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Commentary

Shaping the future of research on chronic pain in children

Anna Huguet, Patrick J. McGrath, Jennifer Stinson, Christine T. Chambers and Jordi Miró

Chronic pain is becoming recognized as a significant health problem in children. Moreover, pain during these early ages is at high risk of persisting into adulthood with associated morbidity. Understanding how pain becomes chronic and disabling is essential in order to identify children at risk, and for implementing effective prevention and interventions. There have been increasing efforts to identify risk factors for the development and maintenance of pain and related disability in children. However, research has traditionally largely been without guidance from relevant specific theoretical models beyond the general biopsychosocial model of pain (Turk, 1996). This commentary will briefly describe the steps which have been taken to gain knowledge from a biopsychosocial perspective by critically evaluating the current state of use of a theoretical approach to guide research and give recommendations for future research.

Introduction

Chronic pain is a significant health problem in children. Recent epidemiological studies in several countries estimate that the prevalence of chronic pain in children ranges from 15% to 38% (e.g. Stanford et al., 2008). Chronic pain can negatively impact all aspects of functioning including physical, psychological, social, and role functioning (Palermo, 2000). Epidemiological studies report that approximately 5% of children in the general population have moderate or severe chronic pain-related disability (e.g. Roth-Isigkeit et al., 2005; Huguet & Miró, 2008), which is defined as difficulty that impairs the ability to perform daily activities due to his/her health status (Walker & Greene, 1991). Chronic pain in children may also negatively affect the child's family (e.g. Jordan et al., 2007) as well as society in general by increasing health care costs and reducing work productivity due to time off to care for the child suffering chronic pain (e.g. Sleed et al., 2005). Understanding how pain becomes a chronic disorder as well as how it causes disability is essential in order to prevent this significant health problem and its negative consequences. Unfortunately, neither the development nor the trajectory of pediatric chronic pain is satisfactorily understood.

State of the art

The biopsychosocial model (Turk, 1996) suggests that chronic pain and related disability result from the interaction of biological, psychological and sociocultural factors, yet it does not provide a detailed and precise account of the underlying mechanisms. Research using this general biopsychosocial model has focused on identifying various factors that are predictive of pediatric chronic pain and related disability. Researchers have examined many risk factors at different levels and units of analysis, with relatively limited attention paid to resilience factors, such as positive expectations about the ability and responsibility to exert control over pain (e.g. Huguet et al., 2009). Table 1 provides a summary of the most commonly studied prognostic factors.

The identification of prognostic factors using an empirical approach is a good starting point for understanding the development and trajectory of chronic pain and pain-related disability. However,

Table 1
Potential risk factors that have been under investigation examining their role in either chronic/recurrent pain or functioning (i.e. defined in terms of functional disability or quality of life)

	Factor domain	Prognostic factors	Examples
Internal personal characteristics of the child	Sociodemographic characteristics	Age, gender, socioeconomic status, ethnicity	Larsson & Sund, 2005
	Pathophysiological characteristics	Heritable and genetic factors	El-Metwally et al., 2008
	Anthropometric characteristics and physical condition	Body mass index, weight, height, fatigue, mobility of joints	Bejia et al., 2005
	Health-related behaviors and lifestyle	Smoking, physical exercise, sleep, use of alcohol or other drugs, sedentary lifestyle	El-Metwally et al., 2004
	Pain characteristics	Pain frequency, pain intensity, duration of pain episodes	Lynch et al., 2006
	Psychological characteristics	Depression, psychological distress, pain coping responses	Gauntlett-Gilbert & Eccleston, 2007
External personal characteristics of the child	Parental and family characteristics	Parental pain, parental stress, family functioning	Logan & Scharff, 2005

this atheoretical approach is not generally helpful in identifying potential mechanisms that would link these factors to the development and maintenance of chronic pain and related disability. Prognostic factors that are identified empirically should be incorporated into new or already existing theoretical pain models to gain a better understanding of the development and trajectory of the problem. This is currently difficult to do because the mechanisms through which a factor increases or decreases the risk of chronic pain or related disability are still unknown. In the past, most researchers have merely observed and collected data on potential prognostic factors derived primarily from cross-sectional studies, without building and testing theory and uncovering (or exploring possible) mechanisms. Such a basic empirical approach is not surprising since the understanding of pediatric chronic pain is at an early stage of development in which little unifying theory exists to serve as a guide (e.g. Palermo, 2000; Stanford et al., 2008).

Atheoretical fact gathering will clearly not improve our understanding of the phenomena or guide clinical practice. For instance, studies have shown that pain is influenced by a child's sex: girls are more likely to suffer chronic pain and related disability than boys (Unruh & Campbell, 1999). However, without considering the underlying mechanisms, we cannot explain the link between pain and a child's sex and provide a focus for treatment. A recent review by Fillingim and colleagues (2009) synthesizes the literature on sex, gender, and pain and suggests a number of potential mechanisms through which social and psychological processes (e.g. coping processes and catastrophizing) are likely to contribute to the repeatedly observed sex differences in pain. This type of sophisticated conceptualization could help

focus our attention more productively on possible sex-linked mechanisms of action.

