

# Understanding chronic pain and disability in young people: a study with Catalan schoolchildren

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## INTRODUCTION

Pain is defined as *an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage* by the International Association for the Study of Pain (IASP, Merskey and Bogduk, 1994; p. 210). Such a definition parallels, to some extent, the widely accepted and used biopsychosocial model of pain; it goes beyond the biomedical perspective which dominated the field years ago. Pain is much more than a simple, straightforward, sensory experience. Pain results from the interaction of multiple factors, physical, as well as emotional, cognitive, and behavioral/contextual.

Pain is unpleasant but necessary. Pain can have biological value, it can be a sign that something dangerous is occurring in the body. That is, pain can warn us of actual or impending tissue damage and motivate the individual to escape or avoid further harm. Indeed, acute pain is defined by the IASP as pain that usually has an identifiable relationship to injury or disease and it has a recent onset and probable limited duration (Merskey and Bogduk, 1994). However, there are times when the pain experience is unnecessary, it happens when pain has lost its value as a signal of danger (see Mitchell and Boss, 2002 for a description of consequences of pain in children). This is what typically occurs in chronic pain problems. Chronic pain is defined by the IASP as pain that persists beyond normally expected healing, as quantified through medical experience (Merskey and Bogduk, 1994); however, it might also occur without a physical injury (American Pain Society, 2001); and it is experienced as repeated (i.e., recurrent) or continuous (i.e., persistent) episodes (American Pain Society, 2001). Chronic pain does not have any useful biological purpose, it can cause an enormous amount of suffering both to the person in pain and to his/her significant others. Chronic pain is considered to be a public health issue (Crombie, et al., 1999). Recent epidemiological studies conducted in Spain (Català et al., 2002 and Miró et al., 2007) with adult and older population - persons 65 years or older- respectively conclude that chronic pain is an important problem.

## 1. CHRONIC PAIN IN CHILDREN

Pain has attracted the attention of humans since the earliest recorded history (see Sabatowski et al., 2004). However, pain in children was not well researched until the 1970s. Fortunately, this situation has changed dramatically. Over the last 25 years, there has been an increased interest in the study of pain in children and adolescents. Experts have come to acknowledge that chronic pain is a real problem for children, as it is with adults. Research specifically considering pediatric chronic pain has increased over the last decades. More resources are also being invested into its treatment, although there is an almost unanimous position claiming that research on pain in children and adolescents has not yet been effectively transferred to clinical practice (Eccleston, 2005).

However, the interest in chronic pediatric pain is not so evident everywhere. For example, as Reinoso-Barbero acknowledged (2003), chronic pediatric pain still seems to be the *poor brother* of adult pain research in Spain. That is, despite the significant number of children in need of treatment, few clinicians and researchers are giving the attention, time and effort, this problem deserves. Moreover, an interdisciplinary approach to the study and treatment of chronic pediatric pain in Spain is almost nonexistent. Thus, many children suffering from chronic pain do not receive the most appropriate treatment because neither researchers nor clinicians are familiar with pediatric chronic pain conditions, much less with recommended assessment tools and intervention strategies for children with chronic pain.

## 2. ASSESSMENT

### 2.1. DOMAINS

A thorough assessment is fundamental to understand a child's pain experience. The assessment process should be guided by solid theoretical grounds.

The Gate Control Theory (Melzack and Wall, 1965), has had a tremendous impact in the study of pain. It has been instrumental in changing the conceptualization of pain, and promoting interdisciplinary treatments. The Gate Control Theory (see Melzack and Casey, 1968; Melzack and Wall, 1965,

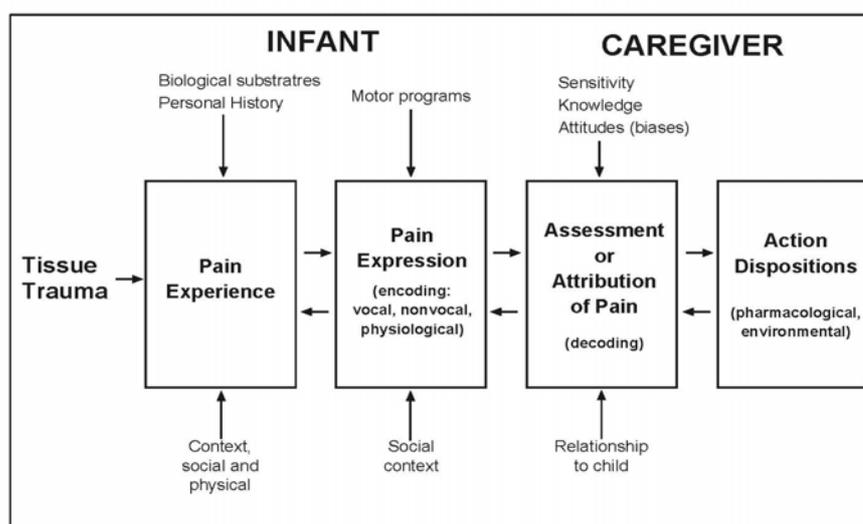
1982) postulated that pain experience needs to be approached from a multidimensional perspective. According to this theory, both somatic sensory input and brain processes are involved in generating the pain experience. The original theory advocated the existence of a spinal gating mechanism which responded to signals descending from the brain by either opening, to allow more somatic sensory input to access the brain, or closing, to reduce the arrival of sensory input to the brain. Psychological factors were thought to be an integral part of pain processing because they were thought to be able to influence sensory input. Cognitive-evaluative factors, motivational-affective factors, and physiological processes were all thought to affect the pain experience. More recently, the Neuromatrix Theory (Melzack, 1999), which has been recognized as an evolution of the Gate Control Theory, postulated that a widely distributed neural network is involved in the pain experience instead of just a gating process.

New psychological theories appeared following the Gate Control Theory, they introduced alternative ways of thinking about chronic pain, and challenged the biomedical model, such as Melzack and Wall had done before. One of these theories was the one proposed by Fordyce (1976). Fordyce's seminal work played a major role in identifying the importance of behavioral factors in the development and maintenance of chronic pain problems. He offered an important distinction between 'pain perception', which is a private experience, and 'pain behaviors' (e.g., limping, grimacing, or groaning), which are observable means of communicating pain and suffering. Fordyce argued that pain behavior, like any other behavior, is subject to the principles of operant conditioning and learning. That is, pain behaviors are influenced by positive and negative consequences, which determine the likelihood of pain behaviors occurring in the future. Pain behaviors that yield positive outcomes are more likely to occur in the future, and vice versa. Fordyce's operant learning theory as applied to pain can help us understand why pain behaviors can persist over time even after the injury has healed.

Turk, Meichenbaum, and Genest (1983) were among the first to recognize the importance of cognitions in the development and persistence of chronic pain problems. Individuals' beliefs about their ability to control pain, their ability to cope with pain, their expectations about coping outcomes, and their

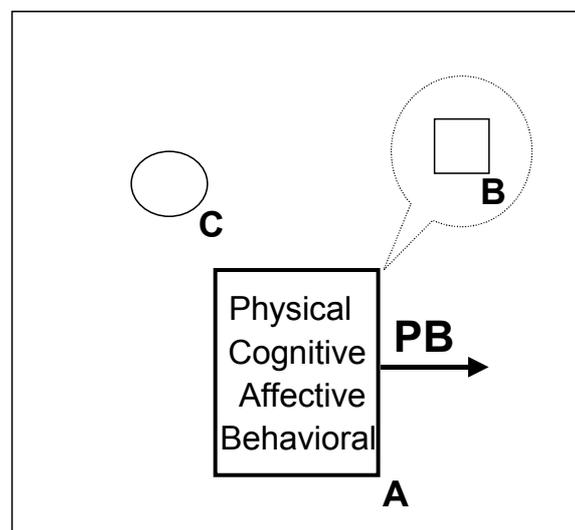
erroneous negative beliefs about pain and pain-related disability were all understood to be factors that influence pain behaviors.

These general and theoretical models have been *translated* into the pediatric field (e.g., Brush et al., 1998). Indeed, there are some conceptual models that address the influence of psychosocial factors on children's pain experience. For example, Kazak (1989), and Kazak and Kunnin-Batson (2001) recognize contextual influences in their social-ecological model outlined by Bronfenbrenner (1979). Kazak and colleagues suggest that multiple interdependent systems in which the child functions have to be considered if one is to understand how children adapt to and cope with a chronic illness. These systems are labelled 'micro' 'meso' and 'exo' and include the child's family, healthcare system, community, and culture. Other authors have also recognised the influence of the context on the children's pain experiences. Craig (see for example, Craig et al., 1996, 2002; Craig and Pillai Riddell, 2003), for example, has developed a social communication model of pain. This model characterizes social influences as experiential, encoding, and decoding. The model encompasses a range of mechanisms responsible for these influences besides the operant conditioning ones. Within this conceptual framework, parental guidance, observational learning (e.g., family role models), promotion of self-regulation, punishment and reinforcement are considered possible modes of intergenerational influence. *Figure 1* displays the model.



**Figure 1. Social influences on childhood pain (reprinted with permission from Craig et al., 2002)**

Therefore, current models do recognize that pain is a multidimensional experience, the result of an interactive set of factors. And, when we want to assess pain experience the perplexity of this experience needs to be addressed. Based on these models, Miró (1998, 2003) suggests that the assessment of people suffering from chronic pain problems should be multicontextual and multimethodological, that is multiple levels and units of analysis should be assessed (see *Figure 2*). The model put forth by Miró was formulated on the basis of Hawkinshire's (1967, 1994) Thoughts-Feelings-Behaviors model and Murray's suggestions about the assessment of human beings (see Murray, 1948). In Miró's model, pain is conceptualized as a multifactorial experience in which factors at the physical, cognitive, affective, and behavioral level interact and determine the characteristics of the pain experience. Following Miró's suggestions, a complete and sophisticated assessment is required in order to comprehensively assess the person, rather than just the 'pain problem'. The American Pain Society has a position statement reinforcing this concept of holistic assessment in the context of pain, see: <http://www.ampainsoc.org/advocacy/pediatric.htm>. For example, the American Pain Society emphasizes the importance of collecting data about recent stressful life events when assessing pain in children.



**Figure 2.** Units (child-individual, dyad, context) and levels of analysis (physical, cognitive, affective, and behavioral) for the multicontextual assessment of children in pain. A, B, C: Social configuration, participants in the child's physical context (e.g., primary care service). A: child; B: health care professional; C: the child's mother, PB: pain behaviors (adapted from Miró (2003), reproduced with permission).

Thus, we need assessment instruments that allow us to study many domains of perception and functioning when we want to understand a child's pain experience. Nowadays, we have a variety of measures; however, there is not a consensus about which domains should be assessed or which instruments should be used when working with children in pain. Without a standardized protocol for pediatric pain assessment, it will be difficult for knowledge about pediatric pain and appropriate pain assessment techniques to be effectively transferred into clinical practice.

Recently, the Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (PedIMMPACT, McGrath et al., 2006) was launched. The mission of this initiative is to develop consensus on recommendations for improving the design, execution, and interpretability of clinical trials for treatment of pediatric pain (both acute and chronic). Academic researchers, government funding and regulatory agencies, and the pharmaceutical industry all participated in this initiative and contributed to determining which domains and measures should be used in pediatric pain assessment (see *Table 1* for their recommendations in chronic pain trials).

**Table 1. Core outcome domains and measures recommended for consideration in clinical trials of chronic and recurrent pediatric pain**

<i>Domains</i>	<i>Measures</i>
Pain intensity	Poker Chip Tool (Hester et al., 1990); Faces Pain Scale-Revised (Hicks et al., 2001), visual analogue scale (Scott et al., 1977)
Emotional functioning	Children's Depression Inventory (Kovacs, 1981), Revised Child Anxiety and Depression Scale (Chorpita, 2000)
Physical functioning	Functional Disability Inventory (Walker and Greene, 1991) PedsQL (Varni et al., 1999)
Role functioning	School attendance, PedMIDAS (Hershey et al., 2001, 2004)
Global judgment of satisfaction with treatment	-- (non-specific measure)
Symptoms and adverse events	-- (non-specific measure)
Sleep	-- (non-specific measure)
Economic factors	-- (non-specific measure)

## 2.2. AVAILABLE MEASURES

In this section, a brief description of the best known measures available to assess children in pain is presented.

There are many pediatric pain assessment measures: physiological, behavioral, and self-report (for extended reviews, see Finley and McGrath, 1998; McGrath and Gillespie, 2001). There are a number of **physiological measures** of pain, for example: heart rate, and blood pressure. Despite the lack of response bias and the apparent objectivity, no single physiological measure has been shown to be ideal. Many physical measures vary not only according to pain but also to emotional states, temperature in the environment and body movement, for example. Furthermore, there is great interpersonal variability on how individuals respond to pain physiologically. Overall, physiological measures are sensitive but not specific, in that they simply cannot discriminate well between the responses to pain and other forms of stress in the body.

**Behavioral measures** of pain are based on the observation of non-verbal clues about specific types of distress behaviors (e.g., vocalization, facial expression, and body movement) that have been associated with pain to estimate the child's pain. Examples of major behavioral measures available for infants and children are: FLACC: Face, Legs, Arms, Cry, Consolability (Merkel et al., 1997), CHEOPS: Children's Hospital of Eastern Ontario Pain Scale (McGrath et al., 1985), PPPM: Parents' Postoperative Pain Measure (Chambers et al., 1996), the COMFORT Scale (Ambuel et al., 1992), the Toddler-Preschooler Postoperative Pain Scale (Tarbell et al., 1992) (for a detailed review of behavioral measures of pain see von Baeyer and Spagrud, 2007) Although these measures provide very valuable information, they are not very appropriate for assessing recurrent and chronic pain because they are vulnerable to habituation, that is, behavioral signs of chronic pain tend to dissipate as time passes, so it is difficult to observe reliably these behavioral signs in cases of chronic pain. Besides, pain behaviors, as any other behavior, are contextually driven, that is, behaviors are influenced by the child's context.

**Self-report measures** of pain tend to be favoured over other types of pain measure for use with children capable of verbal communication. There are several advantages in using self-report measures. Pain is a subjective

experience and self-report measures ask for the individual to articulate their pain experience themselves. Moreover, they are methodologically convenient. However, self-report measures also have limitations. First, they are dependent on the child's cognitive and communicative competence. Second, child's reports are also influenced by their context. In this line, it is possible for the informant to respond in a biased fashion (e.g., minimize their pain). Third, they can provide data based on the assessor's interests. Finally, they may lead to memory bias since children might be requested to recall pain experience in the past.

The next section presents several specific self-report measures in greater detail.

The sensory intensity component of pain (pain intensity) is typically the most frequently used pain descriptor when pain is assessed. Several self-report measures of pain intensity are available: (1) visual analogue scales (e.g., Coloured Analogue Scale, McGrath et al., 1996), (2) faces scales (e.g., Wong-Baker FACES Pain Rating Scale, Wong and Baker, 1988; FPS-R, Bieri et al., 2001), (3) numerical rating scales, (4) verbal rating scales; (5) symptom checklists; and (6) item-based rating scales (e.g., Poker Chip Tool, Hester et al., 1990) (For a review of self-report measures of pain intensity see Stinson et al., 2006). In general terms, children are asked to rate how much pain they feel when they are in pain on a scale.

Several self-report measures for the assessment of factors relating to pain (i.e. pain-related emotions, cognitions, and behaviors) have been also developed. For example, the Pain Coping Questionnaire (Reid et al., 1998), the Coping Strategies Questionnaire (Gil et al., 1991, 1993), and the Waldron-Varni pediatric pain coping inventory (Varni et al., 1996), have been developed to assess pain coping. Scales developed for use in the adult population to assess other relevant factors have also been adapted for use with youth, for example, the Survey of Pain Attitudes (see Engel et al., 2007), which assesses general pain beliefs, or the Pain Catastrophic Inventory (see Crombez et al, 2003), which is used to assess extreme negative predictions about pain and its consequences.

Measures designed to assess domains other than the specific pain experience are also available to better understand the lives of children with chronic pain problems. For example, chronic pain affects various aspects of a

child's well-being, such as his/her physical, psychosocial and social life (Hunfeld et al., 2001; Ostkirchen et al., 2006; Palermo, 2000), and many different instruments are available for measuring health-related quality of life in children (see Creemens et al., 2006; Davis et al., 2006; Eiser and Morse, 2001; Harding, 2001 for some recent reviews). The measures of quality of life cover different areas of functioning of the child (e.g. physical, emotional, social, or school functioning). These measures are becoming increasingly popular as outcome measures in the health field. Moreover, pain related disability measures are also available (e.g., the Functional Disability Inventory, Walker and Greene, 1991). They assess how much difficulty the children have had to deal with their daily activities due to their physical conditions.

Because of the importance of contextual influences (generally speaking, the influence of the family is most often studied) on pediatric pain, clinicians and researchers are developing measures to elucidate some of the modes of influence of parents on a child's pain experience. At this point, there are a few self-report measures available to assess parents' reactions to child's pain: for example, the Illness Behavior Encouragement Scales (Walker and Zeman, 1992), the Adult Responses to Children's Symptoms (Van Slyke and Walker, 2006).

A fundamental issue in pediatric pain assessment is selecting a measure that is accurate. When conducting treatment, clinical judgment about the therapeutic approach and treatment effectiveness is often informed by results obtained with pediatric pain assessment measures. When conducting research, conclusions are also drawn from results obtained through these measures. Thus, researchers and clinicians do strongly support the need to evaluate the accuracy of the measures available (Cohen et al., 2006). The criteria required for a pain measure to be deemed 'accurate' are similar to those required for any measuring instrument. In order to be accurate, a pain measure should be reliable, valid, practical and versatile (Johnston, 1998). That is to say, the measure must unequivocally provide consistent and trustworthy pain ratings regardless of time of testing, the clinical setting, or the evaluator. The measure must unequivocally assess a specific dimension of pain. Moreover, the administration of the measure to assess the child's pain should be as easy as possible.

So far, none of the pediatric pain measures described above have been adapted for use with Catalan-speaking children. This is an important empirical and clinical issue that needs to be dealt with in order to ensure that Catalan-speaking children receive the best pain assessment and treatment possible.

### **3. EPIDEMIOLOGY**

Conducting epidemiological studies is one way to contribute to our understanding of children's experiences of chronic pain. Epidemiological studies allow us to estimate the prevalence of a problem in a given population, describe unbiased relationships between a problem and an outcome, as well as to describe causal relationships, if longitudinal designs are used.

If we review available epidemiological research, there can be no doubt that chronic pain in children and adolescents is a common problem. Its wide prevalence, as well as its impact on children's quality of life makes it a significant public health problem.

The prevalence of chronic pain in children is estimated to range from 15% to 30% (e.g. Bandell-Hoekstra et al., 2001; Brattberg, 1994; Laurell et al., 2005; Perquin, 2000; Roth-Isigkeit et al., 2004). Difference prevalence rates across existing studies may be explained by methodological differences (for a detailed explanation about these issues see Goodman and McGrath, 1991 or McGrath, 1999). Headache, abdominal pain, and low limb pain are the most common pain problems among children (Perquin et al., 2000; Roth-Isigkeit et al., 2005). However, it is also common for a child to have more than one type of pain (Boey and Goh, 2001; Egger et al., 1999; Ghandour et al., 2004; Larsson and Sund, 2005). More girls report suffering from chronic pain than boys, and as girls become older, they are more likely to report chronic pain (Martin et al., 2007; Merlijn et al., 2003; Perquin et al., 2000; Ramchandani et al., 2005).

Most published epidemiological studies report whether or not children have chronic pain, but they do not systematically assess the severity of chronic pain on children's lives. The mere presence (or absence) of chronic pain is not a satisfactory measure, given the high prevalence of recurrent and chronic pain problems in healthy schoolchildren. Research shows that the impact of chronic

pain cases varies widely across individual children. For example, studies conducted on clinical samples, which are small and highly selective groups of children with chronic pain, show that chronic pain can produce considerable disruption on children's daily activities. These children often miss school, give up or reduce the time spent on their extracurricular activities and with friends, suffer sleep difficulties, as well as show anxiety and depressive symptoms (Palermo, 2000; Walters and Williamson, 1999). In these cases, pain also negatively impacts on the whole family (Eccleston et al., 2004; Smart and Cottrell, 2005), which reciprocally affects children's pain experience. For example, parents of these children, can feel highly distressed and by their inability to alleviate their child's pain. Fortunately, not every child in the general population suffering from chronic pain reports such a dysfunctional life; although, in general, it is true that the functioning of children with chronic pain is worse than the functioning of their healthy peers (Connelly and Rapoff, 2006; Hunfeld et al., 2001). To date, however, there is not a single accepted standard system to describe the severity of pain syndromes for children and adolescents.

While there are many studies assessing the personal costs of chronic pain problems, there are just a handful focusing on the economic costs of chronic pain. However, the existing ones estimate that recurrent and chronic pain costs to society can be quite significant. Slead et al. (2005), for example, examined a clinical sample of adolescents (attending to either specialised pain management units or outpatients clinics). They estimated that the cost-of-illness of an adolescent with chronic pain to United Kingdom is approximately 8000 pounds per year.

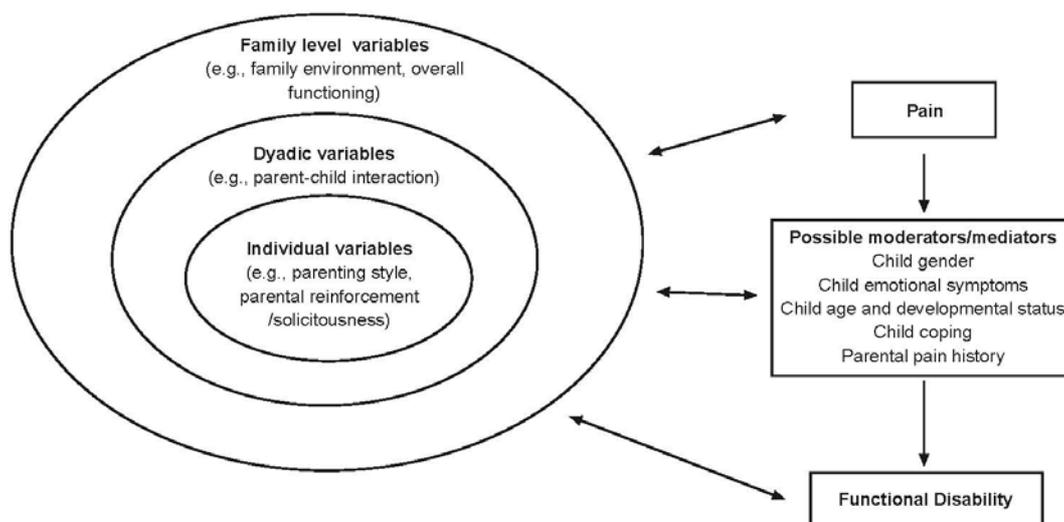
There is very little data on the prevalence of recurrent and chronic pain in Spanish children. As far as we know, two studies have been published reporting some epidemiological data. Kovacs et al. (2003), in a study with schoolchildren (ages 13-15 years) on the island of Mallorca (n=16394), showed that the lifetime prevalence of low back pain was 50.9% for boys and 69.3% for girls. Point prevalence (7 days) was 17.1% for boys and 33% for girls. Barajas et al. (2001) reported that 27% of a convenience sample of 571 schoolchildren aged between 6- and 15-years-old reported chronic pain.

## 4. PREDICTORS OF CHRONIC PAIN AND FUNCTIONING

Given the high prevalence of chronic pain in children, and the fact that it is often associated with disability, it is critical to investigate what factors can predict chronic pain and disability in children. Such information will be extremely helpful in the understanding and the prevention of these problems.

The development and maintenance of chronic pain and pain-related disability in children and adolescents are not completely and satisfactorily explained. Miró's biopsychosocial model can be used as a template to guide new research in this area, and parsimoniously integrate the available knowledge (see *Figure 2*). According to Miró's suggestions, different levels (physiology, emotions, thoughts, and behaviors) and units of analysis (individual, dyad, context) must be analysed, and their interactions studied (see Miró, 2003).

Given the complexity of Miró's model, in order to empirically support it, we need to test components of the model in a step-by-step fashion. This method of developing support for a broad theoretical model can be supported by recent research in the field. In the past few years, a few psychosocial models that address specific domains relating to pediatric chronic pain and its effects have been put forth. For example, Palermo and Chambers (2005) provided an integrative conceptual model for understanding the influences of parental behaviors (e.g., parenting style, parental reactions to the child's pain behaviors), parent-child dyad behaviors (e.g., parental-child interaction), and family behaviors (e.g., family environment, family functioning) on a child's pain experience and pain-related disability (see *Figure 3*).



**Figure 3. Integrative model of parent and family factors in chronic pediatric pain and related disability (reproduced with permission from Palermo and Chambers, 2005)**

Walker (1999) suggested a conceptual model focused on the role of a few cognitive and behavioral factors (child and parent) on the course of recurrent abdominal pediatric pain in children. According to this model, children may become caught in a self-perpetuating cycle. The child's and family's threatening interpretations of the child's symptoms increase parents care behaviors towards the child (e.g., privileges and attention) increasing the dependence of the children on parents. These interpretations increase the risk that children will restrict their normal daily activities (i.e., these children may now be easily excused from activities that might promote feelings of personal failure). All these consequences unintentionally reward the children's sick role, and might foster passive coping, skill deficits, and feelings of perceived inferiority since children lose opportunities to successfully accomplish normative developmental tasks. The model is displayed in *Figure 4*.



Turk's diathesis-stress model (Turk, 1996), a general model to explain the progression from acute pain to chronic disabling pain in adults. The model suggests that injury is a possible trigger of initial pain symptoms, but chronic pain and disability are the result of a complex interaction of biological, psychological, and social and cultural factors. In particular, this model stresses anxiety sensitivity to pain symptoms, fear of further pain and harm, catastrophizing cognitions, feelings of low self-efficacy, and learning factors through operant processes as major predictors of the development of adult chronic pain and the maintenance of disability in the long term (see *Figure 5*).



**Figure 5. Diathesis-stress model of chronic pain and disability following trauma (reprinted with permission from Turk, 1996).**

Vlaeyen et al. (1995) suggested a more narrow model (the fear-avoidance model), and more concrete mechanisms that illustrate the relationship between some prominent cognitions, emotions and behaviors of the individual, and its relation to the adult pain experience. More specifically, the fear-avoidance model illustrates the relationships between catastrophic thinking (cognitions), fear of pain (emotions), and avoidance behaviors. As can be seen in *Figure 6*, two different possible pain reaction pathways exist depending on the individual's interpretations of their pain symptoms. When pain symptoms are interpreted catastrophically, this leads to higher levels of distress and fear reactions. These emotional reactions are linked to avoidance behaviors and guarding behaviors in order to prevent further pain and harm, which in turn are responsible of physical disuse, depression, and disability; contributing to the maintenance of a pain problem. In contrast, when pain is perceived as non-

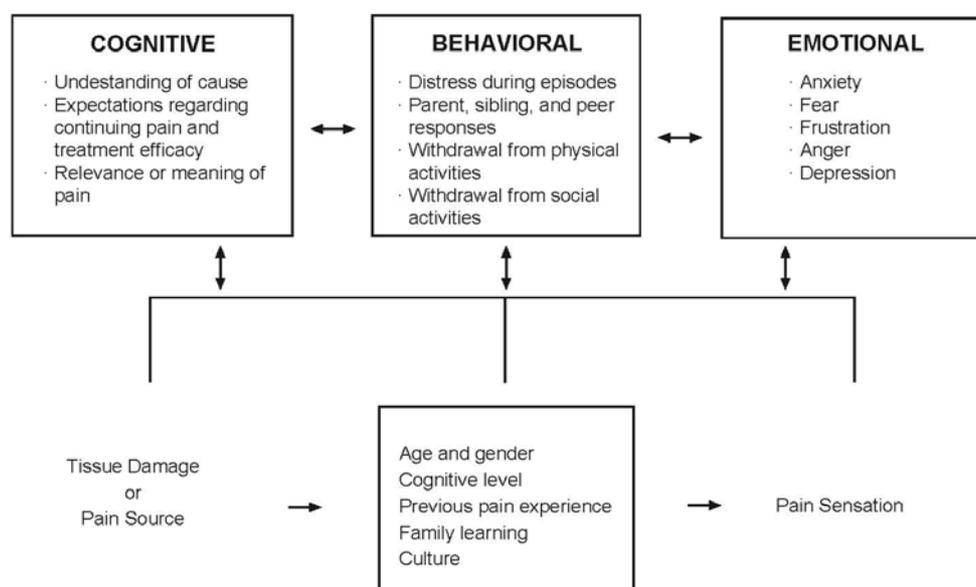
threatening, it encourages confronting rather than avoiding daily activities, and the likelihood to functional recovery increases. The relevance of the 'pain-related fear' factor on pain experience has been repeatedly reported across different stages of pain (e.g., it has been reported as a vulnerability factor for the inception of acute pain, as a risk factor for the development of chronic pain, and as a maintaining factor of chronic pain, see Boersma and Linton, 2005).



**Figure 6. The fear-avoidance model (reproduced with permission from Vlaeyen and Linton, 2000)**

Another way to gather additional insights into what factors predict chronic pain and related disability is to look at factors that are thought to play a significant role in acute pain. In fact, the general model for the factors involved with pediatric acute pain proposed by McGrath and Gillespie (2001) is consistent with Miró's model for chronic pain. McGrath and Gillespie's model suggests that psychological factors, including cognitions, behaviors, and emotions, affect children's perception of acute pain. Beyond these general psychological factors, the model also recognizes the influence of child's age, sex, cognitive level, and relevant situational factors on the pain experience (see *Figure 7*). As the authors state, *what children understand, what they do, and how they feel have a profound impact on their pain experience* (McGrath and Gillespie, 2001, p. 98). This model was formulated in relation to acute pain; however, factors identified might also be relevant for chronic pain problems as we have seen previously.

To summarize, we still have a long way to go to see Miró's framework filled with empirically supported relevant factors that explain the development and maintenance of chronic pain and related disability in children and adolescents. Experts are working on different models, some of which have been presented in the preceding pages; these models are awaiting for confirmation and empirical validation. Quite reasonably, available models are narrow in scope, but we do need a comprehensive model that could help explain the different chronic pain issues that are encountered in the clinic. Moreover, conceptual models are not always specific enough; for example, it is important to understand what specific child, parent and family factors (e.g., what pain thoughts, what pain coping strategy, or what parental pain reactions) have an influence on the children's chronic pain and/or functioning. Palermo and Chambers suggest that parental style plays an important role in pediatric pain behaviors, but they do not provide specific information about different styles in their model. They also highlight the importance of pain coping strategies, but do not focus on specific strategies in their model.



**Figure 7. A model of the cognitive, behavioral, and emotional factors that affect a child's pain experience. Reproduced with permission from McGrath and Gillespie, 2001.**

A growing literature on the role of psychological factors in chronic pediatric pain and disability is being published. A few biopsychosocial risk factors have been isolated in relation to the development and course of chronic pain and/or functioning. In *Appendix 1* potential predictive factors for either chronic pediatric pain or functioning as a result of pediatric chronic pain are presented (i.e., functioning defined in terms of quality of life or pain-related functional disability).

In general, the units of analysis that have been more widely examined in pediatric pain research are the child and parents. In relation to the ***child as the unit of analysis***, multiple levels (i.e. physical, behavioral, affective, and cognitive) have started to be examined. At *the physical level*, demographic factors (i.e., sex and age) have been the most studied. While neither factor seem to be predictive of the child's level of functioning (e.g., Gauntlett-Gilbert and Eccleston, in press; Keogh and Eccleston, 2006; Konijnenberg et al., 2005; Logan et al., 2006; Logan and Scharff, 2005; Merlijn et al., 2006; Peterson and Palermo, 2004), child sex and age may exert some influence on the child's risk of suffering chronic pain (e.g., Egger et al., 1998; Kristjansdottir and Rhee, 2002; Larsson and Sund, 2005; Mikkelsen et al., 1998; Prista et al., 2004; Sjolie et al., 2002; Vikat et al., 2000). Factors related to the child's health status have also been studied. In general, children with more physical symptoms (e.g., fatigue, constipation, nausea) are more likely to suffer chronic pain and to report lower levels of functioning. For example, research by Vikat et al. (2000) showed that the presence of psychosomatic symptoms (e.g., trembling hands, feeling weak) was an important predictor of recurrent neck, shoulder and low back pain among a large community sample of children. Further, research by Claar et al. (1999) showed that the physical symptoms of irritable bowel syndrome in children with recurrent abdominal pain were an important predictor of disability. The influence of anthropometric factors (e.g., Kovacs et al., 2003; Sjolie et al., 2002, 2004; Watson et al., 2003), physical condition (e.g., Feldman et al., 2001; Kovacs et al., 2003; Mikkelsen et al., 1998), and mechanical factors (e.g., Bejia et al., 2005) have also received some attention in recent research. However, most studies conclude that these factors are not relevant neither for predicting chronic pain, using cross-sectional or prospective designs (Bejia et al., 2005; Harreby et al., 1999; Jones et al., 2003; Szpalski et al., 2002).

Besides physical factors, general ***affective, behavioral, and cognitive factors***, have also been studied in relation to the child unit of analysis. As for affective factors, it is noteworthy that psychological distress, depression and anxiety may predispose the child to develop more disabling pain (Anttila et al., 2002; El-Metwally et al., 2006; Feldman et al., 2001; Gauntlett-Gilbert and Eccleston, in press; Hotopf et al., 1998; Jones et al., 2003; Kashikar-Zuck et al., 2001; Larsson and Sund, 2005; Schanberg et al., 2005). As for behavioral factors, it seems probable that health-related behaviors may contribute to increase pain and/or effect daily functioning in children with chronic pain. Sleep, physical activity and smoking are specific health-related behaviors that have been most well researched. Research has shown that sleep problems are common in children with chronic pain problems (see Walters et al., 1999). However, a causal relationship (e.g., sleep leading to greater pain) is not consistently reported (El-Metwally et al., 2006; Kristjansdottir and Rhee, 2002; Laurell et al., 2005; Szpalski et al., 2002). In terms of physical activity, it is possible that activity increases the occurrence of pain problems. A prospective population-based cohort study conducted by Jones et al. (2003a) found that children who undertook high levels of physical exercise were at greater risk for future low back pain problems. However, this effect is not consistently reported across studies (Feldman et al., 2001; Laurell et al., 2005). Additionally, other factors related to physical activity, such as the amount of practice/experience, intensity, or type of physical activity have been investigated and found non-consistent relationships (Bejia et al., 2005; Ghandour et al., 2004; Vikat et al., 2000). The literature is controversial about the role of smoking and its relation to chronic pain (Feldman et al., 2001; Ghandour et al., 2004; Kovacs et al., 2003; Kristjansdottir and Rhee, 2002). As for cognitive factors, very little research has investigated the role of general cognitive factors (Odsson et al., 2006).

Along with general affective, behavioral, and cognitive factors, pain-related factors are starting to be considered. Several studies have shown that the severity of pain (i.e., intensity, frequency, duration, pain-related disability) and maladaptive pain coping strategies (e.g., use of emotion-focused avoidance) are related to chronic pain and level of functioning (Gauntlett-Gilbert and Eccleston, in press; Larsson and Sund, 2005; Merlijn et al., 2003;

Mikkelsen et al., 1998; ), however these relationships are not reported across all studies (El-Metwally et al., 2005; Flato et al., 1997; Perquin et al., 2003).

In comparison to research on factors related to the child as the unit of analysis, there are fewer studies that examine **parents as the unit of analysis**. The most well supported findings in regards to parental factors and chronic pain in children can be synthesized as follows: (1) neither parents' occupation nor parents' educational background can account for the presence of chronic pain in children (Anttila et al., 2002; Bener et al., 2000; Kristjansdottir and Rhee, 2002, Vikat et al., 2000); (2) poor parental support is common among children with chronic pain (Ghandour et al., 2004; Kristjansdottir and Rhee, 2002); and (3) parental pain is associated with chronic pain in children (Boey and Goh, 2001). The most well supported findings in regards to parental factors and level of functioning in children with chronic pain are: (1) parental psychological distress does not seem to affect to the levels of functioning among children with chronic pain (Gauntlett-Gilbert and Eccleston, in press; Logan and Scharff, 2005); (2) a distressed adolescent-parent distressful does not affect functioning in adolescents with chronic pain (Gauntlett-Gilbert and Eccleston, in press; Logan et al., 2006); (3) the association between parental pain and disability among children with chronic pain is not consistently reported across studies (Logan et al., 2006; Lynch et al., 2006; Merlijn et al., 2006; Reid et al., 1997); and (4) the potential effect of the solicitous parental behaviors (e.g., Brace et al., 2000) in response to child's pain is still uncertain (Merlijn et al., 2006; Peterson and Palermo, 2004; Reid et al., 1997).

It is also important to note that the contextual stress in children's lives might be positively related to the occurrence of chronic pain problems in children (Boey and Goh, 2001; Feldman et al., 2002; Ghandour et al., 2004).

The identification of predictive factors is a good starting point to understand both chronic pain and pain-related disability. By learning what the key factors are, we will be in a good position to formulate and test predictive models of pediatric chronic pain and disability. These models will help experts to design and test specific prevention and treatment programs. In this line, the present research examines many factors that may be important to pediatric chronic pain and functioning, such as socio-demographic factors, anthropometric factors, pain characteristics, physical activity, sedentary activity,

stressful life events, as well as psychosocial factors related to pain (i.e., pain coping strategies, pain catastrophic thinking, pain-related affect, pain beliefs, parental reactions to child's pain behaviors, and parental pain) will be also examined. Thus, in the present research, we are more interested in the role of several psychosocial factors related to pediatric pain (e.g., pain catastrophic thinking, pain-related affect) than in the role of other general psychological factors (e.g., anxiety, depression), because specific factors related to pain may be more helpful to predict chronic pain problems and levels of functioning than general factors (De Vlieger et al., 2006).

There are already a handful of studies that connect several child and parent factors with pediatric chronic pain and/or level of functioning. However, more research is necessary and the improvement of research methods in the field is also fundamental. Clearly, the early stage of development of this research field as well as the methodological flaws have hampered our arriving to firm conclusions regarding predictors of pain and disability. For example, research on predictive factors is often based on cross-sectional designs instead of prospective ones. Available studies often work with samples that are very small in size, and not necessarily representative of the general population (for example, they often examine convenience samples extracted from clinical settings). Also, these studies often examine different types of recurrent or chronic pain (e.g. musculoskeletal pain, headache, or back pain), and it is already known that different types of pain co-occur very often. Studies focused on a same pain problem often differ on how to define the pain problem. Many studies select different types of outcome variables to measure level of functioning (e.g., they focus on a general outcome, such as quality of life, or they focus on more specific outcomes, such as physical functioning). Available studies also use different assessment measures to assess the same outcome (for example, they use different instruments to assess quality of life). Moreover, different assessment procedures are used (e.g., self-reports vs. parent-proxy reports), and we already know, for example, that the concordance between self-reports and parents-proxy reports is doubtful (e.g., Chambers et al., 1998). Available studies consider different age ranges, and it might lead to different results since predictors might be dependent to individual's age. Finally, studies might differ in data analysis and data presentation, and it might cause that

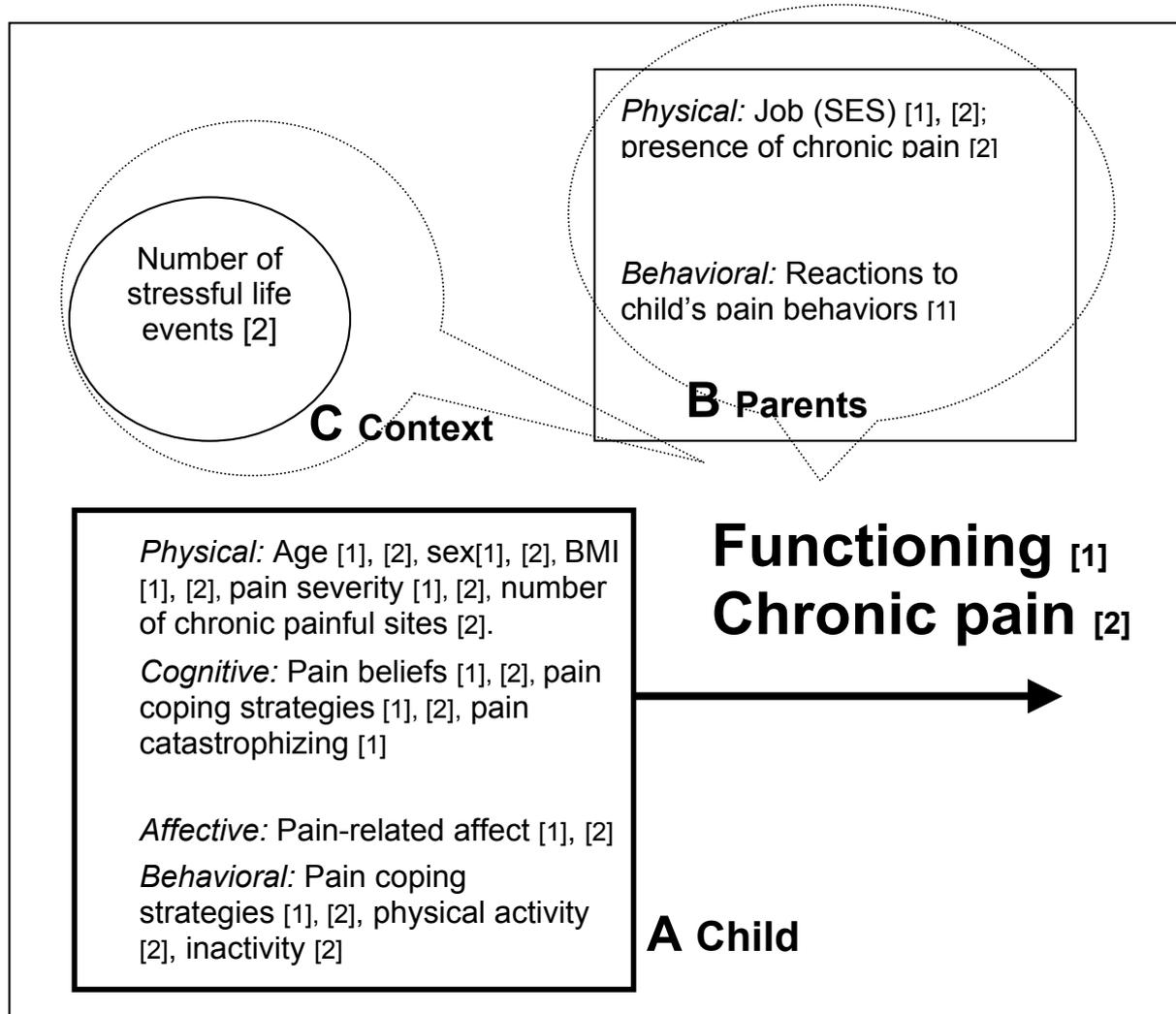
results and conclusions extracted be different. Thus, the conclusions exposed above are still very preliminary. Our study improves on previous studies by also using a population-based approach, a large sample, longitudinal level methods, and by not restricting the research to only one type of pain.

## 5. PURPOSE, SPECIFIC AIMS, TASKS, AND HYPOTHESIS

The *purpose of our study* is to gather additional information that could contribute to a better understanding about pain in youth, and we attempted to do so by means of three complementary **general objectives**:

1. To adapt and develop assessment measures of pain and pain related constructs to be used with Catalan speaking children. More specifically, in this thesis we present studies whose aims were to:
  - a. Adapt and examine the psychometric properties of a Catalan version of the Faces Pain Scale-Revised to assess pain intensity in children aged 7 to 15 years.
  - b. Adapt and examine the psychometric properties of a Catalan version of the Pediatric Quality of Life Inventory<sup>TM</sup> to assess the children's quality of life when it is self- and interviewer-administered to children aged 9 to 17 years.
  - c. Develop and examine the psychometric properties of a measure to assess parents' reactions to the child's pain behaviors.
2. To study pain problems in Catalan children aged 8 to 16 years. More specifically, we wanted to:
  - a. Estimate the prevalence and the course of chronic pain complaints over time.
  - b. Describe the characteristics of chronic pain problems (e.g., severity, localization).
  - c. Analyse the impact of chronic pain problems on children's quality of life.
3. To identify psychosocial prognostic factors of children's functioning, and to identify psychosocial prognostic factors of the course of pain complaints. More specifically, we wanted to:
  - a. Learn what the literature says about prognostic factors of chronic pain and disability.
  - b. Learn what professionals think about prognostic factors of chronic pain and disability.

- c. Investigate the role of several psychosocial factors on children’s functioning, and on the onset and maintenance of chronic pain over time. In this thesis, we have tested some relevant areas from Miró’s model, those that are displayed in *Figure 8* (Miró, 1998, 2003).



[1] Factors under investigation; these were studied in order to determine whether they are predictors or not of the level of functioning of the child; [2] factors under investigation in order to know whether they were predictors or not of the onset or the persistence of chronic pain over time (i.e., at 1-year follow-up).

**Figure 8. Model depicting levels and units of analysis.**

To accomplish our general objectives, we have been working during the last five years on the following avenues:

A. *Pain assessment* (first objective): As we mentioned in earlier parts of this work, valid and reliable pain measures and outcome measures are fundamental to understand and manage pain problems. In our country, there are very few assessment instruments that have been tested in children. One of the cheapest and most useful ways of obtaining psychometrically sound instruments is to translate and validate those that are already available and have proved their worth. This is why we have been working intensively on the validation of already existing instruments that measure pain, and pain related issues. For example, (1) pain intensity (see Miró et al., 2006, 2007; Miró and Huguet 2004), (2) coping; (3) pain attitudes; (4) pain related affect; or (5) quality of life (see Huguet and Miró, in press). At the same time, we have been working in the development of new assessment tools too (e.g., IRPEDNA, a measure to assess parents' reactions to children's pain behaviors; see Huguet et al., in press).

The results of these works are beginning to receive recognition among researchers and clinicians. For example, some of the measures that we have adapted for Catalan speaking children have been requested, and adopted in major hospitals in Barcelona. Moreover, last year, one of our works (i.e., *The evaluation of reliability, validity, and preference for a pediatric pain intensity scale: The Catalan version of the Faces Pain Scale-Revised*; Miró and Huguet, 2004) received the Catalan Society of Behaviour Research and Therapy award for best paper.

B. *Systematic review of the literature* (third objective): Two systematic reviews are being carried out to collect evidence about: 1) psychosocial factors that predict either ability or disability among children and adolescents who suffer from pain (Miró et al., in preparation), and 2) psychosocial factors that predict the transition from an acute pain problem to a chronic pain problem (Huguet et al., in preparation). Cross-sectional and longitudinal studies published in the last 10 years that provide such information have been reviewed. Two manuscripts with the

results obtained from these reviews are being prepared, and they are expected to be submitted soon.

- C. *Learning what professionals' think about prognostic factors of chronic pain and disability* (third objective): Since the available evidence is inconclusive and deficient, we decided to ask what professionals thought about these issues, and conducted a Delphi poll to gather relevant information (see Miró et al., in press).
- D. *Epidemiology of pain* (second and third objective). We are conducting a longitudinal epidemiological study with a large school sample. This study began in 2003, and it is still in progress. The objective of this study is, on the one hand, to understand the most common problems of pain in our country (Huguet and Miró, 2007) and, on the other, to empirically test potential prognostic factors in the onset and persistence of chronic pain, and in the functioning of children (Huguet et al., 2007; Miró et al., in preparation).

Simultaneously, and in an attempt to help our society being aware of these problems, we have been disseminating the results of our research to different audiences. More specifically, we have participated in several national scientific meetings, and published some of our works in Spanish scientific journals (e.g., Miró et al., 2006, 2007); we have arranged presentations for residents of cities where the schools participating in our studies are located. We have also reported the results of our epidemiological study through the media (newspapers, radio, and television). We have got involved in training courses for pediatric clinicians, and at this moment we are preparing a formal report to be presented to politicians suggesting specific actions to help better manage chronic pain problems from their onset.

This research work has been supported, in part, by both private and public agencies (i.e., *Agència de Gestió Acadèmica d'Universitats i Recerca de la Generalitat de Catalunya*, *Ajuntament de Reus*, *Ajuntament de Tarragona*, *Fondo de Investigaciones Sanitarias del Ministerio de Sanidad y Consumo*, *Ministerio de Educación y Ciencia*).

**Hypothesis** for the objectives presented above are as follows: As far as the *first general objective* is concerned, when measures were adapted for Catalan-speaking population, we expected to find similar psychometric properties than those found for the original measures. As far as the *second general objective* is concerned, it was expected that chronic pain would be common in Catalan children, and severe chronic pain would be reported by a small proportion of children. It was expected that the rate of chronic pain would increase with age, and be more common among females. We expected to find that children with chronic pain would have lower levels of functioning than children without chronic pain. As far as the *third general objective* is concerned, it was hypothesized that socio-demographic, anthropometric, life-style, contextual, and psychosocial factors related to children's pain (i.e., pain beliefs, pain coping strategies, affect related to pain, catastrophic thinking about pain, parents' chronic pain problem, and parents' reactions to child's pain) would account for a significant portion of variance of the level of functioning and course of pain over time for children with chronic pain.

Some of the tasks listed above are *still in progress*. At this point, we list and present those studies that are finished or almost finished. You will see that there are manuscripts that have been published, or have been accepted for publication, but still are in press (papers #1, #2, #3, and #5). There are other manuscripts that are under review (papers #4, #6) and another that is almost ready to be submitted for publication (paper #7). We have added these last two categories of manuscripts ("under review" and "in preparation"), so that the tribunal may have a broad comprehensive view of all the work accomplished to date.



## LIST OF PAPERS

### *Assessment*

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1. Miró J, Huguet A. (2004). Evaluation of reliability, validity, and preference for a pediatric pain intensity scale: the Catalan version of the Faces Pain Scale-Revised. *Pain*, 111, 59-64.
2. Huguet A, Miró J. (In press). Development and psychometric evaluation of a Catalan self- and interviewer-administered version of the Pediatric Quality of Life Inventory™ Version 4.0. *Journal of Pediatric Psychology*.
3. Huguet A, Miró J, Nieto R. (In press). The Inventory of Parent-Caregiver Responses to the Children's Pain Experience (IRPEDNA): Development and preliminary validation. *Pain*.

### *Epidemiology of pain*

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4. Huguet A., Miró J. (Submitted for publication). The severity of chronic pediatric pain: An epidemiological study in the Mediterranean region of Catalonia. *Journal of Pain*.

