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**COPING AND ADJUSTMENT IN
CHILDREN'S PAIN**

**Processes of adaptation to illness and
develop effective
interventions for pain management**

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To my wife Valentina

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INTRODUCTION

My PhD project mainly focused on understanding how a child or adolescent copes with pain resulting from a disease, intended in a broader sense (i.e. procedures, treatments and disease-related).

The rationale behind the development of this research was to provide a workable protocol to facilitate measures for clinical practice in pain management: not only in first assessment, but also throughout the disease.

The “Fondazione Trentina per la Ricerca sui Tumori - (FTRT)” and the “Associazione Italiana contro le Leucemie-Linfomi e Mieloma - (AIL sezione Trentino), generously funded this project and allowed, with their support, a further clinical outcome: the planning of an effective training module for Health Professionals on non-pharmacological techniques for pain management.

The starting point is that pain-coping skills in a child’s repertoire may mediate the impact of pain perception on a particular child’s adjustment (Siegel & Smith, 1989). Given that pain experience is multidimensional and dynamic rather than linear, (it seems crucial to understand etc.) understanding the individual differences observed in children’s response to pain within a broader theoretical framework beyond the well-known cognitive-behavioral’s one, seems crucial.

As Mikail, Henderson & Tasca (1994) remind us, cognitive-behavioral theory differs from psychoanalytic theory in that it focuses on current internal processes, through a more descriptive stance of intrapsychic functioning compared to psychoanalytic theory’s interpretive-dynamic framework. By moving between two big approaches aimed at facing illness and pain, I tried to combine the constructs underlying these theories guide and develop an integrative conceptual framework.

This doctoral thesis tried to prove an innovative perspective that can help understand the wide variation in children’s pain experience, by considering intra-interpersonal influences and contextual factors, although the latter appear to remain secondary to the intrapsychic factors that focus on needs, defenses, and self-structure.

Overall, the whole project involved three pediatric units in Italy: the pediatric wards of Trento and Rovereto hospitals and the pediatric clinic of San Gerardo hospital, Monza (Milan).

Four are the chief goals I tried to achieve:

1. Providing a selective overview on current relevant topics in the pediatric pain research and state of the art regarding the existing models of pediatric pain. I then concluded with a preliminary conceptualization of a new theoretical model (chapters 1-2);
2. Developing a multi-dimensional protocol with an intra-method design for the assessment of pediatric pain in several chronic illnesses, by using also a battery of projective tests (drawings) to screen the emotional adjustment (chapter 3);
3. Validating the protocol by extending the methodology of projective drawings' scoring with a control group and adding other assessment variables on a single and larger cohort of patients to test the new model that I developed. Quantitative analysis phase preceded qualitative analysis phase within the same framework to yield a parallel mixed analysis (chapter 4);
4. Planning specific training modules about pain management, starting from a bottom-up process concerning the local health professionals' needs. I investigated these training needs through a series of open-ended questions, analyzed by a thematic analysis method. The modules' focus should be on psychological treatments available for pediatric population, which range from procedural pain to chronic pain, placing special emphasis upon interventions for which a credible evidence base exists (chapter 6);

No less important is the secondary aim related to the skills that I have gained during my research externship in the United States. I was involved in a clinical research project to evaluate treatment's feasibility, acceptability, and satisfaction of a problem-solving skills training for parents of children who have received an intensive pain rehabilitation from one pediatric pain rehabilitation program (Seattle Children's Hospital). I provided a methodological contribute within the mixed-method approach (statistical analysis and grounded theory) that was used to assess treatment acceptability and satisfaction of the program (chapter 5).

In the concluding section, the results are presented all together and their implications are discussed in a clinical perspective, since the rationale of this dissertation is that effective pain assessment must be multidimensional and at the same time feasible and practical. Furthermore, as Petovello (2012) claims in her review of literature, pain management should be multidisciplinary, proactive, anticipatory, and formed to meet each pediatric patient's needs.

This thesis also includes part of the following papers:

Chapter 3

Failo, A., Beals-Erickson, S. E., Venuti, P. Coping strategies and emotional profiles in children with disease-related pain. *Submitted to European Journal of Pain*.

Chapter 4

Failo, A., Nichelli, F., Venuti, P., Jankovic, M. Cope with pain in children/adolescents with malignant hematologic cancers. *In manuscript*.

Failo, A., (2015). Can drawings assist us in understanding better children's pain? *Pain News*, 13, 235-238.

Chapter 5

Law, E. F., Fales, J. L., Beals-Erickson, S. E., Failo, A., Logan, D., Randall, E., Weiss, K., Durkin, L. M., & Palermo, T. M. (2016). A Single-Arm Feasibility Trial of Problem-Solving Skills Training for Parents of Children with Idiopathic Chronic Pain Conditions Receiving Intensive Pain Rehabilitation. *Journal Of Pediatric Psychology*, 1-12.

CHAPTER 1

PSYCHOLOGICAL ASPECTS OF PEDIATRIC PAIN: CURRENT DEVELOPMENTS AND NEW PROSPECTIVES

*“Children’s pain matters — for the child, for the family,
and for society”*

~ IASP

This chapter is focused on a selective overview on the rationale for psychological approaches to assessment and treatment of pediatric pain, current developments and some new prospective.

Overall, I place these topics in the clinical practice’s context, which is the start-off point for this thesis.

Specifically, I will examine the following topics. The first issue is: why managing pain in children matters? In this respect, we will consider acute-procedural pain, disease-related pain and chronic pain, which are factors influencing the expression of pain and the short and long-term effects of unrelieved pain on children, from pain-applicable psychodynamic theories standpoint.

1 Why children’s pain matters

Of all the problems that children with a serious illness face, pain is often the most feared one. Pain is one of the most misunderstood, under diagnosed, and under treated/untreated medical problems, particularly in children. In fact, recent epidemiological studies still suggest that among hospitalized children the moderate to severe pain rates are high (27% to 94%) (Birnie, McGrath, & Chambers, 2014; Groenewald, Rabbitts, Schroeder, & Harrison, 2012).

Of course, pain is a universal experience all humans share but it is unique in how it is expressed. This seems to be even more important with the pediatric patients because the pain expression in children is often mediated due to difficulties in conveying their discomfort (Birnie et al., 2014; Kortetluoma & Nikkonen, 2006). Pain intensity is the most investigated marker of clinical significance in pain research and practice but it is one facet of the pain experience (Birnie et al., 2014, Turner & Turk, 2008). Actually, this is not the whole picture. Pain experience is also influenced by several dynamic

factors close to the children's world, including family, community and culture (Birnie et al., 2014, Twycross & Finley, 2013).

The current body of literature reflects many of the challenges in managing pain. Despite the plentiful knowledge and development of several effective evidence-based strategies to manage children's pain, there is a sharp gap between knowledge and practice. Actually, what we know is not being effectively implemented and a number of myths in this area of practice partially continue to exist (Petovello, 2012; Twycross, Dowden, & Stinson, 2014, Twycross, 2007).

Furthermore, in childhood, the powerful influence of parents in pediatric pain as well as the dynamic interplay between individual (e.g. child age, sex, psychological characteristics), dyadic (e.g. parent-child interactions), and contextual (e.g. family environment) factors in influencing child pain have been widely shown (Palermo & Chambers, 2005).

While the total eradication of pediatric pain may not be realistic (Walco et al., 1994; Pate et al., 1996) there is, however, an urgent need to improve children's pain management in the clinical practice, through strengthening children's and caregivers' ability to fight potential deleterious consequences as they may contribute to their disability and suffering later in life (Palermo, Valrie, & Karlson, 2014; Petovello, 2012; Walco et al., 2003).

2 Looking for definition of pain

Pain is now acknowledged as a complex psychological experience and it is generally accepted that infants and children can feel pain (McGrath, 2005). People use the term pain in relation to their traumatic experiences, usually subjective. Plus, being considered as an unpleasant sensation in one or more parts of the body pain is an emotional experience (Menossi, Lima, & Corrêa, 2008). On the other hand, speaking pain without injury, Ramachandran & Blakeslee (1998) maintain that "Pain is an opinion on the organism's state of health rather than a mere reflective response to an injury" (Ramachandran & Blakeslee, 1998, p.54).

According to a conceptualization shared by several researchers "Pain perception in children reflects the complex moment-to-moment integration of affective, behavioral, cognitive and physiological components within a developmental trajectory and sociocultural context" (Lioffi, 2006, p.2). This is coherent with the widely endorsed definition of pain promulgated by the International Association for the Study of Pain (IASP) describing it as "An unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage or described in terms of such damage. Pain is always subjective. Each individual learns the application of the word through experiences related to injury in early life" (IASP, 1979, p.249). As Twycross, Downen, & Stinson (2014) remind us, the original IASP definition has neglected those unable to communicate verbally, including neonates, young children and cognitively impaired children. This issue was

subsequently addressed and this amendment was made: “The inability to communicate in no way negates the possibility that an individual is experiencing pain and is in need of appropriate pain-relieving treatment” (IASP, 2001, p.2, as cited by Twycross, Downen, & Stinson, 2014). One notable feature of this overall and shared definition is the observation that pain is associated with potential damage, leaving room for multiple causes, mediators and moderators.

3 Several meanings behind the term “pain”

Children and adolescents experience pain from a number of different sources and reasons. From “pain” definition we should be able to recognize and address all types of pain.

A useful classification in this sense and, in my opinion, pretty clear as an overall overview, has been proposed by Varni, Blount, Waldron, & Smith (1995) that subdivided into four categories the types of pain that children may experience: (a) pain associated with chronic diseases (e.g. arthritis, sickle cell disease, cancer); (b) pain related to observable physical injuries or traumas (e.g. burns, lacerations, fractures); (c) pain not associated with a well-defined or specific chronic disease or identifiable physical injury (e.g. migraine and tension headaches, recurrent abdominal pain syndrome); and (d) pain associated with medical and dental procedures (e.g. lumbar punctures, bone marrow aspirations, surgery, injections, extractions).

Among the different types of pain, acute pain is one of the most common adverse stimuli experienced by children, which is associated with increased anxiety, avoidance, somatic symptoms, and also increased parent distress (Srouji, Ratnapalan, & Schneeweiss, 2010). Acute pain experiences subside with physical recovery, but they may involve also a complex interaction of biological, psychological and social processes thus becoming chronic (Katz, McCartney, & Rashiq, 2008).

An area that has gained more attention is that of procedural pain, an acute pain category. Diagnostic and monitoring procedures are the most feared and painful events in pediatric population (Cummings, Reid, Finley, McGrath, & Ritchie, 1996; Kortessluoma & Nikkonen, 2004; Lioffi, 2002; Petovello, 2012, as cited by Cummings, 2015).

Needle-related procedures (vaccine injections, venipunctures and venous cannulation) are the most common source of procedural pain and distress that children encounter (Uman, Chambers, McGrath, & Kisely, 2008, Ortiz, López-Zarco, & Arreola-Bautista, 2012).

In their review of eight Canadian pediatric hospitals, Stevens et al. (2012) on a sample of 3822 children found that more than 78% of them underwent painful procedures over 24-hour period. Procedure-related pain usually refers to a single episodes, but it can become recurrent when occurs at least three times over a 3 months period and interferes with daily activities (von Baeyer & Walker, 1999). In addition, it might become a kind of chronic pain or a disease-related pain condition as well.

Although no real consensus exists on its definition, chronic pain is typically defined as pain persisting longer than three months or beyond the expected healing time (Merskey & Bogduk, 1994; Treede et al., 2015). Chronic pain in children is significantly impactful on most domains of life and can occur continuously or on a recurrent basis. By comparing data across studies, we see that the pain frequency that is required for being considered as recurrent varies considerably from about once a month (von Baeyer, 2007) up to at least once a week (Riddell, Racine, Craig, & Campbell, 2014).

The prevalence rates of pain during childhood and adolescence vary greatly and are difficult to estimate because of the lack of congruence and especially because chronic pain depends on pain frequency.

Epidemiological studies show that 11-40% of children and adolescents experience chronic or recurrent pain, with numbers increasing over time (King et al., 2011; Stanford, Chambers, Biesanz, & Chen, 2008). Also, approximately 27% of community-dwelling children and adolescents report current chronic pain (Fuss, Pagé, & Katz, 2011) and a small but significant portion of the youth with chronic pain (3-5%) are severely disabled by their pain problem (Huguet & Miro, 2008).

Recent studies have shown that mild to moderate intensity pain is common in children with JIA and occurs on a weekly basis for several children (Anthony & Schanberg, 2003; Schanberg, Anthony, Gil, & Maurin, 2003, as cited by Hoff, Palermo, Schluchter, Zebracki, & Drotar, 2006). Research indicates that approximately 25 % of patients in pediatric rheumatology clinics are diagnosed with a pain syndrome such as fibromyalgia, complex regional pain syndrome, localized pain syndrome, or low back pain. Disease-related pain is still the primary cause of hospitalization for children with sickle cell disease (Dobson & Byrne, 2014). Also, the clinical manifestations of “growing pains”, are the most common forms of episodic childhood musculoskeletal pain (Gill et al., 2012). However, considerable inter-individual variability has been observed in relation to pain frequency and intensity as well as limitations extent in daily functioning experienced regardless of disease type or severity (Hagglund, Schopp, Alberts, Cassidy, & Frank, 1995; Ilowite, Walco, & Pochaczewsky, 1992, as cited by Hoff et al., 2006). It should be reminded that chronic pain experience occurs within a complex biopsychosocial framework, with an emphasis on social context (Simons & Basch, 2016).

4 Individual differences

Individual differences in pain experiences are crucial in pain diagnosis and treatment. The pain response is individual and learned. Evidence exists for genetically determined factors and environmental influences (Franck, Greenberg, & Stevens, 2000; Ross & Ross, 1989, as cited by Young, 2005).

The Diagnostic and Statistical Manual for Primary Care (DSM-PC), Child and Adolescent Version (American Academy of Pediatrics, 1996) claims that the pain stimulus is interpreted based on context

or pain meaning to the individual, as well as individual's psychological state, culture, previous experience, and a host of other psychosocial variables. Indeed, the same noxious stimulus may cause different amounts of pain in different individuals based on personal characteristics.

Although the principles of pain evaluation and management apply across human lifespan, infants and children present unique challenges that necessitate consideration of the child's age, developmental level, cognitive and communication skills, previous pain experiences, and associated beliefs (Morton, 1997). As Young (2005) affirmed, some pain response determinants are fixed (e.g. sex, temperament), some may be changed over time (e.g. anxiety, pain coping style), and some may be modified just before or during the procedure (e.g. procedure environment, parental interactions, type of interventions).

Conversely, in childhood-related chronic pain conditions the interrelations between individual factors (physical, affective, cognitive) and social factors that influence pain and related disability are highlighted (Palermo & Chambers, 2005; Palermo, 2012). In fact, children's psychological functioning (e.g., coping, mood, anxiety) can be both chronic pain contributing factor as well as an outcome and can be predictive of chronic pain maintenance in adulthood (Horst et al., 2014).

4.1 The roles of age, developmental stages, sex and gender

Age was recognized as a consistent key contributor to children's pain experiences in the literature (Blount, Piira, & Cohen, 2003; Czarnecki et al., 2011; Karakaya & Gözen, 2016; Kortessluoma & Nikkonen 2006; Young, 2005), but it is also necessary to take child's developmental stage into consideration because age is strongly correlated with development and both affect a child's ability to understand and cope with a painful event (Young, 2005). Even children's definitions of pain tend to follow developmental patterns: by school-age years (age 5/6) children are able to provide significant information on their pain to caregivers and health care providers (Azize, Humphreys, & Cattani, 2011, as cited by Palermo, Valrie & Karlson, 2014; Craig, McMahon, Morison, & Zaskow, 1984; Ross & Ross, 1988, as cited by Gedaly-Duff, 1991). More complex are pain assessment and management in children with developmental delay due to misinformation and preconceptions regarding their capability to feel pain (Boerner, Gillespie, McLaughlin, Kuttner, & Chambers, 2014). Some studies have shown that these children have a diminished observable response to painful events and engage in less attention- or help-seeking behaviors when they experience pain (Gilbert-MacLeod, Craig, Rocha, & Mathias, 2000; Oberlander, Gilbert, Chambers, O'Donnell, & Craig, 1999, as cited by Boerner et al., 2014). Despite these differences between different developmental trajectories, it is well known that pain experienced during infancy can have long-lasting consequences across the lifespan (Grunau & Tu, 2007).

Children's ability to report on past pains may be influenced by developmental factors (age, cognitive ability) within other important variables such as contextual ones (mood state, language used by

significant others), affective and pain-related factors (Jaaniste, Noel, & von Baeyer, 2016). Indeed, several studies have shown that retrospective worst pain ratings are both difficult and influenced by current pain (e.g. Jensen, Wang, Potts, & Gould, 2012; Stone, Broderick, & Schwartz, 2010). In addition, they may be influenced by age: in their study, Zonneveld, McGrath, Reid, & Sorbi (1997) found that older children had a more accurate recall of their worst pain intensity compared to younger children.

Both experimental and clinical pain studies showed that age can be also considered as a moderating factor in pain perception due to the included age range (9–17), which covers the transition from childhood to adolescence, commonly associated also with the emergence of sex differences in pain (Fillingim et al., 2009). For example, a study of Tsao et al. (2013) on conditioned pain modulation (CPM), conducted with a sample of 133 healthy children (aged 8–17 years), showed that older children (12–17 years) exhibited greater CPM compared to younger children (8–11 years). A study of Schmitz, Vierhaus, & Lohaus (2013), conducted with a sample of 1021 healthy children (aged 9-17) and using the cold pressor task has attempted to investigate pain tolerance in children and adolescents separately. The authors found that female and male adolescents may develop their pain tolerance in different directions when reaching puberty and up to the age of 14, the pain threshold and endurance increase in boys and girls are similar.

Given that both healthy and chronically-ill children underwent several needle procedures as part of routine medical care including vaccine injections, venipunctures and venous cannulations, needle-related pain can be considered as a pain “prototype” because it is one of the most commonly occurring and feared procedures (Uman et al., 2013; Taddio & McMurtry, 2015).

Qualitative data provided by Kortetluoma & Nikkonen’s (2004) study involving 4- to 11-year-olds’ experiences of venipunctures for blood sampling, provides valuable insight into children’s perceptions of pain. One of their primary findings was the clarity with which these children were willing and able to talk about their pain experiences. Children primarily used sensory words to describe their pain, and metaphors to explain their experiences. A meta-analysis on effects of distraction on children's pain and distress during medical procedures in children aged 3 to 15 years shows that younger children report and exhibit greater anxiety and pain than older children (Kleiber & Harper, 1999).

The developmental processes consideration is even more important in the chronic pain conditions to guide understanding of children’s experiences and reactions to pain or treatment efforts. Actually, recurrent pain may interrupt normal developmental processes and the parental factors may differentially influence adjustment to chronic pain during distinct developmental periods and in the transition from pediatric to adult health care (Palermo, Valrie, & Karlson, 2014). Several studies showed that chronic or recurrent pains most often occur in adolescence rather than in childhood

(e.g. McKillop & Banez, 2016; Sperotto, Brachi, Vittadello, & Zulian, 2015) suggesting the potential relationship between pain and developmental stage.

Developmental factors are unique to the experience of pediatric pain and are also related to social factors that emerge or are consolidated around the time of puberty (Caes et al., 2016; Unruh, 1996). To explain social factors' relevance in the pain experience context, gender can be viewed as a central explanation. Indeed, gender socialization theories have suggested that through modeling and reinforcement, children learn gender-appropriate pain responses, and that identifying strongly with a gender role is associated with more gender-conforming pain behaviors (Myers, Riley, & Robinson, 2003; Pool, Schwegler, Theodore, & Fuchs, 2007 as cited by Boerner, 2016).

Nevertheless, the research literature on the role of sex (biological/physiological characteristics) and gender (behaviors considered socially appropriate) in children's pain perception shows mixed results. Earlier works showed that sex and gender shaped the experience of pain while a few recent studies showed that these results are not completely shared (Unruh, 1996; Berkeley, 1992, 1997; LeResche, 1997, as cited by Moon & Unruh, 2013).

For example, in a longitudinal study no differences were found between boys and girls in cortisol level during infancy and toddlerhood (Grunau et al., 2004, 2007, as cited by Grunau, 2013) but in another longitudinal study, girls showed greater pain sensitivity than boys among both preterm and full-term adolescents (Buskila et al., 2003, as cited by Grunau 2013). In addition, studies with children and adolescents do not consistently show that females report higher pain ratings and display lower pain tolerance than males. However, sex differences appear to emerge beginning in adolescence (Martin, 2012). Girls tend to ascribe their pain to emotional distress to a greater extent than boys, who are more inclined to attribute their pain to a physical cause (Roth-Isigkeit, Thyen, Stoven, Schwarzenberger, & Schmucker, 2005).

In general, most chronic pain conditions appear to be more prevalent in girls than in boys: in a recent epidemiological review, King et al. (2011) reported higher rates of headache, musculoskeletal pain, recurrent abdominal pain, and, to a lesser degree, back pain in girls compared to boys. Bartley & Fillingim (2013), affirmed that as women enter adulthood, they are at an increased risk for a number of clinical pain conditions and show higher experimental pain sensitivity relative to men.

It seems therefore that demographic factors, not isolated, but combined with other variables, are crucial for improving our understanding of risk pathways that lead to a progression from acute to chronic pain (McKillop & Banez, 2016).

4.2 Temperament, fear and anxiety

Although results from available researches are conflicting, some evidence supports the suggestion of the relationship in children between temperament and response to painful procedures (Rocha, Marche, & von Baeyer, 2009). A literature review on pediatric temperament and pain responses

conducted by Ranger & Campbell-Yeo (2008), identified only few studies that addressed pain-related temperament. Conversely, more concordant studies examined the role of temperamental aspects that may be relevant to the experience of chronic pain.

It can be stated that general anxiety and temperament levels provide an experiential context through which the painful stimulus is processed and later recalled (Rocha, Marche, & von Baeyer, 2009).

As Conte, Walco, & Kimura, (2003) have found in their study on children with juvenile primary fibromyalgia syndrome, it is most likely the interaction, as individual risk factors, of temperamental instability, sensitivity to pain, vulnerability to stress, psychological adjustment, family context, and parental psychopathology that could explain the breadth of symptoms as well as its severity associated with this pain syndrome.

Previous research has found that certain aspects of children's temperament are associated with pain responses to medical procedures (Lilley, 1995). More recently, Chen et al. (2000) extend these findings by demonstrating that a pain-sensitive temperament is related to children's self-reported distress with acute lymphoblastic leukemia (ages 3 to 18) during LPs.

Temperament is believed to influence sensitivity and reactivity to painful situations (Rocha, Prkachin, Beaumont, & Hardy-Zumbo, 2003, as cited by Wolff, 2011). Specifically, child's temperament might predict response to adverse events and may affect children's perception of pain and the observable behavior (Helgadóttir & Wilson, 2004, as cited by Wolff, 2011). Indeed, child's temperament has been previously identified as a risk factor for maladaptive pain's behavioral changes following surgery (Fortier, Del Rosario, Rosenbaum, & Kain 2010).

Furthermore, child's temperament, if intended as a highly emotional behavioral style (i.e. child gets upset or cries easily), may be related to a poorer ability to forget some aspects of pain-related experiences (Jensen & Stjernqvist, 2002). However, in a subsequent study by Rocha, Marche, & von Baeyer (2009), the prediction that the emotionality dimension of temperament is related to pain recollection was not confirmed.

In their review, Racine et al., (2016) who examined 77 studies, have found that children with preexisting anxiety and a difficult temperament were more likely to have anticipatory distress in painful procedures. Moreover, parent's anxiety and previous experiences with pain are also important predictors of anticipatory distress.

As Asmundson, Noel, Petter, & Parkerson, (2012) have reported, fear of pain is a response to an immediate pain-related threat (e.g. initiating a painful activity), while pain-related anxiety is more a response that occurs in anticipation of pain-related threat (e.g. expecting an activity to be painful). In the persistent pain context, the fear or anxiety focus is often on specific objects or experiences, but can also extend to perceptions of self-worth and identity formation (Morley & Eccleston, 2004).

Psychological symptoms of anxiety (feeling tense, worried, fearful, crying spells and inability to “turn off” one’s thoughts) can be difficult to sort out in children/adolescents who face uncertainty about their health status as a recurrent pain condition. Also, if anxiety, which manifest in two ways, state or trait, becomes an anxiety disorder, the consequences in children/adolescents with chronic and or life limiting medical illnesses can be severe (Pao & Bosk, 2011).

There is a growing literature on an interesting relatively new concept, the “anxiety sensitivity” (i.e. the tendency to be specifically fearful of anxiety-related sensations, thus causing distress), that has been proposed as an explanation for individual differences regarding pain-related fear in children and adolescents (Norton & Asmundson, 2004). In healthy adolescents, anxiety sensitivity has demonstrated a strong link to fear of pain (Muris et al., 2001), while in children and adolescents with chronic pain, pain-related anxiety and fear of pain are predictive of pain-related disability and pain-related anxiety at the time of surgery (Asmundson, Noel, Petter, & Parkerson, 2012; Page, Stinson, Campbell, Isaac, & Katz, 2013).

Further specific relations provided by Asmoudson et al., (2012) in a study of children and adolescents receiving treatment for chronic pain, are that this kind of sensivity-trait is related to psychological well-being and social functioning, but not physical disability. Among adolescents with recurrent pain, those with higher levels of pain-related anxiety have been found to be more disabled by their pain and perceive themselves as having poorer social functioning (Caes, Fisher, Clinch, Tobias, & Eccleston, 2014).

Muris et al., (2007) showed that, in adolescents with chronic pain, especially the personality factor and the temperamental trait of fear are associated with high levels of pain catastrophizing. Further, adolescents with chronic pain are more vulnerable in terms of neuroticism, negative fear of failure, and (less) experienced social acceptance (Merlijn et al., 2006).

Several studies suggest that fear of pain plays a crucial role in the transition from acute to chronic pain (Asmundson, et al., 2012; Pao & Bosk, 2011). Together with stress and anxiety, chronic worry may increase the severity of the pre-existing physical illness (Pao & Bosk, 2011).

Given the difficulty in distinguishing the relationship between children’s fear and pain, (although related, they are qualitatively different affective states), specially with younger children who do not have the cognitive ability to differentiate between the two states (Melhuish & Payne, 2006; Noel, Chambers, McGrath, Klein, & Stewart, 2012), the multidisciplinary interventions to manage pediatric pain are most effective when they consider and address together the child’s fear and pain (Petovello, 2012).

5 Emotion regulation process and pain: relations between attachment dimensions, internal working models, object relations and defense mechanisms

Several studies have demonstrated that the relationship between stress and pain is not straightforward, thus suggesting the need to understand the emotional processes associated with stressful events, as well as the person's responses to these emotions. Indeed, the problems in regulating and expressing emotions are linked to pain and distress increase (Keefe, Lumley, Anderson, Lynch, & Carson, 2001).

An important aspect of emotion regulation is learning which strategies to use when confronted with an emotionally arousing, stressful situation, as is the pain (Drake & Winner, 2013), but some of these strategies can also be activated unknowingly. Surely, illness, hospitalization and invasive painful procedures deeply influence a child's psychological well-being. Some studies report that these children display anxious, withdrawn, fearful, restless and angry behaviors during hospitalization (Salmela, Salanterä, & Aronen, 2009; Small & Melnyk, 2006, as cited by Dolidze, Tchanturia, & Smith, 2013).

Hospitalization, but also recurrent pain, can be viewed as an ongoing transaction between the person and his/her environment or between normal sensations to abnormal ones, which results in an emotional appraisal.

The appraisal process and accompanying feelings build an integrated and dynamically changing behavioral pattern (Gaynard et al., 1998, as cited by Dolidze et al., 2013).

Based on these considerations, I would synthesize an overview of the Inner World's psychological theories: object relations theory, attachment patterns, pertinent object relations units and main defense mechanisms, all serving as a potential tool to discuss their implications for interpreting children pain behaviors.

Young children only slowly acquire the capacity to understand painful experiences and consciously engage in the self or social control (Craig, 1997) but it is also true that the relatively high occurrence of painful incidents provides the child with plenty of opportunities for learning something about pain (Walsh, McGrath, & Symons, 2008).

For this reason, parents or other caregivers who frequently interact with children experiencing stress and pain, should provide an important source of comfort and learning about managing pain. The attachment processes in early relationships between child and caregiver serves as protective mechanisms during encounters with danger and threat, including pain. These interactions, generalized over many incidents in a child's memory, are encoded in the "internal working models" of relationships (Mikail, Henderson & Tasca, 1994) and are substantially involved in the regulation of emotion (Bowlby, 1980; Cassidy 1994; de Rosnay & Harris, 2002; Main, Kaplan, & Cassidy 1985, as cited by Walsh, McGrath, & Symons, 2008).

In summary, internal working models define the individual's interpersonal style of relating, coping mechanisms, and world view. Furthermore, they are mental representations of the behavior of the self as well as of interactions with other significant, acting as a filter to interpretative modalities for any situation, which, in turn, which, in turn, affects the way we feel (Bowlby, 1980, as cited by Bretherton & Munholland, 2008). These intrapsychic and external factors are interactive and relatively stable, and therefore predate development of a chronic pain condition. Therefore, it could be useful to investigate how the intrapsychic variables and the concomitant environmental context serve as vulnerable or protective factors (Mikail, Henderson, & Tasca, 1994; Walsh, McGrath, & Symons, 2008).

As the attachment system is activated by the threat of pain, several studies have investigated the role attachment may play in adjustment to chronic pain (Laird, Preacher, & Walker, 2015). In their review on adolescent and adult population studies, Meredith, Ownsworth, & Strong (2008) report that, compared to individuals who are securely attached, insecurely attached people with chronic pain engage in more emotion-focused and less problem-focused coping, describe their pain as more threatening and themselves as less capable of dealing with it and report greater pain intensity and disability. Insecurely attached individuals also report more pain-related distress, a higher number of physical symptoms, higher levels of pain-related stress, anxiety, depression and catastrophizing and lower pain self-efficacy. As Porter, Davis, & Keefe (2007) have found in their literature review, several studies suggest that attachment style is meaningfully related to indices of adjustment to persistent pain.

The core concept is that interpersonal relationships and self-worth perception have a crucial role in the ability to cope with difficult situations such as chronic pain. Besides this self-representation, about how they experience their body self-integrity, the themes embedded within the pain conditions, resonate broadly with principles of object relations theory, i.e. the way they feel embedded in their social environment and how they experience their relationships to 'the other': parents, siblings, peers, or another significant person called "object" that means the target of another's feelings or intentions in order to stabilize their own equilibrium (Daniel, 2016; Di Gallo, 2001).

So, when a child is in pain, it is the family who is responsible for the initial pain assessment and seeking appropriate evaluation and care (Birnie, Boerner, & Chambers, 2013).

As Daniel (2016) reminds us, the object relations are understood to result from an interaction of introjections and projections, and are present from birth. In attachment theory, the emphasis is upon the environmental cause, ie caregiver unavailability: from this standpoint the family environment could be investigated as a vulnerable or protective factor in children pain behavior.

Another interesting field of research within the dynamic approach, much close to the previous two, is the notion of ego defense mechanisms, defined as unconscious mental processes that are used to protect the self from painful emotions, ideas and drives and may be used to regulate emotions experienced in relationships. The psychological development level, and the level at which the child comes into contact with the other, provide valuable hints in the assessment of defensive functions.

In the child, disease induces anxiety about the condition of his/her body especially when the disease is serious. As a result, primitive patterns of adjustment and response are activated (e.g. mechanisms of isolation, repression, projection and regression).

