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Is it worth it? Is an intensive rehabilitation program making a difference for youth with pain-related disability?

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Thèse présentée à la Faculté de médecine et des sciences de la santé
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SOMMAIRE

Évaluation d'un programme de réadaptation intensif pour les jeunes souffrant de douleur chronique en Alberta

Par
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Thèse présentée à la Faculté de médecine et des sciences de la santé en vue de l'obtention du diplôme de philosophiae doctorat (Ph.D.) en recherche en sciences cliniques, Faculté de médecine et des sciences de la santé, Université de Sherbrooke, Sherbrooke, Québec, Canada, J1H 5N4

Résumé

La douleur chronique pédiatrique est un enjeu important de santé. Les traitements interdisciplinaires intensifs (TII) et les traitements multimodaux (TMM) sont les deux principaux modèles de services reconnus. Malheureusement, les descriptions de ces interventions sont peu détaillées dans les écrits et les études comparatives sont rares. Cette recherche évaluative comprend deux objectifs principaux: 1) Analyser les fondements théoriques du programme TII, et 2) Analyser les effets du TII et les comparer à ceux d'un TMM. Pour analyser les fondements théoriques du programme TII, un groupe aviseur composé de 13 parties prenantes a été recruté et une analyse logique a été effectuée. Pour évaluer et comparer les résultats et les effets du TII et du TMM, six domaines de résultats ont été priorisés par le groupe aviseur. L'analyse des effets a été réalisée par une collecte de données pré-post auprès de jeunes participants au TII et ceux inscrits au TMM. Par la suite, des entretiens, facilités par des chronologies narratives, ont été menés auprès d'un sous-ensemble de jeunes participants et de leurs parents. Les résultats font ressortir le bien-fondé des TII. Les TII et les TMM ont tous deux des effets positifs sur les jeunes souffrant de douleurs chroniques, chaque traitement ayant des avantages et des désavantages. Les résultats permettent de mieux comprendre quels traitements fonctionnent, et pour qui. Cette étude a généré des connaissances essentielles pour soulager la souffrance et améliorer la qualité de vie des jeunes souffrant de douleur chronique et de leurs familles. Elle a aussi établi une nouvelle norme quant à l'engagement des parties prenantes dans le processus d'évaluation des interventions complexe en réadaptation.

Mots clés : Évaluation, traitement intensif interdisciplinaire de la douleur, adolescents atteints d'une douleur chronique, engagement des parties prenantes, analyse logique, analyse des effets, ligne du temps, méthode mixte

SUMMARY

Is it worth it: Is an interdisciplinary pain rehabilitation program making a difference for youth with pain related disability?

By
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A thesis presented to the Faculty of Medicine and Health Sciences in partial fulfillment of the requirements for the degree of Doctor of Philosophy in Clinical sciences, Faculty of Medicine and Health Sciences, Université de Sherbrooke, Sherbrooke, Québec, Canada, J1H 5N4

Summary

Pediatric chronic pain is an important health concern worldwide. Intensive interdisciplinary pain (IIPT) and multimodal treatment (MMT) are the two main service models. Unfortunately, descriptions of these published interventions lack the details necessary to reproduce them in different contexts, and comparison studies between the models are rare. This study had two main objectives: 1) analyse the theoretical foundation of the IIPT program, and 2) analyse the effects of a day-hospital donor funded IIPT compared to those of an outpatient MMT. To evaluate the IIPT theoretical foundation, a 13-member stakeholder advisory group was recruited, and a logic analysis process was used. To evaluate the effects of the IIPT, six outcome domains were prioritized by the same advisory group using a nominal group technique. An effect analysis was then conducted, using a pre-post study design, collecting data on youth participating in an IIPT and those enrolled in an MMT at the same facility. Interviews using a narrative timeline then followed with a subset of youth and parent participants. The results highlighted the sound theoretical foundation of the IIPT. Furthermore, positive effects were demonstrated in youth participating in both the IIPT and MMT, with each treatment revealing advantages and disadvantages. Merging findings and comparing the results provided an opportunity to gain a deeper understanding of what treatments work, and for whom. Some insight into why these treatment options work and suggestions for intervention improvements also emerged. Not only did this study generate the knowledge pivotal to alleviating the suffering and improving the lives of youth with chronic pain and their families in Alberta, in Canada, and beyond, it set a new standard of patient engagement for evaluation research of complex rehabilitation intervention.

Keywords: Evaluation, Intensive Interdisciplinary Pain Treatment, Adolescents with Pain-Related Disability, Stakeholder Engagement, Logic Analysis, Effect Analysis, Narrative Timelines, Mixed Methods

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LIST OF ABBREVIATIONS

ACH	Alberta Children's Hospital
ACT	Acceptance and Commitment Therapy
ARCS	Adult Responses to Child Symptoms
BAP-PIQ	Bath Adolescent Pain-Parental Impact Questionnaire
BAPQ	Bath Adolescents Pain Questionnaire
BOT-2	Bruininks Oseretsky Test, Version 2
BPI	Brief Pain Inventory
CADTH	Canadian Agency for Drug and Technology in Health
CDI	Child Depression Inventory
CES-DC	Centre for Epidemiology Depression Scale for Children
CHQ-CF87	Child Health Questionnaire
COPM	Canadian Occupational Performance Measure
CPAQ-A	Chronic Pain Acceptance Questionnaire, Adolescent Version
CPC	Complex Pain Clinic
CRPS	Complex Regional Pain Syndrome
CSI	Child Somatization Inventory
FDI	Functional Disability Index
FOPQ-C	Fear of Pain Questionnaire, Child Version
FOPQ-P	Fear of Pain Questionnaire, Parent Version
HADS	Hospital Anxiety and Depression Scale
HRQoL	Health Quality of Life
ICF	International Classification of Function, Disability and Health
ICF-CY	International Classification of Function, Disability and Health- Child and Youth Version
IIPT	Intensive Interdisciplinary Treatment
IPR	Intensive Pain Rehabilitation
JIA	Juvenile Rheumatoid Arthritis
JFM	Juvenile Fibromyalgia
LEFS	Lower Extremity Functional Scale
LMM	Longitudinal Mixed Models
MAS-C	Multidisciplinary Anxiety Scale for Children
MCID	Minimal Clinically Important Difference
MDT	Multidisciplinary Treatment
MMT	Multimodal Treatment
MPI	Multidimensional Pain Inventory Interference Scale
MSPQ	Modified Somatic Perception Questionnaire

NGT	Nominal Group Technique
NRS	Numeric Rating Scale
OT	Occupational Therapy
PAIRS	The Pain and Impairment Relationship Scale
PCQ	Pain Coping Questionnaire
PCS-C	Pain Catastrophizing Scale for Children
PE	Participatory Evaluation
PEM-CY	Participation and Environment Measure for Children and Youth
PedIMMPACT	Pediatric Initiative on Methods, Measurement and Pain Assessment in Clinical Trials
PedsQL	Pediatric Quality of Life
P-PE	Practical Participatory Evaluation
PII	Pain Interference Index
PIPS	Psychological Inflexibility in Pain Scale
PPI	Pediatric Pain Interference
PPFQ	Pain Psychological Flexibility Questionnaire
PRI	Pain Response Inventory
PRO	Patient-Reported Outcomes
PROMIS	Patient-Reported Outcome Measurement Information System
PRS	Pain Reactivity Scale
PRCQ-C	Pain-related Cognition Questionnaire for Children
PSC-C	Pain Catastrophizing Scale, Child version
PSOCQ	Pain Stages of Change Questionnaire
PT	Physiotherapy
PTSD	Post-Traumatic Stress Disorder
QoL	Quality of Life
RCMAS	Revised Children’s Manifest Anxiety Scale
RCT	Randomized Control Trial
SBE	Stakeholder-Based Evaluation
SCAS	The Spencer Children’s Anxiety Scale
SF-36	The Short Form –36 Health Survey
SPOR	Strategy for Patient-Oriented Research
TII	Traitement intensif interdisciplinaire
TMM	Traitement multimodal
T-PE	Transformative Participatory Evaluation
TSK-11	Tampa scale of Kinesiophobia
UEFI	Upper Extremity Functional Index
VAS	Visual Analogue Scale
WHO	World Health Organization

To all the participants in this project without whom
this dissertation would not have been possible.

Listen. People start to heal the moment they feel heard.
Cheryl Richardson

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INTRODUCTION

Chronic pain is a complex clinical problem in the pediatric population. Pain is defined as a distressing experience associated with actual or potential tissue damage with sensory, emotional, cognitive, and social components (Williams & Craig, 2018). It is considered chronic if persistent longer than 3 months (Aydede, 2017). It may be caused by injury (e.g. burns), an underlying condition (e.g. fibromyalgia), or have no specific identifiable cause (McGrath *et al.,* 2008). The most common pediatric chronic pain conditions include headaches and migraines (69.0%), abdominal pain (16.3%), and back or limb pain (13.2%) (Zernikow *et al.,* 2012). Recently, chronic pain has been recognized as its own disease state (Nicolas *et al*., 2019; Treede *et al.,*2019).

It is estimated that 15-46% of children and adolescents experience chronic pain worldwide, and its prevalence has rapidly increased over the past several decades (King *et al.,* 2011; Stanford *et al.,* 2008). Although reasons for this increasing prevalence is unclear, multiple factors including increased pain identification, stress and anxiety, poor role modeling, and maladaptive pain behaviours and attitudes may provide part of the explaination (Landry *et al*., 2015). In many cases, this pain is unresponsive to conventional treatments. For approximately 8% of youth, this pain can lead to pain-related disability, such as psychological distress and sleep disturbance, important activity limitations, and participant restrictions (Lewondowski *et al.,* 2013; Scharff *et al.,* 2005; Tham *et al.,* 2013). Activity limitations include dependence for personal hygiene and mobility disturbances. Participation restrictions are comprised of school absenteeism and cessation of recreation and leisure activities. Emerging literature also suggests that, like other chronic conditions, chronic pain can be detrimental to family functioning (Palermo *et al.,* 2014), social interactions (Forgeron *et al.,* 2010), and to long-term mental health for both youths and their families, and for which consequences can persist into adulthood (Noel *et al.,* 2016).

As a result of frequent and repeated use of healthcare resources (e.g., acute, emergency, diagnostic services, specialist consultation (Ho *et al.,* 2005; Groenewald *et al.,* 2014; Mahrer *et al.,* 2018), chronic pain is recognized among the highest costing chronic health conditions in childhood, with an estimated annual cost of $19.5 billion USD (Groenewald et al., 2014). If not appropriately treated, 35 to 70% of these youths continue to have chronic pain into adulthood (Dengler-Crish *et al.,* 2011; Walker *et al.,* 2010), also recognized as costly in terms of health services and lost productivity costs (Schopflocher *et al.*, 2011). Identification of treatment approaches that improve these troublesome outcomes is imperative (Risko, 2018).

There is strong evidence to support targeted psychological and physical rehabilitation interventions for pediatric chronic pain (Harrison *et al.*, 2019). However, a variety of clinical and family-focused factors may influence specific treatment recommendations and decisions. Typically, youth whose function and participation are less impacted, or for those who, despite deteriorating function in other domains, school absenteeism has remained at a moderate level, are guided to multimodal treatment (Hechler *et al.*, 2011; Wager *et al.*, 2014). Although treatment frequency may vary (e.g. 1 to 3 times per weeks), most MMT includes two or more of the following disciplines providing services on an outpatient basis: medical (e.g. nerve blocks), pharmacological (e.g. new medication or dosage change), psychological (e.g. individualized or group-based cognitive-behavioural therapy), and physiotherapy interventions (Hechler *et al.,* 2011; Lee *et al.,* 2002; Simons *et al.,* 2013). Interventions typically incorporate the implementation of multidisciplinary home recommendations and provide re-evaluation and adjustments to pain management strategies as needed (Hechler *et al.,* 2011; Lee *et al.,* 2002; Simons *et al.,* 2013). These types of MMT programs have been deemed efficacious in reducing pain, pain-related disability, fear of pain, and school absenteeism (Lee *et al.,* 2002; Wicksell *et al.*, 2009; Soee *et al.* 2013; Trans *et al.,* 2017). For some youth, multimodal outpatient treatment may be sufficient, while for others, admission to intensive pain rehabilitation programs may be indicated (Hechler *et al.,* 2011; Hechler *et al.,* 2014a; Simons *et al.,* 2013; Simons *et al.,* 2018).

It is current practice to refer youth more highly disabled by their pain to intensive interdisciplinary pain treatment programs (IIPT), where available, in an effort to interrupt the vicious cycle of pain interference in everyday life, the emotional distress and escalating school absenteeism (Hechler *et al.*, 2009; Wager *et al.,* 2014). Many IIPT around the world (e.g. United States, in Europe, and Australia) have demonstrated promising initial results, with ameliorations reported in pain-related disability, anxiety and depressive symptoms, and school attendance (Banez *et al.,* 2014; Eccleston *et al.,* 2003; Hechler *et al.,* 2009; Hechler *et al.,* 2014a; Hechler *et al.,* 2014b; Hirschfeld *et al.,* 2013; Logan *et al.,* 2012a; Simons *et al.,* 2013). These programs are defined as concurrent treatment provided by a minimum of three health disciplines (medicine, psychology and physical therapy), 6-8 hours per day over several weeks (3-12 weeks) in either an inpatient unit or a day hospital setting (Dobe *et al.,* 2013; Stahlschmidt *et al.,* 2016). Some programs also include occupational therapy (OT), recreation therapy, nutritional counselling, music therapy, and art therapy, as well as some type of school programming (Banez *et al.,* 2014; Eccleston *et al.,* 2003; Hechler *et al.,* 2015; Logan *et al.,* 2010; Maynard *et al.,* 2009; Sherry *et al.,* 2015; Weiss *et al.,* 2013). Parent inclusion and school reintegration are also particular requirements of these interventions (Stahlschmidt *et al.,* 2016). Although many similarities in admission criteria exist between IIPT programs, the exclusion criteria vary widely (Stahlschmidt *et al.*, 2016). IIPT programs are considered time-intensive, and costly (>$30,000 USD per annum) and are often offered as a last hope for families (Evans *et al.,* 2016; Simons *et al.,* 2018). Variability in response to treatment rates have been documented, depending on the variable measured. The inconsistent outcome domain choices across effectiveness studies has impeded comparison between IIPT program models, despite the availability of recommendations for a core set of outcome domains (McGrath *et al.*, 2008).

Moreover, the IIPT and the MMT have rarely been compared empirically and the value of one compared to the other has yet to be clarified. Program variations, as described above, and the poor descriptions of the treatment components in the published literature (e.g., treatment approach, intensity, frequency) have complicated the comparison between programs and their implementation into other jurisdictions (Stahlschmidt *et al.*, 2016).

For over a decade, pediatric outpatient MMT has been available in several Canadian jurisdictions (Peng *et al.,* 2007), one of which is the Alberta Children's Hospital (ACH), in Calgary, Alberta. The ACH MMT encompasses a multimodal intervention program of pain education for youth and their parents, 1-2 one-hour weekly individual physiotherapy sessions, focused on functional stretching, strengthening, and endurance training, 1-2 one-hour individual cognitive behavioural focused psychology sessions weekly, medical treatment (e.g. medication), and home program recommendations (e.g. relaxation practice). Discharge from programs is based on youth and families identified intervention needs.

No intensive rehabilitation programs existed until recently in Canada. A new donor-funded day-hospital intensive pain rehabilitation program was recently introduced at ACH. The IIPT is a 3-6–week interdisciplinary program including physiotherapy, psychology, occupational therapy, recreation, art and music therapy, along with academic, medical and nursing support. Youth spend 6 hours daily, 5 days per week, in individual, group, and family treatment sessions designed to enhance their pain management skills, facilitate emotional adjustment, improve functioning, and return to normal age-appropriate activities. Youth also receive hospital-based academic support. Self-management is emphasized by all disciplines throughout and within all program activities. Discharge is individualized based on youth and parent identified functional goals.

Due to its current funding profile, the IIPT program requires an evaluation to assist provincial decision makers in determining its future, and its impact. This study aims to evaluate the IIPT and compare it to the MMT at the same facility with the purpose of providing decision makers with evidence on its effectiveness. Unlike typical effectiveness studies, the plausibility of the IIPT program in achieving the anticipated outcomes will first be assessed. This study presents a unique opportunity to expand the scientific evidence in specialized pediatric pain rehabilitation and its delivery. In particular, this study uses multiple, unique, and underutilized research approaches in the field of pediatric chronic pain. It provides an opportunity to build a shared understanding among program stakeholders (e.g., youth and their parents, clinicians) about the program design and its empirical foundation, and about how these programs should be evaluated. This information is not only critical for local and provincial policy makers in determining appropriate rehabilitation services for youth living with this disabling condition but is also pivotal in improving the lives of youth with chronic pain and their families in Canada and beyond.

CHAPTER 1. LITERATURE REVIEW

1.1.Pediatric Chronic Pain Prevalence

Defining pediatric chronic pain is not easy, as inconsistencies in terminology exist in the field. The International Association for the Study of Pain (IASP) defines pain as an unpleasant sensory and emotional experience associated with actual or potential tissue damage or described in terms of such damage (Merksey *et al.,*1979). Recently, chronic pain has been recognized as a new diagnosis in the International Classification of Disease, version 11 (ICD-11). Moreover, a temporal criterion (i.e., pain lasting or recurring for longer than 3-months) was chosen to clearly operationalize its definition and align it with widely used criteria in other health fields (Kumar & Elvarasi, 2016; Treede *et al.,* 2015).

Prevalence estimates vary widely depending on geographical location and medical diagnosis. Global prevalence estimates are most often reported based on pain location and fail to provide a broad overview of chronic pain. For example, in the general population, the prevalence of headaches is reported to be between 8 and 83%; abdominal pain 4 to 53%; musculoskeletal, 4 to 40%, generalized pain 4 to 49%, and other types of pain 5 to 88%, with a median prevalence of 11-38% in community surveys (King *et al.,* 2011). Multi-site chronic pain has been reported as being more prominent than single-site pain among adolescents (Hoftun *et al.,* 2011; Swain *et al.,* 2014). In Canada, the prevalence of chronic headaches is 11.4%, backache 8.1%, abdominal pain 4.0% and multi-site pain 20.5% (Gobina *et al.,* 2018).

Evidence suggests that children and adolescents with chronic pain waiting for or assessed by tertiary services present with more significant impairments and limitations than those identified in the aforementioned population-based samples. For example, in a group of 2239 German youths presenting to tertiary care, 30% reported constant pain, with an alarming median maximum self-reported pain intensity of 9 out of 10 on the numeric rating scale (0=no pain, 10=worse imaginable pain) (Zernikow *et al.,* 2012). Chronic pain at this severity level may be further exacerbated through inappropriate or inadequate medical treatment, self-medication, or inadequately monitored of pharmaceuticals (Hechler *et al.,* 2010b; Pereira *et al.,* 2007).

Substance abuse, overdose, and adverse medication side effects are not uncommon in youths with chronic pain (Coffelt *et al.,* 2013). These youths are commonly referred to specialists such as neurologist, orthopedic surgeons, rheumatologists, gastroenterologists, psychiatrists, which due to wait-times can delay treatment. By the time many reach a tertiary pain centre, they have typically consulted at least four physicians (Zernikow *et al.,* 2012). Finally, admission and readmission rates to hospital are substantive, the number of diagnostic evaluations (including invasive procedures) are higher than average, and the observed length of stay is consistently longer than as compared to similar (Coffelt *et al.,* 2013).

This group of children and adolescents also experience other negative impacts as a consequence of their pain. These include: sleep and appetite disturbances (Palermo, 2000; Perquin *et al.,* 2000), severely impaired daily activities (Coffelt *et al.*, 2013; Zernikow *et al.*, 2012), a considerable amount of missed school (Kashikar-Zuck *et al.*, 2002; Logan *et al.,* 2008; Simons *et al.,* 2010b; Zernikow *et al.*, 2012), and failure to engage in extracurricular activities, such as athletic, recreation, and/or social events (Simons *et al.,* 2010b). They also experience emotional distress (i.e., anxiety and depression) (Kashikar-Zuck *et al.*, 2001; Kaczynski *et al.,* 2011) and report poorer quality of life in response to their pain (Hunfeld *et al.*, 2001; Gold *et al.,* 2009). These functional difficulties are reported in a variety of contexts, including home, school, and the community setting. The lack of common terminology across the field of pediatric chronic pain is recognized as an important contributor to delayed treatment and negative outcomes in this population (Basch *et al.*, 2015).

1.2. Pediatric Pain-Related Disability

Across studies from the USA, Europe and Canada, approximately 3-8% of youth with chronic pain reported substantial interference with functioning, including school, social and relationship functioning, and family relationships (Palermo, 2000; Palermo *et al.*, 2004). These consequences not only affect the child, themselves, but also their family, and the larger community (Cofflet *et al.,* 2013; Palermo, 2000; Palermo & Eccleston, 2009; Palermo *et al.,* 2004; Zernikow *et al.*, 2012). These youths are very difficult to treat (Maynard *et al.*, 2010), and their long-term prognosis is poor (Walker *et al.,* 2010).

The term "pain-related disability" was first defined as the difficulty that impairs the youth's ability to perform daily activities due to his/her health status (Walker & Greene, 1991). The term has been used to describe a chronic pain condition in which youth suffer increased social isolation, resulting in depression and withdraw from normal activities, regardless of location of the pain or its cause (Palermo, 2000). More specifically, functional disability has been defined in the context of pediatric chronic pain as limitations in a person's ability to perform age-appropriate activities relevant to daily life and social participation as a result of their health status (Stein & Jessop, 1990; Walker & Green, 1991). The definition was initially focused on adult chronic pain, and related to the individual's work performance, and was later expanded to include virtually all aspects of an individual's functioning within their environment such as personal care, social interaction, household activities, recreation, as well as work (Palermo, 2000). This expanded definition was then applied to childhood functional disability by Palermo, 2000. Once adopted, this definition led to the adoptation of the World Health Organization's (WHO) *International Classification of Impairment, Disability, and Handicaps* (1980) as the initial guidance for a broader psychosocial assessment of functioning in youth with chronic pain. Based on this model, child's pain experiences were defined across four levels: 1) the underlying disease or disorder (the physical cause of pain); 2) an impairment (pain itself and its symptoms); 3) a disability (activities that are restricted as a result of pain); and 4) a handicap (impairment or disability that impacts normal social functioning) (Kashikar-Zuck *et al.*, 2001; Palermo, 2000). This model had important implications for improving the assessment of functioning in children and adolescents with chronic and recurrent pain, clearly separating the measurement of pain systems from the assessment of functional consequences (Palermo, 2000). Moreover, it provided guidelines for evaluating the severity of the child's functional problems and disability resulting from pain and began to raise awareness of the importance of behavioural, cognitive, and affective factors that contribute to the onset and maintenance of disability in this population (Kashikar-Zuck *et al*., 2001; Palermo, 2000).

As with many conditions, the *International Classification of Impairment, Disability, and Handicaps* (WHO, 1980) was useful in the field of pediatric chronic pain in significantly advancing the conceptualization of disability. It did so by: 1) theorizing disability as a consequence of an underlying condition, attributable to a disease or injury, 2) differentiating these consequences at three different levels of the human experience; that of the body, the person, and society; and 3) emphasizing the multiple dimensions of disability, in the form of impairment, performance limitations, and the experience of disadvantages (Simeonsson *et al*., 2003). However, a need for revisions to the *International Classification of Impairment, Disability, and Handicaps* (WHO, 1980) arose due to policy and concepts challenges. More specifically, from the policy perspective, the framework needed to shift from a medical based model to one that reflected the social and cultural dimensions of disability (Simeonsson *et al*., 2003; Simeonsson, 2009). Conceptually, this policy shift paralleled a recognition of the crucial role of the environment in defining human functioning, the failure of the *International Classification of Impairment, Disability, and Handicaps* (WHO, 1980) model to recognize the dynamic nature of disablement, and a growing negativity surrounding the term 'handicap' (Simeonsson *et al*., 2003).

As a result, a biopsychosocial conceptualization of function emerged referred to asthe *International Classification of Functioning, Disability and Health* (ICF) (WHO, 2001). As evidenced by its revised content, this internationally recognized framework of function and disability acknowledges functioning as a human experience and encompasses the contribution of individual attributes, the disease process, and environmental facilitators and barriers to the performance of personal activities, and levels of participation in daily communal life (Sinclair *et al.,* 2016; WHO, 2001). Moreover, it distinguishes functional and structural impairments from limitations in personal activities and restrictions in social participation, observing that these limits can occur as a result of contextual factors and not strictly because of the impairment (Swanson *et al.,* 2011). It acknowledges functioning as the product of the five dimensions, i.e., body structure and functions, activity, and participation, environment and personal factors

(WHO, 2001). Participation is defined as involvement in life situations affecting health and well-being (WHO, 2001) and for youth may involve aspects of daily life such as education, work or employment, community living, and healthy interactions with family and peers (WHO, 2007). Degrees of impairment, activity limitations, and restriction in participation are particularly recognized in the disability experience, the disablement process and the impact on the individual's quality of life (Swanson *et al.,* 2011). Furthermore, the framework provides a particular focus on the influence of the environment on functioning, an important acknowledgement in the disablement process, particularly for youth when their environments are dramatically changing (Bronfenbrenner & Ceci, 1994), and where child's subsequent interactions the context frames their skill acquisition (Simeonssen *et al.,* 2003; Simeonsson, 2009).

Unfortunately, since its emergence, the ICF framework (WHO, 2001) has not been applied to the field of pediatric chronic pain. However, distinct chronic diseases for which pain is an important symptom (e.g., Ethler Danlos, Juvenile Idiopathic Arthritis (JIA)) have embraced it and applied it in many ways. For example, Houghton (2015) suggested that the ICF provides an excellent framework for the measurement of health and disability in youth with JIA. Applying the framework illustrates that a child with JIA may demonstrate restricted activity and participation in leisure activities due to impairments in body structure and functions (e.g., joint swelling, joint pain, muscle weakness, fatigue), which may be influenced by the child's personal factors (e.g., age, sex, motivation). Environmental factors (e.g., family functioning, peer support) may act as facilitators or barriers to fostering or hindering the child's participation in the activity (Houghton, 2015). Another example provided by Scheper and colleagues (2013), applied the *ICF-Child and Youth version* (WHO, 2007) to general joint hypermobility syndromes (e.g., Ethler Danlos). In doing so, they tease out the factors associated with those common to the condition (i.e., laxity of the connective tissue, chronic pain), from ensuing psychological distress (e.g., fear of movement and pain), and those attributable to the environment. As a result, they were able to better explain the observed functional limitations (e.g., physical fitness), and participation restrictions (e.g., school and social functioning). In both JIA and generalized hypermobility conditions, the development of clinical core datasets using the ICF *Child and Youth versions* has been proposed (Houghton, 2015; Scheper *et al.*, 2015). These core datasets could lead to a better understanding of the potential effects of the condition on youth activities and participation, inform the development of clinical measures, improve condition monitoring, and identify profiles and trajectories within these populations (Houghton, 2015; Scheper *et al.*, 2015).

The adoption of a framework specific to pediatric rehabilitation and one that provides a conceptualization of "disability" and "function" could be useful in youth with chronic pain, in particular the subset of those with pain-related disability. For instance, the *ICF for Children and Youth* (WHO, 2007) would allow for the conceptualization of the well-documented non-linear relationship associated between changes in youth's pain symptoms and functional impairments (Palermo, 2000). Furthermore, it brings to the forefront the importance of the environmental interactions and the influences of personal attributes that have been underscored in pediatric chronic pain (Vinall *et al.*, 2016), which, to date, have been difficult to represent with other models. It offers a common language for professionals in health, rehabilitation, social services, and education to use in their description of youth's function and disability, across setting and disciplines, which could be pivotal in supporting service provision based on functional profiles, instead of categorical or medical diagnoses (Simoeonsson, 2009). Finally, it may also provide a valuable tool in better defining the pain experience, not limiting it to pain intensity and frequency but instead to its disabling consequences, and subsequently assist in identifying appropriate outcomes to better capture the devasting daily and longitudinal life impact of the condition on youth, their families and their communities.

1.3 Contributors and Consequences of Pediatric Chronic Pain

The understanding of the mechanisms contributing to chronic pain in pediatrics has expanded exponentially in the past two decades (Caes *et al.,* 2018). Pain is best understood through its various influences, including biological, social and psychological factors, and should take into account the individual's personal attributes and the complex context that surrounds it (Logan *et al.,* 2012c). It is the interaction among factors, attributes, and context that creates the meaning to the pain experience (Butler and Moseley, 2013). In keeping with this understanding, the next section will present the best evidence associated with the contributing factors to pediatric chronic pain and pain-related disability. It should be highlighted that some pain contributors are thought to be bidirectional. Therefore, when appropriate, some will be presented as factors influencing the pain experience as well as consequences.

***1.3.1. Biological factors**:* Chronic pain can be caused by a myriad of physiological and biological factors and attributes, which may include disease mechanisms, neurophysiological processes to the stress response, genetics, sex, and hormones. Chronic pain includes persistent and recurrent pain in children from underlying health conditions (e.g., JIA, sickle cell disease), as well as being a recognized disorder itself (e.g., primary headaches, complex regional pain syndrome) (Friedrichsdorf *et al*., 2016). Pain becomes chronic when it loses its adaptive function, no longer serving its warning role (Simons *et al.,* 2014). Chronic pain affects the entire nervous system, and the term central sensitization is used to describe this increased neural responsiveness to painful and non-painful stimuli, which contributes to the development and maintenance of many types of chronic pain (Woolf, 2011). Chronic pain is thought to arise from nociceptive stimulation in neuropathic conditions, peripheral and central sensitization processes, along with the reorganization of the nervous system (Aronoff, 2016). The neuronal sensitization theories of pain suggest that when exposed to repeat pain sensations, memory traces on a neuronal level will develop, increasing the sensitivity to further sensation, and a typical benign sensation can be interpreted as pain (vanRavenzwaaij et al., 2010). Larger portions of the brain can be subsequently rewired, and hypersensitivity occurs. Neuroimaging studies have demonstrated changes to various brain structures (Geraghty and Buse, 2015, Vinall *et al.*, 2016). For example, reduced grey matter in the primary motor cortex, premotor cortex, supplementary motor cortex, midcingulate cortex, orbitofrontal cortex, dorsal lateral pre-frontal cortex, posterior cingulate cortex, precuneus, basal ganglia , thalamus, and hippocampus have been shown with magnetic resonance imaging in youth with CRPS (Erpelding *et al,* 2016). Due to the incomplete neurological maturation in children and adolescents, these effects may be even more profound in these age groups than initially anticipated (Mathew *et al.*, 2014).

The physiological stress response underlies many postulated biophysiological pain theories and its chronification. For example, with stress, an immune system response may signal a cytokines release hypothesized as a contributor to chronic pain (Vinall *et al.,*2016) Prolonged and chronic cytokines releases can lead to mood disorders, amplify the perception of pain, cause sleep disturbances, and fatigue (Irwin, 2011). Dysregulation of the hypothalamus-pituitary axis within the endocrine system and its ability to regulate the body's stress response have also been linked to chronic pain. Irregularities in the release of cortisol, a hormone released with stress, have been shown in some chronic pain patients (vanRavenzwaaij *et al.,* 2010). Chronic increases in cortisol levels have been shown to affect some brain structures, including the hippocampus, amygdala, and hypothalamus (Walco *et al.,* 2016). In addition, the involvement of some neurotransmitters has been questioned. For example, decreased levels of serotonin, a widely distributed neurotransmitter, key to modulating the stress response, have been found in individuals with chronic pain, which may be related to the activation of the hypothalamus-pituitary axis (Vinall *et al.*, 2016). Although it is clear that the hypothalamus-pituitary axis plays a role in the pediatric chronic pain mechanisms, the direct clinical implications are not yet fully understood (Walco *et al.,* 2016).

Genetics have also been implicated in chronic pain. Some researchers have theorized that specific individuals may present with an underlying genetic vulnerability predisposing them to developing chronic pain (Nyman *et al.*, 2011). Evidence also exists that genetic variants can lead to variability in inflammation, which can trigger the aforementioned immune or endocrine system response (Irwin, 2011). Furthermore, in addition to direct pathways, genetics may also play a role in the expression of other factors, such as anxiety and depression, that co-occur with chronic pain (Soltani *et al.*, 2019; Vinall *et al.*, 2018).

Finally, age and sex are important biological attributes of pediatric chronic pain. Increased age, typically early adolescents, is associated with various pediatric chronic pain disorders; however, variation exists. For example, localized chronic head and backaches tend to increase during adolescence, whereas the prevalence of abdominal pain decreased with age (Gobina *et al.,* 2018). Some researchers have also suggested that pubertal development during adolescence may be a better chronic pain determinant than age (Janssens *et al.,* 2011). Studies have also highlighted that girls have higher rates of pain conditions than boys across all subtypes (Standford *et al.*, 2008), while others have identified sex patterns (Gobina *et al.,* 2018). Several studies have also recognized a potential relationship between age, sex and puberty, with a strong predictive relationship demonstrated between pediatric chronic pain conditions, sex and attaining puberty particularly in girls (LeResche *et al.*, 2005; Sperotto *et al.*, 2015).

However, adopting a strictly biological approach to chronic pain leads to a purely disease focused understanding of the condition and which may be very different from the youth and parent's experience (Mondivais, 2011; Risko, 2018). It is acknowledged that an array of psychological, social, and developmental variables come into play in modulating pain, affecting the development and maintenance of chronic pain (Butler & Mosley, 2013; Walco *et al.*, 2016). To adequately treat youth with chronic pain, one must move away from exploring solely its biological and physiological basis and adopt a dynamic conceptualization and an awareness of the modulating factors with which the pain experience is associated (Walco *et al.*, 2009; Walco *et al.*, 2016).

1.3.2. Psychological Factors and Functioning: Multiple comorbid psychiatric conditions such as anxiety, depression, and post-traumatic stress syndrome (PTSD), have been associated with pediatric chronic pain. The co-occurrence of these disorders is thought to be bi-directional, with psychological symptoms identified as both contributors and consequences of pediatric chronic pain (Nelson *et al.,* 2017; Kozlowska *et al.,* 2008). Children and adolescents with persistent pain express higher levels of anxiety and depression, particularly among girls and those with generalized pain (Simons *et al.,* 2012b). As a central construct to pain-related disability, anxiety has shown to be associated with poorer functional outcomes regardless of the pain intensity. More specifically, Cohen and colleagues (2010) noted that at high anxiety level, pain intensity was not predictive of functioning, however, at low anxiety level it hindered function. Pain-specific anxiety, such as fear of pain, has demonstrated more direct associations with pain-related disability. For example, pain-specific anxiety has been associated with poorer physical function, depression and poorer family functioning (Eccleston *et al.,* 2005). In addition, pain-related fear reportedly accounts for 40% of the variance in pain-related disability (Martin *et al.,* 2007). Pain catastrophizing, or extreme cognitive worry related to pain, is a recognized predictor of pain intensity and decreasing function overtime (Benore *et al.,* 2015;

Vervoot *et al.,* 2010). These findings are however inconsistent across pediatric chronic pain studies (Engel *et al.,* 2012).

Depressive symptoms have consistently been found to predict disability in adolescents with chronic pain, including poorer physical and social/adaptive functioning (Gauntlett-Gilbert & Eccleston, 2007). Moreover, depressive symptoms in pediatric pain have been linked to more school impairment, and perceptions of higher pain intensity (Logan *et al*., 2008). Furthermore, both sexes are at risk of developing depressive symptoms in musculoskeletal pain (Egger *et al.,* 1999). Depression has also been linked to sleep disturbances, a common co-morbidity found in this population (Palermo & Kiska, 2005; Valrie *et al.,* 2013). Characterized by a difficulty falling asleep, staying asleep, poor subjective quality of sleep, short sleep duration, poor sleep hygiene or habits (Valrie *et al.,* 2013), these disturbances may be associated with underlying disease-related mechanisms (e.g., inflammation), intervention regimens (e.g. medication side effects) or hospitalizations (Lewandowski *et al.,* 2011; Lewin & Dahl, 1999). Poor sleep is associated with compromised physical, social, emotional, and cognitive functioning in adolescents with persistent pain, above and beyond the effects of pain (Motivala & Irwain, 2007; Palermo & Kiska, 2005).