### *Predictors of chronic pain and disability*

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5. Miró J, Huguet A, Nieto R. (In press). Predictive factors of chronic pediatric pain and disability: A Delphi poll. *Journal of Pain*.
6. Huguet A, Eccleston C, Miró J, Gauntlett-Gilbert J. (Submitted for publication). The influence of pain and pain related factors on the functioning of 8-16 year old children. *Pain*.
7. Miró J, Eccleston C, Huguet A, Nieto R. (In preparation). Predictive factors of the course of pain in schoolchildren.



**Paper #1:**

**Miró J, Huguet A. (2004). Evaluation of reliability, validity, and preference for a pediatric pain intensity scale: the Catalan version of the Faces Pain Scale-Revised. *Pain*, 111, 59-64.**





## Evaluation of reliability, validity, and preference for a pediatric pain intensity scale: the Catalan version of the faces pain scale – revised

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### Abstract

The main objective of this research was to determine the initial psychometric properties of the Faces Pain Scale – Revised (FPS-R) as a measure of pain intensity for use with Catalan children and adolescents. Results of the Catalan version of this scale (FPS-R-C) are similar to those obtained with the original instrument. In order to assess the validity and reliability of the FPS-R-C, two different samples were studied. The first sample contained 124 hospitalized children and adolescents (mean age 10.86; SD 2.5). They were asked to rate their affective state on the Faces Affective Scale (FAS) and the intensity of their pain on the FPS-R-C and the Coloured Analogue Scale (CAS). The pain intensity ratings reported with FPS-R-C and CAS were very similar, correlations ranging from 0.83 to 0.9. The relationship between the intensity of pain experienced and children's affective state was also statistically significant ( $r = 0.32$ ,  $P < 0.01$ ). The second sample contained 247 schoolchildren (mean age 9.43; SD 1.55), who were asked to imagine themselves in eight hypothetical painful situations and rate the degree of pain using the FPS-R-C and the CAS (correlations ranging from 0.83 to 0.96). Test–retest correlations on this questionnaire (Painful Events Inventory) ranged from 0.26 to 0.70. Overall, these results provide preliminary evidence of the reliability, and convergent and criterion-related validity of the FPS-R-C. Moreover, all participating subjects were asked to choose the pain scale they preferred the most. Our data suggest that, regardless of their age and/or gender, the subjects prefer the FPS-R-C to the CAS.  
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*Keywords:* Pediatric pain measurement; Faces Pain Scale – Revised – Catalan Language Version; Validity; Reliability

### 1. Introduction

The Faces Pain Scale – Revised is a self-report scale using facial expressions to assess the intensity of pediatric pain. It has shown several advantages over other available faces scales (see reviews by Champion et al., 1998; Kuttner and LePage, 1989; McGrath and Gillespie, 2001). First, it has no smiling face and/or tears. It is well known that scales with smiling 'no pain' faces produce relatively higher pain ratings than the more neutral ones (Chambers et al., 1999; see also Wong and Baker, 2001). Clearly, the FPS-R avoids the confounding of affective distress with pain intensity (Chambers and Craig, 1998; Champion et al., 1998, 2000). Second, the FPS-R has been reduced from seven to six faces. It has the advantage of being suitable for use with the most widely used scoring metric (0–10) (Hicks et al., 2001). And third, besides having a true 0 point, the intervals on the scale are equal (Spagrud et al., 2003).

At present, several non-English versions of the FPS-R have already been developed, and are currently being tested. To our knowledge an Arabic, Bulgarian, Chinese, Dutch, French, German, Hebrew, Italian, Persian (Farsi), Portuguese, Spanish, Swedish and Thai versions are available (see Von Baeyer and Piira, 2003). This is another asset of the scale, further evidence of the interest and positive expectations that the FPS-R has awakened among pain researchers and clinicians.<sup>1</sup>

Translating questionnaires into other languages is a common procedure today (Turk and Melzack, 2001). The rationale being that it is better to use existing instruments that successfully measure pain and/or related variables than develop new tools with all the time consuming procedures that this involves. In fact, a carefully translated instrument makes it possible to compare different

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<sup>1</sup> The interested reader might want to visit [www.painsourcebook.ca](http://www.painsourcebook.ca) where instructions are currently available free of charge in 16 languages (instructions in English, and Catalan are also available).

populations across cultural settings, for example, on treatment effects.

In this process, however, translating the instruments is not enough. The psychometric properties of the instrument transposed into a new language must also be evaluated. Clearly, one cannot assume that an instrument shown to be valid and reliable for one population has equal characteristics in another sample with a different cultural background and/or language. Accordingly, the importance of testing any assessment instrument before recommending that it be used in a language different from the original, has been greatly emphasized (Boureau and Paquette, 1988; Melzack and Katz, 2001; Miró, 1997).

The objectives of this study were to (1) determine the initial psychometric properties of the Catalan<sup>2</sup> version of the FPS-R (FPS-R-C), and (2) compare patients' opinion of the FPS-R-C with another very common self-report instrument for the assessment of pain intensity: namely, the Coloured Analogue Scale (CAS).

## 2. Method

### 2.1. Validity testing

The construct validity and the criterion validity of the Catalan version of the Faces Pain Scale – Revised (FPS-R-C) were examined. It was assumed that, on the one hand, scores on FPS-R-C would be highly correlated with a scale assessing the same construct (*convergent validity*). In this study, ratings on the FPS-R-C should be highly correlated with ratings on the Coloured Analogue Scale (CAS), a scale that is also used to assess the intensity of pain. On the other hand, scores on FPS-R-C will also highly correlate with scores on a scale measuring a variable that can be predicted from pain intensity, such as the subjects' affective state (*criterion-related validity*). In this study, ratings on FPS-R-C should be highly correlated with ratings on the Facial Affective Scale (FAS), which is used to assess the affective dimension of the pain experience. Moreover, the magnitude of the correlations between the FPS-R-C and another pain intensity scale should be greater than between the FPS-R-C and a scale measuring the affective characteristics of the pain experience (*discriminate validity*). In this study, the correlations between the scores on the FPS-R-C and CAS should be higher than correlations between the scores on the FPS-R-C and FAS.

### 2.2. Reliability testing

Subjects were asked to rate pain intensity on the basis of eight different hypothetical events at two points in time.

<sup>2</sup> Catalan is a language spoken by well over 6 million people in eastern Spain, southwestern France, and the Island of Sardinia (located in the middle of the western Mediterranean).

It was expected that estimated intensity of pain at time 2 would essentially be the same as estimated intensity of pain at time 1. If the FPS-R-C is a reliable measure, then two different estimations made at two different points in time ought to be similar.

## 3. Subjects

Two samples of children and adolescents for a total of 371 individuals participated in this study. Of these, 124 were hospitalized patients (sample 1) and 247 were schoolchildren (sample 2).

Patients in sample 1 were hospitalized for different surgical and non-surgical painful conditions. All children and adolescents aged between 7 and 15 hospitalized during the months of June, July or August in 2003 in three different institutions were asked to participate in the study ( $N = 138$ ). In 41 cases (29.7%) the reasons for hospitalization were for surgical treatment which included appendectomy, varicocele, pectus, among other problems. All the other patients (70.3%) were hospitalized for non-surgical painful conditions. Those patients who were between 7 and 15 years old, who had no cognitive impairment, who were able to understand the Catalan language, and whose parents or guardians gave informed consent, were finally included in the study ( $N = 124$ ). Gender distribution for this sample was as follows: 43% girls and 57% boys. The mean age was 10.86 (SD 2.5).

The subjects in sample 2 were recruited from a primary school in Tarragona. As in sample 1, exclusion criteria were: (1) having some cognitive impairment, and (2) being unable to understand Catalan. Two hundred and fifty schoolchildren were approached, and informed consent from parents or guardians for each participant was obtained. Three subjects had to be excluded because of exclusion criteria. Thus, there were 121 girls (49%) and 126 boys (51%) in sample 2. Sample age distribution ranged from 7 to 12 years (mean age 9.43; SD 1.55). Two hundred and nine of these were retested after 4 weeks. Data from 38 schoolchildren (15%) is not available, since they did not attend school on our second visit (retest).

Only children between 7 and 12 years old took part in testing the reliability of the study. Although the FPS-R has proved to be appropriate for use in children above 5 years old (Hicks et al., 2001), the task that we required them to perform with the Painful Events Inventory is too cognitively complex for children under the age of 7, and clearly more complex than the one used by Hicks et al. (2001).

## 4. Instruments

The *Faces Pain Scale – Revised* (FPS-R, Hicks et al., 2001) is a self-rated-administered scale designed to measure children's level of perceived pain intensity. It is a six-point

scale that is presented horizontally. The children's task is to choose the face that best reflects the intensity of the pain they have experienced. A numeric value from 0 to 10 (0–2–4–6–8–10) is assigned to each face. The end points were explained as 'no pain' and 'very much pain'. The scale was validated by Hicks et al. (2001).

The *Coloured Analogue Scale* (CAS, McGrath et al., 1996) is a visual analogue scale that measures the severity of pain experienced. The CAS consisted of a mechanical device with a plastic slider over a 143 mm long tetragon varying from narrow (10 mm) and white at the bottom labelled 'no pain' to wide (30 mm) and dark red at the top labelled 'most pain'. The CAS was scored from 0 to 10 in increments of 0.25.

The CAS has been shown to be a reliable and valid measure in a sample of children and adolescents aged 5–17 years (McGrath et al., 1996).

The *Facial Affective Scale* (FAS, McGrath et al., 1996) is a self-report measure used to assess the unpleasantness of a child's pain experience. It is composed of nine face drawings showing gradual increases in distress. The child is asked to choose the face that best fits his or her affective state from the set of faces, the specific instructions were as follows: 'now I am going to show you some faces and I want you to choose the face that looks like how you feel. Choose the face that looks like how you feel deep down inside, not just the face you show on the outside'. The FAS is scored on a 0–1 scale, where the maximum negative affective value equals 1 and the maximum positive one is 0. The endpoints were explained as 'the happiest feeling possible' and 'the saddest feeling possible'. The FAS has been validated in a sample of young people aged 5–17 (McGrath et al., 1996).

The *Painful Events Inventory* (PEI) was developed for this study. It is a self-report questionnaire that lists eight different hypothetical painful events. Subjects had to report, for each event, 'how much it would hurt them, if they were in that situation' (see Table 2).<sup>3</sup> Cronbach's alpha coefficient for PEI is acceptable ( $\alpha = 0.66$ ). A maximum-likelihood factor analysis suggested that the one-factor model is also acceptable ( $\chi^2_{(20)} = 25.95$ ;  $P = 0.16$ ). Therefore, the PEI can be considered as essentially unidimensional.

## 5. Procedure

### 5.1. Translation of the instrument

The back-translation technique described by Berkanovic (1980) was used to translate the instrument into Catalan, preserving the denotation and connotation of the instrument's instructions. First a bilingual psychologist,

<sup>3</sup> Using verbally administered hypothetical pain situations to validate pain scales is an approach originally developed by Dr Patricia A. McGrath (see McGrath, 1990).

who had never seen the instrument before, translated the instructions into Catalan. Then the translated instructions were back translated from Catalan into English by a native English speaker who was also ignorant of the original version of the instrument. Then, the new English version (back-translation version) was sent to the instrument's author to check whether the back-translation was appropriate. Some minor changes were made in the Catalan instructions following the recommendations of the author, to solve slight discrepancies between the back-translated English version and the original.

This study was approved by the Ethics Committees of the Rovira i Virgili University and the participating hospitals. Once they had granted permission that the protocol could be followed, participants were recruited and informed consent was obtained. Two slightly different procedures were followed depending on the sample and the objectives of the study.

In sample 1 (hospitalized children), all the subjects who met the sampling criteria were approached to participate. The sample was a convenience sample based on consecutive patients. After the parents' had given their consent, all the children were interviewed in their room by the same person for about 15 min. The interview was carefully coordinated with nurses and doctors to avoid interrupting their work. First of all, the interviewer collected some basic demographic information and then measured patients' affective state and pain intensity. Using the Facial Affective Scale, children were asked to choose the face that represented how they felt at the time. The intensity of their current pain was measured with two scales: the Faces Pain Scale – Revised (Catalan version) and the Coloured Analogue Scale. Finally, children were asked to choose the scale they preferred to measure pain intensity.

In sample 2, the participating children were recruited from a primary school in Tarragona. The parents and legal tutors whose children met the inclusion criteria were asked for their permission to participate in the study, and an informed consent for each participant was obtained. Individual interviews took place during school time. All interviews were individually conducted by the same researcher and lasted about 5 min each.

To introduce the task and test whether the children understood the procedure, they were asked to rate the intensity of their present pain using the FPS-R-C. Once it was clear the children understood the procedure and were able to use the scale, they were asked to imagine themselves in eight common painful situations and rate the degree of the pain intensity they would experience in each one (see Table 1). For the purposes of this study, children rated their pain with two different formats, one based on the FPS-R-C and the other on the CAS format. Finally, all the children were asked to indicate which format they preferred. One month later, the children answered the PEI again (retest), but this time only the format of FPS-R-C was used.

Table 1  
 Items in the Painful Events Inventory

Event 1	You are stung by a bee.
Event 2	You shut your finger in the door.
Event 3	You have a tooth taken out.
Event 4	You burn your hand.
Event 5	You have some stitches.
Event 6	You are given an injection.
Event 7	You fall over and scrape your knees.
Event 8	You put your fingers in a socket and get an electric shock.

## 6. Results

### 6.1. Construct validity

Tables 2 and 3 present the Pearson correlations between the FPS-R-C and the CAS for all age groups and samples.

The correlations are high and positive, and the scatter plot suggested that the relation was essentially linear. Moreover, the correlations for all age groups exceeded a common conventional standard in both samples ( $r > 0.70$ ). Fisher's  $z$ -transformation was used to compare the correlation coefficients, and none of the comparisons was statically significant at the 0.01 level. Thus, the correlation structure appears to be invariant across age. As can be seen in Table 2, the children estimated item 6 'the nurse vaccinates you' as the least painful event, but item 4 'you have burnt your hand' as the most painful one.

### 6.2. Criterion validity

Affective state and pain have shown to be correlated (Miró and Raich, 1992; Turk et al., 1983; Weisenberg, 1975). In this study, and as it was previously mentioned, subjects' affective state was used as a criterion for pain intensity. The validity of the FPS-R-C in predicting affective state was assessed by the Pearson correlation between FPS-R-C and FAS scores. The coefficient was statistically significant ( $r = 0.32$ ,  $P < 0.01$ ). The correlation is appropriate if it is taken into account that both scales are single-item instruments and, therefore, expected to have a high amount of measurement error. As expected, the magnitude of the correlation coefficients

Table 2  
 Means, standard deviations and correlations between FPS-R-C and CAS ratings for hospitalized children present pain

Age groups (years)	N	FPS-R-C (0–10)		CAS (0–10)		$r_{(FPS-R-C, CAS)}$
		Mean	SD	Mean	SD	
7–9	43	1.72	2.29	1.79	2.24	0.90
10–12	43	2.33	2.37	2.68	2.46	0.83
13–15	38	2.00	2.13	2.48	2.43	0.89
Total sample	124	2.02	2.27	2.31	2.39	0.87

All coefficients are statistically significant at  $P < 0.001$ .

Table 3  
 Means, standard deviations and correlations between FPS-R-C and CAS ratings for schoolchildren based on their responses to the Painful Events Inventory

Age groups (years)	Item events	FPS-R-C (0–10)		CAS (0–10)		$r_{(FPS-R-C, CAS)}$
		Mean	SD	Mean	SD	
7–9 ( $n = 124$ )	1	6.12	2.79	5.48	2.80	0.90
	2	6.90	2.58	6.01	2.60	0.88
	3	4.78	3.13	4.54	2.80	0.92
	4	7.95	2.34	7.27	2.42	0.85
	5	7.34	2.82	6.75	2.78	0.86
	6	4.18	2.99	3.98	2.89	0.96
	7	4.72	2.48	4.37	2.46	0.87
	8	7.28	2.83	6.77	2.78	0.92
10–12 ( $n = 118$ )	1	5.35	2.59	4.48	2.44	0.88
	2	6.64	2.24	5.65	2.20	0.86
	3	4.91	3.01	4.37	2.55	0.94
	4	6.95	2.73	6.31	2.68	0.92
	5	6.14	2.89	5.45	2.80	0.88
	6	3.23	2.89	3.09	2.66	0.92
	7	3.27	2.00	3.06	1.99	0.83
	8	6.24	3.02	5.44	2.87	0.93

All coefficients are statistically significant at  $P < 0.001$ .

between scores on the FPS-R-C and CAS is greater than that of those on the FAS (*discriminate validity*).

### 6.3. Reliability

The reliability of the FPS-R-C was assessed by schoolchildren completing the PEI twice within a 4-week time interval. As can be seen in Table 4, the test–retest reliability coefficients were between 0.26 and 0.70. Since they are single-item coefficients, these results suggest that the FPS-R-C scores have an acceptable degree of temporal stability. The retest reliability coefficient for the full scale is 0.63.

### 6.4. Scale preference

Our results show that more children prefer the Faces Scale to assess pain. In sample 1 (hospitalized children),

Table 4  
 Test–retest reliability coefficient for each item in the Painful Events Inventory

Events	7–9 ( $N = 99$ )	10–12 ( $N = 90$ )	$N = 209$
1	0.46	0.54	0.50
2	0.55	0.54	0.55
3	0.54	0.41	0.49
4	0.51	0.44	0.42
5	0.54	0.60	0.61
6	0.67	0.70	0.68
7	0.47	0.26 <sup>a</sup>	0.43
8	0.47	0.70	0.57

<sup>a</sup> This correlation is statistically significant at  $P < 0.05$ . All other correlations are statistically significant at  $P < 0.01$ .

82 subjects preferred the FPS-R-C (66%), whereas 42 preferred the CAS (34%). In sample 2, 168 schoolchildren preferred the FPS-R-C (68%) and 79 schoolchildren preferred the CAS (32%). The proportion of children who reported a preference for the Faces Scale was significantly higher than the proportion of children that might be expected randomly in both cases ( $z_{\text{sample1}} = 4.87$ ,  $z_{\text{sample2}} = 2.78$ ;  $P < 0.01$ ).

## 7. Discussion

The main objective of this work was to determine the initial psychometric characteristics of the Catalan version of the Faces Pain Scale – Revised (FPS-R-C). It revealed similar statistical properties to the original. The FPS-R-C has strong validity and reliability properties. Thus, it seems to be an appropriate instrument for measuring pain intensity with Catalan children and adolescents.

Besides these robust psychometric properties, the FPS-R-C also proved to be a practical measure. The FPS-R-C was easy to use at the bedside, and consequently it can easily be integrated into practice. Measuring the intensity of pain by the FPS-R-C is simple and it does not take too much of the health care providers' time.

Our data also show that children and adolescents in these samples clearly preferred the FPS-R-C to the CAS, to report their pain intensity. This is also consistent with previous studies on children preferences for faces scales over such other scales as visual analogue scales, numeric scales or descriptive scales (see, for example, Champion et al., 1998, 2000; Johnston, 1998; Wong and Baker, 1988).

Although these initial results are positive, further studies are warranted to provide additional evaluative data regarding the psychometric soundness of the FPS-R-C with children and adolescents. For example, future work should test the FPS-R-C in children under 7 years old to determine the suitability of the measure for these younger children. Similarly, future studies should apply this instrument more widely, using it with children suffering from pain associated to medical conditions other than the ones included in this study. One other important avenue of research will be that of assessing changes, improved clinical outcomes, due to the regular use of the FPS-R-C in the measurement of pediatric pain.

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**Paper #2:**

**Huguet A, Miró J. (In press). Development and psychometric evaluation of a Catalan self- and interviewer-administered version of the Pediatric Quality of Life Inventory™ Version 4.0. *Journal of Pediatric Psychology*.**



## DEVELOPMENT AND PSYCHOMETRIC EVALUATION OF A CATALAN SELF- AND INTERVIEWER-ADMINISTERED VERSION OF THE PEDIATRIC QUALITY OF LIFE INVENTORY™ VERSION 4.0

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### ABSTRACT

*Objective:* The purposes of this study were to develop and to assess the psychometric properties of a Catalan self- and interviewer-administered version of the Pediatric Quality of Life Inventory Generic Core Scales with a sample of schoolchildren, and to examine the equivalence between both versions.

*Methods:* 511 schoolchildren aged between 9 and 17 years old participated in the study. In addition to completing a Catalan self-administered version of the PedsQL, each child was interviewed individually.

*Results:* Confirmatory factor analysis did not support the original Varni's proposal. Instead, a short 12-item version was derived. The higher-order scales for both versions were internally consistent. Moreover, relationships between ratings of children's quality of life were generally significant; both versions were also found to be related with another measure of quality of life supporting their validity.

*Conclusions:* A new administration form for the PedsQL is presented in this study. The psychometric properties of both self- and interviewer-administered short 12-item versions are reassuring albeit with a few areas of improvement. Further studies are needed to investigate whether self- and interviewer-administered versions can really be considered to be comparable.

**Keywords:** quality of life, Pediatric Quality of Life Inventory Generic Core Scales, interviewer-administered version, self-administered version.

## 1. Introduction

Many different instruments are available for measuring health-related quality of life in children (see Creemens et al., 2006a; Davis et al., 2006; Eiser and Morse, 2001; Harding, 2001 for some recent reviews). Among them, the Pediatric Quality of Life Inventory Generic Core Scales (PedsQL; Varni et al., 2001) is one of the most frequently used instruments (Clarke and Eiser, 2004, Rajmil et al., 2004a) and it has been strongly recommended in many reports (e.g. McGrath et al., 2007, Wallander et al., 2001). This is hardly surprising since it is claimed to have many of the positive features that experts believe a high-quality instrument must possess (Eiser and Morse, 2001, Matza et al., 2004): namely, (1) appropriateness of the conceptual and measurement model (Varni et al. 2005); (2) reliability (e.g. Varni et al. 2003, 2006), (3) validity (e.g. Varni et al. 2003, 2006), (4) responsiveness (Varni et al. 2002), (5) feasibility (e.g. Varni et al. 2003, 2006), (6) availability of alternative forms (i.e. child self-report and parent proxy reports) (Creemens et al. 2006b), (7) availability and equivalence of cultural and language adaptations (Chan et al. 2005; Felder-Puig et al. 2004, Upton et al. 2005, Reinfjell et al. 2006), and (8) interpretability, precision, and respondent and administrator acceptability (e.g. Varni et al. 2001, 2005) (see Fitzpatrick et al. 1998, Scientific Advisor Committee of the Medical Outcome Trust, 2002). The PedsQL 4.0 is a brief general measure of quality of life which covers four areas of interest: the physical, emotional, and social functioning of the child, which are the key components of health as defined by the World Health Organization (1948), and the child's school functioning. Disease-specific modules, which focus on specific issues of certain illnesses, are available that can be administered along with the PedsQL (i.e. asthma, cancer, cerebral palsy, brain tumour, fatigue, cardiac problems, rheumatology problems). A child self-report form, and a parent proxy parallel report form, appropriate to the child's age, are also available. So far, 122 papers have been published on the use of this generic instrument (see [www.pedsq.org](http://www.pedsq.org) for a complete list of PedsQL publications). The PedsQL has been mostly used to describe the quality of life of children suffering from various illnesses (e.g. diabetes mellitus, extreme obesity, cancer, etc.) and to study the factors associated with the quality of life of these populations (see appendix 1). The PedsQL is translated into many languages. However, there are only four papers published which investigate the adequacy of the psychometric properties of these translations (i.e., Norwegian, Chinese, UK-English, and German versions).

The PedsQL was created as a self-administered instrument. However, it would be beneficial to have this measure available in other formats such as in personal interviewer format, to be applied to children who are unable to understand written questions, to reduce the burden of readability, or to be applied when children are too fatigued, too ill, or too young to complete the questionnaire by their own (Matza et al., 2004). In all these circumstances, unless an interview format is implemented to complete the PedsQL, the information about the child's quality of life must be limited to what the parents/caregivers say. However, it has been extensively documented that parent-proxy reports of children's quality of life may be different from the children's self-reports of quality of life (e.g. Creemens et al. 2006b; Theunissen et al., 1998). Thus, the information provided by the interview might be fundamental to complement the

parent-proxy reports. Nevertheless, and although this alternative mode of administering the PedsQL has already been used (i.e., Palmer and her colleagues (2007) used this mode only in a few cases who were not capable of completing the self-administered questionnaire), no data has been made available about the psychometric properties of the PedsQL when administered in this way.

The purpose of this study was twofold. The first purpose was to describe the development of a self-administered and an interviewer-administered version of the Pediatric Quality of Life Inventory Generic Core Scales in Catalan\*. These versions were administered to a sample of schoolchildren. From these data, the internal structure of both versions was investigated. It was expected that both versions would have four factors corresponding to their four scales (i.e. physical, emotional, social and school functioning) and two higher-order factors (i.e. physical and psychosocial health summary). Reliability and validity of both versions were also examined. Given the inevitable differences when health is assessed from the parents' and children's perspectives, the validation against subjective measures was mainly based on information collected from children. Specifically, we hypothesized that both versions would allow us to differentiate healthy children from children with any medically identified illness, children who had suffered from any minor illness in the month previous to the assessment, and/or children who suffered from chronic pain (*construct validity*). Both versions would also correlate with another established measure of quality of life (*convergent validity*), and they would correlate with measures of children's functioning and indicators of interference of the children's health status on parents' daily routines (*predictive validity*). The second purpose was to provide some preliminary data about the concordance between these two Catalan versions after being validated.

## **2. Method**

### **2.1. Participants**

A representative sample of schoolchildren in grades 4 to 11 in Tarragona (a province of Catalonia, Spain) were invited to participate in this study (N=902). They were recruited from non-special primary and secondary schools. Exclusion criteria for the study were as follows: (1) the child had a cognitive impairment, and (2) the family was illiterate or unable to understand Catalan. Of the total sample invited, 511 decided to participate (61.09% child participation rate). The parents of 449 of these children were also involved (49.78% parent participation rate). The participating children did not differ from the eligible children who were invited but did not participate on school grade or sex. The age of the participating children age ranged from 9 to 17 years (mean age=11.73; SD=1.93) and 51.7% of the sample were males. Table 1 presents the relevant socio-demographic and medical data of the participating schoolchildren.

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\* Catalan is a language spoken by well over 10 million people in eastern Spain, south-western France, and the Island of Sardinia (located in the middle of the western Mediterranean).

## **2.2. Procedure**

The protocol followed was reviewed and approved by the Department of Education of the Government of Catalonia, and by the boards of the participating schools. A total of 40 classes from grades 4 through 11 of 20 primary and secondary schools were randomly selected using a stratified sampling procedure from all eligible non-special education schools in Tarragona. All the schools approached agreed to participate, except for one which was involved in several other research projects at the time. After the schools had given their agreement, all children attending these classes and their parents were invited to participate. The parents were sent a letter in which they and their children were asked to participate. Children whose parents provided them with informed consent were individually interviewed by two female interviewers at their school; they were asked to provide information about various domains of their health status and about the impact of their health status on their daily life.

Because the interview was concerned with past experiences (i.e. children were requested to recall their pain experiences over the three months before the interview and they were also asked to make judgment about their quality of life in the month previous to the interview), memory enhancing techniques were used (Orbach and Lamb, 2000). For example, interviewers used open-ended questions, and important events as end point times, in order to prevent interviewer bias from affecting children's reports, and to increase the accuracy of the information. Several other quality control measures were implemented during data collection. First, the interviewers attended standardization courses in which they were instructed and trained on how to conduct the interview. Second, data sheets were carefully checked immediately after the interviews, and further monitored centrally to check for missing data or inconsistencies.

The parents of the participating children were surveyed separately. They were requested to provide some additional socio-demographic data (see below), and information about the child's health status.

### *Linguistic validation of the PedsQL™ 4.0*

A Catalan version of the Pediatric Quality of Life Inventory Generic Core Scales (PedsQL, Varni et al., 2001) was developed using a back-translation procedure according to the guidelines suggested by the Mapi Research Institute (2002) and Dr. J. W. Varni. First, two native Catalan psychologists who had a good command of English translated the questionnaire into Catalan. Then, a professional translator, who was not familiar with the original version of the questionnaire, back-translated this Catalan version into English. Then, the new English version (back-translated version) was sent to the questionnaire's author to check whether the back-translation was appropriate (i.e. whether it preserved the denotation and connotation of the original). Some minor changes were made in the Catalan version following the recommendations of the author, to solve slight discrepancies between the back-translated English version and the original. Then this second version was tested on a sample of 10 children.

An interviewer-version of the Catalan version of the PedsQL was also created. Because consistent wording between modes can lead to inconsistent administration if the questions are not easy to administer in the given mode, the original statements of the self-administered version were formulated as questions (i.e., questions that were developed to be read by the respondent were changed to questions that would be heard by the respondent). After being translated by a professional translator, this new version was sent to Dr. Varni for his approval.

Both versions of the PedsQL™ 4.0 were answered by all the participants with one day between administrations, in order to avoid memory bias as much as possible. Thus, the same participants completed both self-administered and interviewer-administered versions. The order of administration of both versions was randomized to eliminate the order effect.

## **2.3. Measures**

### *2.3.1. Socio-demographic measures*

Information about age, gender, and school grade was collected at the time of interview. Based on the work of Domingo-Salvany et al. (2000) on the measurement of socio-economic status, the highest occupation of the two parents was the one used to rate the socio-economic status (SES) of the family and the child.

### *2.3.2. Quality of life*

The Pediatric Quality of Life Inventory Generic Core Scales (PedsQL, Varni et al., 2001) was used to assess the children's perception of their quality of life. The PedsQL is a 23-item generic instrument that encompasses four multidimensional scales (physical, emotional, social and school functioning) and three summary scales (total, physical health summary, psychosocial health summary). Developmentally appropriate versions (i.e. ages 8–12 and 13–18) were used. The child was asked how much of a problem each item had been in the past month. Items were scored on a 5-point Likert scale, from 0 (never a problem) to 4 (almost always a problem). Items were reverse-scored and linearly transformed to a 0-100 scale (0=100, 1=75, 2=50, 3=25, 4=0), so that higher scores indicated a better quality of life. The feasibility, reliability and validity of the instrument against, for example, different health conditions have been demonstrated in both clinical and school populations (Varni et al. 2001, 2006).

The Questionnaire for measuring the health-related quality of life in children and adolescents-revised version (KINDL, Ravens-Sierberer and Bullinger, 1998) was also used to assess children's perception of their quality of life. The KINDL is a 24-item generic instrument which yields six scales: (1) the Physical Well-being Scale, (2) the Emotional Well-being Scale, (3) the Self-esteem Scale, (4) the Family Scale, (5) the Friends Scale, and (6) the School Scale. Two different forms, developed for different age ranges (i.e. ages 8-11, and 12-16), were used. The KINDL has shown suitable internal consistency and construct validity against self-perceived health status in schoolchildren living in Catalonia (Rajmil et al. 2004b).

### 2.3.3. Pain

We asked the participating schoolchildren whether they had been experiencing pain in the preceding 3 months to the interview, and if they had, additionally they were asked for how long they had been experiencing that pain.

In this study, *chronic pain* was defined as pain lasting for at least three months (see Elliott et al., 1999), that was experienced continuously or recurrently. A recurrent pain problem was one occurring once or twice at least each month (see Mikkelsen et al. 1997 for a similar procedure).

### 2.3.4. Medical history

Information regarding the child's use of health care was provided by the parents. More specifically, parents were asked whether their children had been hospitalised or received any surgical treatment during their life. If so, they were asked how many times, and the reason for their being in hospital or receiving surgical treatment. They were also asked how many times their child had visited a health care professional in the preceding year.

Parents were also requested to provide additional information regarding their children's current medical condition: that is, whether they had any illnesses at the time of interview. In order to gather this information, a list of illnesses was presented to them to choose from. Parents were asked to give a "Yes" or "No" response to each illness; if the appropriate illness was not on the list, parents were asked to report it. They were also asked whether their children had suffered from any acute and minor illness in the previous month to the assessment.

### 2.3.5. Functional disability

The Functional Disability Inventory (FDI; Walker and Greene, 1991) was also used in this study. The FDI assesses 15 activities in a variety of contexts including school, home, recreation, and social interaction. The children are asked how much difficulty they had doing these activities because of their physical health, using a 5-point severity scale ranging from 0 (no trouble) to 4 (impossible). The total score is obtained by summing the ratings. Thus, the greater the final score, the greater the level of functional disability is. The internal consistency of the scale is high. As to validity, the scale correlates with other measures of school-related disability, and somatic symptoms, for example, in clinical and healthy samples (Walker and Greene, 1991, Claar and Walker, 2006, and Huguet et al., 2007).

### 2.3.6. Impact of the children's health status on their parents' daily routine

Parents were asked how much their child's health status had interfered with their own daily activities in the preceding month. An 11-point numerical scale was used, with the following anchors: 0 = It did not interfere at all; 10 = It interfered a lot.

#### **2.4. Data analysis plan**

To examine the factor structure of the self-administered version of the PedsQL a confirmatory factorial analysis (CFA) was performed using LISREL version 8.5; its factor structure had already been validated (e.g. Varni et al., 2001). Each item was assumed to load only on one factor such that items of the same subscale loaded on the same factor, the factors were allowed to correlate, and all residual correlations between items were zero. In the maximum likelihood approach to CFA, the chi-square test statistic was used to assess the goodness of fit of the model (Cole, 1987). However, because of the limitations associated with this statistical test as a result of the sample size effect (Marsh et al. 1988), several additional indices were also analyzed to better evaluate the goodness-of-fit of this model: namely, the Root Mean Squared Error of Approximation (RMSEA; Steiger, 1990), the Non-normed Fit Index, and the Comparative Fit Index (NNFI and CFI; Bentler and Chou, 1987). Since the model testing the factor solution recommended by Varni et al. (2001) was not supported by the data even after addressing some areas of misfit in the model, a new series of factor analyses were required. A cross-validation procedure was then performed. That is, the sample was randomly divided in two. Exploratory factor analysis (EFA) using a principal axis factoring procedure and oblimin rotation were performed on the first half to explore an alternative solution. The following aspects were considered in the building of a better solution: (a) the conceptual model and the original factors suggested by Varni et al. (2001); (b) the content of the items; and (c) the results provided by EFA. After that, CFA was applied on the second half of the sample to verify the EFA solution. To investigate the factor structure of the interviewer-administered version of the PedsQL, a confirmatory approach was used.

For both versions of the PedsQL (i.e. self-administered and interviewer-administered versions) the internal consistency analysis for the total PedsQL score, and for each of the subscales, was completed with Cronbach's alpha. For both versions, support for the *construct validity* was also sought by examining whether there were substantial differences between groups whose quality of life was known to differ. We used the t-test for independent samples, and analysis of variance; the effect sizes were calculated to determine the magnitude of the differences on the quality of life between groups (healthy children vs children with any diagnosed illness; healthy children vs children who had suffered from any minor illness in the month previous to the assessment; children with no chronic pain vs children with chronic pain). The *predictive validity* of both versions of the PedsQL was checked against functional disability and the impact of the children's health status on their parents' daily routines through correlations.

Finally, we examined the concordance between the self-administration procedure and the interviewer-administration procedure by looking at correlations, and the difference of means test. We also examined the effect of the child's age and gender on this concordance.

### 3. Results

#### 3.1. Descriptive characteristics of participating schoolchildren

##### 3.1.1. Medical history

According to the information reported by participating parents, 15% of the children experienced a complicated birth, 11.7% were premature (i.e. less than 38 weeks of gestation), and 7.8% were overdue (i.e. more than 42 weeks of gestation). See Table 1 for additional related information.

Table 1. Characteristics of the children and adolescents

	Scale range	Total sample mean (SD)
<b>Socio-demographic data</b>		
Age (years)	9-17	11.73 (1.93)
Gender (% , N)		
Boys	--	51.7 (264)
Girls	--	48.3 (247)
<b>SES<sup>a</sup> (% , N)</b>		
High	--	40.1 (180)
Medium	--	32.2 (145)
Low	--	27.6 (124)
<b>Medical history</b>		
Number of hospitalizations in their lives <sup>b</sup> (% , N)		
0	--	59.7 (259)
1	--	28.3 (123)
2	--	8.3 (36)
3	--	2.1 (9)
4	--	1.2 (5)
5	--	0.5 (2)
Number of surgical procedures in their lives <sup>c</sup> (% , N)		
0	--	75.9 (334)
1	--	19.1 (85)
2	--	3.6 (16)
3	--	1.1 (5)
4	--	0.2 (1)
<b>Current general health status</b>		
Medical diagnosis <sup>d</sup> (% of the total sample, n)		
Diabetes	--	0.7 (3)
Allergy	--	13.2 (58)
Asthma	--	3.2 (14)
Cardiology problems	--	2.3 (10)
Psychiatric/psychological problems	--	1.1 (5)
Others	--	5.2 (23)
Number of visits to healthcare professional during the preceding year <sup>e</sup> (% , N)		
0-2	--	62.3 (225)
3-5	--	29.9 (108)
6-10	--	5.8 (21)
>10	--	2.1 (7)

<sup>a</sup> Information missing in 62 cases

<sup>b</sup> Information missing in 77 cases

<sup>c</sup> Information missing in 130 cases

<sup>d</sup> Information missing in 72 cases

<sup>e</sup> Information missing in 150 cases

### **3.1.2. Current general health status**

According to the schoolchildren's reports, acute pain episodes were quite usual: 82.4% of the sample reported that they had experienced at least one episode of acute pain in the three months previous to the assessment, whereas 30.1% had suffered from chronic pain; these figures are very similar to those published in previous studies (e.g. Bloom et al., 2006; Perquin et al., 2000). Overall, participating schoolchildren reported a high level of quality of life (mean total KINDL score=77.03, SD=10.40), which is similar to the findings of several other previous studies (e.g. Rajmil et al. 2004b, Serra-Sutton et al. 2006). The positive self-perception of their health was also supported by reports from their parents. That is, the parents rated their child's quality of life very positively (mean=8.44, SD=1.71; 0-10 rating scale), and reported that the child's health status interfered little with their daily routine (mean=0.69, SD=1.36; 0-10 rating scale). The functional disability levels related to physical health reported by participating children were quite low (mean FDI score = 6.00, SD = 7.20), particularly if they are compared with the results from other studies with clinical samples (see for example, Eccleston et al., 2004; Kashikar-Zuck et al., 2001; Lynch et al., 2006; Peterson and Palermo, 2004). See Table 1 for information provided by participating parents about children's current general health status.

## **3.2. Psychometric properties of the PedsQL – Catalan version**

### **3.2.1. Feasibility**

#### **3.2.1.1. Self-administered version**

Missing values were examined to determine the feasibility of the self-administered version. When the PedsQL was self-administered, 0.24% of the items went unanswered. Floor effects were almost inexistent, and ceiling effects were highest for the Physical Health Summary and Social Functioning (see Table 5).

#### **3.2.1.2. Interviewer-administered version**

When the PedsQL was interviewer-administered, all items had a response. The percentage of participants scoring at the floor and ceiling of each PedsQL scales were similar between both modes of administration (see Table 5).

Both versions took very little time to be answered (less than 5 minutes).

### **3.2.2. Confirmatory factorial analysis**

#### **3.2.2.1. Self-administered version**

A confirmatory factorial analysis was performed to examine the validity of the second-order PedsQL factorial structure suggested by Varni et al. (2001). Although five first-order factors emerged from the EFA conducted by Varni and his colleagues (2001), the measure was modelled as a second-order structure whose first-order factors assessed four components instead of five (i.e. physical, emotional, social and school functioning) as dictated by their theoretical model. The estimation of this model resulted in goodness of fit statistics that indicated that the model represents a poor fitting model to the data (see Table 2). In an attempt to find a good fitting model, items with primary factor loadings below 0.30 were removed from

the model (i.e. items # 5, 6, 22 and 23<sup>1</sup>), and a large theoretical sense modification index suggested by LISREL was used (i.e. item # 8 was related to emotional functioning instead of physical functioning). Turning to the goodness-of-fit results, although improvement in overall fit was seen along the series of CFA, all parameter values still remained in an inappropriate range (see Table 2). Additionally, taking into account the internal structure with five first-order factors derived from EFA by Varni and colleagues' work (2001) as well as the relationship between item # 22 and 23 which loaded poorly with their hypothesized primary factor in our data, a model of five first-order factors was also tested. The overall fit of the model, however, was not good (see Table 2).

*Table 2. The goodness-of-fit measures for self-administered and interview-administered version of the PedsQL.*

	Models	Chi-square	df	RMSEA	NNFI	CFI
Self-administered version	Original 23-item version: Four first-order factors load on two higher-order factors	868.30**	226	0.078	0.77	0.79
	Removal item #5	810.84**	205	0.080	0.77	0.80
	Removal item #6	730.09**	185	0.080	0.78	0.81
	Removal item #23	559.34**	166	0.071	0.83	0.85
	Removal item #22	474.77**	148	0.069	0.85	0.87
	Item #8 related to emotional functioning	398.70**	148	0.060	0.87	0.92
	Five first-order factors load on two higher-order factors (23 items)	681.72**	225	0.066	0.83	0.85
Interviewer-administered version	Short 12-item version: Four first-order factors load on two higher order factors	86.63*	50	0.056	0.92	0.94
	Original 23-item version: Four first-order factors load on two higher-order factors	902.19**	226	0.077	0.72	0.75
	Short 12-item version: Four first-order factors load on two higher order factors	113.43**	50	0.050	0.93	0.95

\* p<0.01; \*\*p<0.001

At this point, the model was reestimated within an exploratory, rather than a confirmatory mode. This exploratory approach was performed with a random first half of the sample who answered in full the self-administered version of the PedsQL (N=231). Consistently with results of the factor analysis reported in previous works (McCarthy et al., 2005; Reinfjell et al., 2006, Varni et al. 2001), a principal axis factor analysis yielded five eigenvalues larger than unity (Factor 1, eigenvalue 6.3, explaining 27.63% of variance; factor 2, eigenvalue 1.87, explaining 8.15% of variance; factor 3, eigenvalue 1.55, explaining 6.72% of variance, factor 4, eigenvalue 1.46, explaining 6.36% of variance; and factor 5, eigenvalue 1.41, explaining 6.15% of variance). The factors were rotated to oblique (oblimin) simple structures. Factors related to school functioning were loaded on two different factors as it is so reported in the original work by Varni and colleagues, and others (McCarthy et al., 2005; Reinfjell et al., 2006, Varni et al.

<sup>1</sup> The authors regret not being able to provide a complete list of items; the PedsQL is a copyrighted instrument.

2001). After deleting two items related to school functioning which loaded on a same factor (i.e. item#22 and 23), we forced the number of factors to be four to be consistent with Varni's model (i.e. physical, emotional, social and school functioning) and the items were rotated again. Then nine items were deleted from the exploratory derived scale because of the low-factor pattern coefficients (i.e., <0.40) with their hypothesized primary factor loading or because of the high-factor pattern coefficients (i.e., >0.30) with secondary factor loading. The factor loadings for the resultant short 12-item version is displayed in Table 3. To examine the higher-order factor structure of the four subscales (i.e. physical, emotional, social and school functioning), a principal axis factor analysis, specifying 2-factor solution, was performed in accordance with the Varni's model. The four subscales were calculated by averaging the items that loaded on each subscales. As it was expected, physical functioning subscale loaded on a higher-order factor labelled as physical health summary; and emotional, social and school functioning subscales loaded on another higher-order factor labelled as psychosocial health summary (see Table 4). Physical Health Summary and Psychosocial Health Summary, which were computed by averaging the items of the subscales that loaded on the two higher-order factors, were positively correlated ( $r=0.39$ ,  $p<0.001$ ).

*Table 3. Exploratory factor analysis: Factors loadings by items of the short 12-item version*

Item	Factors			
	Health functioning	Emotional functioning	Social functioning	School functioning
Item #1	-0.202	-0.012	<b>0.659</b>	0.016
Item #2	0.142	0.057	<b>0.836</b>	-0.045
Item #3	0.249	-0.018	<b>0.741</b>	0.062
Item #10	-0.120	0.221	0.073	<b>0.654</b>
Item #11	-0.012	0.066	-0.027	<b>0.830</b>
Item #12	0.223	-0.144	0.021	<b>0.659</b>
Item #14	0.157	<b>0.646</b>	0.017	0.120
Item #15	-0.084	<b>0.760</b>	-0.003	0.090
Item #16	0.079	<b>0.881</b>	0.008	-0.105
Item #19	<b>0.728</b>	0.014	0.128	0.090
Item #20	<b>0.732</b>	0.006	-0.055	0.210
Item #21	<b>0.828</b>	0.126	0.007	-0.119

Extraction Method: Principal axis procedure. Rotation Method: Oblimin.  
 The solution converges in 11 iterations.  
 Bold indicates highest factor loading per item.  
 Sample size: N=236.

*Table 4. Exploratory factor analysis: Higher-factor loadings amongst the scales of the short 12-item version*

Scales	Factors	
	Physical health summary	Psychosocial health summary
Health functioning	<b>0.951</b>	-0.008
Emotional functioning	0.123	<b>0.753</b>
Social functioning	-0.180	<b>0.886</b>
School functioning	0.363	<b>0.551</b>

Extraction Method: Principal axis procedure. Rotation Method: Oblimin.  
 The solution converges in 6 iterations.  
 Bold indicates highest factor loading per scale.  
 Sample size: N=231.

The second half of the sample (N=236) was used to cross-validate the internal structure of the short 12-version derived from EFA. This model showed an acceptable fit to the data (Chi-square<sub>(50)</sub>= 86.63, p<0.01; RMSEA=0.056, NNFI=0.92, and GFI=0.94). Figure 1 shows the relationships amongst the items and the factors in the model by the standardized factor loadings.

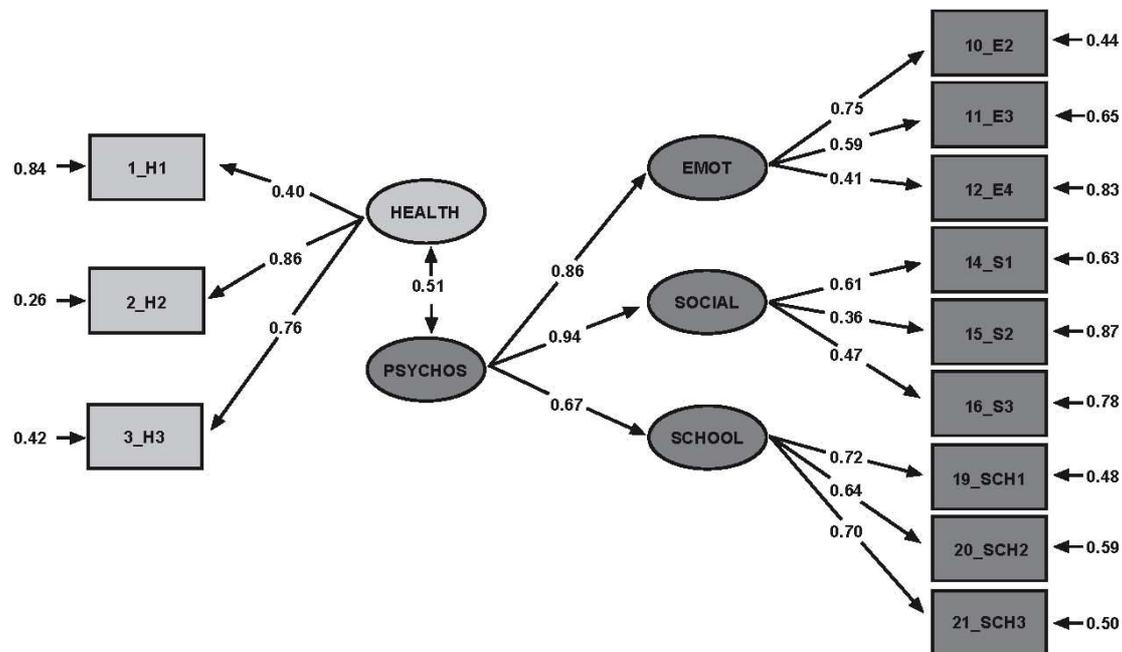


Figure 1. Second-order model of the self-administered short 12-item version of the PedsQL with standardized parameter estimates

### 3.2.2.1. Interviewer-administered version

As the transformations made to adapt the original self-administered version to be suitable for a new mode of administration (e.g., an interview) can fail to be effective, the factor structure of this new short 12-item version was evaluated through CFA. Both the original factor solution suggested by Varni et al. (2001) and the short 12-item version resulting from our data were examined. As can be seen in Table 2, the goodness-of-fit indices for our short 12-item version suggested a relatively good adjustment (Chi-square<sub>(50)</sub>=113.43, p<0.001; RMSEA=0.05; NNFI=0.93; GFI=0.95). Figure 2 shows the relationships amongst the items and the factors in the model by the standardized factor loadings.

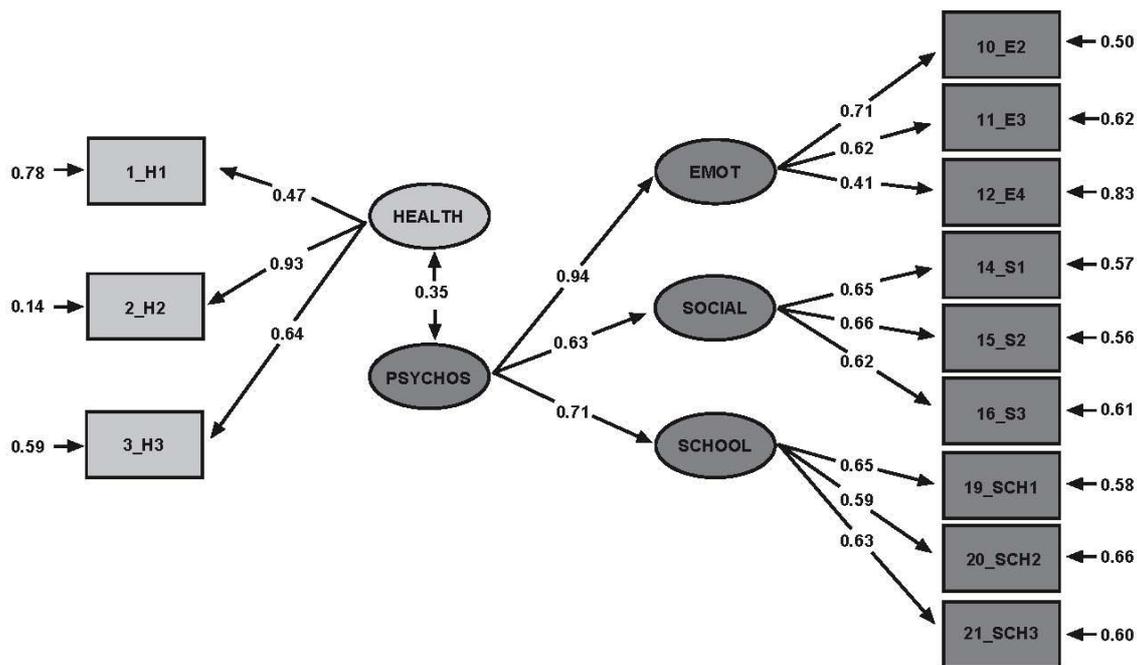


Figure 2. Second-order model of the interviewer-administered short 12-item version of the PedsQL with standardized parameter estimates

### 3.2.3. Internal consistency reliability

#### 3.2.3.1. Self-administered version

Although the internal consistency reliability for the total scale score was found to be good ( $\alpha=0.76$ ) when the measure was self-administered, not all the PedsQL subscales reached the minimum Cronbach's standardized coefficient alpha recommended for comparing groups (i.e.  $\alpha=0.70$ ), these subscales were mostly the second-order scales (see Table 5).