Some of these coping strategies, termed also as psychological defense mechanisms, help the child to constructively defend him/herself against and thereby handle otherwise intolerable pain (Gustafson & Kallmèn, 1990). The elaborated psychological defense mechanism of isolation is described as the split of emotion from cognition which keeps the emotion isolated from any kind of cognitive or verbal processing (Sjoback, 1973) and could explain the difficulties in communicating verbally. Children have difficulties in handling their emotions cognitively: as a consequence, some other primitive patterns of adjustment and response of regression may activating a process of emotions somatization.

Other typical mechanisms activated are repression (e.g. aggressive thought about parents or health professionals) and projection (e.g. thoughts projected onto another person, like siblings). Although this is a way to cope with distress, which is supposed to help the individual to a certain extent, it is nonetheless inappropriate and insufficient to handle the experienced stress over long term (Gustafson & Kallmèn, 1990).

Learning to handle pain based on internal or external cues constitutes a fundamental and highly adaptive process aimed at self-protection (Elsenbruch & Wolf, 2015). The defense mechanisms related to a pain condition, are an essential component of this self-protection response, which is formed by associative and learning processes, either unconscious or not fully conscious.

Considering children with cancer, Di Gallo (2001), said that the defense mechanisms of these children are important ego functions for survival: "the defense mechanism has a mainly adaptive and functional character, yet where it is used in a predominantly destructive way, it may lead in a direction which impedes any dialogue. The child shows us, in the strategies he adopts to cope with physical, psychic and social strains and restrictions, how he deals with his own feelings and where we can support him to find a way out of his isolation" (Di Gallo, 2001, p.200).

6 Ways of coping and adaptive processes

Identification and management of pain of any etiology and how youth cope with their pain is crucial to better understand the negative effects on everyday functioning that are not only harmful and

distressing independently, but may also delay or prevent recovery and contribute to poor long-term prognosis (Rajapakse, Lioffi, & Howard, 2014). While the extant literature on the use of specific coping strategies includes many studies of patients with chronic pain as their primary presenting problem, there is a much smaller body of research related to pain within the serious medical illness context. Furthermore, research regarding the impact of specific coping assistance strategies on child outcomes in children's disease-related pain is limited.

In general, the study of coping is fundamental to understand how stress affects people. In the broadest sense, ways of coping are the basic categories used to classify how people actually deal with stress, whether it is a momentary pain condition or a chronic pain syndrome instead (Skinner, Edge, Altman, & Sherwood, 2003). Categories describe what is happening on the ground during pain episodes coping (e.g. support-seeking, problem-solving, cognitive restructuring, distraction) and are typically classified into a smaller number of higher order categories (e.g. approach versus avoidance, active versus passive) and examined in relation to health outcomes (e.g. disabilities, residual abilities) (Skinner et al., 2003). It seems likely that coping can be conceptualized on several levels, ranging between instances of coping and adaptive processes (Skinner et al., 2003, 2007, as cited by Ellen & Zimmer-Gembeck, 2016), but the commonly used term is "coping strategies" and may grouped into two large families described in terms of adaptive and maladaptive styles. The role of adaptive coping styles is to promote a sense of self-control within the person in response to negative thoughts, behaviors, and feelings (McCracken & Eccleston, 2006). The ways in which children cope with pain-related stressors appear to differ from the coping strategies of adults. In fact, children emotionally appraise threats and their consequences differently from adults, and in turn (as a result), choose different coping strategies (Eccleston, Fisher, Law, Bartlett, & Palermo, 2015).

It is useful to briefly remember that while cognitive coping strategies are internal ways to handle a stressor, the emotion-focused coping is aimed to reduce and manage the intensity of the negative and distressing emotions.

Some studies have suggested that adolescents may use more strategies compared to younger children (and be more effective in doing so), which maximize accommodation to stressful situations; furthermore, older children also seem to use a greater number and variety of cognitive coping strategies (Schmidt, Petersen, & Bullinger, 2003; Zimmer-Gembeck, & Skinner, 2011). In general, older children use more cognitive coping strategies and secondary strategies to control (used to maximize their ability to cope with a stressful situation), than younger children (Rudolph, Dennig, & Weisz, 1995; Compas, Jaser, Dunn, & Rodriguez, 2012).

Compas, Connor-Smith, Saltzman, Thomsen, & Wadsworth (2001), have found that coping involves engagement with the stressor and one's emotional reactions is generally associated with better functioning and fewer emotional, behavioral, and health problems. Overall, passive coping strategies

(self-isolation, catastrophizing, activity avoidance, behavioral disengagement), have been found to be related to higher pain, somatic symptoms, depression and anxiety as well as functional disability (Compass et al., 2006; Kaczynski, Simons, & Claar, 2011; Kaminsky, Robertson, & Dewey, 2006; Reid, Gilbert, & McGrath, 1998).

For children suffering from chronic pain, the experience of pain itself constitutes a significant stressor (Compas & Boyer, 2001) and the process of coping with such stress is an important feature of the process of adaptation to illness (Compas, Jaser, Dunn, & Rodriguez, 2012). For example, the ways in which children cope with episodes of recurrent abdominal pain is related to their emotional distress, somatic symptoms, and pain episodes (Compas & Boyer, 2001).

In youth with sickle cell disease, Gil, Wilson, & Edens (1997) have found that greater coping skill use was related to greater personal and family capacity to manage pain effectively at home. In youth with cancer, previous research has consistently found that these patients and their families face significant physical (including pain), emotional, and psychosocial challenges during treatment (Hildenbrand, Clawson, Alderfer, & Marsac, 2014).

One important factor that requires further study and may be involved in coping strategies differences, including pain as primary factor due to the treatments, is time since diagnosis. Some studies have been focused on psychological adjustment of children with cancer, suggesting that pediatric patients experience reduced quality of life (QoL) in the immediate stages of oncological treatment and improved QoL as time progresses (Landolt, Vollrath, Niggli, Gnehm, & Sennhauser, 2006; Sawyer, Antoniou, Toogood, Rice, & Baghurst, 2000). Indeed, each phase of a chronic illness can present children and their families with significant challenges and stressors (Compas et al., 2012). However, there is evidence that chronic conditions may exert greater psychological and physical stress compared to acute illnesses that resolve quickly (Marin, Chen, Munch, & Miller, 2009), since the adverse effects of chronic stress include physical and psychological wear (Juster, McEwen, & Lupien, 2010, as cited by Compas et al., 2012).

It is useful to remind that the ways of coping depend on the circumstances in which they are used and may be useful in the short term, but probably not over long-term.

Skinner et al. (2003) suggested that at the highest level there are sets of basic adaptive processes that intervene between stress and its psychological, social, and physiological outcomes. We can then consider as “strategies of adaptation” those mechanisms through which coping has short-term effects on the stressor resolution as well as long-term effects on mental and physical well-being.

When we talk of adaptive processes (or adaptive coping), we refer to reach self-adaptive functioning and growth outcomes by minimizing risk processes (e.g. pain catastrophizing) and enhancing resilience processes (e.g. pain-related self-efficacy) in order to enhance positive thinking and limiting maladaptive pain-related cognitions (Cousin, Tomlinson, Cohen, & McMurtry, 2016)

Children's beliefs about self-efficacy and control over pain may at least partially determine their coping efforts and then a long-term adaptive adjustment (Liossi, 2002). In particular, in children, emotional awareness might enhance the likelihood of finding a more adaptive approach to the situation (Rieffe et al., 2007): thus, poor emotion awareness might result in using maladaptive coping strategies. By contrast, an increase in the feeling of control may assist a child in managing pain and contribute to a greater use of adaptive coping strategies.

Social support or strong social ties may mitigate the detrimental effects of the pain experience on school functioning and might have a protective role for children with chronic pain (Cousin et al., 2015, Simons, Logan, Chastain, & Stein, 2010).

A good family functioning is an additional step in finding new adaptive process that may promote pain-related coping, management and adjustment (Cousins, Kalapurakkel, Cohen, & Simons, 2015; Palermo, Valrie, & Karlso, 2014). Among youth with chronic headache, self-efficacy with pain acceptance were associated with less disability, better school functioning, fewer depressive symptoms and less functional disability (Carpino, Segal, Logan, Lebel, & Simons, 2014; Kalapurakkel et al., 2015).

Harper, Penner, Peterson, Albrecht, & Taub (2012), examining the relationships between children's positive dispositional attributes (i.e. resilience, temperament, persistence, adaptability), parents' empathic responses and children's pain/distress responses to treatment procedures, have found that children's resilience was significantly and positively associated with parents' empathic affective responses and negatively associated with children's distress and responses to painful cancer treatments.

According to Eisenberg, Fabes, Guthrie, & Reiser (2000), in children, higher resilience is associated with the ability to adapt and flexible use of problem solving strategies and this seems a key factor to face chronic pain conditions.

In a nutshell, the aim the support administered to a child affected by disease-related pain or procedure-related pain or chronic pain should be based on, is the evaluation of the resources and also the coping and adaptive mechanisms that are available to the child in his social environment.

7 Short and long-term effects of pain in children

Consequences of unrelieved pain can be permanent and may have an impact on memory storage, especially during acute pain episodes and medical procedures. By causing on children physical and psychological trauma, these painful interventions may lead to maladaptive situations throughout their lives, also because during painful procedures children have fewer resources available to accurately store and "encode" the experience into their memories, thus leading to poorly organized or fragmented memories (Noel et al., 2010, as cited by Potovello, 2012).

Since memories of pain are subjective and multidimensional and include several aspects - somatosensory (e.g. pain intensity), affective (e.g. fear, unpleasantness), and contextual (e.g. people, time, place) (Ornstein et al., 1999, as cited by Noel, 2012), it becomes increasingly clear that ensuring that pain is effectively managed is a top-priority.

Previous studies have documented facts, like memory for stressful events, for which infants and children, who experience pain in early life, show long-term changes in terms of pain perception and related behaviors (Brennan-Hunter, 2001; Young, 2005).

Thus, early negative learning experiences related to pain procedures, convey important information to children and adolescents about the appropriateness of specific behavioral responses such as avoidance of future health care (Goubert, Vlaeyen, Crombez, & Craig, 2011; Pate, Blount, Cohen, & Smith, 1996) and may lead to medical nonadherence and other comorbidities (Pao & Bosk, 2011). As Pao & Bosk (2011) highlighted, memories of painful procedures can cause youth to 1) have anticipatory fear and anxiety before subsequent procedures, and 2) develop anxiety disorders such as specific fear of blood or needle phobia.

For instance, in children who must undergo frequent, painful procedures such as bone marrow aspirations and lumbar punctures for the treatment of cancer or other serious immune deficiencies, the memory of a painful procedure early in the course of treatment can affect the pain and psychological distress associated with future procedures (Wall & Womack, 1998, as cited by Pao & Bosk, 2011).

A review of von Baeyer, Marche, Rocha & Salmon (2004), highlighted the individual variability in children's pain memories and resulting health behaviors as well as the need of early interventions that could prevent the consequences of negatively exaggerated pain memories.

Research has continued to demonstrate both short-term and long-term effects from inadequately treated pediatric pain, suggesting that early painful stimuli might permanently alter the neuronal circuits that process pain in the spinal cord, leading to lower pain threshold or central sensitization, especially in younger children (Blount et al., 2003; Cumming, 2015; Czarnecki et al., 2011; Hatfield, 2014).

High levels of pain and distress during medical procedures also increase fear, pain and distress in future procedures (Rocha, Prkachin, Beaumont, Hardy, & Zumbo, 2003; Wolff, 2011).

An important retrospective study conducted by Pate, Blount, Cohen, & Smith (1996) examined gender and other distal factors to determine how childhood medical distress was linked to adults' reports of pain and fear regarding medical events: factors related to previous medical experiences, including child pain, child fear, and child coping effectiveness, were the best predictors of adult reactions to medical procedures.

Given the many research suggesting that untreated pain may have long-term negative and permanent repercussions on pain sensitivity, immune functioning, and memory, an accurate assessment of children's pain is needed to diagnose conditions and to guide pain management interventions (Cohen et al., 2008; Noel, Palermo, Chambers, Taddio, & Hermann, 2015; Young, 2005).

8 Caregivers and Professionals' role

Several hospitals have shifted the focus on a family-centered model, allowing and encouraging caregivers to attend many painful procedures next to health professionals (Kuttner, 2004). All health professionals should be educated to deliver patient-centered care and be trained in pain measurement and treatment techniques that are suitable for infants and children (Srouji, Ratnapalan, & Schneeweiss, 2010).

Health care professionals should have a responsibility to relieve patients' pain, anxiety and provide comfort as much as possible while maintaining their safety. This balance may be hard to achieve for several barriers to pain management in children, such as myths, personal values and beliefs. As a result, preventing adequate identification and alleviation of pain for all children could be difficult and fail to provide children with satisfactory pain relief (Srouji et al., 2010; Twycross, Dowden, & Stinson, 2014). Nevertheless, as Petovello (2012) reported in a literature review, there is a clear preference by both children and caregivers for the caregivers to remain with the child during painful procedures (Blount et al., 2003; Kuttner, 2004; Young, 2005) and the reported effect varies, maybe depending on caregiver anxiety levels and coping skills as well as health professionals' interactions with the caregiver (Blount et al., 2003; Steward & O'Connor, 1994; Young, 2005, as cited by Petovello, 2012).

Arguably, both family and cultural beliefs impacts, affect how children learn to react to pain and the subjective perception on the same level of pain. Recent research has demonstrated that parent - child behavior interaction has implications for children's development and possible consequences related to children's self-efficacy beliefs in managing their pain (Page & Blanchette, 2009) and may reinforce particular behaviors thought their own responses to their child's pain, (Twycross, Dowden, & Stinson, 2014). In particular, for procedural pain, studies of parental presence impact during their child's procedure have been mixed, and there are many factors that determine the influence on a child's pain response, such as presence during procedures (Blount et al., 2003), caregiver assessment of children's pain, coping behaviors they encourage children to utilize during the pain experience (Kuttner, 2004, as cited by Potovello, 2012) and good explanations that focus on positive or negative aspects of the pain experience helping to decrease children's distress level (Cohen et al., 2001; Jacobsen et al., 1990; von Baeyer et al., 2004).

Parental behaviors have an important impact upon child and adolescent pain outcomes, just because relationships between parent and child/adolescent factors are likely dynamic and may change as

pain transitions from acute to chronic (Asmundson et al., 2012). It is in fact well known that family is an important context for understanding, assessing, and managing pediatric chronic pain and functional outcomes, such as disability (Chambers, 2003; Palermo & Chambers, 2005).

Parent pain acceptance, parent psychological flexibility, pain acceptance, emotional acceptance, and pain willingness, have initial support as promising resilience mechanisms (Cousin et al., 2015; Wallace, McCracken, Weiss, & Harbeck-Weber, 2015; Smith, Sieberg, Odell, Randall, & Simons, 2014) that exert a powerful influence on developing a maintenance of chronic pain (Palermo & Chambers, 2005).

In addition, the temperamental difficulties, maternal somatic symptoms, symptoms of anxiety, and parenting stress are all independent risk factors, meaning that they have a powerful impact on the meanings attributed and how to manage pain (Wolff et al., 2010). As Page and Blanchette have found in their review (2009), parents' anxiety impact on child distress is highlighted by a number of studies. For example, higher levels of parental state anxiety have been associated with higher levels of pain, anxiety, and distress in children undergoing anesthesia induction prior to surgery (Davidson et al., 2006).

Overall, clear communication, well-defined roles, cohesion, adaptability, effective problem solving, and affect regulation can contribute to adaptive family functioning (Palermo et al., 2014). If all this is combined with health professionals' correct attitudes and competences regarding pediatric pain management, we could seek the quadrature of the circle.

CHAPTER 2

CONCEPTUAL MODELS AND PRACTICE FRAMEWORKS OF PEDIATRIC PAIN: STATE OF THE ART

*“The essence of knowledge is, having it, to apply it; not
having it, to confess your ignorance”*

~ Confucio

In this chapter I will present the current state of the knowledge state-of-the- regarding the existing biopsychosocial models for pediatric pain and I will examine common key factors across different theoretical conceptualizations. Finally, by complementing the dynamic aspects introduced in the previous chapter with the state of the art presented here, I will try to outline the points still to be addressed in future research through an integrative model.

1 Why many models conceptualize several theories

Theories have been conceptualized to answer the question, ‘Which framework is required to understand the interplay among the wide range of factors that influence pain?’ (Holm, 2013).

I believe it is useful to provide an overview of the conceptual models and practice frameworks for understanding pediatric pain and better explaining the framework that I adopted. Despite differences related to specificity regarding behavioral, cognitive, emotional, social influences on pain, all biopsychosocial models share a common focus: their relevance to the current vision of specific pediatric pain condition and implications for clinical interventions. Importantly, the primary outcome of interest differs from one model to another (e.g. pain behaviors, pain, experience, chronic pain and disability) according to the basic construct’s importance that we wanted to investigate (Asmundson, Gomez-Perez, Richter, & Carleton 2014; Pagè, Huguet, & Katz, 2013).

2 Pain physiology theories

Several theoretical frameworks have been proposed to explain the physiological basis of pain, although none yet completely accounts for all aspects of pain perception (Moayedi & Davis, 2013).

Here, I provide an overview of the two major contributions: Gate Control Theory of pain and the Neuromatrix Theory of pain.

2.1 Gate Control Theory

The understanding of pain complexity and the acceptance of pain as a multidimensional experience is now well accepted. The first attempt to develop an integrative model of pain is the Gate Control Theory of pain, which provides a scientific framework for understanding the complex mechanisms associated with the experience of pain (Melzack & Wall, 1965, as cited by Welkom, Cohen, Joffe, & Bearden, 2009). This theory introduced the notion of a gating mechanism in the spinal cord that could be opened or closed based on input from higher brain centers responsible for psychological processes, such as cognitions (thoughts, beliefs and expectations) and emotions (such as anxiety and depression).

The Gate Control Theory has highlighted the role of central nervous system mechanisms, providing a physiological basis for psychological factors' central role, such as subjective experience of pain (Liossi, 2002). I started from this theory instead of the previous ones, because the contemporary definition of pain used by the IASP is based on the multidimensional definition proposed by Melzack and Casey (1968) from the Gate Theory. Interacting with each other, these dimensions include the sensory-discriminative (intensity, location, quality, and duration), the affective-motivational (unpleasantness and the subsequent flight response), and the cognitive-evaluative (appraisal, cultural values, context, and cognitive state) dimensions of pain.

2.2 The Neuromatrix Theory

Neuroscience studies have grown significantly over the past decades and have led Melzack (1999) to propose an update of the original gate-control theory: the concept of body-self neuromatrix, a widely distributed neural network, as a neurosignature pattern of nerve, including parallel somatosensory, limbic and thalamo-cortical components that sub serve the sensory-discriminative, affective-motivational and evaluative-cognitive dimensions of pain experience (Melzack, 1999).

The neurosignature pattern is partially modulated by sensory inputs, but also by cognitive events such as psychological distress (Melzack, 2001). The Neuromatrix Theory of pain maintains that pain is caused by the synthesis and processing of inputs from brain neurons network mentioned above and draws attention to the fact that pain is a major psychological stressor that alters homeostasis and triggers stress regulatory processes that can further influence pain. With their emphasis on the role of cognitions, emotions, and stress, the Gate Control and Neuromatrix theories have generated considerable interest in psychological perspectives on pain (Keefe & Somers, 2010).

3 Biopsychosocial approach of pain

Starting from the consideration that pain is not simply a neurophysiological phenomenon, but also involves social and psychological factors, the theories from the last 30 years have stated that factors like culture, family, nociceptive stimuli and environment influence pain perception and thus ultimately affect a person's emotions, behaviors and cognition. Over the years, there has been increasing understanding of the close and complex inter-relationship with the psychological processes going on in the individual and that individual's interaction with their social environment and the biological underlying aspects. The central role of cognition has been recognized by most biopsychosocial conceptualizations of pain. Several existing pain management approaches are based on the biopsychosocial model and the model application provides guidance in research and clinical practice.

3.1 Models of acute pain

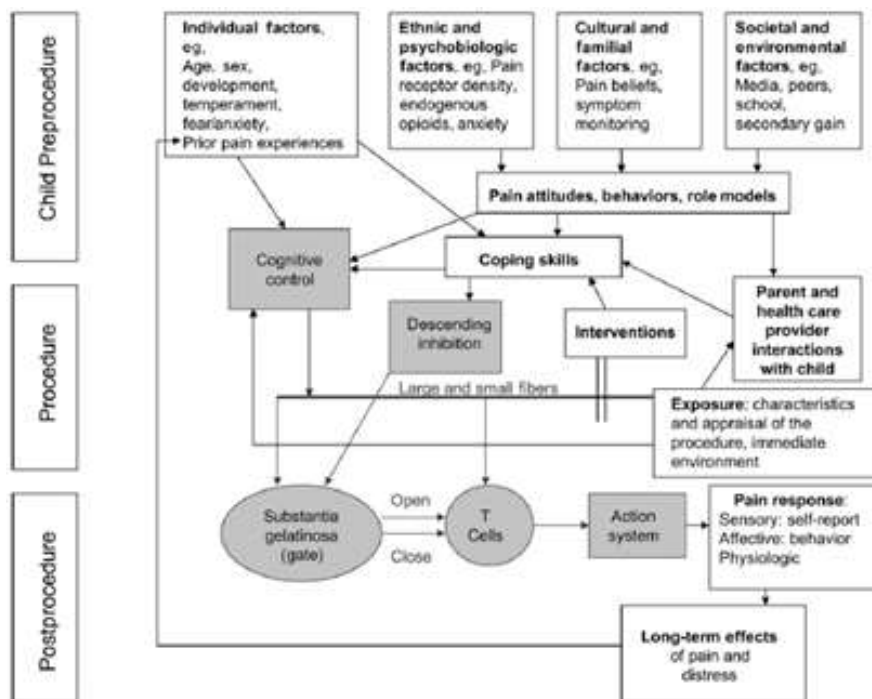
Much of the early research in this area has focused on correlations between variables related to pain responses (Blount, Bunke, & Zaff, 2000). A good explanation of acute pain is the procedure-related pain: Blount et al. (2003) claim that theoretical underpinnings of pediatric procedural pain are well developed through both classical and operant conditioning: a medical procedure and subsequent pain act as unconditioned stimulus and response, respectively. Conditioned stimuli include anxiety and distress that emerge either emotively or vicariously when being told of the procedure or pain and the conditioned response involves the distress, fear and anxiety that precedes the procedure. Generalization across settings and situations can occur via classical conditioning. More recently, clinical researchers have begun implementing more treatment outcome research on interventions designed to reduce pain and distress in children undergoing medical procedures. In all acute or procedural pediatric pain models, relationships have been proposed and interplay between variables are presented in the context of a particular painful episode (Riddell, Racina, Craig, & Campbell, 2014).

3.1.1 A Model for conceptualizing and studying pediatric procedural pain

In the biopsychosocial model framework, Young (2005) has proposed Model for understanding and studying children's pain responses specifically to procedures, but should be apply to acute pain arising from injury (Fig.1). The model incorporates Melzack and Wall's original Gate Control Theory and contextualizes pediatric procedural acute pain by splitting the time sequence of a procedural pain event into three phases: (1) pre-procedure, (2) procedure, and (3) post-procedure, which have also, a cumulative, mutual impact. This framework acknowledges that while acute pain occurs in a distinct episode of time (procedure), it is substantially influenced by pre-procedural factors, such as the mindset the child brings to the procedure (memories of past pain, fear/anxiety, temperament),

biological dispositions (such as pain receptor density, endogenous opioids), and social influences (such as parental modelling, cultural beliefs, media attention, peer influences). By this model, Young, (2005) has stated that it is necessary to improve children’s immediate procedural experiences to mediate the possible responses to future pain events and to reduce the long-term effects of procedural pain and distress. This is possible by modifying pre-procedural factors; improving parent and health care provider interactions with the child; using environmental, non-pharmacologic, pharmacologic, and multidimensional interventions.

Figure 1. Model for pediatric procedural pain (from Young, 2005, p.167, - scaled-down, low-resolution)

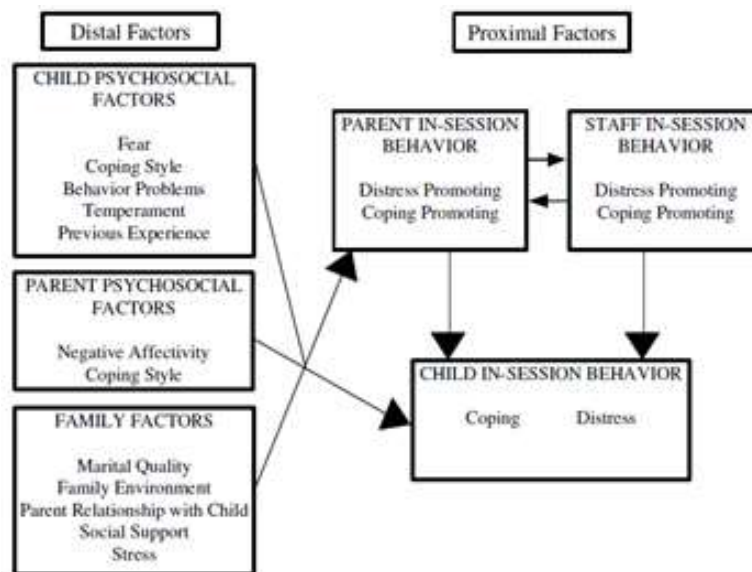


3.1.2 Proximal-Distal model of children’s coping and distress during acute painful medical procedures

The Proximal-Distal model of children’s coping and distress during acute painful medical procedures was developed to better understand and explain possible relations among the variables in the medical treatment room (Blount, Bunke, & Zaff, 1999, 2000; Varni, Blount, Waldron, & Smith, 1995, as cited by Manimala, Blount, & Cohen, 2000). This model incorporates the findings of previous explicative research identifying proximal and distal factors related to children’s pain responses during medical procedures (Fig.2). The distal factors, ie the child’s temperament, coping style, age, gender, previous medical experiences, parental presence, parent anxiety, and parent behavior have been associated with pediatric procedure-related pain and anxiety to varying degrees (Blount, Bunke, & Zaff, 1999; 2000, as cited by Page & Blanchette, 2009). The proximal factors include parent and staff behaviors that promote coping behavior or elicit distress behavior. Indeed, the parent in-session

behavior has a direct impact on children’s in-session coping and distress behavior. Distal factors are less apparent variables that may be related to child distress and/or coping behaviors, yet they may be unchangeable. In this model, distal variables tend to be trait-like and are temporally removed from the procedure. From a behavioral perspective, they become the factors that are not typically seen as factors affecting the immediate context (Blount et al., 2003). Conversely, proximal variables are more state like or situational in nature, and represent the primary behaviors of interest during treatment (Page & Blanchette, 2009).

Figure 2. Proximal-Distal model of coping and distress during acute painful medical procedures (from Varni et al., 1995, p.115, - scaled-down, low-resolution)

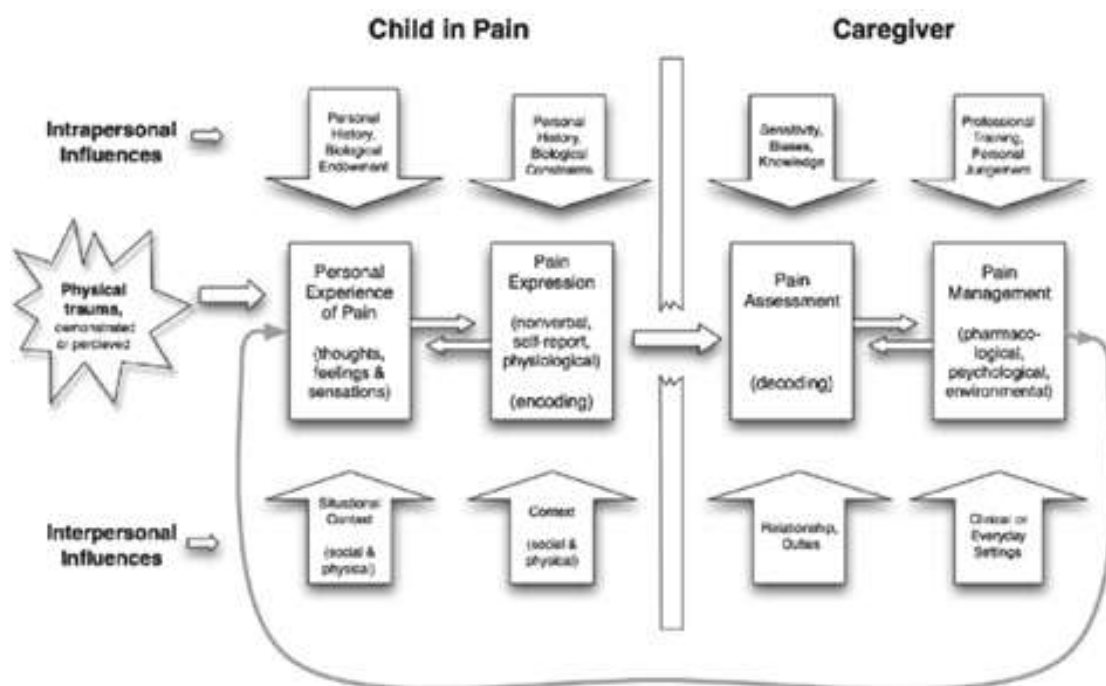


3.1.3 The Social communication model of children’s pain

Identified by Craig, Lilley, & Gilbert (1996), the Social communication model of children’s pain, is directly connected to the social learning theory and is consistent with biopsychosocial conceptualizations (Fig.3). A comprehensive understanding of pain as a social phenomenon requires thorough consideration of social or communicative features and the role of social learning theory in the current understanding of development, expression, and maintenance of pediatric procedural pain is crucial. Actually, this kind of approach contributes not only to our theoretical understanding of pain, but also provides insight into the mechanisms of effective treatment (Page & Blanchette, 2009). The complexities become substantial when considering the several interacting determinants of each stage: in fact, this approach recognizes that biological mechanisms are fundamental to the psychological processes engaged during pain experience and expression and it also emphasizes the focus on the social process of decoding expressive pain behavior and responding to the person in pain (causes and consequences of pain experience and the manner in which pain is expressed) (Craig, 2015). According to the model, a painful event precipitates a complex interaction among the noxious

stimulus, behavioral and emotional expression of pain, parent appraisal of the event and treatments (Craig & Pillai, 2003, as cited by Page & Blanchette, 2009). Individual and interactional factors are both key elements of the pain behavior. Furthermore, the model explains that while the parent and child provide the proximal environment to the pain event, the influence of family, community, and culture further complicate the expression of pain (Craig et al. 1996, as cited by Page & Blanchette, 2009).

