedicated to the understanding of RAP.

### **Interventions for Pediatric Chronic Pain**

Preliminary research reveals that cognitive-behavioral and relaxation interventions can be highly effective for pediatric chronic pain (Eccleston, Yorke, Morley, Williams, & Mastroiannopoulou, 2003; Holden, Deichmann, & Levy, 1999; Janicke & Finney, 1999; Weydert, Ball, & Davis, 2003). The 2001 American Pain Society (APS) position statement on pediatric chronic pain (American Pain Society, 2001) called for research to expand the literature on evidence-based treatments in chronic pediatric pain, with specific attention paid to issues, such as

Careful descriptions of the study samples, detailed documentation of the interventions, the inclusion of family variables, and outcome variables that include both pain and functioning. The position statement also emphasizes the need for patient and family interventions that optimally address underlying pain mechanisms, symptom-focused management of pain, sleep disturbance, and affective symptoms, and pain-associated disability. Since the publication of the position statement, there has also been increasing access and interest among children and adolescents in computer-based activities, creating unprecedented opportunities to test Internet and CD-ROM based interventions that require minimal therapist contact and potentially allow for outreach to geographically remote locations.

This issue includes the results of three cognitive-behavioral intervention studies that move the field forward by providing many of the details called for in the APS position statement, by including caregivers in the assessments and interventions and by testing two novel computer-based treatments. All three interventions resulted in decreased reports of pain and the “in-person” intervention also had a significant impact on functioning.

Treatment outcome studies with adults with fibromyalgia have underscored the efficacy of cognitive-behavioral strategies, and yet there has been very little research with pediatric fibromyalgia samples. Degotardi and colleagues (this issue) describe the development of and report on the efficacy of an 8-week cognitive-behavioral intervention for juvenile fibromyalgia. Their well-described, manualized intervention is geared toward children with fibromyalgia and their parents and targeted pain management, psycho-education, sleep hygiene, and activities of daily living. Specific skills were taught to children, including cognitive restructuring, thought stopping, distraction, relaxation, and self-reward. The investigators collected both parent and child data and measured multiple outcomes, including pain, somatization, anxiety, fatigue, sleep quality, quality of life, and both physical and psychosocial functioning. Posttreatment improvements were detected on all measures of physical symptoms, psychological symptoms, and functioning.

Breaking new ground, Hicks, von Baeyer, and McGrath (this issue) compared the efficacy of a 7-week distance treatment delivered via Internet and telephone to a standard medical care waitlist. Youth with chronic headaches and/or abdominal pain were randomized to a web-based cognitive-behavioral treatment that included pain education, relaxation techniques and cognitive strategies, and positive lifestyle choices. Parents received

web-based education on encouraging healthy behavior. Also included were relaxation tapes and email and telephone contact. The investigators collected both parent and child data and measured pain and quality of life outcomes. They found significant between-group differences with more of the children in the web-based intervention experiencing clinically significant improvement in pain at the 1- and 3-month follow-up assessments.

Also breaking ground, Connelly and colleagues (this issue) compared the efficacy of an adjunctive 4-week CD-ROM pain management program to a standard medical care waitlist. Participating children attending a neurology clinic for recurrent nonmalignant headache; all followed the advice of their neurologists and were subject to randomization to the computerized intervention, which included modules on education, relaxation, thought changing, and pain behavior modification. They were also contacted by telephone weekly. The pain behavior modification module required the child, along with a caregiver, to develop and implement an active pain-coping plan. The investigators collected both parent and child data and measured pain and headache-related disability outcomes. They found greater reductions in headache variables among the treatment group children through the 3-month follow-up.

Taken together, the above studies advance our database for evidence-based treatments in chronic pediatric pain. They also pose a challenge to interventionalists to develop a computer-based treatment that also measurably impacts patient and family functioning and quality of life. The emerging intervention research supports the APS position statement, 2001 (American Pain Society, 2001), that calls for an end to mind-body dualism in favor of a biopsychosocial framework for the assessment and treatment of pediatric chronic pain.

## Conclusion

Pediatric chronic and recurrent pain presents a dilemma for the assessment and treatment of the many children in pain who are caught in a biomedical model-driven health care system. After trying to come up with a “cause” for the pain based on organ system, infectious, immune, or metabolic pathology, as examples, many physicians assume that the pain is “psychological.” Thus, referrals are made to the mental health provider to treat this presumed psychological etiology for the pain. However, unless the biopsychosocial nature of pain is successfully explained and a paradigm shift in parents’ understanding of pain is created, the parents who do not accept this pain model will seek medical care elsewhere

(Crushell et al., 2003). Thus, the child with chronic pain, especially when function is poor, may undergo evaluation by multiple physicians and many medical tests. These are the children often referred to pediatric pain clinics. When a successful referral is made and a psychological intervention results in a reduction in the child's pain and/or increase in functioning, the physician often assumes that the psychological "diagnosis" is correct. Perhaps for this reason, the adoption of biopsychosocial diagnostic explanations by primary and specialty care pediatricians to describe chronic pain has lagged behind the research supporting this model.

The dualism of mind versus body has also had an enormous negative impact on our health care system, which still operates to a large degree within the biomedical model rather than the biopsychosocial model. This limited view of pediatric pain results in inattention to the many psychological risk factors for both morbidity and mortality, especially when traditionally defined "disease" is identified, and typically ignores the psychosocial pathways that lead to over-utilization of medical evaluations and unnecessary intervention. The biomedical model of health care has advanced to the point of hospitals systematically assessing pain with faces or rating scales to maintain accreditation. However, this pain assessment system focuses only on the immediate amount of pain intensity. As the reports in this issue indicate, this limited view of pain assessment results in an inadequate picture of the full experience of pediatric pain and its impact on the child and family. That is, our health care system makes little use of the developing tools, as demonstrated in this issue, to assess pediatric chronic pain. The pediatric health care system also does not fully utilize, as an integral part of the medical care of the child, the many well-documented psychological interventions for chronic pain. In addition, pathways to the development of chronic pain are confusing within the biomedical model and are best understood within the biopsychosocial model. This special issue provides support for understanding pathways to chronic pain, evidence-based models of pediatric pain assessment, and novel interventions for chronic pain. As Roth-Isigkeit et al. (2005) demonstrates, there are many children with chronic or recurrent pain whose parents seek medical help and many more whose parents do not, but who suffer nonetheless. Dissatisfaction with medical care for children with chronic pain has motivated many parents to look to the lay press for answers (Zeltzer & Schlank, 2005). This issue provides further support for abandoning mind-body dualism in favor of the biopsychosocial model of evaluation and treatment of children with chronic pain.

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