#### 3.2.3.2. Interviewer-administered version

When the measure was self-administered, the coefficient alphas for each subscale were quite similar (see Table 5).

Since the psychosocial subscales were not reliable enough, their validity was not examined.

Table 5. Descriptive scale, and internal consistency reliability values

	N. of items	N	Mean (SD)	Min.	Max.	Ceiling effects N (%)	Floor effects N (%)	Cronbach's alphas		
								Total sample	Ages 9-12	Ages 13-18
Self-administered version	Total score	474	81.53 (11.57)	25	100	14 (3)	0	0.76	0.80	0.76
	Physical Health Summary	490	88.26 (14.15)	33.33	100	211 (43.1)	0	0.69	0.69	0.68
	Psychosocial Health Summary	475	79.33 (12.98)	19.44	100	17 (3.6)	0	0.75	0.77	0.78
	Emotional Functioning	489	72.36 (18.11)	0	100	44 (7.7)	1 (0.2)	0.60	0.57	0.57
	Social Functioning	483	88.66 (14.45)	16.67	100	209 (43.3)	0	0.68	0.63	0.72
	School Functioning	485	76.65 (17.67)	0	100	62 (12.8)	2 (0.4)	0.65	0.74	0.70
	Total score	509	83.67 (11.10)	31.25	100	17 (3.3)	0	0.79	0.76	0.75
Interviewer-administered version	Physical Health Summary	510	89.49 (14.37)	25	100	246 (48.2)	0	0.69	0.71	0.63
	Psychosocial Health Summary	510	81.70 (12.78)	16.67	100	24 (4.7)	0	0.77	0.74	0.75
	Emotional Functioning	511	76.78 (18.49)	8.33	100	87 (17)	0	0.60	0.60	0.63
	Social Functioning	511	89.38 (14.85)	8.33	100	251 (49.1)	0	0.64	0.66	0.75
	School Functioning	510	78.90 (17.15)	0	0	87 (17.1)	1 (0.2)	0.74	0.61	0.70
	Total score	509	83.67 (11.10)	31.25	100	17 (3.3)	0	0.79	0.76	0.75
	Physical Health Summary	510	89.49 (14.37)	25	100	246 (48.2)	0	0.69	0.71	0.63

Floor effect: % of children and adolescents (for whom a score could be computed) who achieved the lowest possible score (i.e. 0); Ceiling effect: % of children and adolescents (for whom a score could be computed) who achieved the highest possible score (i.e. 100).

### 3.2.4. Validity

#### 3.2.4.1. Self-administered version

The *construct validity* of the self-administered version of the PedsQL was evaluated by the known-groups method. The comparisons between mean group scores are shown in Table 6.

First, schoolchildren with a medical diagnosis (such as asthma, allergy, diabetes, cardiovascular disease, epilepsy, or migraine) were compared with healthy children. The hypothesis that healthy children, as a group, would score significantly higher on the PedsQL inventory was partially confirmed. In comparison with healthy children, although children with a medical diagnosis did not have lower total scores ( $t=1.69$ ,  $p=0.09$ ), they had lower physical functioning ( $t=2.11$ ,  $p<0.05$ ), and the effect size was small to medium. Their psychosocial functioning was not lower ( $t=1.16$ ,  $p=0.24$ ).

Second, the PedsQL scores from those schoolchildren who had had an acute or minor illness in the month previous to the interview (such as influenza, gastroenteritis or cough) were

compared with the scores from those children who reported not having suffered any illness. Children with a minor illness tended to have lower scores on physical functioning than those without (mean= 85.70, and 88.93, respectively), but contrary to our hypothesis, these differences did not achieve statistical significance ( $t=1.83$ ,  $p=0.06$ ).

Finally, we compared the scores of schoolchildren who reported chronic pain and those who did not and our hypothesis was confirmed. Namely, the mean total scores on PedsQL differed significantly such that children with chronic pain had a lower quality of life than children without ( $t=5.35$ ,  $p<0.001$ ), and both the physical and psychosocial dimension of the quality of life had a significantly lower score ( $t=3.63$  and  $t=4.85$ ,  $p<0.001$ , respectively). Significant moderate effect sizes were found for all the domains examined.

*Table 6. Mean scores and standard deviations for each group, mean difference tests, and effect size for differences in means*

Groups	Self-administered version						Interviewer-administered version					
	Total score		Physical Health Summary		Psychosocial Health Summary		Total score		Physical Health Summary		Psychosocial Health Summary	
	N	Mean (SD)	N	Mean (SD)	N	Mean (SD)	N	Mean (SD)	N	Mean (SD)	N	Mean (SD)
<i>Current medical diagnosis</i>												
No diagnosis	312	82.28 (11.06)	325	89.00 (13.39)	313	80.11 (12.59)	341	84.71 (10.50)	341	90.71 (13.33)	342	82.67 (12.31)
Diagnosis	93	79.97 (13.20)	94	85.02 (16.78)	93	78.31 (14.42)	96	81.42 (12.28)	97	84.54 (17.22)	96	80.38 (13.91)
Mean diff. T score		1.69		2.11*		1.16		2.60*		3.26**		1.57
Effect size		0.21		0.30		0.14		0.31		0.46		0.19
<i>Minor disease in the last month</i>												
No disease	267	82.01 (11.73)	277	88.93 (13.98)	268	79.80 (13.41)	288	84.24 (10.82)	289	89.91 (14.08)	289	82.31 (12.72)
Disease	85	81.30 (11.95)	88	85.70 (15.52)	85	79.77 (12.72)	90	82.52 (12.02)	90	85.65 (17.03)	90	81.48 (12.64)
Mean diff. T score		0.48		1.83		0.02		1.28		2.15*		0.54
Effect size		0.06		0.23		0.00		0.16		0.30		0.06
<i>Chronic pain</i>												
No chronic pain	329	83.37 (10.73)	340	89.88 (13.25)	329	81.21 (12.03)	356	85.47 (10.14)	356	90.73 (13.82)	357	83.68 (11.57)
Chronic pain	145	77.37 (12.33)	150	84.61 (15.44)	146	75.09 (14.03)	153	79.48 (12.10)	154	86.63 (15.23)	153	77.08 (14.25)
Mean diff. T score		5.35***		3.63***		4.85***		5.36***		2.97**		5.05***
Effect size		0.56		0.40		0.51		0.59		0.30		0.57

\*  $p<0.05$ ; \*\* $p<0.01$ ; \*\*\* $p<0.001$

*Convergent validity*, which supports the construct validity of the self-administered version of PedsQL, was assessed through correlations with the KINDL. The interrelationships among the scales of both quality of life measures are presented in Table 7. The PedsQL total score correlated with the KINDL total score ( $r=0.36$ ,  $p<0.001$ ), the Physical Health Summary was significantly related to the Physical Well-being Scale of the KINDL ( $r=0.32$ ,  $p<0.001$ ), and the Psychosocial Health Summary was moderately related to the scales of the KINDL, which assess different psychosocial aspects of the quality of life, with the exception of two cases (see Table 7). As far as the first exception is concerned, there was no relationship between scores on the Psychosocial Health Summary and scores on the Self-esteem Scale ( $r=-0.11$ ,  $p=0.21$ ), but self-esteem is a psychological component that is not assessed by the PedsQL. As far as the second exception is concerned, there was a small correlation between scores on the Psychosocial Health Summary and scores on the Family Scale ( $r=0.21$ ,  $p<0.05$ ), but family functioning is not directly assessed by the PedsQL. Moreover, as could be expected, the correlations between the two quality of life measures tended to be higher on the subscales assessing similar domains (e.g. the Physical Well-being Scale of the KINDL with the Physical Health Summary of the PedsQL).

Table 7. Pearson correlation coefficients between PedsQL scales and KINDL scales

		Total score	Physical Well-being Scale	Emotional Well-being Scale	Self-esteem Scale	Family Scale	Friends Scale	School Scale
Self-administered version	Total score	0.36***	0.29*	0.35*	-0.10	0.25**	0.36***	0.42***
	Physical Health Summary	0.26**	0.32***	0.17	-0.04	0.23**	0.30***	0.24**
	Psychosocial Health Summary	0.34***	0.26**	0.37***	-0.11	0.21*	0.33***	0.41***
Interviewer-administered version	Total score	0.48***	0.42***	0.50*	-0.06	0.34***	0.40***	0.43***
	Physical Health Summary	0.28***	0.34***	0.30***	-0.03	0.22**	0.21*	0.17*
	Psychosocial Health Summary	0.47***	0.38***	0.49***	-0.07	0.32***	0.40***	0.46***

\* $p<0.05$ ; \*\* $p<0.01$ ; \*\*\* $p<0.001$

The *predictive validity* of the self-administered version of the PedsQL was supported by examining the association between quality of life (PedsQL) and the impact outcomes: interference with parents' daily routines due to the child's state of health and functional disability associated with physical status. As was expected, quality of life had a negative relationship with the impact on parents' daily routines ( $r=-0.23$ ,  $p<0.001$ ). Those children whose parents reported higher levels of interference in their life due to their child's health status had significantly lower Physical and Psychosocial Health Summary scores ( $r=-0.14$ ,  $p<0.01$ , and  $r=-0.22$ ,  $p<0.001$ , respectively).

The functional disability outcome variable correlated significantly with both physical and psychosocial dimensions and, as was predicted, the strongest correlations were with physical functioning rather than with psychosocial functioning. Those children who reported higher levels of functional disability had lower physical functioning scores ( $r=-0.28$ ,  $p<0.001$ ), and lower psychosocial functioning scores ( $r=-0.21$ ,  $p<0.001$ ).

#### 3.2.4.2. Interviewer-administered version

Similar results for interviewer-administered version of the PedsQL were found. As far as construct validity data is concerned, there were significant differences between children with a medical diagnosis and healthy children as it was expected. That is to say, the ones with a medical diagnosis reported lower levels of quality of life ( $t=2.60$ ,  $p<0.05$ ) and lower levels of physical functioning ( $t=3.26$ ,  $p<0.01$ ). There were also significant differences in physical functioning between children who had had an acute or minor illness in the month previous to the interview and children who had not ( $t=2.15$ ,  $p<0.05$ ). And there were significant differences between children with chronic pain problems and children without chronic pain in all quality of life domains ( $p<0.01$ ) (see Table 6).

As far as convergent data is concerned, although the correlations among the scales of interviewer-administered version of the PedsQL and the KINDL tended to be a bit higher than the correlations among the scales of the self-administered version of the PedsQL and the KINDL, data converged on a similar correlational pattern. The correlations are displayed in Table 7.

Finally, the results obtained from analyses conducted to examine the predictive validity of the interviewer-administered version also supported our hypotheses. Total, Physical Health Summary, and Psychosocial Summary scores were significantly related to both interference with parents' daily routines due to the child's state of health ( $r=-0.2$ ,  $p<0.001$ , and  $r=-0.11$ ,  $r=-0.19$ ,  $p<0.05$ ; respectively) and functional disability associated with physical status ( $r=-0.36$ ,  $r=-0.34$ , and  $r=-0.28$ ,  $p<0.001$ ; respectively).

### 3.3. Equivalence between the self-administered version and the interviewer-administered version of the PedsQL

The total scores, as well as the Physical, and Psychosocial Health Summary scores on the self-administered and the interviewer-administered versions were highly correlated ( $r_{\text{Total scores}}=0.73$ ,  $r_{\text{Physical Health Summary}}=0.65$ , and  $r_{\text{Psychosocial Health Summary}}=0.72$ ). However, there were significant differences in the PedsQL scores between the self-administration and the interviewer-administration of the inventory. Lower levels of quality of life were reported when participating schoolchildren completed the self-administered version of the PedsQL. Gender also affected the concordance between the self-administration and the interviewer-administration: agreement between both versions was greater in girls than in boys. The pattern of equivalence between the two versions was the following: boys and girls tended to report lower quality of life and lower psychosocial functioning when the PedsQL was self-administered, but not lower physical functioning. As far as the impact of the child's age is concerned, when

children completed the interviewer-administered version, while the older group while the older group scored higher on total quality of life, physical and psychosocial functioning, the younger group scored higher on total quality of life and psychosocial functioning when they answered the interviewer-administered version (see Table 8).

*Table 8. Comparison between scores on the interviewer-administered version and scores on the self-administered version for the total sample, and according to the child's gender and age groups.*

	Total sample							
	Mean (SD)		$t_{(gII)}$	r				
	Interviewer-administered version	Self-administered version						
Total score	83.81 (11.03)	81.59 (11.56)	5.85**	0.73				
Physical Health Summary	89.48 (14.43)	88.32 (14.10)	2.15*	0.65				
Psychosocial Health Summary	81.94 (12.73)	79.34 (12.99)	5.92**	0.72				
	Boys				Girls			
Total score	84.54 (10.43)	81.68 (11.43)	5.16***	0.69	83.05 (11.59)	81.49 (11.72)	3.01**	0.77
Physical Health Summary	90.64 (13.79)	89.19 (13.44)	1.88	0.59	88.26 (15.01)	87.41 (14.74)	1.13	0.70
Psychosocial Health Summary	82.50 (12.25)	79.15 (12.98)	5.34***	0.70	81.36 (12.13)	79.54 (13.01)	2.97**	0.75
	Younger group (9-12 years old)				Older group (13-17 years old)			
Total score	84.51 (11.09)	82.52 (11.58)	4.64***	0.73	81.23 (10.48)	78.12 (10.86)	3.76***	0.70
Physical Health Summary	88.95 (15.15)	88.40 (14.08)	0.89	0.66	91.29 (11.48)	88.06 (14.24)	3.08**	0.65
Psychosocial Health Summary	83.01 (12.57)	80.55 (12.94)	5.01***	0.72	77.94 (12.62)	74.86 (12.20)	3.20***	0.70

\* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$

#### 4. Discussion

A Catalan interviewer-administered version of the originally self-administered paper-and-pencil instrument was constructed after an accurate translation and cross-cultural adaptation. Both the self-administered and the new interviewer-administered versions of the PedsQL were tested with a large sample of Catalan-speaking schoolchildren aged between 9 and 17 years old. After collecting data, the factor structure of both versions was examined, the original 23-item version was rejected in both cases and a shorter 12-item version suggested instead. The results from factor analyses of the original 23 items in the current study resembled some findings related to problematic items of previously published works (see McCarthy et al. 2005, Reinfjell et al. 2006, Varni et al. 2001), which is specially relevant considering the limited number of published works reporting the factor structure of the PedsQL (i.e. Bastianansen et al., 2004, McCarthy et al., 2005, Reinfjell et al., 2005, Varni et al., 2001). Taken together, both self- and interviewer-administered short 12-item versions showed comparable reassuring statistical properties albeit with a few areas needing improvement.

The internal consistency indices for the three summary scales of both measures were satisfactory for group comparison, but sometimes insufficient for the second-order scales. This may be partly due to the small number of items in each of these scales. The construct validity of both versions was supported by their ability to discriminate children with different health status; specifically, between healthy children and children with a medical illness, and between children with chronic pain problems and children with no chronic pain. These results echo those reported in previous studies (e.g., Connelly and Rappof, 2006, Hunfeld et al. 2001, Ostkirchen et al., 2006, Palermo 2001, Varni et al. 2003, 2004, 2006). However, we found a lack of ability to distinguish between children who had had a minor illness in the month before the interview and children who had not in the self-report version of the PedsQL. This finding might be partially explained by the high ceiling effect on the higher-scoring children on physical scale, which restricts the ability to discriminate differences amongst higher-scoring children on this scale. In fact, skewed score distributions towards the end of the PedsQL scales have already been reported (e.g. Felder et al., 2004). The significant correlations between the PedsQL scales and most of the KINDL scales provided some additional evidence of the construct validity of both versions. The low correlations between the Self-esteem subscale and the Family subscale of the KINDL and the PedsQL scales can be attributed to the discrepancies between both quality of life measures. That is to say, although both PedsQL and KINDL assess quality of life, they address different domains (see Rajmil et al. 2004a). Apart from that, the fact that the KINDL scales correlated more with the interviewer-administered PedsQL scales than with the self-administered PedsQL scales may be attributed to the mode of administration: the KINDL scales were administered face-to-face. Some evidence of predictive validity of the self- and interviewer-administered versions was also provided by our data. Thus, the 12-item self-administered version performs as well as the original 23-item version in terms of construct, convergent, and predictive validity.

Further work on both 12-item versions of the PedsQL is necessary. First, the ceiling effects need to be reduced, possibly including items of greater difficulty. Second, the internal consistency for the second-order scales needs to be improved by developing new items with good psychometric properties. Third, additional studies need to be done to replicate these results, and to investigate the temporal reliability of both versions. Fourth, in order for these versions to be useful in clinical evaluations, studies with clinical samples also need to be conducted.

On the basis of our results, it is questionable whether the self-administration and the interviewer-administered short 12-item version of the PedsQL can really be considered comparable because lower levels of quality of life were reported in the self-administered version of the PedsQL. Therefore, clinicians and researchers within the context of pediatric health should be cautious about comparing quality of life information within-subjects and between-subjects using different forms of the questionnaire. If these results on the non-equivalence are confirmed in the future, additional studies should be conducted so as to determine which factors might be responsible for the discrepancies. Although mainly through adult studies, there are

some reports which support the non-equivalence between different modes of administration of quality of life measures (e.g., Beigner et al., 1981, Jörnagarden et al., 2006, Lyons et al., 1999, Perkins and Sanson-Fisher, 1998, Schunemann et al., 2003, Weignberger et al., 1994). At this point we can not conclude whether the discrepancy between responses obtained from both versions is due to the mode of administration, to the minor wording differences between both versions, or both of the above. At any event, our data are consistent with the data reported by previous studies showing a certain effect of the mode of administration in responses to health-related questions. These previous studies have suggested that response within an interview context shows a more social desirability bias than response collected from self-report measures. Thus, this issue could explain why interviewer-version of the PedsQL yielded a more positive quality of life from the participating children than the self-administered version did. This social desirability hypothesis has also been pointed out in studies focused on health-related behaviors such as drugs and alcohol consumption or sexual behavior (e.g. Aquilino and Losciuto, 1990). Moreover, our study showed that gender and age may also mediate these effects (see Jörnagarden et al., 2006 for similar results). Specifically, on one hand, since only female interviewers were involved, the answers from male children could have been more altered. Moreover, the tendency of boys to be more resistant than girls to verbally sharing information about their status of well-being, could also have had an effect. On the other hand, younger children might have been less affected by social desirability than older children when they rate their physical aspect of quality of life. In any case, future studies will have to focus on what can influence the results, and provide rationales so that clinicians and researchers can take informed decisions about how to best administer the PedsQL in each case.

There are some limitations within this study. First, the medical information reported here was based exclusively on parental reports, and sometimes participating parents did not provide the requested information. Second, the children answered both self- and interviewer-administered versions with a short interval between administration, consequently children's responses from the second administration may have been biased by a potential learning effect.

In conclusion the results of the present study suggest that both the self-and interviewer-administered short 12-item versions of the PedsQL are reliable and valid but they are not equivalent. Although the child's participation rate was not as high as other studies report due to time consumption, the large sample size, the randomized study design, and the consistencies between our findings with those from previous published works, provide some guarantee that our results may be fairly generalizable to the population of Catalan-schoolchildren aged between 9 and 17 years.

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## **Appendix 1. Pediatric Quality of Life Inventory Generic Core Scales publications classified according to their use**

<b>1. Publications studying the psychometric properties of PedsQL™ Pediatric Quality of Life Inventory Generic Core Scales</b>	
General population	Varni et al., 2007b, Varni et al., 2007c, Varni et al., 2006a; Varni et al., 2003a, Varni et al., 2001; Upton et al., 2005.
Clinical population	Varni et al., 2007b, Varni et al., 2007c, Varni et al., 2006b; Varni et al., 2004b; Varni et al., 2003b; Uzark et al., 2003; Varni et al., 2002a; Varni et al., 2002b; Varni et al., 2002c; Varni et al., 2001; Varni and Burwinkle, 2006; Goldstein et al., 2006; Connelly and Rapoff, 2006; McCarthy et al., 2005; Bastiaansen et al., 2004a.
<b>2. Publications studying the psychometric properties of PedsQL™ PedsQL Modules</b>	
	Varni et al., 2007a, Varni et al., 2006b; Palmer et al., 2007; Chan et al., 2005; Varni et al., 2004a; Varni et al., 2004b; Varni et al., 2003b; Uzark et al., 2003; Varni et al., 2002a; Varni et al., 2002b; Upton et al., 2005.
<b>3. Publications studying the psychometric properties of different forms of the PedsQL™ Pediatric Quality of Life Inventory Generic Core Scales</b>	
	Chan et al., 2005; Varni et al., 1999, Sherman et al., 2006.
<b>4. Publications developing different cultural or language versions of PedsQL™ Pediatric Quality of Life Inventory Generic Core Scales</b>	
	Reinfjell et al., 2006; Upton et al., 2005; Chan et al., 2005; Felder-Puig et al., 2004.
<b>5. Publications attempting to describe quality of life</b>	
General population	Mansour et al., 2003.
Clinical population	McCarthy et al., 2006; Zeller et al., 2006; Felder-Puig, 2006; Thomas et al., 2006; Youssef et al., 2006; Zeller and Modi, 2006; McCarthy et al., 2006; Parekh et al., 2006; Pinhas-Hamiel et al., 2006; Lau et al., 2006; Ismail et al., 2006; Favreau et al., 2006; McKenna et al., 2006; Johnson et al., 2006; Goyal et al., 2006; Bhat et al., 2005; Sawyer et al., 2005; Youssef et al., 2005; Eiser et al., 2005a; Eiser et al., 2005b; Pongwilairat et al., 2005; Moorthy et al., 2005; Sheppard et al., 2005; Talarska, 2005; Alpert et al., 2005; Lindman et al., 2005; Biard et al., 2005; Meeske et al., 2004; Bastiaansen et al., 2004a; Crabtree et al., 2004; Powers et al., 2003; Friefeld et al., 2004; De Clercq et al., 2004; Curran et al., 2004; Schwimmer et al., 2003; Powers et al., 2003; Bucuvalas et al., 2003; Laffel et al., 2003, Curran et al., 2003.
<b>6. Publications using the inventory to assess the effectiveness of a treatment</b>	
	Seid et al., 2006; Razzouk et al., 2006, Hays et al., 2006; Valenzuela et al., 2006; Riddle et al., 2006, Rudolf et al., 2006; Hicks et al., 2006; Wigal et al., 2005; Mangione-Smith et al., 2005; Hinds et al., 2005; Powell et al., 2005; Eiser et al., 2006; Rose et al., 2005; Chase et al., 2005; Schwimmer et al., 2005; Soo et al., 2005; Alpert et al., 2005; Packman et al., 2005; Sallee et al., 2004; Razzouk et al., 2004; Youssef et al., 2004; Poretti et al., 2004; Packman et al., 2004.
<b>7. Publications using the inventory to examine the relationship between variables</b>	
PedsQL™ 4.0 as a predictor	Seid et al., 2005; Seid et al., 2004.
PedsQL™ 4.0 as an outcome measure	Berrin et al., 2006; Oddson et al., 2006; Hassan et al., 2006; Meeske et al., 2006; Mah et al., 2006; Valenzuela et al., 2006; Brunner et al., 2006; Thomas et al., 2006; Pirpiris et al., 2006; Zeller and Modi, 2006; Slomine et al., 2006; Grilli et al., 2006; The Pediatric Obesity Prevention Study, 2006; Ungar et al., 2006; Williams et al., 2005; Bastiaansen et al., 2005a; Bastiaansen et al., 2005b; Sawyer et al., 2005; Chan et al., 2005; Brunner et al., 2005; Casey et al., 2005; Moorthy et al., 2005; Brunner et al., 2004; Sawyer et al., 2004; Friefeld et al., 2004; Windle et al., 2004; Curran et al., 2004; Mansour et al., 2003; Bucuvalas et al., 2003; Laffel et al., 2003; Burdette et al., 2003.
<b>8. Publications using the inventory for other purposes</b>	
	Varni et al., 2005, Cremeens et al., 2006; Wysocki and The Diabetes Research in Children Network Study Group, 2006; Kolotkin et al., 2006; Varni et al., 2005; Young et al., 2005; Seid and Varni, 2005; Seid et al., 2004; Eiser et al., 2003.





**Paper #3:**

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# The Inventory of Parent/Caregiver Responses to the Children's Pain Experience (IRPEDNA): Development and preliminary validation

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## Abstract

This paper describes the development and preliminary validation of a self-administered instrument designed to measure parents/caregivers' responses to children's pain episodes. For empirical validation purposes, a 60-item inventory was answered by 401 adults whose children's ages ranged from 6 to 16 years (mean = 10.44, and SD = 2.25 years). Factor structure and item analyses led to a 37 item inventory with three interrelated scales, namely: solicitousness ( $n = 15$  items), discouragement ( $n = 10$  items), and promotion of well-behaviors and coping ( $n = 12$  items). The three scales had good internal consistency, with coefficient alphas of 0.87, 0.83 and 0.87, respectively; they also showed good criterion-related validity.

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*Keywords:* Pediatric pain; Assessment; Parent/Caregiver Responses to the Children's Pain Experience; IRPEDNA

## 1. Introduction

External social events and environmental contingencies are hypothesized to play a critical role in children's pain experience and suffering (see Craig and Riddell, 2003), and evidence supports a relationship between parental behavior and children's pain experiences, both in experimentally induced pain (Chambers et al., 2002) and clinical pain (Chambers, 2003).

The research performed to date in acute pain has focused mostly on parent–child interactions during painful medical procedures. For example, Sweet and McGrath (1998) found that increases in infants' pain behaviors during immunization were related to mothers' reassuring behavior. Similarly, Manimala et al. (2000) reported that more children required restraint during immunization if their parents provided reassurance (see also von Baeyer, 1997; Blount et al., 2001).

Research examining the impact of parental behavior on children's chronic pain experience, however, is sparse and much less conclusive. Walker et al. (1993) found that parents of children who suffer from recurrent abdominal pain provide more positive responses, hypothesized to have a reinforcing impact, after their children display pain behaviors. This finding has not, however, been supported by more recent work (see Brace et al., 2000; Merlijn et al., 2003). There also are studies reporting that parents of children who have high levels of functional disability related to chronic pain problems more often discourage their children's adaptive coping behaviors, and reward their children's pain behaviors, in comparison to parents whose children with chronic pain have low levels of functional disability (Dunn-Geier et al., 1986; Peterson and Palermo, 2004; Merlijn et al., 2006). But again, the influence of the parents' behaviors on the child's disability is not consistently supported by published reports (Reid et al., 2005).

Despite lack of agreement in existing findings, it is reasonable to hypothesize that parents are likely to have a substantial impact on children's pain experience. It is pos-

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sible that some of the lack of consistency in previous findings has to do with the lack of valid and reliable comprehensive measures of parental responses to child's pain behaviors.

One of the most significant limitations of the extant research on parental reactions in response to children's pain behavior has been the focus on one type of reaction; that of solicitous attending to children's signs of pain and distress (see Walker and Zeman, 1992). But other types of parental responses may also influence children's pain behavior and ability to cope (see Craig, 1978; Chambers et al., 2002). Moreover, available measures have been developed taking into account reactions from parents of children suffering from chronic disabling problems. But parental responses to different levels of pain severity need to be studied, if we are to understand how parental reactions influence pediatric pain experience; the development and maintenance of chronic pain and related disability in children. The aim of this study was to develop and validate a comprehensive instrument specific to the measurement of parents' and other caregivers' responses to children's pain that would allow for a more thorough evaluation of the role that parental responses might play in children's adaptation to pain.

## 2. Method

### 2.1. Subjects

Parents of 601 school-aged children and their child, recruited from non-special primary and secondary schools (from grade 1 to 11) in Tarragona (a province in Catalonia; Spain), were approached and invited to participate. Exclusion criteria for the study were as follows: (1) the child had cognitive impairment, and (2) the family was illiterate or unable to understand Catalan language. Before inviting the parents and their child, the authors identified, with the children's teachers, those who were not eligible for participation on the basis of the exclusion criteria described. Only 9 participants were excluded. In 8 of these cases, parents were excluded because they did not have sufficient command of Catalan language to complete the questionnaire. The remaining case was excluded because the child had an intellectual disability. Of the 601 parents invited to participate, 401 of them, along with their child, participated in the study (participation rate was 66.7%). Reasons for non-participation are unknown.

The questionnaire was completed in 53.6% of the cases by the mothers, in 9.5% of the cases by the fathers, in 24.7% of the cases by both parents (father and mother answered altogether the same questionnaire), in 0.3% of the cases by guardians, and in 11.9% of the cases the informant's identity was missing. Participating children were between 6 and 16 years (mean age = 10.44; SD = 2.25 years), 58% of them male. Ninety-two percentage of participating parents were married or co-habiting, 6.2% were separated or divorced, and 0.7% were widowers. 41.4% of the participating families had a high socio-economic status (SES), 31% had a medium SES, 22.3% had a low SES, and in 5.2% of the cases this information was missing.

### 2.2. Measures

#### 2.2.1. Parent measures

**2.2.1.1. Sociodemographics.** A questionnaire completed by participating parents was used to gather information about the age, current occupation, marital status, and sex of the informant. Based on the work of Domingo-Salvany et al. (2000) on the measurement of socio-economic status based upon the Spanish Classification of Occupations CNO-94, the highest occupation of the two parents was used to rate the SES of the family and the child.

**2.2.1.2. Parental response to the child's pain behaviors.** The Inventory of Parent/Caregiver Responses to the Children's Pain Experience or IRPEDNA for its Catalan name: *Inventari de Respostes dels Pares-Cuidadors davant l'Experiència de Dolor de Nens i Adolescents*, is a self-administered questionnaire with three subscales: (1) solicitousness, (2) discouragement, and (3) promotion of well-behaviors and coping. The structure, that is the three scales, as well as its content, that is the items included, are based on the *social learning model* of parental/caregiver influence supported in this paper. This model has been useful to investigate the influence of contextual forces in adults' pain experience, for example, to study the influence of significant others on other's pain behaviors (e.g., Flor et al., 1987; Romano et al., 1995), and provides an appropriate framework to investigate the interactions and reciprocal influence between parents and children. The model suggests that modeling and reinforcement of pain behavior and actions by adults, both positive reinforcement (such as attention or "solicitous" behavior by others in the social environment), and negative reinforcement (e.g., removal of normal responsibility when a child complains of pain), will contribute to children's responses to acute pain experiences and to the development and maintenance of chronic disabling pain problems (see Fordyce, 1976; Craig, 1978; Turk et al., 1983).

All the items reflect actions and behaviors that parents/caregivers may enact after observing the children's pain behaviors (see below for a detailed description of the development of the inventory). The respondents were asked to answer how often they enacted each of the reactions included in the inventory when their child, who was also participating in the study, was in pain [i.e., "How often do you... (item)... when your child is in pain?"]. Participating parents rated each item using a five-point response scale (0, never; 1, hardly ever; 2, sometimes; 3, often; 4, always), and both the father and the mother were asked to answer together the questionnaire.

#### 2.2.2. Child measures

**2.2.2.1. Sociodemographics.** Information about age and gender was collected. Days of school absence due to the child's health problems in the month preceding the interview were also reported by the parents.

**2.2.2.2. Pain.** The *Faces Pain Scale-Revised* (FPS-R, Hicks et al., 2001) was used to gather information about the children's usual and highest *pain intensity* in the preceding three months; in the case of children who reported to be suffering from more than one pain problem, they provided this information in relation to the most bothersome one. The FPS-R is a

six-point scale designed to measure children's level of perceived pain intensity. The child's task is to choose the face that best reflects the intensity of the pain she/he has experienced. A numeric value from 0 to 10 (0-2-4-6-8-10) is assigned to each face. The end points are explained as 'no pain' and 'very much pain'. We used the Catalan version of the scale, validated by Miró and Huguet (2004).

*Pain frequency* was measured by means of a categorial scale: more than once a week, about once a week, about once or twice a month, and once or twice in the last three months. The use of pre-coded categories is a usual strategy to assess the frequency of pain in pediatric populations (e.g., Perquin et al., 2000).

Some information about the *pain duration* was also recorded. Specifically, we asked the participating schoolchildren to report whether they had been experiencing pain for more than three months.

*Pain location* was coded according to a list based on the axis I of IASP Classification of Chronic Pain (Merskey and Bogduk, 1994a,b). This information was gathered through an open question about where their pain was located. At the same time, and based on the work of Mikkelsen et al. (1997), children were requested to mark and paint the painful area on a body figure, so as to avoid potential misunderstandings.

In this study, chronic pain was defined as a pain lasting for three months at least, that was experienced continuously or recurrently. A recurrent pain problem was one occurring once or twice at least each month (see Mikkelsen et al., 1997 for a similar procedure).

**2.2.2.3. Children's distress.** The negative affect scale of the child version of the Positive and Negative Affect Schedule (PANAS-C) (Lauren et al., 1999) was used to assess children's pain related distress. The PANAS-C is a standardized measure which consists of 20 adjectives, ten assessing positive affect (e.g., excited), and ten assessing negative affect (e.g., upset). These adjectives describe different feelings and emotions. The measure has shown good psychometric properties in a sample of Spanish children (Sandín, 2003). In the present study, children were asked to respond on a five-point scale, from 1 which means "not at all or almost not at all" to 5 "a lot", the extent to which each adjective describes their feelings when they are in pain, as opposed to describing their feelings in the last few days (as it so asks the original version of the instrument). An adapted Catalan version was used in this study (Huguet et al., 2007). Internal consistency (Cronbach's alpha) for the negative affect scale of the PANAS-C in this study was 0.79.

### 2.3. Procedure

Thirty-two classes, in grades 1 through 11 of 16 primary and secondary schools, were randomly selected using a stratified sampling procedure from all eligible non-special education schools in the Tarragona area (an area in the south east of Catalonia, Spain). All approached schools agreed to participate, except for one due to their involvement in several other research projects at the time. Following their approval, both the children attending these classes and their parents were invited to participate. Parents received a letter explaining the purpose of the study, and requesting their participation as well as the participation of their child. Parents were asked to provide sociodemo-

graphic data, and information about the child's school attendance, they were also requested to complete IRPEDNA and return all that information to the school teacher.

Children with an informed consent from their parents to participate in the study were individually interviewed, they were asked to provide information about pain problems experienced during the preceding three months, they were also asked to report about pain related affect. Those reporting having had pain problems in the last three months were asked to provide additional information concerning location, frequency, duration, and intensity of each pain complaint, and to identify the most bothersome pain site. The interviews took place at the children's school.

The protocol followed was reviewed and approved by the Department of Education of the Government of Catalonia, and by the participating school boards.

#### 2.3.1. Development of the questionnaire

The development of the measure followed different stages. First, a list of 60 statements referring to responses that adults (parents/caregivers) may enact in relation to their children's pain experience was generated by the two first authors (JM, AH). Items were derived from two sources: (1) existing questionnaires with similar objectives, available for the pediatric (the Illness Behavior Encouragement Scales, IBES; Walker and Zeman, 1992) and the adult population (the Significant Other Response Scale of the West-Haven Yale Multidimensional Pain Inventory, WYHMPI; Kerns et al., 1985; Okifuji et al., 1999), and (2) semi-structured interviews with 10 fathers and mothers of schoolchildren aged between 6 and 16 years, in which they provided information on how they reacted to their children's pain experience. One of the objectives was to have as much information as possible about how parents react to their children's pain behaviors. We did not want to limit the inventory to just include items from parents whose children were already suffering from severe chronic pain, we wanted IRPEDNA to be useful with acute and chronic pain problems (in fact, with chronic pain experiences with different levels of related disability).

All the items reflect actions and behaviors that parents/caregivers may enact after observing the children's pain behaviors, namely: positive (solicitously attend to the child's pain behaviors by giving her/him something pleasant), and negative reinforcement (eliminating something unpleasant) of pain behaviors, punishment of well-behaviors (providing something unpleasant when the child behaves appropriately), and positive reinforcement and encouragement of well-behaviors and coping (promote more adaptive behaviors).

Once the list was completed, and before the inventory was used, the authors double checked whether items were appropriately written (e.g., avoiding double negative sentences/items), all items underwent a face validity check. Once this had been done, items were randomly ordered. The investigators then administered this first draft of the inventory to a community sample.

#### 2.4. Statistical analysis

Once the measure had been completed by parents, quantitative analyses were carried out using the SPSS Statistical Package for Windows 12.0.

First of all, missing data were explored, and afterwards an exploratory factor analysis was conducted, using a maximum-likelihood procedure with oblique rotation because we expected that the hypothesized factors would be negatively related to some extent. Our hypothesis was based on the assumption that the way the parents deal with the child's pain complaints is in part determined by their interpretation of the symptoms, and of the influence of their behavior on the child's health. Thus, most of the parental responses would be congruent amongst themselves, and tend to reflect one predominate type of parenting reaction in detriment of the others.

Two criteria were used to determine the number of factors in the inventory: the scree-test and comprehensibility, and to select the most reliable items we followed the guidelines provided by McDonald (1985). Different procedures were used to achieve the best fitting solution and evaluate its goodness of fit (see below).

Available evidence suggests that age, sex, and temporal characteristics of pain problems (i.e., acute *vs.* chronic pain) may influence parental reactions to their children's pain behaviors (see Walker and Zeman, 1992; Craig and Riddell, 2003). Since the inventory was developed to be used with children and adolescents suffering from acute or chronic pain, the stability of the factor solution across these different populations (i.e., by child's sex, age, and type of pain complaint) was examined using the coefficient of congruence.

Subsequently, item analyses allowed to study the effectiveness of the items. Item means and item-total correlations were the parameters under consideration. The Kolmogorov-Smirnov tests, and central moments of the distribution, were also used to test whether or not the sample of data from each subscale was consistent with a normal distribution function.

Finally, internal consistency was assessed, and correlations between IRPEDNA's scales with PANAS's negative affect subscale scores were calculated to study the criterion validity of the inventory.

### 3. Results

#### 3.1. Pain characteristics of participating children

86.7% of the participating children had experienced at least one pain episode in the last three months. Lower limb pain was reported as the most troublesome pain complaint (29.4%), followed by abdominal pain and headache (29.4% and 21.1%, respectively). The most common intensity of the most troublesome pain over the last three months was 3.47 (SD = 2.40, range 0-10), and the highest intensity was 5.22 (SD = 3.20). According to the criteria used in this study to define chronic pain, 37.1% of participating children suffered from it.

Pain problems were not a common reason for missing school amongst this sample, most of the participating children (75%) attended school every day in the month previous to the interview, 17.1% missed school between one and three days. The mean number of days missed from school during that month due to any illness was 0.76 (SD = 1.88, range = 0-20).

Parents' responses to IRPEDNA were examined for missing data. In 27% of the cases, we observed missing data (the number of participants who completely answered the inventory was 292). Fifty-one percentage of them skipped one item, and 34% skipped between two and three items. Cases with missing values were deleted from the main analyses for different motives: (1) there was not any difference in any of the variables studied (i.e., SES, child's age, sex, type of pain complaint, pain intensity) between cases with and without missing data ( $p < 0.05$ ); (2) the missing values were not concentrated in a few variables; and (3) the original data set was large enough. Afterwards, this substantial loss of subjects due to missing values can be considered quite understandable, granted the length of the original version of the IRPEDNA (i.e. 60 items), and the fact that participating parents were requested to complete the questionnaire by themselves at home without receiving any compensation for their participation.

#### 3.2. Exploratory factor analysis

An exploratory factor analysis using a maximum-likelihood procedure and oblique rotation was performed. The factor analysis yielded 16 factors with eigenvalues greater than 1.00. However, the scree-test suggested a three-factor solution (see Fig. 2).

Beside the scree-test plot, the other criterion for determining the number of factors to be included was comprehensibility (i.e., factors whose dimensions of meaning were readily comprehensible). Moreover, goodness-of-fit measures were used to confirm the three-factor model, with data showing some robust characteristics.

The estimated parameters of the factor analysis for this three-factor solution looked acceptable (NNFI-non-normal fit index = 0.83; RMSEA-root-mean-square-error of approximation = 0.055;  $\chi^2$  goodness-of-fit test = 1051.66 (df = 558;  $p < 0.001$ )). As well, the fit of various factor models of the inventory was compared by the hierarchical  $\chi^2$  goodness-of-fit test. Table 1 displays results after comparing models. These analy-

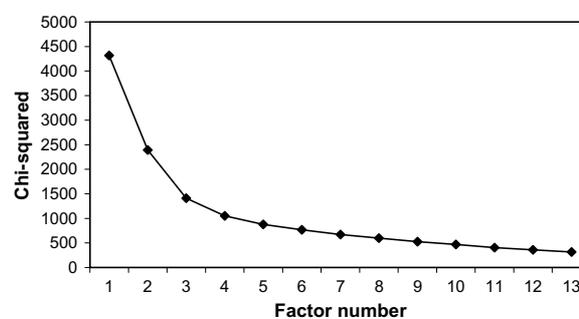


Fig. 1. Hierarchical GOF  $\chi^2$ -difference statistic plotted against the number of factors.

Table 1  
 $\chi^2$  tests used to compare models

	$\chi^2$	df	<i>p</i>
Model 0: Non-factor exploratory model	4318.75	666	0.00
Model 1: One-factor exploratory model	2394.43	629	0.00
Model 2: Two-factor exploratory model	1410.20	593	0.00
Model 3: Three-factor exploratory model	1051.66	558	0.00
Model 4: Four-factor exploratory model	877.45	524	0.00
Model 5: Five-factor exploratory model	769.86	491	0.00
Model 6: Six-factor exploratory model	672.05	459	0.00
Model 7: Seven-factor exploratory model	598.16	428	0.00
Model 8: Eight-factor exploratory model	526.37	398	0.00
Model 9: Nine-factor exploratory model	468.10	369	0.00
Model 10: Ten-factor exploratory model	402.92	341	0.01
Model 11: Eleven-factor exploratory model	360.55	314	0.03
Model 12: Twelve-factor exploratory model	316.47	288	0.12
	$\Delta\chi^2$	df	<i>p</i>
Difference between model 0 and model 1	1920.32	37	0.00
Difference between model 1 and model 2	988.23	36	0.00
Difference between model 2 and model 3	358.54	35	0.00
Difference between model 3 and model 4	174.20	34	0.00
Difference between model 4 and model 5	107.59	33	0.00
Difference between model 5 and model 6	97.81	32	0.00
Difference between model 6 and model 7	73.89	31	0.00
Difference between model 7 and model 8	71.79	30	0.00
Difference between model 8 and model 9	58.27	29	0.00
Difference between model 9 and model 10	65.18	28	0.00
Difference between model 10 and model 11	42.37	27	<0.05
Difference between model 11 and model 12	44.07	26	<0.025

ses revealed a wide range of good-fitting models. Because of the large size of the sample, several complex models were suggested. Nevertheless, we examined a plot of the hierarchical  $\chi^2$  goodness-of-fit indices to guide a selection of a model that provided an optimal balance of good fit statistics and substantive clarity (see see Fig. 1). The three-factor model seemed to be the best one.

Thus, a factor analysis was performed that yielded three factors. Initially, key items selected to be included in the inventory were those whose item-corresponding factor correlation was higher than 0.40 and was at least 0.15 higher than item-other factor correlations' (McDonald, 1985). After the item elimination procedure, the instrument included 37 items grouped in three non-orthogonal domains. All selected items loaded on the factor which had been previously hypothesized. The solution converged in less than 10 iterations which means that the solution was quite clear.<sup>1</sup> The results of this factor analysis are shown in Table 2. Some of the eliminated items were related to the promotion of well-behaviors and coping (e.g., I encourage him/her to prac-

tice sports), which is one of the subscales of the inventory. However, some other eliminated items were related to dimensions that did not fit well within the learning model used here as a template; for example, some referred to using medications or seeking medical help (e.g., I give her/him medication, I take her/him to the doctor), whereas some other ones referred to parents showing/expressing their emotions to the child (although one of such items is already included in the solicitousness scale: item #32).

The relationships between factors were  $-0.09$  (between factor 1 and 2),  $-0.40$  (between factor 1 and 3), and  $-0.17$  (between factor 2 and 3).

The three factor solution explained 42.36% of the total variance. The first factor (22.83% of the variance) has 15 items, the second factor (13.25% of the variance) has 10 items, while the third factor (6.28% of the variance) has 12 items (see Table 2). These three factors were named: (1) solicitousness, (2) discouragement, and (3) promotion of well-behaviors and coping, respectively. They are scored by averaging the items that loaded on each one.

### 3.3. Analysis of the model comparing different groups

The factor solution was compared in relation to the child's age, gender, and pain complaint (non-chronic

<sup>1</sup> Interested readers may obtain copies of the inventory, both in the Catalan and the Spanish version, from the authors. The Spanish version of IRPEDNA is currently being tested. Our first analyses are showing quite promising results, its factorial structure is equal to the Catalan original version one.

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Table 2  
 IRPEDNA's three-factor model: factor loadings by item

Items	Factors		
	Solicitousness	Discouragement	Promotion of well-behaviors and coping
3. Stay home from work to provide him/her with as much care as possible	<b>0.55</b>	0.05	-0.07
6. Accept that, in these circumstances, he/she need not do his/her homework	<b>0.57</b>	0.02	0.15
11. Try to make his/her surroundings as comfortable as possible (for example, minimum noise and little light)	<b>0.45</b>	-0.00	-0.21
12. Monitor the evolution of the problem by regularly asking him/her how he/she feels	<b>0.51</b>	-0.11	-0.19
13. Make him/her go to bed earlier	<b>0.49</b>	0.13	-0.02
16. Try to make up for his/her suffering by paying him/her more attention	<b>0.56</b>	0.04	-0.22
18. Tell his/her teachers how he/she is feeling so that they are aware of the problem during school hours	<b>0.45</b>	0.02	-0.06
19. Suggest that he/she stays at home, with a relative, or with someone else (for example, a child minder) while the problem lasts	<b>0.55</b>	0.06	0.17
20. Arrange for others to take over his/her duties and responsibilities at home while he/she does not feel well	<b>0.59</b>	-0.01	-0.11
21. Show him/her that you are very concerned	<b>0.42</b>	0.00	0.06
23. Help him/her to do certain things (for example, get dressed, do homework)	<b>0.56</b>	0.00	-0.07
24. Spend as much time with him/her as possible	<b>0.64</b>	-0.08	-0.15
29. Stop what you are doing to do something that he/she likes (e.g. play)	<b>0.54</b>	-0.00	-0.16
36. Not do your leisure activities so that you can be with him/her	<b>0.65</b>	-0.12	-0.13
37. Try to arrive home as early as possible	<b>0.45</b>	-0.26	-0.20
1. Think that it cannot really be so bad	-0.26	<b>0.55</b>	-0.19
5. Wonder how he/she can complain so much	0.03	<b>0.45</b>	-0.06
8. Not believe him/her	0.08	<b>0.57</b>	0.03
9. Not listen to him/her	0.07	<b>0.58</b>	0.08
14. Think it does not matter because you believe the suffering will make him/her stronger	0.06	<b>0.40</b>	-0.02
22. Tell your child that it is not so bad	-0.08	<b>0.67</b>	-0.15
25. A little bit angry, tell him/her not to complain so much	0.03	<b>0.66</b>	0.01
27. Tell him/her that adults do not complain	0.07	<b>0.46</b>	-0.08
31. Tell him/her that he/she may be exaggerating	0.02	<b>0.72</b>	-0.06
34. Make light of the problem because I think he/she is exaggerating a little	-0.16	<b>0.64</b>	0.12
2. Tell your child that he/she can cope with the problem using expressions such as "you are strong" "you can cope with this and more"	-0.13	0.24	<b>-0.52</b>
4. Try to explain the reasons for his/her complaint so that he/she does not worry so much	-0.05	-0.06	<b>-0.53</b>
7. Advise your child to relax and breathe deeply	0.07	0.07	<b>-0.56</b>
10. Try to distract him/her by suggesting attractive activities (e.g. plans for the weekend)	0.05	0.19	<b>-0.46</b>
15. Try to entertain him/her so that he/she does not think about the problem	-0.02	0.14	<b>-0.56</b>
17. Advise him/her to concentrate on other things (e.g. listen to music, watch the TV) so that he/she does not think about the pain	-0.00	0.16	<b>-0.61</b>
26. Use humor to take his/her mind off the discomfort	0.17	0.10	<b>-0.54</b>
28. Tell him/her not to worry because it will soon go away	0.13	-0.01	<b>-0.57</b>
30. Tell him/her that you think everything will be all right	0.20	-0.14	<b>-0.64</b>
32. Try to get him/her to be optimistic about the pain	0.01	-0.05	<b>-0.76</b>
33. Tell him/her not to be afraid	0.19	-0.04	<b>-0.46</b>
35. Try to encourage him/her	0.20	-0.26	<b>-0.52</b>

Extraction method: Maximum-likelihood procedure. Rotation method: Oblimin.

The solution converges in nine iterations.

Sample size:  $N = 292$ .

Bold indicates highest factor loading per item.

vs. chronic pain), using the coefficient of congruence. To examine if the solution was invariant across child's age, the sample was split into two groups: group 1 included parent/caregiver responses to children from 6 to 10 years ( $N = 170$ ), and group 2 comprised the parent/caregiver responses of children aged between 11 and 16

( $N = 122$ ). To examine if the solution was invariant across child's gender, the sample was split into two groups: 175 boys and 117 girls. Finally, to examine if the solution was invariant across pain complaint, the sample was split into two groups: 108 chronic pain sufferers and 184 non-chronic pain children. Table 3 pre-

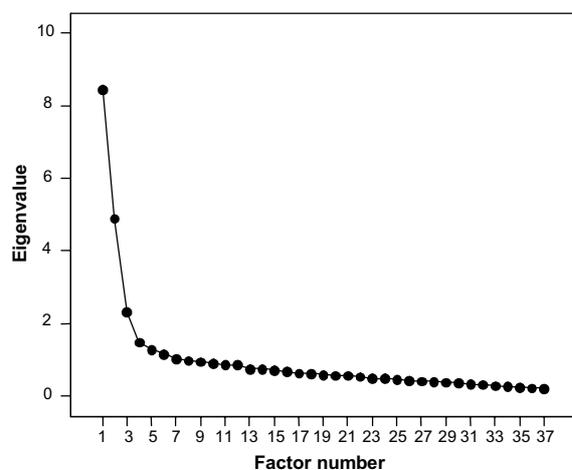


Fig. 2. Scree plot of the IRPEDNA.

sents the coefficients of congruence for each factor comparing factor loadings of male children with those of female children, factor loadings of children aged 6–10 and 11–16 years, and factor loadings of children who suffered from chronic pain and children who suffered from non-chronic pain. The outcomes of the comparisons indicate that the congruence between factors is high, indicating that the three-factor model is stable across children age, gender, and type of pain complaint.