Figure 3. The Social communication model of pain (from Craig, 2015, p.24, - scaled-down, low-resolution)



3.2 Bridge models between acute pain, disease-related pain and chronic pain

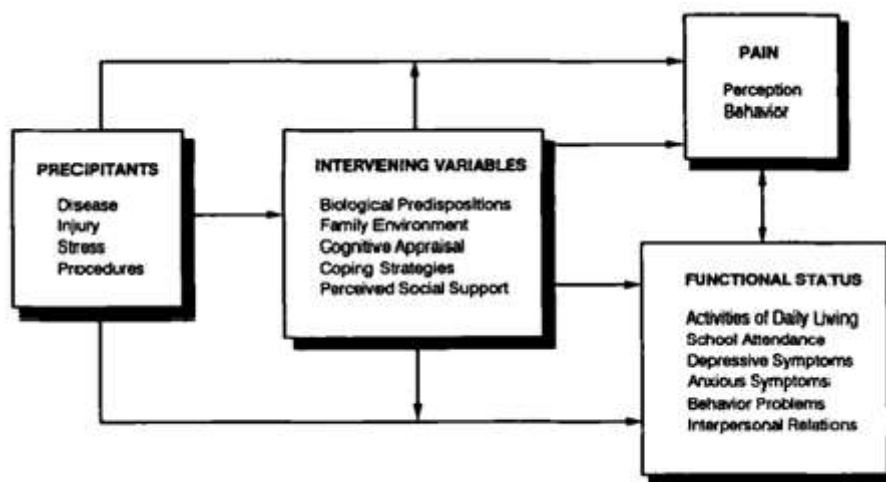
Few authors have considered the transition between pain conditions, also because it is relatively hard to combine different constructs within the same theoretical model. Below I will present two models that, in my opinion, represent a sound bridge between various dimensions of pain.

3.2.1 Biobehavioral model of pediatric pain

The transition from acute to chronic pain is a complex process involving biological, psychological and social factors. The multidimensional Biobehavioral model of pediatric pain (Varni 1989, 1995, as cited by Varni et al., 1996) was developed starting from the wide variability of pediatric pain perception and behavior to facilitate the identification of potential pain predictors in children and to model the relationship between pain and HRQOL (Varni et al., 1996) (Fig.4). This conceptual model includes a number of potentially modifiable factors that contribute to pediatric pain perception and (a) child's associated functional status and health-related quality of life (Varni et al., 1995 as cited by Vetter,

McGwin, Bridgewater, Madan-Swain & Ascherman, 2013). This model has proposed four main categories of precipitants, which include pain associated with chronic illness, pain associated with physical injury, pain not associated with a specific cause, and pain associated with acute medical procedures (Welkom, Cohen, Joffe, & Bearden, 2009). This multidimensional model has successfully been applied in a wide number of diseases: musculoskeletal pain, rheumatic diseases (Varni et al., 1996, Sawyer et al., 2005); headache and abdominal pain (White & Farrel, 2006) Cancer pain (Varni, Burwinkle & Katz, 2004) and a variety of chronic pain disorders (Berrin et al., 2007; Vetter et al., 2013). As a multivariate approach to understanding pediatric pain, this model has laid the groundwork for the implementation of behavioral interventions aimed at relieving children's acute pain experiences (Welkom et al., 2009).

Figure 4. The Biobehavioral model of pediatric pain (from Varni et al., 1996, p.517,- scaled-down, low-resolution)

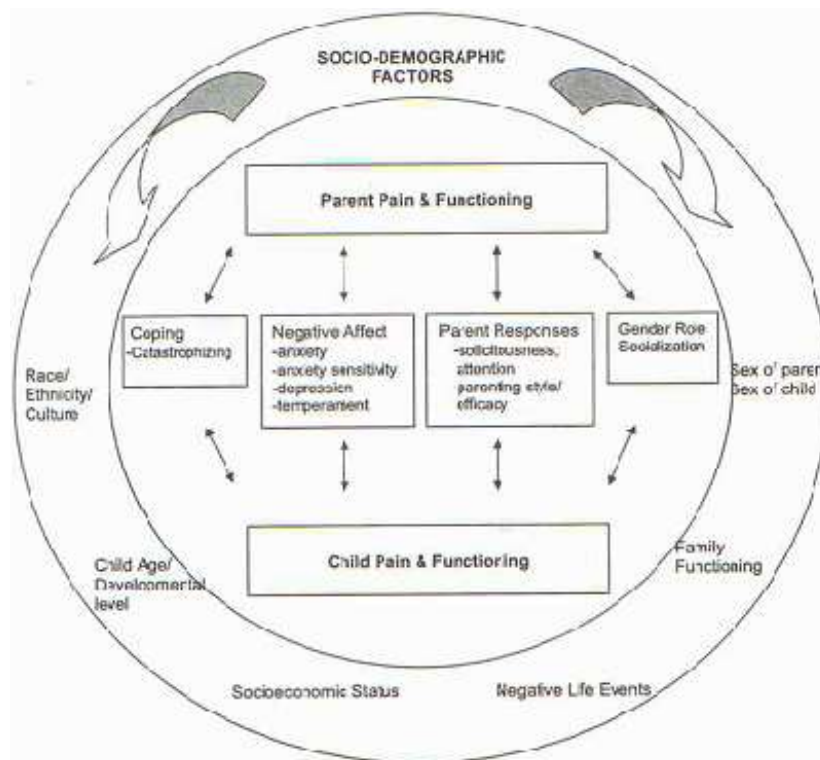


3.2.2 Conceptual model of psychosocial mechanisms linking parent and child pain

This model identifies parent and child characteristics that, singly or in combination, may influence parent-child pain relationships, within chronic and acute pain settings (Evans, 2008). This conceptual model incorporates several parent and child specific characteristics, such as parental responses, coping and gender role socialization as well as broader socio-demographic factors such as parent and child age and sex, family functioning, socioeconomic status, and race/ethnicity (Evans, 2008) (Fig.5). Considering both chronic pain and responses to acute pain, this model led to understanding of pain relationships across situations and for the development of a general parent-child pain model. Children may learn an entire repertoire of pain responses and the presence of pain models in the family environment has been associated with increased risk for child pain and pain-related disability and the analysis of these variables may explain parent-child pain relationships mechanisms. It will also lead the development of interventions designed to prevent and ameliorate child pain sensitivity when it correlates with poor adaptation to pain. Research has shown that the parent-child

relationship affects the child's perception and his/her ability to cope with pain (Reid et al., 2005; Logan et al., 2006, as cited by Twycross, Dowden, & Stinson, 2014). The effect of parent responses to children's symptoms can significantly change complaints and parenting stress may well emerge as an important variable in understanding the clinical parent-child pain relationship (Evans, 2008).

Figure 5. Conceptual model of psychosocial mechanisms linking parent and child pain (from Evans et al., 2008, p.12, - scaled-down, low-resolution)



3.3 Models of pediatric chronic pain

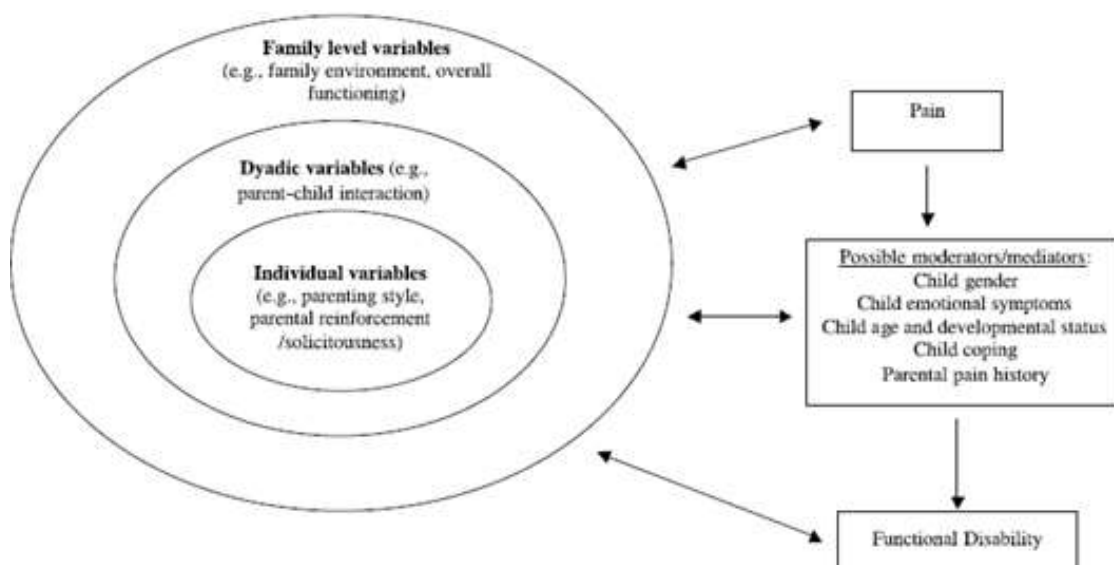
The assessment and treatment of pediatric chronic pain should emphasize the patient's personal experiential perspective as well as parent's own experiential perspective and, in turn, the interaction and dynamics within the parent-child dyad (Vetter et al., 2013). From this prospective, several biopsychosocial models have been put forth to explain how chronic pain develops and becomes maintained during childhood and adolescence.

3.3.1 Integrative model of parent and family factors in pediatric chronic pain and associated disability

Since the dynamic interplay between the individual and the social environment in which pain emerges determines exposure to pain, pain expression, and related disability (Hadjistavropoulos et al., 2011), Palermo & Chambers (2005b) have developed the Integrative model of parent and family factors in pediatric chronic pain and associated disability. The family factors may play a role in children's pain and accompanying disability and it is also important to understand how individual

factors versus dyadic and family level factors may impact pain and functional outcomes. The integrative approach proposed by Palermo & Chambers (2005), has improved understanding of the influence of parent and family factors in pediatric chronic pain. This model situates individual parenting variables (e.g. parenting style, parental reinforcement) within a broader context of dyadic variables (e.g. quality of parent–child interaction), which is further embedded within the more global familial environment (e.g. family functioning) (Fig.6). The model implies that, as it relates to pain, parental behavior must be considered in the context of other dyadic and family variables, emphasizing the importance of multiple levels of family assessment and the integration of operant/behavioral theories of parent behavior within the broader family systems theories. A complex relationship is proposed between family factors and pain, which highlights the reciprocal influence of the pain itself and its accompanying levels of disability, as well as child and/or parental factors (e.g. child age, gender, parental pain history), in interacting with all levels of individual, dyadic, and family variables as they relate to pain according to child’s age and developmental status. Subsequently, Palermo (2012) has expanded upon this earlier model by developing a more comprehensive model that involved several advanced features compared to the original one, including the addition of biological factors (e.g. pubertal development, pain modulation) as well as health habits (e.g. sleep, substance use, health beliefs, physical activity) and broader social factors instead of parents and family only (e.g. school environment, culture, social-peer interactions).

Figure 6. Integrative model of parent and family factors in pediatric chronic pain and associated disability (from Palermo & Chambers, 2005, p.3, - scaled-down, low-resolution)

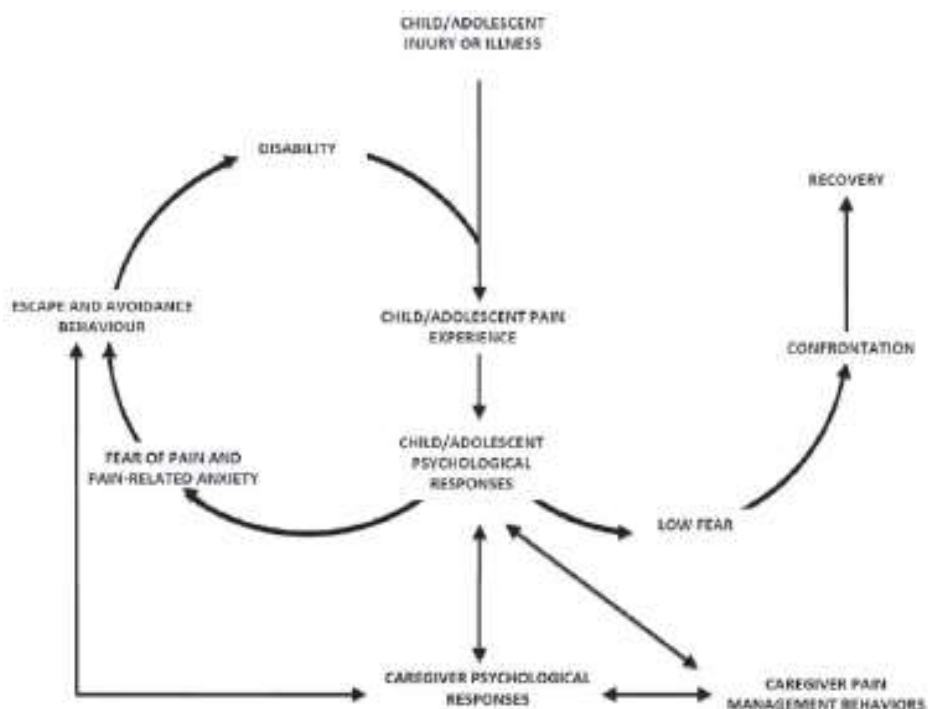


3.3.2 Pediatric Fear-Avoidance model of chronic pain (when acute pain becomes chronic)

To understand the possible role of fear-avoidance factors in the development and maintenance of chronic pain in childhood and adolescence, Asmundson et al. (2012) extended the Fear-Avoidance

model (FA) of chronic pain to pediatric populations (Fig.7). Initially developed for chronic musculoskeletal pain in adult patients, the fear-avoidance model, has become conceptualization of the processes and mechanisms through which acute pain can become chronic (Asmundson et al., 2012). The model has its concepts in behavioral and cognitive work and has led to the development and evaluation of conceptually related treatment options. In addition, it appears to be well applicable for pediatric patients with some modification to account for developmental differences across childhood (Simons & Kaczynski, 2012). In fact, the model adds to the adult fear avoidance model child/adolescent factors and parental variables. More specifically, the bidirectional relationships between parent pain management behaviors (e.g. protectiveness, solicitousness), parent psychological responses (e.g. parent catastrophizing about child pain, anxiety sensitivity, general distress) and child/adolescent psychological responses (e.g. catastrophizing, acceptance, anxiety sensitivity) will interact and influence child psychological response and escape/avoidance behaviors (Asmundson et al., 2012, Pagè, Huguet, & Katz, 2013). In summary, the fear avoidance model, understands the fear conditioning as a potential cause for the acquisition of pain-related fear in chronic pain patients (Vlaeyen & Linton, 2012). The fear has the potential to lead to avoidance behaviors and eventually disability: when an individual perceives pain as threatening, manifested as catastrophic thinking, he responds with pain-related fear and avoidance behavior, which in turn results in declines in functioning (Simons & Kaczynski, 2012). Like other pediatric models, developmental context is emphasized and relationships are conceived of as being dynamic.

Figure 7. Pediatric Fear-Avoidance model of chronic pain (from Asmundson et al. 2012, p.400, - scaled-down, low-resolution)

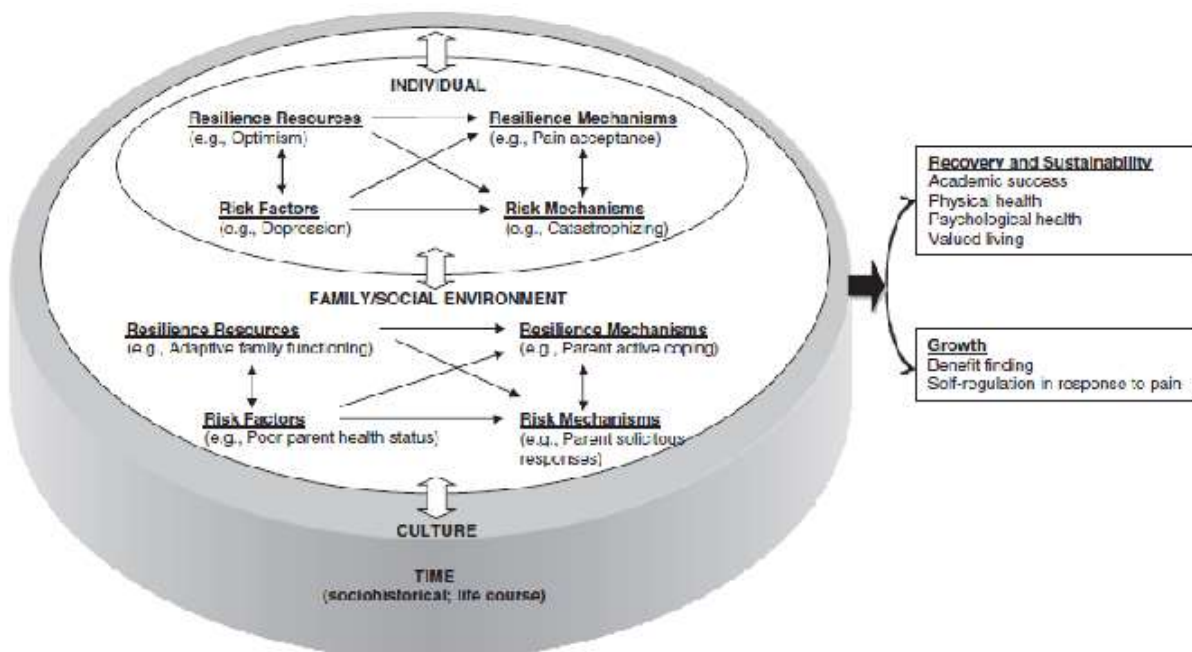


3.3.3 A Resilience-Risk model for pediatric chronic pain

Positive psychological factors (e.g. self-efficacy and acceptance) comprise a growing area of pediatric pain research (Cousins, Tomlinson, Cohen, & McMurtry, 2016). In children with cancer pain, optimism has been related to fewer problematic communications with health professionals and lower pain as well as higher quality of life and psychological, emotional, and behavioral functioning (Mannix, Feldman, & Moody 2009). In adolescents with sickle cell disease pain, higher optimism was related to more coherent medication use with reported pain intensity (Pulvers & Hood, 2013). These studies therefore suggest that optimism minimizes prominent risk mechanisms in pediatric chronic pain.

Cousins, Kalapurakkel, Cohen, & Simons (2015), starting from predominant risk resilience model in adult chronic pain (Sturgeon & Zautra, 2013, as cited by Cousin et al., 2015) have developed risk-resilience models in pediatric pain (Fig.8). Although a well-defined model did not exist previously, this conceptualization has been applied to different pediatric chronic illness populations (e.g. diabetes and asthma), and these models identify multilevel processes that promote health, illness management, adaptability, and psychosocial functioning (Hilliard, Harris, & Weissberg-Benchell, 2012; Koinis-Mitchell et al., 2012, as cited by Cousin et al., 2015). In summary, if the pediatric pain research has predominantly focused on risk factors and maladjustment with this model, the focus here is on protective factors and mechanisms of resilience to promote adaptive outcomes and minimize risk factors.

Figure 8. A Resilience-Risk model for pediatric chronic pain (from Cousin et al., 2015, p.2, - scaled-down, low-resolution)



3.3.4 The Attachment-Diathesis model of chronic pain

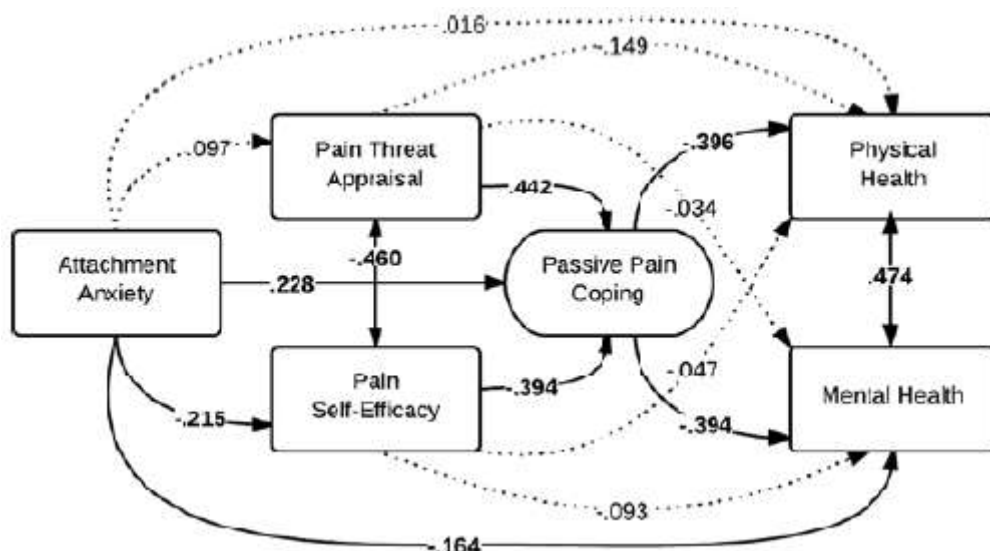
This model is a heuristic model of adult attachment and chronic pain based on current empirical research. However, it was also tested in a sample of adolescents and young adults with a history of childhood functional abdominal pain (Laird, Preacher, & Walker, 2015)

The model tries to understand the potentially differential impact of attachment as a dispositional vulnerability factor for the occurrence of chronic pain conditions, or as a factor affecting their progression and severity (Schroeter et al., 2015) (Fig.9). When applied to pain, Bowlby's attachment theory suggests that pain serves as a signal of threat and consequently activates a sequence of attachment behaviors, which, in turn, will influence the pain experience (Pagè, Huguet, & Katz, 2013).

In the Attachment-Diathesis model of chronic pain, appraisals of pain and the ability to cope with it joined to the availability of support, influence responses to appraisals and adjustment outcomes (Meredith, Ownsworth, & Strong, 2008).

According to this model, attachment factors occur prominently in the primary and secondary appraisals of stress and therefore can influence perceptions of how well one is coping with illness. In the study by Laird et al. (2015), the predictions of the Attachment-Diathesis model of chronic pain in adolescents and young adults with a history of childhood functional abdominal pain were investigated. Results showed that among children with functional abdominal pain, those with anxious attachment were at a higher risk for poor physical and mental health. Probably, there are other factors acting as mediators for pain beliefs and coping. The relation between anxious attachment and health outcomes would be an effective target for further intervention in chronic pain in children and adolescents (Pedersen, 2016).

Figure 9. The Attachment-Diathesis model of chronic pain in adolescents and young adults (from Laird et al., 2015, p.153, - scaled-down, low-resolution)



4.1 The theoretical model used in this dissertation

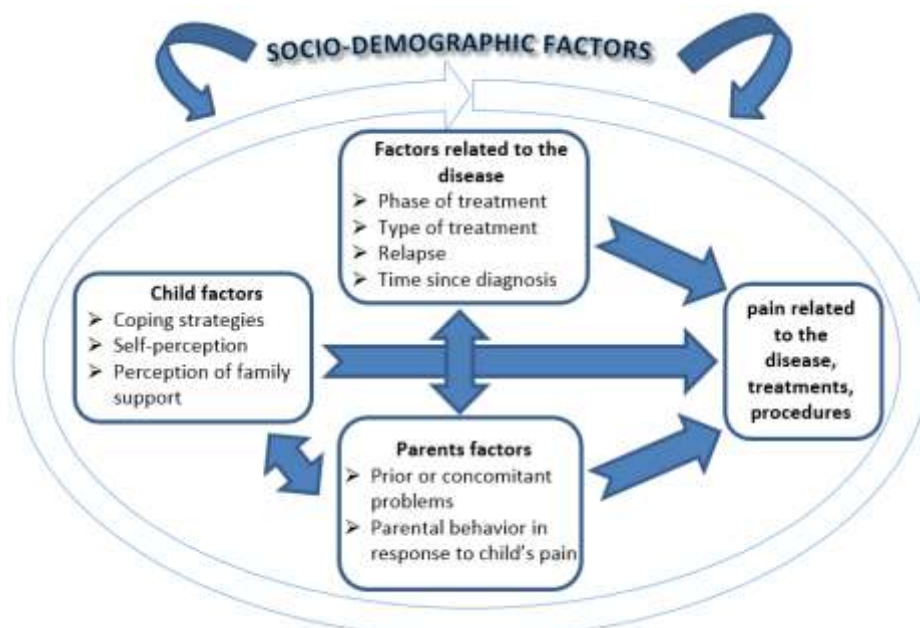
The theoretical model that I used in the first study examined in this dissertation (Chapter 3), is the multidimensional Biobehavioral model of pediatric pain (Varni, 1989, 1995, 1996) that I introduced previously, just because it was the first to consider the transition from acute to chronic pain keeping in mind also psychological and social factors. Given that this dissertation project has attempted to extend existing knowledge on psychological determinants of disease-related pain, I tried to use a new theoretical model to gain more information from projective drawings and other sources that I will present below. I applied this model in the study n° 2 (Chapter 4).

Then I will try to explain an initial conceptualization of a heuristic model guided by the clinical and theoretical literature inspired by the Integrative model of parent and family factor in pediatric chronic pain (Palermo & Chambers, 2005) and by the Attachment-Diathesis model of chronic pain in adolescents and young adults (Laird, Preacher, & Walker, 2015).

Important features of the model I am presenting lie in the fact that it provides the foundation for an integrative approach, by selectively highlighting both cognitive and dynamic constructs as well as socio-demographic influences (child age and sex, socio-economic status), which concludes with a summary of important considerations in assessment planning for a specific disease.

The model incorporates both child factors (pain-coping skills, self-perception, perception of the family support) and disease-related factors (phase and type of treatment, if there has been a relapse, time since diagnosis) as etiological determinants of pain that can be mediated from the parent factors such prior or concomitant problems (other diseases, divorce, any psychiatric problems, problems related to the siblings) and also the parental behavior in response to child's pain (parent acceptance of child's emotions and parent advice's quality when child had to face pain) (Fig.10).

Figure 10. The main model used in this dissertation (Chapter 4)



CHAPTER 3

COPING STRATEGIES AND EMOTIONAL PROFILES IN CHILDREN WITH DISEASE-RELATED PAIN

“To heal does not necessarily imply cure. It can simply mean helping people achieve a way of life compatible with their individual aspirations to restore their freedom to make choices - even in the presence of continued disease”

~ René Dubos

In this chapter I will present a study that aimed to understand how children and adolescents with chronic disease cope with pain and adapt to illness. Understanding what strategies chronically ill children use to cope with pain and stress are important for clinical intervention. Disease-related pain and the consequences of pain on children and families are important areas for further research inquiry. The current study used both self-report measures and projective drawing techniques in a novel way to assess the emotional functioning in a mixed-cohort of youth with disease-related pain. This protocol was administered to 47 children, ages 7 to 14, recruited from pediatric wards of hospitals of Trento and Rovereto. The results showed that the influence of the diagnosis and family support perceptions were predictive of overall coping strategies children use. When analyzing the emotional profiles (from the projective techniques), disease-dependent differences were found.

1 Background, aims and hypotheses

An area within disease-related pain of particular import is how youth with chronic illnesses cope with their pain. A number of existing studies describe pain coping and related outcomes in specific disease populations. (e.g. juvenile idiopathic arthritis in Kaak, 2010; cancer in Hildenbrand, Clawson, Alderfer, & Marsac, 2011). One possible psychological mediator of pain perception is the use of pain coping strategies (Thastum, Zachariae, & Herlin, 2001). For this reason, it is important to identify developmental patterns of children’s coping, including aspects of coping that are stable as opposed

to changing with development (Compas, Jaser, Dunn, & Rodriguez, 2012). There is a lack of consensus regarding age-related differences across the illness-related coping literature (e.g. Aldridge & Roesch, 2007; Campbell et al., 2009), highlighting the need for additional studies for clarity about coping in pediatric cancer research (Compas et al., 2012), and even more for other less-researched illnesses like rheumatic diseases and cystic fibrosis. Few studies have been focused on coping trajectories over time, and only on long-term coping in childhood cancer survivors (Bauld, Anderson, & Arnold, 1998; Boman & Bodegard, 1995). For the current study, we applied the conceptual framework from the Biobehavioral model of pediatric pain (Varni et al., 1996a) to disease-specific pain coping. Within this model, coping strategies are hypothesized as intermediary variables affecting the relationship between youths' emotional profiles and functioning. In order to expand the literature on disease-related pain coping, we utilized projective drawings in a novel way to assess the emotional profiles and coping strategies of youth with serious medical conditions and related pain. Indeed, the use of projective drawings, are useful as an externalized cognitive-emotional structure (Vass & Vass, 2011) and well established in clinical practices (Matto, Naglieri, & Clausen, 2005; Skybo et al., 2007); however, for how often drawings are used, there is relatively little research utilizing drawings to assess children's coping. This study investigated the relationship between children's emotional profiles (as assessed from their projective drawings), their experience of pain, and the strategies they employ to cope with pain.

The first goal of this study was to understand how coping strategies were related to projective drawing qualities across disease groups and age groups. Few studies directly compare unique disease populations; as such, we planned an exploratory aim to discern the coping differences between these groups. We hypothesized that emotional profiles coded as adaptive would correspond to more effective coping, and that projective drawing techniques would be a viable tool for evaluating youths' emotional responses to pain.

Our second aim was to compare main differences in coping strategies used to deal with pain among diseases and treatment-related factors. Based on previous literature, we hypothesized that pre-adolescents/older children would use more coping strategies in general and more active coping strategies than younger children (Compas & Boyer, 2001; Compas et al., 2012). We also hypothesized that the time since diagnosis would be related to different coping trajectories among the different disease cohorts, considering that the previous literature is not exhaustive or concordant.

2 Method

2.1 Participant selection and enrollment

The study was conducted at the pediatric wards of hospitals of Trento and Rovereto (Trentino, Italy). Eligible participants were children and adolescents with serious illnesses and pain conditions identified from lists of outpatient clinic schedules and recruited during outpatient clinic visits. Inclusion criteria for the participants were: (a) between ages 7-14 years and currently undergoing an evaluation in one of the two major hospitals in Trentino; (b) a chronic illness diagnosis for at least 1 month and no more than 5 years post-remission of symptoms; (c) pain was related to a chronic disease (rheumatic, cancer, cystic fibrosis); (d) did not have evidence of a developmental disability, cognitive, or neurological deficit (e.g., mental retardation, organic brain dysfunction) that would prevent them from comprehending and completing the self-report assessment questionnaires; and (e) the parents and child could read and speak Italian. The total pool of potential candidates who met the basic inclusion criteria was about 75-80 children (from the total number of these children seen in hospital programs of Trento and Rovereto over 2 year period). Initial contact was established with 59 patients. Of the 59 patients who could be contacted, 47 children (80%) were enrolled into the study. The most frequently cited reasons for not participating were difficulty scheduling the study visit (n=2), lack of interest/worries (n=9) and withdrawal of consent after the participation (n=1).

2.2 Procedures

The study was approved by the university's and hospital's Human Subjects Research Committees. All eligible children being seen in the hospital from 2012-2014 were offered the opportunity to participate in a study examining how children cope with pain in their illness. They were approached by myself about the project during a clinic visit or while they were in day hospital. If the family indicated interest, the informed consent (and child assent) process was obtained according to institutional guidelines. Once informed consent had been obtained, children completed self-report measures with items read aloud by a researcher. Upon completing the questionnaire, they completed projective drawing tasks. Data was collected in one session.

2.3 Measures

2.3.1 Demographics

Health professionals providing care for the participants completed demographic questions including the child's age and sex and the disease characteristics of the children (type, hospitals admissions, current ongoing medical intervention).

2.3.2 Pain intensity

Pain intensity was rated by the children to assess their pain at present and in general (average of all pain when in hospital for any visit, including outpatient). Children rated their pain using two scales with the following prompts: “How much pain do you have now?” and “How much pain did you have as an average of all time in the hospital?” We used both a numerical rating scale (NRS-11) and the Wong-Baker faces Scales (WBS). The NRS-11, which is scored from 0 “no pain” to 10 “worst pain,” is the scale most frequently used for children older than 8 years of age (Pagè et al., 2012; von Baeyer et al., 2009), however, its use requires numeracy, the ability to think and express oneself in quantitative terms, and verbal communication skills. For this reason we also used the Wong-Baker faces Scale (WBS), because the task can be handled by simply matching how one feels to one of the faces, which is presumed to be easier than quantitative estimation. Although there is debate about the optimum design of the facial expressions, the literature suggests that they are the preferred method of pain reporting by children over 3 years of age (Garra et al., 2010; Loy, 2002).