Finally, a connection between trauma (physical, sexual) and chronic pain has also emerging (Egger *et al.,*1999; Youssef *et al.,* 2008). Recently significantly higher levels post-traumatic stress syndrome (PTSD) have been reported iin youth with chronic pain and their parents, as compared to the regular population (Noel *et al*., 2016).

***1.3.3. Social Factors and Functioning**:* Much like the biological and psychological determinants of pediatric chronic pain, social factors interact with the social context and across the biological and psychological contributors. These contextual social systems play an important role in shaping the pain experience, with family (including parents and siblings), school and peers being the predominant networks of influence in this population (Logan *et al.,* 2012b, Logan *et al.*, 2012c).

1.3.3.1. Family factors: Irrespective of the health care system, children can experience inequalities in accessing pediatric pain services, due to family social differences and poverty

(Ruhe *et al.,* 2016). Low socio-economic status has been associated with greater functional disability due to pain (Palermo *et al.*, 2008) and more limited access to treatment (Ruhe *et al.,* 2016). An extra financial burden, associated with medical assessments, diagnostic, treatment and management of recurrent and chronic pain, along with the cost associated with time off work, transportation, and additional childcare for siblings has also been underscored (Sleed *et al.,* 2005; Le *et al.,* 2019).

Family influences on pediatric chronic pain operate at many different levels. These influences can include individual (e.g., parental protective responses to pain), parent-child relationship, and family environmental factors, each influencing and being influenced by the children's pain experience (Logan *et al.,* 2012c; Palermo *et al.,* 2014). According to a systematic review on the family factors in pediatric chronic pain, the most studied contributing factors include family history of chronic pain, parental emotional distress, parental thoughts/behaviours related to pain (e.g., parent pain catastrophizing), parent-child interactions and overall family environment and functioning (e.g., the extent of conflict) (Lewandowski *et al.,* 2010). This systematic review concluded that the best evidence exists related to the association between family functioning and pain-related disability, with better family functioning associated with lower levels of youth pain-related disability (Lewandowski *et al.,* 2010). A recent qualitative study by Le and colleagues (2019) further expanded on these familial impacts, to include disruptions of family goals and commitment, the inability of the family to engage in valued and healthy coping activities, and the control the child's pain exerted on the whole family.

1.3.3.2. Parental factors: Parent emotions, behaviours, and health play a role in children's pain experiences. Overly protective parent behaviours, increased distress, and history of chronic pain are important parent-level influences (Hoftun *et al.,* 2013; Palermo *et al.,* 2014). Research has demonstrated that a number of parents of children with chronic pain experience negative emotions, mental health problems, and negative social outcomes (Jordan *et al.,* 2007; Jordan *et al.,* 2016; Maciver *et al.,* 2010; Noel *et al.,* 2016). Parents have also reported increased emotional distress, including anxiety and depressive symptoms, role stress, frustrations related to coping and adapting to the child's pain condition, fear, helplessness, exhaustion and care burden associated with their child's pain (Jordan *et al.,* 2016; Maciver *et al.,* 2010).

Furthermore, important lifestyle, health and quality of life consequences have also been underscored in several studies (Gaughan *et al.,* 2014; Le *et al.,* 2019).

A child learns foundational behavioural skills from and within a familial context (Guite *et al.,* 2011) and the existence of a reciprocal relationship between parental behaviour, and child pain coping and functioning have been demonstrated (Palermo *et al.,* 2014). Parental pain catastrophizing is an important behaviour that has been extensively studied. Maternal pain catastrophizing has demonstrated a significant relationship to children's pain intensity but has yet to be associated with pain-related disability (Hechler *et al.,* 2011). However, this behaviour has been associated with child activity limitations, which may subsequently impact on youth's functioning (Caes *et al.,* 2011). Overt attention to children's pain behaviours (i.e. protective response) has been related to increased disability (Claar *et al.,* 2008; Claar *et al.,* 2010), maladaptive coping (Logan & Scharff, 2005), and increased school absenteeism (Logan *et al.,* 2012b). Although gender differences in parental responses have been recognized, little is known about the psychological functioning of fathers in this population (Palermo *et al.,* 2014).

Finally, parental health status can also influence youth's chronic pain. A recent study identified that 50% of parents of youth with chronic pain reported chronic pain themselves (Beveridge *et al.,* 2018). Many types of pain conditions cluster within families (Antillia *et al.,* 2000; Boey & Goh, 2001). A correlation between parental chronic pain and non-specific and multisite pain in adolescents does exist, with the presence of maternal and paternal chronic pain increasing the odds of chronic pain in adolescents (Hoftun *et al.,* 2013). These connections could highlight a genetic vulnerability to chronic pain conditions, or indicate that modelling of pain coping behaviours, the environment, and the family structure are significant contributors to the emergence of chronic pain in youth (Hoftun *et al.,* 2013).

1.3.3.3. Sibling factors: Although a plethora of parent-focused research exists, siblings have received little attention in the pediatric chronic pain literature. In a recent scoping review on this topic, Sckinkel and colleagues (2017) noted that studies have primarily focused on genetic vulnerability suggesting a prominent focus on the biological component of pediatric chronic pain. However, secondary interests in siblings' mental health and the psychological factors related to pain have also emerged. Study findings suggest a general negative influence of pediatric chronic pain on siblings, with reports of poorer functioning, including increased anxiety and depressive symptoms, along with peer and social difficulties in siblings of children with chronic pain (Guite *et al.,* 2007; Schinkee *et al.,* 2018). These findings align with research in other pediatric chronic conditions suggesting that a sibling's experience of illness can adversely influence a child's psychological functioning, their quality of life, and their academic participation and performance (Gan *et al.,* 2017; Limbes & Skipper, 2014; Vermaes *et al.,* 2012). Research has also focused attention on the different perspectives related to family processes and responsibilities which arise between siblings with and without chronic pain. Siblings of those with chronic pain report more significant changes in various aspects of family life, including roles and responsibility, the quality of their relationship with the affected family members and the influences of pain on the family system (Gorodzinsky *et al.,* 2013).

1.3.3.4. Peer factors: Many pediatric chronic pain conditions are exhibited in mid-adolescence, a critical developmental period for social development. Adolescence is typically a time where autonomous behaviours emerge, dependence of parents is reduced, and an increase reliance on peers occurs. In adolescents with chronic pain, social deficits and parental protectiveness can hinder the development and progression of social functioning (Walker *et al.,* 2002; Forgeron *et al.,* 2010). Youth with chronic pain have been reported to judge themselves to be less socially developed than their peers (Eccleston *et al.,* 2008). However, Eccleston and colleagues (2008) also suggest that strong peer relationships may also play a protective role, in youth perception of social development including improved independence, emotional adjustment, and identity formation.

Nevertheless, youth with chronic pain report fewer friendships, experience increased level of victimization and social isolation, and receive fewer positive ratings by peers compared to children without chronic pain (Forgeron *et al.,* 2010). Furthermore, evidence suggests that chronic pain may interfere more extensively with peer relationships compared with other types of chronic conditions (Konijnenberg *et al.,* 2005). Fatigue, low mood and self-esteem, and the cognitive demands associated with chronic pain, may be additional contributors to the lower levels of social competence seen in this population (Forgeron *et al.,* 2013; Jordan *et al.,* 2017). Youth with chronic pain also perceive non-supportive social situations with close friends as more distressing than their peers, feeling left out and different (Forgeron *et* *al.,* 2011). Youth with chronic pain often choose not to disclose their pain condition, to hide their differences, and are unsure on how to secure relationship needs (Forgeron *et al.,* 2013; Geraghty & Buse, 2015). This may lead to avoidance of social situations, further isolation, social distancing, and ultimately contribute to further negative social outcomes not only with their peers, but also with romantic partners (Forgeron *et al.,* 2011; Fales & Forgeron, 2014). Social functioning plays an important role in the functioning of youth with chronic pain, mediating the relationship between pain and school impairment (Simons *et al.,* 2010b).

1.3.3.5. School functioning: School is often cited as the work of childhood and an important setting for development (Logan *et al.,* 2012b; Jones *et al.,* 2018). It is well known that children with chronic pain have impaired school functioning (Logan *et al.,* 2008). In a recent study, Agoston and colleagues (2016) highlighted that as a group, youth with chronic pain reported more school absences, lower quality of life in the school setting, and more visits to the school nurse as compared to adolescents with juvenile idiopathic arthritis-related pain and healthy peers. School functioning research in pediatric chronic pain has particularly focused on the impact of chronic pain on school attendance, with many children missing extensive amounts of school, up to 50% or more in some cases (Logan *et al.,* 2008; Sato *et al.,* 2007). Prolonged school absenteeism is one of the most disruptive events in a child's development (Kearney, 2001). Not only do these students miss valuable academic time, they also miss the opportunity to engage in other important school experiences such as developing independence and their self-identify, as well as interacting and connecting socially with others (Forgeron *et al.,* 2013). Negative effects of chronic pain on school performance (Logan *et al.,* 2008; Sherry *et al.,* 1991) and the sense of academic confidence have also been recognized (Gorodzynsky et al., 2011). Studies have found lowering grades since chronic pain onset in this population (Logan *et al.,* 2008), and lower reading and numeracy scores as compared to peers (Kosola *et al.,* 2017). Lower self-reported academic competence has also been reported and its association with negative functioning has been demonstrated (Claar *et al.,* 1999; Guite *et al.,* 2007; Logan *et al.,* 2008).

Although some evidence of cognitive and school functioning does exist, it is limited. Research has found specific processing deficits related to working memory and divided attention in youth with chronic pain, despite intact global intellectual functioning (Dick &

Pillai Riddell, 2010; Mifflin *et al.,* 2016). Clinically relevant impairments in overall executive functioning and attentional control have also been demonstrated (Hocking *et al.,* 2011). Sleep deprivation, negative interactions with school personel, and difficulties coping with school and academic demands have been postulated as contributing factors (Gorodzinsky *et al.,* 2011).

Like family and peer interactions, interactions with teachers may also play a role in shaping youth pain experience and how they cope in the school environment (Castarlenas *et al.,* 2015). Although the body of knowledge is still limited, some interesting findings should be considered. Teachers report uncertainty associated with the legitimacy of youth chronic pain complaints. For example, if related to a medical diagnosis, teachers have been found to be more supportive, willing to provide school-based accommodations, yet still struggle to appropriately help youth with chronic pain in the classroom (Logan *et al.,* 2007; Solé *et al.,* 2018). Solicitous responses, i.e., providing a reinforcing response when youth display pain behaviours (e.g., relief from responsibilities), although often viewed to be positive, have been consistently associated with, and shown to contribute to, greater dysfunction in youth (Caes *et al.,* 2011; Claar *et al.,* 2008; Walker *et al.,* 2002). These responses have primarily been studied in the parent-child relationship. However, in a recent study, Casterlenas and colleagues (2015) found that teacher's solicitous responses were quite likely to occur for children with chronic pain and may inadvertently contribute to the development and maintenance of chronic pain behaviours, and disability. As important contextual differences between the home and school environment, the effects of these responses may be different and should be further explored (Casterlenas *et al.,* 2015).

1.3.4. Personal Attributes and Traits: The perception and interpretation of one's pain have been acknowledged as determinants in the pain experience. Early studies demonstrated that personality traits and characteristics of individual's thinking process were indicators of how each person processes pain. In the context of pediatric pain, fearful temperament has been identified as a predictor of long-term pain complaints (Wolff et al., 2010; Rocha & Prkachin, 2007). Furthermore, adolescents with chronic pain are more vulnerable to experience a broad range of negative feelings (e.g., distress, worry) which have been associated with a heightened pain experience (Merlijn et al., 2003). Optimism, the general tendency to expect a positive outcome, has recently received increased attention in the context of pediatric chronic pain, being linked to lower pain intensity and improved mental health in theseadolescents (Mannix *et al.,* 2009; Williams *et al.*, 2010). Finally, perfectionism, broadly defined as a personality trait that involves overly subjecting one's self to high standards along with self-criticism, has been clinically observed among youth with chronic pain for some time, yet empirical evidence is still in its infancy (Randall *et al.,* 2018). Although these aforementioned studies suggest that personality and temperament traits have a direct effect on pain experience, others have proposed that they should be included as variables within comprehensive pediatric chronic pain models (Cousins *et al.,* 2015).

1.3.5. Developmental Perspective*:* The developmental aspect and processes of pediatric pain-related disability are what distinguishes it from adult chronic pain, and shape the pediatric pain experience (McGrath, 1975; Palermo et al., 2014). Adolescence is of particular interest in this developmental trajectory as it is a particularly critical period characterized by rapid change in social, psychological and cognitive domains, and is the time at which the prevalence of pediatric chronic pain conditions is known to spike (Eccleston et al., 2008; King et al., 2011). In addition to the previously discussed impact on social development, chronic pain may also influence identity development, another milestone of adolescence. When examined, youth narratives revealed feeling different from their peers and perceived pain as a barrier to undertaking future goals (Meldrum et al., 2009). Furthermore, Jordan and colleagues (2018) showed that adolescent development of identity, autonomy and emotional maturity were invariably affected by the interruption associated with the chronic pain experience. Developmental expectations were transformed, with accelerations noted in some domains (e.g., emotional maturity, health system navigation), halted in others (e.g., smaller social networks), while others involved a fundamental change in trajectory (e.g., identity: future goals) (Jordan et al., 2018). Furthermore, these authors also highlighted that identity development was a personal phenomenon for some, while others described it as viewed through a broader external social lens (Jordan et al., 2018).

Youth with pain-related disability are at high risk of experiencing continued pain and associated disability into adulthood (Leino-Arjas *et al.*, 2018; Walker *et al.*, 2010). Like in many pediatric chronic conditions, the period of late adolescent and early adulthood, where important developmental milestones may not have been reached resulting in lifelong individual and societal consequences has recently received some attention (Rosenbloom et al., 2018). In addition to neurobiological, psychological and behavioural changes, attainment of societal milestones, adjustment to contextual life change (e.g. living independently) and the emergence of risk behaviours (e.g. substance use, decreased medication adherence) are hallmarks of this developmental period (Copeland et al., 2013; DiRezze et al., 2016). All these expected changes may have important consequences on chronic pain management. Evidence suggests that in some chronic pain conditions (e.g., juvenile-onset fibromyalgia), youth and young adults are less likely to achieve higher education or gain full-time employment compared to their pain-free peers (Kashikar-Zuck et al., 2014). Chronic pain has also been shown to significantly impact transition to independence from parents during early adulthood, as a result of a lack of personal and financial resources, disability, low levels of autonomy, and not progressing to higher education (Rosenbloom et al., 2018). In addition to limited socialization with peers, adolescents with chronic pain have been shown to spend less time with and have more negative experiences involving romantic partners than peers without pain (Pilapil et al., 2015). Finally, older adolescents and young adults have been reported to experience difficulty accessing healthcare (Stinson et al., 2013). This is likely due to a combination of factors, including the transition from pediatric to adult care. However, unlike other childhood chronic conditions, acknowledgement of the challenges of this transition phase is just beginning in this population. Emerging findings suggest that although these young adults may share challenges similar to other youth with chronic conditions, they also experience some that are unique (Higginson et al., 2019). Formalized transition processes are not well established in many centres that serve young people with chronic pain, which can lead to negative outcomes (Higginson et al., 2019). Furthermore, a need for an earlier initiation of the transition process and the inclusion of a number of important providers within the youth's current and future health system context (e.g. general practitioner) were recognized as paramount in the transition of care of youth with chronic pain (Higginson et al., 2019).

In summary, research has focused on identifying various factors predictive of pain chronification, and those contributing to shaping the consequence inherent to the pain experience. As a result, a vast amount of evidence exists that support a number of biological, psychological, and social risk factors and contributors to the chronic pain experience in children and youth. Stress and its associated biophysiological responses, along with sleep problems account for many of the factors investigated within the biological domain. Fear, catastrophizing anxiety, depression, and trauma created the foundation in the psychological domain. Parental behaviour research dominates the findings in the social domain. However, chronic pain is also perceived and interpreted by individuals through personal factors (i.e., individual attributes) and environmental facilitators and barriers existent within their social and developmental context.

1.4. Pediatric Pain-Related Disability Interventions

Clearly, chronic pain in children and adolescents is multifactorial and multidimensional, and multidisciplinary intervention approaches for addressing it are critical (Harrison *et al.,* 2019; Landry *et al.*, 2015). These approaches have primarily focused on medical (i.e., pharmacological) psychological, and physical interventions (Caes *et al.*, 2018; Walco & Goldstein, 2008). Emerging evidence regarding the lack of effectiveness in medication-based treatments alone has further emphasized the role of comprehensive multidisciplinary approaches and has helped to expand the understanding of the significant challenges of managing chronic pain in children and adolescents (Landry *et al.,* 2015). While treatment interventions can be successful when delivered in isolation, specialized multidisciplinary pain treatments have gained increased support as the treatment of choice for chronic pain conditions (Odell & Logan, 2013; Stinson *et al.*, 2016). This portion of the literature review will focus on specialized treatment interventions.

Specialized multidisciplinary pain interventions are within the realm of rehabilitation, where a shift away from symptom reduction and pain elimination is replaced by the idea of pain 'management', i.e., living well with pain, and optimizing one's quality of life (Kamstrup *et al.*, 2019; Lotze & Mosley, 2015). Initially created for adults with chronic pain, these programs have demonstrated cost effectiveness, even more so than pure medical treatments, and better associated outcomes, such as reduced medication and health service utilization, higher rates of return to daily function and work, and increased closure of disability claims in adult clients (Stanos, 2012; Turk, 2002). This treatment model has also gained increasing favour in pediatrics with multidisciplinary treatments reportedly viewed as more acceptable to parents and youth than a purely medical focus (Gorodzinski et al., 2012; Harrision *et al.,* 2019; Odell& Logan, 2013).

A variety of options crossing several levels of care and intensity exist worldwide (Harrison *et al.,* 2019; Odell & Logan, 2013; Stahlschmidt *et al.,* 2016). The primary goal of these programs is to improve youth's physical functioning and facilitate their re-engagement in age-appropriate activities (e.g., school attendance and social involvement) (Hechler *et al.,* 2015; Odell & Logan, 2013; Stahlschmidt *et al.,* 2016). Multidisciplinary teams are often composed of providers from several different specialities who work together to develop a treatment plan for the youth and their family (Harrison *et al.,* 2019: Odell & Logan, 2013).

1.4.1. Specialized Multidisciplinary Pain Rehabilitation Program Components:

Multidisciplinary interventions typically share common components, which may include medication, pain education, psychological, PT, as well as OT interventions (Caes *et al.*, 2018; Clinch & Eccleston, 2009; Harrison *et al.*, 2019). The evidence for each will be reviewed below.

Often the first form of treatment for chronic pain, pharmacological interventions are often used to treat pain directly, yet have not produced the desired effects in youth (Caes *et al.,* 2018). Most often medications are used off-label due the lack of research in the pediatric population (Mathew *et al.*, 2014). Guidelines based on expert consensus have been developed. For example, opioid use should be limited to life-limiting conditions and should not be prescribed to youth with chronic pain, as they can lead to chronification of some pain conditions (Berde *et al.*, 2012; Landry *et al.*, 2015). Non-opioid medication prescriptions have also demonstrated limited effectiveness. A recent Cochrane review concluded that there was no evidence to support the prescription and use of non-steroidal anti-inflammatory drugs in this population (Eccleston *et al.*, 2017). Local anaesthetic, anticonvulsants, antidepressants, muscle relaxants, and selective serotonin reuptake inhibitors are among those medications reported to help some, but not all youth with chronic pain (Eccleston *et al.*, 2017). Careful individualization of the pharmacological regimen should be considered and recognized as supportive but is rarely adequate as the sole component of a treatment plan (Landry *et al.*, 2015: Mathew *et al.,* 2014).

Psychoeducation plays an important intervention role providing the child and their family with an explanation between acute and chronic pain and should be a part of all comprehensive treatment plans. It focuses on underscoring the non-protective nature of chronic pain compared to acute pain and explaining how psychological interventions can effectively address pain and associated disability (Simons & Basch, 2018). More recently, pain science education has been introduced to the pediatric pain field and integrated into treatment, providing a foundation for understanding the biopsychosocial mechanisms of pain, including how the brain produces pain and that pain may be present without tissue damage (Harrison *et al*., 2019; Pas *et al.,* 2018; Robins *et al*., 2016). It is thought that this approach may prepare children and adolescents with pain-related disability for biopsychosocial treatment and has been employed with children as young as 6-12 years (Pas *et al.,* 2018). Although evidence remains scarce on the effectiveness of psychoeducation in pediatric chronic pain, initial findings are promising (Andias *et al.,* 2018).

Psychological interventions for pediatric chronic pain focus on the self-management of pain and disability, with the ultimate goal of returning to baseline functioning (Palermo, 2012). Psychological intervention components may include, but are not limited to, relaxation training, identifying and addressing negative cognitions, acceptance and value-based activities, behavioural exposure and parent coaching (Harrison *et al.*, 2019). Recent meta-analyses of psychological treatments for children and adolescents with chronic pain highlight that current approaches such as cognitive behavioural or acceptance and commitment therapy significantly decrease pain intensity, yet their effect on disability post treatment is small (Eccleston *et al.,* 2015; Fisher *et al.,* 2014). This suggests that psychological treatment approaches alone do not address pertinent contributors to pain-related disability in children and adolescents (Sinclair *et al.,* 2016). Rooted in the Fear Avoidance model, behavioural exposure interventions aim to improve youth's functioning by exposing them to activities they currently avoid due to fear of pain (Simons *et al.*, 2012a). Although few studies have reported outcomes using this intervention only, when coupled with other disciplines (e.g., physiotherapy) and with other approaches (e.g., acceptance and commitment therapy) improvements in many functional domains have been demonstrated (Wisksell *et al.,* 2009).

With the increased acknowledgement of their role in managing pain, maintaining and improving function (Guite *et al.,*2018a), parents are often involved in interventions. At its most basic, parents are often taught relaxation techniques and cognitive skills in conjunction with the child or through a parents-only session, with the aim of supporting the application of these strategies at home (Palermo, 2012). A focus on shifting parent attention and behaviour towards encouraging function, coaching and promoting coping in the presence of pain has been found to improve parents' behaviours (Eccleston *et al.,* 2015), but not the child's outcomes (Law *et al.,* 2014). Parental emotional distress related to pediatric chronic pain and the role of interventions in improving parental mental health (Law *et al.,* 2017b) has resulted in the development of interventions targeting the parents. These interventions aim to support parents in managing their own distress, promote their resiliency, thus reducing caregiver burden (Guite *et al*., 2018b; Law *et al.,* 2017a; Sieberg *et al.,* 2017). These parent-only group sessions have incorporated pain education, parent-adolescent communication, parenting, coping, problem-solving and behaviour management skills (Guite *et al*., 2018b; Law *et al.,* 2017a; Sieberg *et al.,* 2018). Initial feasibility and acceptability findings were supportive of these interventions and have demonstrated initial benefits in decreasing parent distress and caregiver burden (Guite *et al*., 2018b; Law *et al.,* 2017b; Pielech *et al.,*2018; Sieberg *et al.,* 2018).

Specialized rehabilitation interventions for youth with chronic pain also include PT and OT. These interventions focus on improving physical function by engaging children in a graded manner to previously avoided activities, using a pain self-management approach (Celedron *et al.,* 2014). The key reported objectives for physiotherapists (PTs) are to foster the adoption of a regular exercise routine, encourage movement exposure, despite the presence of pain, and educate families on pain, exercise and activity, addressing any associated misconceptions (Ayling Campos *et al.,* 2010: Eccleston & Eccleston, 2004). Specific PT interventions shift away from passive modalities targeting pain reduction (e.g., massage, electrical modalities), to more active interventions with a focus on improving strength, flexibility, joint stability, tolerance to weight bearing, exercise endurance, coordination, balance, proprioception, posture and movement biomechanics (Ayling Campos *et al.,* 2010: Eccleston & Eccleston, 2004; Revivo *et al.,* 2019). Although early evidence suggested that for some pain conditions, individualized intensive physiotherapy is considered best (e.g., complex regional pain syndrome), a recent review concluded that the stand-alone value of physiotherapy (as a mono disciplinary treatment) is difficult to determine, as it is typically prescribed in conjunction with psychological and medical interventions (Biolocerkowski & Daly, 2012). In addition, an important lack of detail emerges associated with the prescription of PT, the modalities used, and the frequency, intensity, and duration of treatment, when contrasted to medical and psychological interventions (Biolocerkowski & Daly, 2012).

Exercise is the most frequently identified physiotherapy modality (Biolocerkowski & Daly, 2012), and is recognized as a crucial component of rehabilitation interventions for youth with pain-related disability (Clinch & Eccleston, 2009; Logan *et al.,* 2010: Sherry *et al.,* 1999). These activities are typically related to functional strengthening tasks such as jumping, running, climbing stairs, coordination and balance skill, and other age-appropriate physical education (e.g., shuttle run, push-up, sit-ups) and sporting drills (Sherry *et al.,* 1999). Variations in training settings (e.g., gym, pool, soccer field) is important for generalizing skills, reducing fear avoidance behaviours and site-specific exercise behaviours

(Eccleston *et al.,* 2006). Furthermore, when increasing physical activity, it is important that a behavioural management approach be adopted, using gradual exposure and activity pacing when increasing the intensity and the exercise tolerance in these youth (Eccleston & Eccleston, 2004; Hechler et al., 2015). A recent systematic review suggests that exercise may be associated with pain reduction in this population (Kichline & Cushing, 2019).

Although not included in all multidisciplinary programs, many authors agree that OT is a vital rehabilitation component (Odell & Logan, 2013). Differing from PT, OT interventions aim to maximize independence and participation in age-appropriate daily activities (e.g., bathing, personal hygiene), fulfilling academics (e.g., learning, writing), and family roles (e.g., chores), and re-engaging in meaningful recreation and leisure activities (Celedron *et al.,* 2014; Odell & Logan, 2013). According to a recent scoping review on the OT contribution in adult chronic pain (Lagueux *et al.,* 2018), OT interventions can be classified based on the person (e.g., youth with chronic pain), occupation (e.g., student), and environment (i.e., school) framework developed by McColl & Law, 2013. Person focused interventions (i.e., skill development, and education) may include psychoeducation, energy conservations and pacing techniques, desensitization and sensory re-education activities; those focused on the environment (i.e., support provision and enhancement) may be comprised of ergonomic assessments and contextual modifications; while methods targeting occupation (i.e., occupational development) may consist of vocational intervention, sleep hygiene, pacing and activity grading. Although this classification has yet to be used in pediatrics, many OT interventions listed in multidisciplinary program descriptions (e.g. developing a schedule; desensitization), mirror those identified in the aforementioned scoping review. Unfortunately, like PT interventions, OT treatments are also poorly described in the pediatric chronic pain literature (Biolocerski & Daly, 2012).

1.4.2. Multimodal versus Intensive Interdisciplinary Treatment Options: Specialized multidisciplinary pain rehabilitation can generally be classified into two types of programs: MMT and IIPT. Although treatment frequency may vary (e.g. 1 to 3 times per weeks), MMT is typically delivered in the outpatient ambulatory care setting and consists of some combination of medical (e.g. medication, nerve blocks), physical (e.g. physical and occupational therapy), and psychological treatment interventions (Simons et al., 2013). More specifically, MMT generally involves initiating or modifying medication regimens, increasing activity tolerance through physiotherapy and exercise routines, and learning new coping strategies through cognitive behavioural approaches (Simons et al., 2010a). Furthermore, most MMT also involve the implementation of home recommendations, and provide re-evaluation and adjustments as needed (Hechler et al., 2011; Lee et al., 2002; Simons et al., 2013). Most commonly MMT interventions are not integrated, communication among providers is limited, and goals for each provider are separate (Gatchel *et al.*, 2014). Aligned with multidisciplinary pain clinics (Harrison *et al.,* 2019), these interventions rely heavily on behaviour change (Simons et al., 2010a) and are thought to be sufficient for patients with a moderate level of functional and emotional impairment (Claar *et al.,* 2013; Hechler *et al.*, 2011; Hechler *et al.*, 2014; Simons *et al.*, 2013; Soee *et al.,* 2012; Wager *et al.*, 2014). On average, multidisciplinary pain clinic treatment services cost approximately $2,200 USD per patient per year (Mahrer *et al.*, 2018). Evidence does however suggest that adherence to MMT recommendations may be suboptimal particularly in relation to medication changes, if new treatments are initiated, or when psychological interventions are added (Ho *et al.*, 2008; Simons *et al.,* 2010a).

IIPT may be delivered in either day hospital, inpatient, or in a combination of both settings (Hechler *et al.,* 2015; Liossi *et al.*, 2019). To be considered an IIPT, the program team must be comprised of three or more disciplines, housed in the same facility (e.g., pain specialist, psychologist, physiotherapist), and working in an integrated manner (Dobe & Zernikow, 2013; Harrison *et al.*, 2019; Hechler *et al.,* 2015). A shared rehabilitation philosophy, common restorative functional goals, daily communication and active youth involvement in all program components (e.g., PT, OT, and psychological interventions) are recognized key ingredients in this treatment (Gatchel *et al.,* 2014; Harrison *et al.,* 2019*;* Hechler *et al.,* 2015; Odell & Logan, 2013). The inclusion of parent and school reintegration are also strongly recommended components yet are not incorporated in every program (American Pain Society-Pediatric Pain Task Force, 2012; Stahlschmidt *et al.,* 2016). IIPTs are acknowledged to be time-consuming, resource-intensive, costly (>$30,000 USD/patient) (Evans *et al.*, 2016; Simons *et al.*, 2018).

Despite some commonalities, variability across IIPT structures, organization and frequency of treatment provided by each discipline exists (Stahlschmidt *et al.,* 2016; Odell & Logan, 2013). The amount of each therapeutic component may also differ between program and among patients. For example, the inpatient program at the German Paediatric Pain Center, consists of 6 treatment modules including psychoeducation, pain coping, cognitive intervention to target emotional distress, family therapy, physiotherapy, and psychological functioning (Dobe & Zernikow, 2013). These inpatient programs are focused on medical, psychological and social treatment methods (Stahlschmidt *et al.,* 2016). By contrast, the Mayo Family Pediatric Pain Rehabilitation Center day-hospital program at Boston Children's Hospital adopts a functional rehabilitation approach. Keeping these children and adolescents out of the inpatient setting assists in emphasizing function, de-emphasizing the "sick role" with these youth and their families, while considerably reducing program costs (Logan *et al.,* 2010). Furthermore, this setting fosters a large dose of intense physical activity (i.e. 3-hours of PT and 1-hour of OT) and associated home programming (Logan *et al.,* 2010; Odell & Logan, 2013).

Other differences between IIPT programs include the length of stay, admission criteria and process. Some programs have a fixed 3-week treatment limit (Hechler *et al.,* 2015), while others have a flexible length of stay, established based on individual youth and family needs and progress (Harrison *et al.,* 2019; Odell & Logan, 2013; Stahlschmidt *et al.,* 2016). Several IIPT programs offer both inpatient and day hospital program with youth triaged to the level that best meets their needs (Celedron *et al.,* 2014; DeBlécourt *et al.,* 2008; Roessler *et al.*, 2015). A combination of inpatient and day hospital also exists (Banez *et al.,*2014).

While admission criteria have been found to be fairly consistent, factors for exclusion vary widely between IIPT worldwide. Inclusion criteria include persistent pain of a high intensity for at least three months (Eccleston *et al.,* 2003; Hechler *et al.,* 2010b; Weiss *et al.,* 2013), and youth and/or parent report of severe limitations of daily activities (e.g. school attendance, participation in sport, recreation or leisure activities) (Eccleston *et al.,* 2003; Gaunlett-Gilbert *et al.,* 2013; Hechler *et al.,* 2010b; Logan *et al.,* 2010; Maynard *et al.,* 2010; Weiss *et al.,* 2013). Although most programs treat all types of pain, some target specific conditions (e.g., CRPS) while exclude others (e.g., headache) (Odell & Logan, 2013; Stahlschmidt *et al.*, 2016). Youth and their parents' motivation and willingness to comply with prescribed treatment are also viewed as important admission criteria (Eccleston *et al.,* 2003; Hechler *et al.,* 2010b; Logan *et al.,* 2010). Youth with specific psychiatric needs requiring targeted treatment are often excluded (Eccleston *et al.,* 2003; Logan *et al.,* 2010; Maynard *et al.,* 2010). Youth for whom medical assessment is not yet complete, or those who have a recognized medical pathology are often not admissible (e.g., malignant cause) (Eccleston *et al.,* 2003; Gaunlett-Gilbert *et al.,* 2013; Logan *et al.,* 2010; Maynard *et al.,* 2010; Weiss *et al.,* 2013).

A host of clinical factors (e.g., cohort timing, resource availability) and family-specific factors (e.g., place of residence, loss of parental work time) can influence the type of treatment recommended and selected for each youth. Clinical recommendations for either IIPT or MMT are typically provided to a family following a comprehensive pain assessment, which includes the evaluation of a current pain situation and the patient's psychological well-being, conducted by a multidisciplinary team (Wager *et al.*, 2014). Youth highly disabled by their pain are typically ushered to IIPT (Hechler *et al.*, 2009), while those whose function and participation are less impacted are guided to MMT (Hechler *et al.*, 2011). However, variations in these clinical practices and guidelines exist and the appropriateness of clinical recommendations for various levels of pain-related disability severity have been questioned. Hechler and colleagues (2011) reported that treatment recommendations were adequate for the level of disability of those presenting in their studied clinical population. On the other hand, Wager and colleagues (2014) highlighted that clinical recommendations did not always align with youth disability needs in a sample they studied. For example, in the case of the subgroup presenting with moderate pain problems and low passive coping, which should typically respond to MMT and for which psychoeducation has been deemed effective, almost half of the study group (i.e., 47%) was recommended IIPT (Wager *et al.*, 2014).

These findings suggest that treatment decision may be influenced by other factors (Simons *et al.*, 2013; Wager *et al.*, 2013). Health insurance coverage in some jurisdictions (Simons *et al.*, 2013) limited availability of IIPT for some families. Travel distances, additional incurred costs (e.g., hotels and meals), and the supplementary time away from work, family and school for youth to attend the program (Peng *et al.*, 2007; Simons *et al.*, 2013) may also affect the clinical team recommendations and family's treatment decisions.

To conclude, there is a sub-group of patients with severe pain-related disability whose needs are undertreated or not met at all. Matching the unique characteristics of each youth with pain-related disability and their family with the appropriate treatment, intensity and subsequent service model is imperative (Harrison *et al.*, 2019; Simons *et al.*, 2018). The rarity of evaluation studies comparing these different service models of specialized multidisciplinary pain rehabilitation has limited the available evidence upon which to base this matching.