### 3.4. Item analysis

The mean and distributions of each item were examined subsequently. As Table 4 shows, all items had optimum mean and variance values, except for a few items loading in the *discouragement scale* which had very extreme means (i.e. item 9, 14, 25, 27). The low values, were somewhat expected; since pain complaints, as a danger sign, are normally attended to by parents and other loved ones. However, we decided not to delete them from the scale, granted that there only were four items with extreme means, moreover their item–total correlations were acceptable.

Table 4 also shows the correlation between each item and the total sum score (excluding that item). In order to maximize item–test correlations, or Alpha coefficient (reliability), it had been decided that (a) those items which correlated less than 0.20 with the subscale score

(corrected item–total correlation) would be deleted (i.e., these items do not measure the same construct as does the subscale or they are confusing or misleading to respondents), and (b) those items that correlated more than 0.80 (corrected item–total correlation) should also be deleted, because they were needlessly repetitive. At this stage, no item had to be deleted as correlations ranged between 0.35 and 0.69 (see Table 4).

### 3.5. Sampling distributions

Since the factorial analysis is unable to determine whether score subscales are normally distributed, first we examined indicators of the shape of the distributions, including kurtosis and asymmetry. Neither kurtosis nor asymmetry of the distribution of the first and second subscales was higher than 1 (see Table 5). Therefore, assuming a normal distribution for the first and second subscales was appropriate. That was not the case for the third subscale. On the basis of data presented in Table 5, a leptokurtic distribution could be assumed for the third subscale. However, the authors also applied the Kolmogorov–Smirnov test, which is a strict method, to test for a normal distribution of a measure. The Z statistics for the Kolmogorov–Smirnov test for subscales 1, 2, and 3 were 0.86, 1.46 and 1.21, respectively. Based on this information, the hypothesis regarding the normal distribution could be accepted for the three subscales (data being significant at the 0.05 level).

### 3.6. Reliability

Cronbach’s alpha coefficient was used to study the internal consistency of the inventory and its subscales. The overall Cronbach’s alpha for the instrument was 0.89. The reliability coefficients for subscales 1, 2, and 3 were 0.87, 0.83, and 0.87, respectively.

### 3.7. Validity

The criterion validity of IRPEDNA’s scales scores was examined by studying its correlation with PANAS’ negative affect subscale scores. Since parental responses to children’s pain behavior may be different in cases of children with chronic pain, when compared to those to children with non-chronic pain, these correlations were

Table 3  
 Coefficients of congruence for the factors of the inventory across children gender, age, and type of pain

Gender	C	Age	C	Type of pain	C
$c_{(g)1'} - c_{(g)1''}$	0.96	$c_{(a)1'} - c_{(a)1''}$	0.99	$c_{(d)1'} - c_{(d)1''}$	0.85
$c_{(g)2'} - c_{(g)2''}$	0.94	$c_{(a)2'} - c_{(a)2''}$	0.99	$c_{(d)2'} - c_{(d)2''}$	0.87
$c_{(g)3'} - c_{(g)3''}$	0.92	$c_{(a)3'} - c_{(a)3''}$	0.99	$c_{(d)3'} - c_{(d)3''}$	0.80

The three factors derived in each group are denoted: Male children:  $c_{(g)1'}$ ,  $c_{(g)2'}$ ,  $c_{(g)3'}$ ; Female children:  $c_{(g)1''}$ ,  $c_{(g)2''}$ ,  $c_{(g)3''}$ . Young children:  $c_{(a)1'}$ ,  $c_{(a)2'}$ ,  $c_{(a)3'}$ ; Older children:  $c_{(a)1''}$ ,  $c_{(a)2''}$ ,  $c_{(a)3''}$ . Children without chronic pain:  $c_{(a)1'}$ ,  $c_{(a)2'}$ ,  $c_{(a)3'}$ ; Children with chronic pain:  $c_{(a)1''}$ ,  $c_{(a)2''}$ ,  $c_{(a)3''}$ .

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Table 4  
 Item means and correlations between each item and the total sum score

Factor	Item	Mean (SD)	$r_{(\text{item-total})}$	
Solicitousness	3. Stay home from work to provide him/her with as much care and attention as possible	2.17 (1.24)	0.53	
	6. Accept that, in these circumstances, he/she need not do his/her homework	1.29 (1.11)	0.46	
	11. Try to make his/her surroundings as comfortable as possible (for example, minimum noise and little light)	2.57 (1.17)	0.53	
	12. Monitor the evolution of the problem by regularly asking him/her how he/she feels	3.12 (0.94)	0.55	
	13. Make him/her go to bed earlier	2.57 (1.13)	0.45	
	16. Try to make up for his/her suffering by paying him/her more attention	2.79 (1.10)	0.63	
	18. Tell his/her teachers how he/she is feeling so that they are aware of the problem during school hours	2.30 (1.28)	0.47	
	19. Suggest that he/she stays at home, with a relative, or with someone else (for example, a child minder) while the problem lasts	1.52 (1.28)	0.40	
	20. Arrange for others to take over his/her duties and responsibilities at home while he/she does not feel well	2.64 (1.24)	0.59	
	21. Show him/her that you are very concerned	1.80 (1.21)	0.35	
	23. Help him/her to do certain things (for example, get dressed, do homework).	2.16 (1.15)	0.57	
	24. Spend as much time with him/her as possible	2.96 (0.99)	0.63	
	29. Stop what you are doing and do something that he/she likes (e.g. play).	2.26 (1.02)	0.57	
	36. Not do your leisure activities so that you can be with him/her	2.81 (1.13)	0.67	
	37. Try to arrive home as early as possible	3.32 (0.93)	0.51	
	Discouragement	1. Think that it cannot really be so bad.	1.47 (1.15)	0.55
		5. Wonder how he/she can complain so much	1.08 (1.19)	0.39
8. Not believe him/her		0.88 (1.16)	0.54	
9. Not listen to him/her		0.73 (1.06)	0.53	
14. Think it does not matter because you believe the suffering will make him/her stronger		0.37 (0.83)	0.39	
22. A little bit angry, tell your child that it is not so bad		1.48 (1.13)	0.62	
25. Get angry and tell him/her not to complain so much		0.64 (0.91)	0.60	
27. Tell him/her that adults do not complain		0.68 (1.01)	0.41	
31. Tell him/her that he/she may be exaggerating		1.21 (1.06)	0.66	
34. Think he/she is exaggerating a little	1.06 (1.08)	0.52		
Promotion of well-behaviors and coping	2. Tell your child that he/she can cope with the problem using expressions such as "you are strong" "you can cope with this and more"	1.89 (1.25)	0.46	
	4. Try to explain the reasons for his/her complaint so that he/she does not worry so much	3.05 (1.00)	0.46	
	7. Advise your child to relax and breathe deeply	2.42 (1.18)	0.55	
	10. Try to distract him/her by suggesting attractive activities (e.g. plans for the weekend)	2.16 (1.12)	0.51	
	15. Try to entertain him/her so that he/she does not think about the problem	2.30 (1.16)	0.55	
	17. Advise him/her to concentrate on other things (e.g. listen to music, watch the TV) so that he/she does not think about the pain	2.41 (1.10)	0.61	
	26. Use humor to take his/her mind off the discomfort	2.54 (1.05)	0.60	
	28. Tell him/her not to worry because it will soon go away	2.82 (1.09)	0.57	
	30. Tell him/her that you think everything will be all right	3.08 (1.00)	0.64	
	32. Try to get him/her to be optimistic about the pain	3.04 (0.94)	0.69	
	33. Tell him/her not to be afraid	2.76 (1.36)	0.50	
35. Try to encourage him/her	3.42 (0.82)	0.52		

Table 5  
 Asymmetry and kurtosis values for IRPEDNA's subscales

	Asymmetry		Kurtosis	
	$a_3$	Error	$a_4$	Error
Solicitousness	-0.29	0.14	0.18	0.27
Discouragement	0.42	0.14	-0.64	0.27
Promotion of well-behaviors and coping	0.68	0.13	1.45	0.26

Table 6  
 Correlations between IRPEDNA scales and PANAS negative affect subscale scores

	Non-chronic pain sub sample	Chronic pain sub sample
Solicitousness	0.27***	0.19*
Discouragement	0.09	0.07
Promotion of well-behaviors and coping	0.23**	0.19*

\*  $p < 0.05$ .  
 \*\*  $p < 0.01$ .  
 \*\*\*  $p < 0.001$ .

Table 7  
 Mean and standard deviations comparisons by children gender, age, type of pain complaint, and informant's gender

	Total sample	Median (SD)							
		Child's gender		Child's age		Type of pain complaint		Informant's gender	
		Male	Female	Younger children (6-10 y)	Older children (11-16 y)	Non-chronic pain	Chronic pain	Father	Mother
Solicitousness	2.42 (0.67)	2.44 (0.64)	2.25 (0.66)	2.34 (0.68)	2.40 (0.62)	2.40 (0.69)	2.28 (0.65)	2.10 (0.74)	2.30 (0.66)
Discouragement	0.94 (0.66)	1.02 (0.60)	0.93 (0.67)	1.02 (0.61)	0.93 (0.66)	0.86 (0.61)	0.98 (0.67)	1.04 (0.72)	0.90 (0.62)
Promotion of well-behaviors and coping	2.65 (0.69)	2.67 (0.61)	2.47 (0.69)	2.61 (0.61)	2.55 (0.71)	2.56 (0.71)	2.57 (0.64)	2.28 (0.72)	2.55 (0.64)

separately computed for children with chronic pain, and children with non-chronic pain. As can be seen in Table 6, there was a positive relationship between the scores on the solicitous response, and the promotion of well-behaviors and coping scales with the levels of children's pain related distress, in both chronic and non-chronic pain subgroups ( $p < 0.05$ ).

### 3.8. Comparison of the scores of IRPEDNA subscales by child's gender, age, type of pain complaint, and informant's gender

The effect of child's gender, age, type of pain complaint, and informant's gender upon the scores of each subscale was examined by ANOVA, and Pearson correlations; Table 7 presents means and standard deviations for the total sample group as well as for each different subgroup. Concerning gender, data showed that parental solicitous reactions to child's pain behaviors were higher for boys than for girls ( $F_{(1,317)} = 6.67$ ;  $p < 0.05$ ); similarly parental promoting well-behaviors and coping were higher for boys than for girls ( $F_{(1,338)} = 7.35$ ;  $p < 0.05$ ). In relation to the child's age, there was a significant negative relationship between parental responses encouraging well-behaviors and the child's age ( $r = -0.13$ ;  $p = 0.05$ ). With respect to the type of child's pain complaint, no differences were found on parental responses to the children's pain behaviors regardless of whether the child was suffering from a chronic or a non-chronic pain problem, as it could be expected from the validity analysis performed on the instrument subscales. Finally, and in relation to the effects of the informant's gender, our data show that scores on the promoting well-behaviors and coping subscale were higher when the mother was the informant ( $t = -2.00$ ,  $p < 0.05$ ) (see Table 7).

## 4. Discussion

The objectives of this study were development and initial validation of an instrument to assess parents/caregivers' responses to children experiencing pain. Specifically, the authors sought to develop a comprehensive

instrument that would be useful in learning about adult interactions with children in pain. Although some assessment instruments have been available for some time (Walker and Zeman, 1992; Manimala et al., 2000; Blount et al., 2001; Reid et al., 2005), they suffer from two main problems. First, they had mainly been focused on the effects of parents solicitously attending to their children's signs of pain and distress. However, other types of adults' reactions are thought to influence children's pain behavior and ability to cope (Fordyce, 1976; Craig, 1978; Turk et al., 1983). Second, they were developed on the basis of parental reactions to children suffering from chronic disabling pain problems. Reactions of parents can be very different in acute and chronic pain problems, and in fact what can be considered helpful reactions in one situation can be considered unhelpful in the other. IRPEDNA was developed so as to avoid these problematic issues; it includes reactions other than simply solicitous attention, as well as reactions from parents of children suffering from acute and chronic pain conditions.

The results of the study indicated that IRPEDNA has three factors that explain 42.36% of the total variance. The first scale assesses the extent to which parents or caregivers solicitously attend to the children's pain behaviors by giving them something pleasant (positive reinforcement) or by eliminating something unpleasant (negative reinforcement) contingent upon pain behavior. The second scale assesses the extent to which parents discourage or show disapproval of the children's pain behaviors, be that by ignoring those pain behaviors, providing something unpleasant, or depriving them of something pleasant (punishment). The third scale assesses the extent to which parents try to promote children's "well-behaviors" (i.e., more adaptive behaviors) and coping.

The data examined in this study showed the stability of the factorial structure of the instrument across age and sex of the children, as well as across chronic and non-chronic pain subgroups.

IRPEDNA has shown sound psychometric properties. The scores of individual items were highly correlated with their appropriate subscale, and the factor

analysis indicated that its internal structure was strong. The validity of the inventory is supported by the statistically significant relationship between IRPEDNA and PANAS negative affect subscale scores. The observed relationship between parental response to the child's pain behaviors and pain related distress may be due to the fact that parental attentive behavior (in the form of solicitousness or promotion of coping) could reinforce greater distress in the child. That is, if a child observes his parents responding attentively to his pain complaints, parental responses may function as signs of parental worries which might cause child's distress (see Craig, 1986). At the same time, high levels of child's distress might also elicit greater attentiveness to the child behavior. Thus, and paradoxically, parental attentive responses might exert detrimental effects on child's adjustment to and coping with acute and chronic pain (see Blount et al., 1989; Whitehead et al., 1994; Sweet and McGrath, 1998). These results need to be replicated, and further studies on IRPEDNA's psychometric properties are warranted (for example, it is necessary to look at other features of construct validity, as well as test-retest reliability and sensitivity).

Our results also showed that parental solicitous reactions and promoting well-behaviors and coping in relation to child's pain behaviors were higher for boys than for girls. One tentative hypothesis has to do with parents being habituated or not to the child pain complaints. That is, granted that females tend to suffer and complain more for their pain problems than males (see Goodenough et al., 1999; Merlijn et al., 2003; Keogh and Eccleston, 2006), parents might have got used to their daughter's pain complaints, which, in turn, might have less credibility than those from their son. Consequently, parents might be more prone and tuned into listening and reacting to their son's pain complaints.

One another interesting result of this study that merits further research and clarification is that mothers tended to be higher in promoting well-behaviors. To some extent, this result seems to go against the more "traditional gender roles" where females tend to be considered as being more "solicitous". We do not have a certain explanation but wonder whether this a contextual finding related to cultural differences between our Catalan Mediterranean sample, and Anglo-Saxon and Scandinavian samples (which is the origin of participating subjects in almost all published studies if not all).

This study is not exempt from limitations. First, IRPEDNA relies on self-reports of parents/caregivers about their reactions to the children's pain. Recent research has shown discrepancies between what parents report they do and what they actually do (Cohen et al., 2000). Behavioral observation procedures can provide an alternative perspective and they should be used to complement self-report (see, for example, the scales developed by Blount and his colleagues for use in the

assessment of parental reactions to acute pediatric pain associated to medical interventions; Blount et al., 1989, 1997, 2001). However, while direct observation can be a rich and valid assessment procedure for collecting information concerning children's pain behaviors (Chambers et al., 2003), its use is often not feasible or practical (e.g., in private contexts). Moreover, observational procedures have their own limitations and problems (e.g., reactivity during observation). Thus, there is no "gold standard measure" for assessing parental and caregivers' responses to children in pain, or for controlling biases in judgment. This, of course, is a common issue needing to be addressed when assessing all aspects of human behavior and functioning. At present multidimensional assessment incorporating both self-report and behavioral observation procedures is to be recommended.

Second, IRPEDNA was essentially constructed to gather information on behavioral reactions of parents following their children's pain behaviors, with the operant model of reinforcement contingencies serving as a template (see Fordyce, 1976). However, one must acknowledge that pain is more than pain behavior (see Turk and Flor, 1987), and that there are other important features of pain (e.g., cognitive reactions) that also have an important impact on pain and pain related disability (see Turk and Okifuji, 2002). Pain is a complex experience, encompassing different levels (behavior, cognition, emotion, and physiology) and units of analysis (individual, dyad, and context). This instrument, however, is focused on tapping just one level of analysis (behaviors). Future work should include and examine other levels and units of analysis, in line with recent integrative models of pediatric pain (see, for example, Palermo and Chambers, 2005).

Third, we were unsuccessful in always attracting the participation of both parents. That is, although we had asked both parents to report their reactions to their children's pain behaviors, most of the responses in this sample came from children's mothers, and our results show some informant's gender related effects. Future studies will need to make a greater effort to collect more fathers' responses. At present, it remains unclear how similar mothers and fathers are in their perceptions of their children's pain behaviors, as well as in their behavioral reactions (Garcia-Munitis et al., 2006). Moreover, maternal and paternal behaviors may exert different influences on their children. Thus, looking into separate parental reports, and the extent to which fathers and mothers influence their children's pain experience, and the impact on the children's overall adjustment (e.g. child distress, child pain coping, child pain experience), would also be recommended.

Fourth, although the measure has been constructed to assess parental reactions to their children's pain behaviors, both in chronic and acute pain problems, in this stage of development it was administered to a com-

munity sample of children with very different pain problems, and although these preliminary results are of interest they are not enough. This is an initial attempt to describe parental responses to pain so that future studies could validate them on clinical pain samples (rather than on general community samples). Moreover, adult participants in this study were volunteers whose children were attending school. These were highly motivated individuals and they may not be representative of the adult population at large (for example, parents who were willing to participate could have been those who were more involved and/or interested in the care of their children, and thus their behaviors could also be different from parents who were not willing to participate).

Despite some limitations, IRPEDNA has shown good psychometric characteristics. The inventory can be used to assess adults' reactions to children's pain behaviors, and would be useful in studying their influence on the experience of pain in children. It is the authors' position that the information collected with this inventory could provide helpful insights to improve current treatment approaches by improving parents' involvement in the management of pediatric pain. Accurate assessment of parental reactions to children's pain behavior seems crucial if we are to come to understand the pain experience of children and provide remedial social interventions where warranted.

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**Paper #4:**

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## THE SEVERITY OF CHRONIC PEDIATRIC PAIN: AN EPIDEMIOLOGICAL STUDY IN THE MEDITERRANEAN REGION OF CATALONIA

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### ABSTRACT

The purposes of this study were to (1) provide information on pediatric pain prevalence, and (2) study the severity and other pain related characteristics of schoolchildren from a Mediterranean region. 561 schoolchildren, aged between 8 and 16 years, participated in this cross-sectional study. Besides collecting information about the presence of pain at the time of interview, and in the preceding three months, several characteristics of participants' pain experiences and several indicators of participants' quality of life were requested. Results showed that 37.3% suffered from chronic pain, but only 5.1% suffered moderate or severe chronic pain problems. Gender and age differences were found in the prevalence of pain conditions. Children who suffered from a chronic pain condition reported a worse quality of life, missed more days from school, and were more likely to use pain medication and seek medical care for pain relief. Our study shows that chronic pain is a highly prevalent condition in the community, one that can exert negative consequence for the child, but the prevalence of severe chronic pain cases is low. New studies are needed to further empirically test the proposed method of grading the severity of chronic pediatric pain.

**Keywords:** chronic pain, epidemiology, schoolchildren, prevalence, pain severity.

### PERSPECTIVE

This article provides information on pain problems among Catalan schoolchildren. It also suggests a new grading system of chronic pediatric problems. This new system could help clinicians and researchers to diagnose pain problems in youth and design treatments suited to patients characteristics and needs.

## 1. Introduction

Epidemiological studies of pediatric chronic pain have increased over the last years.<sup>15,16,27,48,51, and 54</sup> It is argued that chronic pain is a common problem among general pediatric population, which negatively affects everyday functioning of children and the family.<sup>25,26,41, and 47</sup> However, most published studies simply report estimations of pain prevalence, and a very few major studies provide additional information, such as intensity or frequency.<sup>42 and 49</sup>

Although some form of chronic pain is common, the severity of these problems varies considerably.<sup>9</sup> For instance, whereas published community studies show that pain impacts the overall health status of children,<sup>24 and 49</sup> clinical population studies, which report information from more severe cases, show that chronic pain children suffer from depression, anxiety, sleep disturbances, and developmental problems in comparison to peers.<sup>12,13, and 34</sup> Children with chronic pain are not a homogenous group. Nevertheless, published epidemiological studies have not provided information about the variation in severity of pain conditions in the general population. The need to create a classification system for chronic pediatric pain that would go beyond defining chronic pain simply by temporal criteria is being advocated by clinicians and researchers.<sup>6,24, and 50</sup> Previous attempts to classify chronic pain conditions have used standard diagnostic criteria,<sup>37 and 46</sup> or have been based in the rather confusing diagnostic of the idiopathic pain syndrome.<sup>30</sup> However, due to the multidimensional nature of pain it is unlikely that pathophysiologic signs or physical symptomatology could properly reflect by themselves the severity of a chronic pain syndrome, and much less be of any use to properly customize treatments to youth grouped under the same diagnosis category.

Epidemiological studies of chronic pediatric pain have been conducted in Anglo-Saxon and Northern European countries, and little is known about pain problems that affect children's lives in other parts of the world. Studies in other cultural environments are needed, because results in geographical areas with different cultures might show differences that could help us better understand pain experiences in children and how the social and cultural context influence pain.

Thus, the purpose of this epidemiological study was twofold: (1) to provide information on pediatric pain prevalence; and (2) to study the severity and other pain related characteristics of schoolchildren aged between 8 and 16 years in Catalonia, a Mediterranean South-European region in the North-East of Spain.

## 2. Method

### 2.1. Participants

A representative sample of 902 schoolchildren were invited to participate. They were recruited from non-special primary and secondary schools (from grades 3 to 10) randomly selected in Tarragona (an area in the southeast of Catalonia). Of these, 337 were not interested or non-compliant (i.e., did not return the parental consent form), and 4 were deemed ineligible (i.e., they were suffering from cognitive impairment as reported by their school's teacher). Thus, the total sample of participating schoolchildren was 561 (62.2% participation rate), parents of

these children were also invited to participate in the study. The participating children did not differ from the eligible children who were invited but did not participate on school grade or sex ( $p>0.05$ ).

The age of the participating schoolchildren ranged from 8 to 16 years (mean age=11.89 years; SD=2.00); 51.7% of the sample was male. Table 1 presents relevant socio-demographic and medical data of participating schoolchildren.

## **2.2. Procedure**

The protocol was reviewed and approved by the Department of Education of the Government of Catalonia, and by the boards of the participating schools. A total of 40 classes from grades 3 through 10 of twenty primary and secondary schools were randomly selected using a stratified sampling procedure from all eligible non-special education schools in the Tarragona area. All the schools approached agreed to participate, except for one which was involved in several other research projects at the time. After the schools had given their agreement, the children attending these classes and their parents were invited to participate. The parents were sent a letter in which they and their children were asked to participate. Children whose parents provided informed consent were individually interviewed at their school; they were asked to provide information about various domains of their health status and about their pain problems.

Since the children were asked retrospective questions, memory enhancing techniques were used.<sup>39</sup> For example, interviewers used open-ended questions, and important events as end point times, in order to prevent interviewer bias from affecting children's reports, and to increase the accuracy of the information. Several other quality control measures were implemented during data collection. First, the interviewers attended standardization classes in which they were instructed and trained on how to conduct the interview. Second, data sheets were carefully checked immediately after the interviews, and further monitored centrally to check for missing data or inconsistencies.

The parents of the participating children were surveyed separately. They were requested to provide some additional socio-demographic data (see below), and information about the child's health status.

## **2.3. Measures**

### **2.3.1. Socio-demographic measures**

Information about age, sex, and school grade was collected at the time of interview. Based on the work of Domingo-Salvany et al.<sup>8</sup> on the measurement of socio-economic status based upon the Spanish Classification of Occupations (CNO-94). The highest occupation of the two parents was the one used to rate the SES of the family.

### 2.3.2. Medical condition

For descriptive purpose of the sample, parents were also requested to provide additional information regarding their children's current medical condition: that is, whether they had any illnesses at the time of interview. In order to gather this information, a list of illnesses (such as asthma, allergy, cardiovascular disease, diabetes, epilepsy, migraine, and psychiatric or psychological problems) was presented to them to choose from; if the appropriate illness was not on the list, parents were asked to report it.

### 2.3.3. Quality of life

The Pediatric Quality of Life Inventory™ Version 4.0 (PedsQL)<sup>57</sup> was used to assess the children's perception of their quality of life. The PedsQL is a generic instrument that encompasses two subscales: (1) Physical Health Summary, and (2) Psychosocial Health Summary, which assess three functioning areas: (a) Social, (b) Emotional, and (c) School functioning. One overall score (Total Scale score) can also be calculated. Developmentally appropriate versions (i.e. ages 8–12 and 13–18) were used. The child was asked how much of a problem each item had been in the past month. Items were scored on a 5-point Likert scale, from 0 (never a problem) to 4 (almost always a problem). Items were reverse-scored and linearly transformed to a 0-100 scale (0=100, 1=75, 2=50, 3=25, 4=0), so that higher scores indicated a better quality of life. The feasibility, reliability and validity of the instrument have been demonstrated in Catalan-speaking schoolchildren.<sup>22</sup>

Parents were asked to report about their child's quality of life in the month before. An 11-point numerical scale was used with the following anchors: 0 = My child has been feeling very bad; 10 = My child has been feeling excellent.

### 2.3.4. School attendance

Days of absence from school due to health problems in the month preceding the interview were reported by parents.

### 2.3.5. Pain

Based on suggestions and recommendations from the initiative known as PedIMMPACT<sup>33</sup>, the *Faces Pain Scale – Revised* (FPS-R)<sup>19</sup> was the instrument used to gather information about the children's usual and highest *pain intensity* in the preceding three months as well as the pain intensity at the time of the interview (see also the recent work by Stinson et al.<sup>53</sup>). The FPS-R is a six-point scale designed to measure children's level of perceived pain intensity. The children's task is to choose the face that best reflects the intensity of the pain they have experienced. A numerical value from 0 to 10 (0–2–4–6–8–10) is assigned to each face. The end points are explained as 'no pain' and 'very much pain'. We used the Catalan version of the scale, validated by Miró and Huguet.<sup>38</sup>

*Pain frequency* was measured by means of a categorical scale: more than once a week, about once a week, about once or twice a month, and once or twice in the last three

months. Pre-coded categories are often used to assess the frequency of pain in pediatric populations.<sup>42</sup>

Some information about *pain duration* was also recorded. Specifically, we asked the participating schoolchildren to report whether they had been experiencing pain for more than three months.

*Pain location* was coded according to a list based on axis I of the IASP Classification of Chronic Pain.<sup>35</sup> This information was gathered by means of an open question about where their pain was located. At the same time, and on the basis of the work by Mikkelsen et al.<sup>37</sup>, children were requested to mark and colour the painful area on a body figure<sup>18</sup>, so as to avoid potential misunderstandings.

In the case of children who reported to be suffering from more than one pain problem, they provided information about these pain characteristics (i.e., intensity, frequency, duration and location) in relation to each pain complaint; finally, they were also requested to mention which pain complaint had troubled them most in the last three months.

#### 2.3.6. *Pain severity*

To our knowledge, there is no validated system to grade the *severity* of a child's pain problem. Therefore, since there is increasing evidence coming from the adult pain literature suggesting that pain intensity and pain-related interference with activities may underlie a single dimension of global pain severity<sup>59</sup>, we decided to follow Von Korff et al.'s<sup>60</sup> suggestions, and the scheme they developed and tested with general population and adult chronic pain samples.<sup>52,58,60</sup> Based upon their work, the severity of a child's pain was defined in terms of three axes: (1) pain persistence (specifically, the presence of chronic pain), (2) pain intensity: average intensity in the 3 months before the interview, that is the mean of the highest and the most usual pain intensity in the last 3 months, and (3) pain impact, that is the level of pain-related disability. The importance of these domains in the study and treatment of chronic pediatric pain has been recently underscored by the Initiative on Measurement and Pain Assessment in Clinical Trials (PedIMPACT).<sup>33</sup> Based on these three axes, pain severity was classified in five grades: (1) *Grade 0*: a non-chronic pain problem; (2) *Grade 1*: low pain intensity (intensity index < 5; range= 0-10) and low pain-related disability (FDI score < 10; range: 0-52), (3) *Grade 2*: high pain intensity (intensity index > 5; range= 0-10) and low pain-related disability (FDI score < 10; range: 0-52); (4) *Grade 3*: moderate level of pain-related disability, regardless of pain intensity (FDI score= 10-20); and (5) *Grade 4*: high level of pain-related disability, regardless of the pain intensity (FDI score > 20).

To proceed in this graded classification, chronic pain was defined as a pain lasting for three months at least<sup>16</sup>, that was experienced continuously or recurrently; a recurrent pain problem was one occurring once or twice at least each month (see Mikkelsen et al.<sup>37</sup> for a similar procedure). Thus, timeframe was not the only criteria for defining chronic pain, if not pain's frequency was also used to screen for chronic pain because in the practice frequent recurrent pain is potentially significant. The *intensity index* [the average value of the highest and

the most usual intensity of the chronic pain complaint] was dichotomized (i.e., high intensity: intensity index higher than 5 vs low intensity: intensity index lower than 5).

Following with the recommendations of PedIMMPACT, the Functional Disability Inventory (FDI) <sup>61</sup> was used as a measure to assess pain-related disability. The FDI is one of the measures most commonly used to assess the impact of chronic pain in children.<sup>11</sup> Published reports<sup>25,50, and 61</sup> have shown that, mean self-report FDI scores with clinical populations are always higher than 10, ranging from 11.25 <sup>4</sup> to 34.5 <sup>10</sup>. Overall, nearly 50% of the examined clinical samples had mean FDI scores between 10 and 20, whereas the other 50% of the samples scored above 20. Thus, in our study, a cut off of 10 was used to define low to moderate disability, and a cut off of 20 for moderate to high disability.

### *2.3.7. Pain medication and pain-related visits to health care professionals*

Children were asked whether they had used pain medication or not, and whether they had consulted with a health care professional for each pain complaint reported within the previous three months.

### 3. Results

#### 3.1. Sample characteristics

Table 1 summarizes the main descriptive characteristics of participating schoolchildren. As can be seen, overall this is a healthy sample of children aged between 8 and 16 years. For instance, 25.8% of them had a confirmed medical diagnosis at the time of the interview with allergies and asthma being the most common reported health problems.

Table 1. Characteristics of the participating children

	Scale range	Total sample mean (SD)
<i>Socio-demographic data</i>		
Age (years)	8-16	11.89 (2.00)
Gender (% , N)		
Boys	--	51.7 (290)
Girls	--	48.3 (271)
SES <sup>a</sup> (% , N) <sup>a</sup>		
High	--	40.3 (191)
Medium	--	32.5 (154)
Low	--	27.2 (129)
Parents' marital status <sup>b</sup> (% , N)		
Married	--	85.2 (473)
Separated or divorced	--	13.5 (75)
Widowers/widows	--	1.2 (7)
Family members at home (% , N)		
Immediate family members	--	86.3 (484)
Extended family members	--	13.7 (77)
<i>Current general health status</i>		
Medical diagnosis <sup>c</sup> (% of the total sample, n)		
Diabetes	--	0.7 (3)
Allergy	--	13.3 (59)
Asthma	--	3.2 (14)
Cardiology problems	--	2.3 (10)
Psychiatric/psychological problems	--	1.1 (5)
Others	--	5.2 (23)

<sup>a</sup> Information missing in 87 cases

<sup>b</sup> Information missing in 6 cases

<sup>c</sup> Information missing in 119 cases

#### 3.2. Prevalence of pain and other related characteristics

18.4% of the children (n=102) were experiencing some pain at the time of interview, 26.5% of them (n=27) had pain in more than one site.

88% of the children (n=493) had experienced at least one pain episode within the three months before the interview. Among them, more than half (59.43%) reported to have experienced pain at more than one site. The most frequently reported pain locations were the lower limb (47%) and the head (43%), and the less common pain locations were the back (11.3%), the neck (5%), the pelvis (2.9%), and the chest (2.3%). Lower limb pain was reported as the most troublesome pain location in the previous three months (i.e., 29.4% of the children reported it). Headache and abdominal pain were more frequently reported by girls (Chi-

square<sub>(1)</sub>=10.1, p<0.01; and Chi-square<sub>(1)</sub>=15.75, p<0.001), and lower limb pain was more frequently reported by boys (Chi-square<sub>(1)</sub>=12.72, p<0.001). Age differences were only found for back pain such that children who reported back pain were significantly older (t=-2.58, p<0.05).

Overall, the intensity for these pain complaints was mild-moderate, that is, the highest intensity of the pain that had troubled them most over the last three months was 5.35 (SD=3.20, range 0-10), and the most usual intensity for the same pain was 3.44 (SD=2.36, range 0-10). No differences in the ratings of pain intensity were found related to gender or age. 38.3% of the children had consulted with a health care professional, and 42.2% had consumed pain medications (e.g., paracetamol, ibuprofen, acetylsalicylic acid) for their most troublesome pain complaint. In comparison with boys, girls had a higher probability of having consumed pain medication (Chi-square<sub>(1)</sub>=9.5, p<0.01), but not of having consulted a health professional for their pain. The age of the child was not related to the use of health care services or medication to get pain relief. Among the different pain problems, headache was the one requiring most medical consultation and medication for pain relief. Specifically, 42.9% of children who reported headache in the 3 months preceding the interview had consulted with a health care professional for their headache complaints, and 75.7% of children with headaches had consumed medication.

Table 2 presents descriptive statistics for children with chronic pain and without chronic pain.

*Table 2. Descriptive characteristics of health status indicators*

	Total sample	Non-Chronic pain	Chronic pain	Grade 0	Grade I	Grade II	Grade III	Grade IV
Quality of life (Mean, SD)								
Total quality of life	81.30 (10.51)	83.05 (9.94)	78.43 (10.81)	83.05 (9.94)	81.34 (9.89)	75.88 (11.14)	73.60 (11.66)	72.83 (7.87)
Physical functioning	80.62 (11.97)	82.55 (11.26)	77.33 (12.45)	82.58 (11.26)	80.59 (12.49)	74.80 (11.02)	70.24 (12.56)	68.22 (8.93)
Psychosocial functioning	81.74 (11.51)	83.44 (10.87)	78.96 (11.99)	83.44 (10.86)	81.58 (10.60)	76.59 (13.37)	75.40 (12.07)	75.28 (12.35)
Pain Medication (%)	44.9	39.3	46.9	39.3	42.3	64.1	80	83.3
Pain-related visits to a doctor(%)	38.3	29.7	53.1	29.7	44.2	59.4	80	83.3
Number of days missed school	0.85 (1.96)	0.77 (1.85)	9.99 (2.14)	0.77 (1.85)	0.77 (1.71)	0.83 (1.73)	1.17 (1.80)	5.40 (5.94)

According to the criteria used in this study to define chronic pain, the prevalence rate was 37.3% (n=209). Almost one third of the children with chronic pain problems (29.3%) reported pain in multiple sites (most frequently, two or three sites). The number of children with chronic pain on one site or multiple sites is shown in Figure 1. Lower limb pain (41.3%), headache (36.1%), and abdominal pain (31.3%) were the most prevalent complaints among chronic pain sufferers. A contrast of proportions showed that chronic pain at the lower limbs was

more likely to be reported as an isolated complaint than headache and abdominal pain were ( $z=2.05$  and  $2.04$ ,  $p<0.05$ ; respectively).

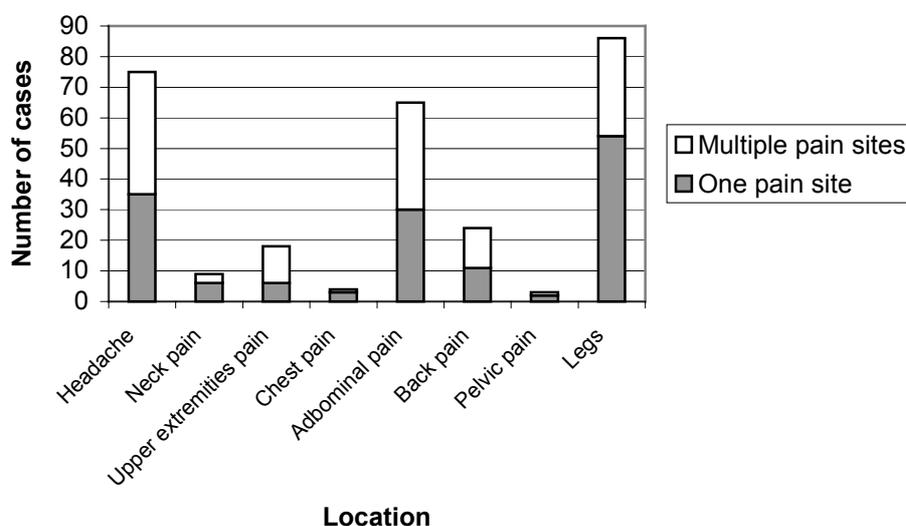


Figure 1. Number of children and adolescents with chronic pain problems suffering from one or multiple pain sites.

The prevalence of chronic pain increased with age, that is, t-test revealed that children with chronic pain were significantly older than children without chronic pain ( $t=-5.67$ ,  $p<0.001$ ). This age difference was more evident among girls than boys ( $t=-5.23$ ,  $p<0.001$ ;  $t=-2.14$ ,  $p<0.05$ , respectively). The prevalence of chronic pain was higher for girls than for boys (Chi-square<sub>(1)</sub> = 20.32,  $p<0.001$ ), irrespective of the child's age, statistically significant differences in Chi-square tests were found in both age groups: 8-12 years (Chi-square<sub>(1)</sub> = 5.83,  $p<0.05$ ), and 13-16 years (Chi-square<sub>(1)</sub> = 16.58,  $p<0.001$ ).

In relation to pain location characteristics, lower limb chronic pain was more common among boys than girls (57% vs. 19.65%; Chi-square<sub>(1)</sub> = 11.48,  $p<0.001$ ), and chronic pain at multiple sites was more common among girls than boys (50.6% vs. 22.2%; Chi-square<sub>(1)</sub> = 6.32;  $p<0.05$ ). No gender differences were found for any other locations. The rate of prevalence of chronic pain was neither associated with child's socioeconomic level (Chi-square<sub>(2)</sub> = 4.97,  $p>0.05$ ) nor with parents' marital status (Chi-square<sub>(2)</sub> = 3.64,  $p>0.05$ ).

The impact of chronic pain on children's physical functioning was significantly higher than the impact of acute and occasional pain (FDI score mean=5.70, SD=6.44, and FDI mean score= 4.42, SD=5.52; respectively;  $t=-2.33$ ,  $p<0.05$ ). Girls and boys with chronic pain reported similar levels of functional disability due to their pain ( $t=-1.61$ ,  $p>0.05$ ), and child's age was not associated with the level of pain related disability.

Specific t-tests indicated that children with chronic pain problems had significantly lower PedsQL scores (both on physical and psychosocial scales) than children who did not report chronic pain problems at the 0.001 level (see Table 2). Boys and girls with chronic pain

problems reported a similar level of quality of life ( $t=0.94$ ,  $p>0.05$ ); and the child's age was not related to quality of life. Parents' reports on their children's quality of life were comparable to the information provided by the children. That is to say, parents rated the quality of life of their children as high (mean= 8.34, SD= 1.92; range= 0-10), but those whose children had chronic pain problems acknowledged lower levels of quality of life for their children, as compared to parents whose children were not experiencing chronic pain ( $t=2.78$ ,  $p<0.01$ ). Similarly, and in agreement with that reported by participating schoolchildren, the quality of life of children with chronic pain was not related to children's age or gender (see Table 2).

Chi-square tests demonstrated that the likelihood to report pain-related visits to the doctor and consumption of medication was significantly higher for children with chronic pain problems than for children without those problems (Chi-square<sub>(2)</sub> = 28.74,  $p<0.001$ , and Chi-square<sub>(2)</sub> = 12.07,  $p<0.01$ , respectively). 53.06% of children with chronic pain problems consulted with a doctor due to their pain complaints, 29.73% of children with no chronic pain problems did so too. 54.6% of children with chronic pain problems had consumed pain medication within the preceding three months of the interview, 39.3%, of children with no chronic problems had done it too.

### 3.3. Severity of pain

The severity of pain syndromes in this population were graded as follows: grade 0: 63.2% (n=350); grade I: 20% (n=111), grade II: 11.7% (n=65); grade III: 4% (n=22); and grade IV: 1.1% (n=6) (see Figure 2). Table 3 presents some pain characteristics for each level of severity.

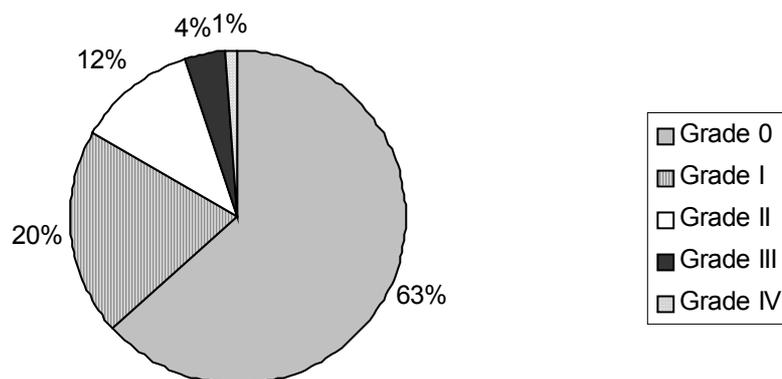


Figure 2. Percentage of schoolchildren in each chronic pain severity grade

*Table 3. Pain characteristics by graded severity*

	Grade I	Grade II	Grade III	Grade IV
<i>Pain frequency (N, %)</i>				
More than once a week	33 (29.7)	28 (43.1)	12 (54.5)	5 (83.3)
Once a week	29 (26.1)	12 (18.5)	3 (13.6)	1 (16.7)
Once or twice a month	49 (44.1)	24 (36.9)	7 (31.8)	0 (0)
<i>Pain intensity (Mean, SD)</i>	3.73 (1.31)	6.41 (1.39)	5.83 (1.99)	6.97 (1.63)
<i>Functional disability (Mean, SD)</i>	3.12 (2.81)	4.69 (3.24)	14.40 (2.26)	31.83 (6.61)

By definition, children at grade I and at grade II differed in terms of pain intensity level (low vs. high pain intensity) but were equal in terms of (low) levels of pain related disability (i.e., FDI scores < 10). However, although children at both grades scored in the lower range of FDI (i.e., FDI scores < 10), we found statistically significant differences between children in those two grades, namely, children at grade I reported lower levels of disability than children at grade II (mean=3.13; SD=2.81 and mean= 4.69; SD=3.24, respectively;  $t=-3.37$ ,  $p<0.001$ ).

Children at grade III and children at grade IV, again by definition, differed in the level of pain related disability (i.e., grade III: moderately disabled vs. grade IV: severely disabled) regardless of their pain intensity. As far as pain intensity, children at both grades tended to report high pain intensity (i.e., intensity index < 5; range= 0-10) (see Table 3), but that was not true for all the cases. Whereas at grade IV 83.3% of children had high pain intensity, at grade III that was true for 59% of children.

The severity of chronic pain syndromes was not graded in terms of frequency of pain episodes. However, it is noteworthy that the frequency of pain episodes showed a tendency to increase with higher pain grades (see Table 3). For example, on average, more than one weekly pain episode was reported by 29.7% of children at grade I, 43.1% at grade II, 54.5% at grade III, and 83.3% at grade IV. However, these differences were not statistically significant ( $\text{Chi-square}_{(9)}= 14.48$ ,  $p>0.05$ ).

The number of cases of schoolchildren distributed in grades of severity according to its location is depicted in Figure 3; percentage of cases in each grade for the most common pain location is depicted in Figure 4. Pain location was related with the severity of chronic pain only for cases of chronic pain in the head and in the lower limbs ( $\text{Chi-square}_{(3)}= 8.64$ , and  $9.57$ ,  $p<0.05$ , respectively). Children with chronic headache were more likely to experience more severe chronic pain syndromes (i.e., 9.9% of children at grade I, 12% at grade II, 36% at grade III, and 33% at grade IV), whereas children with chronic lower limb pain were more likely to suffer less severe chronic pain syndromes (i.e., 27% of children at grade I, 34% at grade II, 4.5% at grade III, and 0% at grade IV). For all other pain problems, the location was not found to be associated with pain severity grade.

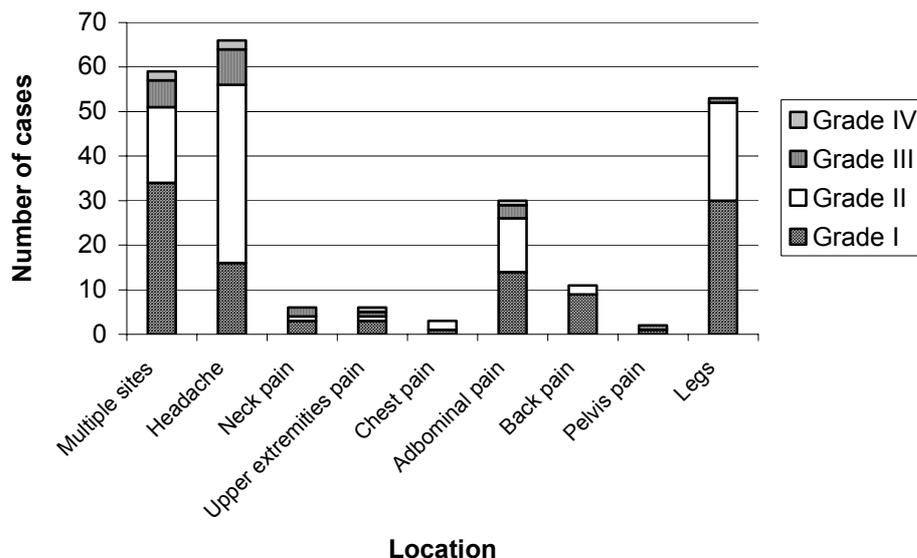


Figure 3. Number of schoolchildren in graded chronic pain severity for location

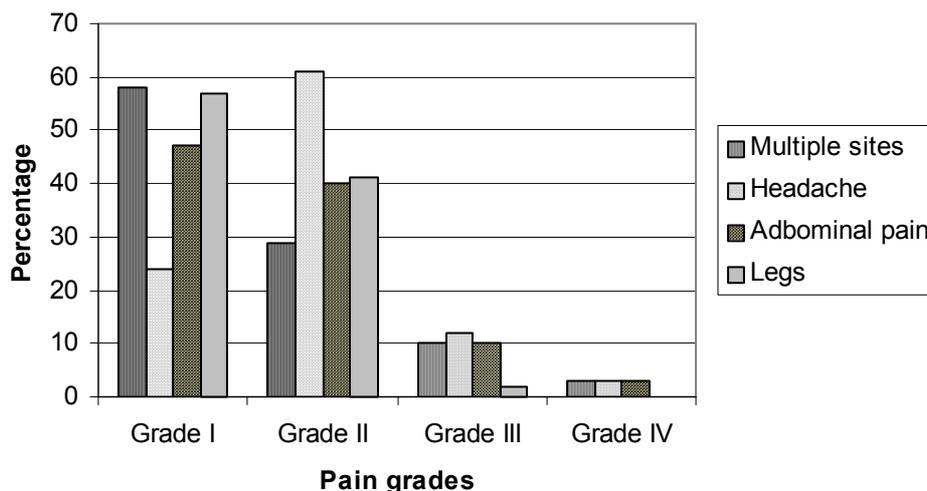


Figure 4. The percent distribution of graded chronic pain severity among headache, abdominal pain, lower extremities pain, and multiple pain sites sufferers

Pain severity grade was associated with age ( $F=3.15$ ,  $p<0.01$ ) as well as with sex (Chi-square<sub>(4)</sub> = 24.93,  $p<0.001$ ). Severe pain syndromes were more common among females (see Figure 5); specifically, children with pain syndrome grades I and III were more likely to be females than males (Chi-squared<sub>(1)</sub> = 20.5,  $p<0.001$  and Chi-squared<sub>(1)</sub> = 7.76,  $p<0.05$ , respectively). No differences were found for the other grades. As far as the child's age group is concerned (see Figure 6), younger children were more likely to report less severe pain syndromes. Specifically, children at grades 0 and I were younger than children at the other grades ( $t=5.36$  and  $-4.98$ ,  $p<0.001$ , respectively). T-tests for independent samples revealed no differences in age for children at higher grades.