2.3.3 Coping

Pain coping strategies were assessed with the Waldron/Varni Pediatric Pain Coping Inventory (PPCI) – Child form, with Italian standardization (Bonichini & Axia, 2000). This questionnaire was developed to systematically assess children’s pain coping strategies (Varni et al., 1996b) and has been shown to have a good reliability and validity in an Italian validation study (Bonichini & Axia, 2000) in a sample of 100 hospitalized Italian children, with a good internal reliability for most subscales: Distractions ($\alpha = .68$), Social Support ($\alpha = .61$), Problem Solving ($\alpha = .57$), and Cognitive ($\alpha = .53$). The questions about catastrophization were removed in the Italian validation because the coefficient was too low ($\alpha = .25$). In our sample (N=47) the total internal reliability Cronbach's alpha for the four subscales was high ($\alpha = .80$) and good for each one of the subscales: Distractions ($\alpha = .60$), Social Support ($\alpha = .70$), Problem Solving ($\alpha = .58$), and Cognitive ($\alpha = .56$). The measure has 29 items scored on a three-point Likert scale [0 = never (not at all), 1 = sometimes, or 2 = often (a lot)]. The scale was developed taking into consideration both the need for scale sensitivity as well as the cognitive developmental abilities of young children. The measure includes four specific subscales, including Cognitive Self-Instruction, Problem-Solving, Distraction, and Seeks Social Support. In addition, at the end of the questionnaire there are four open-ended questions with the stem “When I hurt...”: “I think; I do; I ask; and I wish”. Higher score on all subscales indicate better coping with pain.

2.3.4 Emotional profile, coding procedures as follows

Projective drawings techniques were used to investigate participants’ emotional profiles with respect to both self-representation [Draw A Person (DAP); Tree Drawing Test (TDT) and family representation (Draw A Family (DAF)]. Numerous efforts to standardize drawings as a projective technique have

been undertaken and have simultaneously been supported and challenged over the years (e.g. Hammer, 1997; Motta, Little, & Tobin, 1993). For this reason we have developed our coding systems on three established projective technique manuals (Lis, 1998; Passi Tognazzo, 2012; Serraglio, 2011). We used a global approach that involved judging all of the features of a drawing and ranking on a 0-4 Likert scale, where 0 is “not adequate” and 4 is “very adequate representations”. The higher the score, the more well-adjusted we judged the self-representation or family perception. Additionally, a clearer sense of the drawings' characteristics was gained through the use of the questions done at the end of the main drawings (Person and Family). See the Table 1 for more drawing coding details.

To interpret the DAP, we combined the two major dynamic approaches to human figure drawing scoring and interpretation: Machover (1949) and Koppitz (1968). Drawings were analyzed and compared on various elements such as the expression or attitude of the figure shown in the drawing, position, size, pressure, stroke and type of gesture, orientation of the figure, the number of details, omissions of body parts, disproportions, symmetry, age difference, and physical differences. Information from post-drawing inquiry (e.g. if the figure is more often sad or happy, and why) was also used. Drawings were coded on both qualitative (content) and quantitative (formal drawing quality) scales. Drawings were administered with the request to “Draw a person”. If the child drew a person of the opposite sex, we asked him/her to draw another one of the same sex. The analysis was on the drawing of the person of the same sex.

To code the TDT (Koch, 1952) various elements were analyzed and compared, such as: location; size; pressure; presence of stereotype (e.g. clichè-like repetition, like previous motifs without any variation); line of development of the design; three-dimensionality; the presence and the type of roots; some characteristics of the trunk such as bark, surface, shape, the base, the ground line; shape of the crown; orientation; type of branches; and leaves, flowers, fruit, other details. We coded both content and structural (formal) features as we did with the DAP. Drawings were administered with the request to “Draw a tree, except not a Christmas tree”.

We used the DAF task (Corman, 1964) to assess participants' perceptions of their families. In making coding decisions, we considered the mutual position of the characters, the direction of the structuring of the design, the composition of the family, the child's place within the drawing, and the enhancement or depreciation of characters. Several standard queries were made after the drawing was completed (e.g. who is the most happy/unhappy; who would you be/not be; is there someone who is missing in the drawing). For the purpose of this study we used only drawing's content analysis with this test. The task was administered with the statement “Draw a family”.

Table 1. Summary of integrative drawing scoring system

Drawn A Person (DAP) legend

<i>Formal-structural aspects</i>	
1 = not adequate	position very decentralized, very small or very large, light or heavy pressure and suddenly insecure or very marked, omissions parts of the body, unlike age and / or physical
2 = partially adequate	position decentralized, small or large, pressure and dashed / fragmented or continuously without separations, possible disproportions and / or omissions, lacking details and / or excessive details
3 = overall adequate	central location on page, pressure and total tract (signs) smooth and continuous, balanced whole and details
4 = adequate	central, well-proportioned size, pressure and stretch (signs) well-modulated, any omission or particular disproportion, well-balanced
<i>Content aspects</i>	
1 = not adequate	strong enhancement one of this: debasement, disparagement, belittlement, depreciation... about his self) and asymmetry
2 = partially adequate	excessive devaluation (one of this: debasement, disparagement, belittlement, depreciation... about his self) and / or use, hands hidden or very large
3 = overall adequate	not special markdown and / or valuations, possible shortage / excess of details
4 = adequate	central, well-proportioned size, pressure and stretch well-modulated, any omission or particular disproportion, well-balanced

Tree Drawing Test (TDT) legend

<i>Formal-structural aspects</i>	
1 = not adequate	very decentralized, very small or very large, pressure and suddenly insecure or very marked, omissions parts of tree, holes and / or strong bumps, branches stretch to be represented with a single line and / or horizontal
2 = partially adequate	position decentralized, small or large, pressure and stretch undecided or very marked possible presence of holes, branches messy and / or pointed, irregularities in the bark, fruits not attached to the branches based very close or enlarged, swollen trunk, no hint of root and / or land line absent
3 = overall adequate	central location designed for that, pressure and total tract smooth and continuous, balanced whole although irregularities with right or left, total fruits attached to the branches
4 = adequate	central location, size well-proportioned and full development, pressure and stretch well-modulated, no particular disproportion, well-balanced and airy

Content aspects

1 = not adequate	strong characterization of regressive elements, bare or dying
2 = partially adequate	possible presence of regressive elements or dominant, with leaves and / or fruits falling
3 = overall adequate	no presence of regressive elements, the possible presence of distress symbolism but exceeded
4 = adequate	balanced elements, no trauma

Drawn A Family (DAF) legend

Content aspects

1 = not adequate	characters very separated / neighbors, family composition invented with omissions and / or added, placing the child away from the family and / or absent, devaluation characters
2 = partially adequate	characters with varying distance, family composition invented or real with possible omissions except himself, enhancement and / or devaluation
3 = overall adequate	characters neighbors, family composition real or invented without omissions, possible valuations
4 = adequate	characters together and smiling, composition real family, no enhancement and / or devaluation

Coding procedures

Drawing tests was part of a full battery of measures as occurs in clinical psychological testing and they was combined with several standard queries were made after the drawing was completed and with data from reports obtained from health professionals. The coding procedure may have lost the richness and depth of information that such drawing tests are capable to yielding, but it was a necessary compromise.

Having said that, each drawing was scored using an overall (global) approach with an integrative scoring system designed to measure representations of psychological functioning of the individual on a scale from 1 to 4. On this scale, 1 equals a poor functional perspective (about the self or the environment), and 4 equals healthy adaptation and well-being. To assure reliability of interpretation, two psychologists, myself and another one with previous training and experience scoring and evaluating projective drawings, independently coded drawings (n = 12; 25% of the sample) until at least a 95% agreement was reached. One of the coders was unaware of the disease of each child and was only aware of each child's age. Subsequently, the two coders met to reach consensus about what integrative features of the drawings influenced their ratings and differentiations.

2.4 Data Analysis

Data were analyzed using SPSS statistical analysis software, version 21.0. (IBM Corp., 2012). Descriptive statistics, including means and standard deviations for continuous data and frequencies and proportions for categorical data, were computed to examine mean scores for child self-report for the pain intensity and PPCI, as well as the coded scores from the projective drawings. Bivariate correlations between pain and projective drawing coding were also examined. In order to address the first study aim to understand how coping strategies, projective drawing qualities, treatment factors, and pain differed across disease groups and between age groups, we used a series of Multivariate Analysis of Variance (MANOVAs) and a series of multi variate analysis of covariance (MANCOVAs) to control for demographic differences like age. Children (7-10 years old) vs. preadolescents (11-14 years) were chosen as the cut-off points for the two age groups based on existing data showing developmental differences in these age groups. To address our second aim, we tested the predictive influence of time since diagnosis, type of diagnosis, and self-/family-representations on children's coping by conducting a series of hierarchical multiple linear regressions analyses. Time since diagnosis groups were created based on initial spans of 6 month increments in the more "sensitive" periods, and after 1 year, intervals were more arbitrary but based on examples from the previous literature (Clawson, Jurbergs, Lindwall, & Phipps, 2013) (months since diagnosis: 1-6 ; 7-12; 13-24; >25; since birth). Effect sizes for all analyses were calculated as partial eta-squared.

3 Results

3.1 Sample Description

Descriptive characteristics of participants are presented in Table 2. The entire sample consisted of 47 children: 27 male and 20 female subjects. Their ages ranged from 7 to 14 years ($M = 10.2$, $SD = 2.5$). All children had been diagnosed with a chronic pediatric illness for at least 1 month before participation in the study. Diagnoses included cancer ($n = 20$), rheumatic diseases ($n = 17$), and cystic fibrosis ($n = 10$). All children were Caucasian and were assessed during an outpatient clinic visit ($n = 34$) or while in day hospital ($n = 13$). Approximately 66% ($n = 31$) were currently receiving active medical intervention for their disease, while 34% ($n = 16$) were not. Children reported low present pain intensity ($M = 0.77$, $SD = 1.49$) but their ratings of prior pain (i.e., average of all pain in hospital) were reported as moderate to severe ($M = 6.09$, $SD = 2.39$) across all three disease cohorts. The correlation between the NRS and WBS scales was very high both when for present pain ($r = 0.785$; $p < .001$) and past pain ($r = 0.834$; $p < .001$). For this reason we only report the values of NRS scale. With respect to the PPCI-C, the average overall score was 26.28 ($SD = 9.37$).

Table 2. Demographic and Medical Characteristics of Study participants (n=47)

Characteristics	Overall	Oncological	Rheumatic diseases	Cystic fibrosis
Age, mean (SD), years	10.2 (2.3)	9.4 (1.9)	10.9 (2.5)	10.7 (3.1)
- 7-10 years	N= 27	N= 14	N= 8	N= 5
- 11-14 years	N= 20	N= 6	N= 9	N= 5
Gender, n (%)				
- Male	27 (57)	15 (75)	7 (41)	5 (50)
- female	20 (43)	5 (15)	10 (59)	5 (50)
Setting, n (%)				
- DH	13 (27)	12 (60)	14 (82)	9 (90)
- ambulatory	34 (63)	8 (40)	3 (18)	1 (10)
Therapy, n (%)				
- off	16 (34)	13 (65)	2 (12)	1 (10)
- on	31 (66)	7 (35)	15 (88)	9 (90)

3.2 Differences in the use of coping strategies and characterization of coping profiles

Similar coping strategies for pain management were reported between the two age groups and across the three disease cohorts (see Table 3). Recency of diagnosis was related to coping strategies used [$F(4,42) = 2.930$; $p = .032$; partial $\eta^2 = 0.218$]. In particular, using a Bonferroni correction for group contrasts, the data showed that children more recently diagnosed with their chronic disease (1-6 months) used fewer cognitive strategies than those children who received their diagnosis between 1 and 2 years prior [$p = 0.043$]. Clinical disease-related factors were also associated with use of coping strategies. There was a significant interaction between disease diagnosis and hospital admission (main inpatient) [$F(2,41) = 3.915$; $p = .028$; partial $\eta^2 = 0.160$]. Children with cancer and a current day hospital admission used fewer coping strategies compared to the group of children with rheumatic disease. Additionally, the overall number of strategies used was influenced by the combination of the type of pathology and time since diagnosis [$F(3,38) = 3.323$; $p = .030$; partial $\eta^2 = 0.208$]. Specifically, youth with cancer and who received a diagnosis one year or more prior reported the highest level of coping strategies compared to youth with rheumatic disease.

Table 3. Results Comparing Group Differences

Variable	Group Ms, SD			MANOVA Results			
	Cancer N=20	Rheumatoid N=17	CF N=10	F	DF	P Value	Partial Eta- squared
Present pain*	0.25 (0.72)	1.41 (1.87)	0.70 (2.68)	3.046	2, 44	.058	.117
Past pain	6.00 (2.47)	6.65 (2.32)	5.30 (2.31)	1.028	2, 44	.366	.045
Cognitive Coping	6.40 (2.26)	6.65 (2.78)	6.20 (3.15)	0.95	2, 44	.910	.005
Problem Solving	6.45 (2.59)	5.71 (2.42)	5.20 (3.30)	.801	2, 44	.455	.036
Social Support	8.70 (3.31)	8.35 (3.76)	7.10 (2.77)	.763	2, 44	.472	.034
Distraction	5.95 (2.80)	5.76 (3.11)	4.60 (3.17)	.720	2, 44	.492	.030
Total Coping	27.50 (8.16)	26.71 (9.67)	23.10 (11.26)	.755	2, 44	.476	.032
Formal-DAP**	1.70 (0.57)	2.41 (0.71)	2.40 (0.52)	7.567	2, 44	.001	.256
Content- DAP***	2.20 (0.77)	2.82 (0.73)	2.80 (0.63)	4.109	2, 44	.023	.157
Formal-TDT	1.95 (0.61)	2.35 (0.61)	2.30 (0.48)	2.513	2, 44	.093	.103
Content-TDT	2.05 (0.78)	2.53 (0.62)	2.60 (0.52)	3.134	2, 44	.045	.132
Formal-DAF	2.40 (0.60)	2.53 (0.72)	2.80 (0.42)	1.259	2, 44	.294	.054

*Bonferroni correction for post-hoc analysis: within oncological and rheumatic $p=.053$ (not significant)

**Bonferroni correction for post-hoc analysis: within oncological and rheumatic $p<.05$ (.003) and within oncological and cystic fibrosis $p<.05$ (.016)

***Bonferroni correction for post-hoc analysis: within oncological and rheumatic $p<.05$ (.03)

3.3 Prediction of coping strategies

Five hierarchical multiple regression analyses were conducted in order to examine if the variables such as time since diagnosis, type of diagnosis, and the perception of the family support derived from projective drawings could significantly predict either individual strategy use (distraction, social support, problem solving, cognitive) or total coping strategies use. Only the models for Distraction Coping and Total Coping Strategies were significant (Table 4, figure 1 and 2). Tests for multicollinearity indicated that a very low level of multicollinearity was present (VIF = 1.000 for time since diagnosis, 1.699 for type of diagnosis and 1.062 for perception of the family support). Time since diagnosis was the first variable entered, followed by type of diagnosis and then the perception of the family support (DAF formal), according to our theory. Results of the regression analysis provided partial confirmation for our hypothesis. For prediction of total coping strategies, beta coefficients for the three predictors of total coping strategies were: time since diagnosis, $\beta = .307$, $t =$

1.725, $p = n.s.$; diagnosis, $\beta = -.433$, $t = -2.420$, $p = .020$; and DAF, $\beta = .312$, $t = 2.205$, $p = .033$. The best fitting model for predicting the total coping strategies was a linear combination of the time since diagnosis, the type of diagnosis, and the perception of the family support ($R = .435$, $R^2 = .189$, $F(3,43) = 3.338$, $p = .028$). Addition of the three variables produced a small but significant increase in the amount of variance explained (R^2 change = .092. $F = 4.861$, $p = .033$).

We also performed with the same hierarchical multiple regression analyses with the individual coping strategy subscales as the dependent variables. All the collinearity statistics (Tolerance and VIF) were within accepted limits and the assumption of multicollinearity was met. For the distraction strategies, beta coefficients for the three predictors were: time since diagnosis, $\beta = .292$, $t = 1.639$, $p = n.s.$; diagnosis, $\beta = -.435$, $t = -2.511$, $p = .016$; and DAF, $\beta = .397$, $t = 2.900$, $p = .006$. The best fitting model for predicting the use of distraction strategies was a linear combination of the time since diagnosis, the type of diagnosis, and the perception of the family support ($R = .490$, $R^2 = .240$, $F(3,43) = 7.195$, $p = .008$, R^2 change = .187).

Table 4. Hierarchical Regression Analyses Predicting Total Coping strategies and Distraction strategies

	Total Coping strategies					Distraction strategies				
	b (T)	SE b	β	R^2	ΔR^2	b (T)	SE b	β	R^2	ΔR^2
Step 1				.009	.009				.010	-.012
Constant	24.010	3.720				4.869	1.182			
Time since diagnosis	.670	1.022	0.097			.215	.325	0.098		
Step 2				.097	.088				.092	.051
Constant	26.596	3.803				5.665	1.212			
Time since diagnosis	2.337	1.274	.339			.728	.406	.333		
Diagnosis	-4.603	2.226	-.383*			-1.417	.709	-.371		
Step 3				.189	.092				.240	.187
Constant	17.126	5.634				1.836	1.732			
Time since diagnosis	2.115	1.226	.307			.638	.377	.292		
Diagnosis	-5.209	2.152	-.433*			-1.662	.662	-.435*		
DAF formal	4.465	2.025	.312*			1.805	.623	.397**		

$\Delta R^2 =$ change in R^2 ; $\beta =$ standardized regression weights; * $p < .05$; ** $p < .01$

Figure 1. Predictive factors are diagnosis and the self-representation of the family as supportive, and dependent variables is Total coping strategies

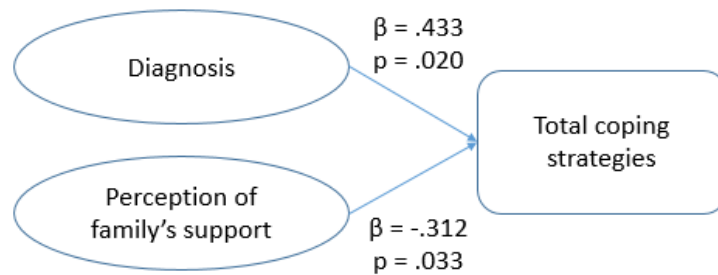
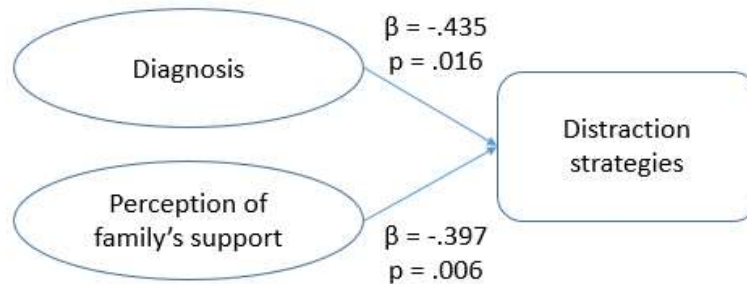


Figure 2. Predictive factors are diagnosis and the self-representation of the family as supportive, and dependent variable is distraction strategies



3.4 Differences in emotional profiles

With respect to self-representation drawings, DAP and TDT scores between both formal ($r = 0.369$; $p = 0.012$) and content codes ($r = 0.321$; $p = .029$) were significantly and positively related. Using MANOVAs (see Table 2), we found differences between disease groups on both structural and content-based codes of the DAP task (formal DAP [$F(2,44) = 7.567$, $p < .001$, partial $\eta^2 = .256$]; content DAP [$F(2,44) = 4.109$, $p = .023$, partial $\eta^2 = .157$]). There were also marginally-significant group differences across disease conditions on the content-based codes of the TDT task (content TDT [$F(2,44) = 3.334$, $p = .045$, partial $\eta^2 = .132$]). With respect to family-representation drawings, DAF scores were not significantly different between groups. Using a Bonferroni correction for group contrasts, the data showed that children with cancer had less adequate self-representations than either children with rheumatological conditions ($p=.003$) or those with cystic fibrosis ($p=.016$).

An additional MANOVA assessed group differences in self- and family-representations based on whether children were current receiving medical treatment or not. Children currently participating in of medical treatment therapy perceived themselves more adequately than those who were not (formal DAP [$F(1,45) = 7.151$, $p = .010$, partial $\eta^2 = 0.137$; Bonferroni correction for group contrast DAP $p = .010$]; content TDT formal-structural [$F(1,45) = 6.55$, $p = .014$, partial $\eta^2 = 0.127$; Bonferroni correction for group contrast TDT $p = .014$]) We found using a multivariate analysis of covariance (MANCOVA) to control for age differences that these significant differences in emotional profile scores between the disease groups [$F(1,43) = 4.408$; $p = .042$; partial $\eta^2 = 0.537$] and also between

those currently receiving medical treatment versus not currently receiving treatment [$F(1,44) = 4.341, p = .043, \text{partial } \eta^2 = 0.531$] remained significant after controlling for age.

4 Discussion

To our knowledge, this is the first study to examine the associations between self-report and projective drawings to understand disease-related pain coping in a mixed-condition pediatric sample. In this study, we examined the relationships between coping strategies and coded projective drawings in a sample of three diseases cohorts: oncological, rheumatic, and cystic fibrosis. Although previous studies have emphasized associations between pain and coping strategies or emotion regulation profiles, the integration of these constructs has received limited attention (Compas et al., 2014).

Our analyses revealed that pain coping strategies and projective drawing profiles differ across various clinical factors. Having more recently received a diagnosis as well as having cancer and more frequent day hospital admissions were related to lower pain coping strategy scores. Having cancer and receiving a diagnosis greater than 1 year prior were related to high pain coping scores. Furthermore, chronic illness diagnostic category and perceived family support were significant predictors of the use of both overall pain coping strategies and distraction-specific strategies. With respect to project drawing profiles, children with cancer diagnosis displayed poorer self-representation, and youth currently receiving active medical treatment displayed higher self-representations.

Our primary objective was to test how the coping strategies were related to emotional profiles as obtained through coding projective drawings of youth with chronic illnesses. We hypothesized that more positively coded emotional profiles with evidence of adaptation and well-being would correspond to greater use of coping strategies. We found a relationship between family-representation and the development of the Overall strategies and the distraction strategies for all three disease cohorts. These findings not surprising given the many illness-related factors, including family support, may impact youth emotional functioning (Palermo, Harrison, & Koh, 2006). In further support of this finding, some authors (Anthony & Schanberg, 2003) proposed a model in which family environmental variables, child psychological variables, and disease variables interact in the development of children's pain experience.

Specific studies on the relationships between anger, coping strategies, mood, and psychosomatic aspects in children and adolescents (Miers et al., 2007) led us to consider the emotional functioning as an important aspect in the management of pain and experiences related to having a chronic illness, such as anxiety, anger, fear, or sadness (Shepherd, Woodgate, & Sawatzky, 2010). In accordance with these previous studies, we have found that self-representation in children with

cancer is less positive than either children with rheumatological conditions or those with cystic fibrosis. A seemingly incongruous finding is that children in medical therapy/treatment perceive themselves more adequately than those that are not. One possible explanation for this finding is that for youth, currently receiving medical intervention may feel like a more proactive way of coping with their illness, while those that are off therapy may perceive their management of their disease as passive or detached. We also explored differences in coping strategies used in different age groups. In contrast to some previous studies (Compas & Boyer, 2001; Zimmer-Gembeck & Skinner, 2011) but in agreement with others (Aldridge & Roesch, 2007; Campbell et al., 2009) in our sample we did not find a significant interaction between coping strategy use and age group, although the average scores for each strategy were consistently lower in the group of the preteens (although not statistically significant).

Our second objective was to understand the different coping strategies among the diseases. In contrast with our previous hypothesis, the elapsed time from diagnosis, in itself is not a factor influencing coping strategy use. However, cognitive strategies in general were lower on average in children assessed within the first 6 months after receiving a diagnosis compared to those who were in a period of between one and two years post-diagnosis. These findings suggest that we should consider each phase of a chronic illness as having potentially different challenges and stressors for the children and their families (Compas et al., 2012). This influence of time since diagnosis on coping was not confirmed in our sample as regression analyses with time since diagnosis, the type of diagnosis, and perceived family support found that only diagnosis and family support perceptions were predictive of both overall coping and the use of distraction coping strategies. Our results are somewhat surprising, given that they suggest an inconsistent pattern of the association across different coping efforts and psychological adjustment over the course of children's treatment and recovery. For example, social, cognitive, and problem-solving coping were not influenced by time since diagnosis or age in our sample, although these strategies have been shown to be beneficial in several studies (e.g. Compas, Banez, Malcarne, & Worsham, 1991). It is unclear why some strategies of coping may be related to poorer adjustment in the different phases of the diseases and better adjustment later, but we did find that children's perceptions of themselves and their family's support could be key intermediary variables, as we found group differences based on diagnosis and current treatment status in how adequately children perceived themselves and their families.

Some limitations within this study should be considered as well. First of all, despite several studies apparently attesting to the clinical and diagnostic usefulness of children's drawings, we still need to improve the reporting of projective tests' reliability and validity (Kortelnuoma, 2009). The criticisms about projective tests have focused on projective tests' lack of reliability and validity (Motta et al., 1993; Lilienfeld, Wood, & Garb, 2000), the weak and inconsistent research base, their

susceptibility to contextual and situational factors (e.g., examiner's mood or biased use of language), and subjectivity in scoring and interpretation (Lilienfeld et al., 2000). We attempted to counteract these concerns by building a structured coding system and using multiple coders with 95% agreement. A further limitation was that our retrospective pain assessment was limited only to pain intensity ratings and did not capture other important dimensions of pain such as memory of pain, duration and frequency of pain, or pain descriptors. Another limit lies in the study of pain coping of children and no data was collected on parents to understand their behavior toward the ill child or their psychological reaction to caring for an ill child. In fact, in the literature it is well-known (i.e. Goubert, Vlaeyen, Crombez, & Craig, 2011, Palermo, Varlie, & Karlson, 2014) the central role of social modeling as a fundamental way to learn adaptive behaviors or maladaptive pain coping that promote disability or recovery, observing the behavior of a pain model (Goubert et al., 2011). It is therefore clear the need to investigate the impact of family and cultural beliefs on how children learn to react to pain (Edwards et al, 2001; Fortier et al, 2009, as cited by Azize et al., 2014). Another limitation is that the study did not follow the power analysis. After the study was completed, a calculation of the size of the post-hoc sample was conducted to determine the effect size of our sample of patients recruited ($n = 47$), using GPower 3.1.9.2 (Faul, Erdfelder, Lang, and Buchner, 2007). Given our sample, the analysis suggested that the evidence would have a statistical power of 0.26 (small effect size).

Despite the limitations, we strongly believe that the proposed method will increase our knowledge by providing specific information on the emotional state and coping styles of these children. Of course, further research is needed in future to confirm our findings and to extend the type of data collected, perhaps with a single specific population, given the pilot study sample size.

CHAPTER 4

COPE WITH PAIN IN CHILDREN AND EARLY ADOLESCENTS WITH MALIGNANT HEMATOLOGIC CANCER

*“One thing you can't hide - is when you're crippled
inside”*

~ John Lennon

From the previous study experience (see chapter 3), in this chapter I will present another study with a single cohort of 30 children with malignant hematologic cancer. The protocol was similar, with several improvements in the scoring methodology of projective drawings, (we also made a comparison with a non-clinical group) and addition of other assessment variables related to this single cohort of patients to test the new model that I developed. The combination of all these tests with a clinical interview and several type of observations within a mixed-method system of analysis have increased the understanding of the coping profiles, the internalizing problems and difficulties faced by children with cancer.

1 Background and main purpose

The risk of developing cancer in childhood is around 1 in 500, with leukemia being the most common diagnosis (Parkin et al., 1998; Ward, DeSantis, Robbins, Kohler, & Jemal, 2014). As a common symptom experienced by children with cancer (Collins et al., 2000; Collins, Stevens, & Berde, 2008; Moody, Meyer, Mancuso, Charlson, & Robbins, 2006), pain is a multifaceted issue and a challenging concern in pediatric patients with hematological malignancies. Pain can originate from several sources, including diagnostic and treatment procedures (e.g. venous punctures, spinal puncture, bone marrow biopsy, but also chemotherapy), underlying malignancy, or other factors not associated with the disease process, beside being strengthened by fear, anxiety and uncertainty that cause emotional distress (Menossi, Lima, & Corrêa, 2008).

All children with cancer experience pain, which is related to their disease or to the treatment, and over 70% of them sometimes present severe pain (WHO, 1998, as cited by Menossi, Lima, & Corrêa,

2008). Data from a wide survey in an heterogeneous population of children with cancer aged 10–18 years, showed that pain was the most prevalent symptom in the inpatient group (84.4%) and was rated as moderate to severe by 86.8% and highly distressing (“quite a bit to very much”) by 52.8% of these children (Collins et al., 2002). Each child experiences this painful feeling in a different way, considering several factors: previous pain experiences, beliefs, sociocultural context, temperament, coping skills, emotional functioning and situation at the time. The direct expression of pain or a partial denial of it, will be a consequence of these particularities (Schechter, Berde, & Yaster, 1993, as cited in Menossi, Lima, & Corrêa, 2008).

The assessment of feeling or emotion,- that is essentially a subjective attribution – is important, because affects primarily cause and consequence of relevant phenomena such as coping, symptoms, social activity and satisfaction among others (Watson, Clark, & Tellegen, 1988, as cited in Estévez-López et al., 2016). In the light of the multiple dimensions involving the phenomenon of pain in pediatric cancer, this study has the purpose to illustrate how children and adolescents with acute leukemia or lymphoma cope with pain.

2 Conceptual framework

The pain's appraisal process and accompanying feelings form an integrated and dynamically changing behavioral pattern and requires both clinical and research practice to shift from seeking information about children to seeking information from them (Docherty & Sandelowski, 1999). The model that we are going to present, considers individual parents variables (e.g. prior or concomitant problems, parental behavior in response to child's pain episodes) as a mediators/moderators within a broader context that consider both child factors (pain-coping skills, self-perception, perception of the family support) and those related to the disease factors (phase and type of treatment, if there has been a relapse, time since diagnosis) which are further embedded within the more broader socio-demographic factors such as parent and child age and sex and socioeconomic status. The following provides a heuristic model guided by clinical and theoretical experiences.

Family and parent factors as possible mediators/moderators. The relationship between parent and child pain is likely characterized by complex, bidirectional influences operating in the parent-child environment (Evans et al., 2008). Parental responses to their children's affective signals and the quality of support provided during painful episodes, and broadly, during the disease, provide the critical context, as an ongoing transaction, within which children organize and regulate their emotional experiences. It is possible that parents' feelings of inefficacy or disinterest, resulting from concomitant problems (separation, sibling with other problems, personal connections with difficult issues) are impacting on their children's efficacy beliefs in managing their pain (Page & Blanchette, 2009, Palermo & Chambers, 2005; Vervoort et al., 2009). Closely linked to these considerations is the

need to understand how the individual factors versus dyadic and family level factors may impact on pain and functional outcomes. Therefore, a complex relationship is proposed among family factors, child factor and factors related to disease and pain.

Child factors and disease-related variables. Adaptive coping skills are functional to effectively manage pain and enhance child emotional distress and Health-Related Quality of Life (HRQOL).