1.5. Program Evaluation

The use of program evaluation in healthcare has seen a steadily increase (Rossi, Lispey, & Freeman, 2004). Evaluation is viewed by many healthcare organizations as a mechanism to systematically determine the effectiveness and efficiency of a program, while ensuring the delivery of safe, appropriate and high-quality care to patients (Long, 2006). In times of fiscal constraints and resource scarcity, evaluation has been valuable in assessing organisational progress, providing critical information to inform program improvement, accountability, and promote organization change (Love, 1983). Finally, evaluation is considered essential to many healthcare decision makers, as it informs the types of programs needed, allows monitoring of program implementation, documents any deviations from the intended process, and promotes evidence-informed decision making on the future continuation of some programs (Health Services Research Group, 1992). Multiple definitions of program evaluation exist. Table 1 presents published examples. A specific component of evaluation is emphasized in each definition. For example, social sciences methods are accentuated in the definition proposed by Rossi and colleagues (2004), the use of evaluation findings underscored by Patton (1997, 2008), and the learning function associated with evaluation highlighted by Preskills and Torres (1999).

Table 1.

Published Definition of Program Evaluation

Source	**Definition**
Beeby, 1977	Program evaluation is the systematic collection and interpretation of evidence, leading, as part of the process, to a judgment of value with a view to action (p. 68).
Poth *et al.*, 2014	Evaluation is the systematic assessment of the design, implementation or results of an initiative for the purpose of decision-making or learning.
Mark *et al.*, 2000	Program evaluation is the systematic inquiry that describes and explains the policies' and program's operations, effects, justifications, and social implications (p. 3).
Patton, 1997	Program evaluation is the systematic collection of information about the activities, characteristics, and outcomes of programs to make judgements about the program, improve program effectiveness, and/or informs decision about programming (p. 23).
Preskill & Torres, 1999	Evaluative inquiry is an ongoing process for investigating and understanding critical organization issues. It is an approach to learning that is fully integrated with an organization's work practices, and as such, engenders a) organization members' interest and ability to exploring critical issues using evaluation logic; b) organization members' involvement in evaluative processes, and c) the personal and professional growth of individuals within the organization (pp. 1-2).
Rossi, Lipsey & Freeman, 2004	Program evaluation is the use of social research methods to systematically investigate the effectiveness of social intervention programs. It draws on the techniques and concepts of social science disciplines and is intended to be useful in improving programs and informing social action aimed at ameliorating social problems (p. 28).
Sciven, 1991	Evaluation refers to the process of determining the merit, worth, or value of something, or the product of the process… The evaluation process normally involved some identification of relevant standards of merit, worth, or value; some investigations or the performance of evaluands on these standards; and some integration or synthesis of the results to achieve an overall evaluation or set of associated evaluation (p. 139).
Weiss, 1972	The purpose of program evaluation is to measure the effects of a program against the goals it sets out to accomplish as a means of contributing to subsequent decision making about the program and improving future programming.

World Health Organisation (WHO), 2013	An evaluation is an assessment, as systematic and impartial as possible, of an activity, project, program, strategy, policy, topic, theme, sector, operational area, institutional performance … focused on expected and achieved accomplishments, examining the results chain, processes, contextual factors and causality, in order to understand achievements or the lack thereof (p. 1).

Modified from Moreau, 2012

Regardless of the differences, all definitions imply that evaluation is systematic, planned, and includes purposeful activities that comprise a methodical collection of data, aimed at addressing a specific inquiry, with the goal of enhancing knowledge for a specific purpose (Preskill & Russ-Eft, 2005). Furthermore, many definitions emphasize the terms “judgment”, which implies a comparison of information (i.e. data gathered for the evaluation), against something else (e.g. baseline observations, control group, external standard). It is this judgment orientation that differentiates evaluation from typical healthcare research, where a judgment may or may not be rendered (Moreau, 2012).

Aligned with the evaluation context and decision makers request, this thesis will adopt the definition of program evaluation put forth by the Poth and colleagues (2014), i.e., the systematic assessment of the design and the results of the initiative for the purpose of decision-making or learning. This definition not only emphasizes the assessment of intervention outcomes, but also the evaluation of the program design. Moreover, it underscores two important purposes of program evaluation: learning and accountability.

The learning process occurs throughout the evaluation process and may provide information to improve an intervention (formative) and may be transformative, where the process is used as a driver of change, transforming a problematic or unjust situation into a beneficial one focused on the well-being of the collective (Brousselle *et al.*, 2011). Through active participation in the program evaluation, involved individuals can increase their competencies in data collection, analysis, and communication (Preskills & Torres, 1999). Furthermore, the process may improve their understanding and appreciation for contextual issues and force them to think more broadly about problems and possible solutions (Preskills & Torres, 1999). Various stakeholders may also come together as a team and collectively examine a program's issues, and through the multiple perspectives, obtain an even more diverse and complex understanding of the problems (Preskills & Torres, 1999). Finally, evaluation processes enable organization to learn, as individuals and teams share their learning with others (Preskills & Torres, 1999). In this sense the evaluation generates new knowledge and facilitates organization multi-level leaning (Moreau, 2012).

With the ongoing political and economic dialogue in health care, many public organizations engage in program evaluation for the purpose of accountability (Chelimsky, 2006). Funding scarcity in the health and social sectors has led to an increase interest and utilization of evaluation to establish program accountability. Commonly referred to as summative evaluation, evaluations conducted for this purpose, typically assess program effectiveness, best practice implementation and the fiscal validity of the program (Clarke, 2006). Effectiveness findings can then be compared to program costs and can be used in decision making related to program continuation (Love, 1983; Dummond *et al*. 2015). Three types of accountability are associated with program evaluation, each of interest to a specific stakeholder group. Focused on whether the appropriate program goals have been identified, goal accountability is a particular interest of upper-level management. Service managers and providers are responsible for process accountability, which explores the implementation and appropriateness of the procedure to reach the program goals. Finally, outcome accountability, which examines the degree to which the program goals were achieved, is also the responsibility of the manager and service providers (Alkin & Christie, 2004). Similar to many public sectors setting, in health, the focus is primarily on outcome accountability.

Limited program evaluation literature exists in the pediatric rehabilitation sector, including in specialized pain rehabilitation. Furthermore, few researchers have reflected on the purpose of program evaluation in the rehabilitation sector (Schnelker & Rumrill, 2001). Of those who have, accountability, fiscal constraints, and accreditation standards have been cited as the principal drivers of evaluation growth in both the institutional and community rehabilitation contexts (Schnelker & Rumrill, 2001). Moreover, funding agencies (i.e., public and private), accreditation and professional regulatory bodies, and patient and family advocacy groups have placed high performance expectations on rehabilitation organizations and the clinicians working within them. Evidence would therefore suggest a strong focus on evaluations for the purpose of accountability in healthcare and rehabilitation settings.

1.5.1. Types of Evaluation: Evaluation approaches and methodologies vary considerably depending on the type evaluation questions, on the type of intervention and as a function of their complexity. An intervention may be subjected to two types of evaluation, either intervention monitoring or evaluation research. Intervention monitoring seeks to examine the intervention components as a function of criteria, norms and standards, and to assess the consistency and conformity of the components in relations to these references (Brousselle *et al.*, 2011). Evaluative research, on the other hand, uses scientific inquiry to analyze the causal relationships between the intervention components, and better understand the how and why surrounding the outcomes produced (Brousselle *et al.*, 2011). Therefore, while evaluation research is considered a research activity, intervention monitoring is not.

Evaluative research can be divided into six different types of analyses, each with its own research methodology. Table 2 presents the different types of analyses, their objectives, and the associated questions they aim to answer.

Table 2.

Evaluative Research Analyses

Types	Objectives	Evaluation Questions
Strategic analysis	To gain an appreciation of the relevance of an intervention	Is it relevant to intervene on this problem, in particular in relation to importance of other issues and the potential impact of the intervention? (technical and social feasibility) Is it relevant to intervene in this way? Are the factors chosen upon which intervening, the most relevant? Is it relevant to target this population? Are the objectives sufficient to resolve the problematic situation and respond to the needs? Is it relevant for those responsible for the intervention to act as they are given their position and role?

Logic analysis	To examine the coherence (i.e., the design and operational validity) of an intervention	Is there a sufficient theoretical foundation to the intervention? Are the quality and quantity of the resources sufficient? Are these resources well organized?
Production analysis	To study the resources utilized and the volume and quality of product (activities) produced by an intervention	What is the quality of the services produced? • Is it possible with the same resources to produce more services or services of higher quality? • Is it possible to use fewer resources to produce the same amount and quality of services?
Effect analysis	To explore the effectiveness of an intervention	What are the observed effects? Are the effects attributable to the intervention or to other factors?
Efficiency analysis	To assess the overall efficiency of an intervention	Is it possible to achieve better outcomes at an equivalent or lesser cost or to achieve the same outcomes at a lesser cost?
Implementation analysis	To observe the interactions between the intervention and the context in which it is implemented	How can the transformation of intervention across time be explained? What is the influence of the context on implementation of the program? In what way did the variations in the implementation influence the observed effects? What is each program components' contribution to producing the effects? What is the influence of the interactions between the implementation context and the intervention on the observed effects?

Created from Brousselle *et al.*, 2011

Effect analysis (or effectiveness evaluation) is the most utilized and requested of these evaluation analyses. It consists of exploring the causal relationship between the observed effects and the intervention and seeks to qualify this effectiveness as a function of the identified research context and the adopted methodology (Brousselle *et al.,* 2011). Effect analysis is interested in capturing all the possible outcomes (i.e., positive and negative) resulting from an intervention (Brousselle *et al.,* 2011).

An array of methods can be used to analyze the effects of an intervention. Design choice is dependent on the purpose of the evaluation, the degree of control available to the researchers on the intervention's delivery and should be based on the principle of minimizing potential bias (i.e. internal validity), while maximizing generalization (i.e., external validity) (Eccles *et al.*, 2003). To generate minimal biased estimates of the intervention effects, randomized control trials (RCTs) are typically viewed as the "gold" standard (Shadish *et al.*, 2002). Their design compares outcomes measured in prospective follow-ups between those randomly allocated to receive an intervention and the control group randomly allocated to receive the comparison condition, generally a currently accepted standard of care (Bonell *et al.*, 2011). The strength of RCTs stems from their capacity to ensure a reasonably just comparison between intervention and control groups, which could be expected to experience comparable outcomes in the absence of intervention (Bonell *et al.*, 2011). This is accomplished by ensuring that intervention and control groups are not systematically different from each other in terms of measured and unmeasured characteristics (Bonell *et al.*, 2012) To do so, RCTs typically randomly allocate individuals or groups of individuals (e.g., from a same school or community) (Eccles *et al.*, 2003).

Random allocation is generally regarded as ethical practice if there is uncertainty about whether intervention confers significant benefits (Bonell *et al.,* 2012). However, considerable challenges exist in conducting RCTs to evaluate the impacts of complex interventions, such as rehabilitation interventions (Bonell *et al.*, 2011; Bonell *et al.*, 2012). They can be logistically difficult to conduct, are time consuming, and can be expensive (Eccles *et al.*, 2003). Stakeholders, such as health providers, may resist RCTs, believing that random intervention allocation unjustifiably denies some of the expected benefits of an intervention, even if those benefits have yet to be demonstrated rigorously through evaluation (Bonell *et al.*, 2011; Bonell *et al.*, 2012). Others, for example patients, may oppose randomization due to treatment preferences or ideological beliefs and values (Bonell *et al.*, 2011). Criticism also arises on their tight inclusion criteria and constraints, which can limit the generalizability of the findings (Eccles *et al.*, 2003). Furthermore, RCT findings associated with complex interventions can be diluted in some cases by 'contamination' effects, if for example participants assigned to a control group participate in intervention group activities or other services similar to those of the intervention under study (Bonell *et al.*, 2012). Moreover, double blinding of the complex rehabilitation intervention (where neither the provider nor patient is aware of the patient allocation) is challenging and often impractical. Thus, information bias in RCTs is more likely. Despite these limitations, RCTs are still viewed as providing the strongest evidence about the causal effects of rehabilitation interventions. However, RCTs have been designed primarily to determine whether or not a specific intervention is effective and have been strongly focused on the internal validity of the trial, addressing the question of efficacy rather than broader questions of reach, effectiveness, adoption, implementation and maintenance (Glasgow et al., 2006).

It has been proposed that randomized trials should only be considered when there is a genuine uncertainty about the effectiveness of the intervention (Eccles *et al.*, 2002). In other cases, quasi-experimental designs where comparison groups are formed based on methods other than randomization, such as 'natural experiments' or statistical matching techniques may be more appropriate (Bonell et al., 2011; Craig et al., 2008). Commonly quasi-experimental designs are used to assess the effects of an intervention on patient outcomes under the usual care conditions (Glasgow et al., 2006). This "real world" evidence is of increasing importance to decision makers (Canadian Agency for Drugs and Technology in Health (CADTH), 2006). "Real-world evidence" is recognized as data derived from clinical practice among heterogeneous sets of patients in real life health practice settings (Liossi *et al.*, 2019). This evidence is increasingly seen as a way to tailor health care decision making more closely to the characteristics of individual patients, and thus as a step toward making healthcare more personalized and effective (Yuan *et al.*, 2018).

Studies of intervention using non-randomized design are inevitably subject to a range of biases that are not present or are less prominent in randomized control trials. More specifically, non-randomized study designs cannot control for unmeasured or imperfectly measured confounders (Bonell *et al.*, 20011; Eccles *et al.*, 2003). Inadequate reporting of the impact of potential confounders can be a deficit in intervention evaluations. Comprehensive matching or adjustment on all potentially important confounders is often challenging, especially when evaluations are relying on routinely collected data from intervention and control groups (Bonell *et al.*, 2011). Confounding can lead to underestimates of effects (e.g., when the greater needs of those receiving the intervention are not sufficiently considered) or, more commonly, lead to overestimates of effects (e.g., when intervention recipients' lesser needs or greater uptake of the intervention are insufficiently considered) (Bonell *et al.*, 2011). Statistical analytical techniques can, however, be used to minimize confounding variables. In the absence of a control group, 'before and after studies' may be possible. However, these studies are vulnerable to confounding due to maturational trends and concurrent influential events, with the extent of their influence being dependent on context (Bonell *et al.*, 20011). Incomparability of data collected before (e.g., routinely gathered data) and after an intervention (e.g., specific and different evaluation surveys) can also create issues (Bonell *et al.*, 2011). Furthermore, 'regression to the mean' can occur when participants are selected at baseline for their increased risk which then returns to a less extreme level regardless of intervention.

Finally, contribution analysis is an alternative research design when a comparator cannot be found and for complex interventions with multiple and diffuse effects, or when an RCT or a quasi-randomized experiment is not feasible (Brousselle & Champagne, 2011, Mayne, 2012). Contribution analysis is useful in situations where the program is not experimental and there is little to no room for varying how it was implemented (Mayne, 2008). For example, most public healthcare programs are funded under the assumption that they will produce desired outcomes, based on some accepted or believed theory of change. The theory of change will set out to render explicit the postulated results chain showing how and why a specific series of activities or processes will result in a sequence of outcomes leading to the anticipated results (Mayne, 2011). Contribution analysis goes two steps further to not only elaborate the theory of change, but to examine and test this theory against logic and available evidence of the results observed (Mayne, 2012). Furthermore, it also considers the various assumptions behind the theory of change and examines other potential influencing factors

(Mayne, 2012). Causality is inferred, based on the following: 1) availability of a reasoned theory of change and the assumptions behind it; 2) the implementation of program activities as outlined by the theory; 3) verification of the theory by empirical evidence; and 4) assessment of the influential program factors for their contribution. This analysis results in a confirmation or verification of the postulated theory of change, or in the suggested revisions to the theory when the theory is not quite correct (Mayne, 2012). The aim of contribution analysis is to reduce the uncertainty the contribution an intervention is making to the results by increasing the understanding of why results did or did not occur (Mayne, 2012). Moreover, the roles played by the intervention and those of other influencing factors are recognized (Mayne, 2012). Contribution analysis argues that if the theory of change can be verified or confirmed by empirical evidence and can account for the major external influential factors, then reasonable conclusion about the difference associated to the intervention can be drawn (Mayne, 2012). The term contribution is thus used deliberately, as the analysis puts forward a claim based on whether the intervention has made the expected difference in light of the multiple influencing factors (Mayne, 2012). Contribution analysis recognized the importance of a well-thought-out conceptualization of a program's theory of change that is embedded in the intervention context, incorporating the perspectives of key stakeholders, intervention users, and existing relevant research (Mayne, 2012). Unfortunately, to date, the implementation and use of contribution analysis have been scarce in the field of health care and absent in rehabilitation (Mayne, 2012).

1.5.2. Conceptualizing and Theorizing the Intervention: An intervention may be conceptualized as an organized system of actions which, in a given context at a specific time, aims to modify the predetermined course of a phenomenon to correct a problematic situation (Brousselle *et al.*, 2011). In all organized action systems, five components can be found: a structure, individual stakeholders and their collective practices, action processes, one or many ends or purposes, and the context (Brousselle *et al.*, 2011). The structure is composed of three interdependent dimensions: 1) the physical dimension, which includes the various intervention resources (e.g., human resources, financial, space, materials); 2) the organizational dimension which is composed of the policies, rules and regulations that determine the distribution of the resources; and 3) the symbolic dimension, which is comprised of the collective beliefs, representations and values, which then allows communication to occur between the various stakeholders vested in the intervention, giving meaning to their actions (Brousselle *et al.*, 2011).

The intervention stakeholders are characterized by their projects, their perceptions and belief about the world around them, their convictions, their resources, and their disposition to act (Brousselle *et al.*, 2011). They interact in both a cooperative and competitive manner to increase their control over critical resources within the action system (Brousselle *et al.*, 2011). The action system is the collection of processes by which the resources are mobilized and used by the stakeholders to produce the goods and services required to achieve the intervention ends (Brousselle *et al.*, 2011). The intervention ends are its objectives and aim to alter one or many phenomena by acting over time on certain causes and determinants to correct a problematic situation. Finally, the intervention context constitutes its physical, judiciary, symbolic, historical, political, economic, and social environment, in which the intervention operates, along with all other action systems with which it intersects (Brousselle *et al.*, 2011, Chen 2014). According to this definition, there is a wide array of interventions in the healthcare system (e.g., medication, care pathways, services, organizations, and even the healthcare system itself) (Brousselle e*t al.,* 2011).

The conceptualization of a specific intervention can be represented by a program theory and illustrated by a logic model. Program theory is defined as the specific actions required to achieve the anticipated effects from the other impacts that could be expected, and the mechanisms by which these effects are produced (Chen, 2014). It is used to define a set of assumptions of the way by which a program is linked to its expected benefits, and the strategy and tactics adopted to achieve its goals and objective (Rossi *et al.*, 1999). A logic model is a visual or graphic representation of these relationships (Rossi *et al.*, 1999). It has been used to guide evaluation (Claphand *et al.*, 2018; Stewart *et al.*, 2014), to identify and explore variations in program implementation in different contexts (Claphand *et al.*, 2018; Zivanni *et al.*, 2011), and to better understanding of the complex and synergistic change processes that occur within an intervention (Livingood *et al.*, 2007). However, for logic models to be useful, program stakeholders must be engaged in their development (Chen, 2014). Stakeholder engagement promotes understanding and agreement among vested parties on program outcomes and its components, thus enhancing buy-in (Chen, 2014).

Unfortunately, in complex interventions, such as specialized pediatric rehabilitation pain intervention, the predominant study focus has been on identifying which interventions worked (Craig et al., 2008). Although the quality of the evidence of these interventions may be highly rated, interventions are often poorly theorized, leading to poorly understood effects, thus hindering a broad real-world implementation. At their core, these interventions are comprised of multiple synergistic components, which when interacting together generate their effect (Medical Research Council, 2000). Furthermore, their effectiveness is also a function of the components' interaction with the context in which they operate (Bonell *et al.,* 2006). This requires both the intervention theory and its contextual interaction to be incorporated into the program theory (Weiss, 1995). The lack of such details may impact the capacity for such interventions to be implemented into an alternative context or may impact the effects generated in different settings (Bonell *et al.,* 2012). RCTs and other study designs rarely sufficiently acknowledge these details.

To better understand the effects of complex intervention, like specialized pediatric pain rehabilitation, theory validation should receive the same emphasis as the study trial design (Bonell *et al.*, 2012). As a means of triangulation, often used in RCTs, the exploration of the plausibility of the causal pathways becomes even more important in quasi-experimental studies. However, such examination requires the intervention to be explicitly theorized and conceptualized (Rychetnik *et al*., 2002).

Logic analysis, a relatively new theory-based evaluation which provides a means of testing the plausibility of a program theory prior to engaging extensively into the evaluation process, not only maps the program's theory but also questions its plausibility based on existing evidence (Brousselle & Champagne, 2011). As a clarification point, logic analysis should not be confused with logic modelling (as previously described). Logic analysis can be useful in providing important insights into the validity of the program theory, in engaging stakeholders in valuable reflections (Page *et al.*, 2007), in improving the understanding of an intervention’s strengths and weaknesses, and in analyzing whether the intervention is designed in such way that it can logically produce the desired results (Champagne et al., 2009). Moreover, it allows for the assessment of strength of the causal links between the intervention and the intended effects, a prerequisite for conducting an effect analysis (or effectiveness evaluation) (Brousselle & Champagne, 2011). Logic analysis can therefore be quite helpful in choosing the appropriate evaluation, best suited for the intervention and its contextual factors (Brousselle & Champagne, 2011). The approach uses scientific knowledge or expert opinion to evaluate the credibility of the intervention theory (direct logic analysis) or in assisting in recognizing promising alternatives to attaining anticipated effects (reverse logic analysis) (Brousselle & Champagne, 2011). Logic analysis evaluation has been used in some health fields, mainly in public health and the healthcare, but to our knowledge, has yet to be applied to pediatric specialized pain rehabilitation intervention evaluation. Engaging stakeholders in logic analysis is crucial in developing a program theory and logic model which consider to be an accurate representation of the intervention, and vital in promoting utilization of the evaluation findings among this group (Tremblay *et al.*, 2013).

1.5.3 Participatory Evaluation: Rather than a specific methodology, participatory evaluation (PE) is an approach designed to engage individuals from a chosen community who have a vested interest in the program under evaluation, known as stakeholders, in the evaluation research process (Cousins & Whitmore, 1998; Cousins & Chouinard, 2012). By working as a team, different knowledge and skills are shared among various non-evaluators/research stakeholders and the evaluator in the evaluation process. While evaluator applies knowledge of logic, methods and an understanding of professional standards to the evaluation process, program stakeholders possess the deep, implicit, comprehension of what the program is expected to do, and how it aims to do it (i.e., program logic). Furthermore, these stakeholders possess an intimate familiarity with the program context (Moreau, 2012).

Cousins and Whitmore (1998) define PE as an evaluation in which evaluators work in partnership with stakeholders to produce evaluative knowledge. Centred on the process dimension, this definition recognizes both the normative development work and the pragmatic of PE’s foundation (Cousins & Chouinard, 2012). Several forms of PE research have emerged and can be distinguished by the level and nature of stakeholder involvement, their rationales, and their ideological penchants (Cousins & Chouinard, 2012). Three principal participatory evaluation streams are recognized: 1) practical participatory evaluation (P-PE); 2) transformative participatory evaluation (T-PE), and 3) stakeholder-based evaluation (SBE) (Cousins &Whitmore, 1998; Weaver & Cousins, 2004).

1.5.3.1. Practical Participatory Evaluation: P-PE central function is to foster the use of evaluation findings and holds as its core the assumption that evaluation is geared towards program, policy and organizational decision making (Cousins & Whitmore, 1998). It has a pragmatic orientation and is based on the empirical observation that stakeholder involvement in the evaluation process enhances its relevance, ownership, and the utilization of its findings (Cousins & Whitmore, 1998; Patton 2008). More specifically, stakeholder involvement renders the evaluation more responsive to the users' needs (Cousins & Whitmore, 1998). The evaluation process and its findings are also more likely to support decision making, with the benefit extending beyond the program itself, into fulfilling education, learning, and change management purposes in the broader organization (Cousins & Whitmore, 1998; Weaver & Cousins, 2005). In P-PE, decision-making is commonly shared between the evaluator and the stakeholders (Weaver & Cousins, 2005). Diverse participation is often limited, as stakeholders are typically primary program users, those vested in the program, or those who are in positions to influence change. (Waever & Cousins, 2005). Participants are involved extensively in a wide variety of inquiry tasks (Weaver & Cousins, 2005). This stakeholder involvement can lead to the acquisition of knowledge and skill associated with systematic inquiry, an appreciation for evaluation, and enhance stakeholders' feelings of self-worth and empowerment (Cousins & Whitmore, 1998).

1.5.3.2. Transformative Participatory Evaluation (T-PE): Based on a more radical ideology than P-PE, T-PE emerged primarily in the developing world as a reaction to positivism models of inquiry, viewed as exploitive and detached from the more pressing social and economic issues of the early 1970s (Cousins & Chouinard, 2012; Cousins & Whitmore, 1998; Weaver & Cousins, 2005). Relating closely to participatory research, participatory action research, community-based research, and participatory rural appraisal, T-PE employs participatory principles and the inquiry process to leverage social change (Cousins & Whitmore, 1998; Cousins & Chouinard, 2012). Framed in the context of power and transformation, T-PE is deeply rooted in community and international development, adult education and the feminist movement (Cousins & Whitmore, 1998). The inclusion of stakeholders is viewed as an emancipatory and liberating process, with the potential of destabilizing the status quo of knowledge production, control, use and ownership of the evaluation process (Cousins & Whitmore, 1998). Through the participatory process, stakeholders acquire a rich understanding of the oppressive forces inherent to the local context and develop their self-determination capacity (Weaver & Cousins, 2005). In T-PE, decision making control is balanced between the evaluator and the stakeholders, with the evaluator adopting more of a facilitator role, once stakeholders have been educated in evaluation logic and inquiry methods (Weaver & Cousins, 2005). A high degree of stakeholder diversity is expected (Weaver & Cousins, 20015). However, given the broad range of diverse perspectives, some level of conflict, logistical challenges, and feasibility questioning should be anticipated (Weaver & Cousins, 2005). As with P-PE, stakeholders should be implicated in a wide range of inquiry tasks, involvement deemed important to the capacity building and empowering tenets of T-PE (Weaver & Cousins, 2005). However, this may or may not be feasible depending largely on stakeholders' expertise and education (Cousins & Chouinard, 2012).

1.5.3.3. Stakeholder-based Evaluation (SBE): SBE was introduced in recognition of an existence of a value conflict arising between stakeholders and a program purpose and goals (Cousins & Chouinard, 2012). Responsive to local context, this evaluation seeks to understand the issues arising from a variety of perspectives (Cousins & Chouinard, 2012; Weaver & Cousins, 2005). Although SBE has goals are similar to P-PE, it is best suited for circumstances where agreement surrounding program goals is lacking among stakeholders (Cousins & Whitmore, 1998). Typically, in SBE processes, the evaluator remains firmly in control of the evaluation and its implementation, while stakeholders play more of a consultative role on debating issues needing resolution and later on the interpretation of the evaluation findings (Cousins & Chouinard, 2012; Weaver & Cousins, 2005). By involving all legitimate groups of stakeholders in the process, the evaluator is able to work on building agreement between those subscribing to different value positions, while retaining a predominately neutral stance (Cousins & Whitmore, 1998). As a result of this consensus building, the evaluation process can become more useful to a broader audience than if only one group were included.

1.5.3.4. Advantages and challenges of participatory evaluation (PE): Like all evaluation approaches, PE espouses advantages and disadvantages. However, in the case of PE, advantages and disadvantages are often situational dependent. The advantages and disadvantages in relation external validity of the evaluation, utilization of the evaluation findings, collaborative stakeholder engagement, the cost of participation and the expected benefit to society will be described below.

PE offers greater external validity to an evaluation. A diversity of perspectives and opinions are expressed by multiple stakeholders engaged in the evaluation, broader relevance is assumed, as is the generalizability of the findings (Patton, 2004). Appropriate use of evaluation methods increases the creditability of the process and further extends the assurance of external validity. However, the involvement of all relevant stakeholders is often not feasible, and a selection process of those interested in participating must ensue (Moreau, 2012). Unfortunately, limited evidence exists to guide this selection procedure (Daigneault & Jacob, 2009). As a result, evaluators often placed in a difficult position when evaluation sponsors exerting intentional or unintentional influence over the evaluation process (Chelimsky, 2008). Furthermore, Plottu and Plottu (2011) suggested that the involvement of program stakeholders, particularly those who benefit either directly or indirectly from the program, may negatively affect the evaluation external validity. This can occur in circumstances where the selected stakeholders only have a partial vision of the program, or their evaluation skills are limited (Plottu & Plottu, 2011).

Generalizability of the findings must also be cautioned, as the evaluation findings are context-bound, i.e., dependent on the specific environmental conditions under which the evaluation is conducted (International Collaboration for Participatory Health Research, 2013). To address these deficiencies, stakeholder selection must aim to be representative and holistic vision of the program (Cargo & Mercer, 2007; Cousins & Chouinard, 2012). The provision of stakeholder training to develop their evaluation skills has been proposed as a means of ensuring evaluation rigour (Brisson, 2007; Mantoura & Potvin, 2012). Finally, careful attention must also be given to providing a meticulous description of the evaluation context and the participant characteristics (including their position in relation to the evaluation) allowing those utilizing the findings to make their own judgments on the transferability of the results (Mantoura & Potvin, 2012).

Evidence surrounding collaborative stakeholder engagement suggests that stakeholders, who have participated in the evaluation, gain a better understanding of the findings, are more likely to integrate these findings into their practice (Cargo & Mercer, 2007), and to adhere to the recommendations (Patton, 2008). Conversely, however, stakeholder participation can also negatively impact finding utilization if a lack of evaluation skill and expertise result in the collection of poor data, analysis, results and conclusion quality (Plottu & Plottu, 2011). The oscillation of control over technical decision-making (e.g., design, data analysis) between the evaluator and the stakeholders should carefully assessed and negotiated to ensure evaluation quality is preserved (Brisson, 2007).

Stakeholder participation in the evaluation reportedly increased costs, effort but also benefit. Through a dialectical process, participatory evaluation renders the evaluative process a collaborative activity (Cousins & Chouinard, 2012). The sharing of values, beliefs, opinions, needs, and differences, the achievement of compromises, and the documentation of non-negotiable positions provide stakeholders with the opportunity to collectively define a problem and offer a chance for collaborative solutions to emerge. It may also lead to a deeper understanding of their motives and drivers. On the other hand, participation can be a barrier to public action and a promoter of the status quo, in particular if stakeholders belong to a representative militant group with longstanding views on a topic and use their position to block changes and stall progress (Plotto & Plottu, 2011). Furthermore, should conflicting perspectives become pervasive and not well managed, a paradoxical situation may arise whereby the vulnerable are silenced and disempowered (Plotto & Plotto, 2011). Because conflicts and tensions are part of all partnerships, effective group processes including operating norms, principles and organisational structure should be put in place (Cargo & Mercer, 2007). Moreover, an appropriate amount of time should be dedicated to the understanding of the context, the stakeholders, their culture and their priorities, along with building reciprocal relationships and developing equitable participation expectations and conflict resolution processes (Cargo & Mercer, 2007; Domecq et al., 2014). These expectations and processes become extremely important when populations whose voices have been excluded from these discussions, such as youth with pain-related disability and their parents, are incorporated. In addition to time, additional core resources and money are typically required, yet frequently underestimated (Domeq *et al.,* 2014). Participants can become frustrated and disengaged due the lengthy process. Realistic timelines should be shared with potential stakeholders to favour a shared understanding of the process (Cornwall & Jewkes, 1995). These challenges may explain why, despite the growing interest in participatory approaches, PE has rarely been applied in the study of pediatric chronic pain.

1.5.4. Program Evaluation of Specialized Pain Rehabilitation Interventions

Similar to other areas of pediatric rehabilitation (Moreau & Cousins, 2011), effectiveness evaluations, using pre-post single group designs, have dominated the study of specialized multidisciplinary pain rehabilitation interventions.

1.5.4.1. Suggested core outcomes and measures: In an effort to streamline evaluation protocols, improve the interpretability and aggregation of data across trials, strengthen publications, and provide assistance to clinicians in treatment decision-making, McGrath and colleagues (2008) identified a core set of recommended outcomes domains for pediatric chronic pain trials, known as the Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinic Trial (PedIMMPACT). To do so, a consensus group, representing a broad spectrum of expertise in pediatric pain (including participants from academic research, government funding and regulatory agencies and the pharmaceutical industry) was assembled, and a 2-stage Delphi poll was employed to focus attention on relevant domains and measures (McGrath *et al.*, 2008). Eight outcome domains were identified and agreed upon by the consensus group: 1) pain intensity; 2) global judgment of satisfaction with the intervention; 3) symptoms and adverse events; 4) physical functioning; 5) emotional functioning; 6) role functioning; 7) sleep; and 8) economic factors. When compared to the multiple consequences previously presented, many domain gaps become evident (e.g., family functioning, school and peer functioning). Furthermore, intervention stakeholders and users, including youth with pain-related disability, their parents, clinicians and managers, were not consulted in the development of these recommendations, leaving the criteria upon which these partners determine the value or worth of an intervention unknown.

Despite the availability of this core outcome set, large variations in the uptake of these outcome domains and the measures to examine the effectiveness of these interventions are apparent. For example, in their systematic review of one randomized control and nine non-randomized trials evaluating the short-term effectiveness (2-6 months) of IIPT programs, occurring in day hospitals or on an inpatients unit, designed for youth with disabling chronic pain (<22 years old), Hechler and colleagues (2015) noted that only five outcome domains, i.e., pain intensity, disability, school functioning, anxiety and depressive symptoms, were measured consistently in the trials. However, upon review of the PedIMMPACT recommendations, only four domains were regularly measured, since anxiety and depression are included in the 'Emotional Functioning' domain of these recommendations. Furthermore, although the PedIMMPACT also provided published measurement recommendations, significant heterogeneity in the measures used to assess these outcomes was reported, hindering data pooling for meta-analysis (Hechler *et al.,* 2015). Recently, Liossi and colleagues (2019) published a systematic review and subset meta-analysis of 19 single group non-randomized and nine randomized trials, evaluating either inpatient or outpatient based interventions, involving the coordination of at least two disciplines, designed for youth with chronic pain (<22 years), and for which pain frequency and/or intensity was measured. Similar to Hechler and colleagues' (2015) systematic review, the heterogeneity of the outcome domain and their associated measures were underscored (Liossi *et al.*, 2019). Apart from pain intensity (which was a study inclusion criterion), the most commonly assessed outcomes were functioning and depression (Liossi *et al.*, 2019).

1.5.4.2. Evidence synthesis of IIPT and MMT effects measured and tools used: For the purpose of this thesis, an evidence synthesis was conducted examining the constellation of effects of specialised interdisciplinary programs reported across studies and to compare them to those suggested by McGrath and colleagues (2008). The search strategy targeted a broad range of databases including: Academic Search Complete, CINAHL, ERIC, MEDLINE, PsycINFO. Google Scholar search engine was employed to uncover any gray literature (e.g. thesis or dissertations) and used the search terms identified in Table 3. The search was restricted to peer-reviewed articles, in English or in French, published between January 1999 to November 2019. In collaboration with an experienced librarian, the search strategies were adapted for each database as needed. The details of the search strategy terminology are provided in Table 1.

Table 3.

Search Strategy Key Words

Population		**Intervention**	**Outcomes**
“chronic pain” OR “persistent pain” OR “long term pain” OR “headach*”	child* OR youth OR adolescen* OR teen* OR pediatric* OR paediatric*	multidisciplinary treatment* OR interdisciplinary treatment* OR rehabilitation OR intensive interdisciplinary pain treatment* OR "inpatient pain treatment* OR day* hospital program*	outcome* OR effect* OR imact* OR consequence* OR disabilit* OR "activit* limitation*" OR function* OR “quality of life*” OR “school attendance” OR participation

To be included studies had to meet the following inclusion criteria: a) target population comprised of youth with pain-related disability; b) any type of pain condition recognized as chronic (i.e., recurrent or persistent, lasting longer than 3-months); c) concurrent treatment with at least three disciplines conducted in outpatient or day-hospital settings; d) outcomes had to be experienced by youth and/or their parents as a result of the intervention; e) primary studies. Studies were excluded if: a) the sample population was composed of adults only (i.e. 24 years and above), b) pain was deemed acute (less than 3 months) and did not impact function; c) intervention description was mono-discipline, symptom-focused (e.g. pharmaceutical management only) or included inpatient IIPT only, d) consisted of a single case study or population-based study, or e) reported only diagnosis, assessment, and/or prevalence. Both qualitative and quantitative studies were included as pain is a subjective experience for which a combination of instruments is often required to assess the widespread effects of the interventions used to treat it (Eccleston et al., 2006; Kempert et al., 2015).