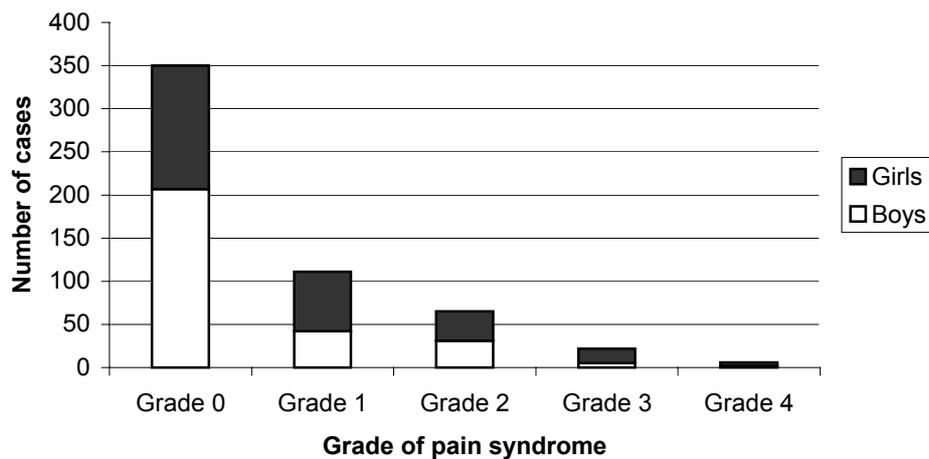


Figure 5. The number of cases distribution of graded chronic pain severity by gender

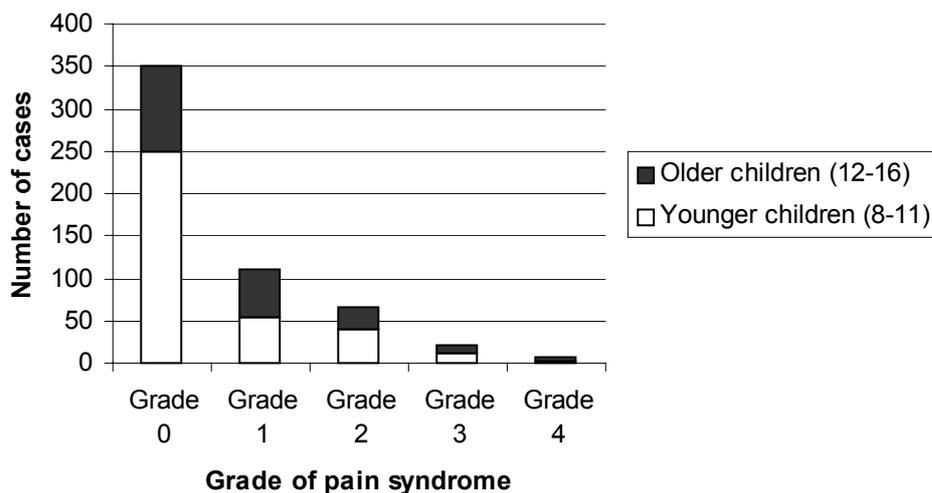


Figure 6. The number of cases distribution of graded chronic pain severity by age

Graded chronic pain severity was related, on the one hand, to child's quality of life ( $F=11.14$ ,  $p<0.001$ ), the level of quality of life (both physical and psychosocial functioning) showed a tendency to decrease with pain grade (see Table 3). No differences were observed between quality of life of children with no chronic pain and children in grade I, but mean differences were found between these children and children with higher grade ( $p<0.05$ ). No differences were found among higher grades. On the other hand, graded chronic pain severity was also related school absence in the month before the interview took place ( $F=7.31$ ,  $p<0.001$ ): the mean number of days absent from school was 0.77 for children with no chronic pain, 0.77 for grade I, 0.83 for grade II, 1.17 for grade III and 5.40 for grade IV. Posthoc analysis for ANOVA analysis revealed, however, that the only significant differences were between children at grade 0 and children at grade IV ( $p<0.001$ ). Finally, graded chronic pain severity was

also related to visits to specialists and medication consumption, that is to say the greater the grade of pain severity the higher the likelihood that children visited a health care professional or used medication due to their pain problems, (Chi-square<sub>(8)</sub>=40.93,  $p<0.001$ ; Chi-square<sub>(8)</sub>=25.56,  $p<0.01$ , respectively). Whereas 39.3% of children with no chronic pain problems had consumed pain medication during the previous three months to the interview for an occasional pain problem and 29.7% had visited a doctor, more than half of children in grade II onwards had consumed pain medication and visited a doctor (see Table 2).

For each pain severity grade, the effect of children's sex and age on the use of pain medication and the use of health care service was also examined. Regarding children's gender, females in grade I and II were more likely to consume pain medication (Chi-square<sub>(2)</sub>=9.45,  $p<0.01$ ; Chi-square<sub>(2)</sub>=6.45,  $p<0.05$ ); however no differences were found between boys and girls in terms of their visits to health care professionals. Regarding children's age, only in grade II children who had consumed pain medication were significantly older than children who had not ( $t=-2.41$ ,  $p<0.05$ ). Pain-related visits to health care professionals were not related to children's age in any grade.

#### 4. Discussion

This is the first work to study the epidemiology of chronic pediatric pain in a Latin country. Results from this study corroborate that chronic pain is an important problem among Catalan schoolchildren, similar to what has been claimed in previous studies conducted in Anglo-Saxon and Northern European countries.<sup>2,3,28,42, and 48</sup> The prevalence of reported chronic pain in this study was higher compared with previous major works, such as Perquin et al.<sup>42</sup>, a study with similar characteristics to this one. Such a discrepancy might be explained by differences in data collection and/or differences in participation rate. On one hand, whereas Perquin and colleagues only collected pain-related information (e.g. pain duration) about the most troublesome pain problem in order to study the prevalence of chronic pain, we collected the same information for any pain experience within the previous three months. The results of our study show that a very intense occasional (acute) pain can be considered as being more interfering with the daily living of the child than a chronic pain problem. Therefore, Perquin and her colleagues could have underestimated the prevalence of chronic pain by just collecting information about the most troublesome problem. On the other hand, the participation rate in our study was lower than in Perquin et al's work (62.2% vs 82%). Due to the special characteristics of our collecting data procedure (i.e., personal interviews that were very time consuming), parents who agreed to participate might have been those that were most concerned about this problem (for example, they might have been worried by their child's pain problems), this might have somewhat contributed to a potential overestimation of chronic pain. However, this hypothesis can not be confirmed because no data is available for non-participants. Also, such a discrepancy in the prevalence of chronic pain could be explained by socio-cultural differences;<sup>7,55,62, and 63</sup> Mediterranean people may be more likely to be aware of

and express physical complaints than people from other cultural environments.<sup>1 and 64</sup> Cross-cultural investigations of pediatric chronic pain are needed to test this hypothesis.<sup>20 and 45</sup>

Lower limb pain, headache, and abdominal pain were the most frequently reported chronic pain problems, as it is the case in previous reports on the epidemiology of chronic pediatric pain;<sup>42 and 49</sup> the co-occurrence of chronic localized pains at multiple anatomic sites was also commonly reported, mainly among girls in our study (see for similar results).

Our data also corroborated the gender and age differences in the prevalence of chronic pain reported in several other available studies.<sup>31,35,42, and 56</sup> Both boys and girls report to be more at risk of suffering chronic pain as they become older. At the same time, girls report to suffer much more often chronic pain problems than boys do. In spite of this general finding related to the child's gender, it might also be true that some types of chronic pain conditions have different gender prevalence patterns. For example, according to some epidemiological studies, our data shows that lower limb chronic pain is more frequent among boys than among girls, and the co-occurrence of multiple sites in pain is more frequent among girls than among boys.<sup>37</sup>

Another remarkable finding in this study is the high proportion of children who consume pain medication when in pain; the likelihood to consume pain medication is greater for children suffering from chronic pain. Future studies should examine both the frequency of pain-related visits to health professionals, and the levels of medical consumption, and their relation to chronic pain severity and related dysfunction.

Our study, not only showed that chronic pain is a common problem in the general population of children, one that often demands medical attention, it also demonstrated, once again, that chronic pain negatively impacts the everyday functioning of the children. For example, our data showed that children suffering from a chronic pain syndrome have a worse quality of life when compared to those without chronic pain.<sup>24,40, and 41</sup> Likewise, chronic pain children reported higher functioning impairments, as a result of their pain, than did children that reported to only suffer occasional pain problems.

This is the first study presenting a classification criteria for grading chronic pediatric pain severity status. It shows that although chronic pain is a prevalent complaint in the general population of children, a smaller proportion (around 5% of the children) suffer from moderate or severe chronic pain syndromes.<sup>9, and 49</sup> That is, most children are able to cope quite effectively with their chronic pain problems, regardless of their pain intensity. The levels of quality of life of these children remain high in comparison to clinical samples;<sup>5</sup> and the same is true in relation for chronic pain related disability: in general, disability levels appear to be on the lower range if compared to clinical populations.<sup>4,10,44, and 61</sup>

Previous studies have rated the severity of chronic pain on the basis of its intensity and frequency characteristics.<sup>42, and 49</sup> However, our results showed that these two parameters are not enough to determine a given pain status. That is, children can report intense and frequent pain episodes for more than 3 months, but this pain is sometimes not interfering their daily functioning (i.e., grade II children), therefore other parameters, besides intensity and frequency,

should be taken into account in order to describe with precision the severity of chronic pediatric pain, and better understand its impact on children's quality of life (see Hunfeld et al.<sup>24</sup> for a related discussion).

The pattern of observed relationships between pain status, pain characteristics, and indicators of health status is somewhat interesting. Children with more severe chronic pain syndromes tend to report worse pain characteristics (i.e., higher pain intensity, more frequent pain episodes, and higher levels of pain related functional disability). Also, children with more severe chronic pain syndromes are more likely to report worse functioning (i.e., worse quality of life, more days absent from school). Finally, children with more severe chronic pain syndromes tend to consume medication and consult a health care professional for their pain problems. Because of the cross-sectional design of this study, it is unclear what is the cause and what is the effect. Longitudinal studies are warranted to clarify the direction of these associations.

As for sex differences in the severity of pain syndromes, girls aged between 8 and 16 years are more likely to suffer more severe chronic pain syndromes than boys. Other published studies would support this finding that girls have significantly higher pain intensity and frequency<sup>24</sup> or that girls are at higher risk for continuing chronic pain after a 3-year period of time.<sup>31</sup> Such sex differences in prevalence of chronic pain could be the reason why girls consume medication much often than boys in spite of the fact that the likelihood that girls visit a doctor for their pain is higher than boys has not been confirmed in our data.

In this study, the severity of a chronic pain syndrome seems to be related to certain pain locations. Lower limb pain is associated with less severe problems, whereas headaches are associated with more severe ones. This could help explain why, in this study, headaches are the most frequent pain-related reason to visit a doctor, and it is in accordance with Hunfeld et al.'s study<sup>23</sup>, which reported that headache sufferers had lower levels of quality of life and larger school absence due to their pain in comparison with sufferers of chronic pain at other sites. On the other hand, it seems paradoxical that lower limb chronic pain that is associated with less severe problems is rated as the most bothersome pain. One possible explanation might be that limb pain, in comparison to other pains, particularly interferes with physical functioning, so children with lower limb pains might be more particularly restricted in physical areas, than children with pain at other anatomical sites.

A systematic examination of epidemiological data with this grading system as a template may be helpful to both basic and clinical pain researchers to generate hypotheses in our search for a better understanding of chronic pediatric pain. The findings of this study provide preliminary support to using Von Korff's proposal to grade chronic pediatric pain severity on the bases of several different variables (i.e., variables related to pain characteristics, variables related to child's functioning, and variables related to medical issues). Nevertheless, few cases with severe chronic pain syndromes participated in this study; this might be the reason for which some analysis did not reach statistical significance. Thus, additional studies need to be conducted to test the suitability of our proposal to grade chronic pediatric pain syndromes on the basis of their severity.

Although the participation rate was not as high as in other epidemiological studies, the large sample size, the randomised study design, the reliable and accurate method of data collection (i.e., individual interviews by highly skilled and trained professionals), as well as the consistency between our findings with those from previous published reports, guarantee that these results may be fairly generalizable to the population of schoolchildren aged between 8 and 16 years.

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**Paper #5:**

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## **PREDICTIVE FACTORS OF CHRONIC PEDIATRIC PAIN AND DISABILITY: A DELPHI POLL**

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### **ABSTRACT**

The purpose of this study was to establish consensus on the factors that predict chronic pediatric pain and pain-related disability. A Delphi poll involving two rounds of data collection was used as a way to reach consensus among professionals with a specific interest in chronic pain in children and adolescents. Amongst the factors that were thought to have the greatest influence on the long-term maintenance of pediatric pain there were: excessive use of health care services, a tendency to somatize, and children's catastrophic thinking about pain. Whereas, amongst the factors thought to have the greatest influence on long-term disability we found: children's self-concept as being disabled, a hesitance to perform exercise because of fear of a potential injury, and children's catastrophizing thinking. These findings may be used to help identify the specific domains that should be assessed in studies seeking to predict which children are at risk to develop chronic pain and disability. If these results are supported by future studies, then they could be used to help develop intervention programs that could effectively prevent long-term pain and disability in children and adolescents who are at risk for chronic pain.

**Keywords:** Delphi poll, pediatric pain, chronic pain, disability, predictive factors, secondary prevention

### **PERSPECTIVE**

A Delphi poll was conducted to reach consensus among professionals with a specific interest in chronic pain in children and adolescents regarding the set of factors most highly associated with the development of chronic pediatric pain and disability. This study could serve as the basis for (1) prospective studies to validate the predictive utility of the variables, and (2) the design of secondary prevention programs.

## 1. Introduction

Over the past two decades, we have moved from a unidimensional view, to a more comprehensive understanding of pediatric pain that conceptualizes children's pain experience as multifactorial. It is now recognized that a variety of factors likely act in combination to contribute to the development and maintenance of chronic pain in children.<sup>2, 15</sup> However, the very complex nature of the experience of pediatric pain is far from being completely understood. On one hand, most of the studies provide some evidence that a few isolated factors are related to the development of chronic pain and/or to chronic pain related disability.<sup>3, 9, 19, 23, 24, 27, 30</sup> On the other hand, there are some methodological problems with the existing research, such as the use of small sample sizes, differences in the definition of the pain problem or in the study population, inadequate study designs and methods of data collection, or even the type of statistical analysis, that limit the interpretation of the data and our ability to reach robust conclusions.

By studying the factors that interact to create this complex so-called *puzzle of pain* (Melzack, 1973)<sup>16</sup>, clinicians and researchers could have better and detailed information on how to design more efficient treatment programs for children and their families. Moreover, treatment programs could be developed that not only address the pain problem when it is already chronic and disabling, but to also, perhaps, to prevent the development and maintenance of chronic pain. Programs that prevent pain from becoming a chronic disabling condition (*i.e.*, secondary prevention) have already been suggested and tested in adults, with some promising results mostly for chronic back pain problems.<sup>4, 12, 20</sup> We are not aware that any such programs have yet been developed and tested for children.

The present study was designed to gather information through consensus among professionals with a specific interest in chronic pain in children and adolescents about factors that might be related to the development of chronic pediatric pain and pain-related disability, that might serve as the basis for the development of preventive interventions. In this attempt to identify predictive factors, it is worth differentiating chronification and disability on purpose because, despite of the fact that they are related, they are not the same; a patient may suffer from chronic pain but may not be disabled at all. A *Delphi method* was selected to serve this purpose.

The Delphi method was developed by the Rand Corporation in 1950s to reach consensus from people by answering a questionnaire in an iterative process.<sup>11</sup> Cumulative research indicates that results from Delphi studies provide the most accurate answers to difficult questions compared to other prognostication techniques, and the judgment of people is recognized as a legitimate and useful input in generating forecasts.<sup>5, 21</sup>

The primary objective of the current study was to reach consensus among professionals with a specific interest in chronic pain in children and adolescents on the factors that predict chronic pediatric pain and related disability, using the Delphi method. There were also two related secondary objectives: (1) to identify whether factors predictive of persistent pain can be

differentiated from those predictive of functional disability, and (2) to study whether there is any factor that professionals consider to be protective, and thus may play a positive or buffering role.

## **2. Method**

### **2.1. Participants: The Delphi panel**

The consensus group was constructed so as to represent the professionals with a specific interest in chronic pain in children and adolescents. To support the content validity of the Delphi study, and to ensure an appropriate panel, we followed suggestions of previous work in the area<sup>28</sup>. Potential participants were identified for the Delphi panel if they met any of the two following inclusion criteria: (1) previous involvement in clinical research on this topic: at least to be the first author of one paper or the second author of two papers focused on prognostic factors of chronic pain and pain-related disability and published in peer-reviewed journals, and/or (2) previous involvement in pain clinical work: at least two years of experience working in a pediatric pain service. Only one of the criteria was required because our intention was to include both researchers and clinicians interested in the field.

According to the inclusion criteria, the recruitment of researchers was based on a review of the literature. We conducted a search limited to child population to identify all published papers on pain indicators, prognostic factors of chronic pain and related disability that appeared in refereed journals during 1995-2005. The aim of this review was twofold. First, to identify prognostic factors of chronic pain and related disability. Second, to identify professionals interested in this topic. For the search in the database, we consulted in Medline. We used *pain* as the Medical Subject Heading and *disability, quality of life, prognosis, prognostic factor, predictors, course, risk\*, natural histor\*, epidemiol\*, prevalen\**, and *treatment* as text words. We also searched reference list of relevant retrieved studies or secondary resources (such as books) to identify additional studies. Titles and abstracts were reviewed for inclusion. When an article which provided information about prognostic factors was identified, a summary form was filled. The whole procedure was independently conducted by two reviewers. The information collected was used to select researchers with an special interest to this topic. Only the authors who met the first criterion were invited to participate in this study.

For the recruitment of clinicians, we also contacted two different subscriber pain lists: the *IASP Special Interest Group on Pain in Childhood*, and the subscribers to the *Pediatric-Pain list serve*, which is an international Internet forum for informal discussion of any topic related to pain in children. Only those people who met the second inclusion criteria were invited to participate.

Additionally, we also asked to identified participants for names of other professionals working on pediatric pain (either in academic or non-academic settings), whom they believed

could be of value to the project. We reviewed each of the recommended potential participant, and requested participation for any that met the inclusion criteria.

Following this procedure, a list of candidates from Australia, Europe, North and South America was compiled, and invited to participate in this Delphi poll. As can be seen in Table 1, the list of candidates was formed by a multidisciplinary international group, with participants coming from clinical practice and research communities, including anesthesiology, epidemiology, nursing, pediatric oncology, pediatric rheumatology, rehabilitation medicine, and psychology.

Table 1. Demographic characteristics of participants

		Invited participants	Participants Round 1	Participants Round 2a(*)	Participants Round 2b(*)	Z <sub>(round 1-2a)</sub>	Z <sub>(round 2a-2b)</sub>	Z <sub>(invited-round 1)</sub>	
Number of participants		159	56	38	28				
Sex	Male	75 (48.7%)	30 (53.6%)	22 (57.9%)	18 (64.3%)	0.41	-0.53	0.62	
	Female	79 (51.3%)	26 (46.4%)	16 (42.1%)	10 (35.7%)	0.41	0.53	-0.62	
Profession	Physician	90 (60%)	35 (62.5%)	27 (71.1%)	23 (82.1%)	-0.87	-1.03	0.33	
	Psychologist	37 (24.7%)	13 (23.2%)	8 (21.1%)	4 (14.3%)	0.24	0.71	-0.22	
	Nurse	20 (13.3%)	5 (8.9%)	3 (7.9%)	1 (3.6%)	0.13	0.72	-0.86	
	Other	3 (2.1%)	3 (5.4%)	0	0	1.46	0	1.27	
Activity	Clinic		19 (33.9%)	15 (39.5%)	13 (46.4%)	-0.55	-0.56		
	Research		5 (8.9%)	1 (2.6%)	1 (3.6%)	0.39	-0.23		
	Clinic + Research		32 (57.1%)	22 (57.9%)	14 (50%)	-0.02	0.64		
North America	United States	53 (33.33%)	20 (35.7%)	12 (31.6%)	9 (32.1%)	0.41	-0.04	0.32	
	Canada	34 (21.4%)	12 (21.4%)	10 (26.3%)	4 (14.3%)	-0.55	1.18	0.01	
South America	Brasil	1 (0.6%)	0	0	0	0	0	-0.59	
Citizenship	Belgium	4 (2.5%)	3 (5.4%)	1 (2.6%)	1 (3.6%)	0.67	-0.23	1.02	
	Finland	1 (0.6%)	1 (1.8%)	0	0	0	0	0.77	
	Italy	1 (0.6%)	1 (1.8%)	1 (2.6%)	1 (3.6%)	0.67	-0.23	0.77	
	Spain	36 (22.6%)	14 (25%)	12 (31.6%)	11 (39.3%)	-0.7	-0.65	0.36	
	United Kingdom	10 (6.3%)	2 (3.6%)	1 (2.6%)	1 (3.6%)	0.67	-0.23	-0.76	
	Europe	Germany	1 (0.6%)	1 (1.8%)	1 (2.6%)	1 (3.6%)	0.67	-0.23	0.77
		The Netherlands	5 (3.1%)	1 (1.8%)	0	0	0	0	-0.53
		Switzerland	1 (0.6%)	0	0	0	0	0	-0.59
	Sweden	3 (1.9%)	0	0	0	0	0	-0.59	
	Norway	1 (0.6%)	0	0	0	0	0	-0.59	
	Austria	1 (0.6%)	0	0	0	0	0	-0.59	
	Denmark	3 (1.9%)	0	0	0	0	0	-1.03	
	Australia	4 (2.5%)	1 (1.8%)	0	0	0	0	-0.31	

Note: Activity data was only available for participants. Sex, profession and citizenship was available for the most part of the invited participants.

(\*) Participants round 2a= Participants answered from 1<sup>st</sup> to 110<sup>th</sup> item.  
 Participants round 2b= Participants answered from 111<sup>th</sup> to 127<sup>th</sup> item.

None of the comparisons was significant at p<0.05

Our goal was to get involved a multidisciplinary international sample of at least 30 research and clinical professionals with a broad range of expertise in chronic pain and pain related disability in children in this Delphi exercise. This criteria followed the rule that a panel of 30 participants is enough in Delphi studies<sup>11</sup>.

Once the list of candidates was ready, however, we were concerned about the issue that candidates could have different levels of expertise in the field, and that could have an effect on the results of our study. Since not all participants had the same level of expertise, we decided to weight their answers. This is a common issue in Delphi studies<sup>11</sup>. Thus, in order to approach this issue, firstly, the level of expertise of participants was estimated. Number of years of clinical experience, as well as quality (based on the impact factor value of the paper), and quantity of published papers in the previous 10 years, were used to estimate participant's level of expertise. Then the data were analyzed by weighting the participants' answers according to their level of expertise: the higher the total level of expertise, the greater impact the participants answers exerted on the report.

## **2.2. List of potential predictors**

In order to know what might predict chronic pain and disability from the point of view of our panel of professionals with a specific interest in chronic pediatric pain, we developed an initial list of potential predictors based on two sources of information. The first source was our review of the literature described above. The second source was an international multidisciplinary group of four academic health researchers who were selected because of their expertise in the field of pain in children. These professionals were requested to add to the list other factors that, although had not been studied yet, they considered important for the prediction of chronic pain and/or related disability. From these two sources of information we prepared an initial list of 28 potential predictors. Most of the predictors were described by using operational definitions available in the literature, so that all the participants worked on the basis of the same idea. For example, "pain catastrophic thinking" was conceptualised, as Crombez and his colleagues suggest<sup>1</sup>, as 'a tendency to dwell on the pain and magnify or exaggerate its threat or seriousness and to be incapable to deal with it'<sup>29</sup>; this is the definition that was presented to the participants for their analysis. On the other hand, eight items were used for pain coping strategies, these eight items defined the eight subscales suggested Reid and his colleagues<sup>25</sup> (see appendix 1).

We generated a list of 110 items that described those 28 potential predictors, and this was the list of items that was presented to the participants in the first round. In the first round participants were asked about additional factors that they considered important and were not present in the initial list. They were also asked to report if there was any item in the list that was not clear enough. Taking into account the participants' comments, 16 new items that assessed 4 new potential predictive factors were added to the list (the procedure followed to elaborate items from factors was the same as described above), and one item was deleted (item 80), because the participants felt that it overlapped with another one (item 127) (see appendix A).

### **2.3. Procedure**

The study was reviewed and approved by the Ethics Committee of the Rovira i Virgili University.

All communication with the panel of participants occurred via the Internet (see Figure 1 for a complete description of the procedures). Once the list of items was developed and made available to participants, then they were asked to indicate which items of the list were related to (*i.e.*, predicted) chronicity, disability, both chronicity and disability, or neither chronicity nor disability. Only for those items that were thought to have an effect on chronicity, disability, or both, the participants were asked to express how and to what extent those items had an influence. Specifically, the participants were asked to provide a value between -100 and 100, depending on whether that item was considered to exert a positive (*i.e.*, protective influence), or a negative influence (that is to say, whether it was considered a potential risk factor) for the development of chronic pain and/or pain-related disability. The value that the participants gave represented both the type and the extent of the association: (1) -100 meant that it was a protective factor that was highly associated with well-being and, therefore, with a decreased likelihood of the development of chronic pain and/or disability, (2) 0, the mid-point on the scale, meant that the item did not predict pain or disability one way or another (*i.e.*, it was neither a protective or a risk factor), and (3) 100 meant that it was a risk factor that was highly associated with the development of chronic pain and/or disability.

To assure understanding of the task as well as the appropriateness of the procedure, a pilot test was completed. Before inviting potential participants, two internationally recognized experts in the pediatric pain field, who otherwise had no direct involvement in the research project, were asked to complete the task once, and report any problematic issues. Their comments and suggestions led to some minor changes.

Detailed information about the coordinating team of investigators, and the research project was available in the study Web page (<http://www.sre.urv.es/delphy/nens/english/index.htm>). Although this information was accessible to anyone who was interested, the on-line list of items which, was requested to be assessed, was only accessible to the participants. In order to access the on-line list, the participants had to use their login and password, provided by us. All the information was available in English, Catalan, and Spanish.

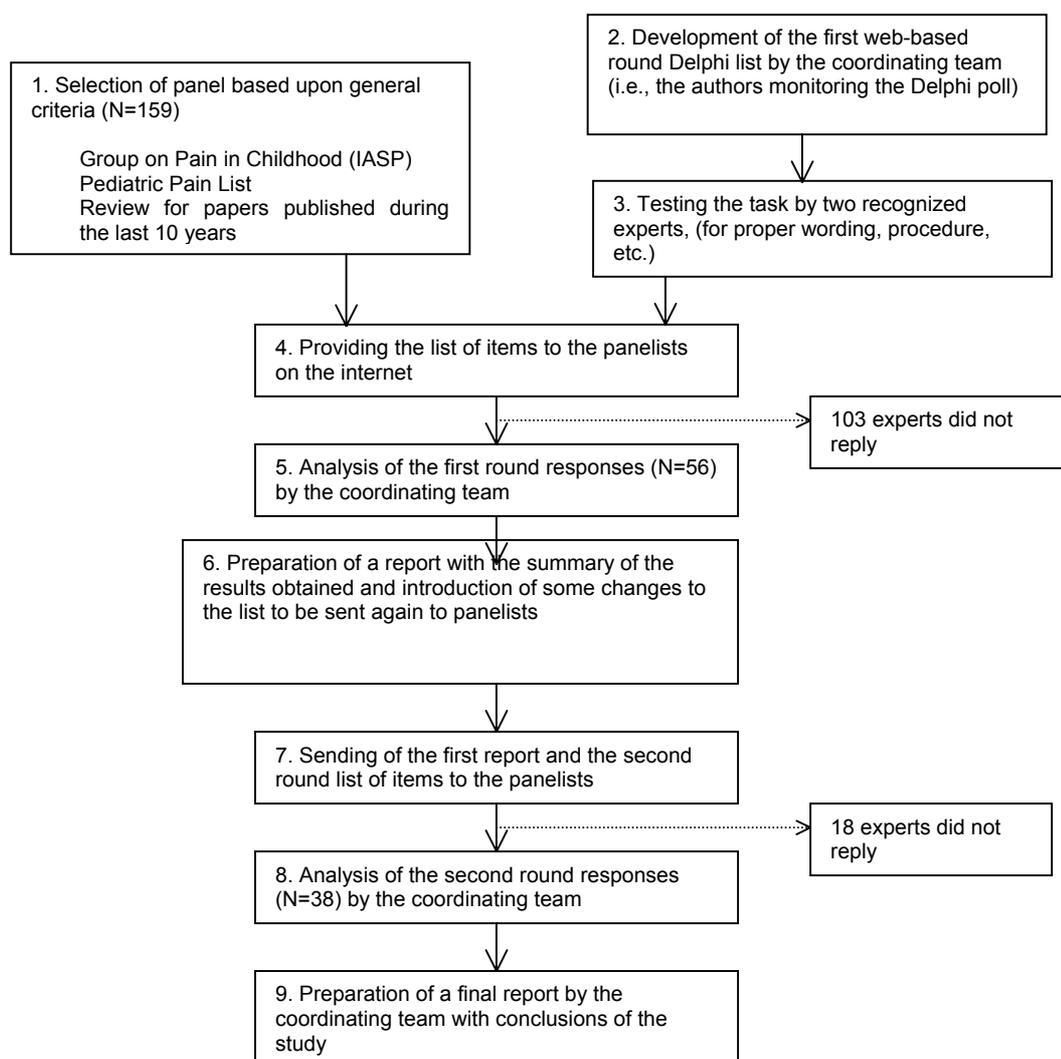


Figure 1. Steps in the Delphi poll.

## Delphi rounds

### Round 1

At the end of March of 2005, 159 potential participants were invited to participate. Participants were assured that their answers would be kept strictly confidential. Each identified potential participant received an electronic message that briefly introduced the research project, and encouraged to take part in it. If the professional invited was interested, she or he was instructed to visit the web of the project where additional information could be found, along with the items that were requested to be assessed. Participants were first asked to provide information about their professional activities. They were then given instructions concerning how to assess each of the items presented to them.

Participants were asked to assess all items within about 3 weeks. During that 3-week time interval, 2 reminders were sent in order to maximize the involvement of as many participants as possible. The task took approximately 45 minutes to be completed. After that 3-week time interval, we analyzed the responses to the list of items. A report describing the

results of the first iteration was then sent to all participants. It included a table and a figure, with the table presenting a summary of the responses of the participating panel as a whole. The mean scores, standard deviations, and 95% confidence interval with the lower and upper values for each item, and the numeric value that the participant had given to rate each item were provided. The figure showed the mean of the participant scores for all the items.

### *Round 2*

At the beginning of April, 2005 the results of the first iteration were emailed to all of the participants who had provided responses to the first list, enabling them to see the position of the participants as a group, as well as the level of agreement among the participants, for each item. The aim of providing this information was to help the participants re-examine their initial responses in light of their colleagues' opinions. After reading the report, they were asked to return to the web page and complete again the same task, that is, to re-assess the list of items. As already mentioned, in this second round, new items were included. These new items described new factors which had been suggested by participants (item 111 to item 127; see Appendix A). Again, the panel of participants were given a 3-week time interval to complete the task. At this point, items 111 to 127 had only been rated once. At the end of this period, the information was again analyzed. A second report was completed and emailed to the participants, who were asked to rate only the new items introduced in the second round for a second time.

The stability in participants' answers was used as the criterion to determine that consensus had been reached, and indicated that 2 rounds were enough to reach stability. This is consistent with the findings of previous Delphi studies<sup>5</sup> (see next section). Figure 1 provides a complete description of the process used in this study.

## **3. Results**

### ***3.1. Participating panel***

A total of 159 potential participants from Australia, Europe and North America was compiled, and invited to participate in this Delphi poll. 103 of these declined to participate (15 reported that they did not have time to take part in the research at that moment, 3 thought that they had not enough knowledge to do what they were required to do, and the remainder did not respond to any of the email messages). Thus, 56 professionals invited responded to the first round; a 35.22% response rate. Thirty-eight of these (67.86%) responded to the second round, and 28 (50%) responded to our request to report on items 111-127 for a second time.

Demographic characteristics of the participants are shown in Table 1. In the first round, 53.6% of the participants were male. Most of the participants (62.5%) were physicians, while 23.2% were psychologists, 8.9% were nurses, and 5.4% had other professions. The first round participants had, on average, 17.20 years of experience (range= 2 - 42 years; SD= 9.15) in the

pediatric pain field (clinical and/or research experience). Most (57.1%) were involved in both clinical and research activities, whereas 33.8% were involved only in clinical work, and 8.9% only in research activity. The participants came from 10 countries (see Table 1 for a description of the characteristics of participants in each round).

As expected, participants had different levels of expertise both in research and clinical activities. We examined whether their level of expertise would make a difference in their responses to the poll. Thus, as previously reported, we analyzed the data by weighting the participants' answers according to their level of expertise (for more details see Tables 2 and 3). No differences were observed between weighted and non-weighted responses. Thus, results reported in this paper are based on the analysis of participants non-weighted responses since the level of expertise did not have any effect on the results.

*Table 2. Formulas for calculating an adjusted level of expertise for participants.*

Level of research expertise
<p>The level of research expertise was based on the quality and quantity of papers published on chronic pediatric pain. We searched for publications indexed in Medline data base in the last 10 years. The procedure used to estimate the level of research expertise was to give a numeric value to publications based on the quality of the journals considering the following:</p> <p>A. Non-reviewed journals = 0.25</p> <p>B. Peer-reviewed journals = 0.75</p> <p>C. Journals with impact factor:                      Journals whose impact factor is under Pc25 (0.94)= 1                      Journals whose impact index is between Pc25 (0.95) and Pc50 (1.84)= 2                      Journals whose impact index is between Pc50 (1.84) and Pc75 (2.66)= 3                      Journals whose impact index is over Pc75 (2.66) = 4</p> <p>* The level of expertise of each participant in the Delphi panel was based on the sum of punctuations of her/his publications (A+B+C). Level assigned to each of the collaborating experts (0-10):                      Participants whose scores were 0 (no publications)= 0                      Participants whose scores ranged under Pc20 (3.4)= 2                      Participants whose scores ranged from Pc20 (3.4) to Pc40 (8.8)= 4                      Participants whose scores ranged from Pc40 (8.8) to Pc60 (19.2)= 6                      Participants whose scores ranged from Pc60 (19.2) and Pc80 (33.85)= 8                      Participants whose scores ranged over Pc75 (33.85)= 10</p>
Level of clinical experience
<p>The level of clinical experience was based on the number of years worked in the treatment of pediatric pain problems. To estimate the level of clinical expertise, we considered the maximum number of years of any participant as an anchorage:</p> <p>Participants that been worked in a clinical setting= 0                      Participants having worked for a period of time less than Pc20 (9.4)= 2                      Participants having worked during a period of time between the Pc20 (9.4) and Pc40 (15)= 4                      Participants having worked during a period of time between the Pc40 (15) and Pc60 (20)= 6                      Participants having worked during a period of time between the Pc60 (20) and Pc80 (25)= 8                      Participants having worked during a period of time more than Pc80 (25)= 10</p>
Total level of expertise
$\text{Total level of expertise} = 5 * + \frac{\text{Level of research expertise}}{4} + \frac{\text{Level of clinical expertise}}{4}$ <p>(*) The minimum expertise level was computed as 5 out of 10 because all invited participants at least accomplish the inclusion criteria.</p>

*Table 3. Means and standard deviations of the participants level of expertise in each round*

	Participants Round 1	Participants Round 2a* <sup>1</sup>	Participants Round 2b* <sup>1</sup>	t <sub>(round 1-2a)</sub>	t <sub>(round 1-2b)</sub>
Level of clinical expertise (0-10) <sup>*2</sup>	5.11 (2.95)	5.57 (2.71)	5.85 (2.60)	-0.03	-0.03
Level of research expertise (0-10) <sup>*2</sup>	4.5 (3.58)	4.63 (3.72)	4.21 (3.58)	-0.01	0.03
Total level of expertise (0-10) <sup>*2</sup>	7.40 (1.18)	7.55 (1.16)	7.51 (0.95)	-0.03	0.01

(\*<sup>1</sup>) Participants round 2a= Participants answered from 1<sup>st</sup> to 110<sup>th</sup> item.  
 Participants round 2b= Participants answered from 111<sup>th</sup> to 127<sup>th</sup> item.

None of the comparisons was significant at p<0.05.

(\*<sup>2</sup>) Table 2 shows the formulas for calculating the level of expertise.

### **3.2. Number of Delphi rounds**

Delphi methodology requires that the procedure be repeated until participant answers reach stability, or when a point of diminishing returns is reached. <sup>6</sup> The stability in the variability of the participants answers was used to determine that the process could end. Paired t-tests were used to detect any significant difference in the variance of the items between consecutive rounds. Results showed that the feedback provided to participants did not significantly change the variability of responses for almost all items (only in 2 items related to chronic pain, and in 6 items related to disability some statistically significant difference was observed in the item's variance, p<0.001<sup>2</sup>). Consequently, there was no need for a third round of ratings, 2 rounds had been enough, as predicted <sup>5</sup> (see Figure 1). Moreover, no statistically significant (p<0.001) difference between the two rounds were observed in how participants categorized the items. That is, 100% of the items were believed to have some kind of effect on the development of chronic pain in both assessments by the same proportions of participants, and 100% of the items were believed to have some type of effect on the disability in both assessments by the same proportions of participants. Paired t-tests for dependent samples were also carried out to determine whether the predictive value attributed to the items, when they were considered predictors of development of chronic pain and when they were considered predictors of disability, differed between rounds. The results of these analyses consistently showed no statistically significant (p<0.001) differences between the first and second rounds for all items.

### **3.3. Professionals consensus on chronic pediatric pain and related disability predictive factors.**

In relation to the first objective, that is, to determine a participant consensus on the factors predictive of chronic pediatric pain and pain-related disability, three different analyses were performed. First, we identified those items that showed the highest consensus among

<sup>2</sup> Due to the large number of analyses in this study (127 statistical tests) it was necessary to lower alpha level required for significance, in order to control for *Type I error* and increase the confidence that findings identified as significant would be reliable. A highly conservative approach, such as the *Bonferroni*, in which the chosen significance level is divided by the number of analyses to yield a p value that must be reached for results to be deemed significant, would result in a p value so low (0.05/127= 0.0004) that it substantially increases the risk of *Type II errors*. Thus, we elected to compromise between these two risks, and choose a p value of 0.001 that must be reached in order for a particular finding to be deemed statistically significant.

participants about their nature (*i.e.*, chronic pain predictors, disability predictors, or factors with no predictive value). In this analysis, the percentage of participants that agreed on the nature of the items was examined.

Items with an agreement equal or greater than the 75% of the participants were considered items with a high consensus about his status as predictors of chronic pain and/or disability. This criterion was established a priori. The confidence interval of the percentage of participants was used to determine consensus, rather than the discrete estimation, because the confidence interval has lower amount of error. Table 4 presents those items for which more than 75% of the participants, at the level of 0.05, agreed about their having some predictive value on the course of the pain problems (specifically on the development of chronic pain and/or pain-related disability). In some items, an agreement higher than 75% was only achieved for one of the two outcomes assessed (chronic pain and disability), while other items achieved an agreement higher than 75% for both outcomes.

*Table 4. Items for which an agreement about their predictive nature between participants is equal or higher than 75%*

		PREDICTOR OF CHRONIC PAIN			PREDICTOR OF DISABILITY			NON-PREDICTOR
		% of participants that consider the item as predictive	Relationship with chronic pain *		% of participants that consider the item as predictive r	Relationship with disability *		% of participants that consider the item as non-predictive
			Risk factor	Protective factor		Risk factor	Protective factor	
CHRONIC PAIN AND DISABILITY	20.The child has a tendency to somatize	92.1 <b>83.24-100</b>	100	0	89.5 <b>79.43-99.57</b>	100	0	0 0-0
	34.The child has a depressive personality.	89.5 <b>79.43-99.57</b>	97.1	2.9	81.6 68.88-94.32	96.8	3.2	5.3 0-12.66
	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.	86.8 <b>75.69-97.91</b>	100	0	86.8 <b>75.69-97.91</b>	100	0	5.3 0-12.66
	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.	89.2 <b>79.01-99.39</b>	100	0	86.5 <b>75.28-97.72</b>	100	0	2.7 0-8.02
	116. The parents are able to identify, assess and alleviate the child's pain	89.3 <b>77.32-100</b>	4	96	96.4 <b>89.18-103.6</b>	3.7	96.3	3.6 0-10.82
CHRONICITY	1.The child has an anxious personality.	86.8 <b>75.69-97.91</b>	97	3	68.4 53.14-83.66	96.2	3.8	7.9 0-16.76
	7.The child uses the health care services excessively for his pain complaints.	94.7 <b>87.34-100</b>	100	0	81.6 68.88-94.32	100	0	0 0-0
	16.The child is subject to stress in his/her immediate environment (for example: family difficulties, problems at school)	97.4 <b>92.18-100</b>	100	0	73.7 59.24-88.16	100	0	2.6 0-7.83
	23.The parents are emotionally unstable.	89.5 <b>79.43-99.57</b>	100	0	76.3 62.34-90.26	100	0	7.9 0-16.76
	30.The child's consumption of medicaments to relieve the pain is not appropriate.	86.8 <b>75.69-97.91</b>	100	0	81.6 68.88-94.32	100	0	5.3 0-12.66
	31.The child has had previous experience of recurrent episodes of acute pain.	86.8 <b>75.69-97.91</b>	97	3	57.9 41.69-74.11	95.5	4.5	10.5 0.43-20.57
	36.The child was exposed, in a prolonged or repetitive way, to nociceptive stimuli in the first stage of life.	86.8 <b>75.69-97.91</b>	97	3	57.9 41.69-74.11	95.5	4.5	10.5 0.43-20.57
42.The child's pain is constant.	86.8 <b>75.69-97.91</b>	100	0	81.6 68.88-94.32	96.8	3.2	7.9 0-16.76	

*Table 4 (cont'd). Items for which an agreement about their predictive nature between participants is equal or higher than 75%*

	PREDICTOR OF CHRONIC PAIN				PREDICTOR OF DISABILITY			NON-PREDICTOR
	% of participants that consider the item as predictive	Relationship with chronic pain *		% of participants that consider the item as predictive r	Relationship with disability *		% of participants that consider the item as non-predictive	
CHRONICITY	68.The child receives attention and/or other privileges immediately after expressing pain.	92.1 <b>83.24-101</b>	97.1	2.9	71.1 56.22-85.98	92.6	7.4	0 0-0
	73.The child has a family history of chronic pain.	94.6 <b>87.18-100</b>	100	0	67.6 52.23-82.97	100	0	2.7 0-8.02
	77.Compliance with the therapy prescribed to treat the pain is low.	86.5 <b>75.28-97.72</b>	96.9	3.1	73 58.42-87.58	96.3	3.7	10.8 0.61-20.99
	108.The child believes that his/her pain will persist over time.	91.9 <b>82.94-100</b>	100	0	73 58.42-87.58	100	0	5.4 0-12.82
	111. The child has previous untreated pain.	89.3 <b>77.32-101.3</b>	100	0	78.6 62.71-94.49	95.5	4.5	7.1 0-17.05
	121. Child and parents think healthcare professionals don't believe his/her pain	89.3 <b>77.32-100</b>	100	0	82.1 67.25-96.95	100	0	7.1 0-17.05
	123. The child and his/ her parents have consulted numerous doctors about the pain but no one has found anything wrong	96.4 <b>89.18-100</b>	96.3	3.7	75 58.22-91.78	95.2	4.8	3.6 0-10.82
	5.The parents believe that pain incapacitates.	65.8 50.22-81.38	92	8	97.4 <b>92.18-100</b>	94.6	5.4	0 0-0
DISABILITY	54.The child tries to do activities that he/she enjoys (for example, playing) in an attempt to feel better.	78.9 65.5-92.3	0	100	86.8 <b>75.69-97.91</b>	3	97	7.9 0-16.76
	61.The parents are disabled as a result of their pain.	84.2 72.22-96.18	100	0	86.8 <b>75.69-97.91</b>	100	0	7.9 0-16.76
	62.The child seeks social support by talking to others (parents or friends) about his/her feelings.	81.6 68.88-94.32	6.5	93.5	86.8 <b>75.69-97.91</b>	9.1	90.9	10.5 0.43-20.57
	66.The child believes he/she is disabled by the pain.	73 58.42-87.58	100	0	94.7 <b>87.34-102.1</b>	100	0	0 0-0
	91.When the child's parents are in pain they believe that they are in danger and that they should avoid exercise as much as possible.	73 58.42-87.58	96.3	3.7	91.9 <b>82.94-100</b>	100	0	5.4 0-12.82
	106.The child believes that pain means he is damaging him/herself and that he/she should avoid exercise.	83.8 71.7-95.9	96.8	3.2	94.6 <b>87.18-100</b>	97.1	2.9	0 0-0
	110.The child constantly avoids activities that involve moving the part of the body that hurts through fear of experiencing more pain.	75.7 61.44-89.96	100	0	91.9 <b>82.83-100</b>	100	0	2.7 0-8.09
117. Parental anxiety is high.	82.1 67.25-96.95	100	0	92.9 <b>82.95-100</b>	100	0	0 0-0	

\* The numbers in this column indicate the percentages of respondents who viewed the items as being a risk factor versus a protective factor for the development of chronic pain or pain-related disability.

We then identified those items that had the greatest predictive power, that is those items that received a mean score higher than the 75<sup>th</sup> percentile. Because the respondents were asked to rate the items using a ratio scale, we determined that it was appropriate to use parametric statistics for analysis. Further support for this decision comes from the fact that the distribution of the scores assigned to each of the items was essentially normal for most of cases.

The results based on ranking items according to their mean scores are summarized in Table 5. These scores were based on participants' responses to a numerical scale with

anchorages: -100 to 100. Only those items whose mean was over 75<sup>th</sup> percentile are included in Table 5.

We also identified those items that had the highest consensus across raters. To identify these factors the coefficient of variation (SD/Mean) was used<sup>31</sup>. The lower the coefficient, the lower the variation among the responses is. Items with a coefficient of variation below percentile 25<sup>th</sup> were considered as items with a high consensus (see Table 5).

*Table 5. The mean value of responses of all participants and standard deviations with its 95% confidence interval*

	PREDICTORS OF CHRONIC PAIN	MEAN (Sd)	C.I.	PREDICTORS OF DISABILITY	MEAN (Sd)	C.I.
RISK FACTORS	7.The child uses the health care services excessively for his pain complaints.	46.34 (26.00)*	37.79-54.89	66.The child believes he/she is disabled by the pain.	50.74 (26.02)*	42.18-59.29
	20.The child has a tendency to somatize	44.32 (25.83)*	35.82-52.80	106.The child believes that pain means he is damaging him/herself and that he/she should avoid exercise.	46.62 (28.41)*	37.15-56.09
	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.	43.79 (31.21)*	33.53-54.05	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.	43.92 (30.11)*	34.02-53.82
	42.The child's pain is constant.	43.42 (28.83)*	33.94-52.90	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.	43.11 (29.89)*	33.14-53.07
	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.	42.40 (29.07)*	32.71-52.10	110.The child constantly avoids activities that involve moving the part of the body that hurts through fear of experiencing more pain.	40.76 (25.58)*	32.23-49.28
	34.The child has a depressive personality.	40.13 (30.36)*	30.15-50.11	20.The child has a tendency to somatize	38.10 (23.26)*	30.46-45.75
	108.The child believes that his/her pain will persist over time.	38.70 (29.66)*	28.81-48.59	7.The child uses the health care services excessively for his pain complaints.	36.76 (25.52)*	28.37-45.15
	16.The child is subject to stress in his/her immediate environment (for example: family difficulties, problems at school)	37.50 (21.14)*	30.55-44.45	5.The parents believe that pain incapacitates.	36.24 (30.74)*	26.13-46.34
	68.The child receives attention and/or other privileges immediately after expressing pain.	35.66 (28.73)*	26.21-45.10	42.The child's pain is constant.	35.39 (27.59)*	26.32-44.46
	123. The child and his/ her parents have consulted numerous doctors about the pain but no one has found anything wrong	35.39 (26.62)*	25.07-45.71	61.The parents are disabled as a result of their pain.	34.79 (24.48)*	26-74-42.84
	30.The child's consumption of medicaments to relieve the pain is not appropriate.	35.00 (29.11)*	25.43-44.57	117. Parental anxiety is high.	34.21 (25.41)*	24.36-44.07
	66.The child believes he/she is disabled by the pain.	35.00 (32.04)	24.31-45.68	34.The child has a depressive personality.	34.08 (28.04)*	24.86-43.30
	1.The child has an anxious personality.	34.97 (27.19)*	26.04-43-91	105. The child's parents tend to dwell on and magnify or exaggerate the threat or seriousness of their pain. They are incapable of dealing with it.	33.51 (27.93)*	24.20-42.83

*Table 5 (cont'd). The mean value of responses of all participants and standard deviations with its 95% confidence interval*

	PREDICTORS OF CHRONIC PAIN	MEAN (Sd)	C.I.	PREDICTORS OF DISABILITY	MEAN (Sd)	C.I.
RISK FACTORS	119. The child has pain to light touch (allodynia)	34.29 (22.51)*	25.56-43.02	64.The child suffers from a problem of mental health.	31.89 (27.46)*	22.87-40.92
	82.The child has frequent pain episodes.	34.00 (28.92)*	24.35-43.64	101.The child's quality of life has been impaired by the pain.	31.89 (26.21)*	23.15-40.63
	61.The parents are disabled as a result of their pain.	33.66 (28.08)*	24.43-42.89	119. The child has pain to light touch (allodynia)	31.32 (23.59)*	22.17-40.47
	23.The parents are emotionally unstable.	33.21 (27.43)*	24.19-42.23	17.The child perceives that pain interferes in his/her daily activities.	31.02 (30.61)	20.96-41.09
	73.The child has a family history of chronic pain.	32.95 (24.36)*	24.82-41.07	91.When the child's parents are in pain they believe that they are in danger and that they should avoid exercise as much as possible.	30.49 (23.59)*	22.62-38.35
	106.The child believes that pain means he is damaging him/herself and that he/she should avoid exercise.	32.84 (28.35)*	23.39-42.29	108.The child believes that his/her pain will persist over time.	29.70 (30.46)	19.54-39.86
	77.Compliance with the therapy prescribed to treat the pain is low.	32.49 (28.88)*	22.86-42.12	79.The parents believe that their child's health is vulnerable.	29.69 (24.00)*	21.57-37.82
	105. The child's parents tend to dwell on and magnify or exaggerate the threat or seriousness of their pain. They are incapable of dealing with it.	31.62 (26.46)*	22.80-40.44	23.The parents are emotionally unstable.	29.39 (29.57)	19.68-39.11
	81.The child has fewer responsibilities or chores (for example, helping with the housework) after expressing pain.	31.49 (27.48)*	22.32-40.65	11.The child's self-esteem is low.	29.37 (27.17)*	20.44-38.30
	125. The child has been abused physically, sexually and/or verbally	31.28 (24.90)*	21.63-40.94	16.The child is subject to stress in his/her immediate environment (for example: family difficulties, problems at school)	28.82 (23.81)*	20.99-36.64
64.The child suffers from a problem of mental health.	31.26 (28.23)*	21.98-40.54	125. The child has been abused physically, sexually and/or verbally.	28.21 (29.91)	16.61-39.81	
PROTECTIVE FACTORS	21.The child has positive thoughts about his/her pain (for example, everything will turn out well).	-38.16 (32.93)*	-48.98- -27.33	54.The child tries to do activities that he/she enjoys (for example, playing) in an attempt to feel better.	-37.37 (31.51)*	-27.01- -47.72-
	54.The child tries to do activities that he/she enjoys (for example, playing) in an attempt to feel better.	-32.63 (31.72)	-43.06- -22.20	116. The parents are able to identify, assess and alleviate the child's pain	-35.36 (33.77)*	-22.26- -48.45
	55.The child actively searches for a solution to his/her pain problem.	-30.37 (31.01)	-40.56- -20.17	21.The child has positive thoughts about his/her pain (for example, everything will turn out well).	-34.71 (29.06)*	-25.16- -44.26
	116. The parents are able to identify, assess and alleviate the child's pain.	-30.21 (32.67)	-42.88- -17.55	102.The child has a good concept of him/herself.	-31.03 (28.73)*	-21.45- -40.61
	102.The child has a good concept of him/herself.	-28.51 (29.46)	-38.33- -18.69	55.The child actively searches for a solution to his/her pain problem.	-30.29 (32.78)	-19.51- -41.06
	41.The child reduced unsuccessfully attempts to avoid or control pain and focus instead on participation in valued activities and the pursuit of personally relevant goals.	-25.79 (43.11)	-39.96- -11.61	41.The child reduced unsuccessfully attempts to avoid or control pain and focus instead on participation in valued activities and the pursuit of personally relevant goals.	-30.00 (40.37)	-16.73- -43.27
	90.The child's parents tend to think positively about their own pain episodes (for example, they believe that everything will turn out well).	-23.24 (27.14)	-32.29- -14.19	38.When the child feels pain, he/she paces his/her activities appropriately.	-27.24 (28.40)	-17.90- -36.57

\* The Person coefficient was under 25<sup>th</sup> percentile (25<sup>th</sup> percentile<sub>(chronic pain)</sub> = 0.91; 25<sup>th</sup> percentile<sub>(disability)</sub> = 0.97)  
 Items included in this table are those whose mean value were over 75<sup>th</sup> percentile

All the items that were identified as predictors of chronic pain and related disability by more than 75% of the participants received mean scores over 75<sup>th</sup> percentile, and all of these items except for one (i.e., item 116) had coefficients below 25<sup>th</sup> percentile. Most of the items identified by more than 75% of the participants as predictors of chronic pain received also a mean score over 75<sup>th</sup> percentile (i.e., 11 out of 15 items; 73.3%), and all of these items had coefficients of variation below 25<sup>th</sup> percentile. An identical situation is found for disability: 8 out of 9 items (88.89%) were identified by more than 75% of the participants, and received a mean score over 75<sup>th</sup>, moreover all of these had coefficients of variation below 25<sup>th</sup> percentile.