In children with acute lymphoblastic leukemia (ALL) and even those with brain tumors, treatment regimens consisting of high-dose chemotherapy and radiation therapy delivered to the brain are often associated with neurocognitive morbidity and are particularly stressful on the entire organism (Askins & Moore, 2008). Neurocognitive sequelae are most apparent in the domains of attention, memory, visuospatial abilities, executive functioning, cognitive processing speed and the highest risk factors for these declines seem to be the younger age at diagnosis and female gender (Fouladi et al., 2005, Lahteenmaki et al., 2007, as cited by Askins & Moore, 2008). However, research findings have been inconsistent regarding the domains affected and the degree to which they are compromised (Campbell et al., 2007).

Even if impaired neurocognitive functioning is one increasingly recognized long-term consequence of childhood ALL treatment, it is uncertain if these impairments affect even social learning processes, and thus, change also the self-perception in term of efficacy and self-esteem and the perception of family support (by parents, siblings or other significant caregivers). Alongside factors highlighted, the possibility of attended school normally and the support from classmates/friends can represent influential variables. Within this context, all these social and emotional aspects, are relevant but maybe difficult to investigate.

Drawing tests used in clinical practice with pediatric patients with cancer: measurement issues. The child's self-report (i.e., what the child says) has generally been considered to be the 'gold standard' for assessments of pain and related emotions despite its obvious limitation such the different level of cognitive development and possible poor language abilities (Chambers & Craig, 1998, as cited by Aminabadi et al., 2011). Further, since medical treatments administered to children with cancer can have effects on neurocognitive function, it can be difficult to isolate a child's pain experience from other emotional states.

From a clinical perspective, the use of a battery of projective drawings to examine family representations and self-perceptions of children who have/have had pain, could offer relevant indicators for identifying specific areas of fragility, i.e. poor adjustment or emotional indicator of distress, which could be directly addressed by an intervention to enhance parenting and/or coping reinforcement (Pace, Zavattini, & Tambelli, 2015).

A recent systematic review of the utility of data collection methods employed when undertaking research with children, teenagers and young people with cancer found that conventional methods of

data collection include questionnaires, one-to-one interviews and focus groups. However, in recent years, more novel methods have been tested, i.e. the use of participatory methods or digital technologies. Video diaries are a way to record and communicate their experiences and the use of art and photography based techniques have also proved successful with younger children with cancer (Flanagan, Greenfield, Coad, & Neilson 2015).

Starting from these considerations, drawing techniques, referring to the type of expressive methods in art-therapy or in diagnostics field as a method based on psychological mechanism of projection (alone or in combination with interview) were used to investigate several areas in this population. For instance, to depict the phenomenon of fear and the efforts to cope with the anxiety induced from all kinds of suffering: physical pain, mental anguish and spiritual agony in child and adolescent cancer patients (Nowak, 2014). To investigate how children, aged 3 to 9 years, undergoing cancer treatment describe their experience of comfort (Ångström-Brännström & Norberg, 2014) while Knighting, Rowa-Dewar, Malcolm, Kearney, & Gibson, (2001) employed them to investigate children's views of cancer and health behaviors (aged 8 to 11 years). Also this method was adopted as a way of enhancing communication around the issue of cancer (Di Gallo, 2001; Oppenheim, & Hartmann, 2003; Rollins, 2005), while Gibson, Richardson, Hey, Horstman, & O'Leary (2005) used it as platform to discuss sensitive issues around cancer with a sample aged 4 to 19 years. Furthermore, it can be helpful for determining the impact of cancer on the family as perceived by the child with cancer (Bossert & Martinson, 1990).

As to the use of projective techniques, there is not a shared agreement about hurt/pain/suffering in cancer patients. For example, Ammen et al. (1996) revised the color/affect pairings that are used with Color-Your-Life (CYL), a technique that involves teaching children color-affect pairs (e.g. black-scared) and instructing them to fill a blank piece of paper with the colors that represent the feelings of their life. In their non-clinical sample of 172 children, (ages 6 to 13), these authors eliminated the feelings "hurt" and "sick" because they affirm that these were physical sensations rather than emotions. Another study that used the same technique (CYL) in a sample of 40 children aged 6 through 12, found differences in style of drawing, percentages of color/feeling represented in the drawings between pediatric cancer patients and a control group, suggesting a different representation of their feelings. Specifically, patients expressed fewer sad feelings (blue), hurt feelings (brown), scared feelings (black), and anxious feelings (orange) in their drawings compared to the control group's members. (Boley, Peterson, Miller & Ammen, 1996).

These reflections highlight the necessity of using new and appropriate self-report measures. Therefore, it seems to be useful the use of "paired tools", such as questionnaires and/or scales, a behavioral observation scale with projective drawings, that, as mentioned earlier, are a nonspecific

test for pain, but its familiarity and simplicity make it an invaluable screening test of general well-being.

As Pace, Zavattini, & Tambelli (2015) claim, this approach could offer relevant indicators for identifying specific areas of fragility, i.e. poor adjustment or emotional indicator of distress, which could be directly addressed by an intervention to enhance parenting and/or coping reinforcement.

Being a part of the projective methods used in dynamic models, drawing tests application, follows the idea that drawing can reveal affective dynamics and is based on the projection principle, i.e. “bringing outside” one’s experiences, notions, aspirations, etc. In their graphic representations, children do not reproduce reality faithfully, nor do they depart from reality on purpose. Like a narrative, the drawing is a “construction”, and is thus a way of “communication” of the inner representational world and its affective implications through internal working models that cannot be assimilated literally with real events (Tambelli, Zavattini, & Mossi, 1995). As Venger & Morozova (2011) point out, specific projective drawings test reflect first not the conscious dispositions of the person, but his or her unconscious impulses and experiences

Finally, speaking about the complexity in scoring drawings, Failo (2015), in his narrative review on the use of drawings for understanding children’s pain better, found that numerous efforts to standardize drawings as a projective technique have been undertaken, yet simultaneously supported and challenged over the years. Despite these controversial studies, the Draw-A-Person, Child Drawing Hospital, House Tree Person test and Kinetic Family Drawing continue to be regularly used as a part of a general psychological test battery in many settings. As regards validity and reliability, it may be affirmed, as Rollins (2005) suggests, that a series of drawings by the same individual demonstrates constant structure and form, although content may vary. Most likely the most reliable method to interpret children’s drawings is in combination with other information such as that derived from interviews or writing tasks (Failo, 2015).

3 Research questions and hypotheses

The first goal of this study was to compare the emotional characteristics of children with malignant hematologic cancer and a control group of healthy children in the Draw-a-Person Test, using the Koppitz's 30 emotional indicators (EIs) (1966, 1968, 1984) and the sense of family as an internalized system of relations, in the Draw-a-Family Test, through a procedure developed by Tambelli, Zavattini & Mossi (1995, 1996)

We hypothesized (assumed) that the different representations of themselves and the levels of coding in the family representation drawing can give us other information concerning the self-perception and perception of the world as a network of interconnected relationships. We have also hypothesized that the number and the type of emotional indicators could be different between the

two groups, as the research with other disorder have shown (Dağlioğlu, Deniz & Kan, 2010; Dolidze, Tchanturia & Smith, 2013; Dutta & Sanyal, 2016; Perets-Dubrovsky, Kaveh, Deutsh-Castel, Cohen & Tirosh, 2010).

All these points could be useful to identify a scoring systems and/or graphic indicators of children's drawings that are indicative of emotional distress and different adjustments.

The second aim was to increase understanding of clinical profiles of the ways pediatric patients with cancer cope with pain during or following period under cancer treatments using a multi-method assessment approach, and to begin to identify factors that place children at risk for both acute and chronic pain. The central hypothesis is that coping factors and emotional well-being quality are associated with children's perception of support provided by parents and during the treatments.

We have also hypothesized that the phases of chemotherapy treatment would be related to different coping skills and emotional traits, considering that literature on these topics is lacking.

4 Method

4.1 Setting, participants and procedures

This study was conducted in the Hemato-Oncology Outpatient Clinic. Department of Pediatrics University of Milan-Bicocca, Monza. Thirty child aged from 7 to 14, served as participants, a sample enrolled in this observational study of a single cohort of pediatric patient with a hematological malignant cancer (Acute Lymphoblastic Leukemia, Acute Myeloid Leukemia, Hodgkin's lymphoma and non-Hodgkin's lymphoma).

The children were chosen through purposive sampling, in which clinical researchers recruited all children meeting the selection criteria for the study. In order to be contacted to participate in the study, children had to be between 7 and 14 years of age with hematological malignant cancer, and accompanied by a parent/guardian. Participants were excluded from the study if they did not speak Italian as a first language or had been diagnosed with specific developmental delays. Following enrollment, no families withdrew from the study and no adverse events were reported.

Participants in the control group (only for the projective test section) were randomly recruited from schools in the city of Rovereto. The inclusion criteria for the control group were that children were healthy with no medical complications and matched the age range of the children from the clinical sample.

The procedure was fully explained to children and their parents. Informed consent was obtained from all participants under the local institutional review board (IRB) approved protocol. Parents were allowed to remain with their child during the interview if desired by either the child or the parent. Children completed a questionnaire, projective drawings battery within a specific

observation during the interview process in a single session. The researcher ensured that the entire protocol lasted between 25 and 55 minutes to minimize fatigue. It was stressed to the parents that whether or not they participated as subjects in the research their treatment would not be affected. All participants had access to psychosocial care according to hospital standards and guidelines.

4.2 Research instrument and measures

In seeking to understand how the recurrent pain affects these children from a psychosocial point of view, we started from the consideration that noninvasive and discrete testing may be beneficial, particularly when attempting to identify emotional distress in children and younger adolescents.

4.2.1 Demographics

All demographic and medical information was extracted from the psycho-social and medical records. Information gathered concerning the child were: age, gender, time since diagnosis, type of malignant hematologic cancer, type and phase of treatment, if there has been a relapse, hospital admission, current ongoing medical intervention. Sociodemographic characteristics of child's mother and father: age, education level, employment status, occupation, other concomitant problem, other siblings). The socioeconomic status (SES) of the parents was calculated with the Four-Factor Index of Social Status (Hollingshead, 1975; Rossi, 1994). For more details see Table 1.

4.2.2 Pain intensity

Children used the numerical rating scale (NRS-11) and the Wong-Baker Faces Pain Scale (WBS) to rate their current pain and average of all pain episodes when they were in hospital. For more details, see the study in Chapter 3.

4.2.3 Questionnaire related to social variables and dyadic interactions during episodes of pain

This questionnaire, specifically developed by the research group, (adapted from a work of Mathews, 2012) was administered to the two psychologists which followed the family during the illness course. The coders' psychologists met to discuss the interactions and eventually coding discrepancies.

The measure included five specific subscales: 1) School attendance on a 3-point scale, which is scored from 1 "yes, with regularity", 2 "not always", 3 "no, just hospital school". 2) Constancy of support from friends and classmates during hospitalization, which is scored on a 3-point scale. 3) Explanations provided by health professionals during the therapies / painful procedures (scored from 1 to 3). 4) Observed parent acceptance of child's emotion during painful episodes. Psychologists rated the parent's reactions (usually mother) to children's emotional expressions on a 5-point scale. Higher scores indicated reactions to child's emotional expressions that were characterized by openness, acceptance, being emotionally available, and validation of the child's emotions and point of view. Lower scores indicated non-accepting reactions to child's emotional

expressions that were characterized by dismissing, or distressed reactions, being emotionally unavailable to the child, and no validating the child's emotions and point of view. 5) Observed quality of parent's advices provided to the child when he/she had to face pain on a 5-point scale. Higher scores indicated parent advice that was characterized by encouragement to actively problem-solve, face the painful situation, and have confidence in his/her ability to effectively cope with the situation. Lower scores were characterized by parent's encouragement of avoidance or lack of encouragement to actively problem-solve and his/her lack of encouraging the child to have confidence in his/her ability to effectively handle the situation. In addition, at the end of the questionnaire there was a space left for comments.

4.2.4 Observation during protocol-interview

During the entire interview process, the researcher took notes to evaluate the kind of behavioral attitude of the child (adaptation from Roberti, 2014). For each child, we observed the attitude assumed when receiving instructions, while doing the questionnaire and while drawing (e.g. refusal, excessive speed, anxiety, concerns etc.). It has been rated in four cluster: 1) Collaborative/exploratory: when the child showed curious and open toward the tests). 2) Collaborative/non exploratory - when he worked quietly, but showed a closure to the recognition of himself (evasive answers, rigid and controlled behavior). 3) Resistant - when the child showed doubts and misgivings towards the test (e.g. why do I need this test, I do not believe in the test). 4) Refusing - child refused to participate and exhibited verbal assault or behaved in a passive-aggressive way.

4.2.5 Pain-coping skills

Pain-coping skills were assessed by the Waldron/Varni Pediatric Pain Coping Inventory (PPCI) – Child form, with Italian standardization (Bonichini & Axia, 2000). The questionnaire features 29 items and four open-ended questions and includes four specific subscales, including Cognitive Self-Instruction, Problem-Solving, Distraction, and Seeks Social Support. The rating scored on a three-point Likert scale [0 = never (not at all), 1 = sometimes, or 2 = Often (a lot)] for the closed-items. The open-ended questions were classified by cluster analysis. For more details see the study presented in Chapter 3.

4.2.6 Emotional adjustment

Adjustment-related emotional well-being was conceptualized using the Draw A Person test (DAP) and Draw A Family test (DAF) as part of the self-concept adaptive mode. Self-concept can affect role functioning and interdependence and can be related to physical pain. The use of projective tests as part of an overall test battery can be useful in helping children overcome communication barriers and make a discrimination between children who need additional well-being adjustment examination (Tielsch & Allen, 2005; Waweru, Reynolds & Buckner, 2008). Two primary methods of interpreting children's drawings are used in this study: one focused on identifying specific features of

the child's drawing and the other adopting a global approach with an integrative scoring system that we have tested in a previous study (see chapter 3 for more details). We believe that the combination of both methods can define an algorithm that can measure representations of the child's psychological functioning. In the conclusive section, we will try to discuss, if it is possible, with drawing, to assess basic defense mechanisms.

The Draw A Family test (DAF) by Corman (1964, 1965, 1967) was conceived and developed for youth "aims to investigate the relationships that the individuals have established with other members of the family group, as well as the problems they have with the group as a whole and/or certain individuals" (Pasta, Prino, & Gastaldi, 2014, p.30).

Two existing scoring methods were combined to create an assessment method based on a global approach to the family drawing aimed at assessing the perception of the Family representation during the phases of chemotherapy treatment as a child's support psychological adjustment.

The first method, the "integrative drawing scoring system", from the Family Drawing Test task was developed by Failo, Beals, Venuti (see the previous chapter) as a global approach that involve judging of all drawing and ranking features on a 0-4 Likert scale, where 0 is "not adequate" and 4 is "very adequate representations" and it was applied to the formal-structural and content aspects. The higher the score, the more well-adjusted (we judged) the self-representation or family perception. This method was tested with a group of 47 individuals aged between 7 and 14 with chronic illnesses. The second scoring method (used), the Family Drawing Test was developed by Tambelli, Zavattini, & Mossi (1995). It consists of 9 scales based on the characteristics of characters, the links between them, and the activities they share in the drawing. Specifically, we proceeded to compare the real family with the family depicted in the drawing (by analyzing omissions and additions), to determine the role of identifications and, in accordance with Corman's "valorization/devalorization" concept, to rank the significance assigned to the characters depicted on three nominal scales (i.e., priority, size and importance) (Pasta, Prino, & Gastaldi, 2014). This information was obtained also from the questions that were asked when the children finished their drawing, which concerned their preferences and identification with the people represented in the drawing (for each person represented, they were asked to indicate name, gender, age, role in the family, what they do and where they are, and who is the most or least liked, happy or sadness. The method was validated among 5,619 healthy children aged between 6 and 13 and was used because it is the most standardized scoring method of drawing and meets at least partially, a number of important criteria regarding validity (Tambelli, Zavattini, & Mossi, 1995).

For its proven effectiveness as a screening tool and its time and cost effectiveness, the Draw A Person (DAP) was chosen as one of the two screening tools for this project.

Also for this graphic method we followed two scoring methods: a global approach (same rating system of the DAF developed by Failo, Beals, & Venuti) and a second one utilizing a list of 30 emotional indicators (EIs) as objective signs that reflect children's worries or anxieties, developed by Koppitz (1968; 1984). According to Koppitz, two or more such indicators are highly suggestive of emotional problems (Koppitz, 1968). Some examples of emotional indicators are poor integration of parts, asymmetry in legs, short arms, small figures, figures without hands or with partial hands (Koppitz, 1968, 1984; Carrol & Ryan-Wenger, 1999).

From this emotional indicator, we have also examined differences in the scores of the emotions/behaviors categories proposed by Koppitz (1984) and repurposed by Dağlioğlu, Deniz, & Kan, (2010) for classifying the indicators according to their presumed underlying emotional symptom (impulsivity, insecurity/inadequacy, anxiety, shyness/timidity, anger/aggressive).

4.3 Analysis plan

The results were analyzed using the Statistical Package for the Social Science (SPSS, V.21.0; IBM Corp., Armonk, NY). In the light of the two main goals of this study, we planned two kinds of analysis: one only on the clinical group and another to compare the clinical group with a control group. Each group was assessed within itself for indicators.

For the analysis between the clinical group and the healthy group, given the number of the participants (30 + 30) and because some cells had less than 5 values, the data were analyzed using non-parametric methods (e.g. Mann–Whitney U-test, Spearman's rho, Fisher's Exact test), because they do not require that the population from which the sample is taken is assumed to be normal (Siegel & Castellan, 1988). To examine the emotional indicators and the object relationships within the family system, both derivable from the drawings in the clinical and in the healthy group, we used a Chi-Square test in crosstabs to estimate the possible socio-demographic differences between the two samples and to better understand the samples comparability. The two groups were matched by age. Descriptive measures were computed for all relevant variables and comparisons were made between the two groups.

The data analysis involved descriptive statistics and content analysis for the themes of the drawings. Data from the semi-structured interviews were linked to the themes of the drawings and were scored independently by two raters and processed through triangulation into quantitative scores

The emotional indicators included in the drawings were correlated with each other and analysed with the Koppitz method (Koppitz, 1968; Tielsch & Allen, 2005). The two raters scores for the emotional indicators were highly correlated (Cohen's Kappa = .80, $p < .05$). The indexes of object relationships were also correlated with each other and analysed with the Tambelli, Zavattini, & Mossi's method (1995, 1996). The Family Drawing Indexes have had a good agreement between the two evaluators as well (Cohen's Kappa = .75, $p < .05$).

To plan the analysis within the clinical group, data were first examined for skewness, kurtosis, outliers and normalcy (Shapiro-Wilk): no transformations were necessary as the distribution was normal for all dependent variables considered.

Descriptive statistics were run. Then, Pearson's bivariate was run to show any existing correlations between variables. A series of ANOVAs was performed to assess differences in self- and family-representations and coping strategies, and also to identify the differences between coping strategies and time since diagnosis, age at diagnosis, pain intensity (present and past), relapse, ages groups and sex.

To test the predictive influence of phase of treatment, medical therapy and other important variables on prior pain (past), a series of Linear regression analysis was conducted. We have also run other regressions to test the hypothesis that if there are other psycho-social variables that can influence coping strategies and emotional indicators of well-being and adjustment.

Thematic content analysis (Boyatzis, 1998) was also used to derive additional information on when child has in pain from the open-ended questions at the end of the PPCI questionnaire.

5. Results

5.1 Sample description

Thirty children were enrolled including 18 males (60%) and 12 females (40%) between 7 and 14 years of age ($M = 10.00$ $SD = 2.28$).

30 children and young adolescents with cancer, aged 7–14, were matched with a control group ($M = 9.84$ $SD = 1.44$).

Parents and children were Caucasian, and middle class for both groups as it can be seen from the S.E.S. ($M = 37.98$, $SD = 14.43$ vs $M = 42.88$, $SD = 14.34$). Most of the parents had completed college or higher education. Conversely, the parents' occupational status changed between mother and father and within groups. The number of siblings was almost the same for both group. Socio-demographic characteristics of parent and child are described in Table 1.

Other descriptive information from the clinical group are that 86.7% ($n = 26$) of children had not faced a relapse. Children were in different phase of treatment: Induction (30%), Consolidation (13.3%), Re-induction (13.3%), Bone marrow transplant (13.3%), Maintenance (16.7%) and Stop treatments (13.3%). Most of the children (73.3%) had concomitant psycho-social problems (e.g. other disease, a disease in the family, death of close relatives, recent divorce of parents).

Children reported low present pain intensity ($M = 1.57$, $SD = 2.06$) but their rating of prior pain (i.e., average of all pain in hospital) was moderate to severe ($M = 6.53$, $SD = 3.32$). Medical Characteristics of the clinical group are described in Table 2.

Table 1. Socio-demographic characteristics of parent and child

Parent and Child Demographic Characteristics	Clinical group N = 30	Healthy group N = 30	χ^2	p
Mother's age (M_{years}, SD)	42.07 (4.89)	47.17 (5.84)		
Father's age (M_{years}, SD)	46.08 (5.49)	45.95 (4.78)		
Mother's Education (%)			6.98	.032
- 5 years of schooling	0	0		
- 8 years of schooling	5 (16.75)	1 (3.3)		
- 13 years of schooling	12 (40)	14 (46.7)		
- >13 years of schooling	13 (43.3)	11 (36.7)		
- Not reported	0	4 (13.3)		
Father's Education (%)			4.33	.366
- 5 years of schooling	1 (3.3)	0		
- 8 years of schooling	7 (23.3)	3 (10)		
- 13 years of schooling	10 (33.3)	13 (43.3)		
- >13 years of schooling	10 (33.3)	9 (30)		
- Not reported	2 (6.7)	5 (16.7)		
Mother's Occupational status (%)			16.58	.002
- Househusband/unemployed	3 (10)	1 (3.3)		
- Blue-collar workers	18 (60)	6 (20)		
- Clerks/professionals	2 (6.7)	10 (33.3)		
- Manager/Doctors	7 (23.3)	9 (30)		
- Not reported	0	4 (13.3)		
Father's Occupational status (%)			3.526	.474
- Househusband/unemployed	2 (6.7)	0		
- Blue-collar workers	12 (40)	10 (33.3)		
- Clerks/professionals	6 (20)	6 (20)		
- Manager/Doctors	8 (26.7)	9 (30)		
- Not reported	2 (6.7)	5 (16.7)		
S.E.S (M_{years}, SD)	37.98 (14.43)	42.88 (14.34)	4.44	.217
- Low - lo thru 26.5 (%)	6 (20)	4 (13.3)		
- Medium - 27 thru 44.5 (%)	15 (50)	11 (36.7)		
- High - 45 thru hi (%)	9 (30)	12 (40%)		
- not calculable	0	3 (10%)		
N° Siblings (%)			2.59	.459
- 0	6 (20)	3 (10)		
- 1	18 (60)	16 (53.3)		
- ≥2	9 (30)	11 (36.7)		

Children's Gender (%)		
- Male	18 (60)	14 (46,7)
- Female	12 (40)	16 (53,3)
Children's Age (M_{years}, SD)	10.00 (2.28)	9.84 (1.44)
- 7-10 years (%)	18 (60)	18 (60)
- 11-14 years (%)	12 (40)	12 (40)

Table 2. Medical Characteristics of the clinical group

Medical Characteristics of the clinical group	N = 30
Present pain (M, SD)	1.57 (2.06)
Past pain (M, SD)	6.53 (3.32)
Age at diagnosis (M_{month}, SD)	103.53 (34.20) range 44-140
Time since diagnosis (M_{month}, SD)	16.50 (16.83) range 2-55
Diagnosis (%)	
- Acute Lymphoblastic Leukemia	23 (76.6)
- Acute Myeloid Leukemia	4 (13.3)
- Hodgkin's Lymphoma	2 (6.7)
- non-Hodgkin's Lymphoma	1 (3.4)
Phase of treatment (%)	
- Induction	9 (30)
- Consolidation	4 (13.3)
- Re-induction	4 (13.3)
- Bone marrow transplant	4 (13.3)
- Maintenance	5 (16.7)
- Stop	4 (13.3)
Relapse (%)	
- Yes	4 (13.3)
- No	26 (86.7)
Concomitant psycho-social problems (%)	
- Yes	22 (73.3)
- No	8 (26.7)

5.2 Differences in emotional well-being and in the internalized system of relations in youth with cancer compared to otherwise healthy children

This section presents the findings of the procedures conducted to understand which are the differences of children's "self-concept" and how they see the family, as a prototype of the closer network of interconnected relationships that a child usually has.

We have investigated these different relationships, in youth with pain due to the illness compared to otherwise healthy children through their drawings of a person (DAP) and of a family (DAF). All results are synthesized in tabular form.

From the observations, most of the children, during the entire interview process and also while drawing have showed a "collaborative/exploratory" behavior (73.3% in clinical group and 86.7% in healthy group). Only 26.7% sick children (and the 13.3% of healthy) have adopted a "collaborative/non exploratory" behavior and nobody showed "resistant" or "refusing" behaviors.

Children self-perception of well-being (DAP)

As regards the general observation during the process of drawing and from questions at the end of the process, we have found a difference between groups, where the clinical one showed more corrections (as a way to erase) ($\chi^2 = 4.043$; $p = .044$) and some stick figures instead of an entire body ($\chi^2 = 4.286$; $p = .038$). This index along with others (see the results (submitted) below), tend to suggest under-use and anxiety surrounding the body (Foley & Mullis, 2008). See Table 3 for more details.

Table 3. Distribution of general observational characteristics in children's DAP in the two groups

Observational Characteristics	Clinical group N = 30	Healthy group N = 30	χ^2	p
Who drew?			5.800	n.s.
- Himself/herself	2 (6.7)	8 (26.7)		
- Father/mother	6 (20)	2 (6.7)		
- Friends/siblings	11 (36.7)	9 (30)		
- Invented	11 (36.7)	11 (36.7)		
Excessive corrections			4.043	.044
- yes	6 (20)	1 (3.3)		
- no	24 (80)	29 (96.7)		
Age difference (with the real one)			2.411	n.s.
- yes	11 (36.7)	17 (56.7)		
- no	19 (63.3)	13 (43.39)		
More happy or sad?			1.964	n.s.
- happy	26 (86.7)	29 (96.7)		
- sad	4 (13.3)	1 (3.3)		
Stick figure			4.286	.038
- yes	4 (13.3)	0		
- no	26 (86.7)	30		

Table 4 presents the age and gender distribution of impulsivity-related characteristics in children's drawing a person. As can be seen, 26.7% of children in the clinical group and 6.7% in healthy group

omitted the neck in their human figures. The difference is statistically meaningful ($p < .05$).

Table 4. Distribution of impulsivity related characteristics in children's DAP in the two groups

Impulsivity	Clinical group		Healthy group	
	n° EI	%	n° EI	%
Poor integration of parts	6	20	3	10
Gross asymmetry of limbs	2	6.7	4	13.3
Transparencies	1	3.3	-	-
Big figure	2	6.7	-	-
Omission of the neck*	8	26.7	2	6.7%
Tot of impulsivity indicators**	19	63.3	9	30

* $\chi^2 = 4.320$; $p = .040$; ** $\chi^2 = 6.70$; $p < .001$

The table below (Tab.5) reveals that 16.7% of children in the clinical group did not completed the hands in their drawings ($p = n.s.$), while the percentages was 13.3% for the tiny head indicator ($p = n.s.$). Despite there is no significance, it is important to consider that none of these indicators was present in the drawing of the children's healthy group.

Table 5. Distribution of insecurity and inadequacy related characteristics in children's DAP in the groups

Insecurity/inadequacy	Clinical group		Healthy group	
	n° EI	%	n° EI	%
Slanted figure	2	6.7	-	-
Tiny head	4	13.3	-	-
Omission of hands	-	-	-	-
Monster or grotesque figure	1	3.3	-	-
Omission of arms	-	-	-	-
Omission of legs	-	-	-	-
Omission of feet	-	-	-	-
Hands cut off	5	16.7	1	3.3
Tot of Insecurity/inadequacy indicators**	12	40	1	3.3

** $\chi^2 = 11.880$; $p < .001$

As it can see in Table 6, no statistical difference was found between drawings' indicators that can reflect anxiety in either of the groups.

Table 6. Distribution of anxiety related characteristics in children's DAP in the two groups

Anxiety	Clinical group		Healthy group	
	n° EI	%	n° EI	%
Shading of face	1	3.3	1	3.3
Shading of body or limbs	1	3.3	1	3.3
Shading of hands or neck	1	3.3	-	-
Legs pressed together	1	3.3	3	10
Omission of eyes	-	-	-	-
Omission of body	-	-	-	-
Three or more figures drawn [‡]	-	-	1	3.3
Tot of Anxiety indicators	4	13.2	6	19.9

[‡] no specific clear note about the interpretation of this indicator in previous research, however it seems belonging anxiety's indicators.

As can be inferred from Table 7, the omission of the nose, which is an indicator of shyness and timidity, occurred more often compared to other characteristics. Although no statistical difference was found, it was noteworthy that the control group have more overall shyness' and timidity's emotional indicators that the clinical group.

Table 7. Distribution of shyness-timidity related characteristics in children's DAP in the two groups

Shyness/Timidity	Clinical group		Healthy group	
	n° EI	%	n° EI	%
Tiny figure	3	10	4	13.3
Short arms	3	10	7	23.3
Arms clinging to body	-	-	2	6.7
Omission of nose	9	30	8	26.7
Omission of mouth	-	-	-	-
Tot of Shyness/Timidity indicators	15	50	21	70

As can be seen in Table 8, no statistical difference was found between drawings' indicators that can reflect anger in either of the groups.

Table 8. Distribution of anger related characteristics in children's DAP in the two groups

Anger	Clinical group		Healthy group	
	n° EI	%	n° EI	%
Crossed eyes	1	3.3	-	-
Presence of teeth	1	3.3	1	3.3
Long arms	2	6.6	2	6.7
Big hands	4	13.3	-	-
Genitals	-	-	-	-
Tot of Anger indicators	8	26.5	3	10

The comparison between the drawings of the clinical and control group related to the frequency of occurrence of emotional indicators, reflects a significant difference (Mann-Witney U = 336.500; p (exact 1 tail) = .042). Also when we tested the same hypothesis with the global approach, we have found significant differences both for formal-structural DAP coding (Mann-Witney U=303.000; p (exact 1 tail) = .009) and for content-DAP coding (Mann-Witney U=279.500; p (exact 1 tail) = .003).

Children's perception of the family (DAF)

There are several factors to be taken into consideration when one examines children's perception of the family within the object relationships theory in relation to the family system. Most of the family drawings (DAF) had similar characteristics to the DAP in terms of pressure and type of stroke and used lines. As regards identification of desire, the response to the interview question: "Who would you like to be?" refers to a desire or tendency at the conscious level: children choose the person who best expresses their "confessable" aspirations. In fact, 40% of the sick children and the 43% of the healthy children, drew themselves, followed by parent or sibling, having the same sex as his or her own. This means that children identifies with the figures "who represent the power, value and models that children would like to make their own" (Abraham, 1976, as cited by Pasta; Prino, & Gastaldi, 2014, p.33). This highlights the sense of sexual belonging and the function it assumes in the organization of the Self. No statistical difference was found between the two groups (see Table 9).

Table 9. Distribution of Identification of desire in children's DAF in the two groups

Identification of desire	Clinical group		Healthy group	
	n°	%	n°	%
Self	12	40	13	43
Same-sex parent	3	10	7	23.3
No same-sex parent	1	3.3	-	-
Same-sex sibling	6	20	3	13.3
No same-sex sibling	3	10	1	3.3
Other/doesn't know	5	16.7	5	16.7

Comparing the actual composition of the families with the one depicted in the drawing, it was found that 60% of the sick children and 42.3% of the healthy children drew all their family without making changes.