Four reviewers, including 3 stakeholders (i.e., one healthcare professional and 2 youth with pain-related disability) trained in research methodology and experienced in qualitative and quantitative systematic synthesis, conducted the selection process. When study relevance was questioned, the complete article was read and if disagreement arose discussion occurred until consensus was reached. A flow chart of the source selection process for each stage is presented in Appendix 1. Selected studies were imported into NVivo and data was extracted using the following headings: author, year, country, design, participants demographics, intervention, data collection time point(s), outcome domains and measures.

Appendix 2 presents a summary of the findings of the 34 studies comprising this review. Light grey shading identifies the day-hospital IIPT studies (n=23), white the MMT studies (n=9), and dark grey shading a comparision study (n=1) included in the review. Thirty-two studies used quantitative methodology. Most IIPT studies employed a non-randomized pre-post designs with a single group, while RCTs were mostly found in the MMT studies. The control group typically involved in the MMT comprised of a group receiving a different therapeutic approach (CBT vs ACT), or dose (e.g., PT once per week or three times per week) of a treatment component. Three qualitative studies were included. Overall, the review identified 64 outcome domains, 43 youth-focused, and 21 parent-focused, for which 79 different measures or methods were used to collect the effect data. Most measures were self-reported, and the data collection timeframes were limited to immediately or shortly after the end of the intervention (i.e., 2-6 months). In addition to specifying the outcomes domains and measures used in each study, this synthesis also compared the effects and measures to those recommended by the PedIMMPACT. The most common outcome domains considered included pain intensity, physical functioning, emotional functioning (depression), while sleep, global judgment of satisfaction with the intervention, and economic factor were rarely assessed. Only one MMT study considered adverse symptoms in all of the studies reviewed.

When considered separately, IIPT and MMT studies have reported promising short-term (at discharge-6 months) and long-term (12-60 months) effects. Most specifically, day-hospital IIPT have demonstrated significant short and long-term reduction in pain intensity (Banez *et al.*, 2014; Benore *et al.*, 2015; Benore *et al.*, 2018; Eccleston *et al.*, 2003;Kemani *et al*, 2019; Kempert *et al.*, 2017; Logan *et al.*, 2012a; Pielech *et al.*, 2018; Randall *et al.*, 2018;

Sieberg *et al.*, 2017; Sherry *et al.*, 1999; Sherry *et al.*, 2015; Simons *et al.*, 2012), in pain-related disability (Benore *et al.*, 2015; Benore *et al.*, 2018; Bruce *et al.*, 2017; Gauntlett-Gilbert *et al.*, 2012; Kemani *et al.*, 2019; Logan *et al.*, 2012a; Pielech *et al.*, 2018; Randall *et al.*, 2018; Risko, 2018; Sieberg *et al.*, 2017; Sherry *et al.*, 1999; Sherry *et al.*, 2015; Simons *et al.*, 2013), in catastrophizing, general anxiety and pain-specific anxiety (Benore *et al.*, 2018; Bruce *et al.*, 2017; Eccleston *et al.*, 2003; Gauntlett-Gilbert *et al.*, 2012; Kemani *et al.*, 2019; Logan *et al.*, 2012a; Simons *et al.*, 2013; Weiss *et al.*, 2013). However, only small to moderate effect have been observed in depressive symptoms (Benore *et al.*, 2015; Bruce *et al.*, 2017; Kemani *et al.*, 2019; Logan *et al.*, 2012a; Sherry *et al.*, 2015; Simons *et al.*, 2013; Weiss *et al.*, 2013). Short and long-term improvements have been noted in school functioning (Banez *et al.,* 2014; Benore *et al.*, 2015; Benore *et al.*, 2018; Eccleston *et al.*, 2003; Gauntlett-Gilbert *et al.*, 2012; Logan *et al.*, 2012a; Randall *et al.*, 2018; Sherry *et al.*, 2015; Weiss *et al.*, 2013). Significant reductions in health care and medication utilization, both in the short and long-term have been underscored (Banez *et al.*, 2014; Bruce *et al.*, 2017; Gauntlett-Gilbert *et al.*, 2013; Logan *et al.*, 2012a). Evidence of changes in parent mood, behaviours and function were noted (Benore *et al.*, 2018; Kemani *et al.*, 2019; Pielech *et al.*, 2018; Sieberg *et al.*, 2017). Finally, parents and youth described positive changes to their skills to self-manage, feelings of hope, empowerment and self-efficacy (Gaughan *et al.*, 2014; Risko, 2018)

MMT evaluation studies have also reported important effects, with the majority being short-term. These include the following: reduction in pain frequency and intensity (Kashikar-Zuck *et* al, 2018b; Lee *et al.*, 1999; Revivo *et al.,* 2018; Soee *et al.*, 2013, Simons *et al.*, 2010; Trans *et al.,*2016; Wicksell *et al.*, 2009), pain-related disability (Lee *et al.*, 1999; Kanstrup *et al.*, 2016; Revivo *et al.*, 2018; Simons *et al.*, 2010; Soee *et al.*, 2013; Trans *et al.*, 2016; Wicksell *et al.*, 2009), depressive symptoms (Kanstrup *et al.*, 2016; Revivo *et al.*, 2018; Trans *et al.*, 2016), general anxiety and pain-related anxiety (Revivo *et al.*, 2018; Trans *et al.*, 2016), along with improvements in physical and social functioning (Revivo *et al.*, 2018) in youth. Some reductions in health care and medication utilization in the short underscored (Simons *et al.*, 2010). Furthermore, short-term improvements in parent emotional functioning (Revivo *et al.*, 2018) and positive behaviour change (Kanstrup *et al.*, 2016) have been noted. Although in existence for the same amount of time as IIPT, few studies have considered the longitudinal effects of MMT.

Comparison of these two intervention models are rare. Only one study was found. Simons and colleagues (2013) examined 100 matched participants, 50 who enrolled in an IIPT day hospital program, and 50 who pursued the recommended multimodal outpatient treatment as recommended following assessment of their pain condition by a specialized multidisciplinary pediatric pain clinic team. Improvements were found in both treatments at discharge, yet youth involved in the IIPT day-hospital program had significantly larger improvements in the outcome domains measured, which included functional disability, pain-related fear, and readiness to change (Simons *et al.,* 2013). As highlighted by these authors, this finding was not surprising given the larger treatment dose received in the IIPT group and the limited ability to verify treatment adherence in the MMT (Simons *et al.,* 2013).

The rarity of program comparison studies may be attributed to the inconsistencies in the outcome domains evaluated and the measurement tools used to assess a same domain, (e.g., pain intensity, emotional functioning (i.e., depression and anxiety) in the studies, as highlighted in this synthesis and previously by others (Hechler *et al.*, 2015; Stahlschmidt *et al.*, 2016). Other outcome domains and measures not included in the original PedIMMPACT recommendations, also emerged with increased consistently across program evaluations (e.g., family functioning, parent behaviours). In developing the PedIMMPACT, McGrath and colleagues (2008) acknowledge that any core set of outcomes and measurements is provisional, dynamic and should be subject to frequent updates. Unfortunately, despite the extensive research in the field and the development of new measures, the PedIMMPACT recommendations have yet to be modernized.

1.5.4.3. Intervention descriptions: Another factor contributing to the rarity of program comparison may be a lack of comprehensive description for each program currently missing in the literature. In the PedIMMPACT development, McGrath and colleagues (2008) recognized that pediatric chronic pain and its interventions require contextualizing in order to better understand this population and their outcomes. Liossi and colleagues (2019) using the Template for Intervention Description and Replication (TIDiER) (Hoffmann *et al.*, 2014), a 12-item checklist developed to improve the reporting of interventions in evaluative studies, highlighted that many of the specialized multidisciplinary rehabilitation studies failed to clearly describe the mode of delivery (individual, group), where the intervention was conducted, which professionals facilitate which intervention component, whether the intervention was personalized, and if so in what ways.

Furthermore, the interactions between disciplinary processes and mechanisms of change within these programs have remained ill-explored (Odell & Logan, 2013; Harrison *et al.*, 2019). Stahlschmidt and colleagues (2016) also proposed that the descriptions of the program's separate components alone may not be enough to capture what components create an effective treatment and suggest that the effectiveness may arise from the non-linear effects of interactions within the context, including those between the program components, the interdisciplinary team and the team, the parents and the patients.

Currently, programs exposed youth participants to a wide array of treatment components, which vary significantly between programs This "kitchen sink" approach is inefficient, in determining who will benefit, and how outcomes are achieved (Simons *et al.*, 2013). A more standardized method of reporting IIPT and MMT treatment components and of rendering explicit the multidisciplinary processes and mechanisms are required (Stahlschmidt *et al.,* 2016). Reporting the null or negative effects of these specialized multidisciplinary pain interventions is also provide valuable insight into the effective and non-effective components of these specialized interventions (Caes *et al.*, 2018).

The literature reviewed provides a synopsis of the various issues surrounding the complexity of pain-related disability, its treatment, and the evaluation of these programs, which helped frame the program evaluation perspective for this study. It also exposed many gaps in the literature. First, while this review identified an increased need to evaluate these interventions, it also identified that those evaluated were poorly described, and inadequately theorized. Not only does this lack of description hinder their implementation into alternative clinical settings, it also impedes program comparisons and whether the effects produced are a function of the intervention or other factors. Secondly, although a core set of outcomes has been developed for for this population, many intervention stakeholders and users, were not consulted in identifying them. The omission of these important perspectives renders the criteria upon which stakeholders determine the value and worth of these programs unknown. Recognizing this unfortunate oversight, the advances in pediatric pain research and measurement development, the core outcome dataset should be updated. Thirdly, although some evidence exists on the effectiveness of IIPT, longitudinal comparisons between interventions and levels of service are rare, in particular for those provided in the same facility. As a result, the value of one program in relation to other intervention options remains unclear. The causal relationship between an appropriately theorized specialized multidisciplinary pain rehabilitation intervention and all its effects (i.e., positive and negative) requires exploration.

1.6. Conceptual Framework

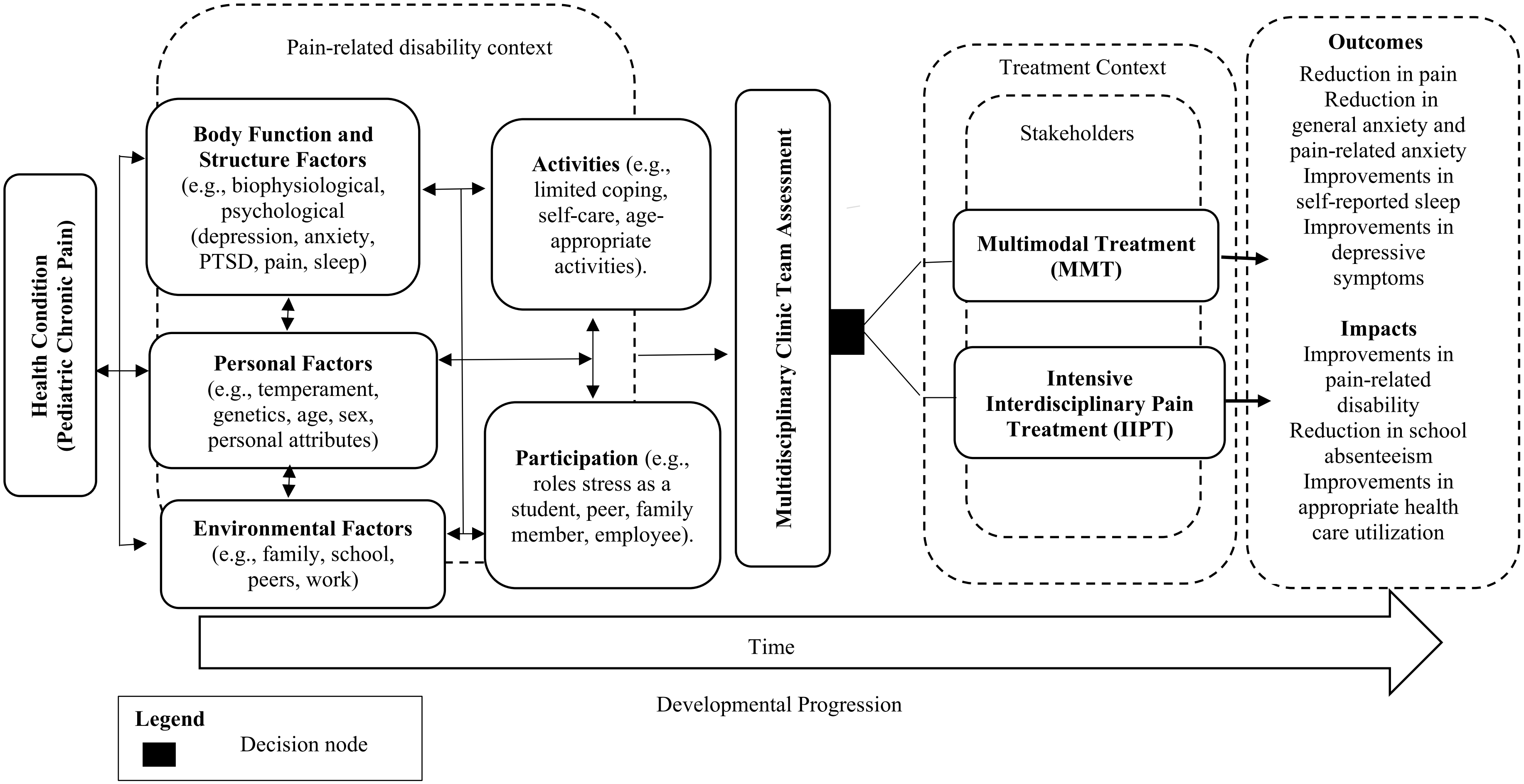
In light of the reviewed literature, a conceptual framework was developed and is presented in Figure 1. It provided the guidance and the structure to this study. Furthermore, it helps identify the theoretical relationships between the pediatric chronic pain contributing factors and its functional consequences within a disability context, and acknowledges the influence of the intervention models, their contexts, and their associated outcomes and impacts. A description of the various components of the model is discussed below, along with the rationale for their selection.

A modified version of the *International Classification of Function* (WHO, 2001) and its category schema was used to represent the contributing factors associated with the development and sustainability of pediatric chronic pain in an attempt to make further links between these contributing factors, the functional deteriorations experienced by these youth, and their resulting disability. More specifically, the biophysiological mechanisms and psychological factors identified in the literature review were included in the Body Function and Structure, and the personal traits and attributes (e.g., gender, sex) were incorporated into the Personal Factors. All social factors were united under the Environmental Factors, including those associated with the family, parents, siblings and peers. The Activity and Participation components, which cover a full range of life areas from basic learning to life skills, were associated with those concepts related to functioning (e.g., coping, school functioning). Within this conceptual model, the pain-related disability context is created by the interaction between the aforementioned contributing factors (i.e., body function and structure, personal factors and environmental factors) and the activity and participation limitations which ensue. The figure represents the timing of the multidisciplinary pain team consultation (i.e., when functional deterioration and disability due to pain escalates) and the role of the team in recommending an intervention option. Once the multidisciplinary assessment is completed, clinicians provide recommendations to families, suggesting the intervention from the options available and the one most likely to improve the youth's functioning. This clinical decision-making point is indicated in the figure by a decision node, where either the IIPT or the MMT is selected.

As previously highlighted, evaluation literature recognized the interplay between an intervention and the organizational factors. These organization factors include the individual stakeholders, their collective practices, and the unique context in which the program operates. As these factors impact the achievement of intervention outcomes, these components have been added to the figure. In illustrating the product of the intervention, the term *outcome* has been used to indicate medium-term intervention results (i.e., those reported in the literature to be achieved within 2-6 months), while the term *impact* has been reserved for those that are longer term. As highlighted in the literature review, the evaluations of IIPT and MMT have produced similar outcomes and impacts and therefore are not differentiated in the figure. Finally, a time and developmental perspective have been included, as represented by the arrow at the bottom. These elements are meant to draw attention not only to the importance of the timing of the intervention within the child's chronic pain trajectory and its effect on the outcomes achieved in relation to the intervention, but also on rapidly changing developmental environments which occurs in adolescence, a reality shared by all pediatric rehabilitation interventions and services.

Figure 1.

Visual Diagram of the Conceptual Framework



1.7. Research Questions and Objectives

The literature reviewed in this chapter provides a synopsis of the contributors and consequences of chronic pain, of the recognized specialised interventions used to treat this population, and various program evaluation concepts that have assisted to frame the evaluation of multidisciplinary pain rehabilitation treatments. It also exposed numerous gaps in the relevant literature.

The evaluative study presented in this thesis seeks to address the gaps identified in this literature and to advance the knowledge related to the evaluation of specialized rehabilitation pain treatment programs. More specifically, the evaluation aims to answer two separate, yet complementary questions articulated below.

> Question 1. Is the IIPT program at Alberta Children's Hospital theoretically sound to produce its anticipated outcomes?
>
> Question 2. Is the IIPT at Alberta Children's Hospital more effective in treating youth living with chronic pain compared to the MMT program provided by the same facility, based on stakeholder prioritized outcomes?

Furthermore, aligned with each of these questions are several objectives. As a result of the evaluating the theoretical foundation of the IIPT program, this study will aim:

> Objective 1. To model the IIPT program theory and test its plausibility in achieving the anticipated outcomes (Article 1).

In assessing the IIPT program effects, this purpose of this study will be:

> Objective 2: To identify and prioritize a set of outcomes meaningful to all program stakeholders upon which the evaluation will be based (Article 2);
>
> Objective 3: To longitudinally compare the change in outcomes of youth with pain-related disability participating in the IIPT to those enrolled in the MMT based on the stakeholder-prioritized outcomes at 3 and 12 months (Article 3);
>
> Objective 4: To explore and compare the impacts of both specialized multidisciplinary pain rehabilitation programs (i.e., IIPT and MMT) based on the experiences and perceptions of youth and parent program participants (Article 4).

CHAPTER 2. STUDY METHODOLOGY AND RESULTS

2.1. Study Context

A program context consists of any factors that can influence, facilitate or hinder a program, including social structures, political conditions, the economy, funding, and a variety of associated stakeholders (Chen, 2014). This study context is comprised of three organizations: a) Alberta Children's Hospital (ACH), a tertiary care pediatric health facility that provides health and rehabilitation services to youth and their families; b) Alberta Health Services (AHS), the provincial-wide healthcare organization that provides all publicly funded health services (including those at ACH); and c) a specialized school, located within the walls of the hospital, and part of a regional board of education. Family-centred care is a principal foundation that units these three organizations and is defined as follows: *children, youth, families, and staff partnered together to provide excellent care and education by treating each other with respect, listening to one other, sharing ideas and information, asking and answering questions, creating a safe environment and making decisions together* (Family and Community Resource Centre, 2019). Alberta Children's Hospital (ACH) is the main provider of specialized pediatric health and rehabilitation services in the following geographical jurisdictions: southern Alberta, south-eastern British Columbia and south-western Saskatchewan. It is home to one of the most comprehensive pediatric complex pain clinical programs in Canada.

This program includes two comprehensive clinics, the Complex Pain Clinic (CPC) and the Headache Clinic, associated outpatient MMT services (e.g., psychology, physiotherapy, medical interventions, psychoeducation), and the day-hospital IIPT. Many of these services (e.g. Headache Clinic, day-hospital IIPT) were created as a result of a clinical expansion that occurred in 2014, thanks to the generosity of a large 5-year philanthropic donation. In addition to coordination and administrative staff, the day-hospital IIPT is comprised of a comprehensive interdisciplinary team, including physicians from a variety of specialities (e.g., anaesthesia, neurology), nurses, a nurse practitioner, physiotherapists, psychologists, a family counsellor all specifically trained in pediatric chronic pain. The program also shares a staff complement with the rehabilitation day-hospital service including a recreation therapist, an occupational therapist, therapeutic assistants, a program coordinator, and its academic resources (i.e., teachers, educational supports, an assistant principal), along with spiritual care and child life services, more specifically art and music therapy. The assistant manager of rehabilitation services oversees the allied health and nursing health human resources compliment. The IIPT aims to foster pain self-management in youth and their families, and improve youth functioning, despite pain. With the end of the donation approaching, the need to evaluate some of these service components arose. Senior decision makers at the program and facility-level requested the evaluation of the IIPT, with the goal of providing information on the effect of the program to assist them in determining the future of these services, in particular the IIPT.

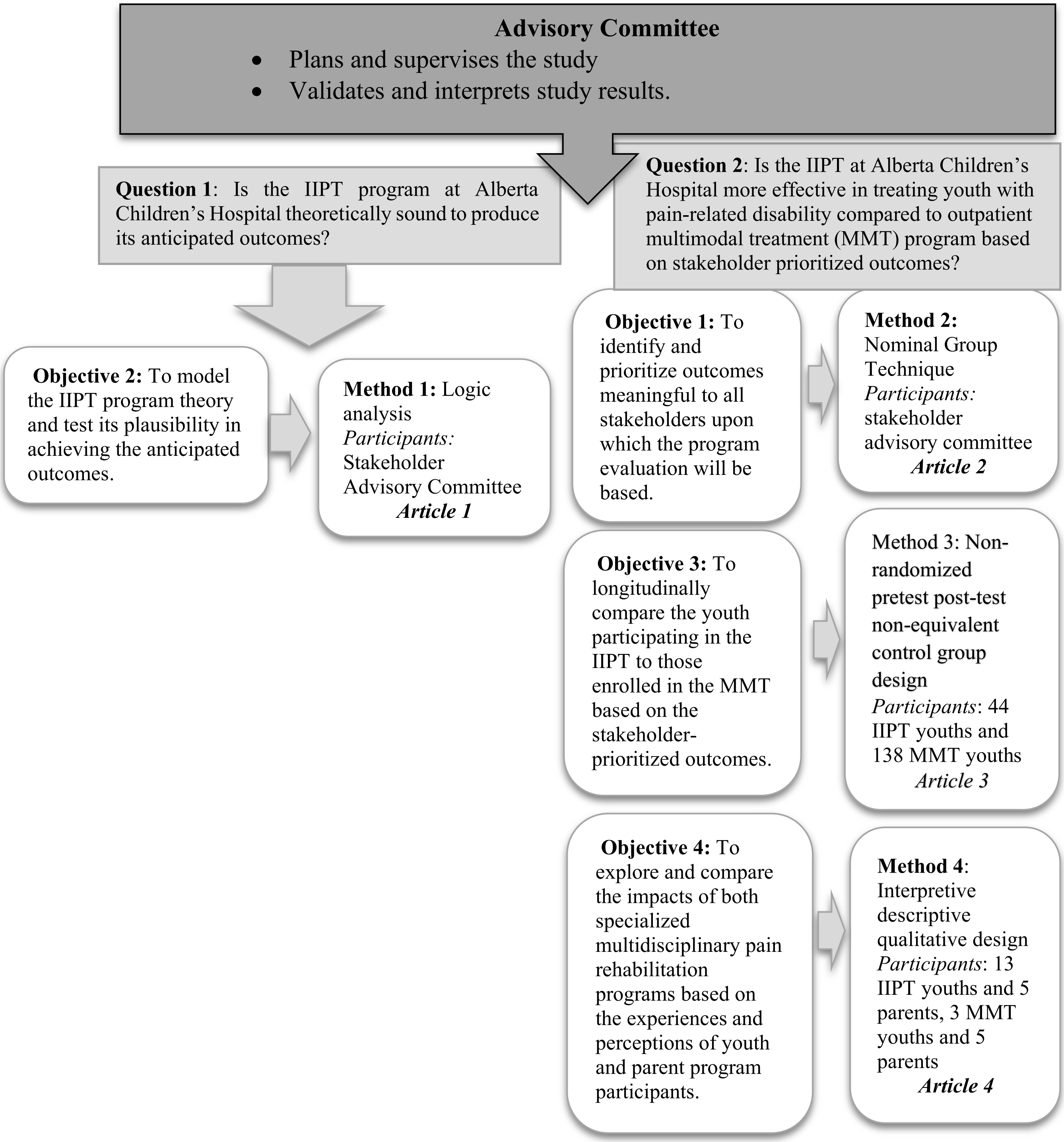
As stakeholder*s* within the context can challenge the establishment of common evaluation outcomes upon which success is judged (Lamontagne et al., 2010), representatives from the diverse partners involved in the pain intervention programs (e.g. youth with chronic pain and their parents, health providers, teachers, and managers) were recruited with the assistance of the hospital leadership and school administrative teams, Complex Pain Clinic. The inclusion of youth and their parents' representatives in the evaluation reflects the family centred care philosophy and collaborative and inclusive nature in which care is provided within the services, across the facility, and across stakeholder organizations.

2.2. Study Design

Figure 2 provides an overview of the methodology used for this study. As illustrated, some of the phases are interlinked and therefore decisions made in one phase affected the methodological procedures in the subsequent phases. Moreover, each phase was guided by a particular research question and/or objective and used specific methodology and participants to address it. A more detailed description of the study design, participants, data collection and analysis procedures, along with the findings of each phase are presented and can be found in the included articles, available in the subsequent chapters as highlighted in the diagram.

Figure 2.

Visual Diagram of the Methodology

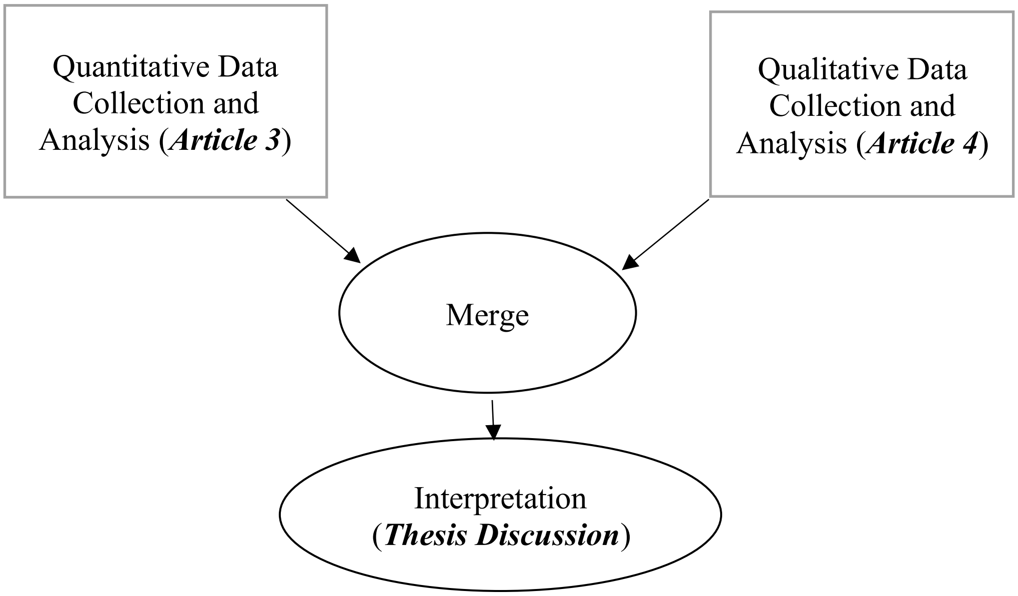


More specifically, Article 1 highlights the collaborative logic analysis process and procedures used with the stakeholder advisory group to theorize and validate the IIPT program theory, and its findings. Article 2 explicates the results from a nominal group technique, where representatives from the stakeholder groups identified and prioritized meaningful outcome domains upon which the evaluation would be based. Article 3 examines the findings from the effectiveness study, using a pre-post repeated measures with a treatment as usual group, used as the control. Patient-reported outcomes, representative of some of the stakeholder-prioritized outcome domains identified in Article 2, were completed by youth with pain-related disability participating in the IIPT and in the MMT and then compared longitudinally over 4 time points (pre-3, 6, 12 months post). Finally, Article 4 qualitatively and longitudinally describes and compares the perceptions of youth and their parent participants on the impacts of the specialized multidisciplinary pain rehabilitation interventions (i.e., IIPT and MMT) using a narrative timeline approach.

In order to comprehensively address our second research question, i.e., whether the IIPT is more effective in treating youth living with chronic pain compared to the outpatient MMT, a convergent mixed method design was used (please see Figure 3). As illustrated in Figure 3, the intent of the convergent design is to merge the qualitative and quantitative data analysis results, providing both a qualitative and quantitative picture and different insight into the impact of the IIPT intervention (Creswell, 2014).

Figure 3.

Visual Diagram of Convergent Mixed Method Design.



The quantitative and qualitative data were collected and analyzed separately (Articles 3 and 4). More specifically, following the presentation of results in Article 3 and Article 4, the findings will then be compared, and the extent to which the quantitative results are confirmed by the qualitative findings will be presented in the discussion of this thesis.

Prior to moving on to the details of these articles, a brief overview of the intervention and the overall study procedures, including the participatory approach employed, and my position within the evaluation context, will be reviewed.

2.3. Treatment Interventions

At the heart of this evaluation rests two specific specialized multidisciplinary pain rehabilitation interventions provided through the Complex Pain Program at Alberta Children's Hospital. A detailed description of each one follows.

2.3.1. Outpatient Multimodal Treatment (MMT)**:** The MMT encompasses a multimodal intervention program of group or individualized pain education for youth and their parents, weekly individual physiotherapy sessions focused on functional stretching, strengthening, and endurance training, individual cognitive behavioural focused psychology sessions, medical treatment (e.g. medication prescription, injections), and home program recommendations (e.g. relaxation practice, strengthening) as appropriate. Discharge from programs is individualized and based on youth and family identified intervention needs and treatment goals.

2.3.2. Intensive Interdisciplinary Pain Treatment (IIPT)**:** The IIPT is a 3-6–week interdisciplinary program including daily physiotherapy, psychology, occupational therapy, recreation, and academics, along with weekly art and music therapy, medical and nursing interactions. Youth spend 6 hours daily, 5 days per week, in individual, group, and family treatment sessions designed to enhance their pain management skills, facilitate emotional adjustment, improve functioning, and return to normal age-appropriate activities. Youth attend the hospital-based school and self-management is emphasized by all disciplines throughout and within all program activities. Discharge is individualized based on youth and parent identified functional goals.

2.4. Overall Study Procedures:

2.4.1. Participatory Approach: This study adopted an overall participatory evaluation approach. As per the evaluation sponsor's request, the approach was concerned with providing support for ongoing program and organization decision-making and to practically problem solve with an aim to foster program improvements. This approach was chosen for its observed benefits of supporting decision making, and for its strategic function. Stakeholder advisory committee members (*n*=13) worked in partnership with the members of the research team members to implement the study and provide oversight for this evaluation. Diversity in participation of non-research members was ensured by its inclusive recruitment strategy and included representatives from the primary users of the evaluation findings (i.e., physician (*n*=1), program coordinator (*n*=1) and health care manager (*n*=1) and those with a vested interest in the program (i.e., clinicians (*n*=4), teachers (*n*=2) youth with pain-related disability (*n*=2) and their parents (*n*=2)). Due to the diversity and inclusiveness of the committee, process decisions could be made quickly as group perspectives were deemed to be exhaustive. Secondly, through the development of a team charter, all stakeholders committed to share expertise, the responsibilities and ownership of the project and its findings, to focus on gaining a better understanding of specialized multidisciplinary pain rehabilitations, to put any new knowledge gained into action, and to enhance the program and improve participants' and their families' well-being. This commitment assisted in conceptualizing this evaluation as a learning process for both the researcher and for the stakeholders, assisting in building evaluation skills and abilities, a respect for all ways of knowing, and a capacity to produce knowledge and interpret it together. A political rationale for using this approach was also an important consideration, in particular in relation to the use of evaluation findings. Involvement of physicians and clinical staff was deemed crucial to the adoption of program improvement recommendations, and the acceptance of program outcomes and uncovered impacts.

Each advisory committee participated extensively in the research process. When examined as a function of the research process, the advisory group validated the research objectives, reviewed and selected the studies included in the literature review, identified and prioritized the outcome domains to be measured, reviewed the outcome measures to be used, assisted in developing the program theory and updating the logic model of the interventions to be evaluated, prioritized the program improvement recommendations following the logic analysis, refined the questionnaire data collection schedule, shaped and piloted the qualitative interview process, provided suggestions for, and assisted with participant recruitment, validated the results, and assisted in disseminated findings throughout the research process to their stakeholder audience, as well as others (e.g., academic community, funders).

The only tasks that were completed solely by the research team with minimal input from the advisory committee were: 1) the scoping review to test the plausibility of the program theory in the logic analysis process, 2) the statistical analysis of the quantitative data, and 3) the initial theming of the interview narratives. As evidenced by their participation, control of the technical decision-making was shared among the stakeholder advisory group members, and evaluation tasks were distributed among the advisory group, yet were dependent on members' availability, time constraints and tasks complexity. Due to the customary hierarchies present within the advisory group, many specific targeted activities aligned with best practices in stakeholder engagement were included in this study, as a means to minimize the power differentials. Firstly, roles, responsibilities and expectations, meeting ground rules, and decision-making and conflict resolution processes were developed collaboratively, documented, and signed off by all members (including researcher) early in the evaluation process. Formal orientation and training sessions were provided and completed by all members, and educational resources were circulated regularly. Routine individual check-ins were conducted to ensure members’ comfort with the experience and that adequate support and training was being provided.

To foster engagement and to maximize inclusive participation, regular meetings were scheduled with 2 weeks’ notice provided and any materials were pre-circulated. A variety of communication platforms were also made available and utilized, including face-to-face interactions, telephone, email, and teleconferencing (e.g., Skype, FaceTime). Despite the scheduling procedures, meetings and discussions with full advisory group attendance were difficult and rare. Typical reasons provided included caseload and work schedules. To further accommodate and manage this logistical glitch, a sequential meeting strategy was adopted where members could choose from a variety of meeting times within a week timeframe. Any previous meeting discussions were summarized, and further discussion progressed from that point onward. However, decisions were not final until all advisory members were consulted. This process did somewhat increase the time and resource investment of the study.

2.4.2. Researcher's Position: An evaluator's attitudes, values, behaviours and characteristics are reported to have a strong influence on the evaluation process (Crisha, 2006). It is also acknowledged that the identity and position of the evaluator are multifaceted evolving notions linked to the conceptualization of the evaluation within a particular setting, the stakeholders involved, and the collaborative strategies adopted (King *et al.*, 2001). To enhance the methodological rigour of this study, I recognize the need to acknowledge my particular viewpoints and experiences within the context of this evaluation and the development of this study (Ritchie *et al.,* 2009). For the purpose of this evaluation, I identify as a career-driven female, who has shared most of my adult life with someone who self-manages chronic pain daily. I am a pediatric physiotherapist with over 25 years of experience working with youth with chronic disabling and painful conditions and their families. My career has included: working as a clinician and member of an interdisciplinary team, as a program facilitator in a pediatric day-hospital, as a program developer designing interventions and services for children with disability and pain, and as a healthcare manager. The philosophy of family-centred care permeates all facets of my practice (i.e., clinical, program design, and managerial), and my research interests. This philosophy, grounded in mutually beneficial partnerships among healthcare providers, patients, and families, recognizes the vital role families play in ensuring the health and well-being of children and other family members (Institute for Patient and Family Centered Care, 2012) and in planning, delivering, and evaluating pediatric healthcare services.