Table 6 presents those factors that appeared to be considered as the most likely determinants of persistence of pain and disability, according to the panel of participating professionals. Every item included in the table met the following criteria: (1) the item was identified as a predictive factor by more than 75% of the participants, (2) its mean score was over 75<sup>th</sup> percentile, and (3) its coefficient of variation was under 25<sup>th</sup> percentile.

*Table 6. Items considered as the most important ones to predict chronic pain and disability problems.*

	UNIT OF ANALYSIS	PREDICTOR FACTORS	Relationship with chronic pain*	ITEMS
CHRONIC PAIN	CHILD	Traits of personality	R	20.The child has a tendency to somatize
			R	34.The child has a depressive personality.
			R	1.The child has an anxious personality.
		Individual history	R	73.The child has a family history of chronic pain.
		Pain catastrophizing	R	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.
		Pain attitudes	R	108.The child believes that his/her pain will persist over time.
	PARENTS (OR FAMILY)	Characteristics of pain problem	R	42.The child's pain is constant.
		Traits of personality	R	23.The parents are emotionally unstable.
	ENVIRONMENT	Pain catastrophizing	R	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.
			R	7.The child uses the health care services excessively for his pain complaints.
		Characteristics of pain treatment	R	30.The child's consumption of medicaments to relieve the pain is not appropriate.
			R	77.Compliance with the therapy prescribed to treat the pain is low.
		Consequences to pain behaviors	R	123.The child and his/ her parents have consulted numerous doctors about the pain but no one has found anything wrong
		Stressful environment	R	68.The child receives attention and/or other privileges immediately after expressing pain.
	DISABILITY	CHILD	Traits of personality	R
R				20.The child has a tendency to somatize
Pain coping skills			R	34.The child has a depressive personality
Pain catastrophizing			R	110.The child constantly avoids activities that involve moving the part of the body that hurts through fear of experiencing more pain.
Pain attitudes			R	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.
PARENTS (OR FAMILY)		Traits of personality	R	66.The child believes he/she is disabled by the pain.
			R	106.The child believes that pain means he is damaging him/herself and that he/she should avoid exercise.
		Pain coping skills	P	117. Parental anxiety is high.
		Pain catastrophizing	R	116. The parents are able to identify, assess and alleviate the child's pain
		Pain attitudes	R	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.
		Disability	R	5.The parents believe that pain incapacitates.
			R	91.When the child's parents are in pain they believe that they are in danger and that they should avoid exercise as much as possible.
			R	61.The parents are disabled as a result of their pain.

(\*) R= Risk factor  
 P= Protective factor

Items included in this table comply with the following criteria: 75% of participants agreement on the predictive nature of the item, the predictive power value attributed to the item lies above 75<sup>th</sup> percentile, and variation coefficient lies below 25<sup>th</sup> percentile.

### **3.4. Factors predictive of chronic pain vs. factors predictive of disability**

As a secondary study objective, we studied whether items that influence the development of chronic pain are different from those that influence the disability process. Most items were considered by the participants as predictive of both chronic pain and disability (see examples in Table 2).

The proportion of participants who categorized each item as predictor of chronic pain and disability were also compared. Contrasts of proportions for independent samples showed no differences at 0.001.

In relation to this secondary objective, we also examined the predictive power that the participants had attributed to the items, using paired-t tests for independent samples to determine whether the items' mean values on the development of chronic pain and disability were different or the same. The results showed that no item had a different predictive power for chronic pain versus disability ( $p < 0.001$ ).

### **3.5. Predictive factors considered to be of a protective nature**

Another secondary objective was to determine whether any of the items in the questionnaire presented to the panel of participating professionals exerted a protective role. That is to say, were any factors thought to be related to a positive prognosis?

The results reveal that 33 items were rated as of having a positive influence; in contrast, 99 items were rated as exerting a negative function. On the other hand, and on the basis of the criteria used in this study (*i.e.*, 75% of participants agreement on the predictive nature of the item, the predictive power value attributed to the item lies above 75<sup>th</sup> percentile, and coefficient of variation lies below 25<sup>th</sup> percentile), just one item could be considered a protective factor. Namely, the factor described in item 116: "Parents are able to identify, assess, and alleviate the child's pain," was rated as being protective of both chronic pain and disability by the participants.

As a whole, there only was one protective factor considered to be important by the participants; protective factors seem to be considered as less important by the participants. Moreover, there also was a lower agreement on their predictive power. That is, the items regarded as protective factors tend to be given lower predictive power than the ones attributed as risk factors; at the same time, their coefficients of variation were larger.

## **4. Discussion**

This Delphi poll resulted in professional consensus on risk factors predictive of chronic pediatric pain and related disability after two consecutive rounds of ratings. It also provided important information about factors that may play a protective role in the development of pain and disability. Concerning the development of chronic pain, we found that the factors deemed most important by the participants could be grouped within the following classes and included (1) child's psychological characteristics: the child's tendency to somatize, depressed personality, and anxious personality; (2) parent's psychological characteristics: parental emotional instability, (3) characteristics of the pain experience: suffering from constant pain, and a family history of chronic pain; (4) characteristics of pain management: an excessive use of

health care services for the child pain complaints, an inappropriate consumption of medicines to relieve the pain, doctor shopping for the pain problem without finding anything wrong, and a low compliance with the health care professionals' recommendations, (5) psychological factors related to the child's pain experience: catastrophic thinking of the child and her or his parents about the child's pain, child's negative expectations about the course of her or his pain problems, that is, the belief that her or his pain will persist over the time, and the presence of positive reinforcements in response to the child's pain behaviors; and (6) a stressful environment.

The most important factors that participants considered to contribute to disability include: (1) child's psychological characteristics: the tendency to somatize, and a depressed personality; (2) parents' psychological characteristics: high anxiety; (3) psychological factors related to the child's pain experience: pain-related fear avoidance (child's and parents' catastrophic thinking about the child's pain, and child's avoidance behaviors due to the fear of experiencing more pain), child's beliefs about her or his being disabled by the pain, and pain modeling factors (parents' avoidance behaviors due to the fear of experiencing more pain, parents' belief that pain incapacitates, and parents own disability because of pain); and (4) parents ability to deal with her or his child pain. To the extent that the opinions expressed by the panel of professionals who participated in this study reflect reality, then these results support the hypothesis that socio-psychological factors are central in the development of chronic pain and disability problems.<sup>10, 13, 14, 17, 22</sup>

According to the panel's opinion, the factors are potentially important both in the development of chronic pain and pain related disability. However, our findings need to be interpreted with caution due to our having to balance between type I and II errors. The proportion of items identified as potential risk factors was higher than the proportion of items identified as potential protective factors for both chronic pain and disability. This difference is consistent with the literature concerning the predictors of both pain and disability.<sup>9, 17, 18, 19, 23, 24, 27, 30</sup>

The Delphi technique is a useful preliminary strategy to reach consensus among people, and this study has provided some relevant data on the factors that may lead to chronic pain and chronic pain-related disability in children, at least as identified by professionals with a specific interest and experience in chronic pain in children and adolescents. However, the study also has some important limitations. First, participants were a multidisciplinary international heterogeneous group of individuals representative of professionals interested in pediatric pain. However, most of the participants were physicians. Although this can be considered as normal, granted that pain is mostly managed by medical doctors, we could not compare among the different responder groups, and there is no way to determine if representatives from different disciplines or settings differ in their responses.

Although the number of participants was appropriate, as recommended for Delphi studies<sup>6</sup>, and participants were representative of the international community studying pediatric

pain, it could be that the statements selected by us and experts may not be an exhaustive list. However as the list that was developed was very long (110 items), and all of the participants were also allowed to add additional factors on the first round we think that the list likely represents most, if not almost all, of the potential factors that influence pain and disability in children.

There was a relatively low recruitment and attrition in the participants, as a result of the duration of the survey process and the time commitment required to assess the list of items. This could rise concerns on the generalization of the results as well as on the statistical power of the analysis. However, we believe that this issue probably does not have had any significant effect on the results of the study. First, the rule that 15-30 people are required for an adequate panel size was preserved <sup>11</sup>. Second, the profile of the participants did not change significantly between rounds when compared to those failing to complete both rounds, in terms of their gender, country of residence, professional activity or level of expertise. Third, there were not significant differences between invited professionals that did not participate and those that in fact participated (see Table 1).

In the second round no consensus was reached for a few items, when they were scored as predictors of chronicity or disability, and despite of that no additional rounds were performed. If we had attempted a third round, this would have required a significant amount of additional time and effort, and it would also likely have resulted in additional expert attrition. We therefore considered that an additional round would have produced more problems than benefits for this study, especially given the high consensus obtained for the great majority of the items. Still, if this study were to be replicated, a focus on these items, in particular, would be useful.

Finally, this study was conducted so as to explore what factors may predispose (or protect) from chronic pain and/or related disability. However, there is no question that there may be specific factors affecting differently different pain syndromes. For example, musculoskeletal pain may have different predictive factors than headache. Therefore, future studies should look into this specific factors, in relation to specific diagnosis or categories.

In summary, the results of this study are consistent with the hypothesis that the development of chronic pain and pain-related disability are the result of a set of interacting forces, both internal and external personal characteristics of the actors. A complete and detailed picture of chronic pediatric pain and related disability should consider different units (individual, dyad, and group: child, parents, and environment), and levels of analysis (physiology, emotions, cognitions, behaviors). Since the study of predictive factors of chronic pain and disability in children is in a relatively early stage of development, we have used the Delphi method as the first step in a comprehensive approach to this area. A next step is to empirically examine the factors identified in this study in relation to actual pain and disability prospectively. If the findings from this study were supported, then they could provide valuable information that could be used in the development of a screening instrument to identify children at risk of suffering chronic pain and disability, as well as in the design and implementation of treatment programs tailored to

impact (or prevent) chronic pain, disability, or both. The methodology used here, although important for identifying factors that should be more closely examined in future research, does not provide data concerning their actual influence on the development of chronic pain and/or pain related disability, since professionals' beliefs may, or may not, predict reality. What this approach provides is basic and essential information which can be used to help to design field studies that will test new hypothesis, or compare already and competing existing ones.

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Appendix A. Items which participants were requested to assess

UNIT OF ANALYSIS	FACTORS	MEASURES	SCALES	Round*	ITEMS
<b>CHILD</b>	PHYS. & BIOL. CHARACTER.			1	3.The child is growing quickly.
				1	24.The child weighs too much.
				1	63.The child's postural hygiene is bad.
				1	65.The child's body mass index is high.
				1	74.The child's pain threshold is low.
	JEWELRY CHARACTER.			1	40.The child is between 12 and 15 years of age.
				1	45.The family's socioeconomic level is low.
				1	57.The child is female.
	TRAITS OF PERSONALITY			1	1.The child has an anxious personality.
				1	9.The child does not find it easy to express his/her feelings and emotions.
				1	11.The child's self-esteem is low.
				1	20.The child has a tendency to somatize
				1	34.The child has a depressive personality.
				1	52.The child has a difficult temperament.
				1	69.The child is a perfectionist and/or demanding
				1	96.The child is shy.
				1	102.The child has a good concept of him/herself.
	JUVENILE CHARACTER.			2	118. The child has disabilities, cognitive impairment, or communication limitations.
	STATE OF WELL-BEING			1	64.The child suffers from a problem of mental health.
				1	75.The child has a sleep disorder.
				1	76.The child has a chronic disease that is not related to the pain experience
				1	79.The parents believe that their child's health is vulnerable.
				1	88.The child is in good physical shape.
				1	98.The child has behavioral difficulties (for example, hyperactivity, disruptive behavior or hidden aggression)
	INDIVIDUAL HISTORY			1	12.The child has had previous physical injuries in the zone that is painful.
				1	31.The child has had previous experience of recurrent episodes of acute pain.
				1	36.The child was exposed, in a prolonged or repetitive way, to nociceptive stimuli in the first stage of life.
				1	73.The child has a family history of chronic pain.
				1	87.The child has a history of treatment and medication for pain.
				2	111. The child has previous untreated pain.
				2	112. The child has a prior inflammation.
			2	125. The child has been abused physically, sexually and/or verbally.	
CHARACT. OF THE PAIN PROBLEM			1	18.The child has a medical diagnosis associated with his/her pain experience	
			1	42.The child's pain is constant.	
			1	47.The child's pain lasts for a long time (do not respond to this item in the chronic pain section).	
			1	82.The child has frequent pain episodes.	
			1	84.When it first appears, the child's pain is very intense.	
			2	119. The child has pain to light touch (allodynia)	
PAIN COPING SKILLS	PCQ <sup>26)</sup>	Externalizing	1	15.The child externalizes his/her feelings when in pain in an improper way (for example, he/she gets angry or shouts)	
		Positive self-statements	1	21.The child has positive thoughts about his/her pain (for example, everything will turn out well).	
		Information seeking	1	32.The child asks others for information about the pain he/she is experiencing.	
		Cognitive distraction	1	46.The child tries to ignore his/her pain.	
		Behavioral distraction	1	54.The child tries to do activities that he/she enjoys (for example, playing) in an attempt to feel better.	
		Problem solving	1	55.The child actively searches for a solution to his/her pain problem.	
		Seeking social support	1	62.The child seeks social support by talking to others (parents or friends) about his/her feelings.	
		Pace	1	38.When the child feels pain, he/she paces his/her activities appropriately.	
		Avoidance	1	110.The child constantly avoids activities that involve moving the part of the body that hurts through fear of experiencing more pain.	

Appendix A (cont'd). Items which participants were requested to assess

UNIT OF ANALYSIS	FACTORS	MEASURES	SCALES	Round*	ITEMS				
PAIN CATALAN TROPICIZING	PCS-C <sup>1</sup>			1	71.The child tends to dwell on the pain and magnify or exaggerate its threat or seriousness. He/she is incapable of dealing with it.				
				PAIN ATTITUDES	SOPA <sup>8</sup>	Solicitude	1	10.The child believes that others, and particularly his/her parents, should be sensitive to his/her pain experience.	
						Emotion	1	37.The child believes that his/her emotions affect his pain experience.	
						Medical cure	1	39.The child believes that his/her pain problems can be cured medically.	
						Medication	1	44.The child believes that medication is an appropriate treatment for chronic pain.	
						Control	1	53.The child believes he can control his/her pain.	
						Disability	1	66.The child believes he/she is disabled by the pain.	
						Harm	1	106.The child believes that pain means he is damaging him/herself and that he/she should avoid exercise.	
								1	108.The child believes that his/her pain will persist over time.
						2	121. Child and parents think healthcare professionals don't believe his/her pain		
						2	122. The child and/or his/her parents believe that medication alone can cure the pain		
				EMOTIONAL REACTIONS OR EMOTIONAL DISTRESS	Emotional Distress Checklist <sup>25</sup>		1	72.The child is quiet or calm when he/she is in pain.	
							1	83. The child is happy when he/she is in pain (he/she not getting attention/secondary gain).	
							1	86.The child is scared when he/she is hurt.	
							1	94.The child is sad when he/she is in pain.	
	1	107.The child is angry when he is in pain.							
	2	127. The child is worried when he/she is in pain.							
	1	58.The child is frustrated when he/she is in pain.							
	1	80.The child feels agitated when he/she is in pain.							
ACCEPTANCE OF PAIN				1	41.The child reduced unsuccessfully attempts to avoid or control pain and focus instead on participation in valued activities and the pursuit of personally relevant goals.				
				1	2.The child often rides on a motorcycle or quad bike.				
				1	29.The child usually drinks alcohol.				
				1	43.The child carries a heavy school satchel (excessive load).				
				1	48.The child usually smokes.				
					50.The child has a sedentary life style (for example, he/she spends many hours in front of the television or computers, playing computer games, doing homework, etc.).				
				1	51.The child does a considerable amount of physical exercise and/or is heavily involved in physically-demanding activities (for example, ballet).				
					59.The child's eating habits are not healthy (irregular and not well balanced).				
					67.The child works during the school year in white-collar jobs (office work and sales).				
					70.The child spends a considerable amount of time playing a musical instrument (for example, the piano or violin).				
					95.The child sleeps little.				
					17.The child perceives that pain interferes in his/her daily activities.				
					101.The child's quality of life has been impaired by the pain.				
				PARENTS (OR FAMILY)					124. The parents are divorced.
								PERSONALITY	
	100.The child's parents are pessimistic.								
	117. Parental anxiety is high.								
STATE OF WELL-BEING		78. The parents are physically healthy.							
	PAIN COPING SKILLS	CPCI <sup>7</sup>	Guarding					1	8.When the parents are in pain they usually restrict the use and the movement of the part of the body that hurts.
Resting			1					14.The parents engage in a resting activity in response to pain: for example: lying down, sitting down, slowing down, or going to a dark or quiet room).	
Seeking social support			1					19.The parents usually seek social support when they are in pain.	
			1					26.The parents usually take medication when they are in pain.	
Relaxation			1					33.The parents tend to engage in a specific relaxation task when they are in pain.	
Exercise/Stretching			1	35.When they are in pain, the parents tend to do stretching exercises and/or physical exercise.					
persistence	1	85.The parents usually ignore pain when there is something they have to do.							

	Coping self-statements	1	90.The child's parents tend to think positively about their own pain episodes (for example, they believe that everything will turn out well).
		1	97.When the child's parents are in pain, they pace their activities appropriately.
		2	114. The siblings do not successfully cope with their pain problems.
		2	116. The parents are able to identify, assess and alleviate the child's pain
PAIN CATASTROPHIZING	PCS <sup>2*</sup>	1	105. The child's parents tend to dwell on and magnify or exaggerate the threat or seriousness of their pain. They are incapable of dealing with it.
		1	109.The parents tend to dwell on and magnify or exaggerate the threat or seriousness of their child's pain. They are incapable of dealing with the child's pain.
PAIN ATTITUDES	Disability	1	5.The parents believe that pain incapacitates.
	Medication	1	22.The parents think that medication is an appropriate treatment for chronic pain.
	Medical cure	1	27.The parents believe that problems of pain can be cured medically.
	Harm	1	91.When the child's parents are in pain they believe that they are in danger and that they should avoid exercise as much as possible.
	Pain Control	1	93.The child's parents believe that they can control their pain.
	Solicitude	1	99.The child's parents believe that others, particularly the family, should be sensitive to his/her complaints of pain.
	Emotion	1	104.The child's parents believe that his/her emotions affect his/her pain experience.
EMOTIONAL REACTIONS			25.The parents feel bad (for example, guilty) about the child's pain.
ACCEPTANCE OF PAIN			4.The parents reduced unsuccessfully attempts to avoid or control the child's pain and focus instead on encouraging his/her son to participate in valued activities and pursuit of relevant goals.
		1	92.The parents reduced unsuccessfully attempts to avoid or control their own pain, if they feel any, and focus instead on participation in valued activities and the pursuit of personally relevant goals.
DISABILITY		1	56.The child's pain interferes in family life.
			61.The parents are disabled as a result of their pain.
ENVIRONMENT	CHARACTERISTICS OF THE PAIN TREATMENT	1	7.The child uses the health care services excessively for his pain complaints.
		1	30.The child's consumption of medicaments to relieve the pain is not appropriate.
		1	49.The child has already been treated for his/her pain.
			77.Compliance with the therapy prescribed to treat the pain is low.
		2	115. The instructions provided by the health care professionals to the child and/or his/her parents are not sufficient.
		2	120. The parents talk for the child even when the child is directly addressed
CONSEQUENCES TO PAIN BEHAVIORS	Positive punishment	1	13.The pain prevents the child from doing and/or receiving what he/she likes (for example, he/she has to stay at home, and cannot go to play in the park).
	Negative punishment	1	28.The child is reprimanded or provokes some other negative response when he/she expresses his/her pain.
	Positive reinforcement	1	68.The child receives attention and/or other privileges immediately after expressing pain.
	reinforcement	1	81.The child has fewer responsibilities or chores (for example, helping with the housework) after expressing pain.
ESSFUL ENVIRONMENT		1	16.The child is subject to stress in his/her immediate environment (for example: family difficulties, problems at school)
CIAL SUPPORT		1	89.The child perceives little social support.
PAIN ATTITUDES			103.Society (health care professionals and the general population) believes that young people do not experience chronic pain.
OTHERS		1	60.The ergonomics of the school furniture (tables and chairs) are not good.
		2	113. Peers/schoolmates do not successfully cope with their pain problems 126. The child and his/her family know other cases who have suffered or are suffering from similar problems.

(\*) 1= Included in the first list of items (factors suggested by the literature and experts).  
 2= Suggested by participants



**Paper #6:**

**Huguet A, Eccleston C, Miró J, Gauntlett-Gilbert J.  
(Submitted for publication). The influence of pain and  
pain related factors on the functioning of 8-16 year old  
children. *Pain***



## THE INFLUENCE OF PAIN AND PAIN RELATED FACTORS ON THE FUNCTIONING OF 8-16 YEAR OLD CHILDREN

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### ABSTRACT

Pain is a common everyday occurrence. However, impaired functioning associated with pain is a much less common event. Relationships between biological, social and psychological variables have rarely been investigated in samples of children reporting pain in non-clinical settings. The aim of this study was to identify the predictors of functioning in a normal school-attending population of children, who report pain as a common experience. 561 schoolchildren aged between 8 and 16 years and their parents participated in a cross-sectional interview and questionnaire study. Data were collected on biological, social and psychological variables. Child functioning was measured in four ways: self-reported quality of life, school attendance, engagement in out of school sporting activity, and engagement in out of school sedentary activity. Psychological variables included measures of child coping with pain, including the extent of beliefs about pain, and catastrophic reaction to pain, and parental attention to children's report of pain. Results showed a variable pattern depending on the specific measure of functioning. In general age and socioeconomic status were related to greater function. Less affective response to pain was associated with greater functioning. Parental behaviour had little effect on measures of function. These results are discussed within a biopsychosocial model of pain-related behaviour. In particular, it is proposed that emotional responding to everyday pain may be an important predictor of function. More study on the relationship between pain and their engagement in active or sedentary behaviour in children is needed.

**Keywords:** schoolchildren, prognostic factors, functioning, severity of pain, psychosocial characteristics

## **1. Introduction**

Pain is a common but usually benign childhood experience. Some children, however, develop disabling chronic pain that severely diminishes the quality of their lives and the lives of their family members (*e.g.*, Smedbraten et al., 1998, Perquin et al., 2000, Hunfeld et al., 2001, Roth-Isigkeit et al., 2004). The typical clinical presentation of a young person with chronic pain is often challenging and can be characterised by the presence of a complex interrelationship of both physical and psychosocial factors (Williamson et al. 2002, Merlijn et al., 2003, Eccleston et al., 2004).

Empirical studies have identified factors that are candidates for explaining why for some children pain interferes with daily functioning whilst for others there is no effect of pain. Key variables examined are different characteristics of pain such as intensity, duration, or frequency have been proposed to be playing a role (*e.g.*, Kashikar-Zuck et al., 2001, Peterson and Palermo, 2004, Logan and Scharff, 2005, Roth-Isigkeit et al., 2005, Merlijn et al., 2006, Logan et al., 2006, Lynch et al., 2006). Similarly, coping styles (Sawyer et al., 2004, 2005, Merlijn et al., 2006), catastrophic thinking (Vervoort et al., 2005, Lynch et al., 2006), and environmental contingencies (Craig and Riddell, 2003, Merlijn et al., 2006; Van Slyke and Walker 2006) have also received some attention and support.

Despite these advances, there exists an important gap in the literature about the obverse and more common situation of children who function well despite pain. We know very little about children suffering from pain who live a “normal” life, and especially about those factors that could predict a good prognosis for pain problems. The study of the normal case, of children’s everyday functioning, and of children who do not become disabled by pain, may offer insight into the poor adaptation to persistent and recurrent pain commonly encountered in clinic.

In this paper we focus on the ability to function despite pain. The aim of this study was to identify what predicts functioning in a normal school- attending population of children, who report pain as a common experience. Functioning was measured in terms of the self-report of quality of life, school attendance, level of engagement in physical activities, and amount of time spent on sedentary activities. Based on previous research (Vervoort et al., 2005, Walker et al., 2005, Lynch et al., 2006, Merlijn et al., 2006), we hypothesised that the greater severity of pain would be associated with less adequate functioning, and psychosocial components of pain would independently influence functioning regardless of pain intensity or frequency.

## **2. Method**

### **2.1. Participants**

902 school-children were recruited from non-special primary and secondary schools (from grade 2 to 8) in Tarragona, a province of Catalonia (Spain). Children were excluded if they were (1) suffering from any cognitive impairment (four children), or (2) unable to understand Catalan (none). 561 children participated (62%). The primary reason for non-participation was lack of parental assent. There were no significant differences in either school grade or sex between those participating and not participating. Child age ranged from 8 to 16 years (mean

age=10.9; SD=2.0). 51.7% of the sample was male. Table 1 gives full demographic and clinical data of the children and parents.

## **2.2. Procedure**

Following governmental and school ethical approval 40 classes of 20 primary and secondary schools were randomly selected using a stratified sampling procedure from all eligible non-special education schools in Tarragona. All invited schools agreed to participate, except for one who gave current involvement in other research as the reason. Participants were asked to provide information about their daily activity and about their common pain experiences. Individual interviews took place at the school and lasted approximately 40 minutes. Because the interview was concerned with past experience, memory enhancing techniques were incorporated (Orbach and Lamb, 2000). Several other quality control measures were implemented during data collection. First, all interviewers attended standardisation classes in which they were trained in interview technique. Second, data sheets were checked immediately after the interviews, and monitored centrally for missing data or inconsistencies.

Parents were surveyed separately. They were instructed to complete written questionnaires at home. These included questions about their child's health and about their own reactions to the child's pain behavior.

## **2.3. Measures**

### *2.3.1. Child measures*

#### *2.3.1.1. Socio-demographic measures*

Information about age, sex, number of siblings, and number of people living at home was collected at the time of interview. Children's SES was rated on the basis of the work by Domingo-Salvany et al. (2000), who support a method for measuring socio-economic status based upon the National Classification of Occupations in Spain CNO-94. Since both parents were requested to provide their current occupation, the "highest" occupation of the two parents was the one used to rate the SES of the family and the child.

#### *2.3.1.2. Anthropometric measures*

Child weight and height were measured post interview.

#### *2.3.1.3. Pain*

The highest pain intensity and usual pain intensity in the last 3 months was measured using the Catalan version of the *Faces Pain Scale – Revised* (FPS-R) (Miró and Huguet, 2004). Pain frequency was measured on a categorical scale following Perquin et al., (2000) and pain duration was recorded in days, weeks and months. Pain location was coded according to axis I of IASP Classification of Chronic Pain (Merskey and Bogduk, 1994). Following Mikkelsen et al., (1997) an open question was used about pain location, and a pain mannequin drawing included (Goodman et al., 1997).

#### *2.3.1.4. Quality of life*

The Paediatric Quality of Life Inventory™ Version 4.0 (PedsQL, Varni et al., 2001) was used to assess children's perception of their quality of life. The PedsQL is a generic instrument that encompasses four multidimensional scales (physical, emotional, social and school functioning) and three summary scales (total, physical health summary, psychosocial health summary). Developmentally appropriate versions (i.e., ages 8–12 and 13–18) were used. The child is asked how much of a problem each item has been in the past month. Items were scored on a 5-point Likert scale, from 0 (never a problem) to 4 (almost always a problem). Items are reverse-scored and linearly transformed to a 0-100 scale (0=100, 1=75, 2=50, 3=25, 4=0), so that higher scores indicate better quality of life. The feasibility, reliability and validity of the instrument has been demonstrated both in clinical and school population (Varni et al., 2001, 2006). Its psychometric properties have been examined in Catalan-speaking school-children population by Huguet and Miró (in press)

#### *2.3.1.5. School attendance*

Days of school absence due to the child's health problems in the month preceding the interview were reported by parents.

#### *2.3.1.6. Physical activity*

Child self-report data were collected on usual amount of time engaged in sports and other physical exercise (out of school) weekly, and usual amount of time spent on sedentary activities (specifically: watching television, using the computer and playing videogames), weekly.

#### *2.3.1.7. Functional disability*

The Functional Disability Inventory (FDI: Walker and Greene, 1991) was employed. This measure assesses 15 activities in a variety of contexts including school, home, recreation, and social interaction. The child was asked how much difficulty she/he had doing these activities because of her/his pain, using a 5-point severity scale ranging from 0 (no trouble) to 4 (impossible). Higher total scores indicate greater functional disability. Claar and Walker (2006) and Walker and Greene (1991) have reported appropriate psychometric properties for the instrument both in clinical and healthy samples.

#### *2.3.1.8. Pain Catastrophizing*

The child version of the Pain Catastrophizing Scale (PCS-C: Crombez et al., 2003) is a 13-item instrument that assesses the child's catastrophic thinking about pain. The child rates the extent to which she/he endorses particular catastrophic thoughts on a 5-point Likert scale ranging from 0 (not at all) to 4 (extremely). The higher the total PCS-C score the more the child engages in catastrophic thinking about pain. The questionnaire has acceptable reliability and validity in both a community and clinical pain sample of children (Crombez et al., 2003; Miró et al., 2006).

### *2.3.1.9. Pain beliefs*

The child version of the Survey of Pain Attitudes (SOPA-C, Engel et al., 2007) was used. This measure was developed from the original scale used to assess adult pain attitudes (see Jensen et al., 1994). The survey includes twenty-nine true-false attitude statements related to pain which underlie seven attitudes: (a) belief in a medical cure for pain (Medical Cure scale), (b) belief in one's ability to control pain (Pain Control scale), (c) belief that it is the responsibility of others to assist people who are experiencing pain (Solicitude scale), (d) belief that one is disabled because of pain (Disability scale), (e) belief that the medications are the best treatment strategy for pain (Medication scale), (f) belief that pain means something is being hurt in the body and consequently, that exercise should be avoided (Harm scale), and (g) belief that emotion affects pain (Emotion scale). The child is asked how much she/he agrees with each attitudinal statement on a 3-point Likert scale (0 = I do not agree with this, 1 = I am not sure, 2 = I agree with this). To obtain a score for each SOPA-C subscale, which may be used as a measure of overall agreement with the belief construct, ratings of the items within each subscale need to be averaged after reversing the score of a few items. The instrument has shown appropriate psychometric properties when used in children with chronic pain and disabilities (see Engel et al., 2007). The Catalan version of the SOPA-C validated by Miró et al (2007) was used.

### *2.3.1.10. Pain coping strategies*

The children's pain coping strategies were assessed with the Pain Coping Questionnaire (PCQ, Reid et al., 1998). The PCQ has 35 items that measure seven coping strategies that the child might use to cope with or manage pain. The strategies include 'approach' coping strategies (information seeking, problem solving, seeking social support, positive self-statements), 'problem-focused' avoidance strategies (behavioural distraction and cognitive distraction) and 'emotion-focused' avoidance strategies (externalizing and internalizing/catastrophizing). On a 5-point Likert scale from 1 (never) to 5 (very often) the child rates how often each of these coping strategies is used when in pain. The score for each PCQ subscale is calculated by summing the ratings given for each item within each scale, and dividing by the number of items in that scale. The reliability and validity of the PCQ in healthy school children has been documented by Reid et al. (1998). The questionnaire has shown good psychometric properties when used with Catalan speaking school-children (see Huguet et al., 2006).

#### *2.3.1.11. Affect associated to pain*

The child version of the Positive and Negative Affect Schedule (PANAS-C) is a standardised measure which consists of 20 adjectives, ten assessing positive affect (e.g., excited) and ten assessing negative affect (e.g., upset). An adapted version was used here (Huguet et al., 2007). Children were asked to respond on a five point scale, from 1 which means “not at all or almost not at all” to 5 “a lot” the extent to which each adjective describes their feelings when they are in pain, as opposed to describing their feelings in the last few days. The measure has shown good psychometric properties in a sample of Spanish children (Sandin et al., 2003).

#### *2.3.2. Parent measures*

##### *2.3.2.1. Socio-demographic measures*

Age, sex, marital status, and occupation status were all recorded.

##### *2.3.2.2. Parental response to child's pain behaviors*

Parents reported their reactions to their child's pain behaviors. For this, we used the IRPEDNA inventory (Huguet et al., in press). IRPEDNA is a self-administered questionnaire that has three subscales: 1) solicitousness, 2) discouragement of pain behaviors, and 3) promotion of well-behaviors and coping. Parents report the frequency of behaviour in response to child pain using a five-point scale (0=never, 1=hardly ever, 2=sometimes, 3=often, 4=always). This instrument has been shown to have adequate reliability and validity among parents of Catalan school children (Huguet et al., in press).

##### *2.2.2.3. Impact of the child's health status on their parents' daily routine*

Parents were also asked how much their child's health status had interfered with their own daily activities in the preceding month. An 11-point numerical scale was used, with the anchors: 0 = It does not interfere at all; 10 = It interferes a lot).

#### **2.4. Data analyses**

Data were analysed using the SPSS Statistical Package for Windows 12.0. Missing data resulted in exclusion of cases from some analyses.

The focus of this investigation was child functioning. Four related concepts of function were identified as the primary dependent variables: 1. quality of life, 2. school attendance, 3. level of engagement in sport or physical exercise, and 4. the average time spent on sedentary activities weekly. The relationship of both pain and related psychosocial variables to each functioning variable was the primary focus of the investigation.

Data were first cleaned. Square root transformations were used for the normalization of the PedsQL scores and the average time spent on sedentary activities (Tabachnik and Fidell, 2001). Because of this normal sample distribution of the school attendance variable was highly negatively skewed so a dichotomous variable was calculated (i.e., Complete school attendance

vs Incomplete school attendance) (MacCallum et al., 2002). Where variable distributions were bimodal dichotomous variables were calculated using natural division (e.g., Level of engagement in sport or physical exercise into: None or almost no involvement vs Involvement) (Tabachnik and Fidell, 2001). Second, associations between variables were explored. Third, the putative effects of pain and related psychological variables on the four concepts of function were tested in a series of separate stepwise hierarchical regression analyses. Two separate hierarchical regression analyses were performed for each dependent variable. Child and parent predictors were analysed in separate equations. In the 'child' equations, variables that correlated with the outcome were entered in blocks. Entered first were descriptive variables (specifically, socio-demographic and anthropometric variables). Entered second were variables related to the severity of the child's pain problems. There is no available classification system to grade a child's pain status, therefore, following the adult example (Von Korff et al., 1990) we measured pain status in terms of the self-report of intensity (the highest and the most common pain intensity in the last 3 months), the persistence of pain (presence of chronicity and recurrence), and pain-related disability (FDI). Entered in a final step were variables related to behavioral, cognitive and affective components of pain (specifically, pain coping skills, pain attitudes, pain catastrophic thinking, and pain affect). Scores on the Internalizing/catastrophizing subscale of the PCQ were not used in the regression analyses to avoid an overlap with the PCS-C scores.

In this study chronic pain was defined as pain of three months or greater duration, experienced either continuously or recurrently. 'Recurrent' was defined as one or two pain episodes at least each month (see Mikkelsen et al., 1997 for similar protocol).

In the second set of analyses on parent variables, a similar stepped approach was used. Descriptive variables and variables related to the severity of pain were entered into the first two blocks, following the same procedure detailed previously. Subsequently, the IRPEDNA scales were entered as a separate block to determine the additional variance accounted for by parents' reactions to their children's pain behaviors given that they correlated with the outcome.

Following each regression analysis, the assumptions of the model were checked. The patterns shown in the plot standardized residuals against standardized predictive values were checked for the assumption of linearity and homocedasticity. Durbin-Watson statistics were used to support the assumption of independent residuals. To exclude the influence of multicollinearity the VIF value for every independent variable was calculated. Moreover, the outliers were also checked in order to be sure that they did not unduly bias the results of the regression equations.

### 3. Results

#### 3.1. Descriptive statistics

Table 1 shows the descriptive statistics for both children and parents.

*Table 1. Characteristics of the study population*

	Scale range	Total sample mean (SD)
<b>Children characteristics</b>		
1. Socio-demographic variables		
Age (years)	8-16	10.89 (2.00)
Sex (n,%)		
Boys	--	51.7 (290)
Girls	--	48.3 (271)
SES (n, %)		
High SES	--	34 (191)
Medium SES	--	27.5 (154)
Low SES	--	23 (129)
2. Anthropometric variables		
BMI	--	19.24 (3.72)
3. Pain severity variables		
The highest pain intensity in the last three months (of the most troublesome pain)	0-10	5.31 (3.23)
The most common intensity in the last three months (of the most troublesome pain)	0-10	3.41 (2.38)
Functional disability	0-52	4.92 (0.27)
Number of pain episodes (in the last three months)		8.87 (17.62)
Presence of chronic pain (n,%)	--	37.3 (209)
Pain catastrophizing	0-52	21.39 (9.16)
Pain coping strategies		
Information seeking	1-5	2.34 (0.85)
Problem solution	1-5	2.84 (0.93)
Seeking social support	1-5	2.96 (1.03)
Positive self-statements	1-5	2.85 (1.05)
Behavior distraction	1-5	3.21 (1.07)
Cognitive distraction	1-5	3.13 (0.99)
Externalizing	1-5	1.64 (0.76)
Internalizing/ Catastrophizing	1-5	2.07 (0.77)
Pain beliefs		
Control	0-2	1.46 (0.50)
Disability	0-2	0.98 (0.75)
Harm	0-2	1.13 (0.57)
Emotion	0-2	0.92 (0.74)
Medication	0-2	1.84 (0.45)
Solicitude	0-2	1.69 (0.52)
Medical Cure	0-2	1.73 (0.40)
Affect related to pain		
Positive affect	0-50	21.46 (6.02)
Negative affect	0-50	16.75 (4.79)

Table 1 (cont'd). Characteristics of the study population

	Scale range	Total sample mean (SD)
<b>Children characteristics</b>		
5. Functioning outcomes		
Quality of life	0-100	81.31 (10.52)
Number of days missing school in the last month	0-20	0.83 (1.95)
Average amount of time spent participating physical activities (minutes)	--	162.46 (151.78)
Average amount of time spent on sedentary activities (minutes)	--	1182.62 (623.40)
<b>Parents' characteristics</b>		
1. Socio-demographic variables		
Age		40.61 (4.30)
Marital status (n,%)		
Married	--	86.3 (484)
Separated or divorced	--	12.4 (70)
Widow/er	--	1.3 (7)
2. Parents' informing about the child's pain		
Parents' reactions to the child's pain behaviours		
Solicitousness	0-4	2.36 (0.67)
Discouragement	0-4	0.91 (0.64)
Promotion of well- behaviors and coping	0-4	2.57 (0.68)

### 3.1.1. Child Pain.

Regarding pain characteristics, all participants reported a mean of 11.40 pain episodes in the last three months (SD=18) and a median of 5. Lower limb pain was reported as the most troublesome pain complaint. The highest intensity of the most troublesome pain over the last three months was 5.31 (SD=3.23, range 0-10). The mean intensity of the most troublesome pain over the last three months was 3.41 (SD=2.38, range 0-10). According to the criteria used in this study to define chronic pain, 37.3% of children and adolescents could be so defined. Almost a third of those (29%) reported pain in multiple body regions. Compared with clinical populations who have completed the FDI, this sample was, as expected, less disabled (e.g. Walker and Greene, 1991, Kashikar-Zuck et al., 2002) (see Huguet and Miró, submitted, for more details).

### 3.1.2. Child Functioning.

1. *Quality of Life*. Participants reported a high level of quality of life (mean total PedsQL score = 81.31, SD = 10.52) which compares favourably to those reported in previous studies (e.g., Varni et al. 2006; Connelly et al 2006). 2. *School attendance*. Most of the participants (74.4%) attended school every day in the month previous to the assessment (number of days: 20). The mean number of days missed from school during that month due to any illness was

0.83 (SD=1.95), and the median and mode were 0. 3. *Engagement in sport or physical exercise.* 32.8% of the participants did not play any sport out of school schedule, 53.9% played one sport, and 13.3% played more than one. The reported mean of time spent weekly doing physical exercise or sport was 2 hours and 43 minutes. Soccer and basketball were by far the most common sports (15.9% and 13.5%, respectively). 4. *Sedentary behavior.* Children spent a mean of 17 hours and 42 minutes weekly on the following sedentary activities: watching TV, using the computer and playing videogames.

### 3.2. Associations with functional outcome

Table 2. Correlations between functioning variables

	Quality of life	Number of days missing school	Time spent on physical activities	Time spent on sedentary activities
Quality of life	--	-0.121*	0.147**	-0.089*
Number of days missing school		--	0.004	-0.032
Time spent on physical activities			--	-0.034
Time spent on sedentary activities				--

\* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$

Table 3. Relationship between functioning variables and socio-demographic, anthropometric, pain severity and psychosocial variables.

	Quality of life		School attendance		Time spent participating physical activities		Time spent on sedentary activities	
	R	F	t	$\chi^2$	t	$\chi^2$	r	F
<b>Children characteristics</b>								
1. Socio-demographic variables								
Age	0.047		-0.700		2.376*		0.338**	
Sex		-0.791		0.259		24.838***		0.004
SES		5.273**		3.358		18.530***		5.746**
2. Anthropometric variables								
BMI	0.026		0.372		0.546		0.166***	
3. Pain severity variables								
The highest intensity	0.391***		3.015**		-1.551		0.069	
The most common intensity	0.331***		2.751**		-0.431		0.011	
Number of pain episodes	0.279***		1.763		-0.127		0.129**	
Presence of chronic pain		-5.221***		0.491		0.940		6.889**
Functional disability associated to pain	0.383***		2.709**		-1.061		0.096*	

Table 3 (cont'd). Relationship between functioning variables and socio-demographic, anthropometric, pain severity and psychosocial variables.

	Quality of life		School attendance		Time spent participating physical activities		Time spent on sedentary activities	
	R	F	t	$\chi^2$	t	$\chi^2$	r	F
<b>Children characteristics</b>								
4. Individual psychological variables related to pain experience								
Pain catastrophizing	0.419***		1.970*		2.449*		0.090*	
Positive affectivity associated to pain	-0.092*		-0.870		-1.053		-0.032	
Negative affectivity associated to pain	0.439***		0.863		1.818		0.178***	
Information seeking	0.143***		1.591		-0.941		0.035	
Problem solution	0.058		-0.892		-1.425		-0.005	
Seeking social support	0.054		-0.168		-1.392		0.036	
Positive self-statements	-0.051		-0.226		-1.686		0.002	
Behaviour distraction	-0.020		-0.714		-0.945		0.081	
Cognitive distraction	-0.096*		-0.112		-2.373*		-0.022	
Externalizing	0.390***		-1.535		0.878		0.177**	
Internalizing/ Catastrophizing	0.450***		0.858		2.098*		0.104*	
Control	-0.189***		0.323		-2.705**		-0.100*	
Disability	0.130**		1.766		-0.977		-0.009	
Harm	-0.003		1.401		1.044		0.021	
Emotion	0.164***		1.127		0.640		0.026	
Medication	-0.055		-0.856		0.185		-0.092*	
Solicitude	0.035		0.882		-1.602		0.003	
Medical Cure	-0.052		0.233		-0.525		-0.057	
<b>Parents' characteristics</b>								
1. Parents' reactions to child's pain behaviors								
Solicitousness	0.086		2.011*		2.331*		-0.117*	
Discouragement	0.049		-0.162		-0.245		-0.091	
Promotion of well-behaviors and coping	0.031		0.626		1.173		-0.036	

\* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$

### 3.2.1. Quality of life

To determine the relationship between quality of life and potential predictors, bivariate analyses were performed. As shown in Table 3<sup>3</sup>, SES was the only descriptive characteristic that exerted some influence on quality of life ( $F=5.27$ ,  $p < 0.01$ ). All pain variables that define severity of pain problems were significantly related with the children's quality of life: pain intensity (both the highest and the most common pain intensity), number of pain episodes and

<sup>3</sup> These results need to be interpreted taking into account that a reflected square root transformation was used for reducing negative skewness of the PedQL scores. Thus, all directions of correlations are reversed.

functional disability were negatively correlated with quality of life ( $r=0.39, 0.33, 0.28$  and  $0.38, p<0.001$ , respectively), as was the presence of chronic pain ( $F=-5.22, p<0.001$ ). Quality of life (PedsQL) also correlated with psychological variables related to pain. Whereas quality of life was closely related with emotion-focused avoidance coping strategies (i.e., externalizing  $r=0.39, p<0.001$ ; and internalizing / catastrophizing  $r=0.45, p<0.001$ ), the relations with approach and problem-focused avoidance coping strategies were absent or weak (i.e., information seeking  $r=0.14, p<0.001$  and cognitive distraction  $r=-0.10, p<0.05$ ). Quality of life also correlated with emotional reaction to pain experience in the direction expected. Positive emotional reaction accounted for less than 1% of the variance in quality of life ( $r=-0.09, p<0.05; R^2=0.008$ ), but negative emotional reaction accounted for 19.27% of the variance ( $r=0.44, p<0.001; R^2=0.19$ ). Quality of life correlated positively with control beliefs ( $r=-0.19, p<0.001$ ) but negatively with disability ( $r=0.13, p<0.01$ ) and emotional beliefs ( $r=0.16, p<0.001$ ). The relationship between type of parental response to the child's pain behavior and quality of life was not significant.

To identify those factors exerting an independent influence on quality of life, a multiple regression analysis was conducted. Descriptive variables were not related to quality of life; however, based on data from studies by Williams et al. (2005) and von Rueden et al. (2006) showing the high relationship between socioeconomic status (SES) and the body mass index (BMI), we decided to force together into the model the SES and BMI information in the first block. After SES and BMI had entered into the model, variables related to pain severity which were related to quality of life were added as a third block to evaluate the association between quality of life and pain severity while controlling for descriptive variables. Finally, those psychological variables that were related to quality of life were added as a fourth block to evaluate the association between psychological variables related to pain while controlling for descriptive and pain characteristics. Table 4 presents the outputs of the regression analyses predicting change in child's quality of life. The results showed that anthropometric and socio-demographic variables only explained 1% of the variation in child's quality of life. Variables in block number two accounted for a much more proportion of variation ( $R^2=0.24, p<0.05$ ). Among variables related to the severity of pain, pain intensity, level of disability associated to pain, and the number of pain episodes were those variables that significantly improved the ability of the model to predict quality of life ( $\Delta R^2=0.16$  and  $0.05, p<0.001; \Delta R^2=0.02, p<0.01$ , respectively). As the highest pain intensity increased, quality of life decreased ( $\beta=0.17, p<0.001$ ). Similarly, as disability associated with the pain increased, quality of life decreased ( $\beta=0.19, p<0.001$ ), and as the number of pain episodes increased, quality of life decreased ( $\beta=0.13, p<0.01$ ). When the psychological variables related to pain were included, the proportion of variance explained increased to 36% ( $p<0.05$ ). Externalizing pain, catastrophic thinking about pain, and negative affectivity associated with pain were significant predictors of lower quality of life ( $\beta=0.19, p<0.001; \beta=0.14, p<0.01$ ; and  $\beta=0.14, p<0.05$ , respectively).

Table 4. Predictors of children's quality of life

Block	Predictor variable	$R^2$	Adj. $R^2$	$\Delta R^2$	$\Delta F$	$\beta$	
Child's predictors	1	BMI	0.000	0.000	0.000	0.115	-0.062
		SES	0.018	0.010	0.017	3.620*	
		Low SES vs High SES					-0.095*
		Low SES vs Medium SES					-0.005
	2	The highest pain intensity	0.175	0.167	0.158	78.474***	0.165***
		Disability associated to pain	0.226	0.216	0.051	26.865***	0.186***
		Number of pain episodes	0.246	0.235	0.021	11.137**	0.127**
	3	Externalizing	0.329	0.318	0.083	50.361***	0.194***
		Pain catastrophizing	0.362	0.350	0.033	21.303***	0.138**
		Negative affect	0.372	0.358	0.010	6.315*	0.136*

$N_{(\text{child's predictors})}=419$ , \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

### 3.2.2. School attendance

T-student and chi-square tests were used to estimate the associations between school attendance and the variables being tested. The results are presented in Table 3. No socio-demographic factors were associated with school attendance. As predicted, pain severity was related to school attendance. Specifically, those children who have missed school during the previous month scored higher on pain intensity (both the highest and the most common pain intensity) and functional disability ( $t=3.02$ ,  $2.75$ , and  $2.71$ ,  $p<0.01$ , respectively). However, neither the presence of chronic pain nor the number of pain episodes were associated with school attendance. Among all the psychological variables related to pain only the score on the Pain Catastrophizing Scale was related to school attendance ( $t=1.97$ ;  $p<0.05$ ). Those children with higher scores on this scale were more likely to have missed school. As far as the contextual factors related to child's pain, for those parents who tend to respond solicitously to a child's pain behavior the likelihood of child's school absenteeism was higher ( $t=2.01$ ,  $p<0.05$ ), but no significant differences were seen for the other types of parental reactions (i.e. discouraging child pain behavior and parental promoting of child well behavior and coping).