In the remaining 40% of cases (56.7% in healthy group), the changes consisted in omitting people, especially siblings (16.7% vs 36.7%). However, on other hand, children without siblings added at least one brother or sister in 20% of cases (for both groups). The added figures, which like the omitted figures are here interpreted as different “identifications” of the subject, rather than as indicating unexpressed drives, are chiefly representative of children with whom there is a fraternal bond (20% for both groups).

No statistical difference was found between the clinical and healthy group (see Table 10).

Table 10. Distribution of omission and added figures in children’s DAF in the two groups

Omission figure	Clinical group		Healthy group	
	n° with gr	%	n° with gr	%
Self	1	3.3	1	3.3
Mother	1	3.3	1	3.3
Father	-	-	2	6.7
Siblings	5 (24)	16.7	11 (27)	36.7
Nobody	23	76.7	15	50

Added figure	Clinical group		Healthy group	
	n° with gr	%	n° with gr	%
Siblings	6	20	6	20
Other/animals	1	3.3	-	-
Nobody	23	76.7	24	80

As for the order (priority index) in which the figures are drawn, the father is often represented first in both groups (53.3% and 35.7%) (see Table 11, Priority).

By assuming that there is a link between the emotional resonance of a family member and the size of that member’s depiction, the father is also the most highly valorized figure in terms of height (79.3% and 60.7%), followed by the mother (24.8%) (see Table 11, Size).

Based on the number of attributes, the father is the figure who receives most attention among the sick children (58.6). Conversely, healthy children depicted themselves first (34.5) followed by the father (see Table 11, Importance).

Table 11. Distribution of priority, size and importance in children's DAF in the two groups

FD INDEXES	Clinical group					Healthy group				
	n°, (reference % with gr.)					n°, (reference % with gr.)				
	Self (100%)	Mother (96.7%)	Father (100%)	Sibling1 (60%)	Sibling2 (20%)	Self (100%)	Mother (96.7%)	Father (93.3%)	Sibling1 (53.3%)	Sibling2 (36.7%)
Priority	7 (24.1)	4 (14.3)	16 (53.3)	3 (15.8)	-	9 (31)	9 (31)	10 (35.7)	2 (11.8)	-
Size	1 (3.4)	5 (17.9)	23 (79.3)	-	-	2 (6.9)	7 (24.19)	17 (60.7)	-	2 (14.3)
Importance	5 (17.2)	4 (14.8)	17 (58.6)	2 (10)	-	10 (34.5)	5 (17.2)	9 (32.1)	1 (5.9)	-

In the response to the question “who is the happiest?” and from drawings observation, a difference between the clinical group was found, where the father is the last one happy, compared to the control group ($\chi^2 = 7.68$; $p < .01$). A generally positive emotional tone was instead found for all the other characters (self, mother, siblings). From the question “who is the less happy” no one difference was observed between and within groups (see below Table 12).

Table 12. Distribution of emotional tone in children's DAF in the two groups

FD INDEXES	Clinical group						Healthy group					
	n°, (reference % with gr.)						n°, (reference % with gr.)					
	Self	Mothe r	Father	Siblings	Everybody	Nobody	Self	Mother	Father	Sibling1	Everybody	Nobody
Happiest*	6 (20)	8 (26.7)	1* (3.3)	8 (26.7)	4 (13.3)	3 (10)	6 (20)	5 (16.7)	9* (30)	6 (20)	2 (6.7)	2 (6.7)
Less happy	5 (16.7)	4 (13.3)	9 (30)	3 (10)		9 (30)	7 (23.3)	7 (23.3)	5 (16.7)	4 (13.3)		7 (23.3)

* $\chi^2 = 7.68$; $p < .01$

Considering depicted closeness as an indicator of the dynamics of the affections, we evaluated “where” children draw themselves with respect to other family members. Most of the sick children (51.7%) and the 31% of the healthy children placed themselves near to one parent at least (see table 13), followed by siblings (20.7% and 27.6%), thus emphasizing the bond of mutual understanding and liking that connects children with their parental figures and siblings (Tab.13).

Table 13. Distribution of closeness in children's DAF in the two groups

Closeness	Clinical group (n=29)		Healthy group (n=28)	
	n° with gr	%	n° with gr	%
Mother	8	27.6	4	13.8
Father	7	24.1	5	17.2
Siblings	6	20.7	8	27.6
Other/animals	1	3.4	-	-
Between parents	1	3.4	5	17.2
Between siblings	2	6.9	1	3.4
Between siblings and parents	4	13.8	6	16.9

In conclusion, the comparison between the drawings of the clinical and control group through global approach (formal-DAF) showed a significant difference (Mann-Witney U = 263.000; p (exact 1 tail) = .001)

5.3 Characterization of pain-coping profiles and emotional well-being adjustments

Which child's pain-coping styles are adopted and which are the associations with pain intensity, disease and treatment factors and family factors?

To answer this question, we have preliminarily run descriptive statistics. Table 14 showed that style of coping with pain mostly used by children with hematologic cancers was seeking social support.

Pearson's bivariate correlations were computed between illness parameters, pain intensity, demographic variables, child's and family dimensions assessed and child's coping strategies.

Not all child's coping strategies were correlated with each other: Cognitive self-instructions strategies were significantly correlated with Problem Solving ones ($r = .409$; $p = .025$) and Social Support ones ($r = .508$; $p = .004$). Cognitive strategies were also negatively correlated with the socioeconomic status (SES) of the parents ($r = -.398$; $p = .033$). Distraction strategies were negatively correlated with pain intensity at present time ($r = -.398$; $p = .029$).

From the thematic content analysis, four overarching themes were examined: what I do, what I think, what I wish, what I ask. The description of the sub-themes represented in the children's writings is given below.

What I do that helps me when I hurts:

Pro-active behavior (n = 11; 36.7% - e.g. play, jigsaw, drawings about what I like, take a shower); nothing/alienation (n = 9; 30% - e.g. nothing, I cry and scream, I stay alone); seeking for social support (n = 10; 33.3% - e.g. I play with mum and pa, I go to mum and I tell her that I hurt).

What I do that helps me when I hurts:

Needs of emotional closeness (n = 22; 73.3% - e.g. I ask my parents to stay close to me and I like receiving cuddles, I ask the proximity of someone, If someone can give me support); passivity (n =

4; 13.3% - e.g. nothing, only if they let me watch a television program); some solutions (n = 4; 13.3% - e.g. medicine and cuddles, how can I stop the pain)

What I wish when I hurts:

Heal (n = 20; 66.7% - e.g. when I will feel better, I think I will heal soon, I will have no more pain soon), consolation / distraction (n = 8; 13.3% - e.g. it would be nice to have a dog with me, stay with friends, forget about it and play); helplessness (n = 2; 6.7% - e.g. nothing, none)

What I think that helps me when I hurts:

Fatalism (n = 10; 33.3% - e.g. it hurts and I can do nothing to stop it, the fact is that it hurts); feel better (n = 9; 30% - e.g. if can I take medicine to feel better, I think I don't feel pain so the aching will stop); think positive (n = 11; 36.7% - e.g. about all my friends, that I can make it, having fun and think it does not hurt so much).

In the treatment active phases (induction, consolidation, re-induction, bone marrow transplant), children report that they asked more solutions compared to those in a no active phase (20% versus 0%) and they had more negativistic attitudes and passive resistance (30% versus 0%).

Table 14. Descriptive statistics for the child's coping strategies

COPING STRATEGIES	N	Mean	SD	Range
Seek for social support	30	7.63	3.26	2-13
Problem Solving	30	6.47	2.93	1-13
Cognitive self-instruction	30	5.67	2.23	1-11
Distraction	30	5.30	3.03	1-12
Overall coping strategies	30	25.10	7.69	10-43

An ANOVA was performed with the dependent variables the time since diagnosis, age at diagnosis, pain intensity (present and past), relapse, age's groups and sex. The different coping strategies were included one by one as fixed factor.

There was a significant interaction between cognitive self-instruction strategies, age at diagnosis [F(9,29) = 2.423; p = .048] and gender [F(9,29) = 2.578; p = .037].

Another significant interaction was found between phase of therapy (2 main groups - active and maintenance) and problem solving strategies [F(10,19) = 3.537; p < .001].

A linear regression model showed that Cognitive self-instructions strategies were associated significantly to Socio-economic status of the parents ($\beta = -.390$, $t = -2.243$, $p = .033$). See Table 15 and Figure 1.

Furthermore, Socio-economic status of the parents was also associated to the quality of parent's advice provided when child had to face pain ($\beta = .382$, $t = 2.190$, $p = .037$). See Table 16 and figure 1

Table 15. Linear Regression Analyses Predicting Cognitive self-instruction to deal with pain

	Cognitive self-instruction to deal with pain				
	b (T)	SE b	β	R^2	ΔR^2
Constant	8.238	1.208		.152	.122
S.E.S.	-1.224	.546	-.390*		

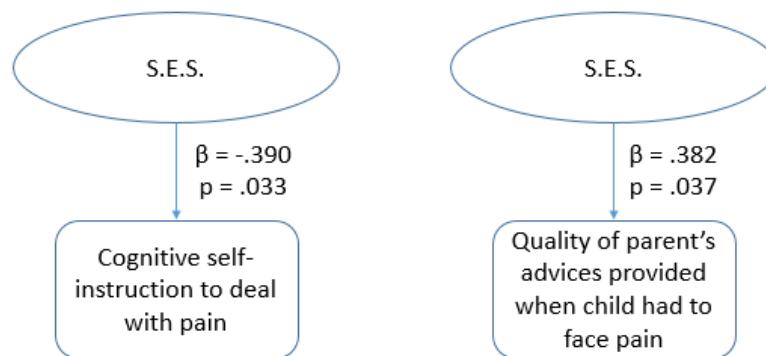
ΔR^2 = change in R^2 ; β = standardized regression weights; * $p < .05$

Table 16. Linear Regression Analyses Predicting Quality of parent's advices provided when child had pain

	Quality of parent's advices provided when child had to face pain				
	b (T)	SE b	β	R^2	ΔR^2
Constant	1.476	.509		.146	.116
S.E.S.	.503	.230	.382*		

ΔR^2 = change in R^2 ; β = standardized regression weights; * $p < .05$

Figure 1. Predictive factors are S.E.S and dependent variables are Cognitive self-instruction and quality of parent's advices provided



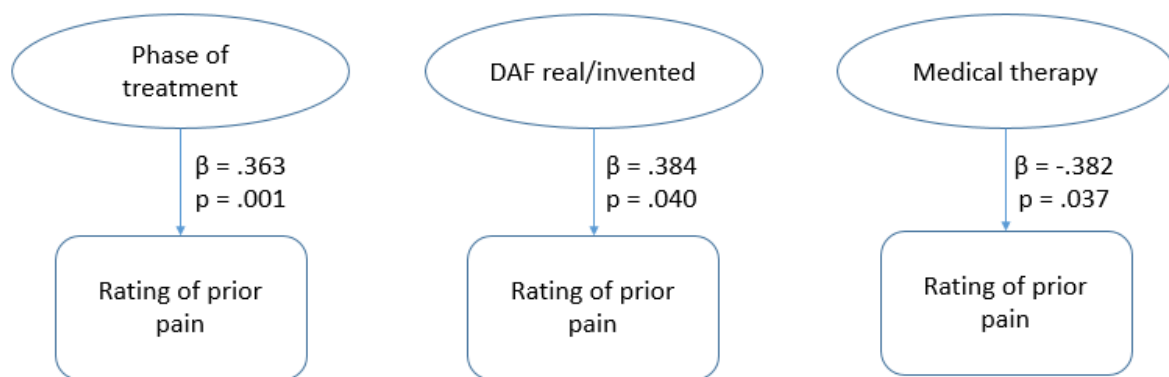
As to other possible variables, we have found that the age at diagnosis was correlated with the pain intensity at present time ($r = .536$; $p = .002$). Furthermore, children currently participating in medical treatment therapy, perceived different quality of parent's advice provided (supplied) when they had to face pain ($r = -.382$; $p = .037$), had a different ratings of prior pain (i.e. average of all pain in hospital) ($r = -.382$; $p = .037$) and different choices regarding identification of desire ($r = -.390$; $p = .033$). The treatment phase was correlated with rating of prior pain ($r = .363$; $p = .049$). A series of regression analysis measured the possible factors associated with prior pain (past). Results of the regression analysis provided partial confirmation for our hypothesis. For prediction of level of prior pain, beta coefficients for the three predictors of past pain (average of all pain in hospital) were phase of treatment, ($\beta = .363$, $t = 1.147$, $p = .001$); medical therapy, ($\beta = -.382$, $t = -2.189$, $p = .037$); and DAF real/invented, $\beta = .384$, $t = 2.162$, $p = .040$). See Table 17 and Figure 2.

Table 17. Linear Regression Analyses Predicting Rating of prior pain

	Rating of prior pain				
	b (T)	SE b	β	R^2	ΔR^2
Constant	4.490	1.147		.132	.101
Phase of treatment	.652	.317	.363*		
Constant	8.444	1.044		.146	.116
Medical therapy (on/off)	-2.730	.1.147	-.382*		
Constant	2.824	1.792		.148	.116
DAF real/invented	2.588	1.197	.384*		

ΔR^2 = change in R^2 ; β = standardized regression weights; * $p < .05$

Figure 2. Predictive factors are phase of treatment, medical therapy, DAF real/invented and dependent variables is rating of prior pain



What happens to the child's psychological well-being and his/her adaptation in relation to child's coping styles?

To answer this question by descriptive statistics, we have run preliminary correlations to see the associations between our variables.

To represent the processes of the child's inner knowingness of the state of his/her body, feelings and relationships, we have used the global approach with the integrative scoring system (in DAP and DAF), which aims to help identify and understand the complexities of this appraisal process and the associated feelings. With respect to self-representation drawings, DAP scores between formal coding and content coding were significantly and positively related ($r = 0.729$; $p < .001$).

In addition, the DAF scores, as an indicator of relationship of the family's perception, were correlated with the DAP formal ($r = .6027$; $p < .001$) and DAP content ($r = .527$; $p = .003$).

It is very important that the EI scores were correlated with all the self-representation drawings: DAP formal ($r = -.486$; $p = .006$), DAP content ($r = -.567$; $p = .001$). This means that the two methods of scoring work well. The DAF were correlated with the kind of behavioral attitude of the child during the protocol ($r = -.366$; $p = .047$).

We have found a correlation with gender and DAP formal ($r = -.554$; $p < .001$); DAP content ($r = .419$; $p = .021$), DAF ($r = -.594$; $p < .001$), Emotional Indicators ($r = .389$; $p = .033$). No significant correlations were found with child's age. See Table 18.

Table 18. Descriptive statistics for the child's indicators of emotional well-being

INDICATORS OF EMOTIONAL WELL-BEING	N	Mean	SD	Range
Formal-DAP	30	2.23	.74	1-4
Content-DAP	30	2.57	.85	1-4
Formal-DAF	30	2.17	.69	1-4
Koppitz' Emotional Indicators (EI)	30	2.03	1.67	0-6

An ANOVA assessed differences in self- and family-representations and coping strategies. There was a significant interaction between DAF formal and cognitive strategies [$F(9,29) = 2.425$, $p = .047$], as well between problem solving strategies and content-DAP [$F(11,29) = 2.472$, $p = .043$] and DAP formal [$F(11,29) = 3.078$, $p = .017$].

A series of hierarchical multiple regression analyses were conducted to examine if variables such gender, age, the various coping strategies from the four subscales of the PPCI questionnaire and also from the open-ended questions at the end of it could significantly predict the emotional indicators.

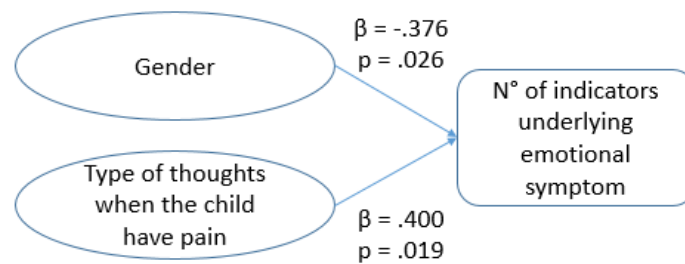
A model has found a significant association between a number of indicators underlying emotional symptom and the following independent variables: gender (step one), gender and type of thoughts when the child has pain (step two). The best model was the second one [$R = .558$, $R^2 = .260$, $F(1,29) = 6.095$; $p = .019$, R^2 change = .260], showing Gender ($\beta = .376$, $t = -2.355$, $p = .026$) and type of thoughts when the child has pain ($\beta = .400$, $t = -2.500$, $p = .019$) as the best factors associated with emotional indicators. See Table 19 and Figure 3.

Table 19. Hierarchical Regression Analyses Predicting N° of indicators underlying emotional symptom

	N° of indicators underlying emotional symptom				
	b (T)	SE b	β	R^2	ΔR^2
Step 1				.152	.121
Constant	1.250	.452			
Gender	.670	.584	.389*		
Step 2				.311	.260
Constant	-.320	.753			
Gender	1.262	.536	.376*		
Type of thoughts when the child have pain	.785	.314	.400*		

$\Delta R^2 =$ change in R^2 ; $\beta =$ standardized regression weights; * $p < .05$

Figure 3. Predictive factors are gender, type of thoughts and the and dependent variables is n° of emotional indicators



6 Discussion

Our primary aim was to test if the drawings of children with hematological cancer and healthy children showed significant differences.

Emotional well-being was conceptualized as part of the self-concept adaptive mode and can affect role functioning and interdependence. These concepts are related to psychological or internalized symptoms that are underlying a pain condition (Collins, 1984; Debiasi, Reynolds, & Buckner, 2012; Goldner & Scharf, 2011). It is known that Koppitz's Els in the drawing of a person, reveals children's underlying attitudes and characteristics, useful in estimating these constructs. Aiming to identify these problems, this study used 5 categories suggested by Koppitz (1968, 1984) and recently revised by Dağlıoğlu, Deniz, & Kan (2010).

In the impulsivity category, the total number of these indicators are manifested more meaningfully in children with cancer than in healthy children (63.3% vs 30%), as well the insecurity/inadequacy's signs (40% vs 3.3%) confirming the results from a previous study that used the same tools (Durualp & Altay, 2012). Conversely, the anger's indicators are different between ill children and healthy children, although not significantly. We have found in both groups low levels of anxiety and high levels of shyness/timidity.

Our results can be compared with the study of Cihangir and Kılıçarslan-Törüner (2007, as cited by Goldner & Scharf, 2012), who showed that children with cancer that underwent acute surgery, have experienced higher rates of emotional indicators such as impulsivity, mistrust, anxiety, shyness, and anger compared with healthy children.

Beside these individual indicators of internal distress (Goldner & Scharf, 2012), the family drawing can be considered a "narrative construction" of the relevant relationship, probably in its unconscious aspects, within the internal models' regulating emotions (De Coro, Tambelli, & Cundari, 2008). In fact, the relationships between feeling pain and reporting pain are highly context-dependent, are central among the relevant factors to the experience of pain, family and culture (Kortessluoma, 2009; Stevens, Hunsberger, & Browne, 1987). In line with these considerations, our data confirms that there are differences in the family drawings of children with cancer versus healthy children, both

with regard to specific markers and for the global evaluation. The differences especially lie in the ways of representing the paternal figure, which is generally represented with greater importance, size and priority compared to the other figures of the family, especially in the children with cancer. On the other hand, father is also considered clearly the happiest person in the healthy group, as opposed to that one of the ill children.

As regards the actual composition of the family, in both groups the family was 40% drawn without making changes and this could be considered as a defense mechanism, maybe the repression to impulsivity thoughts or insecurity feelings about themselves with parents. Repression might be linked to family reaction towards child's internalized problems, which may be present to a different extent, in both groups. However, we should be cautious in attributing psychological defenses mechanism before examining the case history of the child and the family.

This question, has found a partial response in the second objective of this study: the characterization of pain-coping profiles and the emotional well-being adjustment of the children with hematologic cancers. In fact, the quality of parent's advice provided when child had pain, was higher when even the socio-economic status was high. Further, to a lower socio-economic status, corresponded greater search of the cognitive self-instructions to deal with pain.

Several factors have influenced high scores of self-ratings of pain in general, (as an average of all pain when the child was in hospital): the phase of treatment, to be in active medical therapy and, precisely, the representation of the family as invented or featuring variations, like additions and / or omissions, instead of their real own family. Another important consideration to be made is that the total score of the indicators underlying emotional symptoms, depended on gender and on type of thoughts the child had when he felt pain: girls usually have fatalistic thoughts, which corresponds to greater emotional distress indicators. The seeking of social support is the most effective strategy used by these children, confirming previous results in a similar Italian population (Tremolada, 2008).

Although we would have wished to test the model conceptualized in this study, analyzing mediators and moderators, the small sample that we managed to collect has precluded us the possibility to conduct additional meaningful analyses. However, our choice of a convergent design, within a mixed analyses paradigm, through the combination of both quantitative and qualitative results, allowed us to gain a more complete understanding than that of each type of data alone (Zheng, 2015).

Our study features certain limitations that must be taken into account when both interpreting our findings and considering future research on this topic.

First, although the sample size was small and was a convenience sampling, we have followed a power analysis. An a priori sample size calculation was conducted to determine the number of patients to recruit, by using GPower 3.1.9.2 (Faul, Erdfelder, Lang, & Buchner, 2007). The calculation determined that a sample size of 30 was required to detect a medium effect size ($d = .50$) with an analysis of

variance ($\alpha = .05$, power = .60). However, despite the sample that we have recruited was sufficient for obtaining a good effect size, it was not enough to test our conceptual model.

Second, as the research literature examining pain in school-age children has involved an examination of parents in some capacity, (i.e. the role of parental behavior in child pain (Chambers, Craig, & Bennett, 2002; Goodman & McGrath, 2003; Moon, Chambers, & McGrath, 2011, as cited by Boerner, 2016) we have measured only dyadic interaction during episodes of pain (parent acceptance of child's emotion during painful episodes and quality of parent's advices supplied to the child), but only as an average over the whole observational period in hospital and not at a specific time. This may have caused biased interpretations intended as a tendency to interpret some situations in a too positive or too negative fashion. Furthermore, we studied only the child's own coping profiles, and not parents'. It would have been useful to understand their behaviors toward the ill child or their psychological reaction to caring for their own child, given the effect that parent functioning has on child adjustment, especially to chronic pain (Palermo & Eccleston, 2009), but also in the relations between family functioning and child acute pain, including pain ratings, coping, and parent-child (Birnie, Chambers, Chorney, Fernandez, & McGrath, 2016).

In fact, the impact of family and cultural beliefs on how children learn to react to pain in general, has been debated and there are more evidence in primary school-age children (Azize, Endacott, Cattani & Humphreys, 2014; Fortier, Del Rosario, Rosenbaum & Kain, 2010).

Despite these statistical limitations and bias risk, this is an ambitious study because it tried to complement different theoretical constructions. In conclusion, the most important aspect, from a clinical standpoint, is that examining the emotional distress and the family representations of children using DAP and DAF can be useful, both with the global system that give us an overall impression through the signs convergence and the analytic approach that offers relevant indicators for identifying specific fragility areas that children find hard to express through verbal communication (Pace, Zavattini & Tambelli, 2015).

However, although it may give an initial picture of the ill child's psychological and physical state and about his/her pain (Stefanatou & Bowler, 1997), it is believed that such an assessment should serve as a guide, rather than as a method of diagnosis and could thus be used as a pre-screening tool (Durualp & Altay, 2012). As Failo (2015) remarked, the most effective method of using drawings as a projective technique is via the use of multiple drawings, at different time points of the evaluation but also of the treatment process, to try and find an internal coherence of the child's self-perceptions and the expression of his/her emotional and contextual experiences of pain. The combination of these information with the clinically meaningful profiles of pain coping strategies appeared very helpful, also during follow-up for better pain management in several respects and in considering the child competent in expressing his/her needs.

CHAPTER 5

ACCEPTABILITY AND SATISFACTION OF A SPECIFIC PROGRAM FOR PARENT OF CHILDREN SUFFERING FROM CHRONIC PAIN

“...You can’t just go to something for one day or a week.... It really has to kind of be a way of life and what you change and what you do after...Well, he’s, he’s a trooper. He’s a good kiddo...”

~ a mum

The aim of this chapter is to evaluate acceptability and satisfaction by using quantitative and qualitative method about a specific problem-solving skills training (PSST) for parents of children receiving intensive pain rehabilitation. The program was implemented in the Seattle Children’s Hospital (U.S.) and my contribution to this study was limited to help in the analysis planning with the Grounded Theory method within the mixed method approach.

This chapter is completely based on the paper: [Law, E. F., Fales J. L., Beals-Erickson, S. E, Failo, A., Logan, D., Randall, E., Weiss, K., Durkin, L., M., & Palermo, T. M. (2016). A Single-Arm Feasibility Trial of Problem-Solving Skills Training for Parents of Children with Idiopathic Chronic Pain Conditions Receiving Intensive Pain Rehabilitation. *Journal of Pediatric Psychology, 1-12*] but it features only a portion of the data obtained from one of the three pediatric pain rehabilitation programs in the United States that have been presented in the entire original article.

1 Background, rationale and purpose of the study

An important consideration to do in relation to the management of children with chronic pain is the bidirectional influences of children’s pain experiences along with parental and family factors (Palermo, Valrie, & Karlson, 2014).

Parenting a child with chronic pain is distressing, indeed models put forth to understand the development and maintenance of pediatric pain problems hypothesize that parents play an integral role in their child's pain experience (Asmundson et al., 2012). Further, psychological distress serves

to fuel the vicious cycle of increased pain, fear, avoidance, and disability (Asmundson et al., 2012). As a consequence, parents, should be critical intervention agents in the treatment of their children's pain problems, given their role of care (Palermo & Eccleston, 2009).

There is growing literature suggesting that increased parent distress, parent protective behavior, and poorer family functioning are associated with poorer emotional and functional outcomes for youth with chronic pain (Palermo, Valrie, & Karlson, 2014). For this reason, any change in parent behaviors is considered as an integral treatment target in many cognitive-behavioral interventions for pediatric chronic pain (Levy et al., 2012; Palermo, Wilson, Peters, Lewandowski, & Somhegyi, 2009).

A series of parent training programs exists throughout United States and Canada with a wide variability in parent-focused interventions in intensive pain rehabilitation programs. Many of these programs are widely used by child pain services to improve the parenting practices of families for child's pain, focusing on teaching parent behavior management strategies to support their child's functional gains (Hechler et al., 2014).

There are no similar programs in Italy, so the only type of intervention offered systematically by health services is pharmacological. Initiatives undertaken by individual wards are sporadic and not guaranteed by guidelines. The only alternative for parents is to turn to private services without having the interventions safety shared with national health policies.

By analyzing the components of evaluated parent training programs, researchers gained valuable information that could be applied to similar programs. For example, in a Cochrane review on this topic, problem-solving skills training (PSST) delivered to parents of children with chronic conditions was found to be effective in reducing the distress (improving parental mental health and behavior) associated with parenting a child with a chronic illness (Eccleston, Palermo, Fisher, & Law, 2012, as cited by Palermo, Law, Essner, Jessen-Fiddick, & Eccleston, 2014). By contrast, there was no evidence for the effectiveness of other cognitive and behavioral therapies in improving parental mental health or behavioral outcomes (Palermo et al., 2014).

Findings demonstrated significant improvements in problem-solving skills and a significant reduction in anxiety and depressive symptoms among mothers who received an eight-session PSST intervention when compared to mothers who received ordinary psychosocial care (Sahler et al., 2005, as cited by Palermo et al., 2014).

PSST is based on the social problem-solving model proposed by D'Zurilla, & Nezu (1999) and uses modeling, behavioral rehearsal, and performance feedback to teach a step-wise approach to solving problems.

A research team (Palermo et al., 2016) has adapted PSST for parents of children with chronic pain receiving outpatient pain treatment, and evaluated the feasibility and efficacy of the intervention in a pilot randomized controlled trial. The program's protocol was a specific adaptation for parents of

children with chronic pain receiving care in an outpatient pain clinic based on a framework for problem-solving called “Bright IDEAS” (Sahler et al., 2002). In a nutshell, the thinking behind the conceptual framework is the optimism about the problem to be solved, the steps to be followed are Identification of the problem, Define the options, Evaluate the options and chose the best one, Act out the solution and See if the solution worked well, and if necessary, try again.

Based on these premises, therefore, I will mention only a portion of the entire study and the paper which I am a co-author of and which this chapter is based on.

The focused aim pursued, was to evaluate treatment acceptability and satisfaction using first a qualitative method, and then a quantitative assessment method.

2 Methods

2.1 Participants

Participants were recruited from the pediatric pain rehabilitation program in the Seattle Children's Hospital (USA). Inclusion criteria were: parent of a child with idiopathic chronic pain between the age of 10-17 (assuming from previous research that chronic pain is the most prevalent type of pain in childhood - King et al., 2011). Exclusion criteria were: 1) parent did not read or speak English, and 2) parent had active suicidal ideation or psychosis (evaluated separately). The Institutional Review Board (IRB) approved this study.

2.2 Procedures

The entire study is briefly described below. In this chapter I focused only on the interviews part.

Before starting the program, potential participants were identified by providers in the pediatric pain rehabilitation program. Informed consent was obtained from all participants prior to initiating study procedures.

After completion of the pre-treatment assessment, parents received up to six individual sessions of PSST over two to four weeks from a postdoctoral psychology fellow who had experience regarding cognitive-behavioral therapy for pediatric pain management. The program protocol was a specific adaptation for parents of children with chronic pain receiving care in an outpatient pain clinic based on a framework for problem-solving called “Bright IDEAS” (Sahler et al., 2002).

Children did not participate in PSST sessions.

After completing the final assessment, participants were invited to complete a semi-structured, qualitative interview by telephone, which is the part that is discussed in this chapter.

Study staff not involved in treatment delivery conducted, transcribed, and coded the qualitative interviews.

2.3 Measures of treatment acceptability and satisfaction

To assess treatment acceptability and satisfaction with PSST, parents completed a modified version of the Treatment Evaluation Inventory-Short Form (TEI-SF) (Kelley, Heffer, Gresham, & Elliott, 1989), a 9-item scale with scores rating from 1 (strongly disagree) to 5 (strongly agree). Total scores above 27 indicate that treatment has achieved moderate acceptability (Kelley et al., 1989).

Select items were adjusted to be specific to a pediatric pain population (e.g., “I find this treatment to be an acceptable way of dealing with children’s pain”). The TEI-SF has demonstrated adequate reliability and validity (Kelley et al., 1989) across a wide range of treatment studies, including in youth with chronic pain (Law, Fisher, Fales, Noel, & Eccleston, 2014; Palermo et al., 2009; Palermo et al., 2016). Cronbach’s alpha in the present study ranged from .82-.94.

To further investigate treatment satisfaction with PSST and get also constructive feedback to improve the program a Semi-structured telephone interview was administered to one of the parents (mother or father)

The interviews were conducted by trained study staff and included a standard set of open-ended questions and probes to guide the conversation and encourage parents to share their experience with the problem-solving therapy intervention. Interviews were audio-taped, transcribed, and coded by trained study staff who did not participate in treatment delivery and who is expert in qualitative analysis.

See Table 1 for Demographic Characteristics and Table 2 for the Track interview details PSST.