The primary goal of a theory (Bacharach, 1989) is to propose and examine how, when and why factors contribute to an outcome such as chronic pain. A constant interplay between empirical work and theory-driven analysis is what is most needed to advance our knowledge (Gelso, 1991). This knowledge would allow scientists and clinicians to design and deliver effective early protective and preventive strategies with children at risk of chronic pain and associated disability. A good theory must be unique and differentiated from other existing theories. It cannot be replaced by another theory unless the new theory is superior in its virtues. It must be generalizable and applicable to different settings. A good theory must be fertile by expanding the area of investigation into new conceptual areas. It must be parsimonious and internally consistent by providing an adequate explanation of all the relationships between the variables. It also must be empirically risky and abstract, that is, it must exist, to a certain extent, independently of time and space (Quine & Ullian, 1980). Very few researchers in pediatric pain have adopted a theory-driven approach by formulating and testing theories in an attempt to understand pediatric chronic pain (Minuchin, 1977; Lewin & Dahl, 1999; Walker, 1999; Palermo & Chambers, 2005). However, there are already promising examples of theory-based accounts of the development and trajectory of chronic pain in the general population using operant conditioning (Fordyce et al., 1968), modeling (Bandura, 1965), fear-avoidance (Vlaeyen et al., 1995), perceived self-efficacy (Bandura, 1977), childhood traumatic stress (Charmandari et al., 2003; Goffaux et al., 2008), attachment (Bowlby, 1969), disuse (Bortz, 1984), and central sensitization (Woolf, 2011). The evidence regarding the validity of these theories is different across theories. Some are well-supported by evidence (the fear avoidance model, theory of primary central sensitization), whereas others are not (attachment theory, theory on childhood trauma). These theories, which are not mutually exclusive, could provide useful templates to further our understanding of chronic pain and related disability in children. None of the theories individually capture the entire complexity of the phenomena. However, they could be combined to better understand chronic pain and related disability, since each theory may contribute a unique piece to the pain puzzle.

Recommendations and directions for future research

We offer the following directions for future research to advance our understanding and build testable theories of chronic pediatric pain.

Testing of existing theories. Testing is suggested for theories that have been shown to be useful with the general population but need to be validated with children to determine their appropriateness, and theories that have already been formulated in the field of chronic pediatric pain and have yet to be fully tested. It is important to reformulate theories that have been shown to be useful in adults in order to make them developmentally oriented (at the biological, cognitive, emotional, or social level) when exploring the applicability of these theories towards a younger population (Walco, 2004). In the process of testing existing theories, both comprehensive, integrated theories and micro theories have merit in guiding research as long as each key construct in the theory is operationalized.

Use of different, more sophisticated approaches and methodologies to test specific mechanisms informed by theories. Studies contrasting theories often will benefit from sophisticated quantitative techniques such as structural equation modeling using longitudinal data from prospective cohort studies (e.g. Nelson et al., 2008). Research that has been conducted in this area has tended to be descriptive and correlational, and therefore, experimental methodology should be used in the future. Theory-based experimental intervention studies such as randomized controlled trials are useful to test and refine theories of the etiology and continuity of chronic pain and disability. These studies can provide valuable information about the mechanisms of change (e.g. how the treatment yields positive outcomes) and about the boundary conditions of effectiveness (e.g. who benefits, who does not, and why).

Increased research in key areas. More research is needed examining what resilience factors protect against chronic pain and pain-related disability. In addition, studies concerning the potential transition from acute to chronic pain are needed, as this is a particularly fertile time period to examine mechanisms of chronic pain and disability.

Systematic reviews of prognostic factors. These can synthesize the most relevant factors available in the literature and be instrumental in elaborating new or better working hypotheses about underlying principles or apparent relationships of the observed phenomena (e.g. Forgeron et al., 2010; Gieteling et al., 2011). Systematic reviews should take into consideration that different types of chronic pain in children and youth may have different prognostic factors, and prognostic factors of chronic pain and disability may also differ (Miró et al., 2007).

To sum up, the application of theory-driven approaches is a step in the right direction to a better understanding of chronic pain and related disability in children.

Anna Huguet, PhD
IWK Health Centre and Dalhousie University, Halifax, NS, Canada
email: anna.huguet@dal.ca

Patrick J. McGrath, PhD
IWK Health Centre and Dalhousie University, Halifax, NS, Canada

Jennifer Stinson, PhD
The Hospital for Sick Children, and Lawrence S Bloomberg Faculty of Nursing, University of Toronto, Toronto, ON, Canada

Christine T. Chambers, PhD
IWK Health Centre and Dalhousie University, Halifax, NS, Canada

Jordi Miró, PhD
Unitat per a l'Estudi i Tractament del Dolor, Centre per a l'Avaluació i la Mesura de la Conducta, Institut d'Investigació Sanitària Pere Virgili, Universitat Rovira i Virgili, Tarragona, Catalonia, Spain

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