In an attempt to identify predictors, the same procedure described above was followed for the regression analyses with school attendance as a dependent variable. First, the relevant pain severity variables (i.e., the highest pain intensity, the most common pain intensity, and functional disability) were entered as the first block to control for any influence that pain severity might have on school attendance. A forward stepwise method was determined. Next pain catastrophizing, the only child psychological variable related to school attendance, was entered as the second block. As can be seen in Table 5, school attendance was only predicted by variables which describe the severity of pain problems: the highest pain intensity and functional disability associated to pain (Wald  $X^2=3.82$  and  $9.69$ ,  $p<0.01$ ). As the highest pain intensity

increased across the sample, the odds of a child attending school daily decreased (OR= 0.92, 95% CI= 0.84-0.99). Also, as the functional disability associated to pain increased, the odds of a child attending school daily decreased (OR= 0.96, 95% CI= 0.92-0.99).

Next, parent predictor variables were examined in a separate equation. In this case, following the standard procedure, the pain severity variables were entered into the first block, and psychosocial variables, more specifically parents' solicitous reactions, were entered as a second block. As can be seen in Table 5, parental solicitous reactions significantly contributed to the predictive power of the model ( $p < 0.05$ ).

Table 5. Predictors of school attendance

	Block	Predictor variable	$\beta$ (SE)	Wald $X^2$	Exp $\beta$ (Odds ratio)	95% CI Exp $\beta$	Block $X^2$
Child's predictors	1	Functional disability	-0.043 (0.021)	4.252*	0.958	0.920-0.998	13.366**
		The highest pain intensity	-0.087 (0.045)	3.818*	0.916	0.839-0.999	
Parents' predictors	1	The highest pain intensity	-0.120 (0.048)	6.298*	0.887	0.808-0.974	5.676*
	2	Solicitous response	-0.514 (0.225)	5.215*	0.598	0.385-0.930	5.450*

DP: 0=Incomplete school attendance, 1=Complete school attendance.

$N_{(\text{child's predictors})}=326$ ;  $N_{(\text{parents' predictors})}=262$ . \* $p < 0.05$ , \*\* $p < 0.01$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

### 3.2.3. Level of engagement in sports or physical exercise

As can be seen in Table 3, bivariate analyses revealed significant differences for age, sex and SES on time spent on physical activities ( $t=2.38$ ,  $p < 0.05$ ;  $\chi^2=24.84$   $p < 0.001$  and  $\chi^2=18.53$ ,  $p < 0.001$ , respectively). Namely, boys, older children and children with higher SES were more likely to be involved in sports than girls, younger children, and children with lower SES. When the pain severity data (*i.e.*, pain intensity, number of pain episodes, presence of recurrent pain, and functional disability associated to pain) were examined, no statistically significant relationship was found. When the psychological variables related to pain (*i.e.*, coping strategies, emotional reactions, beliefs, and catastrophic thinking) were examined, those children who were engaged in physical activities had lower catastrophic thinking about pain, used cognitive distraction as a pain coping strategy less often, and perceived themselves as having relatively more control of their pain than those children who were not engaged or almost not engaged in sports ( $p < 0.05$ ).

Finally, when contextual variables (*i.e.*, parental reactions to child's pain) were examined, parents whose child was engaged in sports scored lower on solicitous reactions to child's pain behaviors than parents whose child was not engaged ( $t=2.33$ ;  $p < 0.05$ ).

Again, we established a logistic regression model to analyse the relative contributions of the severity of pain problems and psychosocial variables related to pain on the child's engagement on physical exercise or sports, results are summarized in Table 6. The first logistic regression showed that sex and SES were related significantly with the engagement in physical activities (Block  $X^2=33.65$ ,  $p<0.001$ ). Boys and children with higher SES tended to be more involved in this type of activities during their time out of school. Also the evaluation of psychological variables showed that pain control attitudes improved the predictive power of the model (Block  $X^2=6.50$ ,  $p<0.05$ ). Children who believed that they were able to control their pain problems were those more likely to be involved in physical activities (OR=1.69, 95% CI= 1.13-2.54).

The second logistic regression analysis to study the relative contributions of the parents' variables showed that the parents' solicitous reaction to child's pain problems did not exert any effect by itself on the fact of being involved in physical activities.

Table 6. Predictors of the engagement on physical activity

Block	Predictor variable	$\beta$ (SE)	Wald $X^2$	Exp $\beta$ (Odds ratio)	95% CI Exp $\beta$	Block $X^2$
1	Sex	-0.819 (0.206)	15.747***	0.441	0.294-0.661	33.648***
	SES					
	Low SES vs High SES	1.024 (0.251)	16.594***	2.785	1.701-4.558	
	Low SES vs Medium SES	0.771 (0.259)	8.837**	2.162	1.300-3.594	
3	Pain control	0.526 (0.207)	6.480*	1.693	1.129-2.539	6.504*

DP: 0=Not engagement or almost not engagement (average amount of time spent weekly= $or>90$ ), 1= (average amount of time spent weekly  $<90$ ).

$N_{(child's\ predictors)}=459$ . \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

### 3.2.4. The average time spent on sedentary activities

Table 3 provides a summary of the relationship between time spent in sedentary activities and its potential predictors. Age and SES were positively associated with the time spent on sedentary activities ( $r=0.34$   $p<0.01$ ;  $F=5.75$ ,  $p<0.01$ , respectively). Sex had no effect. Children who reported suffering fewer pain episodes, with lower levels of disability associated with pain, and those without chronic pain spent significantly less time on sedentary activities ( $r=0.13$ ,  $p<0.01$ ;  $r=0.10$ ,  $p<0.05$ ; and  $F=6.89$ ,  $p<0.01$ , respectively). Pain intensity was not related to time spent on sedentary activities. As regards child psychological variables related to pain catastrophic thinking about pain, externalising and internalizing coping, and negative affect were all positively associated with greater involvement in sedentary activity. The greater the emotional response the greater the sedentary behaviour (PCS  $r=0.09$ ,  $p<0.05$ ; PCQ-Ext  $r=0.18$ ,  $p<0.01$ ; PCQ-Int/Cat  $r=0.10$ ;  $p<0.05$ ; NA  $r=0.18$ ;  $p<0.001$ ). Finally, the stronger the belief that

pain is controllable ( $r=-0.10$ ,  $p<0.05$ ) and that medicine can be trusted ( $r=-0.09$ ,  $p<0.05$ ), the less sedentary activity. Among the parental variables, a less solicitous parental response to child pain behaviour was associated with greater child sedentary behaviour ( $r=-0.12$ ,  $p<0.05$ ).

Multiple regression analyses were required to predict time spent on sedentary activities from the examined variables. Two socio-demographic variables and one severity variable had a significant effect on time spent on sedentary activities: age ( $\Delta R^2=0.09$ ,  $p<0.001$ ), SES ( $\Delta R^2=0.03$ ,  $p<0.001$ ), and number of pain episodes ( $\Delta R^2=0.01$ ,  $p<0.01$ ). Psychosocial variables related to pain had no significant effect. The complete child's equation explained 12% of the variance for this outcome. Results are summarized in Table 7.

Table 7. Predictors of the time spent on sedentary activities

	Block	Predictor variable	$R^2$	Adj. $R^2$	$\Delta R^2$	$\Delta F$	$\beta$
Child's predictors	1	Age	0.090	0.088	0.090	43.440***	0.294***
		SES					
		Low SES vs High SES	0.119	0.115	0.029	14.300***	-0.170***
	2	Number of pain episodes	0.127	0.121	0.008	3.882*	0.089*

$N_{(\text{child's predictors})}=441$ . \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

#### 4. Discussion

Four related aspects of "function" were measured in children (aged 8-16 years) who experienced pain in the three months prior to testing. In line with previous research (e.g., Perquin et al., 2000; Roth-Isigkeit et al., 2004) pain was found to be a common experience. Impaired function was, however, uncommon. Functioning as measured by self-reported quality of life was predicted by low pain, low levels of disability associated to the pain complaints, low recurrence of pain, and the absence of fear-related cognition. Functioning as measured by school attendance was predicted by low levels of disability associated to the pain complaints and a low number of pain episodes. Child psychological variables related to pain did not help to explain the variance around the score on school attendance, but parental solicitous reactions to child's pain behavior did: the more attention given to pain by parents the lower the school attendance. Functioning measured by engagement in physical and sporting activity was related to male sex, high SES, and high pain control. Finally, younger age, high SES and fewer pain experiences were related to less sedentary behaviour.

The pattern of findings for identified predictors is not consistent across the several outcomes used. This might be due to the non-equivalence of the outcomes (see Table 2). A comprehensive picture emerges of child functioning in the context of pain, when different aspects of functioning are sampled. In this large sample of children we find that pain is common and occurs without major disability. However, in general, across many aspects of functioning

the severity of pain complaints affects the ability to function normally. A low number of pain episodes, low intensity of pain, and low disability were all consistently associated with better function. Although this is perhaps unsurprising the role of these variables has not been definitively examined amongst a non-clinical school attending population of children. Our knowledge arises largely from studies with samples of children who suffer from chronic pain (Kashickar-Zuck et al., 2001, Frare et al., 2002, Thachuk et al., 2003, Merlijn et al., 2006, Gauntlett-Gilbert and Eccleston, in press).

Of significant interest in this study was the finding that across most aspects of functioning the influence of psychological variables was also found. Importantly, children with low fear of pain consistently function better. It is unclear what drives these relationships. One can consider that positive coping and low catastrophic thinking about pain may inoculate one from pain related disability. Theoretically, one potentially helpful way to conceptualise the relevant psychological variables is within the context of a psychosocial model of pain related disability. Vlaeyen and Linton (2000), for example, in studying adult disability, argue that low catastrophic interpretations of pain, low pain-related negative affect, and low uncontrollable expression of emotions related to pain (i.e. externalising), would decrease pain-related fear and promote child engagement in activity (such as physical activities), thereby improving quality of life. Although at a relatively early stage of development, a greater focus on the role of specific variables of fear will be helpful in this field because of the equivocal clinical findings (e.g., Vervoort et al., 2005; Lynch et al., 2006; Gauntlett-Gilbert and Eccleston, in press).

We found little support for the role of parental context on child functioning (see Merlijn et al., 2006 for similar results). Although solicitous parental response is related to the majority of the aspects of the child's functioning examined, its influence only emerged as important for the child's school attendance behavior. The finding that school attendance behaviour does not depend on child psychological variables, but is related to parental factors may be partly explained by the fact that parents have a legal obligation to ensure that their children attend school.

These findings, in conjunction with those reported in clinical samples (Walker and Zeman, 1992; Walker et al., 1993) can be taken as support for Peterson and Palermo's hypothesis (2004) that child psychological distress moderates the relationship between parental response to children's pain behavior and functional disability. That is, for those children who are more distressed, greater parental solicitousness might lead to greater functional disability. From this perspective, studies focused on clinical samples, which are a more distressed population, are more likely to find that parental solicitous response impacts on the child. Further more detailed investigation focussing on specific relationships is necessary. For example, for the community population it will also be interesting to investigate the influence of peers (see Merlijn et al. 2006). Amongst healthy youth, the influential role of peers might be higher than the influential role of parents.

There are a number of limitations to the current study. First, what we gained in the breadth of sampling and measurement we have lost in the detail. For example, we included a

measure of self-reported sedentary behaviour that produced interesting results. However, we are not able to give context to the meaning of the sedentary behaviour. Although physically sedentary, children could be mentally active (school-work at home, etc). In support of this, note in Table 2 that time spent in sedentary activity correlated negatively with quality of life, but had no association with time spent on physical activity or school attendance. Future studies will need to further sample the activities children are engaged in when sedentary. Second, we found little support for the role of parental context on child behaviour. This may be due to inadequate measurement. Conversely, it may be that the effects of parental solicitousness are only important in clinical populations, and there should be a focus on variables more relevant to the active normally functioning child such as peer solicitousness. Third, for some measures, because of the distribution of the data, we created dichotomous variables from continuous scales (*i.e.*, school attendance, level of engagement on physical activities, and functional disability). Although a necessary manoeuvre it may have created a considerable loss of information and statistical power leading to an inflated risk of type II error. Finally, our conceptualisation of function was pragmatic and based on available instruments. Some aspects of function went unsampled. For example, we measured school presence but not academic performance, sport engagement but not enjoyment, parental influence but not peer influence. For a truly comprehensive and integrative approach we may have to invoke multiple levels of measurement and units of analysis (e.g., individual, dyad, family, group) (Palermo and Chambers, 2005).

This study adopted exploratory methods where variables were examined without specific hypotheses. We suggest a number of avenues of further investigation. New approaches to model-based analysis based on the available results are needed because understanding how children experience everyday pain, and the relationship between everyday pain and functioning, are crucially important to the prevention of chronic pain and disability. These data highlight the potential protective function of psychosocial variables such as positive self-regard, the lack of a strong belief in the disabling consequences of pain, and a non-fearful interpretation of pain. Interestingly, these data also raise questions about the role of other variables such as sex, physical fitness (including BMI) and social and economic status. Finally, a focus on positive outcomes such as acceptance of pain and social engagement, and a focus on positive psychological variables, such as adaptive coping and pain fearlessness, are uncharted territory in pediatric pain. We need to develop a theoretical understanding and a predictive model to explain why most children who commonly experience pain do not develop chronic distress and disability. Such a model will require not only the measurement of key variables and relationships previously missing from the literature, but also an explicit focus on the development of beliefs about, and behaviour in response to, everyday pain.

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**Paper #7:**

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Predictive factors of the course of pain in  
schoolchildren**



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## **PREDICTIVE FACTORS OF THE COURSE OF PAIN IN SCHOOLCHILDREN**

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### **ABSTRACT**

The study of psychosocial factors related with the onset and persistence of chronic pain in children and adolescents has recently attracted the interest of experts. The evidence collected until now is deficient. The aim of this study was to examine the influence of several psychosocial variables in the onset and maintenance of chronic pain problems among children. A prospective study of schoolchildren aged between 8 and 16 years was conducted in Catalonia (a region in the North-East of Spain). 561 children and adolescents and 223 parents participated in this study. Relevant information was collected through individual interviews with the children. Children were interviewed twice: at baseline, and at one-year follow-up. Parents were only requested to provide some information at baseline. Data were collected on biological, social and psychological factors. Child pain experience was also assessed. Potential predictors included age, sex, pain characteristics, coping with pain, pain-related affect, beliefs about pain, parental pain, parental reactions to children's report of pain, level of physical activity, level of sedentary activity, and number of stressful events in the previous 12 months.

The average annual incidence rate of chronic pain was 9.25, and the average annual recovery was 14.96. Results showed that chronic pain presence at baseline was predicted by older age, sex (female), high catastrophizing and low belief in a medical cure for pain. Onset of chronic pain at one-year was predicted by catastrophizing coping, time spent on sports or physical activity out of school and number of stressful events in the year before the follow-up. Finally, chronic pain persistence at 1-year follow-up was predicted by age and generalized chronic pain at baseline. As far as parents' factors concerned, neither the presence of chronic pain nor their reactions to the child's pain behaviors were related with any outcome.

The results of this study strongly support a multifactorial cause responsible for the course of pain in youth.

**Keywords:** schoolchildren, prognostic factors, chronic pain, onset, persistence, psychosocial characteristics

## 1. Introduction

Chronic pain in youth is a common experience (Perquin et al., 2000; Roth-Izigkeit, 2005; Huguet and Miró, 2007), in some cases it might be quite severe and disabling (Palermo, 2000; Kashikar-Zuck et al., 2002; Konijnenberg et al., 2005). The interest in the assessment and treatment of chronic pediatric pain has increased in the last 20 years, and important advances have been made (e.g. Eccleston et al., 2003; 2006). However, neither the development of chronic pain nor its impact are well-understood yet. With the advent of biopsychosocial models to study chronic pain in children (Walker, 1999; Zeltzer et al., 1997; Palermo and Chambers, 2005), we are gaining a better understanding about their occurrence.

Various factors, other than the traditional biomedical ones, have been identified and suggested to play a significant role in the development and perpetuation of pain. Unfortunately, a clear picture of the contributing factors and the interactions among them is far from clear. Thus far, several factors have been studied, for example, socio-demographic characteristics (e.g. Bener et al., 2000; Boey and Goh, 2001; Bejia et al., 2005; Laurell et al., 2005), anthropometrical and physical conditions (e.g., Harreby et al., 1999; Szpalski et al., 2002; Kovacs et al., 2003; Sjolie, 2004; Bejia et al., 2005), mechanical factors (e.g., Jones et al., 2003; Watson et al., 2003; Prista et al., 2004; Bejia et al., 2005), health-related behaviors (e.g., Vikat et al., 2000; Feldman et al., 2002; El-Metwally et al., 2006), general health conditions (e.g., Flato et al., 1997; El-Metwally et al., 2004), psychosocial factors (e.g., Brusgaard et al., 2000; Perquin et al., 2003; Jones et al., 2004), lifestyle issues (e.g., Kristjansdottir and Rhee, 2002; Ghandour et al., 2004), and pain related characteristics (e.g., Perquin et al., 2003; El-Metwally et al., 2005; Larsson and Sund, 2005). The evidence about the relative contribution of each of these factors still is unknown. Most reports about prognostic factors related to the onset and course of recurrent or chronic pain come from cross-sectional studies (Balagué et al., 1999; Jones and Macfarlane, 2004) where the temporality of the relation is doubtful. Although there exist a few prospective studies, they typically investigate the onset or persistence of specific types of chronic pain problems (for back pain see, for example, Bejia et al., 2005).

There is a need for prospective works that study risk factors for pain becoming chronic, as well as for the persistence of chronic pain (Goodman and McGrath, 1991; Larsson and Sund, 2005). These studies would have to be conducted on samples drawn from the general population, and not restricted to any specific pain problem; since it is recognized that different pains frequently co-occur together these studies would have to be addressed to pain in general (Perquin et al., 2003).

Learning about these factors contributing to chronic pain syndromes would help us improve current treatment programs. But what could be considered even more important is that this knowledge would be fundamental to identify children that are at risk for developing chronic disabling pain problems, and to implement preventive strategies.

This study was designed to explore the contribution of a set of child's and parents' factors on the course of children's pain over a 1-year period. Specifically, the aim was to determine what factors predicted (a) chronic pain, and (b) the persistence of chronic pain at 1-

year follow-up. Based on previous research, it was hypothesized that the course of pain would be predicted by a set of demographic characteristics, pain related factors (both physical and psychosocial parameters), and life style issues.

## 2. Method

### 2.1. Participants

902 school-children and their parents were recruited from non-special primary and secondary schools (from grade 2 to 8) in Tarragona, a province of Catalonia (Spain). Children were excluded if they were (1) suffering from any cognitive impairment (four children), or (2) unable to understand Catalan (none) as reported by their teacher. 561 children participated (62%). Most children did not participate due to lack of parental consent. There were no significant differences in either school grade or sex between participants and non-participants. Child age ranged from 8 to 16 years (mean age=10.9; SD=2.0). 51.7% of the sample was male. Parents of 223 children participated and provided all the information requested (see Figure 1). Table 1 gives demographic and clinical data of the participating children and parents. All participating children were approached again after one year of the initial contact (a median of 365.79 days; range 404-271 days).

*Table 1. Characteristics of the study population at baseline*

	Scale range	Mean (SD)
<b>Children's characteristics</b>		
1. Socio-demographic variables		
Age (years)	8-16	10.74 (1.93)
Sex (n,%)		
Boys	--	51.7 (290)
Girls	--	48.3 (271)
SES (n, %)		
High SES	--	40.1%
Medium SES	--	32.3%
Low SES	--	27.6%
2. Anthropometric variables		
BMI	--	10.09 (3.6)
3. Pain severity variables		
The highest pain intensity in the last three months (of the most troublesome pain)	0-10	5.24 (3.19)
The most common intensity in the last three months (of the most troublesome pain)	0-10	3.37 (2.34)
Functional disability	0-52	4.23 (5.7)
Number of pain episodes (in the last three months)		11.37 (17.88)
Presence of chronic pain (n,%)	--	208 (37.3)

Table 1 (cont'd). Characteristics of the study population at baseline

<b>Children characteristics</b>		
4. Psychological variables related to pain		
Pain coping strategies		
Information seeking	1-5	2.33 (0.84)
Problem solution	1-5	2.82 (0.93)
Seeking social support	1-5	2.96 (0.84)
Positive self-statements	1-5	2.86 (0.93)
Behavior distraction	1-5	3.21 (1.03)
Cognitive distraction	1-5	3.14 (1.04)
Externalizing	1-5	1.63 (1.07)
Internalizing/ Catastrophizing	1-5	2.00 (1.00)
Pain beliefs		
Control	0-2	1.47 (0.49)
Disability	0-2	0.97 (0.75)
Harm	0-2	1.12 (0.57)
Emotion	0-2	0.,92 (0.74)
Medication	0-2	1.85 (0.44)
Solicitude	0-2	1.70 (0.52)
Medical Cure	0-2	1.74 (0.39)
Pain related affect		
Positive affect	0-50	21.54 (6.01)
Negative affect	0-50	16.59 (4.75)
5. Functioning outcomes		
Quality of life	0-100	81.83 (10.11)
Average amount of time spent in physical activities (minutes)	--	166.07 (151.79)
Average amount of time spent on sedentary activities (minutes)	--	1146.31 (594.65)
<b>Parents' characteristics</b>		
1. Socio-demographic variables		
Age		40.61 (4.30)
Marital status (n,%)		
Married	--	86.3 (484)
Separated or divorced	--	12.4 (70)
Widow/er	--	1.3 (7)
2. Parents' informing about the child's pain		
Parents' reactions to the child's pain behaviors		
Solicitousness	0-4	2.36 (0.67)
Discouragement	0-4	0.91 (0.64)
Promotion of well- behaviors and coping	0-4	2.57 (0.68)
3. Presence of chronic pain (n,%)		
Father		74 (29.5)
Mother		138 (44.7)

Note: SES: Socio-economic status

## 2.2. Procedure

Following governmental and school approval, 40 classes of 20 non-special primary and secondary schools in Tarragona (a province of Catalonia, Spain) were randomly selected by means of a stratified sampling procedure and invited to participate. All invited schools agreed to participate, except for one who claimed current involvement in other research projects as the

reason for non-participating. Children were interviewed individually in the school, the interview lasted approximately 40 minutes. Children were asked to provide information about their daily functioning and about their common pain experiences. Once the interview finished, the children's weight and height were rated. Parents were surveyed separately; they were instructed to complete written questionnaires at home which included questions about their common pain experiences, and about their reactions to the child's pain behaviors. One year later, participating children were approached again, the same questions about their common pain experiences were asked. This time, the children were also asked to recall stressful life events occurring in the previous 12 months.

Because the interview was concerned with past experiences (i.e. children were requested to recall their pain experiences over the three months before the interview and they were also asked to make judgment about their quality of life in the month previous to the interview), memory enhancing techniques were used (Orbach and Lamb, 2000). For example, interviewers used open-ended questions, and important events as end point times, in order to prevent interviewer bias from affecting children's reports, and to increase the accuracy of the information. Moreover, several other quality control measures were implemented during data collection. First, the two interviewers involved in the study attended standardization classes in which they were trained in interview technique. Second, data sheets were collected during the interviews and they were further monitored centrally to check for missing data or inconsistencies after each interview.

## **2.3. Measures**

### *2.3.1. Child baseline measures*

#### *2.3.1.1. Socio-demographic characteristics*

Information about age, sex, number of siblings, and number of people living at home was collected at the time of interview. Children's socioeconomic status (SES) was rated on the basis of the work by Domingo-Salvany et al. (2000). Since both parents were requested to provide their current occupation, the "highest" occupation of the two parents was the one used to rate the SES of the family and the child.

#### *2.3.1.2. Anthropometric characteristics*

Child's weight and height were measured at the end of the interview.

#### *2.3.1.3. Pain characteristics*

Children were asked to report any pain that they had experience in the last three months. If they had experienced pain in the last three months, further information about these experiences was requested.

Following Mikkelson et al., (1997) an open question was used about where the pain was located, and a pain mannequin drawing included (Goodman et al., 1997). *Pain location* was

coded according to a list based on the axis I of IASP Classification of Chronic Pain (Merskey and Bogduk, 1994).

*Pain frequency* was measured on a categorical scale following Perquin et al., (2000) work. Some information about *pain duration* was also recorded. Specifically, we asked the participating schoolchildren to report whether they had been experiencing pain for more than 3 months, and pain duration was recorded in days, weeks and months.

The highest and usual *pain intensity* in the last 3 months was measured using the Catalan version of the *Faces Pain Scale – Revised* (FPS-R) (Miró and Huguet, 2004). The FPS-R is a six-point scale designed to measure children's level of perceived pain intensity. The children's task was to choose the face that best reflects the intensity of the pain they have experienced. A numerical value from 0 to 10 is assigned to each face. The end points are explained as "no pain" and "very much pain".

In the case of children who reported to be suffering from more than one pain problem, they provided information about these pain characteristics (i.e., intensity, frequency, duration and location) in relation to each pain complaint; additionally, they were also requested to mention which pain complaint had troubled them most in the last three months.

In this study, chronic pain was defined as a pain lasting for three months at least, that was experienced continuously or recurrently (see Elliott et al., 1999); a recurrent pain problem was one occurring once or twice at least each month (see Mikkelsen et al., 1997 for a similar procedure). Thus, timeframe was not the only criteria for defining chronic pain, the frequency of pain was also used to screen for chronic pain.

The *Functional Disability Inventory* (FDI; Walker and Greene, 1991) was used as a measure to assess pain-related disability. This measure assesses 15 activities in a variety of contexts including school, home, recreation, and social interaction. The children were asked how much difficulty they experienced doing these activities because of their pain, using a 5-point severity scale ranging from 0 (no trouble) to 4 (impossible). The total score is obtained by summing the ratings, therefore the greater the final score, the greater the level of functional disability. Claar and Walker (2006) and Walker and Greene (1991) have reported appropriate psychometric properties for the instrument both in clinical and healthy samples. The Catalan version of the measure has shown similar psychometric properties to the original version (Huguet et al., 2007).

#### 2.3.1.4. Chronic pain severity

Pain severity was classified in five grades: (1) *Grade 0*: a non-chronic pain problem; (2) *Grade 1*: low pain intensity (intensity index < 5; range= 0-10) and low pain-related disability (FDI score < 10; range: 0-52), (3) *Grade 2*: high pain intensity (intensity index > 5; range= 0-10) and low pain-related disability (FDI score < 10; range: 0-52); (4) *Grade 3*: moderate level of pain-related disability, regardless of pain intensity (FDI score= 10-20); and (5) *Grade 4*: high level of pain-related disability, regardless of the pain intensity (FDI score > 20) (for additional information about this grading system see Huguet and Miró, 2007).

### 2.3.1.5. *Quality of life*

The *Paediatric Quality of Life Inventory™ Version 4.0* (PedsQL, Varni et al., 2001) was used to assess children's perception of their quality of life. The PedsQL is a generic instrument that encompasses four multidimensional scales (physical, emotional, social and school functioning) and three summary scales (total, physical health summary, psychosocial health summary). Developmentally appropriate versions (i.e., ages 8–12 and 13–18) were used. The children were asked how much of a problem each item has been in the past month. Items were scored on a 5-point Likert scale, from 0 (never a problem) to 4 (almost always a problem). Items are reverse-scored and linearly transformed to a 0-100 scale (0=100, 1=75, 2=50, 3=25, 4=0), so that higher scores indicate better quality of life. Its psychometric properties have been examined in Catalan-speaking school-children by Huguet and Miró (in press)

### 2.3.1.6. *Pain beliefs*

The child version of the *Survey of Pain Attitudes* (SOPA-C, Engel et al., 2007) was used. Engel and colleagues developed the questionnaire from the original scale used to assess adult pain attitudes (see Jensen et al., 1994). The survey includes twenty-nine true-false attitude statements related to pain which underlie seven attitudes: (a) belief in a medical cure for pain (Medical Cure scale), (b) belief in one's ability to control pain (Pain Control scale), (c) belief that it is the responsibility of others to assist people who are experiencing pain (Solicitude scale), (d) belief that one is disabled because of pain (Disability scale), (e) belief that the medications are the best treatment strategy for pain (Medication scale), (f) belief that pain means something is being hurt in the body and consequently, that exercise should be avoided (Harm scale), and (g) belief that emotion affects pain (Emotion scale). As suggested by Engel et al (2007), children were asked how much they agreed with each attitudinal statement on a 3-point Likert scale (0 = I do not agree with this, 1 = I am not sure, 2 = I agree with this). To obtain a score for each SOPA-C subscale, which may be used as a measure of overall agreement with the belief construct, ratings of the items within each subscale need to be averaged after reversing the score of a few items. The instrument has shown appropriate psychometric properties when used in children with chronic pain and disabilities (Engel et al., 2007). We used the Catalan adapted version of the SOPA-C validated by Miró et al. (2007).

### 2.3.1.7. *Pain coping strategies*

Children pain coping strategies were assessed with the *Pain Coping Questionnaire* (PCQ, Reid et al., 1998). The PCQ has 35 items corresponding to seven coping strategies that the child might use to cope with or manage pain. The strategies include 'approach' coping strategies (information seeking, problem solving, seeking social support, positive self-statements), 'problem-focused' avoidance strategies (behavioural distraction and cognitive distraction) and 'emotion-focused' avoidance strategies (externalizing and internalizing/catastrophizing). On a 5-point Likert scale from 1 (never) to 5 (very often) the children rated how often each of these coping strategies is used when in pain. The score for each PCQ subscale is calculated by summing the ratings given for each item within each scale,

and dividing by the number of items in that scale. The reliability and validity of the PCQ in healthy school children has been documented by Reid et al. (1998). The questionnaire has shown good psychometric properties when used with Catalan speaking school-children (see Huguet et al., 2007).

#### *2.3.1.8. Pain related affect*

The child version of the *Positive and Negative Affect Schedule* (PANAS-C) is a standardised measure which consists of 20 adjectives, ten assessing positive affect (e.g., excited) and ten assessing negative affect (e.g., upset). An adapted Catalan version was used here (Huguet et al., 2007). Children were asked to rate on a five point scale, from 1 which means “not at all or almost not at all” to 5 “a lot” the extent to which each adjective describes their feelings when they are in pain, as opposed to describing their feelings in the last few days. The measure has shown good psychometric properties in a sample of Spanish children too (Sandin et al., 2003).

#### *2.3.1.9. Physical activity*

Children also provided information about their usual amount of weekly time engaged in sports and other physical exercise (out of school), and usual amount of time spent on sedentary activities (specifically: watching television, using the computer and playing videogames).

### **2.3.2. Child follow-up measures**

#### *2.3.2.1. Pain*

We collected the same information as in baseline.

#### *2.3.2.2. Stressful events in the previous 12 months*

The questionnaire EAVI (Canalda et al., 1992) was used to assess stressful life events in the previous 12 months. EAVI is a checklist of 84 possible stressful situations (e.g., parents divorce, death of a familiar member). Children’s task was to indicate whether they had or not experienced each of the situations listed in the preceding 12 months.

### **2.3.3. Parent measures**

#### *2.3.3.1. Socio-demographic characteristics*

Age, sex, marital status, and occupation status were recorded.

#### *2.3.3.2. Pain characteristics*

Parents were asked to report if they had experienced any pain in the last six months. They were also asked to indicate the localization using a categorization based in the axis I of IASP Classification of Chronic Pain (Merskey and Bogduk, 1994). In case that they reported to have had more than one pain problem, they were requested to mention which of those had interfered them most in the previous 6 months. And, additional information about the

characteristics of the most bothersome pain complaint was requested. Specifically, they were requested about the course of the most bothersome pain (categories: continuous, recurrent), and in case that they reported that the course of their pain was recurrent, they were requested about the frequency of pain episodes (categories: more than once a week, once a week, once or twice a month, five or less times each year). Finally, pain duration was registered.

Those subjects with any pain complaint that persisted for more than six months were considered to have chronic pain (Merskey and Bogduk, 1994).

#### *2.3.3.3. Parental response to the child's pain behaviors*

Parents reported their reactions to their child's pain behaviors by responding to IRPEDNA (see Huguet et al., in press). IRPEDNA is a self-administered questionnaire that has three subscales: 1) solicitousness, 2) discouragement of pain behaviors, and 3) promotion of well-behaviors and coping. Respondents were asked to answer how often they enact each of the reactions included in the inventory when their child is in pain. Parents rated each item using a five-point response scale (0=never, 1=hardly ever, 2=sometimes, 3=often, 4=always). This instrument has shown to have adequate reliability and validity characteristics (Huguet et al., in press).

#### **2.4. Data analysis plan**

Data were analyzed using SPSS for Windows. Initially, for descriptive purposes, changes in chronic pain status over a year were described, and children were grouped in four categories: (1) children with no chronic pain over 1-year period, (2) children that did not report chronic pain at baseline but were found suffering from chronic pain at one year follow-up assessment, (3) children for which pain persisted at 1-year follow-up, and (4) children who reported chronic pain at baseline, but were recovered from it when interviewed at 1-year follow-up.

As a first approach, to examine which baseline factors (child and parents factors) were related with the course of pain status over time, bivariate analysis were conducted to compare baseline characteristics between the four groups outlined above. Next, three sets of multivariate regression analyses were conducted to identify prognostic factors of: 1) chronic pain at baseline, 2) pain becoming chronic at one year follow-up, and 3) persistence of chronic pain at one year follow-up. In each set of the multivariate analyses two separate hierarchical logistic regression analyses were performed because child and parent predictors were analysed in separate equations. Bivariate associations between each of the three outcomes and potential prognostic factors were examined by the Spearman's rank (Spearman's  $\rho$ ) correlation coefficient before performing multiple regression analyses. Only factors significantly related to outcomes in bivariate analyses were entered into multivariate models. In the 'child equations', we entered demographic and anthropometrical variables (*i.e.*, age, sex, and body mass index) in the first block. The second block included child's psychological variables related to pain (*i.e.*, pain-related affect, pain coping strategies, and pain attitudes), and the third block included those

variables related to the child's lifestyle (*i.e.*, physical activity index, inactivity index, and number of stressful events occurred in the year before follow-up). Only when persistence of chronic pain was used as an outcome, variables related to chronic pain characteristics at baseline (*i.e.*, severity of chronic pain, and number of painful sites) were entered as another block, and before entering the child's psychological variables related to pain. In the 'parent's equations' sociodemographic variables were entered in the first block, whereas occurrence of chronic pain in parents, and IRPEDNA scales' scores were entered later.

The assumptions of the model were checked for each regression analysis performed. The patterns shown in the plot standardized residuals against standardized predictive values were checked for the assumption of linearity and homocedasticity. Durbin-Watson statistics were used to support the assumption of independent residuals. To exclude the influence of multicollinearity the VIF value for every independent variable was calculated. Moreover, the outliers were also checked in order to be sure that they did not unduly bias the results of the regression equations.

### **3. Results**

#### **3.1. Response rate**

Figure 1 shows the flow diagram of participants throughout the study. Of the 902 children that were invited to participate, 561 children and the parents of 223 of those children were completely involved in the study at the baseline, participation rate was 62.19% and 25%, respectively. The reasons for children non-participation were: (1) non compliance with the inclusion criteria ( $n=4$ ), and not giving in the written informed consent from parents ( $n=337$ ). Just a few comparisons between participating and non-participating children at baseline could be performed due to the limited data available on eligible non-participants. The results showed that there were no significant differences in either school grade or sex.

Of the initial 561 participating children, 50 were not interviewed at 1-year follow-up (participation rate at follow-up=91.09). The reason for losing participants at follow-up included: declined to participate ( $n=2$ ), moved to another city ( $n=7$ ), changed school ( $n=40$ ), and a long stage in hospital ( $n=1$ ). Attrition analyses indicated that the few children who were lost at follow-up only differ significantly from those who provided data at both assessments on age (mean age= 12.48 and 10.74, respectively;  $t=6.06$ ,  $p<0.001$ ). This is not surprising granted the fact that, in Catalonia, when students finish 5<sup>th</sup> grade they sometimes have to move to a different school to continue their education. Thus, children around 12 years old were expected to be more likely to be lost during follow-up than any other age group in our sample of participating children. However, no other differences were found on any of the variables considered important. Thus, besides this low attrition rate (attrition rate at follow-up=8.91%), our data suggest that there has not been any systematic bias that would compromise the generalizability of our findings.

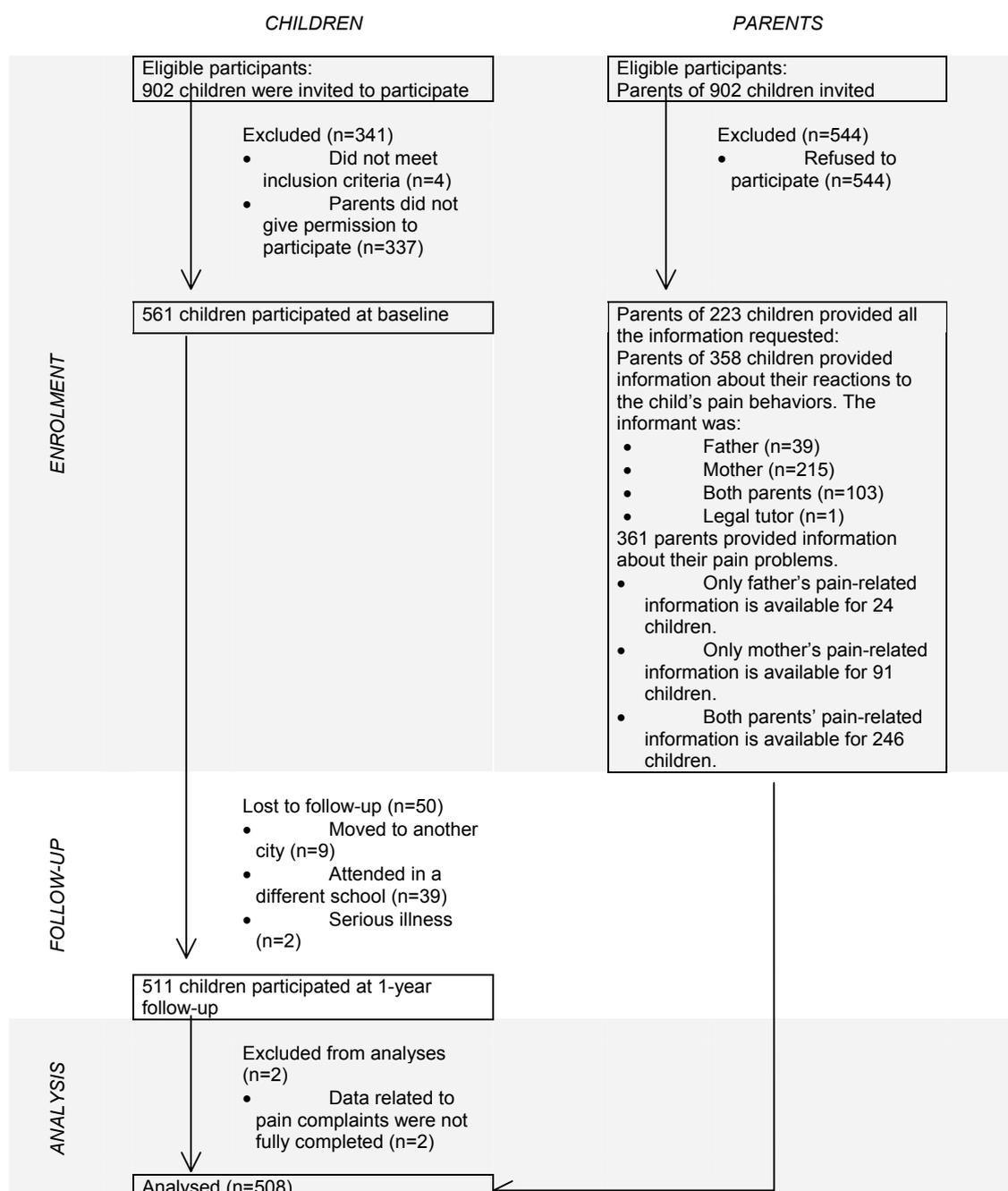


Figure 1. Diagram of flow

### 3.2. Characteristics of participants at baseline:

Pain complaints were quite usual among children. At baseline, children reported a mean of 11.74 (SD=18) pain episodes in the previous three months, and a median of 5. Lower limb pain was reported as the most troublesome pain complaint. The highest intensity of the most troublesome pain over the previous three months was 5.31 (SD=3.23, range 0-10), and the most common intensity of the most troublesome pain was 3.41 (SD=2.38, range 0-10).

According to the criteria used in this study to define chronic pain, 37.3% of children suffered from it. Almost a third of those (29%) reported pain in multiple body regions. Compared with clinical populations, participants in this study reported lower disability rates (e.g. Walker and Greene, 1991; Kashikar-Zuck et al., 2002). Our participants' quality of life (mean score =81.30, SD=10.52) was almost the same as that reported by other healthy samples (see Varni et al., 2006), and higher than the one reported by clinical chronic pain samples of the same age (see Connelly et al., 2006).

### 3.3. Changes in pain status over 1 year:

Table 2 shows the change in pain status between baseline and 1-year follow-up. Of the 326 children (64.17%) who did not report chronic pain at baseline, 47 (14.42%) reported chronic pain at 1-year follow-up. Among the 182 children (35.82%) who reported chronic pain at baseline, 106 (58.24%) still suffered from chronic pain at 1-year follow-up. The average annual incidence rate of chronic pain was 9.25, and the average annual recovery rate was 14.96.

*Table 2. Change in chronic pain status between baseline at 1-year follow-up.*

Chronic pain status at baseline	Chronic pain status at 1-year follow-up		Total
	Non-chronic pain	Chronic pain	
Non-chronic pain	279 (85.58%)	47 (14.42%)	326 (100%)
Chronic pain	76 (41.76%)	106 (58.24%)	182 (100%)
Total	355	153	508

### 3.4. Variables related to pain status

Means or percentages of potential predictors of the course of pain status are reported in Table 3, as well as comparisons among the four different groups (i.e., children with no chronic pain over 1-year follow-up, children with chronic pain onset at 1-year follow-up, children with persistence of chronic pain over 1-year period, and children who had recovered from chronic pain at 1-year follow-up). As far as demographic variables are concerned, children with chronic pain that persisted over 1 year were significantly older ( $p < 0.001$ ). There was a higher proportion of males among those who did not report chronic pain at any time (59.9% males vs 40.1% females), and a higher proportion of females among those who reported that chronic pain persisted at 1-year follow-up (71% females vs 35% males). As far as chronic pain characteristics are concerned, there was a significant difference in the number of painful sites at baseline ( $F = 12.85$ ,  $p < 0.001$ ). As far as psychological variables related to pain are concerned, and as it was hypothesized, the group means differed significantly in some scores. Specifically, those children who reported persistence of chronic pain over 1 year scored significantly higher on externalizing and internalizing/catastrophizing coping compared to those children who did not report chronic pain at any time ( $p < 0.01$ ,  $p < 0.05$ , respectively). Children who reported onset of chronic pain at 1-year follow-up also scored higher on internalizing/catastrophizing than those who did not report chronic pain at all ( $p < 0.001$ ). Children for which chronic pain persisted at 1-year follow-up perceived themselves as having less control over their pain, perceived that the medication was less effective to deal with pain, and trust less in a medical cure for pain than did

those who did not report chronic pain at any time ( $p<0.01$ ,  $p<0.05$ ,  $p<0.001$ , respectively). These children scored significantly lower on positive pain related affect than those who did not report suffering from chronic pain problems at any time, and those who reported being recovered from chronic pain at 1-year follow-up ( $p<0.01$ ,  $p<0.05$ ). Moreover, those with persistent chronic pain scored significantly higher on negative pain related affect than those without chronic pain ( $p<0.01$ ). Regarding the children life style issues, comparatively with those without chronic pain at both point times, those with onset of chronic pain or persistent chronic pain at 1-year follow-up reported to have experienced a significantly higher number of stressful life events during the year before the follow-up ( $p<0.001$ ). Finally, and unexpectedly, no differences were found between the groups regarding any of the parents' variables evaluated.

Table 3. Characteristics of the study population based on the chronic pain status over the time.

Potential predictors	Scale range	NoCP-NoCP	NoCP-CP Development of CP	CP-CP Maintenance of CP	CP-NoCP Recovery from CP	Test	
						F	X2
<b>Children's related factors</b>							
<b>Socio-demographic characteristics</b>							
Age (years)	8-16	10.36 (1.82)	10.79 (1.61)	11.69 (2.10)	10.71 (1.76)	13.12***	
Gender (% , n)							22.70***
Boys	--	59.9 (167)	46.8 (22)	33 (35)	51.3 (39)		
Girls	--	40.1 (112)	53.2 (25)	67 (71)	48.7 (37)		
<b>Physical condition</b>							
BMI		18.85 (3.37)	18.92 (2.60)	19.63 (3.84)	19.33 (4.48)	1.34	
<b>Chronic pain characteristics at baseline</b>							
Chronic pain grade	1-4	--	--	1.60 (0.79)	1.55 (0.78)	0.15	
Number of pain sites	1-9	--	--	1.49 (0.66)	1.17 (0.47)	12.85***	
<b>Psychological variables related to pain</b>							
<b>Pain coping strategies</b>							
Information seeking	1-5	2.32 (0.84)	2.48 (0.91)	2.29 (0.93)	2.30 (0.80)	0.62	
Problem solution	1-5	2.79 (0.95)	2.91 (0.87)	2.82 (0.91)	2.86 (0.92)	0.28	
Seeking social support	1-5	2.94 (1.02)	2.96 (1.02)	3.02 (1.09)	2.94 (0.99)	0.19	
Positive self-statements	1-5	2.84 (1.04)	2.80 (0.95)	2.81 (1.01)2.	2.99 (1.15)	0.53	
Behaviour distraction	1-5	3.15 (1.18)	3.04 (1.05)	3.18 (1.22)	3.26 (1.15)	0.34	
Cognitive distraction	1-5	3.18 (1.00)	3.08 (0.99)	3.04 (1.04)	3.18 (0.98)	0.61	
Externalization	1-5	1.53 (0.66)	1.74 (0.84)	1.81 (0.90)	1.68 (0.76)	4.14**	
Internalization / Catastrophizing	1-5	1.91 (0.76)	2.26 (0.70)	2.30 (0.96)	2.09 (0.83)	7.23***	
<b>Pain beliefs</b>							
Control	0-2	1.51 (0.47)	1.50 (0.45)	1.32 (0.56)	1.48 (0.47)	3.91**	
Harm	0-2	1.10 (0.58)	1.16 (0.52)	1.19 (0.54)	1.09 (0.61)	0.80	
Medical cure	0-2	1.80 (0.34)	1.73 (0.45)	1.61 (0.48)	1.72 (0.38)	5.94***	
Solicitude	0-2	1.68 (0.54)	1.77 (0.43)	1.68 (0.57)	1.77 (0.41)	0.96	
Medicine	0-2	1.90 (0.37)	1.79 (0.55)	1.76 (0.53)	1.84 (0.47)	2.94*	
Emotion	0-2	0.87 (0.73)	1.08 (0.77)	1.03 (0.73)	0.83 (0.75)	2.37	
Disability	0-2	0.96 (0.73)	1.04 (0.77)	0.98 (0.73)	0.97 (0.81)	0.14	
<b>Pain related affect</b>							
Positive affect		22.16 (5.88)	20.72 (5.47)	19.84 (5.64)	22.24 (6.88)	4.53**	
Negative affect		16.01 (4.40)	17.17 (4.76)	17.77 (5.40)	16.63 (4.72)	3.85**	
<b>Life style issues</b>							
Physical activity index (minutes)		158.72 (150.77)	215.96 (169.25)	156.18 (147.47)	175.40 (147.48)	2.16	
Inactivity index (minutes)		1094.47 (580.70)	1159.13 (514.52)	1253.35 (656.81)	1164.67 (586.89)	1.86	
Number of stressful life events		7.24 (3.74)	10.12 (3.71)	9.06 (5.03)	1164.67 (4.08)	10.04***	
<b>Parents' related factors</b>							
<b>Parents with chronic pain problems</b>							
Chronic pain in any of both parents (% , n)	--	61.7 (87)	77.3 (17)	65.1 (41)	62.2 (23)	2.09	
Father with pain (% , n)	--	28.1 (38)	33.3 (7)	28.6 (16)	34.2 (13)	0.69	
Mother with pain (% , n)	--	41.6 (69)	56 (14)	49.3 (36)	40.9 (18)	2.83	
<b>Parents' reactions to the child's pain behaviors</b>							
Solicitousness	0-4	2.42 (0.66)	2.31 (0.87)	2.23 (0.63)	2.34 (0.67)	0.95	
Discouragement	0-4	0.86 (0.61)	0.84 (0.61)	0.99 (0.67)	0.96 (0.67)	0.69	
Promotion of well-behaviors	0-4	2.57 (0.70)	2.43 (0.64)	2.58 (0.62)	2.58 (0.66)	0.33	

\*p< 0.05, \*\*p<0.01; \*\*\*p<0.001

### 3.5. Predictors of chronic pain at baseline

Table 4 shows the correlations between potential predictors and outcomes for chronic pain status at baseline. The occurrence of chronic pain at baseline was significantly related with age, sex, use of externalising and catastrophizing pain-coping strategies, pain related affect, beliefs about the ability to control pain, beliefs in medication as an appropriate treatment for pain, beliefs in a medical cure for pain, and average time spent on sedentary activities.

Table 4. Correlations between potential predictors and chronic pain status at baseline and the course of chronic pain status over 1-year follow-up.