Table 1. Parent and Child Demographic Characteristics

Parent Demographic Characteristics	N = 9
Gender (% female)	77.8%
Age (M_{years} , SD)	47.22 (5.74)
Race (% Caucasian)	88.9%
Marital Status (% married)	88.9%
Education	
Vocational School/Some College	22.2%
College	33.3%
Graduate/Professional School	44.4%
Household Annual Income	
\$10,000-29,999	11.1%
\$50,000-69,999	11.1%
\$70,000-100,000	44.4%
>\$100,000	33.3%

Employment Status	
Full time	55.6%
Part time	11.1%
Not working	33.3%
Child Demographic Characteristics	
	N = 9
Gender (% female)	55.6%
Age (M_{years} , SD)	14.82 (2.10)
Race (% Caucasian)	66.7%
Primary Pain Location	
Headache	11.1%
Stomach	11.1%
Musculoskeletal	55.6%
Missing	22.2%
Pain Frequency	
1 time per week	11.1%
Daily	77.8%
Missing	11.1%

Table 2. Follow-up Interview Questions

1. What did you like about the Problem Solving Skills Training program?
2. What did you not like about the program?
3. Since finishing the program, have you used any of the skills that you learned? If yes, what have you tried?
4. Since finishing the program, have you send any of the materials in your treatment manual?
5. Would you be interested in receiving training in problem solving skills online or via Smart Phone? What do you think about having the worksheet available via Smart Phone in addition to or instead of the manual?
6. Have you taught these skills to your family? How did you do that?
7. What did your spouse think about the program?
8. Did you come downtown for session? How easy or difficult was that for you?
9. Did you complete session by phone? What did you think about completing sessions by phone vs in person?
10. How can we provide better support to parents and families who are going through the Pain Rehabilitation Program?

2.4 Data analysis plan

Concurrent mixed method data collection strategies have been employed to validate one form of data with the other form, to better understand the inherent complexity resulting from different type of data: the quantitative ones, from 9-items- scale TEI-SF and the depth of the data obtained from the interviews.

Quantitative analyses were done by using IBM SPSS v.21 (IBM Corp., 2012), and qualitative data analysis was conducted with NVivo v.10 (QSR International, 2012).

Descriptive statistics were used to summarize demographic characteristics of the sample as well as quantitative ratings of treatment acceptability and satisfaction. Qualitative interviews were coded using the constructivist Grounded Theory approach (Charmaz, 2006). The analytical process consistent with this methodology consisted in a transformation into text and a verbatim transcription of the interviews, followed by a focused coding to inductively identified themes (without a pre-defined conceptual framework or theory) and concluded with a theoretical coding in order to find relationships with the sub-categories and to be capable of bring out the core categories.

To implement a proper qualitative data analysis, we followed a series of fundamental steps (Wu, Thompsom, Aroian, McQuaid, & Deatrck, 2016):

- Kept a track about concepts emerging from the data and how data were coded to documented decisions and definitions of codes during the coding process (field notes and theoretical memos)
- Codes and themes were compared across interviews participants, also to understand how they were developed to identify similarities and differences (iterative coding process)
- Weekly meetings within the research team to discuss data and its interpretation in detail as the codes were developed (shared analysis of categories)
- Triangulating and validating findings, by comparing thoroughly the qualitative data with our quantitative ratings of treatment acceptability and satisfaction (diminish bias in the data).

Consistent with the constructivist approach of the Grounded Theory method (Charmaz, 2006) data analysis began with the research team reading all transcripts to become familiarized with relevant data. All data were transcribed and analyzed by two researchers.

Each protocol was initially coded line-by-line and divided into meaning segments for analyses. A meaning segment represents a separate idea, thought, theme, issue, observation, or process identified by the interview participants (Angus & Rennie, 1989; Charmaz, 2006).

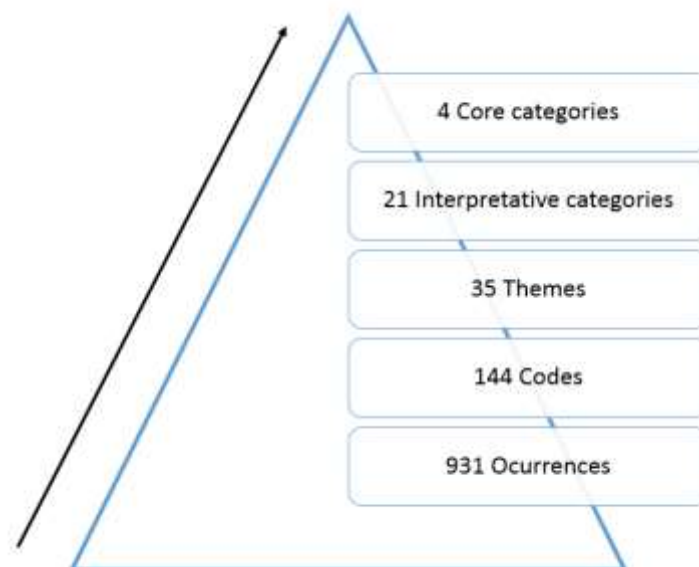
Two primary coders then created the initial codes by organizing segments of text into meaningful groups as expected by focused coding procedure using NVivo program: 931 occurrences and 144 codes emerged from this initial coding stage (carried out by two independent coders).

The primary coders compared their codes application and disagreements were resolved in weekly coding meetings with the research team.

Initial coding continued in an iterative fashion until the research team agreed that the coding scheme accurately captured the data. The segments of text were sorted into thematic clusters which united (grouped) the properties into descriptive categories. Each identified segment of text was placed into several appropriate or related subcategories, thus preserving as much variation in the data as possible, and helping to identify linkages between subcategories. In this focused coding 35 themes were identified inductively, defining the 21 interpretative categories. In the theoretical coding, these categories were better described in their properties and relationships in order to find 4 core categories

By using open coding, themes included the data were identified and all of the coded segments of text were reviewed to ensure that the data comprising the themes was coherent. This iterative process was needed for categorizing and describing the themes through systematic consideration by each participant to find similar concepts and continued until the themes accurately represented the data. This coding was followed by the process of axial coding whereby already identified codes (interpretative or sub-categories) were related to each other. The data were then reviewed again and coding refined until the research team agreed that further refinements would no longer lead to additional understanding of the data, then until theoretical saturation has been achieved with the core categories identified (see the focused coding process in Figure 1).

Figure 1. Focused coding process



3 Results

3.1 Sample description

In total, 9 parents participated in the study, including 7 mothers and 2 fathers ($M = 45.22$, $SD = 5.7$), primarily of Caucasian race, and middle-high class as indicated by annual household income between \$70,000 and \$100,000 (44.2%). Most of the parents had completed college or had higher education. Their children ranged in age from 10 to 17 years ($M = 14.82$, $SD = 2.10$). Pain conditions of the children included musculoskeletal pain (55.6%), headache (11.1%), and abdominal pain (11.1%). Overall, 84.6% reported experiencing pain daily. Demographic characteristics of the sample are presented in Table 1.

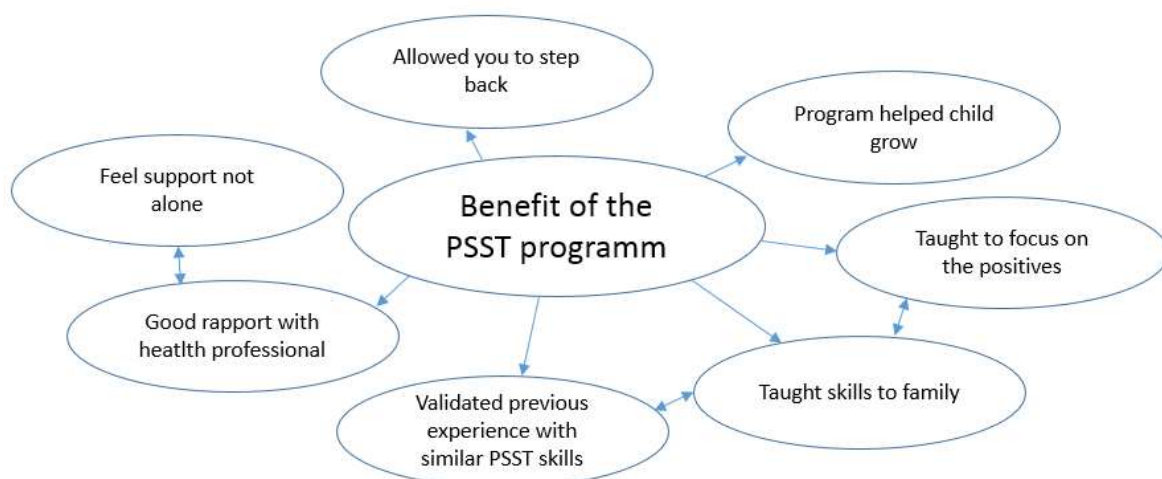
3.2 Treatment acceptability and satisfaction

Quantitative ratings from the 9-item TEI-Short Form (TEI-SF) with a 5-point scale, indicated that parents found the intervention to be highly acceptable and satisfactory ($M = 32.60/45$). The qualitative data collected was generated from a total of the nine parent interviews. The categories resulting from the coding, which are the core of inductive analysis, describe participants' satisfaction with PSST (both positive and negative), their experience using PSST skills, and their ideas to improve the intervention.

There was a total of 931 meaning segments, 144 codes, 35 themes and 21 subcategories (interpretative categories) as a whole. Four main categories emerged from the analysis, which led to the construction of an interpretative feasibility model to deliver PSST to parents during intensive pediatric pain rehabilitation (see Figure 2 for details of the interpretative categories).

The model includes benefits and limitations, which are the background of each step of analysis. However, what emerged was the treatment delivery mode that grounded the use of PSST skills post-treatment in parents (Figure 3).

Figure 2. Interpretative categories behind the core categories



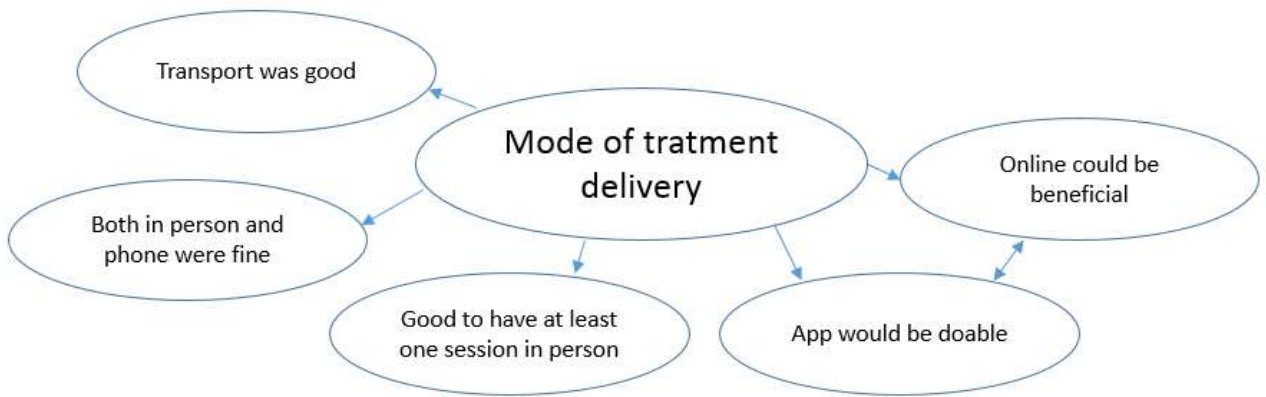
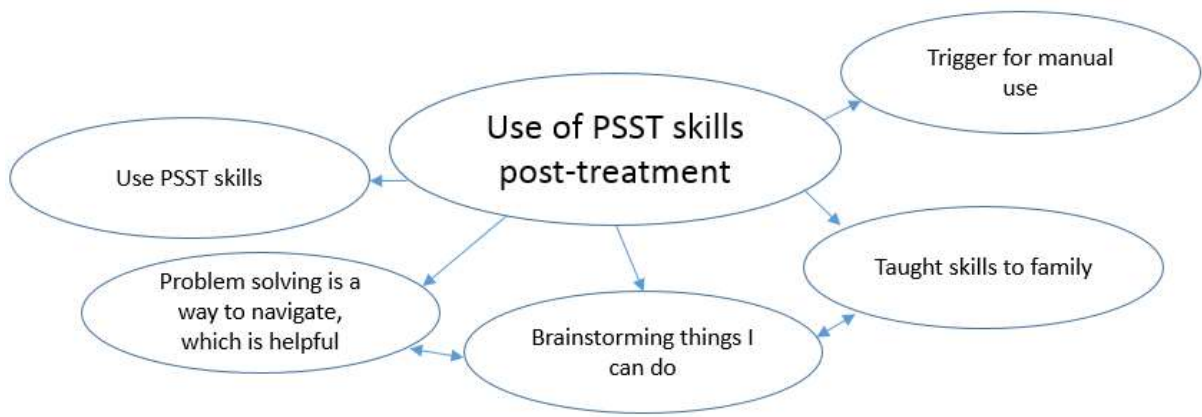
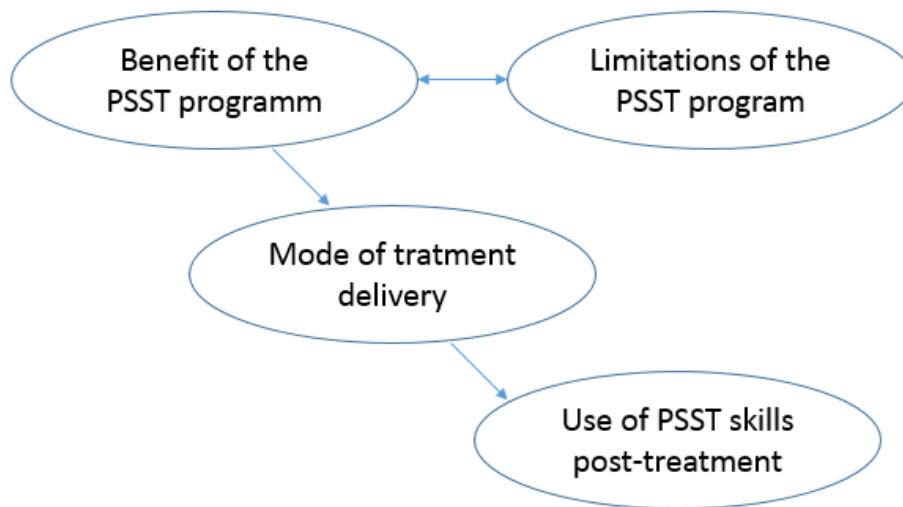


Figure 3. Interpretative model of to deliver PSST to parents



Qualitative interviews were completed by nine parents. Four primary themes, as a “core categories” were identified from the interviews: benefits of the PSST program, limitations of the PSST program, use of PSST skills post-treatment, and mode of treatment delivery.

Benefits of the PSST program

Many parents indicated that the PSST program validated their personal experiences and was helpful in their everyday lives. In particular, parents appreciated that the PSST program accounted for the unique experiences of parents participating in intensive pediatric pain rehabilitation.

“I felt kind of guilty about having those thoughts...‘This is exhausting, this is horrible, why is my child like this?’ Reading notes of other parents who felt the same way...was really reassuring...because you know, we’re really isolated...out in our own world. So that was really comforting to know I wasn’t alone in feeling that way, and it was okay to feel that way”.

“I really liked the skills that I learned. I mean, it helped me not only in solving her problems or helping her, but...in many areas of my life in general it helped me”.

Several parents had previous experience using problem-solving skills in their work settings, and reported that the PSST intervention helped them to apply these tools in a new area of their lives.

“It validated some of the military training I had, which was a side benefit for me, because then I could say, ‘Look, it works in real life!’ to my guys, but it also allowed me to take a step back and look at [child’s] situation and make sure that I was approaching it to be helpful”.

Limitations of the PSST program

Parents shared their perspectives on features of the PSST intervention that needed improvement. For example, one parent raised a concern that the PSST materials were not specific enough to the chronic pain population.

“Not all of the phrasing dealt specifically with chronic illness...occasionally...I kind of went, ‘This doesn’t quite fit’. And obviously there’s some similar things, but I think in some ways there’s major differences because a lot of us...that are dealing with chronic pain,...the doctors keep trying all these things and we don’t really know what causes it, why he’s even got it, and so...there’s a lot more uncertainty”.

Other parents pointed out that the problem-solving skills were rudimentary, and that they would have preferred more advanced training.

“It was just so basic. I’ve been working on problem-solving for, you know, how many years? Where we’re at in our relationship with [child]...the problem-solving wasn’t an issue so much”.

Parents also indicated that they wished there would have been additional support beyond the 4-6 treatment sessions to help them apply the problem-solving skills in the long-term.

“I couldn’t call and say ‘Help!’ [laughing]. When you get to a point, it’s like ‘Okay, I’ve exhausted everything you’ve taught me...and we’re still at a stalemate. Do you have any ideas or suggestions?’ I think that would be key”.

Use of PSST-skills post-treatment

In order to assess acceptability and generalization of PSST skills, parents were asked about their use of PSST skills since completing the intervention. Many families indicated that they continued using the PSST skills in their daily lives.

“We’ve used [the problem-solving skills], as a family, and myself in particular. I’ve been trying to use the books for coming up with solutions to something when somebody...gets pushy with me. ‘Let’s see if we can find another way...because this isn’t working’ ”.

Several parents taught the problem-solving skills to other family members.

“I took [child] and I told her,...‘This is for [solving problems]’ and we went through the steps, and I actually showed her the steps in the book, and then [for husband], I actually had him read it and then I explained, ‘This is what the study [is about]...and it really works!’ He actually applied a little bit of it, too!”.

While most families indicated continued use of skills, some responded that they have not applied the PSST skills.

“I haven’t [used the problem-solving skills since the study]. I thought about, you know, actually [using] some of the visuals in it. I’m kind of thinking sometimes when my son’s whining, ‘Oh, I need to...’ It was helpful at the time, I guess”.

Mode of treatment delivery

Parents appreciated the flexibility to participate in PSST sessions either in-person or via telephone, highlighting the importance of individualizing the PSST protocol to particular families’ needs.

“We did, I think, one of the [sessions] when we were still in the area...and then we did the rest of

them over the phone I believe. It was nice to have that flexibility and not have to come in...to do it every time".

We also assessed parents' interest in a future version of the PSST intervention that would be delivered on the Internet or via mobile application. Some parents expressed a preference for a web-based intervention that included human support.

"I don't mind doing things, you know, online or on my phone or whatever, but...if I can actually have a human being to connect with or something, you know, along the way, I feel like...I get a little bit more out of it".

Others indicated that they would appreciate the accessibility of an entirely web-based intervention.

"That would be cool because then...I could do it on my phone or at work. It would be handy no matter where I was".

4 Discussion

We chose to employ a concurrent mixed methods research design to validate one form of data with the other form (Creswell, Plano Clark, Gutmann, & Hanson, 2003), because this integration provides a better understanding of acceptability and satisfaction of problem-solving skills training (PSST) program for parents of children receiving intensive pain rehabilitation.

Overall, parents rated the program as having a pretty high acceptability and satisfaction. Furthermore, from the interviews it emerged that parents felt emotionally validated and found the problem-solving skills valuable and applicable to their daily lives. Indeed, of the nine parents interviewed, eight parents (88.9%) reported continued use of PSST skills after completing treatment and five of them (55.6%) reported continued use of PSST materials. An important result is that six parents (66.7%) have taught PSST skills to their family members as a personal decision.

A further strength underlined by parents, was the flexibility in scheduling treatment sessions, either in-person or by telephone, of the intervention protocol. Moreover, we have found that parents seem interested in receiving PSST via an Internet program or mobile application and this paves the way to new, even more flexible treatment modes.

As for the improvement, it seems that parents would like additional PSST advanced training in problem-solving skills as well as access to therapist's support following termination of the program.

About the limitations regarding the qualitative interviews, as Willing (2013) suggested, ideally, the process of data collection and data analysis in grounded theory continues until theoretical saturation has been achieved. However, theoretical saturation functions as a goal rather than a reality because even though we may reach saturation of our categories, categories modification or changes in perspective are always possible.

In conclusion, it is important to underline that intensive pain rehabilitation is increasingly common for youth with chronic pain in United States and the main outcome is that PSST have the potential to improve both parents' and child's skills. However, none of these programs exists in Italy, but the findings of this study may have important clinical implication also for other countries, considering the cultural differences as well. The next step could be quite close.

CHAPTER 6

FIRST STEPS TOWARDS DEVELOPING A MULTIDISCIPLINARY PEDIATRIC PAIN MANAGEMENT PROGRAM IN TRENTO

“The unreasonable failure to treat pain is viewed as an unethical breach of human rights”
~ Brennan, Carr & Cousins, 2007

This chapter addresses the need of building a shared educational project for healthcare professionals on pain management. We think of what we can do if we consider that pain relief is related to the application of the knowledge produced in an interdisciplinary clinical practice. In order to meet this purpose, a specific open-ended questionnaire was administered to doctors, nurses, midwives, nursery attendants and physiotherapists from two different care services: the neonatal unit and pediatrics unit of the hospital of Rovereto. Thematic content analysis method was used to derive the major themes and subthemes emerged from the perceptions of skills needs by 32 healthcare professionals.

1 Background about the educational needs in pain management

Several are the key factors that need to be explored, as a concise but clear introduction for the reasons that led us to promote this clinical survey.

Several countries have seen the development and expansion of pediatric pain management services within pediatric health care institutions that have integrated research-based guidelines into clinical practice (Dowden, McCarthy, & Chalkiadis, 2008). As to our country, Italy's national health service (Servizio Sanitario Nazionale - SSN), in 2001 had launched the "Pain-free hospitals" project, ("Ospedale senza dolore"), the first program in Italy for hospitals aimed at providing appropriate solutions for people with pain, with an agreement between the Ministry of Health and Regional Authorities. However, from the last available Report on the Health Status in Italy 2009-2010, "[...] [the program] did not achieve the expected results and there is no information available on the number of Pain-free Hospital Committees established" (Ministry of Health, 2011, p.83).

Today the situation is not too different, although, the health care system of the Provincia Autonoma di Trento (Italia), is specifically committed to the local Project “Pain Free Hospital” that promoted training for doctors and nurses, through special training events focused on the clinical recognition and assessment of pain and also public awareness campaigns.

Despite these good premises about efforts to identify the limits to effective pain management and attempts to modify the, through quality improvement programs, apparently the barriers to relieving pain remain unchanged, and still pain management in newborns and children is far from optimal (Comley & Banks, 2000; Cesado Gómez et al., 2015).

While recent studies have reported decreasing pain prevalence among hospitalized children (Groenewald et al., 2012; Birnie et al., 2014) many others reported that pain in infants and children, despite this data, has often been under-treated and often mismanaged (Dowden McCarthy & Chalkiadis, 2008; Srouji, Ratnapalan & Schneeweiss, 2010; Schechter, Berde, & Yaster, 2003) also because the pain-management medications are often used in the wrong way, which complicates the management of pain in the ambulatory setting (in outpatients) (Vadivelu, Kai, Kodumudi, & Berger 2016). Further, it should be remembered, that usually most of health care practitioners manage pain without considering any complementary approaches (and no “alternative” approaches that would be out of medical scope).

It is evident that it is not possible to prevent all pain, but when individual interventions are combined, as appropriate, a significant pain relief can be achieved (Taddio et al., 2015).

Given that children and adults react differently when it comes to pain, clinicians are often so focused on determining what are the causes of a child’s pain that they fail to perform appropriate pain management (Mathews, 2011). This is particularly apparent in the ambulatory setting, when time is so precious and the ability to make a distinction between several pain types that children may have is so crucial. Therefore, developing a systematic approach to pain that considers the individual and subjective differences is central and paramount in the healthcare (system) (Vadivelu et al., 2016).

Another important point to underline are the existing discrepancies between reported beliefs and knowledge among health professionals, influencing the treatment of children’s pain (Dowden, 2008; Twycross, Dowden, & Stinson, 2014; Young, 2005), not only in relation to treatment but also to prevention, as an essential human right, regardless of its consequences in the short or long term (Gómez et al., 2015).

The complexities surrounding pediatric pain management stresses that health professionals should do constant updates. Unfortunately, knowledge of current pain management practices by health care staff is lacking and consequently they do not apply latest research findings (Petovello, 2012).

This should be even truer for nurses and midwives, as they would have the opportunity to manage pain through multimodal means including non-pharmacological approaches to reduce fear and distress associated with medical procedures and hospitalization and minimization of the potential for long-term trauma (Dowden, 2008).

Therefore, an effective clinical inquiry on the field is required to determine healthcare professionals' educational needs so that they can directly help patients and provide support to parents so that they can manage more effectively the pain of their children, both in hospital and at home.

Starting from this standpoint, probably, we should create and promote an individual and collective accountability in the healthcare professionals, for pain management, as a crucial element of any strategy to improve the quality of this care, including its coordination and continuity.

2 Aims and purpose of our clinical survey

The purpose of this study was to identify the existing skills and the learning needs for doctors, nurses, midwifery and physiotherapists to plan a shared educational project for health professional on pain management, specially stressing out the integration with the psychological approaches

The hypothesis is that when common objectives, different skills and needs are considered, the construction of a collective project is really feasible.

The following research questions were developed to guide this purpose:

- What are the basic skills that doctors, nurses, midwifery and physiotherapists have regards pain management? (both clinical application of pharmacological and non-pharmacological skills)
- What are the barriers to use these skills, if any?
- What are the areas that need more improvements? (e.g. acute/procedural pain, chronic pain, disease-related pain)
- What are the most effective educational approaches to learning and what are the preferred methods for these health professionals? What is the best time for learning?

3 Methods

3.1 Study design, setting and sample

An institution-based, cross-sectional study was conducted in one of the two referral hospitals in Trentino through a prospective consultation of the clinical staff of two care services: the neonatology and the pediatrics units of the hospital of Rovereto.

Data were collected from a convenience sample including doctors, nurses, midwives, nursery attendants and physiotherapists who agreed to participate. Exclusion criteria was: other kind of professionals who were not directly involved in patients care.

3.2 Procedures

The approval for the study was obtained from the Head of the hospital and the Head of the two units. Eligible healthcare provider received written and oral information about the study upon recruitment, included their right to refuse participation without penalty. The head nurse distributed a self-administered specific open-ended questionnaire to the participants in the respective units.

Specially developed after a series of meetings between the Head nurse, the Head of Units and the clinical researcher, this tool contained some information about each health professional, including qualifications, years of experiences, service unit and 8 open questions aimed to provide a series of recommendations based on the qualitative findings from the responses (see table 1)

To encourage genuine answers and promote confidentiality, all participants were asked to write no other personal information, including name or date of birth, and to return the questionnaire in a box. Questionnaire anonymity guarantee had a crucial role in the success of this kind of survey. We have chosen this instead of the classical interview, because we deemed it more neutral with the person generally less influencing the answer and a better cost-benefit rapport.

3.3 Analysis process

An inductive approach was ensured through a regular verification (checking) of emerging themes with initial data and always within their context to preserve intentionality and meaning.

All data were collected prior to analysis. Answers were transcribed *verbatim* and analyzed thematically according the five steps suggested by Lagerløv, Rosvold, Holager, & Helseth (2016):

Step 1: Two members of the research team read the content very carefully and condensed all the questionnaire, staying close to the clinical staff descriptions of the broad topics of the questions within the emerging themes' framework.

Step 2: In this step the results were described for each participant within the broad topics emerged during step 1. An inductive approach was ensured through a constant verification (comparison) of emerging themes with initial data and always within their context to preserve intentionality and meaning.

Step 3: Entering step 2 results into NVivo v.10 (QRS International, 2012) software, we were able to perform a thematic analysis across the individuals, looking for similarities and differences in their descriptions. During this process, patterns emerged across the participants, suggesting that similarities within one topic (e.g. barriers in pain management) were followed by similarities in the other topics (e.g. how often and which types of non-pharmacological intervention are used).

Step 4: Participants with similar descriptions on all topics were then grouped to describe the skills and the needs of the Healthcare professionals in the two units

Step 5: Discussing findings and building a statement describing the characteristics and the improvement of each units.

4 Results

4.1 Sample description

The data collection was carried out between December 1, 2015 and February 1, 2016.

Data were processed with NVivo v.10 (QRS International, 2012) software program.

In this survey, a 32 persons sample from the clinical staff was recruited, with a 51% response rate of the entire staff of the pediatric unit and the neonatology unit of the local hospital of Rovereto. The questionnaires were filled and returned correctly. Demographic characteristics of the sample professionalism are presented in Table 1.

Working experience varied from less than 1 to 40 years, with an average of 12.25 (SD, 11.78) years. Almost 25% has 1 years or less of experience in pediatric or neonatal units (n = 8), 37.5% from 2 to 10 years (n = 12) and 37.5% more than 10 years (n = 12).

Females accounted for 96.8% of the participants (N = 31) and most of the time their activities were carried out by 21.8% in emergency room (n = 7), 18.7% in general ward (n = 6) and 15.6% in an ambulatory setting (n = 5), the rest of the staff ranged regularly its work setting.

Table 1. Demographic characteristics of professionalism of Study participants (n=32)

Staff by disciplines	Pediatrics Unit	Neonatal Unit	Total participants (% compared staffing with the Staff)
Nurses	8	7	15 (53)
Midwives	-	10	10 (77)
Nursery attendants	-	3	3 (50)
Doctors	2	0	2 (15)
Physiotherapists	2	-	2 (67)
Total	12	20	32 (51)

4.2 Emerging themes

This research aimed at investigating the actual clinical staff's standpoint and the thematic analysis, in cases like these, could be considered the best choice due to its flexibility and theoretical freedom to give an exploratory account of the data. Below the emerged themes are presented (see Table 2 for a summary). Five main themes were identified from the analysis. Supportive quotes are provided for each theme.

Table 2. Emerged areas from the answers of the open-ended questionnaire:

- | |
|---|
| <ol style="list-style-type: none">1. pain education and management areas (needs and enhancement requests)2. limits and barriers in the implementation of pain management (perceived from the clinical staff)3. preferred methods of receiving pain education (define which timetable and models)4. personal point of view on non-pharmacological techniques5. how often and how types of non-pharmacological intervention are used (in the two units) |
|---|

1. Pain education and management areas (needs and enhancement requests)

This theme captures participants' needs and enhancement requests for what they could achieve from an effective education.

- Training-application request for the non-pharmacological techniques (e.g. *"I would know a clear procedure with all techniques described in detail, with application examples"; "I would deepen non-pharmacological approaches"; "The knowledge of practical techniques is crucial", "I need more theoretical insights and insights on sensory saturation methods"; "In the application of psychological techniques"; "I would deepen my knowledge about the use non-pharmacological techniques in the prevention and treatment of pain"*).
- Need to expand their knowledge on the most appropriate approach to acute and procedure-related pain (e.g. *"The most lacking area of knowledge is on the procedural pain and even more the acute one"; "I need other information regarding the best practices about blood samples"; "I need to improve my knowledge to implement the usual procedures in the ambulatory setting such as heel lance, venipuncture and intramuscular injection"; "If I have to decide, I will definitely opt for the procedural one"*).
- Implementation of knowledge in all areas of pain and need for general education (e.g. *"Generally all"; "On all kind of pain and particularly in what regards premature infants"; "In all these areas knowledge should be implemented: the acute, chronic and total pain - palliative care"; "Across all areas, both in outpatient admissions and in hospitalization"; "In all areas of interest"; "The assessment and rational classification of pain in an operational scheme and treatment"*).

2. Limits and barriers in the implementation of pain management (perceived from the clinical staff)

This theme captures participants' fragmented opinions about the limits and main barriers, but there is a general agreement about historical preconceptions and organizational difficulties.