As the program designer and most recently as the manager of the program understudy, my past roles as a leader and decision maker within this group may have created the perception that I was an insider researcher (Richie *et al.*, 2009). These roles did foster a shared identity, language, and experiential base between the study participants and me (Asselin, 2003). As such, some stakeholder representatives trusted me and were more open with some of their responses, while others perceived an alignment with management and were subsequently more reserved. At the inception of the study, I had resigned from my managerial position 2 years prior, rendering me more of an outsider researcher (Dwyer & Buckle, 2009), with my limited access to potential study participants, resources, and data. Regardless, I have to be conscious and self-aware of how these roles could have impacted data collection (e.g., the questions asked) as well as my interpretation of the findings. Three reflective methods fostered this consciousness and minimized potential biases. Firstly, an informal critical friend approach was used. A critical friend is defined as a trusted person who asks provocative questions, provides examination of data through another lens, and offers critique of my work as a friend with the aim of assisting in critical reflection (Costa & Kallick, 1993). This role was assumed by a researcher at ACH, who assumed the role of site coordinator for the duration of the study. Secondly, study journal and field notes were also kept during and/or following all group discussions. The content of the journal tracked the reasoning behind the specific research decisions and my subjective research process. Thirdly, while conducting the analysis, bracketing was used in conjunction with journaling and the critical friend approach (i.e. bracketing interviews) (Tufford & Newman, 2010). Bracketing, a qualitative method, aimed to mitigate the potential detrimental effect of unacknowledged preconceptions or biases related to the research (Tufford & Newman, 2010). This method was chosen as I acknowledge my close relationship to the study topic, the program being evaluated, and a recognized interest in parent and youth experiences within the healthcare system. Journal notes encompassed my emotions, preconceptions, presuppositions and assumption about the program and the potential experiences of stakeholders with it.

Within my pragmatic approach, I recognize the existence of natural and social worlds, and value them both (Johnson & Onwuegbuzie, 2004). This fuels my curiosity for what is and what might be, aligning with a mixed method design. During some of steps of the logic analysis (e.g., scoping review) and effectiveness evaluation (i.e., qualitative research phases), I held assumptions about the natural world that were aligned with realistic ontology, where I accepted that things and events may be independent of thought (Dewy, 1998) and that some knowledge about the world is fact. However, in theorizing the IIPT intervention, prioritizing meaningful stakeholder outcome domains, and in exploring participants' perception of intervention impacts (i.e., more qualitative phases), I tended to hold a more relativist ontology stance, where nature of being is dependent of an individual’s or a group’s perspectives and interpretation of reality (Dewey, 1998). By holding a pragmatic view, I was able to draw on multiple research assumptions (i.e., quantitative and qualitative, and mixed method designs) to better understand the problem, with the assumption that knowledge could be constructed based on the live human experience (Johnson & Onwuegbuzie, 2004). In terms of axiology, a value-oriented approach was used. As such, recognition and acknowledgement that my own values affected my investigation choices, my observations, and my interpretation (Johnson & Onwuegbuzie, 2004). My values were especially important in developing a fair and respectful engagement process associated with the advisory group throughout the study, in ensuring the meaningfulness of the prioritized outcomes to all stakeholders, and in developing qualitative data collection procedures that would foster authentic responses. Furthermore, they also played a role in my understanding, description, and interpretation of the participants’ perspectives. Consequently, throughout the results and discussion sections of this thesis, instead of using the pronoun “I” as is typical practice, the pronoun “we” will be used to reflect the collective contributions and the vital role stakeholders fulfilled in this study.

In summary, this study aims to theorize and validate the IIPT program theory, examine IIPT participant stakeholder-prioritized outcomes and explore its perceived impacts as compared to those enrolled in the MMT. To do so, a participatory evaluation approach was adopted, where multiple phases, each with its own study methodology and participant group, aimed to address a research objective. As the researcher’s position can impact this type of study, my position within the research environment, along with my ontology, epistemology, and axiology stance were presented. As noted above, the next four articles will describe each of the phases in more detail, including the participant sample, data collection and analysis procedures, as well as the findings of each.

Article 1

Preamble

Using collaborative logic analysis evaluation to test the program theory of a pediatric specialized rehabilitation intervention for youth with pain-related disability

Authors: Karen Hurtubise, Astrid Brousselle, Chantal Camden

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Karen Hurtubise's contribution to this manuscript included its conceptualization, the literature and scoping review, the data analysis, creation of the conceptual framework, the presentation of the findings and the writing of this manuscript. The co-authors (Astrid Brousselle and Chantal Camden) supervised and provided validation and guidance during the various stages of the study, reviewed the results and provided feedback on the manuscript prior to submission.

The two authors authorized the integration of this article into Karen Hurtubise's thesis.

Résumé: Les programmes de réadaptation pédiatrique sont complexes et impliquent de nombreux acteurs des secteurs la santé et des services sociaux. La modélisation de ces programmes, une exigence en évaluation de programme, peut être difficile et nécessite l'engagement des acteurs. Cependant, les données probantes sont limitées quant à la meilleure façon de les impliquer dans le processus de théorisation. L'analyse logique, une évaluation théorique qui teste la plausibilité d'une théorie de programme en utilisant des connaissances scientifiques et expérientielles, semble une avenue prometteuse. Son utilisation est rare en réadaptation pédiatrique et dans les programmes sociaux et communautaires. Ainsi, peu de détails sur l'approche méthodologique sont disponibles. Cet article décrit l'utilisation d'une méthodologie d'analyse logique collaborative dans le contexte de l'évaluation d'un programme de réadaptation visant à outiller des jeunes atteints d'un handicap lié à la douleur dans l'autogestion quotidienne de leur condition. Un processus d'analyse logique direct en trois étapes a été utilisé. Un groupe d'experts composé de 13 membres incluant des cliniciens, des enseignants, des gestionnaires, des jeunes souffrant d'un handicap lié à la douleur et de leurs parents, a été impliqué à chaque étape. 1) Un modèle logique a été construit au moyen d'une analyse de documents, de sondages auprès de groupes d'experts, et de discussions de groupe. 2) Une revue de portée, portant sur l'autogestion chez l'adolescent, le développement de l'auto-efficacité, et la promotion de la participation a permis de créer un cadre conceptuel. 3) La comparaison du modèle logique et du cadre conceptuel a permis de formuler des recommandations pour l'amélioration de l'intervention. Dans l'ensemble, le processus d'analyse logique collaborative a permis : 1) de faire connaître les hypothèses des intervenants sur les mécanismes de causalité du programme ; 2) d'inclure les éléments les plus valorisés par les jeunes et leurs parents ; 3) d'identifier les caractéristiques du programme appuyées par les connaissances scientifiques et expérientielles, ses lacunes et les données probantes émergentes dans le domaine. L'analyse logique collaborative est une méthodologie accessible et réalisable qui permet de centrer l'analyse sur les besoins des utilisateurs et de tester la plausibilité de la théorie du programme d'interventions pédiatriques en santé, en sciences sociales et communautaires. Elle constitue une méthode prometteuse pour impliquer les acteurs dans l'évaluation de programmes de réadaptation pédiatrique.

Abstract: Intensive interdisciplinary pain treatment (IIPT) involve multiple stakeholders. Mapping the program components to its anticipated outcomes (i.e., its theory) can be difficult and requires stakeholder engagement. Evidence is lacking however on how best to engage them. Logic analysis, a theory-based evaluation, that tests the coherence of a program theory using scientific evidence and experiential knowledge, may hold some promise. Its use is rare in pediatric pain interventions, and few methodological details are available. This article provides a description of a collaborative logic analysis methodology used to test the theoretical plausibility of an IIPT designed for youth with pain-related disability. A 3-step direct logic analysis process was used. A 13-member expert panel, composed of clinicians, teachers, managers, youth with pain-related disability and their parents were engaged in each step. First, a logic model was constructed through document analysis, expert panel surveys and focus-group discussions. Then, a scoping review, focused on pediatric self-management, building self-efficacy, and fostering participation helped create a conceptual framework. An examination of the logic model against the conceptual framework by the expert panel followed, and recommendations were formulated. Overall, the collaborative logic analysis process helped raise awareness of clinicians' assumptions about the program causal mechanisms, identified program components most valued by youth and their parents, recognized the program features supported by scientific and experiential knowledge, detected gaps and highlighted emerging trends. In addition to providing a consumer-focused program evaluation option, collaborative logic analysis methodology holds promise as a strategy to engage stakeholders and to translate pediatric pain rehabilitation evaluation research knowledge to key stakeholders

Key Words: Logic analysis, intervention theory, theory-based evaluation, logic model, pediatric chronic pain, interdisciplinary pain rehabilitation program.

1. INTRODUCTION

Pain-related disability affects eight percent of youth.[1,2] Within the pediatric pain context, pain-related disability is defined as pain which impairs youth's ability to perform age-appropriate activities relevant to daily life.[3,4] Due to the complexity of these impairments, intensive interdisciplinary pain treatment (IIPT), a specialised multidisciplinary rehabilitation intervention, is viewed as the treatment of choice.[5-9] To be considered an IIPT program, three or more disciplines (e.g., pain specialist, psychologist, physiotherapist) must work together, in an integrated manner, guided by a shared rehabilitation philosophy.[7,10,11] The aim of IIPT intervention is self-management, whereby youth and their parents actively engaged in managing pain, and resume participation in age-appropriate activities.[12] Although these programs exist worldwide, their comparison and reproducibility are complicated by poor descriptions of the intervention components, and a lack of transparency in how the components produce the anticipated outcomes.[12,13] (. Moreover, stakeholders' perceptions of the value of these programs are missing from the evidence, rendering judgment of their worth difficult.

Integrated knowledge translation (IKT) is a model of collaborative research, where researchers and stakeholders engage together to produce mutually beneficial research and optimize healthcare delivery.[14] Stakeholder engagement is increasingly recognized as essential, and believed to increase accountability, broaden the underlying value base, and enhance the relevance and utilization of the research findings.[15,16] (However, how best to engage stakeholders is less well-known. To date, stakeholder engagement in the evaluation of interventions, like IIPT, has been limited.[16-19]

Interventions like IIPT are recognized as complex. According to the Medical Research Council, a complex intervention is described as one that contains several interacting components, requires various behaviors to be exhibited by both those delivering and those receiving it, incorporates different groups and organizations, includes many different outcomes, all the while exhibiting flexibility or tailoring.[18] The interaction of these multiple components can be represented as a program theory, defined as the specific activities by which an intervention achieves its anticipated outcomes.[20] Furthermore, it can be illustrated by a logic model, a visual map of this theory.[21] Stakeholders have unique experience and knowledge of the contextual factors, and how these may have influenced the implementation of an intervention.[22] Without creating an in-depth understanding of how complex interventions work and under what condition, treatment outcomes become difficult to explain and are poorly understood.[23] Currently, an explicit theorization of IIPT and its context is lacking in the pediatric pain-related disability intervention literature.[12]

Theory-based evaluation is an approach that may facilitate stakeholder engagement.[24] It aims to explain how and why programs work (or fail) in different contexts and for different stakeholders.[24] Logic analysis, a relatively new theory-based evaluation methodology, theorizes a program by mapping the links between the intervention components and the anticipated outcomes (i.e., program theory), highlights contextual influences, and evaluates the plausibility of the program theory against existing evidence and experiential knowledge.[25,26] Logic analysis uniqueness lies in its theoretical examination of the core intervention characteristics, which must be present to achieve the desired outcomes, and in its identification of the critical conditions necessary for implementation and production of these outcomes.[25] It is useful in uncovering causal pathways that may be discernible but not always perceptible.[27] Furthermore, it helps reduce uncertainty about the program theory inherent to complex interventions, provides a preliminary evaluation of the theoretical and empirical foundation of the intervention, and is valuable in recognizing the strengths, weaknesses and areas of improvement in the program theory.[25,26,28] Evaluations, using logic analysis, have yet to be applied in pediatric health or rehabilitation interventions, such as IIPT. Furthermore, some methodological gaps exist, including how to engage stakeholder.[29]

In an attempt to broaden the application of this evaluation approach in pediatric health and rehabilitation, this article aims to provide details on the logic analysis methodology including the strategies targeting stakeholder inclusion, the data collected, and the analyses used. To do so, we will present an example of its application in a preliminary evaluation of an implemented IIPT for youth with pain-related disability and share the findings assessing whether this IIPT was theoretically designed to achieve its desired outcomes.

2.METHODS

2.1. Study context

With funding from a large philanthropic donation, the IIPT in Western Canada was conceived in response to a growing number of youth presenting with pain-related disability. This cohort-based IIPT was influenced by the day-hospital model described by Logan and colleagues.[9,30] The six-hour daily IIPT operated five days per week in a day-hospital setting, and included individual, and group psychology, physical, family, occupational, art, music, and recreation therapies, as well as classroom time with a qualified teacher. Weekly nursing and physician consultations were also incorporated. All providers had specific training and experience working with youth with pain-related disability. Activities emphasized self-management knowledge acquisition and skill development, with a focus on restoring function and returning to age-appropriate activities. Treatment intensity and frequency, the disciplines involved, and the discharge timeframe were individualized and contingent on the achievement of patient identified goals established at treatment commencement. Participants received on average 119 hours of scheduled treatment, with an average length of stay of 5-weeks. Once implemented, an evaluation was requested by decision makers to determine the program's value, and to identify any improvement recommendations.

2.2. Study design

To determine if the core intervention components and critical contextual conditions were present to produce the desired outcomes, a direct logic analysis was used.[26,27,29] This evaluation was part of a larger participatory study for which ethical approval was obtained.

2.3. Participants

An expert panel of representatives from stakeholders involved in the treatment designed for youth with pain-related disability, was identified by facility leadership and recruited via email invitation. The 13-member panel consisted of five clinicians, a program coordinator, and healthcare manager, all of whom had experience (range 2-15 years) treating youth with pain and/or disability (e.g. pain-related disability, cerebral palsy). Also included were two teachers with over ten years of experience academically supporting youth with an array of physical and mental health conditions, two youth managing pain-related disability, and their parents. As no standards exist to guide the appropriate number of stakeholders to engage in the panel, guidance was gleaned from the consensus building literature, where a diverse group of 5 to 15 participants is recommended.[31-33]

2.4. Procedures

To foster an environment conducive to stakeholder engagement, several activities preceded the evaluation process. First, a charter of the role and responsibilities was created, and once agreed upon, was signed by all expert panel and research team members. Additionally, educational resources and training sessions associated with the logic analysis methodology were provided (e.g., logic model creation, scoping review processes). The 3-step logic analysis process described by Brousselle and Champagne[26] was then followed (see Figure 1).

Figure 1.
Association Between the Logic Analysis Steps and Results

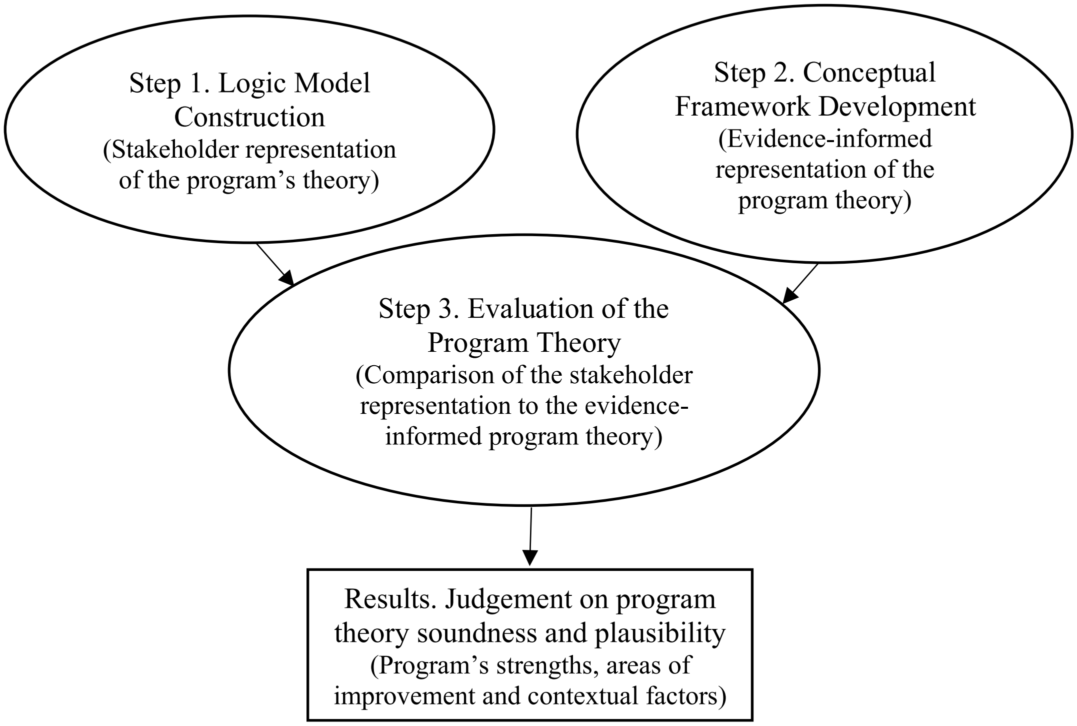

Table 1 provides a summary of the processes and procedures used in each sequential step. Additional details for each step are provided below.

Table 1.
Summary of Logic Analysis Steps, Processes and Procedures

Logic Model Methodology		
Steps	**Process**	**Procedures**
1. Logic Model Construction: Create a representation of the intervention's program theory and the links between resources, activities processes and anticipated outcome, using diverse data sources.[26,29]	Review of all historical program document	Deductive analysis using data extraction form based on logic model components by research team
	Expert panel electronic survey	Deductive analysis using data extraction form based on logic model components by research team
	Draft logic model created by research team using data gathered in documents and surveys	
	Group Discussion 1.Validate the primary program objective 2. Review and modify anticipated outcomes (short, medium and long-term) 3. Review and modify resources, activities, and processes 4.Review and modify reach and important contextual factors 5.Establish perceived links between components and anticipated outcomes 6.Achieve agreement on final logic model	Updates of the draft logic model after each meeting by research team Each subsequent draft returned to expert panel members for further discussion and detailing until agreement achieved
	Agreement reached by the expert panel members on the logic model representation	
2. Conceptual framework development: Identify and examine the evidence and document the mechanisms similar to those attributed to the intervention, providing a representative synthesis of the most recent knowledge in the most relevant and meaningful fields of research.[26,29]	Scoping Review Framework[36] i) Identify research question ii) Identifying relevant studies	Expert panel discussion conducted to identify and achieve agreement on the research question and the study inclusion and exclusion criteria
	iii) Study selection	Studies identified by the research team. Final selection presented to expert panel for approval
	iv) Charting the data	Data extracted and deductive analysis completed by research team using a form based on the

	v) Collating, summarizing, and reporting the results	logic model components and the primary program objective. Draft conceptual framework created by research team and presented to the expert panel for discussion and validation
	vi) Consultation	Expert panel consulted throughout the scoping review process and assisted in the re-interpretation of the findings in the context of IIPT
	Agreement reached by the expert panel on the interpretation of the conceptual framework	
3. Evaluating the program theory: Review of the logic model in light of the evidence contained in the conceptual framework, highlighting the intervention's strengths, weaknesses and recommendations for improvement.[26,29]	The logic model was compared to the evidence contained in the conceptual framework for convergence (i.e., IIPT strengths) and divergence (i.e., IIPT weaknesses and gaps)	A list of strengths, weaknesses and gaps of the IIPT were identified by the research team, IIPT improvement recommendations formulated, and presented to the expert panel for discussion. Following discussion, only improvement recommendations upon which consensus among the expert panel members was achieved were presented to the hospital leadership team.

2.4.1. **Step 1. Logic model construction**

In this first step of the 3-step logic analysis methodology, three data collection methods were used to generate the data required to construct a stakeholder representation of the logic model. These included: document analysis, stakeholders surveys and group discussions. All available historical documents (see Table 2 for full list) were analysed. A stakeholder survey was developed by the research team guided by the semi-structured interview question for constructing a logic model proposed by Gugiu and Rodrigez-Campos[34] (see Supplementary 1). Once developed, it was distributed electronically to the expert panel to supplement the document data. A form, founded on the logic model components and their definitions, was used for data extraction of the documents and a deductive analysis followed.[35] The same process was then repeated for the survey data. The extracted data from the document and the survey analysis were used to populate the various components (i.e., resources, research, activities, process, outcomes, contextual factors) of a draft logic model. Six group meetings with the expert panel, facilitated by a member of the research team, were held for the purpose of gathering missing information about logic model components and to clarify inconsistencies. Using various communication strategies (e.g., face-to-face, Facetime, telephone, and email), all expert panel member participated in all six discussions. More specifically, at the first meeting, the program goal and objectives were discussed. A dialogue updating each logic model component, the linkages between the components and the influential contextual features followed in the five subsequent meetings (see Table 1). New iterations of the logic model, based on expert panel feedback, were distributed between meetings, and the iterative process continued until agreement was reached. The sixth iteration was adopted.

2.4.2. **Step 2. Conceptual framework development**

The purpose of the conceptual framework, the second step of the 3-step logic analysis methodology, is to examine the intervention's main components, and determine if the optimal conditions have been assembled to achieve the desired outcomes. The aim is not to complete a systematic synthesis of the literature, but instead to create a representative synthesis of the most recent and meaningful evidence across various fields upon which the scientific validity of the logic model is examined.[26,29]. To develop the conceptual framework, the 6-stage scoping review process described by Levac and colleagues[34] was followed, and included the stages outlined in Table 1. A scoping review was the evidence synthesis method chosen as it summarizes a range of evidence in order to convey the breadth and depth of a field.[36] As suggested in logic analysis methodology, review studies were favoured.[26] Further details about each scoping review stage are provided below.

Identifying the research question: The research question identified by the expert panel was founded in the primary objective of the IIPT, as identified in Step 1 of the logic model methodology. More specifically, the following question guiding the search: "What components should an IIPT designed for youth with pain-related disability adopt to promote self-management, self-efficacy and participation in age-appropriate meaningful activities?"

Identifying relevant studies: Medline, CINAHL, and PsychInfo electronic databases were consulted using the following key words: chronic pain; pain-related disability; chronic conditions; disability; pediatric* or pediatric*, self-manag*; self-efficacy; participation. The target population was broadened to include youth with chronic conditions and disabilities for which pain is an important symptom, along with those with pain-related disability. It has been argued that youth with chronic conditions and disability share more comparable challenges than differences, and that disease specific orientations minimizes the efficiency with which solutions for these challenges can be identified.[37]

Study Selection: To be included, studies had to incorporate youth, aged 12–18 years (as per the age inclusion criteria of the evaluated IIPT), be related to self-management, self-efficacy, and/or participation in meaningful activity (i.e., leisure, recreation or activities that promote productivity (e.g. school, work)), and have a multi- or interdisciplinary focus. Retrieved titles and abstracts were screened by two reviewers for relevance. Entire manuscripts were then examined. Reference lists were inspected, yet no additional studies were identified. Once completed, original manuscripts cited in the review studies were scanned for additional relevant information.

Charting the data: A data extraction form (as per the categories outlined in Table 3) and procedures were developed and validated by the research team. Once consensus was achieved, the extraction process was completed by KH.

Collating, summarizing and reporting the results: Data were coded, categorised, themed and then culminated into a table format (see Table 4). An initial draft of the conceptual model was presented and discussed with the expert panel to explore the meaning, clarity and consistency of the thematic interpretation.

Consultation: As identified in Table 1, the expert panel members were involved in the scoping review in the initial three stages of the review, provided consultation throughout the process, and assisted in the re-interpretation of the data in the context of IIPT.

2.4.3. **Step 3. Evaluation of the program theory**

The third and final step of the logic analysis methodology consisted of comparing the constructed logic model with the developed conceptual framework.[26]. Moreover, this comparison examined the scientific validity of the program theory,[29] identified program gaps, and highlighted potential program improvements.[26] This step was completed collaboratively with the expert panel. It began with rereading of the program logic model, the appraisal of its components, and the examination of their relationship with those identified in the conceptual framework. Discrepancies and connections were initially identified by two members of the research team. Prior to the expert panel meeting, a compiled list of identified program strengths and weaknesses, copies of the logic model and the conceptual framework were distributed electronically to members. At the meeting, the discrepancies were debated in relations to the members' experiential knowledge. Recommendations upon which consensus was achieved, were then shared with hospital leadership.

3.FINDINGS

3.1. Logic model construction

3.1.1. **Program documents**

Fifteen key program documents and 13 stakeholder surveys were used to construct the draft logic model. Although the documents contained many important program details, when closely compared, inconsistencies emerged (see Table 2). Different program objectives were noted across documents. For example, stated goals/objectives focused on youth returning to age appropriate activities, or on the resumption of participation in social roles in various contexts (e.g. students at school); some specified goal achievement, despite pain, while others promised a decrease in pain over time.

Program resources, related to clinical disciplines, also varied. Program activities were described as a function of these disciplines, which, in some cases, varied depending on the cohort and the chosen service model (e.g. individual-focused versus group-based). Although program outcomes were present in select documents, they were not linked to the program activities or resources, and their relationships with the program objectives were unclear. The anticipated causal mechanisms between the activities and the expected program outcomes were unidentifiable. Finally, contextual factors were scant.

Table 2.

Document and Survey Analysis

		Program Logic Model Components							
Data Sources	Document Title (year)	Program Goals	Program Objectives	Reach	Eligibility	Program Resources	Program Activities	Program Outcomes	Program Context
Program Documents (n=15)	Initial Program Description (2013)	Not consistent	Absent	Not consistent	Absent	Not consistent	Absent	Not consistent	Absent
	Program Curricula (2015–2018)	Absent	Absent	Absent	Absent	Not consistent	Not consistent	Absent	Absent
	Program Goals and Objectives (2016)	Not consistent	Not consistent	Absent	Absent	Not complete	Not complete	Absent	Absent
	Program Implementation Evaluation (2016)	Not consistent	Not consistent	Not complete	Absent	Not complete	Not consistent	Not consistent	Absent
	Program Referral Guide (2017)	Absent	Absent	Absent	Complete for youth only	Not complete	Absent	Absent	Absent
	Program Information for Patients and Families (2016)	Not consistent	Not consistent	Not complete	Absent	Not consistent	Not complete	Absent	Absent
	General Information for Youth and Families (2016)	Not consistent	Absent	Not complete	Complete for youth & families	Not consistent	Not complete	Absent	Absent
	Overall judgment after	Not consistent	Not consistent	Complete for youth & families		Not consistent or complete	Not consistent	Not consistent	Absent

	document analysis			or complete		
Stakeholder surveys (n=13)	Survey questions	What are the goals & objectives of the IIPT?	Who should the program target? No further information required	Who and what help accomplish the objective(s) of the program?	What are the effects of the program?	Context Analysis
	Overall judgment after survey analysis	Still not consistent	Complete for youth & families	Not consistent	Priority setting	Not consistent
Stakeholder focus groups (n=6)	Focus group guiding questions	Is each component representative of the current program?				
	Overall judgment after focus groups	Complete	Expanded to include school personnel	Causal mechanisms clarified	Validated	Complete

3.1.2. **Expert panel surveys**

Survey responses assisted in further elaborating the logic model components, although discrepancies remained. A synthesis of the program resources, activities, causal mechanisms, and expected outcomes as perceived by the expert panel revealed that, similar to the document analysis, most expert panel members (i.e., clinicians) described program activities as a function of the disciplines (see Supplementary Material 2).

Furthermore, perceived mechanisms varied and were considered unique to each activity. The service model (i.e., group-vs. individual-based), the program intensity, as well as pre-program activities were viewed to be important contributors by some. Despite these added details, the relationship between the mechanisms and outcomes remained ambiguous (see Table 2). Contextual factors were also identified in the survey responses (see Supplementary Material 3). Internal factors were linked to program structure and team dynamics, while external factors related to building community-based partnerships and securing future program funding. Although these factors helped to further understand the context and the conditions deemed essential for success, questions remained.

3.1.3. **Group meetings**

At the first expert panel group meeting, a new program objective drafted and distributed prior to the meeting, was validated. The program objectives became “To provide youth with pain-related disability and their parents the knowledge, skills, and tools to self-manage their pain, build their self-efficacy, and promote their participation in meaningful activities, despite their pain”. Furthermore, based on expert panel discourse as per the member below, the program reach was extended to include school and community personnel.

> “*Our target population should include parents and the school, but also others in their community environment*” (Clinician 1).

Some activities and processes were omitted, while others were added, or further detailed. Program activities which provided support, most valued by parents and youth were underscored.

> “*I think two things are absolutely fundamental in this program: the education group sessions and the connections you have with the other participants*” (Youth 2).

Youth also recognized activities that should be added to further improve their outcomes. Such activities focused on self-advocacy, and the need to facilitate their transition back to their community following the program. The expected outcomes were adjusted and further elucidated based on panel member's experience.

> "*In terms of long-term outcomes, it should be how much knowledge is retained. Because if you can refine the application of that knowledge, and once you build routines, you've found a way to make it work for you*" (Youth 1).

Finally, contextual factors believed to be essential for program success were discussed, and agreement was reached. These factors were associated with the pre-program screening, access to specialized health human resources, and participant characteristics. Figure 2 illustrates the final agreed upon logic model.

3.2. Development of the Conceptual Framework

3.2.1. Scoping review results

Table 3 outlines the details of the 19 articles selected for the conceptual framework development and the deductive framework used to extract the data. All population samples included children and adolescents with a variety of disabling conditions for which pain is an important symptom.

3.2.2. **Conceptual framework summary:** Table 4 synthesizes the salient evidence of the conceptual framework, its relationship with both the logic model components, and the themes supportive of the program's key objectives. Further description is provided below.

***Promoting Self-Management**:* Self-management, a person's ability to acquire and apply the skills and knowledge to manage their symptoms, is learned with the support of family, community members (e.g. friends, peers, teachers, coaches), and healthcare professionals.[38] Chronic conditions are experienced within the perspective of everyday life contexts (i.e. peers, family, school, occupation, leisure, community)[38,39]. Although medical management is important, emotional coping and role (social participation, occupation) management should also be considered.[40]

Figure 2.

Expert Panel Agreed Upon Logic Model

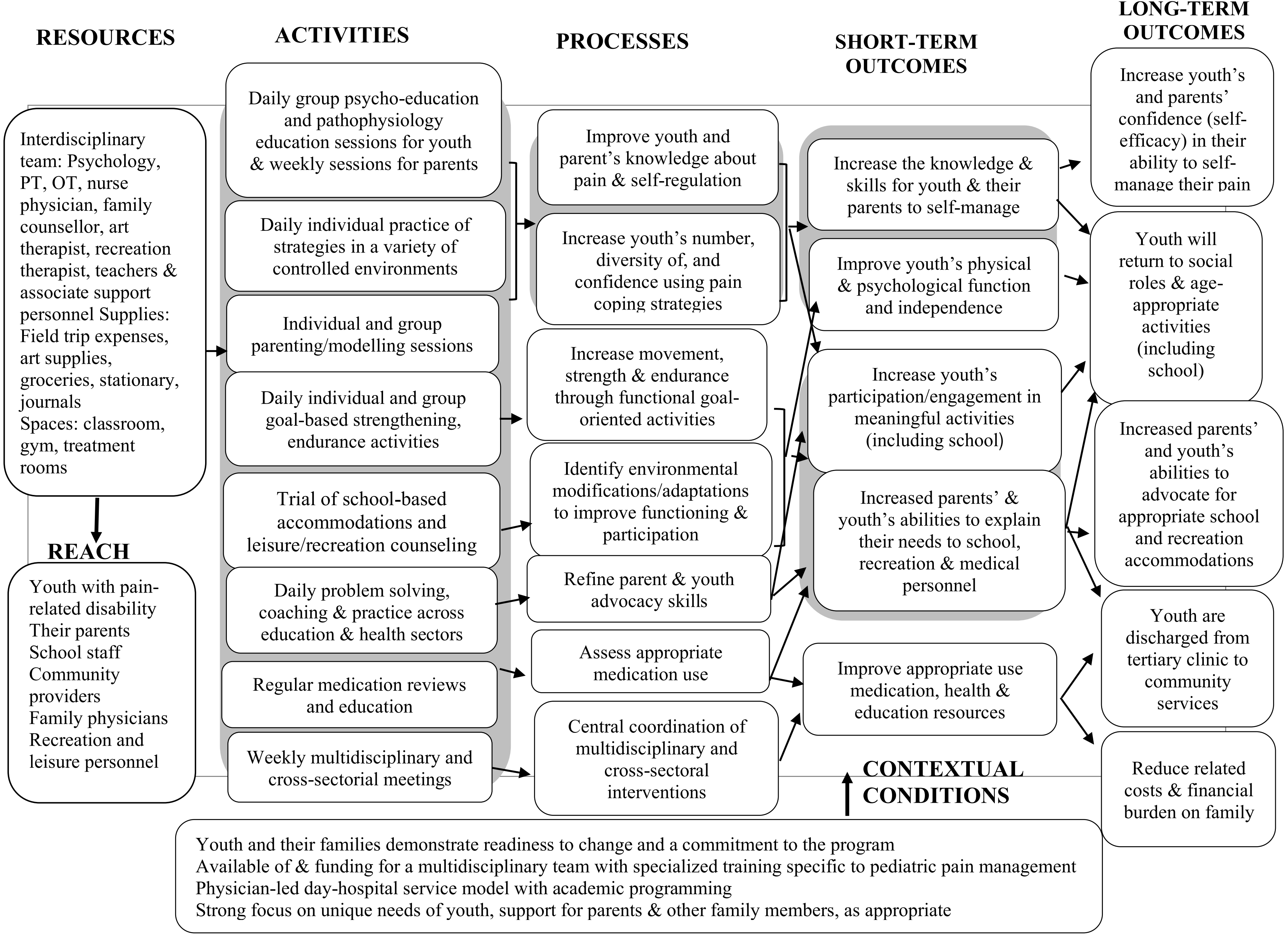

Table 3.

Summary of Studies Retained for Conceptual Framework

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
Self-management Interventions (SMI)						
Stinson *et al.* (2008)[41]	Canada	Systematic Review	To critically appraise the evidence on effectiveness of internet SMI on health outcomes in youth with chronic conditions.	Children and adolescents (6–18 years). Asthma, recurrent pain, encopresis, traumatic brain injury, obesity	7 randomized control trials, 1 pilot randomized control trail, and 1 quasi-experimental study	Internet-based SMI have demonstrated some evidence improving symptoms and disease self-management yet are inconclusive in whether as effective as in-person individualized or group interventions.
Lindsay *et al.* (2011)[40]	Canada	Integrative Review	To synthesise findings from empirical studies examining influential factors of adolescents' self-management of chronic illness.	Adolescents and young adults (12–20 years). Diabetes, asthma, spina bifida, inflammatory bowel disease, juvenile idiopathic arthritis	34 studies, 16 qualitative, 14 quantitative and 4 mixed methods designs.	Psychosocial factors (e.g. self-efficacy), parent involvement, knowledge about illness are important facilitators. Youth self-management skills should be assessed, along with their social and developmental context to identify supports.
Lindsay *et al.* (2014)[39]	Canada	Systematic Review	To systematically assess the effectiveness of SMI for school-aged children with physical disabilities.	Adolescents and young adults (13–24 years) Children and adolescents (2–18 years) Spina bifida, juvenile rheumatoid arthritis, juvenile idiopathic arthritis.	2 randomized control trials; 4 before and after designs	Intervention components should include knowledge about condition, medication management, psycho-social factors (e.g. self-efficacy). Parental involvement can be a barrier to self-management and should be carefully assessed.