Potential predictors	Chronic pain status at baseline <sup>a</sup> (NoCP vs CP)	Onset of chronic pain at 1-year follow-up <sup>b</sup> (NoCP-NoCP vs NoCP-CP)	Recurrence of chronic pain at 1-year follow-up <sup>c</sup> (CP-NoCP vs CP-CP)
<b>Children's related factors</b>			
<b>Socio-demographic characteristics</b>			
Age	0.227***	0.110*	0.228**
Sex	0.191***	0.093	0.184*
<b>Physical condition</b>			
BMI	0.075	0.041	0.066
<b>Chronic Pain severity</b>			
Chronic pain grade	--	--	0.032
Number of painful sites in cases of chronic pain sufferers	--	--	0.288***
<b>Psychological variables related to pain</b>			
<b>Pain coping strategies</b>			
Information seeking	-0.015	0.070	-0.015
Problem solution	0.037	0.055	-0.034
Seeking social support	0.020	0.013	0.044
Positive self-statements	0.022	-0.023	-0.078
Behaviour distraction	0.010	-0.051	-0.018
Cognitive distraction	-0.019	-0.031	-0.064
Externalization	0.120**	0.091	0.067
Internalization / Catastrophizing	0.128**	0.186**	0.109
<b>Pain beliefs</b>			
Control	-0.092*	-0.019	-0.139
Disability	0.005	0.034	0.015
Harm	0.035	0.034	0.074
Emotion	0.054	0.096	0.133
Medication	-0.108*	-0.088	-0.108
Solicitude	0.026	0.040	-0.036
Medical cure	-0.174***	-0.038	-0.113
<b>Pain related affect</b>			
Positive affect	-0.112**	-0.086	-0.173*
Negative affect	0.115**	0.089	0.097
<b>Life style issues</b>			
Physical activity index (minutes)	-0.014	0.129*	-0.057
Inactivity index (minutes)	0.103*	0.064	0.062
Number of stressful life events	--	0.272*	0.025

*Table 4 (cont'd). Correlations between potential predictors and chronic pain status at baseline and the course of chronic pain status over 1-year follow-up.*

Potential predictors	Chronic pain status at baseline <sup>a</sup> (NoCP vs CP)	Onset of chronic pain at 1-year follow-up <sup>b</sup> (NoCP-NoCP vs NoCP-CP)	Recurrence of chronic pain at 1-year follow-up <sup>c</sup> (CP-NoCP vs CP-CP)
<b>Parents' related factors</b>			
Chronic pain in any of both parents	0.010	0.111	0.029
Father with chronic pain	0.032	0.039	-0.060
Mother with chronic pain	0.032	0.098	0.082
Parents' reactions to child's pain behaviors			
Solicitousness	-0.078	-0.017	0.077
Discouragement	0.082	-0.003	-0.024
Promotion of well-behaviors	-0.008	-0.073	-0.032

<sup>a</sup> 0=Non-chronic pain, 1=Chronic pain  
<sup>b</sup> 0=Non-development of chronic pain, 1= Development of chronic pain  
<sup>c</sup> 0=Non-maintenance of chronic pain, 1= Maintenance of chronic pain  
 \*p< 0.05, \*\*p<0.01; \*\*\*p<0.001

A logistic regression analysis with chronic pain status at baseline as an outcome was performed to study the unique contribution of these variables on the child's chronic pain status. The results of the regression analysis are shown in Table 5. The model contained variables from blocks 1 and 2. Chronic pain status at baseline was predicted by age (Wald  $X^2=17.3$ ;  $p<0.001$ ), sex (Wald  $X^2=15.06$ ,  $p<0.001$ ), the use of catastrophizing pain-coping strategies (Wald  $X^2=9$ ;  $p<0.001$ ), and the belief in a medical cure for pain problems (Wald  $X^2=5.34$ ;  $p<0.05$ ). That is, the girls were more likely to have chronic pain (OR=2.08, 95% CI= 1.44-3.02), and. As the child's age increased, and the use of catastrophizing pain coping strategies increased, the odds of chronic pain occurring increased (OR=1.22, 95% CI= 1.12-1.34). As the use of catastrophizing pain coping strategies increased, the odds of chronic pain occurring increased too (OR=1.45, 95% CI= 1.34-1.85). However, as the belief in a medical cure increased the odds of chronic pain decreased (OR=0.58, 95% CI= 0.36-0.92). The variables related to the child's lifestyle did not make any significant contribution to the predictive power of the model, in spite of the fact that the inactivity index (i.e., time spent on sedentary activities) was related to experiencing chronic pain.

The effects of parents' variables on the status of the child's pain complaints was also examined. However, neither the presence of chronic pain in any of both parents, nor the type of reactions to the child's pain behavior were significantly related with chronic pain status at baseline.

Table 5. Predictors of chronic pain at baseline

	Block	Predictor variable	$\beta$ (SE)	Wald $X^2$	Exp $\beta$ (Odds ratio)	95% CI Exp $\beta$	Block $X^2$	
<b>Child's predictors</b>	1	Age	0.201 (0.048)	17.300***	1.222	1.112-1.343	46.107***	
		Gender <sub>(1)</sub> <sup>a</sup>	0.734 (0.189)	15.056***	2.083	1.438-3.017		
	2	Pain catastrophizing	0.373 (0.124)	9.000**	1.452	1.138-1.852		
		Medical cure	-0.550 (0.238)	5.340*	0.577	0.362-0.920		
								13.999***

DP: 0=Non-chronic pain, 1=Chronic pain.

$N_{(\text{child's predictors})}=538$ . \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

<sup>a</sup> Codification: 1=Girl; 0=Boy

### 3.6. Predictors of onset of chronic pain at 1-year follow-up

In order to examine the role of the variables being tested on the onset of chronic pain at 1-year follow-up two subsamples were compared: children with onset of chronic pain at 1-year follow-up vs children without chronic pain at both assessment time moments. Table 4 displays the relationship of the onset of chronic pain at 1-year follow-up with each of the variables examined. Onset of chronic pain was associated with age, catastrophizing pain-coping strategies, average time spent in doing physical exercise or sports out of school, and number of stressful life events during the year preceding the follow-up assessment.

A forward stepwise logistic regression analysis was performed to determine which child's variables independently contributed to the onset of chronic pain at 1-year follow-up. In a first block, age was entered. In a second block, catastrophizing coping was entered. And, in a third block, physical activity index (i.e., time spent in doing physical exercise), and number of stressful events experienced in the previous year to the 1-year follow-up were entered. The results of this logistic analysis are shown in Table 6. A significantly higher odd ratio of chronic pain at 1-year follow-up was found for those children who tended to use catastrophizing strategies to cope with pain (OR=1.62, 95% CI= 1.03-2.54), for those who spent more time performing sports or physical activity out of school time at baseline (OR=1, 95% CI= 1.00-1.01), and for those who had experienced higher number of stressful events in the previous year to the follow-up (OR=1.20, 95% CI= 1.10-1.23).

As far as the parents' variables, again neither the presence of chronic pain nor reactions to the child's pain behavior were related to the onset of chronic pain at 1-year follow-up.

*Table 6. Predictors of development of chronic pain at 1-year follow-up*

	Block	Predictor variable	$\beta$ (SE)	Wald $X^2$	Exp $\beta$ (Odds ratio)	95% CI Exp $\beta$	Block $X^2$
<b>Child's predictors</b>	2	Pain catastrophizing	0.480 (0.232)	4.281*	1.616	1.026-2.547	7.860**
	3	Number of stressful events	0.177 (0.041)	18.781***	1.194	1.102-1.294	24.806***
		Time spent on sports	0.003 (0.001)	8.067**	1.003	1.001-1.005	

DP: 0=Non-development of chronic pain, 1= Development of chronic pain.  
 $N_{(\text{child's predictors})}=317$ . \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

### **3.7. Predictors of the course of chronic pain over one year**

To evaluate the relationship between the potential predictive factors examined and course of chronic pain problems over 1 year, two other subsamples (i.e., children with persistent chronic pain over 1 year vs children recovered from chronic pain at follow-up) were compared. The relationship between the course of chronic pain and child's and parents' variables collected at baseline are displayed in Table 4. Analyses revealed that persistence of chronic pain at 1 year was related with age, sex, number of body sites in pain, and pain related positive affect.

A logistic model was used to predict the course of chronic pain over 1-year. We followed a stepwise procedure, same as described above. The results are displayed in Table 7. Only age and number of painful sites were significant predictors. Older children who reported chronic pain at baseline were more likely to still report chronic pain at 1-year (OR=1.25, 95% CI= 1.07-1.48). Moreover, children who reported more generalized chronic pain at baseline were more likely to still suffer from chronic pain 1 year later (OR=2.77, 95% CI= 1.43-5.35).

Again, parents' variables did not exert any effect on the course of chronic pain problems.

*Table 7. Predictors of maintenance of chronic pain at 1-year follow-up*

	Block	Predictor variable	$\beta$ (SE)	Wald $X^2$	Exp $\beta$ (Odds ratio)	95% CI Exp $\beta$	Block $X^2$
<b>Child's predictors</b>	1	Age	0.227 (0.083)	7.456**	1.255	1.066-1.476	10.367**
	2	Number of painful sites	1.019 (0.336)	9.208**	2.771	1.435-5.353	11.363***

DP: 0=Non-maintenance of chronic pain, 1= Maintenance of chronic pain.  
 $N_{(\text{child's predictors})}=179$ . \* $p<0.05$ , \*\* $p<0.01$ , \*\*\* $p<0.001$ . The reported standardized  $\beta$ s are those from the final step in the regression analysis.

#### 4. Discussion

The results of this population-based study show that chronic pain in youth is a frequent problem (for similar results, see Perquin et al., 2000). In spite of being frequent, in concordance with previous studies (Magni et al., 1987; Guidetti and Galli, 1998; Mikkelsen et al., 1999; Perquin et al., 2003; El-Metwally et al., 2004), chronic pain at this development stage might be a very irregular condition because a significant percentage of children with chronic pain fully recover from their problem. The annual percentage of new cases with chronic pain in our sample is similar to the percentage of new cases occurring in adults, but the percentage of children who recover from chronic pain is higher (see Elliot et al., 2002 for comparisons). Longer-term follow-up studies are recommendable to further investigate this issue. The instability of the course of chronic pain problems in children population emphasizes the importance to study what factors contribute to the course of chronic pain in children.

We tested the influence of several factors in a schoolchildren sample. Occurrence of chronic pain at baseline was predicted by age, sex, pain catastrophizing, and expectancies about recovering from pain. Onset of chronic pain at 1-year was predicted by pain catastrophizing, level of physical exercise, and level of stressful life events. Finally, persistence of chronic pain was predicted by age and number of pain sites.

Some of our findings have already been reported in the literature but mainly in cross-sectional studies. For example, existing studies have shown that differences in pain prevalence rates are related to age and sex (e.g., Unruh and Campbell, 1999; Perquin et al., 2000; Merlijn et al., 2003), with older children and girls suffering more frequently chronic pain problems. These cross-sectional relationships have been confirmed in our study. However, we demonstrated, prospectively, that only age was a risk factor for the persistence of chronic pain. Previous works have also examined the role of a few characteristics of chronic pain on the onset and persistence of chronic pain (Flato et al., 1997; Perquin et al., 2003). Our research confirms that the co-occurrence of multiple pains is a risk factor for the persistence of pain (Flato et al., 1997). Moreover, we have examined for the first time the role of the severity of chronic pain problems described in terms of pain intensity and disability. Somewhat surprisingly this factor was not found to have any effect on the course of chronic pain problems at one year. The reduced number of participants with severe chronic pain problems might be the reason for the analysis not reaching statistical significant differences.

Physical exercise has also been examined as a factor related to the presence of recurrent or chronic pain (e.g. Vikat et al., 2000; Kovacs et al., 2003; Ghandour et al., 2004; Laurell et al., 2005; El-Metwally et al., 2006). As in Jones et al.'s (2003) study, our results showed that intense exercise out of school time exerts an effect on the future onset of chronic pain. This relationship might be explained by the fact that children who exercise to a high level might have an increased risk of serious injuries, and that can be considered a risk factor for future chronic pain problems. This relationship, however, is not consistent across studies (Feldman et al., 2001).

Our results are in agreement with those of Boey and Goh (2001), and support the American Pain Society recommendations (see APS home page Web: <http://www.ampainsoc.org/advocacy/pediatric.htm>), namely: adequate assessment of children in pain must include an evaluation of stressful life events, since it has shown to be a risk factor for chronic pain.

The only psychological factor which has been related with chronic pain is emotional-focused avoidance coping, which encompasses externalising and catastrophizing coping (Merlijn et al., 2003). However, our study is the first which examines longitudinally the effect of this and several other psychological factors related to pain on the course of pain complaints in children. Firstly, our findings are consistent with Merlijn et al.'s (2003) findings. Our results partially support the fear-avoidance model outlined by Vlaeyen et al. (1995). The model suggests that subjects with high levels of catastrophizing are supposed to have more fear-avoidance beliefs, and in turn more disability and depression that increases pain, a vicious circle that perpetuates the experience of pain. Our study showed that catastrophizing is also a predictor of the onset of chronic pain in children. However, the intermediate steps of the model should be tested in order to examine the applicability of the model to chronic pain in youth. Therefore, these results show the powerful effects of these pain-related cognitive processes only on the transition from acute to chronic pain problems. If these findings hold true in future studies, then health professionals should pay close attention, and intervene on these cognitive processes as soon as children start to complain about pain in order to stop the vicious cycle described above. Secondly, our results are also partially consistent with those from the adult literature on individual's expectancies of outcome (i.e., outcome about treatment results, see Goossens et al., 2005). A poor expectancy about recovering from pain predicted chronic pain problems at baseline, but it did not act either as a risk factor for the development of chronic pain at 1-year follow up nor as a risk factor of the persistence of chronic pain (at 1-year follow-up). Therefore, on the basis of our findings, it is unclear whether this cognitive factor is a cause or a consequence of chronic pain but in a period shorter than a year. On the one hand, adherence to medical prescriptions might be worse for those who do not believe in a medical solution for their problem, and it might have negative consequences on the results of the medical intervention that they are receiving. But on the other hand, the lack of positive results from medical intervention (i.e., recovery from pain) might lead to these negative expectancies.

The results of this study show that neither parents chronic pain problems nor parental reactions to the child's pain behavior did influence the course of the child's pain problems. Peterson and Palermo's (2004) hypothesis, that child's distress moderate the relationship between parental responses to recurrent pain and functional disability, might be applicable to other parents' factors as well as to other child's pain outcomes. That is, parents' reactions to child's pain behaviors or parents' chronic pain might act as a more important risk factor for chronic pain syndromes when children are distressed (Mikail and von Baeyer, 1990; Walker and Zeman, 1992, Osterhaus et al., 1998), but be much less important when we explore these issues in the general population, because they typically are non-distressed (see Szpalski et al.,

2002; Merlijn et al., 2003; Jones et al., 2004 for similar results). Detailed investigations on the role that parents reactions exert on children's pain experience are needed. Moreover, among healthy youth, the influential role of peers might be higher than the influence exerted by their parents, therefore it will also be interesting to investigate the influence of peers on children's pain experience (see Merlijn et al. 2003).

Our work provides some relevant data on factors related to the onset and persistence of chronic pain in youth, but it has some limitations. First, although the prospective design provides evidence regarding the effects of the factors, many variables have been explored without a specific guiding model. At this early stage of the research, these analyses are all of an exploratory nature. Thus, they do not help us in theoretically clarifying either the relationship between separate entities that underlie the processes involved in the chronicity of pain, or the overlap between the examined independent variables. Moreover, observation over longer time as well as careful attention to differences in developmental stages are necessary to understand the course of pain over the lifespan (for further discussion, see Walco, 2004). Second, all data were collected through self-report, and although this is an appropriate assessment procedure (McGrath, 1990) no other alternative methods to collect information were used; future studies should be based on a multimethod approach to pain assessment. Third, although it is interesting to consider chronic pain without taking into account its body location, we acknowledge that certain predictive factors might be specific for a pain complaint, studies examining a specific chronic pain syndromes are also necessary. Fourth, caution should be taken in the fact that IRPENDA was answered sometimes by mothers, sometimes by fathers, and sometimes by both, because father and mother may behave in different ways and these different reactions may exert different effects on the child's pain experience. Finally, the participation rate of parents was low, thus concerns regarding the generalization of the data about the role of parental factors arise.

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## DISCUSSION

The purpose of this thesis is to help gain a better understanding about pain problems among children and adolescents.

*In relation to the first main objective of this thesis*, the psychometric properties of three assessment instruments have been tested for their use with Catalan-speaking schoolchildren: 1) the *Faces Pain Scale-Revised* (FPS-R), 2) the *Pediatric Quality of Life Inventory* (PedsQL), and 3) the *Inventari de Respostes dels Pares davant el Dolor dels Nens i Adolscents* (IRPEDNA).

The Catalan version of the FPS-R has shown to be an appropriate measure to assess pain intensity in Catalan-speaking children, with psychometric properties similar to the original. The FPS-R-C has not only showed sound psychometric properties, but has also shown other practical characteristics: 1) it is feasible, and consequently it can be “easily” integrated for routinely assessment in a clinic setting; and 2) it is preferred by children and adolescents compared to the Coloured Analogue Scale. However, further study on the psychometric properties of the FPS-R-C is necessary. Specifically, its psychometrics properties should be tested in younger children (under 7 years of age), and for other painful conditions other than the ones in our study.

The PedsQL is a fundamental measure to assess treatment outcome in pain; it is widely used (Clarke and Eiser, 2004; Rajmil et al., 2004). Our results failed to replicate the original structure of the Catalan version of the PedsQL. A shorter 12 item-version was proposed instead, a version that showed sound psychometric properties. One limitation of this short version is the low internal consistency of some of the second-order scales. The development of new items to improve the internal consistency of these scales is needed. An interviewer-administered version was also developed in that study. Its psychometric properties are similar to the self-administered version. However, further studies are recommendable to examine the equivalence between reports provided by self- and interviewer-administered versions. If the non-equivalence was confirmed, then practitioners and researchers should use systematically the same version in order to be able to explore inter and intra-individual differences in the child's answers to the PedsQL. Our results need to be replicated in

clinical populations, and the temporal reliability of both versions needs to be addressed in future studies too.

Finally, IRPEDNA is a measure that we developed to assess the parental (adults) reactions to children's pain behavior. Preliminary psychometric properties support a three factorial solution and support a good validity and reliability. Further research is needed in order to test other psychometric characteristics (such as construct validity, test-retest reliability, and sensitivity).

*The second objective of this thesis* was specifically addressed to examine the impact of pain in this population. We have conducted a longitudinal epidemiological study in a sample of 8-16 year-old schoolchildren. This study provides an in-depth description of chronic pain among this general population. This study supports that pain is a very important health, social, and economical problem. The results corroborate that pain is a common experience amongst schoolchildren in our country, and for many of them the experience is of a chronic long-lasting nature: 88% of the population had experienced at least one pain episode in the preceding 3 months to our interview, and 37.3% of them reported chronic pain when they were interviewed. One year later, of the schoolchildren reporting chronic pain, 58.24% still reported chronic pain. And, of the schoolchildren pain free at baseline, 14.42% reported chronic pain. Those schoolchildren with chronic pain at baseline reported significantly lower levels of quality of life than those without chronic pain. Clearly, these results are the opposite of the extended belief in our society that pain is not a problem in children. Children in pain should receive special attention from clinicians, health researchers, and politicians.

These epidemiological data are in accordance with previous studies reporting about the situation in other parts of the world. However, the prevalence found here is slightly higher than in other reports (see for example, Perquin et al., 2000). Such a discrepancy might be a result of differences in the studies' design. On the one hand, our study was not restricted to a specific pain complaint (it was neither restricted to the most bothersome complaint nor to a specific location). And, on the other hand, the methods implemented to gather this information were also different. While in most of the epidemiological studies children have been requested to fill-out self-report questionnaires, in our study

children were individually interviewed by two widely trained interviewers. The interview may offer some advantages in front to self-report questionnaires. First, children's memories can be enhanced when they are interviewed; it is not easy for children to remember things as they happened in the previous three months (e.g., pain experiences). Second, children are more positively engaged in the task when they are interviewed. Finally, misunderstandings might be corrected if children are interviewed. However, we acknowledge the limitations associated to this method. Since it requires a lot of work, two interviewers were involved in the collection of data. This could be a bias source. In order to avoid it as much as possible, both interviewers were intensively trained and instructed to follow a standardized protocol very closely. Also, the presence of an interviewer might also influence the children's answers. Be as it may, however, it is uncertain whether different methods of collecting data produce different or equivalent data; such a controversy is still open to discussion (for further discussion see paper #2). Most of the studies available support that different methods of data collection can influence outcomes, but this conclusion is not always supported. Staes et al. (2000), for example, compared self-administration and face-to-face interview in the investigation of low back pain and they did not find too many differences.

Besides being interested in learning about the epidemiology of chronic pain, we were prompted to study differences among chronic pain cases: not every child reporting chronic pain suffers the same. We were able to observe that there are important differences in the impact of chronic pain in children, the extent to which chronic pain interferes with the children's life, as well as in their parents' lives. Thus, we decided to work on a grading system to rate the severity of chronic pain problems in children. We are confident that this grading system will be a positive asset in the study of the impact of chronic pain in peoples' lives. Although we acknowledge that it needs further work to assure the minimum standards of reliability and validity criteria, this grading system preliminarily points out that, in most cases, chronic pediatric pain is not severe (at least in our sample of schoolchildren). Most of the participating schoolchildren with chronic pain lead relatively normal lives, only 5% of them suffered from severe chronic pain.

One other important area of interest to us is *the secondary prevention of chronic pain and related disability*. That is, we thought about looking into the alternatives of helping children who are at greater risk for developing chronic disabling pain. We firmly believe that “an ounce of prevention is better than a ton of a cure”. We are well aware that the prevention of acute pain in some cases is not possible, and in others it may simply be not desirable (pain is often a sign of alarm, an indication that something is not right). But suffering, disability, and chronic pain, may be *optional* and could be prevented. This new conceptualisation would be not only beneficial for children with pain and their parents, but also would allow society to save a lot of personal and collective suffering, as well as a lot of money. These preventive strategies should not be applied to everybody, but to those children who are at risk. We are convinced that the best way to use the scarce resources available for the management of pain in children is to use them for prevention (that is, for secondary prevention of chronic pain and related disability and suffering), at least part of them. In this line, we are working in the identification of (1) predictive factors of disability in children, and (2) predictive factors of the onset (and persistence) of chronic pain.

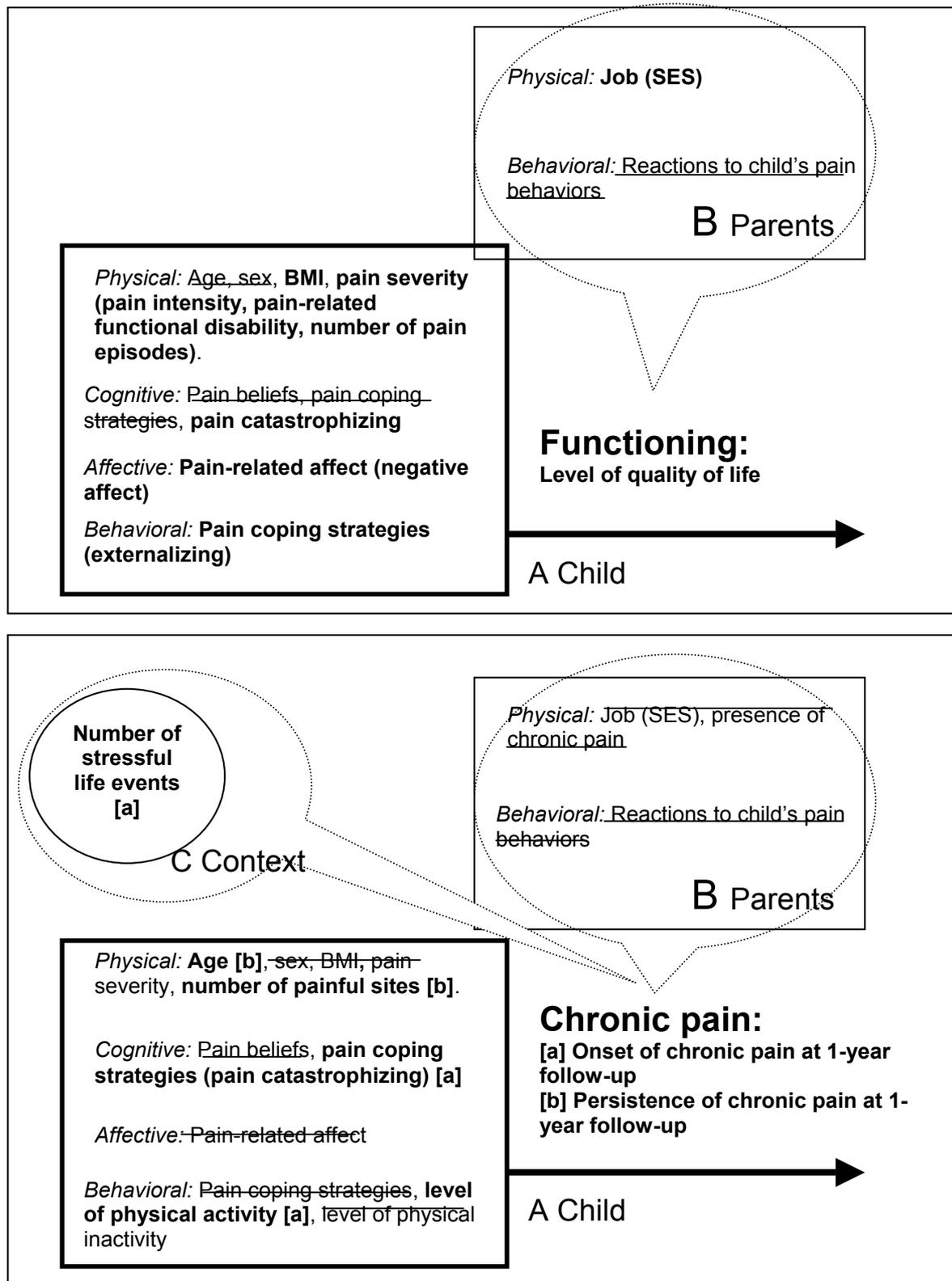
Therefore, after learning the point of view about the predictive factors of chronic pain and disability from a group of professionals with a special interest on chronic pain in childhood, we evaluated the role of a few of them; our work was based on a general framework based on the work of Miró (2003). As for the first issue related to predictive factors of children’s disability, we decided to start our search by using a *positive approach*. If we want to understand why a small amount of children develop disabling pain, whereas others do not, different alternative approaches can be pursued. For example, we can learn from the minority of children who often seek for help, as it is typically done in the literature, or we can learn from the majority of children who are able to function normally in spite of their pain. And we decided to start by following this second alternative, that is, studying children who are highly functioning despite of pain. In general terms, our results showed that pain related characteristics are not the only contributors that interfere children’s functioning, rather there are several socio-psychological factors playing an important role here too. More specifically, pain severity characteristics, and variables related to pain-related fear are

indicators of children's functioning (see *Figure 9*). These factors were evaluated, however, using cross-sectional data.

As for the second issue related to predictive factors of the onset and persistence of chronic pain, psychological factors are also relevant to explain the course of pain problems over time. Specifically, our results showed that age, pain catastrophizing, child's expectancies about a medical cure for pain, number of stressful events in the child's life, and a high level of physical exercise are predictors of the occurrence of chronic pain in the present and/or in the future (see *Figure 9*).

From a biopsicosocial perspective, pain is defined as a multidimensional experience that is influenced by individual, social and contextual factors (Craig and Riddell, 2003; Miró, 2003). A finding that was unexpected is that the examined parental factors were not related to either the children's pain experiences or his/her functioning.

In sum, the results from our studies related to two of our main objectives (the impact of pain, and the predictive factors of chronic pain and disability in children) demonstrated that pain needs to be addressed from a holistic approach. Only by working from such a position it will be possible to design effective strategies to prevent children from suffering chronic pain and disability.



Note. (1) Quality of life is being used here to illustrate the model, it has been chosen because among all the outcomes used this one might be the best indicator of functioning, besides we have shown the validity of the PedsQL too (paper #2).

**Figure 9. Model tested for the level of child's functioning and the onset persistence of chronic pain (based on the results presented in this thesis)**

There are several recommendations for future directions:

1. It is essential to keep working on providing appropriate assessment instruments for clinicians and researchers working with Catalan speaking children. Furthermore, we have to be able to help clinicians to incorporate those assessment instruments, that meet the validity, reliability and sensitivity highest standards, to their working routines. Only by using the appropriate instruments, that is, instruments adapted to our children population, clinicians will better understand and treat presenting pain syndromes.
2. Given the cross-sectional design in the study #6 (i.e., The influence of pain and pain related factors on the functioning of 8-16 year old children), caution in interpreting the direction of causality between child's functioning and identified "predictors" is advised. It is necessary a replication of the results in a prospective longitudinal study.
3. Miró's model although initially suggested for adult pain problems, might be useful to understand chronic pain in children and adolescents. Furthermore, it can be useful to understand the functioning of these children. However, we still are far away of filling in the gaps of the template, additional studies are needed in order to have a clear picture on both areas. Nowadays, research about the predictors of chronic pain and disability is emerging. Available studies are primarily exploratory, only a limited number of pieces in Miró's model have been analysed. An example of this type of research is part of the work presented into this thesis. Our research revealed a number of significant psychological factors related to pain. This new information along with already available information together with that from future exploratory studies is fundamental to start testing specific explicative models about certain mechanisms underling these relationships. For example, our results partially support the fear-avoidance model of chronic pain (Leeuw et al., 2007; Vlaeyen and Linton, 2000; Vlaeyen et al., 1995). However, additional empirical evidence needs to be collected to test this and other models. Besides, since relationships obtained through regression analysis are not enough to support theoretical models, it is necessary to validate these models using structural equation modelling techniques.

4. An instrument to assess the effects to parental reactions has been designed and tested. However, the results showed that parental reactions did not exert any influence in pain experience. This issue needs to be further investigated in clinical populations because, as it has been suggested by Peterson and Palermo (2004), parental reactions could be more important in those subjects with high levels of distress whereas parental reactions could have little influence in general population. IRPEDNA should be used with clinical samples to further our knowledge about its psychometric properties, and study the relationship between parents' responses and children's pain experiences. Moreover, other environmental influences, beyond parental reactions, need to be studied. Especially in teenagers, the influence of peers should be addressed because peers could act as models in this developmental stage.
5. In this attempt to understand chronic pain problems, we acknowledge that it is also important to investigate different types of pain (e.g., musculoskeletal pain, headache or abdominal pain) because there might be different mechanisms underlying different types of pain.
6. We have determined the value of several factors related to the development of chronic pain problems and level of functioning. A second step in the identification of children at risk should be to develop a screening questionnaire. It should be a brief questionnaire to be applied to all children with an acute problem to ascertain their risk to have a long-lasting pain problem and/or disability. Following the stepped-care approach suggested by authors like Linton et al. (2002), and Miró (1998), those children at risk should receive special attention. Preventive strategies should be based on knowledge gained in these studies, and programs addressed to reduce or eliminate risk factors, and promote and reinforce well known protective factors.
7. Following von Korff's Chronic Pain Grade Scale, we have presented a proposal that could be used as a brief and easy method to grade the severity of chronic or recurrent pain problems in children. This proposal could be very useful for surveys in the general population as well as for studies with pain patients receiving treatment. However, its psychometric

properties should be analysed further. Specially, it is needed to test this classification in a sample in which all severity grades are represented. In our epidemiological study most children with chronic pain only experienced mild chronic pain syndromes.

8. In order to understand better the development of pain and disability, it is necessary to conduct additional studies with clinical populations. Only by examining these problems in both contexts, general and clinical populations, we will be able to fully understand the complex puzzle of pain, and design effective preventive strategies.
9. Once predictors would be studied both in clinical and general populations, we will be able to design secondary preventive programs. The effectiveness of these programs should be tested in clinical trials.

The information gathered in this project lends us to highlight the following suggestions:

A) As far as children's functioning is concerned:

- (1) Respond promptly and adequately to their daily pain complaints in order to promote a better quality of life: pain relief should be always ensured.
- (2) Reduce variables related to pain-related fear: we should reduce the emotional distress of children when they are in pain, we should help the children gain control over strong emotions while in pain, and we should reduce their catastrophic thinking.

B) As far as the course of the children's pain is concerned:

- (3) Modify the child's catastrophic thinking in order to prevent pain experiences of becoming chronic.
- (4) Promote positive child's expectancies about a medical cure for pain.
- (5) Pay special attention to the older children, they are more prone to suffer persistent pain problems.
- (6) Take into account the environment around the child, and pay special attention to those who are suffering stressful life events: stressful environmental situations can be an obstacle for recovering from acute pain episodes.

(7) Pay special attention to the pain complaints of those who are highly engaged on physical activities.

Finally, it is our hope that health care professionals working in our country will be able to profitable use the assessment instruments presented into this thesis to understand, assess, and manage better pain problems and functioning in children.

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**Appendix 1. Potential predictive factors that have been under investigation for examining their role on either chronic pain or functioning (i.e. defined in terms of functional disability or quality of life) in children and adolescents**

UNIT	LEVEL	TYPE OF FACTOR	FACTORS	PREDICTOR OF CHRONIC PAIN (Occurrence, onset or persistence)	LEVEL OF FUNCTIONING
CHILD	PHYSICAL	Socio-demographic factors	Age	Bejia et al., 2005; Boey and Goh, 2001a; Brusgaard et al., 2000; Egger et al., 1998; El-Metwally et al., 2004, 2005, 2006; Feldman et al., 2001, 2002a, b; Flato et al., 1997; Ghandour et al., 2004; Harreby et al., 1999; Kristjansdottir and Rhee, 2002; Larsson and Sund, 2005; Jones et al., 2003b; Mikkelsen et al., 1998, 1999; Prista et al., 2004; Stahl et al., 2004; Szpalski et al., 2002; Vikat et al., 2000.	Gauntlett-Gilbert and Eccleston, in press; Logan et al., 2006; Konijnenberg et al., 2005; Logan and Scharff, 2005; Merlijn et al., 2006; Roth-Isigkeit, 2005; Tkachuk et al., 2003; Vervoort et al., 2005.
			Sex	Bejia et al., 2005; Boey and Goh, 2001a; Brusgaard et al., 2000; Feldman et al., 2001, 2002a, b; Flato et al., 1997; Egger et al., 1998; El-Metwally et al., 2004, 2005, 2006; Harreby et al., 1999; Hotopf et al., 1998; Jones et al., 2003b; Kristjansdottir and Rhee, 2002; Kovacs et al., 2003; Larsson and Sund, 2005; Mikkelsen et al., 1998, 1999; Prista et al., 2004; Sjolie, 2002; Stahl et al., 2004; Szpalski et al., 2002; Vikat et al., 2000.	Claar et al., 1999; Gauntlett-Gilbert and Eccleston, in press; Keogh and Eccleston, 2006; Konijnenberg et al., 2005; Logan et al., 2006; Lynch et al., 2006; Merlijn et al., 2006; Tkachuk et al., 2003; Vervoort et al., 2005.
			Ethnicity	Boey and Goh, 2001a; Ghandour et al., 2004; Laurell et al., 2005.	
		Anthropometrical factors and physical condition	BMI	Bejia et al., 2005; Feldman et al., 2002b; Harreby et al., 1999; Jones et al., 2003a; Kovacs et al., 2003; Sjolie, 2002; Szpalski et al., 2002; Vikat et al., 2000; Watson et al., 2003.	
			Weight	Bejia et al., 2005; Jones et al., 2003a; Sjolie et al., 2004b; Szpalski et al., 2002.	
			Height	Bejia et al., 2005; Feldman et al., 2002b; Jones et al., 2003a; Sjolie et al., 2004b; Szpalski et al., 2002; Vikat et al., 2000.	
			Growth		
			Mobility of joints	Harreby et al., 1999; Sjolie et al., 2004b; El-Metwally et al., 2004, 2005, 2006; Mikkelsen et al., 1998.	
			Strength of the part of body hurts or other parts of the body	Feldman et al., 2001.	
			Spine profile	Kovacs et al., 2003; Szpalski et al., 2002.	
			Different leg length	Kovacs et al., 2003.	
			Painful palpation of certain back sites	Szpalski et al., 2002.	
			Muscular flexibility	Feldman et al., 2001; Harreby et al., 1999.	
			Laboratory variables	Flato et al., 1997.	
			Wearing glasses or lenses	Vikat et al., 2000.	
			Puberty	Jones et al., 2003a; Laurell et al., 2005; Vikat et al., 2000.	

Health status	Physical fitness	El-Metwally et al., 2004, 2006; Kristjansdottir and Rhee, 2002; Sjolie, 2002.	
	General health	Flato et al., 1997; Szpalski et al., 2002.	Konijnenberg et al., 2005.
	Number of psychosomatic or physical symptoms	Crushell et al., 2003; Laurell et al., 2005; Vikat et al., 2000.	Clair et al., 1999; Konijnenberg et al., 2005; Tkachuk et al., 2003.
	History of injuries	Bejia et al., 2005; Kristjansdottir and Rhee, 2002; Laurell et al., 2005.	
	Recurrent illness	Vikat et al., 2000.	
	Comorbidity of chronic disease	Bener et al., 2000; Perquin et al., 2003; Kristjansdottir and Rhee, 2002; Vikat et al., 2000.	
	Fatigue	Boey and Goh, 2001a; El-Metwally et al., 2004, 2005, 2006, Kristjansdottir and Rhee, 2002; Mikkelsen et al., 1998, 1999; Szpalski et al., 2002; Watson et al., 2003.	
	Constipation	Boey and Goh, 2001a.	
	Nauseas	Boey and Goh, 2001a.	
	Physical functioning*		Konijnenberg et al., 2005.
	Referral status to pain specialized service		Konijnenberg et al., 2005.
	Emergency department visits because of pain		Tkachuk et al., 2003.
Mechanical factors	Right or left handed	Bejia et al., 2005.	
factors	Other pain complaints	Anttila et al., 2002; Boey and Goh, 2001a; El-Metwally et al., 2004, 2005, 2006; Flato et al., 1997; Jones et al., 2003b, Mikkelsen 1998, 1999; Stahl et al., 2004; Vikat et al., 2000; Watson et al., 2003.	
	Intensity	Perquin et al., 2003.	Kaskikar-Zuck et al., 2001; Gauntlett-Gilbert and Eccleston, in press; Konijnenberg et al., 2005; Logan et al., 2006; Logan and Scharff, 2005; Lynch et al., 2006; Merlijn et al., 2006; Odsson et al., 2006; Roth-Isigkeit et al., 2005; Tkachuk et al., 2003.
	Course of pain (continuous vs recurrent)	Perquin et al., 2003.	Merlijn et al., 2006.
	Localized vs generalized pain	El-Metwally et al., 2005.	
	Number of painful sites	Flato et al., 1997.	
	Duration of pain problem	Flato et al., 1997; Perquin et al., 2003.	Merlijn et al., 2006; Tkachuk et al., 2003.
	Frequency of pain episodes	El-Metwally et al., 2005; Larsson and Sund, 2005; Perquin et al., 2003.	Oddson et al., 2006; Merlijn et al., 2006; Peterson and Palermo, 2004; Roth-Isigkeit et al., 2005; Tkachuk et al., 2003.
	Duration of a pain episode	Larsson and Sund, 2005.	Gauntlett-Gilbert and Eccleston, in press; Konijnenberg et al., 2005; Logan et al., 2006; Logan and Scharff, 2005; Merlijn et al., 2006; Roth-Isigkeit et al., 2005.
	Pain diagnoses / Pain location	Perquin et al., 2003.	Konijnenberg et al., 2005; Logan et al., 2006; Merlijn et al., 2006.

EMOTIONAL		Frequency and duration of other pain	Larsson and Sund, 2005.		
		Pain threshold	Mikkelsen et al., 1999.		
		Trauma induced pain	El-Metwally et al., 2005.		
		Age at onset		Gauntlett-Gilbert and Eccleston, in press.	
		Yunus criteria	Mikkelsen et al., 1999. Mikkelsen et al., 1999.		
		Recent hospitalisation of the child himself or an immediate familiar member	Boey and Goh, 2001b		
	Psychological factors		Emotional problems	Jones et al., 2003a; Larsson and Sund, 2005; Perquin et al., 2003; Watson et al., 2003.	
			Depression	Anttila et al., 2002; El-Metwally et al., 2004, 2005, 2006; Larsson and Sund, 2005; Mikkelsen et al., 1998, 1999.	Gauntlett-Gilbert and Eccleston, in press; Kashikar-Zuck et al., 2001; Oddson et al., 2006.
			Psychiatric disorders	Egger et al., 1998.	
			Personality traits (i.e., neuroticism, introversion)	Hotopf et al., 1998.	
			Anxiety	Hotopf et al., 1998.	
			Psychological distress	Brusgaard et al., 2000; Feldman et al., 2001, 2002a, b; Jones et al., 2003a, 2004.	Logan et al., 2006; Schanberg et al., 2005.
			Emotional instability		Vervoort et al., 2005.
			Vulnerability		Merlijn et al., 2006.
			Stress		
			Social competence		Claar et al., 1999.
		Athletic competence		Claar et al., 1999.	
		Academic competence		Claar et al., 1999.	
	Functioning		Psychosocial functioning	Flato et al., 1997.	
			Social functioning*		Konijnenberg et al., 2005.
		General well-being	Sjolie, 2002.		
		General happiness	Sjolie, 2002; Szpalski et al., 2002.		
	Calmness	Sjolie, 2002.			
Psychological factors related to pain		Locus of control			
		Positive self-statements		Kashikar-Zuck et al., 2001.	
		Cognitive distraction		Kashikar-Zuck et al., 2001.	
	Internalizing / Catastrophizing		Kashikar-Zuck et al., 2001; Lynch et al., 2006; Vervoort et al., 2005.		

	Pain-specific anxiety		Gauntlett-Gilbert and Eccleston, in press.
Health-related behaviors	Smoking	Bejia et al., 2005; Feldman et al., 1999, 2001, 2002a, b; Ghandour et al., 2004; Harreby et al., 1999; Kovacs et al., 2003; Kristjansdottir and Rhee, 2002; Vikat et al., 2000.	
	Practise or not practise of physical activity	Ghandour et al., 2004; Szpalski et al., 2002.	
	Level of frequency of physical activity	El-Metwally et al., 2004, 2005, 2006; Feldman et al., 2001, 2002a, b; Harreby et al., 1999; Jones et al., 2003b; Kovacs et al., 2003; Kristjansdottir and Rhee, 2002; Laurell et al., 2005; Mikkelsen et al., 1998; Prista et al., 2004; Sjolie 2004a; Vikat et al., 2000; Watson et al., 2003.	
	Level of physical education at school		
	Intensity of physical activity	Vikat et al., 2000; Kristjansdottir and Rhee, 2002.	
	Sleep	Kristjansdottir and Rhee, 2002; Szpalski et al., 2002; Laurell et al., 2005; El-Metwally et al., 2004, 2005, 2006; Mikkelsen et al., 1998, 1999; Brusgaard et al., 2000	
	Use of alcohol or other drugs	Ghandour et al., 2004; Kristjansdottir and Rhee, 2002; Kovacs et al., 2003; Laurell et al., 2005; Vikat et al., 2000.	
	Caffeine intake	Ghandour et al., 2004; Kristjansdottir and Rhee, 2002.	
	Eating habits	Bener et al., 2000; Kristjansdottir and Rhee, 2002; Laurell et al., 2005.	
	Regular meals	Kristjansdottir and Rhee, 2002	
	Fast food	Kristjansdottir and Rhee, 2002	
Functioning	Prosocial behaviors	Jones et al., 2003a; Watson et al., 2003	
	Behavior problems	Jones et al., 2003a; Larsson and Sund, 2005; Watson et al., 2003.	
	Hiperactivity	Jones et al., 2003a; Watson et al., 2003.	
Life-style	Time involved in computer games, TV watching	Bejia et al., 2005; Bener et al., 2000; Jones et al., 2003b; Kovacs et al., 2003; Kristjansdottir and Rhee, 2002; Laurell et al., 2005; Prista et al., 2004; Sjolie, 2004a; Szpalski et al., 2002; Watson et al., 2003.	
	Time spent outdoors	Laurell et al., 2005 Hotopf et al., 1998	
	Time playing with friends	Laurell et al., 2005.	
	Having a job	Feldman et al., 2002b; Jones et al., 2003a; Watson et al., 2003.	
	Number of hours worked	Feldman et al., 2001, 2002a; Jones et al., 2003a; Kristjansdottir and Rhee, 2002.	
Mechanical factors		Bejia et al., 2005; Feldman et al., 2002b; Vikat et al., 2000.	
	Type of work (e.g. heavy work)	2003a	
	Playing a musical instrument	Feldman et al., 2002b.	
Pain factors related to pain	Functional disability	Sund, 2005; Laurell et al., 2005; Mikkelsen et al., 1998, 1999; Perquin et al., 2003;Stahl et al., 2004.	Gauntlett-Gilbert and Eccleston, in press.

		variables related to pain	Taking pain medication	Laurell et al., 2005; Stahl et al., 2004; Szpalski et al., 2002. Laurell et al., 2005; Perquin et al., 2003; Szpalski et al., 2002.		
			Approach coping		Keogh and Eccleston, 2006; Merlijn et al., 2006.	
			Problem focused avoidance coping		Keogh and Eccleston, 2006; Merlijn et al., 2006.	
			Emotional focused avoidance coping			
			Information seeking		Kashikar-Zuck et al., 2001.	
			Problem solving		Kashikar-Zuck et al., 2001.	
			Seeking social support		Kashikar-Zuck et al., 2001.	
			Behavioral distraction		Kashikar-Zuck et al., 2001.	
			Externalizing		Kashikar-Zuck et al., 2001.	
			Strive to rest and be alone			
PARENTS	PHYSICAL	Socio-demographic factors	Parents occupation	Anttila et al., 2002; Boey and Goh, 2001a, Hotopf et al., 1998; Kristjansdottir and Rhee, 2002; Larsson and Sund, 2005; Laurell et al., 2005; Vikat et al., 2000.		
			Parents' educational background	Bener et al., 2000; Boey and Goh, 2001a, ; Flato et al., 1997; Ghandour et al., 2004; Vikat et al., 2000.		
		Health status	Parents fatigue		Reid et al., 1997.	
			Parental health	Hotopf et al., 1998.		
	EMOTIONAL	Functioning	Parental pain	Boey and Goh, 2001a.	Lynch et al., 2006; Logan et al., 2006; Merlijn et al., 2006.	
				Ghandour et al., 2004; Kristjansdottir and Rhee, 2002; Vikat et al., 2000.		
		Psychological factors	Parental psychological distress		Gauntlett-Gilbert and Eccleston in press; Logan and Scharff, 2005.	
			Parental stress		Gauntlett-Gilbert and Eccleston, in press.	
			Parents depression			
			Parents anxiety	Hotopf et al., 1998		
			Personality traits (i.e., neuroticism)			
			Adolescent-parent distressful relationship			
		COGNITIVE	factors related to the child's pain	Belief in the helpfulness of a psychiatrist consultation		
				Belief in the helpfulness of a medical consultation	Crushellet al., 2003.	
	Belief in a physical cause of the child's pain			Crushellet al., 2003.		
	BEHAVIORAL	Psychological factors related to the child's pain	Parents rewarding		Merlijn et al., 2006.	
		Psychological factors related to the parents' pain	Approach coping			
			Problem focused avoidance coping			

			Emotional focused avoidance coping		
FAMILY	Socio-demographic factors		Family income	Boey and Goh, 2001a.	
			Family composition	Vikat et al, 2000.	
			Residence (i.e., level of urbanization)	Jones et al., 2003b; Kristjansdottir and Rhee, 2002; Laurell et al., 2005; Prista et al., 2004; Vikat et al, 2000.	
			Housing condition	Laurell et al., 2005.	
	PHYSICAL	Health status		Family size	Logan and Scharff, 2005.
				Recurrent disease or health complaints in family members (i.e., colds)	Hotopf et al., 1998; Laurell et al., 2005.
				Chronic illness in family	Boey and Goh, 2001a; Perquin et al., 2003.
			Parents health		
	Stressful events		Family members with pain	Anttila et al., 2002; Flato et al., 1997; Bejia et al., 2005; Bener et al., 2000; Laurell et al., 2005; Jones et al., 2004; Perquin et al., 2003; Szpalski et al., 2002.	
			Death of a family member		
			Change in occupation of an immediate family member		
				Boey and Goh, 2001b.	
					Logan and Scharff, 2005.
					Gauntlett-Gilbert and Eccleston, in press.
	EMOTIONAL			Bener et al., 2000.	
			Unhappy home		
	BEHAVIORAL	Pain factors related to pain	Impact of pain on family	Perquin et al., 2003	
			Family conflicts	Logan and Scharff, 2005.	
			Family independence	Logan and Scharff, 2005.	
			Family functioning	Gauntlett-Gilbert and Eccleston, in press.	
SCHOOL	PHYSICAL	Mechanical factors	School satchel weight	Bejia et al., 2005, Jones et al., 2003a; Szpalski et al., 2002; Watson et al., 2003.	
			Method of carrying schoolbag (type of schoolbag)	Jones et al., 2003a; Kovacs et al. 2003; Watson et al., 2003.	
			Journey home-school	Bejia et al., 2005; Szpalski et al., 2002.	
			Walking minute to school	Prista et al., 2004.	
	Stressful events		Change of address or school	Boey and Goh, 2001b.	
			Beginning of academic year	Bener et al., 2000.	

OTHER CONTEXTUAL UNITS (e.g. peers, work)	EMOTIONAL		Happiness to school	Bener et al., 2000; Laurell et al., 2005.	
		BEHAVIORAL		Examinations and much homework	Bener et al., 2000; Kristjansdottir and Rhee, 2002; Laurell et al., 2005.
			School problems (i.e., disobedience, being a disciplinary problem in class, lying)	Bejia et al., 2005; Hotopf et al., 1998.	
			School performance (failure)	Bejia et al., 2005; Boey and Goh, 2001b; Hotopf et al., 1998; Kovacs et al., 2003; Kristjansdottir and Rhee, 2002; Laurell et al., 2005; Vikat et al, 2000.	
			School absence	Hotopf et al., 1998.	
	PHYSICAL	Stressful events		Hot climate	Bener et al., 2000.
				Strong light	Bener et al., 2000.
				Loud noise	Bener et al., 2000.
				Childcare	Feldman et al., 2002b.
	EMOTIONAL	Functioning		Friends support	Ghandour et al., 2004; Kristjansdottir and Rhee, 2002; Laurell et al., 2005.
				Adults support	Bener et al., 2000; Ghandour et al., 2004; Kristjansdottir and Rhee, 2002.
	BEHAVIORAL	Functioning		Peer problems or bullying	Boey and Goh, 2001b; Ghandour et al., 2004; Jones et al., 2003; Laurell et al., 2005; Watson et al., 2003;
				Psychological factorss related to the child's pain	Peers rewarding Merlijn et al., 2006.

The studies cited in this table meet all the following inclusion criterion: (1) the study population consisted of children aged between 8 and 16 years old without any cognitive handicap; (2) the study population concerned children with idiopathic chronic pain (e.g. recurrent abdominal pain, musculoskeletal pain); (3) the article assessed any psychosocial prognostic factor; (4) the outcome was assessed through self-report measures or objective measures; (5) the intent of the analysis is prediction and consequently the relationship between potential prognostic factor and outcome was explored through regression analyses; (6) the article was published in English or Spanish from January 1996 to March 2007.