- Time limited and logistical difficulties (e.g. *"The concerns may arise in cases of urgency/emergency or, as for the emergency room, when there are many patients to be treated"; "The*

rush to perform the examination it is a deterrent"; "Same poor time management practices in the hospital environment"; "When performing a capillary puncture on an infant while the baby is at the breast which often causes an abrupt withdrawal of breastfeeding"; "Lack of time available, the main obstacle is related to the "haste"; "My work pace is always very high during the working hours, they don't help to create the right space to be able to best implement the procedures in pain management"; "Another obstacle, perhaps, is the excessive haste, and not being able to devote time during the procedure, also because often the same procedure is interrupted by phone calls, several requests etc..."; "Time-assistance and the workload that can adversely affect the monitoring "; "Lack of time, both for methods and the type of medication").

- *Underestimation of pain by health-care providers (e.g. "Pain is often underestimated", "often in the medical records, prescriptions for painkillers and also EMLA anesthetic are missing"; "I believe that there are common beliefs, such as the child has not real pain, that he has to get used or that pain will not remembered"; "There are barriers that arise primarily from the correct detection of pediatric pain").*
- *Someone affirmed, that, in general, there are no limits or deterrents in pain control (e.g. "In our clinical situation we overcome the deterrents and positively control the child's pain with different techniques"; "From the nursing staff there are no deterrents in order to control the pain of the child").*
- *There are limits due to the child him/herself (e.g. "The baby may be hungry, can be disturbed by a colic, he is dirty and he does not feel quiet for many other reasons"; "The child's personality, intended as the ability to concentrate and the self-control"; "Limitations due to child's age and the consequent inability to indicate and quantify pain"; "The child is unable to express himself adequately and you can't really understand").*
- *There are limits due to parents (e.g. "Sometimes parents are uncooperative and this can become an obstacle"; "The mother, we should let her know and participate in the maneuvers"; "Mother must be convinced of what we have done, quietly"; "Too much parental anxiety").*

3. Preferred methods of receiving pain education (define what timetable and models)

In this area a clear consensus about which kind of training or timetable should be implemented did not emerge.

- *Combining theory and practice (e.g. "Theoretical concepts and, if it is possible, practical exercises"; "Theoretical insights related to the cause and manifestation of pain, as well as no pharmacological techniques to help pain control"; "3-5 hours of theory and another 3-4 hours*

of practical exercises"; "Theoretical and practical concepts, both together and separate"; "Something that could be useful for both theory and practice").

- Clinical cases (e.g. *"Geared primarily on clinical cases and practical exercises", "Explore several topics in small groups within clinical cases")*).
- Duration up to 8 hours (e.g. *"Targeted training on weekdays, 8 hours"; "About 8 hours - 1 working day"; "3-5 hours of theory lessons and another 3-4 hours of practical exercises"; "I think: a training day spitted into 2 or 3 hours of theory lessons and 3 hours of practical exercises, could make up for my shortcomings related to the topic")*).
- Training in the afternoon or in the morning.

4. Personal point of view on non-pharmacological techniques

This theme captures participants' observations and reflections regarding the impact of the non-pharmacological approach. Participants also shared their experiences and beliefs.

- Non-pharmacological pain management therapies are useful and effective (e.g. *"I think that are very effective"; "Overall, they are very useful"; "In my experience they are effective. I believe that are effective techniques, because are simple strategies, useful to visibly reduce pain in newborns"; "I think that are part of my gold standard approach to the child"; "I think they are my duty, as a part of a gentle approach to the little patient"; "Certainly they are very effective for children of all ages, of course, modulating needs and different approaches" , "Very effective if they done properly, if motivated")*).
- Synergistic integration and intervention with pharmacological approach (e.g. *"Next to them, however, there are also the needs to use pharmacological techniques for a holistic and sensible approach to child well-being", "Through the synergistic action, the pain can be controlled well enough")*).
- Varied responses (e.g. *"We have and we need sufficient skills in pharmacological analgesia"; "EMLA is sufficient and can be used before performing screening at the heel"; "They are not always effective"; "They are only tricks"; "It is more important the use of observational instruments to assess pain than other stuff")*).

5. How often and how types of non-pharmacological intervention are used (in the two Units)

This theme captures participants' experiences on the non-pharmacological techniques. Several aspects of these techniques were noted to have positive impacts in patient care.

- Non-pharmacological treatments used in combination with drug therapies (e.g. *"I use these techniques sometimes also in combination with pharmacological ones"; "If possible, I guarantee the combination of these psychological techniques accompanied by the application*

of the prescribed medical therapy"; "...are effective as a completion of pharmacological treatment").

- They use only Non-pharmacological treatments (e.g. *"Usually, we do not use drugs with newborns, only sometimes with infants who were born suffering due to vacuum device use"; "In my daily work I don't use any medication to manage pain, only soothing techniques "; "Yes!, I use non-pharmacological treatments, avoiding pharmacological ones"*).
- Non-pharmacological treatments used in procedural operations (e.g. *"I believe that the sensory saturation is effective, for example, during blood sampling"; "Yes, for example, I use sensory saturation during sampling with micro-method"*).
- Non-pharmacological treatments used few times (e.g. *"They are techniques that I had the opportunity to practice in few cases"; "I used them rarely in my clinical practice and not rationally"*).

We believe that the themes presented above can define a possible path to follow in relation to the real needs and demands the members of the clinical staff of the two units face every day.

4.3 Differences between Health Professionals of the Neonatal unit and the Pediatric unit

There are no significant differences on limits and main barriers perceived between the two clinical staffs, as well as about the preferred methods of receiving pain education. Differences were found in the best known and most used psychological techniques: neonatology is more focused on breastfeeding, containment such as Kangaroo Parental Care, solutions with higher concentrations of sucrose/glucose, Sensorial Saturation (the "3Ts" stimuli: touch, talk, taste), structured parent involvement and finally, distraction techniques.

By contrast, the pediatric unit is focused primarily on Relaxation and sensory saturation, distraction and shifting attention, general parent involvement and other techniques (physical contact, containment, desensitization, magic glove, breathing, visualization) (see Table 3).

We have found also different need in priority areas of training: the neonatology's staff gives top priority to the needs to gain knowledge in the application of specific non-pharmacological techniques. Conversely, the pediatric staff prefers the improvement of the knowledge about all areas of pain (procedural, chronic, palliative care) without any specification on what techniques are to be used (see Table 4).

Table 3. Differences in the use of psychological techniques (ordered by frequency)

Neonatal unit	Pediatric unit
1. Breastfeeding	1. Relaxation and sensory saturation
2. Containment	2. Distraction and shifting attention
3. Sucrose/glucose solutions	3. General parent involvement
4. Sensorial Saturation	4. Several other techniques
5. Structured parent involvement	
6. Distraction techniques	

Table 4. Different needs in priority areas of training (ordered by frequency rate)

Neonatal unit	Pediatric unit
1. Application of specific non-pharmacological techniques	1. Knowledge about all areas of pain
2. Acute procedure-related pain	2. Application of the non-pharmacological techniques
3. Knowledge about all areas of pain	3. Acute, procedure-related pain
4. General education and training	4. How to combine pharmacological and non pharmacological approach

5 Knowledge translation interventions in the Neonatal unit and Pediatric unit

The results presented above, show that a good work has already been done in the pediatric and neonatology unit in the pain management area. However, without the guidance of a cohesive plan for current and future service demand, this work is done without being able to provide lasting effects. To achieve this goal not only additional resources are required, but also a transformation in the way in which health professionals understand and deal with pain. This process is called "Knowledge translation (KT)", a relatively new term that implies an interactive and engaged process between the research and systems of care and primarily pertains to the assessment, review, and utilization of scientific research (Jacobson, Butterill, & Goering, 2003). Indeed, the process of putting knowledge into action as a translation of research findings into the clinical practice is not direct and simple and continues to be a significant barrier to improved quality of care and patient outcomes (Graham et al., 2006).

A recent and focused systematic review by Gagnon, Hadjistavropoulos, Hampton, & Stinson (2016), on knowledge translation initiatives directed to health care providers in pediatric pain (98 article selected), highlighted that most of the initiatives were most frequently multidisciplinary (53%) and took place in hospitals or community medical centers (85%). Several areas of training were targeted

and nearly one-third were related to procedural pain (28%) followed by the ones related to disease illness/injury pain (18%). The remaining initiatives either combined both procedural and disease pain-related issues or were broad in nature (53%). Most of the initiatives used several and multicomponent approaches. Among the format used, in-service or workshop training was implemented in a large subset of studies (88%), followed by informal education (such as bedside training, discussions during chart reviews, feedback or reminders), but also with several strategies targeted at unit-wide changes to protocols on the unit or changes to chart formats, prescription forms and/or unit materials. A further element that should be given careful consideration seems to be related to the likelihood of patient outcomes that results from KT intervention in pediatric pain. All initiatives should implement a follow-up period (6 - to 12-month) to evaluate whether gains are sustained over time. These evidences for KT success in pediatric pain, along with the data generated from our clinical survey, will facilitate the planning of specific educational interventions addressed to health professionals.

The project title is "Non-pharmacological Pain Management Therapies for Infants and Children - Education program for healthcare providers" (16 hours). The curricular content is subdivided into 3 modules and is planned over three half- days. Each module consists of 4-6 contact hours and is coordinated and supervised by the Head of both Units. Teaching methodologies include lectures, problem-based discussions, video or audio examples and practicum. Pre-test was conducted (T0) and post-testing will be done (T1: after the program and T2: again following 6 months). At the moment we have started the first module (December 12th, 2016) and the other two will be held probably in February and in April, 2017.

The Pediatric Non-pharmacological Pain Education Program was offered to all Rovereto's hospital healthcare providers working with neonates/children. Staff were invited through the Head of both units to register to the ECM (Continuing Medical Education) system. A total of 14 inter-professional staff have started the module 1.

The title of the module 1 was "The role and theoretical basis of multidisciplinary non-pharmacological pain management and the Evidence-Based Clinical Practice" (4 hours). The content was decided through university-hospital partnerships fulfilling clinical and education needs, which comprised a multi-disciplinary team, including a clinical psychologist with expertise in pain research, a clinician with expertise in pediatric palliative care, the Head of the two units and the Head nurse (see Table 5). The modalities and implementation of the other two modules (2 and 3) are planned without detailed scheduling (see Table 6). Both modules will probably involve other healthcare professionals, as experts in addition to those that have participated in the first module. Also, participants could be more, since each health professional is free to attend to even only one module (despite the modules are considered as a connected path).

In fact, even though the complete program is not mandatory, the goal that guided us from the beginning, was to plan a flexible "path" able to integrate and enhance performance in clinical practice, research, and education to achieve a good operational and service level.

Table 5. Module 1: The role and theoretical basis of multidisciplinary non-pharmacological pain management and the Evidence-Based Clinical Practice

Module 1: program and outline done (interactive lecture, audio and video files) – 4 hours
<ul style="list-style-type: none"> • Dealing with pediatric pain in Italy - 38/2010 Law: state-of-the-art • Psychological influences on children’s acute pain and the importance of interventions to prevent chronic pain development • Appropriate use of pain assessment tools through proper protocol for pain management • Guidelines and recommendations based on an extensive review of evidence-based research literature primarily on management of acute and procedural pain (infants and children) • The approach and gradual non-pharmacological pain management techniques in children (video and audio examples)

Table 6. Module 2 & 3: planned – Integrating non-drug treatments in the standard clinical practice

Module 2: outline plan (testimonials, video and interactive education meetings) – 6 hours
<ul style="list-style-type: none"> • Testimonials & parents and children’s experiences • Video presentations to learn some tips about how to use and implement nonpharmacological techniques (different approaches for different ages & stages of child development) • Examples of innovative ways to use the techniques, such as links to previous experiences in the wards along with relevant case studies (as a references prototypes) • Presentation of the proposal of a partnership model between pain researchers and clinicians to better address pain management challenges (e.g. models for studying strategies for the prevention of chronic pain following acute)
Module 3: outline plan (small-group tutorials, problem based interactive sessions using case scenarios) – 6 hours
<ul style="list-style-type: none"> • Experiences in pain control in Neonatal Pathology Units of the general hospitals of Rovereto and Trento during the last two years • Planning and increase of multidisciplinary interventions for future implementations • Face to face activities (exercise simulations in neonatal and pediatric age) • Effects of different interventions on pain management (e.g. reduction of anxiety and distress, decrease fear, provide patients with a sense of control) • Integrate non-pharmacological treatment in practice

6 Constructive discussion

The purpose of this study was to investigate the clinical staff's point of view from neonatology and pediatric unit of a local Hospital.

The starting point that guided the aim and the methodology is the consideration that when common objectives and the different skills and needs are considered, the construction of an effective project is more feasible and accepted as part of integrated best practices.

Through the open-ended questionnaire and the qualitative analysis used to survey the opinion of 32 clinical professionals from a range of disciplines, several factors that can actually contribute to build effective pain management have emerged.

The results show that the most frequently perceived barriers to the adoption of an effective pain management care are insufficient time and organizational difficulties, but also the underestimation of pain has achieved general consensus. Participants reported that the knowledge of procedure-related pain management, is the one that requires more insight than others types of pain, because is the one that they face more frequently.

As for an approved training, the preferred methods for receiving helpful pain education t are the practical and concrete practices, combining theory with several clinical cases.

These results can be reached by a duration training up to 8 hours for each module: pharmacological and non-pharmacological interventions. Further, training should raise awareness among operators towards a real pain measurement and an integrated pain treatment: there are still deterrents within the units and limits that should be better understood and dammed.

As regards the use of psychological techniques, the main topic that has been investigated by this survey, it should be said that they are quite well known and considered among health professionals, in particular in the neonatal unit. However, as these techniques are used indiscriminately, it should be better define modality and utility specifications of each technique and / or a focused pool of techniques.

As the common medications used to treat or reduce the symptoms of Pain are well known, psychological treatments are not so well defined.

However, all in all, each healthcare professional uses this kind of approach, although not always consciously.

Indeed, as Lioffi (2006) reminds us, the Nonpharmacological Treatments, are defined as a verbal interaction between a health care professional and a child that leads to changes—from a less adaptive state to a more adaptive state—in the child's pain-related thoughts, feelings, and behaviors. In the context of wards, neonatal and pediatric pain management, includes the perspectives, interpretations and interactions of those involved.

Nurses and midwives, the major professional categories represented in this study, have conceptualized pain management as a part integrated with their interactions with children but also with parents.

Perhaps this feature inherent to the relationships, should be the one that will guide future training.

About strengths and limitations: as said, if on one hand the involvement of those working with children who suffer in some way could be considered as the strength of the study, on the other hand, clinicians were only two and this may have affected findings representation and generalization in the clinical units.

Obviously, these results are derived from a relatively small sample, but highlight many key components that health professionals look for in an educational module on pain management, that need a better planning or additional resources.

In accordance with the Head of the Units and the Head Nurse, it seems that this information was taken into consideration in the planning of a Non-pharmacological Pain Education Program that we are administering to Rovereto's hospital healthcare providers to better manage pain in infants and children.

We should remember that continuing the research for improvement measures, through listening those who regularly work with children, is also the best choice to promote healthcare professionals' motivation.

GENERAL DISCUSSION AND CLINICAL IMPLICATIONS

This research project, in its transversal complexity, offers important contributions to the field of pediatric pain management, particularly, the process of adaptation to illness and effective interventions. The various types of pain caused by their illness and treatments children and adolescents face, affect not only the direct aspects of pain, (e.g. the intensity), but also their psychological well-being and adaptation abilities.

This research offers insight into several coping and adjustment factors that influence the different trajectories of pain experiences in children with serious diseases.

In addition, the theoretical and clinical implications, the methodological contribution of this project have been discussed in-depth, highlighting that no single method can adequately provide adequate research results when issues involve the complexity of clinical insights. Therefore, in the presented clinical studies, I have used the triangulation strategy to seek convergence, inconsistency and contradiction by investigating data and methods in order to better answer to the search questions (Cox & Hassard, 2005).

Several theoretical models have been proposed in an attempt to better understand the complexity inherent in pediatric pain. Surely, specific models both for pediatric chronic pain and responses to acute pain allow a better explanation for the various underlying processes. However, the starting point of this thesis was to propose a general pain model by assuming that large part of the risk factors for acute and chronic pain are the same and underpinned by aspects not completely clear at conscious level.

To achieve this goal, two studies have been performed: the first with three cohorts of children with chronic disease and the second with a single and specific cohort.

In the first study, we found that the type of diagnosis (cystic fibrosis, rheumatic diseases, and the group with Hematological and Solid Tumor) and perceived family support, as an internal working model of the typical environment tied to psychosocial well-being, were two predictors for both overall pain-coping strategies and the use of the specific distraction coping strategies. With respect to drawing profiles on self-efficacy and self-esteem, children with cancer diagnosis displayed poorer self-representation. Taken together, these results highlighted the importance of building a sound perception of family support as a significant factor that may also influence self-esteem and self-

efficacy and thus better pain-coping strategies. Regarding pain coping trajectories across the course of chronic illness, it is important to understand how children cope with pain during the whole period of their disease. The coping strategies that are adaptive could be changed over time and depending on disease progression. However, the influence of time since diagnosis on coping was not confirmed in our sample.

In the second study, on a sample of children with hematologic malignancies, we have found that despite some children achieving good psychological adjustment, children with cancer have shown significantly higher scores for impulsivity behaviors and insecurity/inadequacy feelings and this, as more than half have drawn the family making changes, it could be considered as the repression defense mechanism, when compared to healthy controls. Furthermore, unlike the healthy children who indicated no difference between family members, they perceive the paternal figure as the one with greater importance attached, but also the last one happy, suggesting that father could be a guide as objective support to containment, but probably not for the emotional part. Examining the case history of the child and the family for the clinical sample, the data supported a model in which a higher socio-economic status may activated in children less cognitive self-instruction skill to deal with pain. Also a higher quality of parent's advice provided when child had pain, suggests less investment in the management of their pain, leaving this aspect to the family, as if it were a dependence, therefore with less emancipation. These findings suggest that ratings of prior pain reported by children are affected by the treatment phase, where the maintenance phase and the stop therapy predict higher pain scores, as a cumulative factor of the average pain when children have been in hospital. Surprisingly, receiving in a current medical therapy predict lower pain scores. Furthermore, higher rating of prior pain is also related with a representation of the real family of these children, instead that one invented. Moreover, our findings showed that emotional distress has a strong influence on the type of thoughts when the child has pain and depends on gender as well.

Taken together, all these results, confirmed our hypothesis that several family and treatment factors affect the perception of pain and how to cope with it. We can also point out the usefulness of the method of drawings in children, that being an intentional process involving projection and introjections by the individual, associated to various sensations, perceptions and emotions with their body, can provide a natural vehicle for the expression of one's needs and conflict about the illness and treatments (Angstrom-Brannstrom & Norberg, 2014; Machover, 1949; as cited by Kortessluoma, 2009).

The underlying theory I support is that many mechanisms described above, play a key role in mediating between conscious and unconscious activations of well-being and in the early identification of those kids at risk of developing a chronic pain, or at least, less skills in pain management. Each factor is an independent contributor, but I believe that a factors' combination (or

interaction), as an emotionally vulnerable child and modeling or reinforcement behaviors by a parent, might pose a risk of developing maladjustments due to the experience with pain.

From the experience gained in the evaluation of the acceptability and satisfaction of the specific program for parent of children suffering with idiopathic chronic pain that was implemented at Seattle Children's Hospital, it emerges the importance of parental cognitions and behaviors. In fact, the results combined from the interview and the scale, support the consideration that specific interventions targeted to parents, might be effective in improving outcomes in children with pain and other symptoms where environmental factors can play a key role in pain expression and ability to cope with symptoms, as well the ability to adaptations and adjustment (Langer, Romano, Mancl & Levy, 2014). Parents should be educated about their child's pain, which will help them to engage in adaptive and functional behaviors, by taking an active role to help their child/adolescent, who, in turn, will let parents feel that they have an important task to fulfill. Findings for this program in USA, thus suggest possible directions for the design of psychosocial interventions also in Italy, where, unfortunately, are currently missing.. It is worth k in mind that parents are a possible means for influencing child pain behavior, his/her adjustment processes and functioning.

Another remarkable achievement in this thesis, is that we have outlined attributes and desirable features of an interdisciplinary pain management that have considered to start a Non-pharmacological Pain Education Program at Rovereto's hospital healthcare providers. We felt that it was important to give voice to health care professionals for understanding their personal point of views about pain management, especially on non-pharmacological therapies. Only after hearing them, it would be easier and feasible to plan an effective and integrated pain education module (or modules) on Evidence Based Practices and individual needs. Although it is true that some gaps remain in health care professionals' knowledge about pain, it is also true that education alone is not enough, as they themselves have said. It should include a mixture of interactive and didactic teaching, developing strategies that help link education to practice, in addition to encouraging reflection, clinical simulations and the use of case studies and also - why not - we-based materials (Twycross & O'Conner-Vonn, 2013). Of course, translating best evidence on pain assessment and management into clinical practice remain a challenge but it seems more credible and feasible if we hold in mind the centrality of the child's care and his family, which are in turn mediated and moderated in their explicit components such as cognition and beliefs on pain on one hand, but also self-esteem, family support perception and the defenses on the other. To gain a new perspective from health professionals it is necessary to attempt to identify the commonalities about pain education through the individual inclinations combined with Evidence-based treatments. These concepts should be part of the educational curriculum of all health

professionals who care for children and used with several types of pain, along a continuum from acute to chronic pain (APS, 2012).

An examples of confluent education methods is suggested by Twycross and O'Conner-Von, (2013) including use of clinical experiences within group, discussion, role play, clinical simulation, case studies, reflection on what individuals have learnt from the sessions and how this learning will be applied in practice.

More specifically about the content, Taddio et al., (2010), in her review on evidence-based clinical practices guidelines to reduce pain during vaccination, suggests to health professionals the importance to encourage breastfeeding or administration of glucose solution (for infants) and the application of topical anesthetics and psychological interventions (for children of all ages). Furthermore, healthcare providers should discuss these additional options with parents and children (as appropriate) and to select the strategies best suited to individual children.

A further central topic to develop effective strategies in pain management is by increasing knowledge of pain during treatments, such those related to cancer and other chronic conditions, that might be used to develop approaches and interventions to prevent the development of chronic pain and provide examples of innovative care delivery models (Palermo, 2012).

Although many studies of specific risk factors may contribute to chronic pain, (e.g. demographic factors such as age and sex, factors related to the individual such as coping style, and familial or environmental factors), the mechanisms that lead acute pain to become chronic, as well as the contribute to chronic pain maintenance are not well understood (McKillop & Banez, 2016).

McKillop & Banez, (2016) proposed that potential variables are more complex and we should take into account: (1) developmental stages; (2) family and peer factors; (3) physiological components; and (4) will integrate the interplay of multiple risk factors.

Anyway, preventing and controlling pain are crucial points that should be the gold standard for any healthcare professional.

Preventive strategies for pain often utilize principles of primary and secondary prevention, including multimodal pre-emptive approaches that are important to prevent people from developing chronic pain, together with a constant monitoring the quality of pain management.

Primary prevention generally refers to the anticipation and avoidance of pain onset, as that from iatrogenic origin, but also refers to the early identification of acute pain and management strategies to ensure the pain does not become chronic (Pagè, Huguet, & Katz, 2013). Cancer is illustrative, because most pain is iatrogenic. Given that at time of diagnosis, almost 50% of children have cancer-related pain and intense pain is more common at the beginning of treatment, procedure- and treatment-related pain are, initially, the major problems (Ljungman, Gordh, Sorensen, & Kreuger,

2000). Thus, reducing distress and anxiety associated with the procedures and treatments, through psychological interventions, are the primary outcome measures (Birnie et al., 2014).

Secondary prevention, insists rather on preventing early chronic pain states from worsening or persisting (Pagè, Huguet, & Katz, 2013). Taking cancer as an exemplificative, it is well know that even if procedural pain gradually decrease, treatment-related pain is constant and dominating (Ljungman, Gordh, Sorensen, & Kreuger, 2000). Therefore, children undergoing cancer treatment are faced with long hospital stays and they need comfort when they are in pain, scared and tired (Angstrom-Brannstrom & Norberg, 2014). A literature review suggests that children with cancer experience more pain from treatments and painful procedures than from the disease itself and in cancer this has been linked to changes in quality of life, depression, and posttraumatic stress (Twycross et al., 2015). People in traumatic situations often oscillate between endurance and emotional suffering to endure means control of emotions to save energy (Morse, 2001, as cited by Griffiths, Schweitzer, & Yates, 2011). In children undergoing treatment for cancer, closeness to family members, friends, and trusting relationships with hospital staff, are important for enduring discomfort and finding comfort, having a positive impact on them (Griffiths et al., 2011). While having a child with cancer can strain relationships and patterns of coping and adjustment, it may also foster family connection, and there are many ways in which family functioning may be either a hindrance or an asset in the context of childhood cancer (Sposito et al., 2015).

For healthcare providers it is therefore important to be aware of their role and to facilitate parents' participation in care, which will help preventing unrecognized or underestimated of pain and the resulting harmful effects. Further, the use of general cognitive– behavioral strategies in this population has the benefit of targeting the umbrella of distress that many children experience around procedures and treatments that involves a mix of anxiety and pain further reducing the risk of develop chronic pain conditions.

To increase dissemination of pain prevention measures, it is important to involve in this process the competencies of pediatric clinical psychologists in the development and implementation of innovative knowledge translation initiatives for procedural pain management to help preventing challenging and negative experiences for many children and their families (Boerner, Gillespie, McLaughlin, Kuttner, & Chambers, 2014). In particular, it could be useful an early use of Cognitive and Behavioral Therapies (CBT techniques), despite they have not yet been effectively used to prevent or provide early intervention for chronic pain (Palemo, 2012).

Instead, psychological interventions are notably effective for reducing the severity and frequency of chronic pain in youth (Eccleston, Morley, & Williams, 2003). Indeed, CBT has been used to treat children and adolescents with chronic and recurrent pain problems.

Given the strongest evidence of the efficacy of psychological interventions in chronic pain reduction in children (e.g., relaxation strategies, parent-training, cognitive strategies), these should be routinely recommended by health providers to supply skills training both to children and their parents, all delivered within a psychoeducational frame(work) (Eccleston, Fisher, Law, Barlett, & Palermo, 2015). The treatment regimens for chronic pain comprise the three P approach. The three Ps include Physical Therapy approaches, Pharmacological approaches (Medications), and Psychological approaches (coping strategies) for managing pain. Another important approach that should be added is the family support, since parents are an essential part of treatment of children with chronic pain, and strategies to teach parents adaptive responses to their child's pain can enhance rehabilitation efforts (McGrath & Ruskin, 2007; Reid, Lander, Scott, & Dick, 2010).

Other areas that need to be developed in teaching pain management to healthcare professionals are: studies of clinical algorithms for pain treatment, plans and evaluations of novel treatment strategies and delivery methods such as computerized delivery of treatment interventions (APS, 2012).

Below I present two algorithms highlighted by the literature: the first aimed at procedural pain and the second for chronic pain.

Algorithm for pain management during procedures (adaptation from Young, 2005; Lioffi, 2002; Taddio et al., 2015).

First of all, keep in mind the need for an individualized preparation and parental presence in order to minimize pain, anxiety and distress. Remember that any medical procedure essentially consists of four phases: anticipatory, preparatory, procedural, recovery.

Prior to the procedure we should discuss the information concerning parents/caregivers and children/teens to reduce anxiety and pain. Allow parent to stay during the entire procedure.

During the procedure, we should minimize pain and anxiety by the most effective combination of pharmacological and psychological techniques. Do not perform these tasks mechanically.

Remember that at the end of the procedure monitoring is crucial and encourage the child to continue to use his/her coping skills and praise the child and the parents because their coping efforts have contributed to a successful procedure.

Below there is a track on which pain management strategies are effective for infants/children/teens.

- Provide information and prepare the parent and child, also during the procedure and be honest
- Give step-by-step information of what will occur during the procedure
- Give sensory information about what the child will see, hear and feel
- Use age-appropriate language and terminology
- Avoid high-anxiety words such as pain, hurt, cut, shot and address children's concerns

- Consider using books describing the procedure the child can read with the parent, useful could be also the use of favorite stuffed animals/robots or blankets
- Allow parents to stay with child
- Do not ask the parent to help restraining the child
- Instruct the parent on coping-promoting behaviors (e.g. distraction) and to avoid distress-promoting behaviors (e.g. reassurance)
- Maintain a quiet, calm environment
- Do not force the child to lie down if he/she does not want to and is not required to
- Consider giving the child a “job” (e.g. holding a gauze)
- Give the child choices to increase the perception of control (e.g. right arm or left)
- For venipunctures and intravenous cannulation in thumb-sucking children, avoid the arm of the preferred thumb
- For long procedures (e.g. burn dressing changes), allow the child “time outs” of a predetermined number and duration
- Allow the child to “count down” from 10 to 1 before a brief procedure
- Use a treatment room; keep the patient’s room/bed as a “safe place”
- Plan and draw all blood samples at once if possible
- If multiple injections are needed, the most painful should be the last
- Positioning: upright/holding/sitting up (depending on the age)
- Use topical anesthetics
- Breastfeeding and/or sweetening agent in infant are necessary
- Deep breathing and distraction. Imagery can be helpful as well
- Vibrating device with cold
- Manual tactile stimulation

An interesting algorithm for children with chronic pain is provided by an adaptation by Twycross, Dowden, & Stinson (2014, p.201) from of a work of Brown (2006) and APS (2012) and it consist of:

1. Evaluation of the child

- Comprehensive medical and pain history, including prognostic factors
- Assess pain location, onset, duration, quality, variability, exacerbating and alleviating factors
- Physical and neurological exam including appearance, posture, gait, growth parameter and vital signs
- Use appropriate diagnostic tests and remember that treatment should also be focused on pain-related disability including impact of pain on daily life such as sleep, school, eating,

social and physical activities, with the goal of child function and comfort enhancement as well as improvement of quality of life

2. Diagnose the primary and secondary causes
 - Current nociceptive and neuropathic components
 - Attenuating physical symptoms
 - Contributing psychological factors, peer and social issues, biological processes
3. Select appropriate therapies to improve overall functioning and quality of life
 - Pharmacological (analgesics, adjunct analgesics, topical anesthetics)
 - Physical (graded exercise program, regular daily activity, pacing)
 - Psychological (relaxation training, imagery, deep breathing, biofeedback, CBT, school reintegration, sleep assessment and intervention, teach parents adaptive responses to child's pain). It is important constant home practice.
4. Implement pain management plan
 - Provide pain diagnosis, feedback on cause and contributing factors and also a rationale for integrated treatment program. Use a continuum technique.
 - Develop mutually agreed treatment goals. Assignments and follow-up.
 - Measure child's pain and functional improvement regularly
 - Evaluate effectiveness of treatment plan, and revise as necessary

Transversely, to ensure most effective methods of treatment and interventions, parental involvement is an important aspect to be considered, through a joint assessment of both child and parent to decide whether it is appropriate to include the parent and his role (Rook & Gauntlett-Gilbert, 2016).

Overall, it can be suggested that when both organizational and team-related conditions are present and barriers have been overcome, developing an effective pain assessment and management is possible. Moreover, awareness of the potential advantage of medical perspectives, psychological over nursing perspectives could reduce the somewhat unilateral attention and contribute to an inter-professionally shared reflection.

A further step in the direction of implementation of pain management programs could be to adapt the inpatient model for use in the pediatric home care setting, especially for those who suffer from recurrent pain (Czopp, Kay, & Cheryan, 2015).

As a final consideration, the set of these findings provide a framework which can be helpful to identify children who are most at risk of developing maladjustments due to their experience with pain and to provide a focused support for their parents through the intervention of trained and motivated health care professionals.

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