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
Sattoe *et al.* (2015)38	Netherlands	Systematic Review	To provide a systematic overview of the SMI for young people with chronic conditions.	Children (7–11 years) and adolescents (12–18 years) Asthma, diabetes, cancer, chronic fatigue, chronic pain, chronic respiratory conditions, inflammatory bowel disease, juvenile fibromyalgia, juvenile idiopathic arthritis, migraine, physical disabilities, sickle cell	45 randomized control trials, 29 cohort studies, 3 cross-sectional studies, 3 qualitative, 5 mixed methods, 1 case-study, 26 pilot evaluations	Role and emotional management should be included in SMI, along with medical management. Parents can either facilitate or hinder youth self-management. Experiential learning, peer-learning for others, and mastery experiences strategies are appropriate pediatric SMI. Developmental factors need to be considered.
Bal *et al.* (2016)	Netherlands	Systematic Review	To systematically explore the effectiveness and effective components of SMI.	Children to young adults (7–25 years) Asthma, diabetes, cystic fibrosis, cancer, HIV, sickle cell, spina bifida, hemophilia, juvenile fibromyalgia	42 randomized control trials	SMI should focus on medical, emotional, and role management in the context of youth's daily lives. Peer support stimulates self-efficacy. Online peer-support could improve self-efficacy, problem-solving and coping behaviors.
Lindsay, Kolne, Cagliostro (2018)45	Canada	Systematic Review	Synthesis and review literature on the impact of electronic mentoring for children with disabilities.	Children to young adults (12 to 26 years). Rheumatic disease, juvenile arthritis, cerebral palsy, spina bifida, muscular dystrophy, pediatric transplant, visual impairments, chronic pain.	3 RCTs, 7 surveys, 1 case study, 1 feasibility study	Electronic mentoring is effective for children and youth with disabilities in improving career decision-making, self-determination, self-management, self-confidence, self-advocacy, social skills, attitude towards disability, and coping with daily life.

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
Self-Efficacy						
Cramm *et al.* (2012)[56]	Netherlands	Cross-sectional study	To investigate the influence on general self-efficacy perceived by adolescents with chronic conditions and parents on quality of life.	Adolescents, and young adults (12–25 year) & their parents Diabetes, juvenile rheumatoid arthritis, cystic fibrosis, urology conditions and neuromuscular disorders	Not applicable	Interventions aimed at improving general self-efficacy should include activities that seek to enhance confidence and the ability to deal effectively with difficult and unexpected events.
Johnson *et al.* (2015)[44]	United States (US)	Cross-sectional study	To determine the preferred methods for health information among youths with chronic conditions and their relationship to health care transition readiness, self-efficacy and medication adherence.	Children and adolescents (6–16 years) Diabetes, musculoskeletal conditions, cerebral palsy, heart disease, neurological and gastrointestinal condition	Not applicable	Youth with chronic conditions receive their health information from physicians/nurses, parents/family, and the internet. A range of health information should be considered to include those that deliver it directly to the patient, the family/parent, including the internet, allowing youth to select their preferred method.
Molter & Abrahamson (2015)[57]	USA	Literature Review	To investigate the relationship among self-efficacy, transition and health outcomes.	Children, adolescents and adults (6–55 years) Sickle cell	20 studies of various unspecified design	Knowledge of condition, body awareness, and spirituality are factors that affect self-efficacy. Journaling, self-awareness, scripture reading, and prayer activities can increase feelings of self-efficacy. Experiences of acting independently and developing patient-health provider partnerships are important. Education, counselling, and advocacy interventions to the broader

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
						public could be used to decrease stigmatization.
Kalapurakkel *et al.* (2015)[42]	US	Cross-sectional study	To examine pain self-efficacy and pain acceptance in relation to functioning in pediatric headache patients.	Children and adolescents (8–17 years); Headache	Not applicable	Higher levels of self-efficacy are associated with improved school functioning, fewer depressive symptoms, and lower disability levels, higher self-esteem and fewer somatic symptoms.
Tomlinson *et al* (2017)[58]	Canada	Literature Commentary	To examine the resilience mechanism of pain self-efficacy.	Children and adolescents	Not specified	Exposure to and mastery of feared activities reinforces self-efficacy. Generalizing prior successes that highlight mastery and increase confidence can enhance pain-self efficacy. Mindfulness and biofeedback are also helpful modalities The identification of valued goals and utilizing graded exposure techniques to previously avoided activities promote self-efficacy.
Participation						
Pinquart & Teubet (2011)[59]	Germany	Meta-analysis	To compare the levels of academic, physical, and social functioning of children and adolescents with chronic physical diseases with those of healthy peers.	Children and adolescents (under the age of 18 years) Arthritis, asthma, cancer, chronic fatigue, cystic fibrosis, cerebral palsy, inflammatory bowel disease, headaches, diabetes,	954 studies designed not specified	Sports and leisure activity counselling should be available to guide these youth. Teachers and coaches should promote participation in sports to improve physical functioning. School functioning can be improved with school accommodations.

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
				hemophilia, epilepsy, sickle cell, spina bifida		Group social skills training provide youth with strategies to deal with teasing and bullying.
Anaby *et al.* (2013)[46]	Canada	Scoping Review	To identify and analyze research evidence regarding the effect of the environment on community participation of children with disabilities.	Children, adolescents and young adults (5–21 years). Cerebral palsy, physical disabilities (with restricted mobility due to neurological or musculoskeletal disorders), acquired brain injury, autism spectrum disorder, Down Syndrome	31 studies; 17 qualitative, 10 qualitative, review 3, 1 mixed-method design	Negative attitudes within the communities can be a barrier to participation. Parental involvement and advocacy can influence on social functioning, participation and friendship development. Peers, and teacher, service provider support fosters participation. Parental over-protectiveness and stress can limit participation. Parental education about recreation activities and advocacy supports participation.
Adair *et al.* (2015)[48]	Australia	Systematic review	To critically appraise studies aimed at improving participation outcomes of children with disabilities.	Children and adolescents with disabilities (5–18 years) such as cerebral palsy, developmental coordination disorder, autism spectrum disorder, arthrogryposis, intellectual disabilities	7 randomized control or non-randomized trials	Tailored programs using both individual and group-based approaches can enhance participation. Coaching approaches focused on mutually agreed upon goals are effective. Practice of desired behaviors in a social context is proven useful.
Forgeron *et al.* (2018)[60]	Canada	Systematic Review	To identify the psychosocial interventions found to be most promising in their effectiveness in	Children and adolescents (5–18 years) with diabetes, epilepsy/seizures, cerebral palsy, spina	13 studies; 10 non-randomized control trials, 3 randomized control trials	Most improvements in social functioning stemmed from interventions that focused on a broad range of social skill

Authors & publication year	Country	Study Design	Study Aim	Population Characteristics	Feature of Included Studies	Key Findings
			improving social functioning outcomes of children and adolescents with a wide range of chronic physical health conditions.	bifida, inflammatory bowel disease, burn scaring, chronic respiratory condition		development rather than solely on communication about condition with peers. Interventions that consisted of more than one session targeting social functioning were more promising. A paucity of evidence exists on effective interventions.
Jones *et al.* (2018)[49]	Canada	Narrative review	To review selected studies that have made an impact on the field of school functioning in children and adolescents with chronic pain.	Children and adolescents (8–18 years) with chronic pain such as abdominal, myofascial, neuropathic, limb, back pain, headache	13 non-randomized control trials.	Evidence suggests that psychological factors (depression and anxiety), social factors (peer-relationships, perception of teachers support, parent protectiveness), physiological factors (sleep disturbance), and cognitive factors (self-efficacy, memory and attention deficits may interact to influence school functioning.
Ideal Context						
Stahlschmidt *et al.* (2016)[12]	Germany	Review	To present an international perspective on the structure and components of pain rehabilitation programs worldwide.	9 different programs from 4 different countries	15 descriptive or non-randomized studies.	Specialized rehabilitation programs for disabling chronic pain conditions worldwide have similar admission criteria, structure, and therapeutic orientation. Differences in exclusion criteria impede program comparability.
Mirò *et al.* (2017)[61]	Spain	Cross-sectional	To identify the features current		Not applicable	Staff should be multi-disciplinary, with research and

Authors & publication year	**Country**	**Study Design**	**Study Aim**	**Population Characteristics**	**Feature of Included Studies**	**Key Findings**
		study design using surveys	chronic pain programs and describe the feature required to achieve an ideal state.	136 pediatric pain experts located in 12 different countries		formal specialty training available. A wide variety of treatment options should be offered and publicly funded.
Harrison *et al.* (2019)[7]	USA, Belgium, Stockholm	Review	To present and overview of rehabilitation interventions for children and adolescents with chronic pain and to inform clinicians on the innovative treatment delivery and patient outcomes.	Not applicable	Systematic review, meta-analyses, clinical trials with sample >20, clearly describing the intervention	Patients who have been unsuccessful at outpatient treatment are targeted. Must include three or more disciplines housed within the same facility (e.g., pain specialist, psychologist, and physical therapist) who work in an integrated manner to provide treatment. Patient must participate in exercise-based therapy and psychological interventions. The aim is to improve function across domains. Variability exists in program structure, organization, frequency of treatment across disciplines, treatment model (inpatients vs. day-hospital), and length of stay.

Effective medical self-management is contingent on youth acquiring independence, knowledge and skills.[41] Psychoeducation and skills training are the cornerstones of self-management programs.[7,41] Parental education and parent-to-parent support are effective in addressing the gradual shift of self-management responsibilities to youth.[39] Support from social networks, including peers, has also emerged as a facilitator.[38-41] Many additional effective activities and promising emerging approaches are presented in the conceptual framework (see Table 4).

Table 4.

Conceptual Framework

	Program Objectives		
Logic Model Components	**Self-management interventions**	**Building self-efficacy**	**Fostering participation**
Program goals and objectives	Role, emotional and medical self-management relative to developmental expectations should be integrated within youth's daily life and relevant social contexts.[38,39,55]		
Program reach and eligibility	Parent involvement should be carefully assessed.[38,39,40] Education should extend beyond youth with chronic conditions and parents, to include peers, teachers.[39,47]		Education initiatives should target peers, classmates, teachers, and community leaders (e.g. coaches).[46,49,59]
Program activities	Psychoeducation, combining information and skills training is the focus of self-management interventions.[38,55] Parent education, parent-to-parent support, and using parent coaching approaches are effective in fostering independence in youth self-management.[39] Experiential approaches, varying delivery methods (group, individualized, internet-based), peers learning opportunities, and skill mastery experiences should be provided.[38-41] Communication, assertiveness and advocacy training is a need identified	Activities that build independence, life and leadership skills should be promoted.[56] Opportunities for youth to create their own patient-professional relationships can be enriching.[44,56] Self-awareness (e.g. journaling), self-directed learning (e.g. web-based resources), and spiritual program activities, using a variety of learning methods and mediums (e.g. health professionals, parents, internet-based modules) should be included.[44,45,57] Biofeedback, self-regulation, relaxation, mindfulness, cognitive behavioural	Individualized and group-based interventions are effective when combined.[48] Physical and leisure activity selection should be guided by mutually agreed upon participation goals and identified though coaching approaches.[48] Training parents and youth on how to advocate for social inclusion and how to adapt and modify the activity and environment are effective strategies to minimize participation barriers.[46] Sport and leisure activity counselling and social

	by youth to promote shared decision-making with professionals.[39,41] Opportunities for youth to create their own patient-professional relationships can be enriching.[41] Peer-to-peer learning and mentoring is an emerging model showing promise.[45]	therapy, value-based goal identification nurture self-efficacy.[58] Successful accomplishment of assigned tasks and generalization of prior successes, and graded exposure to fear-eliciting activities are also beneficial.[58]	skills training should be available.[48] Coaching on how to communicate about the condition and the supports required may be beneficial for this population in peer and school settings.[46,48,49] More complex age-specific in-person sessions expanding social skills training to peer interactions, conflicts (e.g. bullying), and intimate friendships may also be beneficial for older adolescents.[59,60]
Program outcomes	Increased knowledge and skills in problem-solving, decision-making and advocacy have been described.[38] Improvements in self-efficacy, psychosocial well-being, and family functioning, along with reduction in social isolation, school absenteeism and pain have been demonstrated.[41] Reduced family and parent burden, reducing healthcare utilization, and improving overall health outcomes and quality of life have also been reported.[38]	Benefits to physical, emotional and school functioning have been recognized.42 Self-efficacy has been identified as a key contributor to chronic disease self-management, to promoting of long-term behavior change, to improving the appropriateness of health care utilization practices, and to enhancing health quality of life.[43]	Participation improved academic performance, social interactions, mental & physical health, and helps develop life purpose and meaning.[46,62]
Creating the ideal context			
Program Resources	Program should be publicly funded.[61] A variety of health disciplines with specific training and expertise in pediatric pain.[7,12,61] A clinical and research training role, along with a public education (e.g. school personnel) and advocacy mandate should be fulfilled by the program.[61] Youth with variety of pain conditions, regardless of the type and origin, and their parents should be targeted.[7,12,61]		

Building Self-Efficacy: Self-efficacy, defined as a youth's confidence in their ability to function effectively while in pain,[42] is critical to self-management, to appropriate healthcare utilization practices, and to enhancing health-related quality of life.[43] Effective activities for building self-efficacy were highlighted in the framework (see Table 4). Appealing to youth's preferred information seeking practices is considered pivotal to the process, with web- and application-based resources holding promise for this population.[44,45]

***Enhancing Participation in Meaningful Activities**:* Participation, defined as one's involvement in life situations (e.g., education, employment, recreation, and community living), is an important pediatric rehabilitation outcome.[46,47] Social supports (e.g., school personnel, peers) are important facilitators to achieving participation.[46] Moreover, effectively communicating about one's condition and requesting the supports required within various contexts (e.g., in school, with peers) are important skills for increasing participation.[46,48,49] Other associated activities are presented in Table 4.

Creating the Ideal Context: Contextual conditions essential for program success were also found in the literature. Admission criteria across IIPT programs worldwide are similar, of which, pain impacting function, and youth and parent allegiance to self-management approach dominate.[7,12] Other contextual factors are highlighted in the conceptual framework (see Table 4).

3.3. **Evaluating the intervention theory**

When detailed IIPT components, their links and anticipated outcomes were systematically compared to the conceptual framework, generally speaking, the scientific evidence supported the program theory plausibility. Furthermore, interconnectivity between the three IIPT program objectives was illustrated. Below the IIPT program strengths are presented, followed by recommendations for improvements.

3.3.1. **IIPT strengths:** Regarding refining the self-management intervention for youth, our IIPT intervention aligned well with the evidence contained in the conceptual framework. As per the evidence, psychoeducation was acknowledged as a valued tenet of the program. Many teaching approaches (e.g. peer learning) recognized as effective were incorporated in the program group activities and included opportunities for practice in real-life environments (e.g. classroom, community field trips). These peer learning moments were highly valued by expert panel parent and youth members and recognized as pivotal in achieving positive outcomes. However, a need to incorporate additional community-focused transition opportunities was underscored by both parents and youth, and by the scientific-evidence reviewed.

In relation to building self-efficacy, our IIPT program also performed well against the scientific evidence of the conceptual framework. In addition to family counselling and individual psychological interventions, many targeted activities identified as beneficial (e.g. self-awareness, self-reflection) in the evidence were already incorporated in the IIPT. Moreover, the inclusion of community-based activities (e.g. field trip, leisure planning) in the IIPT, designed to foster problem-solving, decision-making, and self-management skills and their generalization to real life, were strongly supported by the scientific evidence, and the experiential knowledge of the youth expert panel members. However, youth panel members also requested even further guidance on the safe return to such activities post-discharge.

With respect to fostering participation in meaningful activity, the IIPT included several components deemed effective in the evidence. Sports, recreation and leisure counseling, advocacy education, and youth and parental training in activity and environment modification were activities already incorporated in the IIPT and for which conceptual framework scientific support existed. Transition meetings with school personnel, part of the current program discharge process, were acknowledged by youth and parent expert panel members as an opportunity to foster collaboration with teachers, which coincided with the conceptual framework evidence. Youth expert panel members not only valued these meetings, they requested additional tools to further facilitate their ongoing advocacy initiatives in this context post-discharge.

Finally, concerning creating an ideal context to achieve the anticipated program outcomes the IIPT fulfilled many of the pre-requisite conditions identified in the conceptual framework. When compared, the IIPT admission criteria, key program features and team memberships shared many similarities with studies included in the conceptual framework.

3.3.2. **IIPT Improvements:** When comparing the logic model to the conceptual framework, three main areas of improvement associated with the reach, activities, and processes of the evaluated IIPT were presented to the expert panel for consideration. First, the importance of adopting a developmental lens to the acquisition of knowledge and skills aligned with the expectations of different age groups was recognized. Although the IIPT integrates school-based, sports, leisure and recreation activities, the evidence supported incorporating sessions addressing topics such as vocation and work, independent living (e.g. housing), and the management of intimate relationships, for older youth (i.e. 16–18 years). Youth expert panel members also advocated for post-program support associated with the quickly changing responsibilities and mounting societal expectations inherent to this age group. To incorporate this empirical and experiential knowledge, the inclusion of developmental goals to the already existing goal setting process was suggested. The conceptual framework also highlighted the need to expand the reach of the program to include youth's broader social networks. Enhancing peer support through educating classmates and school personnel on pain-related disability and on how to support to those suffering from this condition was recommended. Expert panel clinicians, youth, and parents' members alike, acknowledged this missing pillar in the IIPT. Finally, the conceptual framework highlighted emerging evidence supporting the use of the web- and application technology. Although the technological trials have been limited to one or two of the IIPT components (e.g., cognitive-behavioural therapy), these technologies hold promise for families for whom access to trained professionals, distance from care facilities, and long waiting times are major barriers. However, web-based expansion of any of our program component was not acknowledged or recognized as a gap by our expert panel. Upon review of these IIPT improvement recommendations and in light of the organisational constraints raised by the health manager expert panel member, the panel provided the following recommendations to the hospital leadership team: 1) expand information provided to older adolescents to incorporate vocation, work, independent living, and relationships; 2) incorporate self-management goals tailored to the developmental spectrum; and 3) broaden the psychoeducation to involve peers and school personnel.

4. DISCUSSION

The purpose of this article was to detail the logic analysis methodology and to share the findings of the program theory testing of an IIPT using this approach. As a collaborative IKT approach, this evaluation methodology proved helpful in many ways. First, logic analysis provided an opportunity to create a shared understanding of the complexity of IIPT among stakeholders, highlighting previously unidentified intervention and context interactive mechanisms. Stakeholder engagement was critical in ensuring the accuracy, validity and the integrity of the implemented IIPT description. Furthermore, stakeholders' reflections, in particular those of youth and their parents, were crucial in establishing those causal mechanisms and activities most valued. Through this value-based process, mechanisms were identified where an interaction between intervention components and the context occurred. Complex interventions, like IIPT, are built on a number of components, which may be dependent and interdependent, and where interactions between the intervention and the context exist.[50] It has been previously suggested that the effectiveness of these interventions may rest in the interaction between the intervention components (e.g., psychoeducation) and the context (e.g., group milieu, staff interactions, real life situation). To date, the explorations of these interactive intervention-context mechanisms had rare.[12] The logic analysis methodology presented a standardized approach which not only helped theorize this complex intervention, but also assisted in acknowledging intervention-context interactive mechanisms (e.g., psychoeducation in peer-supported environments), as a result of the engagement of the target population.

Secondly, the logic analysis process assisted in unveiling health professionals' beliefs about the causal mechanisms thought to contribute to the achievement of the anticipated outcomes. It provided an opportunity to weigh these assumptions against two important sources of validity: scientific evidence and youth and caregivers' experiential knowledge and values. More importantly, both these sources failed to confirm clinicians' assumptions of discipline and activity specific mechanisms. In evaluation research, it has been recognized that the mechanisms of change are not so much linked to the interventions per se, but instead to the participants reasoning and responses generated by the activity, and the context which lead to the outcomes of interest.[51] Further exploration of youth and their parents' reasoning and responses to IIPT activities and the program as a whole, and within different daily contexts (e.g., school, home), may represent valuable new avenues of research in this field.

Thirdly, the conceptual framework used a recognized evidence review method and presented a synthesis of current evidence to expert panel members. This evidence-informed framework stimulating practice reflection and comparison with experiential knowledge and values. As such, logic analysis presented an innovative way to integrate IKT, addressing the persisting knowledge-to-practice gap in pediatric rehabilitation. Discovering scientific evidence to support many of the causal mechanisms of the evaluated program and gaining awareness of those components most valued by youth and their families were noted by clinician expert panel members to be most enlightening part of this collaborative process. Whether this reflective process and increased awareness of the evidence prompt behaviour and practice change in clinicians will require further investigation.

Engaging stakeholders in logic analysis has been previously recommended.[29] Particularly unique in our application of this methodology was the involvement of patients (i.e. youth with pain-related disability) and their caregivers. The premise of engaging patients beyond the level of research subjects reflects a growing desire for more ethical, democratic and moral practices.[52] However, the absence of parent and youth voices in the published evaluation of pediatric pain rehabilitation interventions, including IIPT, is a gap recognized by many.[17,19,38,53] In our evaluation, their engagement resulted in identifying youth and their parents' program expectations, as well as recognizing their ongoing challenges following program discharge. Also noteworthy was the causal mechanisms identified by youth and parent expert panel members (i.e. their experiential knowledge) was acknowledged in the scientific evidence incorporated in the conceptual framework. Building this shared understanding within the expert panel proved valuable in later prioritizing program refinements. Furthermore, organisation constraints highlighted by the health manager provided important insight in selecting recommendations that were feasible to implement within the program context.

Specific evidence-informed practices and strategies to foster stakeholder engagement were incorporated into this logic analysis methodology. Targeted activities included (a) choosing a sample of parents and youth who have used the services,[19] (b) creating clearly defined roles, responsibilities, and expectations for the expert panel members and research team,[54] (c) engaging stakeholders early and throughout in the evaluation process,[16,54] (d) providing training on evaluation principles,[53,54] (e) ensuring regular interactions with panel to foster mutual understanding amongst members,[15] (f) embracing a variety of communication technologies to promote participation and discussion,[19] and (g) distributing discussion materials prior to the meeting.[15]

Despite our best efforts, this study should be interpreted with some limitations in mind. First, the non-equivalent numbers in each of our stakeholder groups on our expert panel may have biased our results and may have created a power imbalance in favor of clinicians in the group discussions. A variety of data collection methods were however used, incorporating anonymous strategies (e.g. electronic surveys) to ensure authentic perspective were expressed by expert panel member, decreasing social desirability biases. Second, despite expansive recruitment efforts, limited diversity was evident in our expert panel membership. Although youth and parents were representative of the population using this program, other recruitment strategies should be explored if this methodology is expanded to interventions servicing a more cultural and ethnic diverse population. Third, the inclusion of expert panel members into the conceptual framework development could be enhanced. In previously described logic analysis processes, the conceptual framework phase was completed by the evaluator only. Although the expert panel members were included in many stages of the conceptual framework construction, incorporating stakeholders in the data extraction and theming processes could be added if appropriate oversight was provided.

Theory-based evaluation provided an opportunity to further detail the causal path of IIPT rehabilitation intervention, lead to a better understanding of these interventions and evaluated the plausibility of the program theory in achieving its identified outcomes. Stakeholders were implicit to this process. The methods presented in this article, where scientific and experiential knowledge were weighed in a similar manner, provided a collaborative, pragmatic and realistic approach, representative of the clinical environment in which most health and social providers conduct evaluation. Engaging stakeholders, including parents and youth, in the logic analysis, represents a catalyst for better understanding complex of pediatric pain rehabilitation interventions, such as IIPT, and their evaluations. Furthermore, it represents a novel IKT method to narrow the ongoing knowledge-to-practice gap existent in the field.

SUPPLEMENTARY MATERIALS

Supplementary Material 1.

Expert Panel Survey

Purpose: This questionnaire includes 8 short answer questions that will assist in providing us with some information about your thoughts on the current intensive rehabilitation program to help us better understand the program what it is designed to achieve, how it achieves it and what are the things that have or could influence it. The information will be used to create a visual representation of the program (also known as a logic model) that will be discussed at our next meeting.

Instructions: Carefully read the question and fill in the space below. Please note there are no wrong answers and all information is welcomed.

Q1. According to you, what is the objective(s) of the intensive pain rehabilitation program? Please give examples on how that objective could be observed.

Q2. According to you, what are the activities of the intensive rehabilitation program that help accomplish this objective? Please be as specific as possible in naming the activity components.

Q3. How do you think these activities contribute in accomplishing the objective(s) of the program? Please describe the links you think exist between what the activities do and why you would expect a specific outcome to result.

Q.4 According to you, what are the strengths of the intensive pain rehabilitation program? *Strengths are aspects of the program you are pleased with and that currently contribute to the quality of the services offered.*

Q.5 According to you, what are the weaknesses of the intensive pain rehabilitation program? *Weaknesses are aspects of the program that need to be improved to increase the quality of the services provides.*

Q6. According to you, what opportunities exist that could positively improve the quality of services of the intensive pain rehabilitation program? *Opportunities are elements within the hospital or in the community at large that favor the development of the intensive pain rehabilitation program and can help to improve the quality of the services provided.*

Q.7 According to you, what are the treats that could negatively impact the quality of service provided by the intensive pain rehabilitation program?
Threats are elements at the hospital or in the community at large that can hinder the quality of service of the intensive pain rehabilitation.

Q.8 Is there anything else you wish to tell us about the program that would help us better understand it?

Thank you for your thoughts!

Supplementary Material 2.

Table S1. Expert Panel Themes about Causal Mechanisms and Program Structure Assumptions

Resources	**Activities**	**Mechanisms**
Physiotherapy	Behavioral activation or physical re-activation Improve physical endurance, tolerance, strength, flexibility and posture Goal-oriented physical activity and home programming Graded increase in physical activity Flare plan development Home program activities	Increases activity and movement Reduces fear avoidance Improves fitness, endurance, and strength Fosters reconditioning Dampens pain signals in the brain Facilitates adherence Promotes improve coping Increases energy reserves Sets expectations for ongoing practice of learning and skills Promotes the establishment of a routine
Psychology	Active pain management and coping strategies Education and training on thoughts, feelings, behaviors, acceptance Individual and group coaching in self-management Sleep education Flare plan development Focus on mindfulness	Improves youth's understanding of their pain at a physiological level Reduces fear, anxiety and depression Increases self-efficacy Teaches youth and their family to think and talk about pain differently Promotes functioning and participation, despite pain Enhances self-regulation
Medicine	Provision of rational for rehabilitation approach & medical closure Medication management Progress updates	Increases youth and parents feeling in control Improves treatment adherence Improves buy-in to rehabilitation approach
Family Therapy	Group and therapy session to coach parents on how to support their children Parental pain education Parental support Identification and management of family issues	Empowers family to support their child Creates a supportive family and transition environment for youth following program Keeps parents informed about what their child is learning
Occupational Therapy	Ergonomic assessment and recommendations Adaptation to activities of daily living Sleep education Pacing	Supports youth in daily activities through to transition back into the community Facilitates an alternate coping strategy Promotes motivation, relaxation or distraction a needed

Music Therapy	Development of music play-lists	Self-expression
		Promotes an alternate coping strategy
		Self-expression of chronic pain
Art Therapy	Alternative to express emotions & chronic pain experience	
		Provides realistic contexts in which to apply self- management knowledge and skills
Recreation	Graded increase in leisure activity and sports	Promotes transition to community activities post-program
	Exploration of alternative sport and leisure activities in community	
		Supports families
Nursing	Parent and adolescent support	
	Medication management	Enables youth to maintain school some level of academic expectations
Academics/School	Assessment of learning needs	Encourages a routine and normalcy
		Assists in identifying academic and social school-based challenges
		Eases transition back to community school setting
		Decreases isolation
		Teaches support of others and of self
Program Structures	Group-based service delivery	Validation of the experience by others
		Creation of a support network
		Peer discussions and learning
		Facilitates empathy towards others
	Intense duration	Allows time for the immediate application of learnings and reinforcement
		Concentrates learning and practice
	Signing of daily attendance expectations and pre-program contract	Sets future expectations for daily school attendance

Supplementary Material 3.

Table S2. Expert Panel Survey Response Context Analysis

	Strengths	Weaknesses	Opportunities	Threats
Program structures & organization	Program intensity • Re-establishes a daily routine • Focused learning and practice on self-management • Less disruptive to family life Decreases stress & anxiety in youth as removed from some environments (e.g. school) Encourages fun Tailored for youth's learning capacity Development of support network for youth and parents Integration of academic component	Lack of a standardized intake process and clear criteria Difficulty recruiting patient Time demands and stress on youth, family and staff • Loss important learning and social opportunities Complexity of intervention Length of program Lack of coordination of program components • Need for clear transition early in the program • Stronger curriculum throughout program Need for additional components/time • Parent education about accommodations • More academic time	Streamlining and/or standardization of program Pan-Canadian recruitment & marketing Integration of other hospital services	Not offering program to all youth who could benefit Competition with other programs
Team members and dynamic	Caring, positive, expert staff Wholistic & comprehensive approach Multidisciplinary approach	Team dynamics and conflict • Communication • Lack of a shared philosophy on program components Too many professionals involved Lack of capacity building opportunities for program staff • Cross-coverage within and between disciplines • Development of additional expertise		Loss of key staff resources
Building partnerships			Development of community partnership Build in volunteer opportunities into program Expand space available, Creation of education and training for personnel	Lack of society knowledge and recognition of pediatric chronic pain
Program funding				Future funding Perceived as expensive

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Article 2

Preamble

What really matters in pediatric chronic pain rehabilitation? Results of a multi-stakeholder nominal group technique study.

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Karen Hurtubise's contribution to this manuscript included its conceptualization, the literature review, the data analysis, creation of the thematic map, the presentation of the findings and the writing of this manuscript. The co-authors (Astrid Brousselle, Melanie Noel and Chantal Camden) supervised and provided guidance during the various stages of the study, reviewed the results and provided feedback on the manuscript prior to submission.

The three authors authorized the integration of this article into Karen Hurtubise's dissertation.

Résumé:

Analyser les effets des programmes qui visent à réduire les conséquences invalidantes de la douleur chronique chez les jeunes implique d'identifier les nombreux effets des interventions. Les variables les plus importantes à mesurer peuvent différer de façon importante selon la perspective des acteurs impliqués. Notre étude vise à identifier et prioriser les variables, selon les perspectives des différents acteurs impliqués dans les programmes pédiatriques interdisciplinaires de réadaptation de la douleur, incluant les jeunes eux-mêmes. Une technique de groupe nominal a été menée avec 13 participants représentatifs des acteurs impliqués dans ces programmes. Une analyse thématique de contenu a permis d'identifier les variables qui ont ensuite été catégorisées par thèmes. Une priorisation a été effectuée en calculant des scores pondérés. Les participants ont priorisé les variables suivantes: 1) activités de la vie quotidienne, 2) participation à des activités significatives, 3) rôles et relations sociales, 4) humeur et affect, 5) engagement scolaire, et 6) auto-efficacité. Des divergences dans le classement des variables sont ressorties selon le type de participant. Les parents et les jeunes ont accordé plus d'importance aux variables reliées à la douleur, au sommeil et à la satisfaction par rapport au programme, tandis que les thèmes de la peur liée à la douleur et au fonctionnement de la famille sont plus ressortis chez les cliniciens et les gestionnaires. Les résultats démontrent l'importance d'intégrer divers points de vue, y compris ceux des jeunes ayant un handicap lié à la douleur, dans la formulation de recommandations sur les variables principales à évaluer pour cette population.

Abstract:

Analyzing the effects of programs aimed at decreasing the disabling consequences of chronic pain on youth implies capturing the numerous outcomes of such interventions, which may differ according to stakeholders' perspectives. Our study aimed to identify and prioritize outcome domains sensitive to the evaluation needs of the various stakeholders involved in paediatric interdisciplinary pain rehabilitation programs, including youth themselves. A nominal group technique was conducted with 13 participants, representative of the various stakeholders of these programs. A content thematic analysis assisted in identifying the outcome domains; domain themes were then ranked. Total priority scores were calculated and weighted. Participants prioritized the following outcome domains: 1) activities of daily living, 2) participation in meaningful activities, 3) social roles and relationships, 4) mood and affect, 5) school engagement, and 6) self-efficacy. Discrepancies in domain ranking existed; parents and youth attributed importance to pain, sleep, and program satisfaction domains whereas clinicians and managers additionally underscored pain–related fear and family functioning. Findings demonstrate the importance of integrating various perspectives, including those of youth with pain-related disability, in generating recommendations about key outcome domains for this population.

Key Words: Pediatric pain-related disability; rehabilitation programs; outcomes; specialized pain treatment; program evaluation

Introduction

Youth living with chronic pain experience significant functional impairments, activity limitations, participation restrictions, social isolation, decreased quality of life, detrimental effects on family functioning and long-term mental health problems, resulting in a pain-related disability [1-4]. As compared with the extensive research available on interdisciplinary adult pain interventions, program evaluation evidence for pediatric interdisciplinary pain rehabilitation (IPR) programs is scarce [5]. The information about program impact is, however, central in guiding further implementation of appropriate rehabilitation programs for this younger population.

The stated goal of most pediatric IPR programs is to improve the child's functioning, despite pain [5]. However, few IPR programs have named the conceptual framework underpinning their program or the theoretical basis for understanding disability and functioning, and the impact on these youth. The International Classification of Function for Children and Youth (ICF-CY) is a widely accepted biopsychosocial framework for describing and categorizing functioning and disability in children and youth worldwide [6]. Its focus is on the impact or effect and not on cause of disability [7], on describing functioning and the health condition from the perspectives of body functions and structures, activities and participation. The umbrella terms 'functioning' and 'disability' are considered to be the result of the dynamic interaction between the health condition and the environmental factors with special consideration given to aspects of child and adolescent development [6]. Currently utilized by service providers, researchers and policy makers in a range of sectors for multiple purposes, the ICF-CY framework offers a shared language known to fosters interdisciplinary and multi-sectorial collaboration [8,9].

Collaboration among diverse stakeholders (e.g. school personnel, sports coaches, choir and band conductors, church group leaders) across multiple sectors (e.g. education, health, recreation and leisure, religion) is required in IPR programs [10]. This diversity can create challenges in establishing common outcomes upon which rehabilitation program success or failure is judged [11]. Stakeholder values, beliefs, and interests strongly impact the perspectives of program worth [11-13]. Moreover, when stakeholders do not completely agree on a shared purpose, the next steps of the collaborative process are at risk, creating conflict about the key indicators, and their interpretation [11]. Engagement of stakeholders at all organisational levels and across collaborative networks is key in achieving agreement on the important outcomes, without overlooking important impacts [14].

Attempts have been made to identify standardized outcome domains in pediatric pain in an effort to streamline study design, protocols, and assist clinicians in decision-making [5,15]. The PedIMMPACT recommendations were foundational in identifying eight core outcome domains and suggesting associated evidence-based measures for pediatric acute and chronic/recurrent pain clinical trials [15]. However, only four of the eight recommended outcome domains are routinely used in IPR program effectiveness studies. These include pain intensity, physical (i.e. disability), emotional (i.e. depression & anxiety), and school functioning [5]. Although, the PedIMMPACT did not recommend measurement of each domain in every study, it did call for those included to be justified to ensure useful interpretation of the published conclusions by researchers and clinicians [5]. With the recent promotion of patient-oriented outcomes, useful interpretation extends beyond statistical difference, to clinical relevance (i.e. the lowest level of change considered relevant by the patients in outcomes that are important to them) [16]. The PedIMMPACT outcome domain selection process did not formally integrate youth with pain-related disability or their parents, nor have they been included in previous effectiveness IPR evaluations. Consequently, little is known about what is clinically relevant or important to this patient population.

Input from other important stakeholder groups is also missing from the PedIMMPACT recommendations. School is central to the lives of adolescents [17]. Although school functioning has been acknowledged as a critical outcome, school attendance is the recommended and is the most frequently used indicator of this domain [5,15]. Evidence suggests that adolescents with chronic pain experience much broader school impairments (e.g. poor self-perception of academic competence, school avoidance, impaired ability to cope with classroom demands, peer-group participation), strongly suggesting that a more comprehensive picture of school functioning is required [17-19]. Contextual knowledge and pedagogical expertise of teachers and school personnel may be critical in advancing the conceptualization of this domain.

Decision makers from various levels of healthcare organizations are another group whose perspective, expertise and influence may be useful, yet has been overlooked to date.

The involvement of organizational leaders and managers in research from the beginning enhances the understanding of the complexities of the organizational context and needs and improves the relevance of the research generated to solving real-world problems [20-22]. In particular, middle managers, strategically located between senior leaders and frontline employees (e.g. providers, booking clerks), have received attention in the implementation of innovation and research due to their ability to bridge associated informational gaps, align resources and incentives, transcend professional barriers, and identify priorities, thus increasing the projects feasibility, outcomes, and sustainability [21-23].

With the evolution of theories about pediatric chronic pain and disability [15], the observable shift toward person- and family-centred care, the recognition of the need for patient-oriented outcomes, and the call for greater stakeholder involvement by the rehabilitation community [24,25], it seems timely to revisit the PedIMMPACT recommendations. Innovative approaches are needed to reconcile the outcomes valued by different stakeholder groups and integrate them into future pediatric IPR program effectiveness evaluations. The purpose of this study is to generate and prioritize outcome domains, sensitive to the needs of various stakeholders involved in IPR programs, including youth with pain-related disability, their parents, clinicians, teachers, and managers. This study also aimed to provide valuable information about the feasibility of using a formalized consensus-building process in selecting pain rehabilitation program outcomes with a very diverse group of stakeholders. Building on multi-stakeholder perspectives, the outcome domains selected will be used in the development of a comprehensive effect analysis of a pediatric interdisciplinary pain rehabilitation program.

Materials and Methods

Study Design

This context-specific study employed a nominal group technique, a common consensus method used to democratically make group decisions [26,27]. Descriptive statistics and a qualitative content thematic analysis [28,29] were used to make sense of the data. This study was an important step in a larger participatory research project aimed at evaluating the effectiveness and efficiency of a new intensive pain rehabilitation program, for which ethical approval was obtained from the appropriate institutional research ethics boards.

Participants

A purposeful sample of stakeholders involved in services aimed at youth with chronic pain at Alberta Children's Hospital were recruited via an email invitation. Individuals were identified by the hospital leadership and school administrative teams, and the Complex Pain Clinic program. To be included, hospital (n=5 clinicians; n=2 middle managers), and school personnel (n=2) were required to be actively involved in providing interdisciplinary pain rehabilitation intervention to youth with pain-related disability and their families for at least 2 years and be committed to the meeting schedule and activities. Youth needed to be 15-21 years of age and have chronic pain, past involvement with the pediatric Complex Pain Clinic for at least 1 year and be discharged from the clinic at the time of recruitment (n=2). Parents had to have a child or adolescent meeting the above-mentioned criteria (n=2). Once interest was established, the consent process was completed. No standard method to calculate sample size has been reported in the literature when using consensus methods. However, 5 to 15 participants have been suggested as a sufficient number to ensure group judgment reliability, while limiting group coordination issues [30-32]. Furthermore, heterogeneity of the group is recommended, reflecting a full range of stakeholders' perspectives, who have an interest in the study results, thus increasing the credibility and the acceptance of the indicators selected [30]. As inviting more participants increases of group judgment reliability and the variety of expertise and perspective, we chose to approximate the higher limit of the number of participants suggested [30].

In order to describe the characteristics and assess participants' representativeness, a demographic questionnaire was distributed, which examined participants' age, sex, and years of experience with pediatric chronic pain. As per Table 1, the recruited participants were predominantly female (77%) and were over 40 years of age (54%). The majority of clinician, manager and teacher participants had at least 6 years of experience with youth with chronic pain (69%). When compared to samples described in the literature, our clinician sample was deemed representative, based on age, sex and years of experience with pain [5,33,34].

Table 1.

Participant Characteristics

Stakeholder Group	Health Manager (n=2)	Teacher (n=2)	Clinicians (n=5)	Parents (n=2)	Youth (n=2)
Age–number of participants (frequency)					
51 years>	-	2 (100%)	-	2 (100%)	-
41-50 years	1 (50%)	-	2 (40%)	-	-
31-40 years	1 (50%)	-	3 (60%)	-	-
<30 years	-	-	-	-	2 (100%)
Gender - number participants (frequency)					
Female	2 (100%)	2 (100%)	4 (80%)	1(50%)	1(50%)
Male	-	-	1(20%)	1(50%)	1 (50%)
Years of working with youth - number participants (frequency)					
10 years>	2 (100%)	2(100%)	3 (60%)	2(100%)	N/A
6-10 years	-	-	2 (40%)	-	N/A
Years of experience with youth with chronic pain–number of participants (frequency)					
10 years>	2 (100%)	1 (50%)	1 (20%)	-	-
6-10 years		-	3 (60%)	1 (50%)	N/A
3-5 years		1 (50%)	1 (20%)	1 (50%)	N/A

Parents and youth participants were from the same family and had at least 5 years of experience managing a pediatric chronic pain condition. Although the invitation to participate in this study was extended to all 16 eligible youth and families and repeated attempts (up to 3) were made to contact those who did not respond, only two youth-parent dyads [13%] agreed to volunteer. Of the 88% of families who were considered non-responders in the overall sample, 6 families did not respond [37%], while the other 8 stated they were unable to participate [50%] due a combination of factors; these included a lack of time (n=8), school demands (n=4) and extracurricular activities commitments (n=2), distance from hospital and transportation issues (n=6).

Procedures

The nominal group technique (NGT): Participants were asked to respond individually to a question, aimed at generating ideas, which was then followed by judgments pooling, consensus achievement, and a voting procedure [26]. The NGT methodology is designed to avoid an individual's viewpoint dominating the discussion [32]. It was chosen for this study, as it has previously demonstrated effectiveness in engaging youth in the evaluation processes

[31], and in achieving consensus on outcome domains with varied stakeholder groups [35-38].

The NGT question was adapted from a previous study [38]. The question was piloted with all participants during the introductory phase of the study to ensure that it was: 1) broad enough to capture a range of outcomes; 2) relevant and meaningful to all participants; and 3) specific enough to stimulate discussion regarding outcomes relevant to pediatric pain-related disability interventions [35]. The final question presented to participants was "What are the effects that you consider most important to measure when evaluating the effects of an intervention for yourself/ your child/ your patient/ your student with chronic pain?"

Table 2 illustrates the NGT steps, the procedures completed at each step, and the coinciding data analysis processes. The NGT process was completed over the course of 4 months (October 2016-January 2017). The 5-step process employed in this study was influenced by the method described by Fink and colleagues [26], and adapted to the rehabilitation context, based on Camden and colleagues [20] recommendations for fostering greater stakeholder engagement and support, and for developing a sustainable, satisfying, mutually beneficial partnership among the varied groups of stakeholders.

Table 2.

Nominal Group Procedures and Data Analysis

Steps	Procedures	Analysis
Step 1. Introduction to a framework to establish a common language	A version of an evaluated online video [8] introducing and explaining the "F-words" in Childhood Disability [39], a series of words beginning with "f" (Fitness, Function, Family, Friends, Fun and Future) representative of the International Classification of Function (ICF) [40] domains, was presented to clinicians, teachers, and managers, and each parent-youth dyad. Relevance of the framework with the group was established by multiple examples of the impact of pain-related disability within each of the domains being generated by all stakeholder group.	
Step 2. Idea Generation	The aforementioned NGT question was distributed to participants via email, who was then asked to generate as many ideas as	Using content analysis, recorded ideas were themed into outcome

	possible related to the question and returned them to the research team.	domains by two research team members; frequencies were then calculated (see Table 3).
Step 3. Idea Recording and Step 4. Idea Clarification	Due to scheduling conflicts, two 90-minute sequential face-to-face meetings were organized; participants were asked to attend one. Representatives from each stakeholder groups were present at the meetings. At the first meeting (including 6 participants), a research team member, experienced in facilitation (KH) presented each generated theme on a flip chart sheet and invited everyone to add clarity. Any additional ideas were recorded. Elimination of items was not permitted; however further theming of the domains occurred when two or more domains represented the same concept. The second meeting (including 7 participants) began with the review of each theme and its summary from the first meeting. Further clarification by participants, and additional idea recording as per the first meeting was encouraged, in conjunction with theming. Domain definitions, emerging during the discussion were recorded during the meeting and finalized by the research team through two processes: 1) a peer debriefing process, where a researcher familiar with pediatric chronic pain outcome literature, yet not in attendance at the participant meetings reviewed the themes and definitions for comprehensiveness and clarity; and 2) a member-checking process, where a participant from the first group meeting reviewed the themes and definitions generated at the end of the second meeting.	All outcome domains themes were linked to the ICF using an established linking procedure [28] (see Figure 2)
Step 5. Voting	An online survey was then created using the identified outcome domain themes and their definitions and distributed to the participants. Participants were given 2-weeks to select and rank outcome domain themes in order of importance, using a 6-point Likert scale (with 1 = least important domain and 6 = most important domain), as participants decided that	

	6 outcome domains would be selected. Evidence suggests that the number of items chosen by participants depends on the topic but ranking of five to six is common [26]. The survey results were presented to the participants at a final meeting to ensure consensus, defined an outcomes domain theme achieving a weighted priority cut-off score of 25 or higher, and ranking highest with 3 groups or more stakeholder groups.	Priority scores were calculated as a product of the number of votes per domain and the domain ranking (from 1 to 6).

Analysis

As the NGT generated both quantitative and qualitative data, the analysis processes used reflected the different types of data collected.

Content analysis: A three-phase content thematic analysis framework guided the analysis of the responses from the NGT questions, and included: 1) condensing the data, 2) displaying the data, and 3) drawing and verifying conclusions [29]. As per the analysis guidelines, following the reading and re-reading of the questionnaire responses, data segments were coded (i.e. a word or short phrase was assigned to summarize the meaning of the segment) in an Excel© spread sheet. Two members of the research team organized these codes into categories or themes; any disagreements were discussed until consensus was achieved. Descriptive statistics (i.e. the total number and frequencies for each theme) were calculated.

International Classification of Function for Children and Youth (ICF-CY) Linking: To ensure the broadness of the outcome domain themes generated and to create a focus on the impact or effect of the IPR programs on the functioning of youth with pain-related disability, the ICF-CY conceptual framework [6] was used to categorize the final list of outcome domain themes. Content analysis themes from Step 2 (i.e. Idea Generations) and those arising from the group discussions in Step 3 (i.e. Idea Recording) and Step 4 (i.e. Idea Clarification) were associated to the ICF-CY using the linking process described by Cieza and colleagues [28], where the themes were coupled with the ICF-CY categories that were deemed most representative. One author performed the coding, with peer debriefing provided by a researcher with significant knowledge about ICF-CY framework, its components and categories. A thematic map was created demonstrating the associations between the content themes and the ICF-CY categories, as well as the relationship of those content themes that did not directly relate to the aforementioned framework.

NGT Ranking: Participants' rankings of the outcome domains were summed. A priority score for domain selection was calculated by adding the importance scores (as per the 6-point Likert scale) given by every participant for each outcome domain listed. For identical rank scores, a domain receiving more stakeholder group votes was listed higher [32]. In order to adjust for varying numbers of representatives in each stakeholder group, the priority scores were weighted to achieve equal representation within each group (i.e. 20% per group); the clinician propriety score was therefore multiplied by 0.44, that of the manager and teacher scores was magnified 2.22 times, and parents and youth scores grew by 1.11 [41].

Results

All participants responded to the electronically distributed NGT question, "What are the effects that you consider most important to measure when evaluating the effects of an intervention for yourself/ your child/ your patient/ your student with chronic pain?" and submitted their ideas. Table 3 highlights the 11 outcome domains that emerged in this second step of the NGT process (i.e. Idea Generation), along with the absolute and relative frequencies with which they were identified, and by which stakeholders.

Table 3.

Emerging Outcome Domains from NGT Question Responses

Outcome Domain Themes	Responses/Type(s) of Stakeholder	Total number of Responses	Frequency
Activities of Daily Living	Clinicians (n=5) Managers (n=2) Youth (n=2) Parents(n=2) Teachers (n=2)	13	100%
Self-Efficacy	Clinicians (n=2) Managers (n=2) Teachers (n=2) Youth (n=1)	7	55%
Quality of Life	Clinicians (n=3) Parents (n=1) Youth (n=1) Managers (n=1)	6	45%
School Engagement	Clinicians (n=1) Teachers (n=2) Parents (n=1)	5	38%

	Youth (n=1)		
Mood and Affect	Clinicians (n=4)	4	31%
Social Roles and Relationships	Clinicians (n=2) Youth (n=1) Parents (n=1)	4	31%
Family Functioning	Managers (n=2) Clinicians (n=2)	4	31%
Pain	Youth (n=1) Parent (n=1) Clinicians (n=2)	4	31%
Participation in Meaningful Activities	Clinician (n=1) Youth (n=1) Parents (n=1)	3	23%
Sleep	Parents (n=1) Youth (n=1)	2	15%
Program Satisfaction	Clinician (n=1)	1	8%

Four additional themes emerged from the third and fourth steps (i.e. Idea Recording and Idea Clarification) of the NGT procedure, during the group meeting discussions; this raised the total to 15 outcome domains. The 15 outcome domains related to the Body Functions and Structures (33%), Activity (13%) and Participation (20%) components of the ICF-CY. Only 6% linked to the Personal and Environmental factors of the framework. Table 4 presents the coding scheme and results of ICF-linking process for theses 15 themes.

Table 4.

Content Analysis Codes, Themes and ICF-CY linkage results

ICF-CY Component & Categories	Themes	Codes
Body Function • Sensory Function and Pain	• Pain	• Intensity, frequency, duration • Impact on life • Focus and described in personal identity
• Mental Function	• Anxiety	• Worry, nervousness, unease
	• Pain-related fear	• Fear, fear of movement, fear of recurrence, trauma
	• Mood & affect	• Depression, energy, posture, demeanour

	• Sleep	• Quality & duration, • Ability to get to sleep & stay asleep
	• Memory	• Ability to recall or remember knowledge and previous acquired skills
Activity • Learning & applying knowledge	• Focused attention & learning	• Maintain focus to complete a task/activity, acquire knowledge or learn/master a new skill
• General tasks & demands • Self-care • Mobility	• Activity of daily living	• Daily routine • Self-care • Ambulation • Community mobility
Participation • Interpersonal interactions and relationships	• Social roles & relationships	• Household chores • Friends and friendships • Relationship with siblings and other family members • Helping others
• Major life areas	• School engagement	• Number of courses • Types of courses • School performance • Involvement in school & extracurricular activities • Plans after graduation
• Community & civic environments	• Participation in meaningful activities	• Vocational exploration • Sporting activities • Leisure activities • Volunteering • Having fun
Personal Factors	• Self-efficacy	• Confidence problem-solving • Self-confidence • Empowerment • Self-advocacy

Environmental Factors • Support and relationships	• Family functioning	• Family stress • Family financial burden • Impact of parent employment • Impact on all family members • Changing parental roles
• Attitudes		• Family focus on pain • Parent acceptance of condition

Based on these electronic responses, the most common outcome domain was Activities of Daily Living*,* identified by all participants [100%] in all stakeholder groups. It was defined as routine activities that people are required to perform during the course of a normal day depending on age and developmental expectations. Included codes related to activities and tasks associated with self-care (e.g. dressing, coiffing, preparing lunches) and daily routines (e.g. preparing for bed). This participant provided these descriptors:

> "*Independence in activities of daily living would initially be the ability to get herself out of bed, brushing her teeth, using her own alarm clock to get herself up, getting herself dressed, ready on time so she could attend school*" [Parent 1].

It was also linked to ambulation (e.g. walking, climbing stairs) and broader community mobility (e.g. transferring in and out of vehicles, using public transit, learning to drive), as described this individual:

> "*Getting out of the house required me, 'cause he could not walk more than a few feet from his wheelchair. We noticed a big step when we went from driving him to school to having him walk down and take the bus. Sounds trivial but it was huge*" [Parent 2].

Self-efficacy, the second most frequently identified outcome domain was defined as one's belief and confidence in their ability to succeed in specific situations. Integrated codes were associated with the ability to self-manage, the confidence to problem solve in varying contexts, as well as youth and parents' abilities self-advocate in different settings (e.g. hospital, school, community). The following participants written response provides an example:

> "*Teens' ability to self-manage their pain flares and get back to activities/life better*

> *than, faster than, without assistance or as much disruption to their routine as prior to the intervention*" [Clinician 1].

The third most popular domain, Quality of Life, encompassed statements about one's feeling about their life, their short-term goals and vision for the future, as stated by this stakeholder:

> "*With quality of life, I can see a shift in how the kids look at the future, as far as their goals, what I'm gonna do this summer. I'm gonna go on a camping trip, I see myself becoming a doctor, going to university, and this is how I am going to get there*" [Clinician 2].

Freedom to do what one wants to do when they want to do it was the definition adopted for Quality of Life domain. Throughout the clarification phases (i.e. Step 4), the complexity of this composite outcome domain theme became evident to the group as this domain could encompass several of the other domain themes such as activities of daily living, mood and affect, sleep, participation in meaningful activities, and social engagement, as expressed by this participant's comment:

> "*Quality of life encompassed all these things. How it is being achieved falls under all these other pieces, such as more sleep, more functioning, more participation*" [Parent 1].

Due to the recognized difficulty in capturing subtle changes within a domain theme that includes so many different subdomains, participants chose to keep the subdomains separate instead of grouping all these domains under the Quality of Life.

The Mood and Affect outcome domain incorporated codes related to depression, as well as changes in energy and posture. This participant response described it like this:

> "*I have witnessed in many of the patients a change in their carriage, posture, and energy; a definite lightness in their presentation and overall a brighter demeanour. Obviously, this is a more intangible quality, but it's apparent to the staff who work with them*" [Clinician 1].

This theme was defined as a state of mind or feeling at a particular time, that may be demonstrated by posture and behaviour. Participants recognized this domain as being strongly linked to the Sleep outcome domain, as explained by this participant:

> "*I think there is a [relationship] with sleep. More sleep gives you a better demeanour you know*" [Parent 2]

More specifically, the sleep domain included codes related not only to the quantity or quality of sleep, but its relationship to daily performance expectations and one's the ability to get to and stay asleep.

The Social Roles and Relationships domain, defined by participants as the expectations, responsibilities, and behaviours adopted in certain situations, which naturally changes with age and development, comprised codes related to relationships and interactions with peers, siblings and other family members, household roles. As this participant reported:

> "*As he began walking longer distances, he began walking around the park. Then he would come home, notice the dogs were getting anxious and would take them out into the park with him. It was a huge help*" [Parent 2].

School Engagement referred to a disposition, willingness, and desire to participate and be successful in meaningful learning processes in the classroom and school-related activities. This domain involved not only codes associated with school attendance, but also school performance (e.g. grades), course load, involvement in school activities and planning for and following high-school graduation. This participant explained it like this:

> "*Engagement in education can be demonstrated not only by changes in school attendance, but also with an increase in number of courses completed during a school year, improvements in marks, the articulation of a post-graduation plan, participation in school-related/extracurricular activities of interest, taking on leadership roles. It's about seeing themselves as students again*" [Teachers 1 and 2].

Another participant worded it this way:

> "*To me there is definitely a difference between showing up at school and just making it through the day, and actually being involved, working on projects, having fun with friends at school, being involved in clubs and things like that. Actually, getting something out of the school day, instead of just trying to get through the day and going home*" [Youth 1].

Family Functioning adopted the definition of the capacity of the family unit to meet the needs of its members. Codes incorporated related to family stress, family members' focus on the pain, the impact of pain on family members (i.e. siblings, parents, grandparents), changes in parental roles, parental acceptance of the condition, and the effects on parents' employment and family finances. This participant scripted this way:

> "*Families report they are functioning despite pain, when pain is not at the forefront, and they are confident in how they effectively support their child in pain*" [Clinician 3].

This participant explained it as follows:

> "*Another family functioning thing is the expenses associated with the condition. For example, [my child] was in a wheelchair, so we had to buy a van. That's expensive! It may seem silly, but it adds up on the things that impact the family*" [Parent 2].

Pain denoted intensity, frequency, type and quality of the pain experience, as highlighted by this participant's quote:

> "*The character of pain to me is the change in the obsession about pain; with pain being the biggest thing in life, the only focus, to being able to focus on other things*" [Parent 1].

Participants also spoke about how pain becomes less part of the youth's identity, as explained by this individual:

> "*I notice how the program participants initially identify or label themselves, like I have pain, I am a pain patient, whereas in follow-up sessions, some of them will instead identify or label the activities they are doing, or I'm a grade X student, I'm training for a certain race, so it changes how they identify themselves*" [Clinician 2]

Involvement in activities that were important and/or enjoyable outside the home such as community recreation and leisure activities (e.g. clubs) and sports, and the freedom to make activity choices were the codes categorized under Participation in Meaningful Activities domain theme, as highlighted in this participant's quote:

> "*To do things on my own, what I want to do, like get a part-time job*" [Youth 1].

This participant states it this way:

> "*One of the first things I had to cut out was training. With modifications, like instead of skiing, I'd go out and do [less physically challenging drills], and still get to hang out with my friends, be outside doing stuff, and at least be part of what I love*" [Youth 2].

Consequently, this domain was defined as engagement in activities that fulfill a personal or culturally important goal or a purpose, in an attempt to improve life satisfaction.

The Program Satisfaction domain was defined as the level of agreement between the youth/parents perceived results of a program with their preconceived expectations of that same program. This domain theme reflected statements linked to the interdisciplinary care received, the quality and rapport established with professionals, and whether parents' and youth expectations, goals, and needs were met by the services provided. One individual put it this way:

> "*It's about comprehensiveness. Bringing all these medical experts to us was very satisfying; everyone sharing the same focus on the pain issue. It also was about keeping parents informed about what was happening as my expectations were built into that*" [Parent 2].

The outcome domains added as a result of the meeting phases of the NGT (i.e. Steps 3 and 4) included: Pain-Related Fear, Anxiety, Focused Attention and Learning, and Memory. Fear of movement, fear of pain, and fear of recurrence associated with pain were codes included in the Pain-Related Fear outcome domain, as summarized by this participant:

> "*One thing I want to put out there is that fear goes along with pain. Because when the pain gets better and then you have a bad day, there is a fear that you might end up back where [you started]. Like for [my child], who was in a wheelchair, maybe it will happen again*" [Parent 1].

Pain-related fear associated with the anticipation of pain related to movement, event or specific situation due to previous experience was the definition attributed to this domain. This domain was closely associated with the Anxiety outcome domain, defined as the degree of feeling of worry, nervousness, or unease, typically about an event or something with an uncertain outcome, as explained by the same participant:

> "*Triggers in these kids are much quicker and easier compared to someone who has not experienced the pain. 'Cause they are trying to get better but they might do something, which may lay me up for a few days but for these kids it's a lot more important, because it is a whole lifestyle to go back to. I think it is a trauma of some sort*" [Parent 2].

Focused Attention and Learning domain related to statements associated with one's ability to focus on things other than pain, for example homework, acquire new knowledge or to learn or master a new skill (e.g. musical instrument) and did not include distracting strategies (e.g. listening to music or playing video games) often utilized as a pain-management strategy.

> "*In the case of homework, the pain disrupts the focus or more the quality of focus where you are doing things that are adding value to you, like how long you can attend to a task, value added-focus. Not like video games, that are like a distraction when you have pain*" [Parent 1].

This domain was defined as the ability to direct and focus to complete any cognitively planned activity, any sequenced action, or any thought process. The Memory domain was also associated with learning but was specifically related to recalling and remembering information, knowledge or skills previously acquired. The capacity to recognize, recall, remember facts, events, impressions, or previous experiences were adopted as its definition. The participants also underscored the importance of considering some outcome domains through a developmental lens, as stated by this participant:

> "*There is a developmental trajectory to all of this. What youth are expected to do and what they want to do is very different at 12 years of age versus 15, and so on into young adulthood*" [Manager 1].

The participant provides another example:

> "*My ability to manage my pain in a way that allows me to continue what I'm doing. It's also important that I'm not limited in what I can do, and that I'm able to operate at or above the level of other people my age*" [Youth 1 with Chronic Pain].

This developmental lens was incorporated into the definition of the outcomes domain themes to which it applied. Figure 1 illustrates how the 15 outcome domain themes linked to the ICF-CY, and as well as the relationship of themes that did not directly relate to the aforementioned framework.In their descriptions of the outcome domains, participants made links between all of the components of the ICF-CY (as indicated by the arrows in Figure 1), as highlighted in this quote:

> *"It is about breaking the vicious cycle. She does not get to see her friends and work out; it impacts her sleep. If her sleep gets impacted, then she does not get to school. She does not go to school, then I say: 'You can't go do your training', and then [it] just keeps cycling*" [Parent 1].

At the fifth step (i.e. Voting) of the NGT procedure, all participants completed the online survey distributed. The following outcome domains were prioritized by all 5 stakeholder groups: 1) Activities of Daily Living, 2) Participation in Meaningful Activities, 3) Social Roles and Relationships, and 4) Mood and Affect. Four out of the 5 groups also focused on Self-Efficacy (all except manager) and School Engagement (all except youth). Analysis by stakeholder groups identified some discrepancies in outcome domain rankings.

Figure 1.

Thematic Map of the Outcome Domain Themes

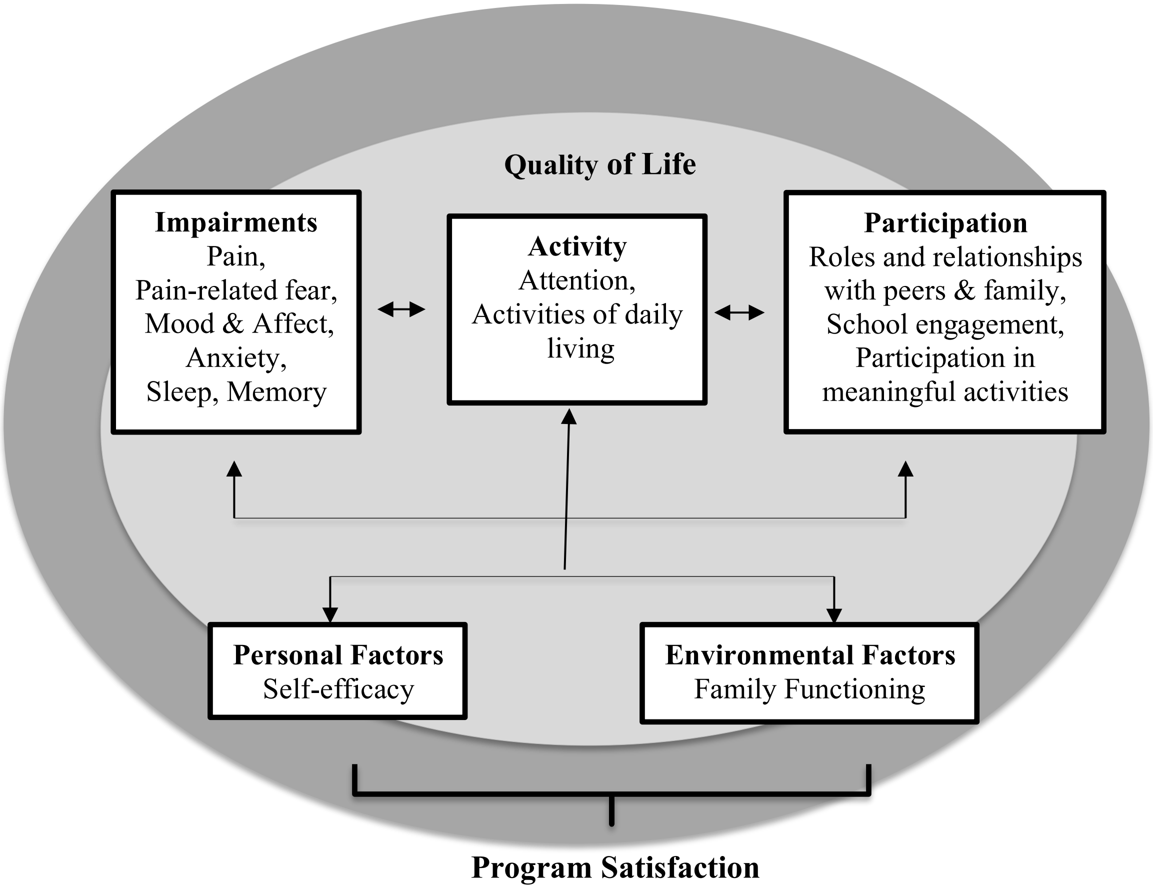
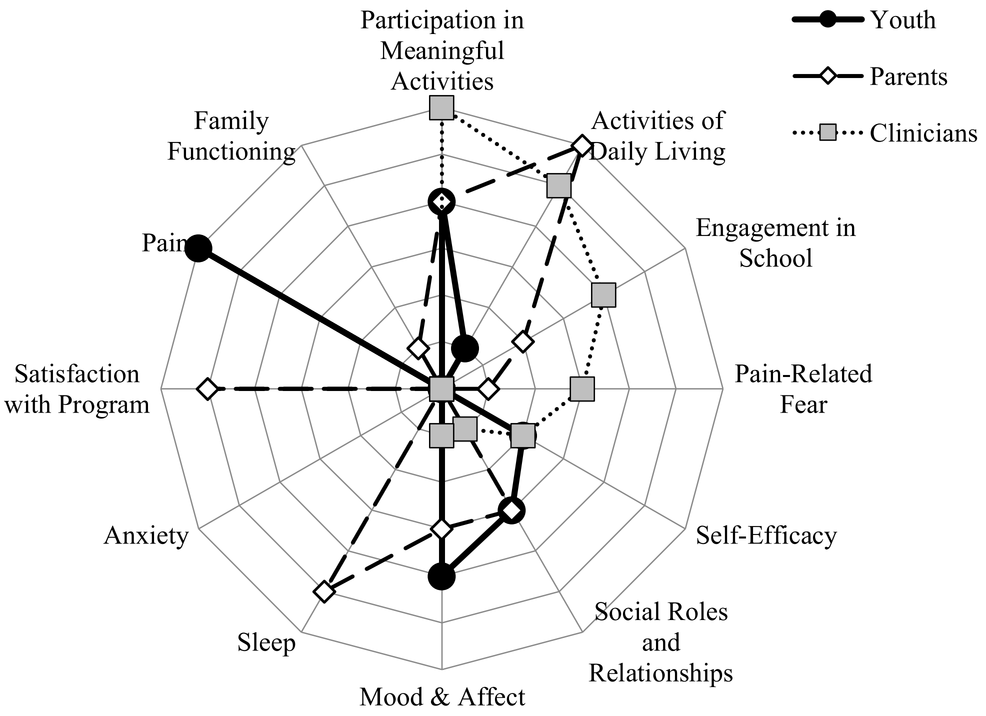


Figure 2 represents, in a two-dimensional format, the multiple outcome domains as prioritized by the youth, parent, clinician, and middle manager stakeholder groups; teachers were not included as they did not prioritize their selection. Twelve outcome domains out of

15 received a ranking score by at least one stakeholder group. Each of these 12 domains forms an individual axis of the graph, which has been arranged radially around a point. The prioritization ranking score for each outcome is depicted by the marker on the axis (also known as a spoke); a line has been drawn connecting the data values for each spoke with a different line style (e.g., dashed, solid) representing each of the stakeholder's (i.e. youth, their parents, clinicians and managers) outcome priorities. The closer the marker is to the outer edge of the spoke, the higher the priority, while the more it approximates the centre, the lower the importance. Overlapping markers indicate similar ranking priority for more than one stakeholder group. For example, parents and youth assigned the same level of importance to Participation in Meaningful Activity and to Social Roles and Relationships domains.

Figure 2.

Highest Ranked Outcome Domains per Stakeholder Group



As illustrated, Pain, Activities of Daily living, Mood and Affect, Social Roles and Relationships, Participation in Meaningful Activities and Self-Efficacy were the outcome domains ranked highest by youth, while parents prioritized, Activities of Daily Living, Sleep, Program Satisfaction, Participation in Meaningful Activities, Social Roles and Relationships, and Mood and Affect. Clinicians underscored the outcome domains of Participation in Meaningful Activity, Activities of Daily Living, Engagement in School, Pain–Related Fear, and Self-Efficacy. While middle managers also emphasized Participation in Meaningful Activity, and Activities of Daily Living, they also highlighted Program Satisfaction, Family Functioning, Engagement in School, and Mood and Affect respectively. Finally, as previously mentioned, teachers assigned the same priority to Participation in Meaningful Activity, Activities of Daily Living, Engagement in School, Pain–Related Fear, Social Roles and Relationships and Self-Efficacy, as in their opinion, all were of equal importance.

Based on the set weighted priority cut-off score (i.e. 25 or higher) and the themes ranked highest by the majority of stakeholder groups (i.e. 3 or more), the six final outcome domains prioritized for future effect analysis studies include: 1) Activities of Daily Living, 2) Participation in Meaningful Activities, 3) Mood and Affect, 4) Social Roles and Relationship, 5) Engagement in School, and 6) Self-Efficacy.

Discussion

This study's primary aim was to generate and prioritize outcome domains, sensitive to the evaluation needs of various stakeholders involved in our IPR programs, including health managers, clinicians, teachers, youth with chronic pain, and their parents. Our stakeholder group identified 15 outcome domains, distributed across the 5 components of the ICF-CY framework. Although differences between stakeholder groups arose, 6 of the domains were prioritized for measurement in a future effect analysis of our context specific IPR programs. The addition of stakeholders' perspectives is deemed particularly important in the development of outcome sets in order to ensure their relevance and meaningfulness [36,43,44]. Moreover, the inclusion of varied perspectives has been found to be effective in identify novel outcomes and provides a unique perspective in their prioritization [42-47].

The novelty and uniqueness in our stakeholder–identified outcome domains were evident when our findings were compared to those of the PedIMMPACT recommendations [15]. Our stakeholder prioritized domains associated with only three of the eight recommendations. More specifically, the Physical Functioning domain was linked by definition to our Activities of Daily Living domain, Emotional Functioning to our Mood and Affect domain, and Role Functioning related to two of our domains, Social Roles and Relationships and Engagement in School. Although our Social Roles and Relationships domain was defined similarly to that of McGrath and colleagues [15], Engagement in School was added. School engagement has been demonstrated as a robust predictor of school performance, attendance, and school completion [48-50], areas requiring further exploration in the pediatric chronic pain population [49].

Our study also highlighted other outcome domains, which were not identified previously; these included Self-Efficacy, Participation in Meaningful Activities, and Family Functioning. The emergence of the Resilience-Risk Model for Pediatric Chronic Pain [51] has led to the suggested inclusion of these outcome domains in effectiveness studies. For example, Zernikow and colleagues highlighted the need to further explore the competencies developed during IPR programs [52]. These competencies, and more specifically, the confidence to perform them, could be assessed through the measurement of self-efficacy. Self-efficacy has demonstrated a strong relationship with physical functioning in adolescents with chronic pain, mediating the relationship between pain and physical functioning [53,54]. Participation in meaningful activities, also suggested as a key outcome (i.e. perseverance with valued activities) in the Resilience-Risk Model [51], is critical to psychological flexibility, pain acceptance, enables commitment to goal-directed action [55,56], and is vital to building competencies in youth [51]. Few studies have considered the impact of IPR programs of the development of adequate competencies, which impact adolescents' abilities to engage in activities, and develop the attributed necessary to manage their pain [57]. Finally, family functioning, an additional resilience resource, has been suggested as a protective factor in promoting higher functioning among youth with a variety of chronic pain conditions [57], and may improve pain self-management, coping, and adjustment in youth with chronic pain [51]. Although the importance of Family Functioning continues to be underscored in the literature [58] and was identified as an outcome domain by our stakeholders, it did not achieve priority status among the majority of the stakeholder groups. This may be due to the recognition of the lack of an appropriate measurement tool able to capture the impact of specific family processes over time or a belief that a longer episode of care than is typically available in IPR programs is required to influence family functioning.

The list of outcome domains generated from our findings could be used in designing future context specific IPR program evaluations, serving as a guide for stakeholders in the prioritization of the outcomes to be measured. However, it must be recognized that the focus of the study (e.g. youth centred versus family centred) and the stakeholders involved may alter the choice of outcome domains.

Our use of the ICF-CY framework allowed a broader conceptualization of the scope of disability and function than traditionally represented in pain-related outcome domains. In addition to underscoring the breadth of our context-specific stakeholder concerns, it confirmed the far-reaching disabling consequences of pediatric chronic pain on youth and their families. Moreover, it suggests a need to describe these effects using a recognized "disability" framework, which could offer further guidance in the formulation of IPR program objectives and targeted outcomes and expands the choice of measurement tools available to capture stakeholder-prioritized outcome domains. Our stakeholders prioritized outcome domains most frequently linked to the Activity (i.e. the execution of tasks or actions) and Participation (i.e. one's involvement in real life situations) components of the ICF-CY. This suggests that stakeholders, despite their background, consider participation in life and its related activities as key desired outcomes of IPR programs; a concept that aligns with other paediatrics rehabilitation studies [7,38]. Recently, Kempert and colleagues identified a lack of specific rehabilitation outcomes to track physical functioning [59]. An array of clinically relevant physical functioning measures exist in pediatric rehabilitation for youth with disabilities (e.g., the School Function Assessment (SFA) [60], Participation and Environment Measure for Children and Youth (PEM-CY) [61]), which, once validated, could be applied to youth with pain-related disability, filling an important measurement gap.

Finally, our study also provides valuable information about the feasibility of using a formalized consensus method in selecting program outcomes with a diverse group of stakeholders. Unlike the Delphi method, the NGT process has been commonly used with lay persons and their carers, requires minimal pre-meeting preparation, is time-efficient, and relies on active input and face-to-face discussion among participants to achieve its goal [32]. Active dialogue in a heterogeneous stakeholder group is critical in establishing a comfort level among lay persons and empowering them, fostering respect for all ways of knowing, developing partnerships, and promoting mutual learning across stakeholder groups

[31,62,63]. However, our findings also demonstrated the importance of incorporating individual anonymous strategies to ensure valid results, reflecting the authentic perspective of each participant and minimizing participant response bias. Frequencies fluctuations in outcome domains generated from the individual responses to the NGT question (Table 2) to those prioritized following the voting process (Figure 2) were apparent. For example, Participation in Meaningful Activities was initially identified by a small number of participants yet achieved a high priority score in the voting process, while Pain*,* although identified by a variety of stakeholders in the initial NGT question responses, decreased in ranking with all stakeholders throughout the process, except for youth with a history of pain-related disability. With the growing consensus about the crucial role of patient and caregiver involvement in improving the value of healthcare research, and quality and safety in the healthcare system [62], and the mounting requests by funding agencies to show evidence of genuine consumer involvement [63], this study suggests that the NGT method engages stakeholders, and allows authentic stakeholder perspectives and opinions to be voiced. However, replication of this method in other contexts is needed to ensure its generalizability to multiple milieus, and the broader application of these outcome domains across IPR programs.

Several limitations exist in this study. First, although deemed a representative sample, the selection of the participants was restricted to those who were identified by hospital leadership and school administrative teams and therefore may limit the variability in our sample. Indeed, individuals approached may have shared similar characteristics and perspectives about what should be valued in such interventions. Secondly, stakeholder groups had an unequal member distribution due to the volunteer nature of study participation, and therefore weighting of our results were required. Although weighting equalized the influence of each stakeholder group limiting the underestimation of outcomes valued by more marginal groups (e.g. healthcare manager), we did assume that any additional participants would agree with the outcome domain priority ranking assigned by their associated stakeholder group representative which may not be the case. Thirdly, in adhering to the recommendations for consensus methods, where the study sample should reflect the perspective of a range of stakeholders interested in the results of the study [26], our participant sample was intrinsically linked to the context of our study. Consequently, generalization to other IPR programs may be limited. Future studies may consider incorporating a sample of community stakeholders (e.g. coaches, peers, and siblings), an equal and larger number of participants in each group, and one with greater geographical variability. However, increasing the sample should be carefully considered, as samples that are too large are reported to diminish the return due to coordination issues that may ensue [31,62].

Core outcome domains and measures for a particular condition have a finite lifespan. With the recent expansion in scientific discoveries in pediatric chronic pain, and the advancing conceptualization of this condition, revisiting previously published recommendations is timely. Our study findings highlight differing perspectives on what is valued as outcomes for youth with pain-related disability, underscoring the need to consult an inclusive group of stakeholders in designing program evaluations targeting this population. It also raises a widening measurement gap in outcome measures sensitive to some stakeholder priorities. Lastly, with the growing focus on patient engagement in research in many countries (e.g., Canada, United States, United Kingdom), the NGT process framed in the ICF-CY framework used in this study holds promise as a method capable of promoting mutual learning among stakeholders, while ensuring the authenticity of their perspectives, when a context-specific group of diverse stakeholders is involved.

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Article 3

Preamble

Is it worth it? A comparison of an intensive interdisciplinary pain treatment and a multimodal treatment for youth with pain-related disability

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Karen Hurtubise's contribution to this manuscript included its conceptualization, the literature review, the presentation of the findings and the writing of this manuscript. Samuel Blais assisted with the data analysis and presentation of the findings. The other co-authors (Astrid Brousselle, Melanie Noel, Frederic Dallaire, Nivez Rasic, and Chantal Camden) supervised and provided guidance during the various stages of the study, reviewed the results and provided feedback on the manuscript prior to submission.

The seven authors authorized the integration of this article into Karen Hurtubise's dissertation.

Résumé

Objectif: Évaluer l'efficacité d'un programme de traitement intensif interdisciplinaire (TII) d'un hôpital de jour en le comparant à un traitement multimodal (TMM) en consultation externe pour les jeunes souffrant de douleur chronique.

Méthodes: Un devis prétest post-test non randomisé avec groupe de contrôle a été utilisé. L'interférence de la douleur, la qualité de vie et les symptômes dépressifs chez les patients ont été mesurés au début du traitement et à 3, 6 et 12 mois après le traitement. Au total, 44 jeunes inscrits à TII et 138 jeunes engagés dans le TMM, avec diverses douleurs chroniques, ont participé à l'évaluation. Les effets des deux programmes de traitement ont été comparés à l'aide de modèles à effets mixtes longitudinaux.

Résultats: Les variables d'intérêt étaient la différence de score des variables par rapport aux valeurs initiales des mesures rapportées par le jeune à trois moments au cours des 12 mois suivants le début de l'intervention. Les participants au TII ont démontré une plus grande amélioration de l'interférence de la douleur par rapport aux participants au TMM, à 3 et 12 mois. Initialement, les scores de qualité de vie sur la santé se sont améliorés de façon similaire dans les deux groupes, mais une amélioration plus marquée a été observée dans le groupe de TMM à 12 mois. Les scores de symptômes dépressifs ne se sont pas améliorés, quelle que soit l'intervention. Seuls les scores d'interférence de la douleur ont atteint des niveaux de différence statistiquement et cliniquement significatifs avec de meilleurs résultats dans le groupe TII.

Discussion: Cette étude documente les effets des interventions de réadaptation spécialisées, TMM et TII, pour les jeunes souffrant de douleur chronique. Les résultats suggèrent également que le TII pourrait avoir un impact bénéfique plus important à long terme pour les jeunes, en particulier ceux présentant des scores élevés d'interférence de la douleur.

Abstract

Objective: Evaluate the effectiveness of an intensive interdisciplinary pain treatment (IIPT) day-hospital program as compared to an outpatient multimodal treatment (MMT) for youth with chronic pain.

Methods: A non-randomised pre-test post-test design with a treatment as usual group, used as a control. A battery of patient-oriented measures assessing pain interference, quality of life, and depressive symptoms was completed at treatment commencement, and 3-, 6- and 12- months post-treatment by 44 youth enrolled in the IIPT and 138 youth engaged in the MMT, with various chronic pain conditions. Data were analysed using longitudinal mixed-effects models.

Results: The main outcomes were the score difference from baseline of patient-oriented measures across three timepoints within 12-months of intervention initiation for both treatment groups. IIPT participants demonstrated greater improvement in pain interference, as compared to MMT at 3 and 12 months. Initially, health quality of life scores improved similarly in both groups, but greater improvement was seen in the MMT group at 12 months. Depressive symptom scores did not improve with either intervention. Only pain interference scores reached statistical and clinical significance difference levels.

Discussion: This study supports the benefits of specialized rehabilitation interventions, including both MMT and IIPT programs, for youth with chronic pain. The findings also suggest that IIPT might have greater long-term impact for helping youth, in particular those with high pain interference scores.

Key words: Pediatric chronic pain, intensive interdisciplinary pain treatment, long-term outcomes, treatment response.

INTRODUCTION

One in four youth is affected by chronic pain and the prevalence is rapidly increasing.[1] The most common pain conditions include headache (69.0%), functional abdominal pain (16.3%), back/extremity pain (13.2%), and generalized pain (1.4%).[2] Three to eight percent of children and adolescents experience pain-related disability including high levels of functional impairment, school absenteeism, restricted extracurricular activities, social isolation, decreased quality of life, and disruption in their emotional, physical, and social wellbeing.[1-7] Many of these youth are effectively treated by an outpatient multimodal intervention (MMT) including physiotherapy, psychological therapy and pharmacological interventions over many months.[8-11] On the other hand, some youth are admitted to intensive interdisciplinary pain treatment (IIPT) programs, which consist of daily treatment provided concurrently by a minimum of three healthcare disciplines including medical, psychological and physical therapy,[12] in either an inpatient unit,[13-18] a day-hospital, [19,20] or a combination of both settings for a short time period (e.g., 4-weeks).[21,22] Studies have demonstrated the short-term improvements following both IIPT and MMT program participants on pain-related disability, anxiety and depression, and school absenteeism.[23-24] IIPT is time-intensive, costly (>$30,000USD per annum), and is often deemed the last hope for families who have not experienced success in outpatient treatment.[25,26] Evidence suggests that clinical recommendations vary, despite patients reporting similar impairment profiles.[26,27] Other factors, such as insurance coverage, accommodation and travel costs, loss of productivity, family commitments, and access to services have also influenced clinicians' recommendations and parent choices.[28]

These treatment models have rarely been formally compared and the value of one compared to the other is not yet clear. Comparing the effectiveness of specialized rehabilitation pain treatment approaches is crucial in helping families, clinicians and decision makers make informed decisions about which treatment model is most effective and in what circumstances, rendering a judgment about their value and worth.[29,30] Only two published studies have attempted such a comparison, yet limitations are apparent in both. Simons and colleagues compared the outcomes at discharge and three months post-treatment of 50 children participating in a day-hospital IIPT with a gender matched group of 50 children pinvolved in an outpatient MMT.[28] The IIPT consisted of intensive daily physical, occupational, and psychological therapies, 8 hours a day, 5 days per week with a typical length of 3- to 4-weeks.[28] Alternatively, the MMT included some combination of medical, physical, and psychological therapies, with participant treatment adherence varying considerably. Youth enrolled in the IIPT demonstrated significantly greater improvements in functional disability and pain-related fear, which could be attributed to a larger treatment dose in this group.[28] Likewise, Hechler and colleagues (2014) compared youth with chronic pain involved in the IIPT, an inpatient multimodal inpatient program, and those participating in an outpatient MMT option at their 1 year follow-up visit.[31] The three-week IIPT consisted of psycho-education, pain-coping strategies for youth, parental coaching, and school-, peer- and family-related interventions as needed.[31] The MMT intervention included an in-depth physical examination, a 1.5-hour session including different education modules tailored to specific patient needs, a treatment plan comprised of medical and psychological treatment recommendations, and a follow-up scheduled at 3-months, with an open invitation to return as needed.[31] Unlike, Simons and colleagues,[28] participant matching was not evident. The study concluded that both IIPT and MMT treatments resulted in substantial changes at 12 months in pain intensity, disability, and school absenteeism, yet the IIPT showed greater improvement, which was attributed to a larger treatment dose.[31] Nevertheless, despite the standardization of MMT, the intervention option described a much smaller treatment dose than is typically received by youth with chronic pain in North America (episodic vs. weekly or twice weekly), rendering the findings of this study ungeneralizable to many pediatric chronic pain settings.[28,32] In Canada, MMT exist in most urban centers, where specialized pediatric pain services are concentrated.[32] With new health funding initiatives, IIPT programs have recently become available in some of these centers, yet none have been empirically evaluated.

Due to the lack of comprehensive details in IIPT to adequately describe the intervention components, duration and mode of delivery, more effectiveness studies have been recommended.[33] Many existing studies assessed outcomes at only one timepoint (i.e., immediately after program discharge) or in the short-term (i.e., 3-months), limiting the understanding of the trajectory of response to treatment.[34] A need exists to assess response to change over multiple time points, to gain a broader understanding of the impact of different multidisciplinary pain rehabilitation programs on youth with chronic pain over time.[56] Finally, recently patient-centered and patient engagement in research have become a focus for funding agencies and health care reform in many countries (e.g. United States, United Kingdom, Canada).[30] A gap exists in pediatric chronic pain research, where the perspectives, interests, and values of stakeholders, including youth with chronic pain and their families are lacking. Previous studies evaluating IIPT have been guided by the PedIMMPACT recommendations.[37,38] Although foundational, these recommendations did not formally integrate many important stakeholders, including youth with pain-related disability or their parents in the consensus-building process.[36] Consequently, little is known about what is clinically relevant or important to this population.[36] Absence of these perspectives hinders our understanding of the real-life impacts of the treatment experience and the judgement they would render on the value of care.[30]

To ensure the relevance and meaningfulness of this evaluation and to address the existing gaps, a consensus building process, involving an advisory committee, composed of stakeholders (e.g., health care managers, clinicians, youth with chronic pain, and their parents), preceded this study to determine outcome domains deemed important to stakeholders.[36] Six outcomes domains were prioritized by the stakeholders and included 1) activities of daily living, 2) participation in meaningful activities, 3) mood and affect, 4) school engagement, 5) social roles and responsibilities, and 6) self-efficacy.[36] This article presents the findings from an effect analysis of an IIPT and MMT programs offered at the same tertiary level chronic pain centre in Canada. Our aim was to evaluate change in stakeholder prioritized outcomes among youth who participated in a day-hospital IIPT program and compare them to youth who participated in MMT at this same facility. In keeping with a stakeholder-focus, the advisory committee selected patient-reported outcomes, which matched with four (i.e., activities of daily living, mood and affect, social roles and responsibilities and school engagement) out the six stakeholder-prioritised outcome domains. Corresponding measures for self-efficacy and participation in meaningful activity outcome domains were not available. We hypothesized that, from both a statistical and clinically significant standpoint, when compared to youth receiving treatment as usual (i.e., MMT), participants receiving the intervention (i.e., IIPT) would demonstrate greater and faster initial improvements in pain interference, overall quality of life, and in depressive symptoms, with improvements being sustained at 12-months post-treatment initiation.

MATERIALS AND METHODS

Design

Our study used a non-equivalent pretest post-test design with control group to assess the effects of each treatment under typical service delivery conditions. This study design was chosen as the study was conducted in a real-world clinical setting where a host of aforementioned clinical- (e.g., cohort timing, resource availability) and family-specific factors (e.g., place of residence, loss parental work time) can influence the type of treatment recommended and selected for each youth, and can render a high quality randomized control trial impractical and ethically questionable.[39] Instead, a self-selection process was favored, in keeping with current clinic and facility procedures, and aligned with best practice family-centred care guidelines for pediatric rehabilitation services.[40]

Study Participants

Potential participants consisted of youth who presented for a tertiary care pain clinic assessment at the chronic pain service in an urban pediatric care facility in Western Canada. To be included in this study, youth had to be between 12-18 years of age, have experienced pain for a duration of three months or more,[41] reported functional impairment as a result of their pain (e.g. decreased extra-curricular activity, school absences), and could understand verbal instructions in English. Participants were excluded if, following a multidisciplinary assessment, concerns were raised by the Complex Pain clinical team about suicidality, or intellectual disability which would interfere with rehabilitation, or of a condition better treated through a non-chronic pain related program (e.g., inpatient psychiatry). Once both treatment options were deemed appropriate, a team member (e.g. physician or nurse) presented the treatment options to families with the required information. Families were asked to choose either the IIPT (the intervention) or the MMT (the control treatment). Both interventions were publicly funded.

Intervention Group - IIPT

The IIPT intervention, inspired by the model originally described by Logan and colleagues,[19,42] is a cohort-based (3-5 patients/cohort) day-hospital program for youth (12-18 years) with chronic pain. It consists of 6 hours, 5 days per week, of collaborative and concurrent treatment by multiple disciplines including physical (PT), occupational (OT), psychological and other associated therapies (e.g. recreation, art, and music) provided individually or in groups with the inclusion of classroom time with qualified teachers. A cognitive behavioral therapy approach predominated. The program includes a psychoeducation component, inspired by the youth component of the Comfort Ability workshop[43,44] curriculum, led by a psychologist, with concepts reinforced by all disciplines throughout the duration of the program. This manualised group intervention is designed to provide a foundation for understanding how psychological interventions can facilitate pain management.[43] It includes an introduction to cognitive behavioural skills, multiple opportunities for in vivo practice of biobehavioral skills (e.g., diaphragmatic breathing, guided imagery, relaxation), and the provision of peer based support.[43] A minimum of weekly nursing and pain physician assessments are also included with a specific focus on overall progress, strategy reinforcement and medication titration. This functionally focused IIPT program aims to provide youth who experience pain-related disability with the tools to self-manage, improve their daily functioning and participation in age-appropriate meaningful activities, despite their pain. Youth complete interactive worksheets throughout the program, creating a personalised pain management plan emphasizing adaptive coping strategies that are preferred and deemed effective for each participant. Similar to other IIPT, [8,12,19,20,21] treatment intensity, the disciplines involved, frequency and intensity of treatment, and discharge are individualized and contingent on the achievement of patient identified goals as determined at the commencement of the program. Youth participants received, on average, 119 hours of scheduled therapeutic programing, of which 53-hours directly related to psychoeducation, psychology, PT, and medical interventions for an average length of stay of 25 consecutive working days. Parents of youth admitted to the IIPT are informed that at least one parent is expected to participate in parent programming. This programming, delivered in groups and/or through family therapy, includes psychoeducation, inspired by the Comfort Ability parent modules[44], and includes information about pain and parenting skills for parents with children experiencing pain (e.g., setting limits, use of consequences) and aims to provide parents with knowledge and coping skills to assist their child in returning to age-appropriate activities (e.g., school) and themselves to healthy parenting roles. On average, parent involvement was a minimum of 11-hours of participation throughout the program.

Treatment as Usual- MMT

The MMT is an ambulatory care service which, similar to IIPT, provides functionally-focused interdisciplinary treatment comprised of a mandatory 1-day Comfort Ability Pain Management Workshop for youth and their parents [43,44]. Once completed, outpatient physiotherapy and psychology sessions, medical intervention (e.g. medication regime and/or interventions), and home programming (e.g. strengthening programs, return to school and family activities, self-regulation training) are offered. The objective of sessions is to improve self-management and function with minimal interruption to daily life. The 1-day (6-hour) workshop consists of a parent group (average 8-12 participants) and an adolescent group (average 8-12 youth) which run simultaneously yet separately.[43] To minimize loss of school time, the workshop is typically delivered on a school holiday or weekend by 2 psychologists, a clinical nurse specialist, and an art-therapist who have received specific training. The duration and dose of the PT, and psychological interventions, and the need for other disciplines (e.g., family therapy) are tailored for each participant with discharge contingent on the achievement of patient-identified goals. Youth participant received on average 51-hours of psychoeducation, psychology, PT, and medical intervention over 10-months, while parents received a minimum of 9-hours of intervention during this same timeframe.

All clinicians in both the IIPT and MMT received special education and training in pediatric chronic pain, had developed extensive knowledge, skills, and experience in providing care to this population, and often alternated between these two programs, gaining experience in both intervention models.

Outcome Measures

Patient demographics and pain condition characteristics: Basic demographic (e.g. age, sex, household income), pain-related variables (e.g. primary pain complaint, chronicity), and reason for treatment choice were collected at program commencement (i.e. baseline).

Pain Intensity: Participants reported their pain intensity over the past 7-days using a valid and reliable 11-point numeric rating scale (0 = no pain to 10 = most pain possible).[45] The pain numeric rating scale has been found to be a reliable and valid measure of pain intensity in children and adolescents.[37,45]

Available patient-oriented outcomes (PRO) were matched to the stakeholder prioritised outcomes domains. PRO's are defined as any reports coming directly from patients about how they function or feel in relation to their health condition and its therapy.[46]

Pediatric Quality of Life Inventory (PedsQL) Generic Core Scales 4.0: The PedsQL, a 23-item standardized self-report measure, was used to assess general health-related quality of life over the past month across four domains i.e., physical, emotional, social, and school functioning.[47] Participants responded to questions on a 5-point-scale ranging from 0 (*never*) to 4 (*almost always*).[47] Each item was then reverse-scored and transformed to a scale of 0-100, from which a score sum is created, with higher scores indicating better health-related quality of life.[47] The PedsQL has shown excellent internal consistency, well-established validity and reliability in youth and has been widely used to pediatric pain research.[34, 48-51]

The PROMIS Pediatric Pain Interference (PPI): The PPI measure short form, part of National Institute of Health's Patient-Reported Outcome Measurement Information System (PROMIS) initiative, was employed to assess the consequences of pain on the relevant aspects of youth's life over the past 7 days, including its impact on engagement in social, cognitive, physical, and recreational activities.[52] Scored on a five-point Likert scale from 1 (*never*) to 5 (*almost always*), the raw scores were converted into a T-score (using the tables found at www.healthmeasures.net) with higher scores suggesting greater pain interference on daily activities. The measure has been found to be reliable and valid for youth with pain-related disability.[53]

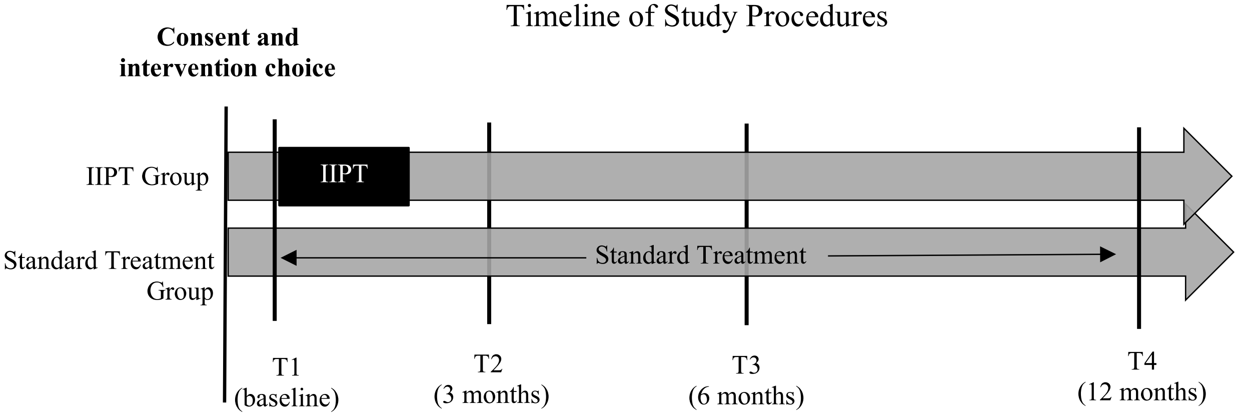
The PROMIS Pediatric Depressive Symptoms: The PROMIS Depressive Symptom measure short form was completed to evaluate depressive symptoms in youth. Youth were asked to rate on a scale from 1 (*never*) to 5 (*almost always*) how often they experienced a particular depressive symptom over the past week, with raw scores transformed into T-scores as recommended.[54] Higher scores are suggestive of a more negative mood, poorer view of one's self and reduced social cognition.[54] This measure has also been found to be reliable and valid with a sample of youth with pain-related disability.[53]

Procedure

Data was collected from participants enrolled in the IIPT between 2014 and 2018. During this same time period, the Outpatient Clinical Outcome Registry, established to collect longitudinal and comparison patient data receiving pain services at our facility, was used as the MMT data source. Demographic, pain-related and questionnaire data were collected using Research Electronic Data Capture (REDCap), a secure online data collection tool[55] using electronic links. For both groups, the battery of previously described patient-reported measures was assessed at baseline (i.e., commencement), 3-months, 6-months and 12-months post-program commencement (see Figure 1). Collecting data from several points in time (usually ≥ 3 time-points) is important to test the long-term strength of intervention effects once the treatment is completed.[56]

Figure 1.

Timeline of Study Procedures



For the data analysis, de-identified data were extracted. The study and database registries were approved by the institutional review boards. Informed consent was obtained prior to study involvement, and all patients who agreed to participate completed an electronic consent form prior to initiating the online questionnaires distribution. Youth and their parents were also asked to indicate their desire for responses to be shared with their treating clinical team. No compensations were offered to participants for the survey completion.

Statistical Analysis

Data analysis were performed using SAS software (version 9.4, Cary, NC, USA) with the overall significance level established at p=0.05. Continuous variables are presented as means (standard deviation) or median (interquartile range) depending on their distribution. Categorical variables are presented as frequencies (percentages). Follow-up outcomes were reported in absolute values and as differences from baseline measure.

Longitudinal mixed models (LMM) were used to test the influence of the treatment group on the mean value and progression rate of score difference from baseline, while correcting for the initial absolute outcome value. Covariates that were potentially clinically important such as sex, age, identified pain condition (e.g. headache), along with initial average pain intensity scores, and baseline outcome scores (i.e. PedsQL, PPI, and Depressive Symptoms) were tested. Variables with a significant influence on the models (e.g., sex, baseline score) were kept and the others discarded (e.g., age, pain condition). Supplemental materials provide further details on the variable selection process. The estimates of the influence of variables on the progression rate were calculated by including an interaction term between the variables and time post-treatment. To provide adequate modeling and a better model fit to the score differences over time, two linear regressions were needed: 1) from baseline to 3-months and 2) after 3 months post-treatment. Although other authors have questioned the linearity of response trajectories in this population, in particular related to IIPT, such hypotheses have yet to be tested.[34,53] We report estimates (β) of the influence of the independent variables of initial mean value (intercept) and on that of progression rate of score differences from baseline (slope). LMM offer several advantages: 1) they allow empirical description of the amount of variance in outcome scores (intercept variance) and the amount of variance in the change across time (slope variance), 2) they account for the correlation of repeated measures on the same patients; 3) they enable utilization of all the available data (individuals can have missing data for one or more timepoints); and 4) the account for unequal time periods between observations.[34,57] Model validation strategy consisted of verifying the assumptions associated with LMM (e.g., heteroscedasticity, linearity, and normal distribution of residuals).

Group-specific estimated mean value, 95% confidence intervals of score difference for each outcome, and adjusted means for other covariates were computed. The group differences were compared at 3 months and 12 months using a student's T-Test. The 95% confidence intervals were calculated using bootstrap resampling of the original sample (bootstrap results are presented in Table S1). More details on model specifications, validations strategy and bootstrap method are available in supplemental materials.

Finally, PRO's should be assessed within a clinically meaningful framework in order to determine treatment effectiveness.[46] Therefore, in addition to the psychometric properties, the minimal clinically significant difference (MCID) represents the smallest difference perceives as beneficial by patients and which mandates a change in patient management was calculated for each outcome. To do so, a triangulation method using two statistical methods, (the standard variation and the effect size methods) and values identified in the literature was employed.[58-60]

RESULTS

A total of 182 participants (44 youth in IIPT and 138 in the MMT group) met the inclusion criteria. The descriptive characteristics of the included study population are presented in Table 1. Slight differences between the two groups were noted with the IIPT group having slightly higher household income, not attending school full-time, and suffered predominantly from neuropathic/musculoskeletal pain.

Table 1.

Participant Sociodemographic and Medical Characteristics

Variables	All	IIPT	MMT
Demographic characteristics			
Age (Median (Q25-Q75))	16 (14-16)	16 (15-17)	15 (14-16)
Female (Frequency)	82%	87%	80%
Caucasian (Frequency)	68%	69%	68%
Married parents (Frequency)	78%	82%	77%
Household income>$90,000 CAD (Frequency)	55%	62%	54%
Full-time School Status (Frequency)	72%	39%	81%
Missed school days in 1 months (Median (Q25-Q75))	3 (1-5)	4 (1-9)	2 (0-5)
Missed Days of work by parent at admission (Median (Q25-Q75)	2 (0-6)	2 (0-10)	3 (0-6)
Pain condition characteristics			
Average pain rating (in the past 7 days) Median (Q25-Q75))	6 (5-7)	7 (6-8)	6 (5-7)
Pain lasting for more than 12 months	90%	94%	78%
Neuropathic pain (Frequency)	19%	41%	14%
Headache (Frequency)	35%	7%	42%
Musculoskeletal pain (Frequency)	21%	30%	19%
Other (e.g., abdominal) (Frequency)	24%	22%	24%

For IIPT parent respondents (n=24), their program selection decision was influenced primarily by their child's condition (100%) and the assessment and recommendations of the clinical team (100%). Other factors included the child's academic & social requirements (20%), the amount of treatment (20%), and the needs of the other children (10%), and family commitments (10%). The majority of participants (70%) completed the questionnaire battery for at least 2 timepoints, with the varying number of observations for each outcome at each time point recorded in Table 2. Retention of the MMT participants at 3 months (i.e., timepoint 2) was 51%, 34% at 6 months (i.e., timepoint 3), and 28% at 12 months (i.e., timepoint 4), which is typical of this treatment option.[53] In the IIPT group, the retention was higher at each of these timepoints with 93%, 62% and 59% participant retention respectively. Unadjusted mean scores for each timepoint according to treatment are presented in Table

Treatment Effect: The general progression and the influence of variables on the slope and the mean value of outcomes score difference from baseline are presented in Table 3. All outcomes showed higher progression rates (i.e., steeper slope) initially following program commencement, with a decreased score difference progression rate noted after 3-months. This non-linear relationship of time was accounted for in our models by computing two different equations, a baseline to 3-month slope, and a 3-month to 12-month slope.Table 2 presents the general progression of score differences for each outcome (intercept and slope) by treatment group and the influence of other covariates on slope and intercept. Pain type and age were initially tested in the model but did not show sgnificant influence on mean values or progression rate and were therefore excluded from final models. The estimated adjusted means for each group and outcome are presented in Table 4.

Table 2.

Outcomes Unadjusted Means, 95% Confidence Intervals, and Number of Observations at Each Time Points

Outcome	Treatment group	Baseline	3 months		6 months		12 months	
			Absolute value	Difference from baseline	Absolute value	Difference from baseline	Absolute value	Difference from baseline
PedsQL	IIPT	50.06 (46.00,56.13) n=36	60.78 (53.46,68.11) n=32	10.00 (3.75,16.25) n=31	61.11 (53.06,69.17) n=24	8.40 (2.44,14.37) n=24	55.39 (47.22,63.56) n=17	5.29 -1.18,11.76) n=17
	MMT	60.67 (56.71,64.63) n=105	68.57 (63.24,73.90) n=56	8.58 (4.08,13.09) n=53	68.98 (62.19,75.77) n=36	12.06 (5.76,18.36) n=34	75.21 (67.04,83.37) n=32	15.00 (6.76,23.24) n=28
PROMIS Pain Interference	IIPT	70.72 (68.65,72.79) n=21	59.62 (55.32,63.91) n=20	-11.14 (-15.13, -7.14) n=20	61.73 (56.07,67.39) n=13	-8.68 (-13.94, -3.42) n=13	56.91 (50.07,63.76) n=14	-9.57 (-16.78, -2.36) n=10
	MMT	53.24 (51.97,54.72) n=109	58.21 (55.62,60.80) n=57	3.54 (1.25,5.84) n=55	57.67 (54.10,61.24) n=37	1.29 (-1.96,4.54) n=36	55.80 (51.54,60.05) n=30	0.85 (-3.00,4.70) n=27
PROMIS Depressive Symptoms	IIPT	61.49 (57.87,65.11) n=21	55.77 (50.14,61.39) n=20	-6.02 (-10.02,-2.01) n=20	59.66 (51.04,68.28) n=12	-1.52 (-8.49,5.46) n=12	57.83 (50.56,65.10) n=13	-4.41 (-10.64,1.81) n=9
	MMT	47.15 (45.41,48.90) n=111	51.45 (48.44,54.45) n=57	4.50 (2.22,6.79) n=56	53.07 (49.20,56.95) n=38	4.95 (2.06,7.84) n=38	46.93 (43.14,50.72) n=33	-0.78 (-5.06,3.49) n=31

Table 3.

General Progression and Parameter Estimates for Longitudinal Mixed Linear Model Evaluation of Outcomes Differences from Baseline to 12 months

Outcomes (number of observations)	**Intercept**		**Slope before 3 months****		**Slope after 3 months****		**Estimates of variables influence on slopes (progression rate)**		**Estimates of variables influence on intercept**		
	IIPT	MMT	IIPT	MMT	IIPT	MMT	Sex $_a$	Baseline Score	Sex $_a$	Baseline Score	Average Pain Score at Baseline
PedsQL Total (n=301)	19.87	17.68	3.10	3.67	1.17	1.74	0.81	-0.03***	1.07	-0.09***	-2.19***
PROMIS Pain Interference (n=289)	-6.12	-2.06	2.48	3.02	1.75	2.30	-	-0.04**	-	-0.10	1.45***
PROMIS Depression (n=298)	-2.56*	1.20*	3.16	2.88	1.58	1.30	-0.66*	-0.04***	-1.09	-0.01	-

Legend: *p<0.05, **p<0.01, *** p<0.001

NOTE: Values are β. Slopes before and after 3 months were significantly different for each outcome but there was no significant difference between groups regarding overall progression rate.

$_a$ =Male. Female used as reference

Table 4.

Outcomes Adjusted Means and Differences Between Treatment Groups at 3- and 12-month Follow-up

	3 months LS Mean (95% CI)				**12 months** LS Mean (95% CI)			
Outcome	**IIPT**	**MMT**	**Difference between IIPT-MMT**	**t-test value** (*p* value)	**IIPT**	**MMT**	**Difference between IIPT-MMT**	**t-test value** (*p* value)
PedsQL Total	9.36 (5.70,13.02)	8.88 (8.85,11.90)	0.48 (-2.70,3.67)	0.30	17.43 (9.8,25.06)	22.06 (15.25,28.87)	-4.63 (-10.84,1.57)	-1.47
PROMIS Pain Interference	-3.77 (-5.58, -0.96)	1.94 (0.20,3.67)	-5.71 (-8.67, -2,75)	-3.81***	-4.52 (-10.28,1.24)	6.13 (2.32,9.94)	-10.65 (-16.72, -4.57)	-3.46***
PROMIS Depression	0.77 (-1.88, 3.42)	3.69 (1.79,5.59)	-2.92 (-5.47, -0.36)	-2.26*	-0.24 (-5.83,5.35)	0.15 (-3.97,4.26)	-0.39 (-5.47,4.69)	-0.15

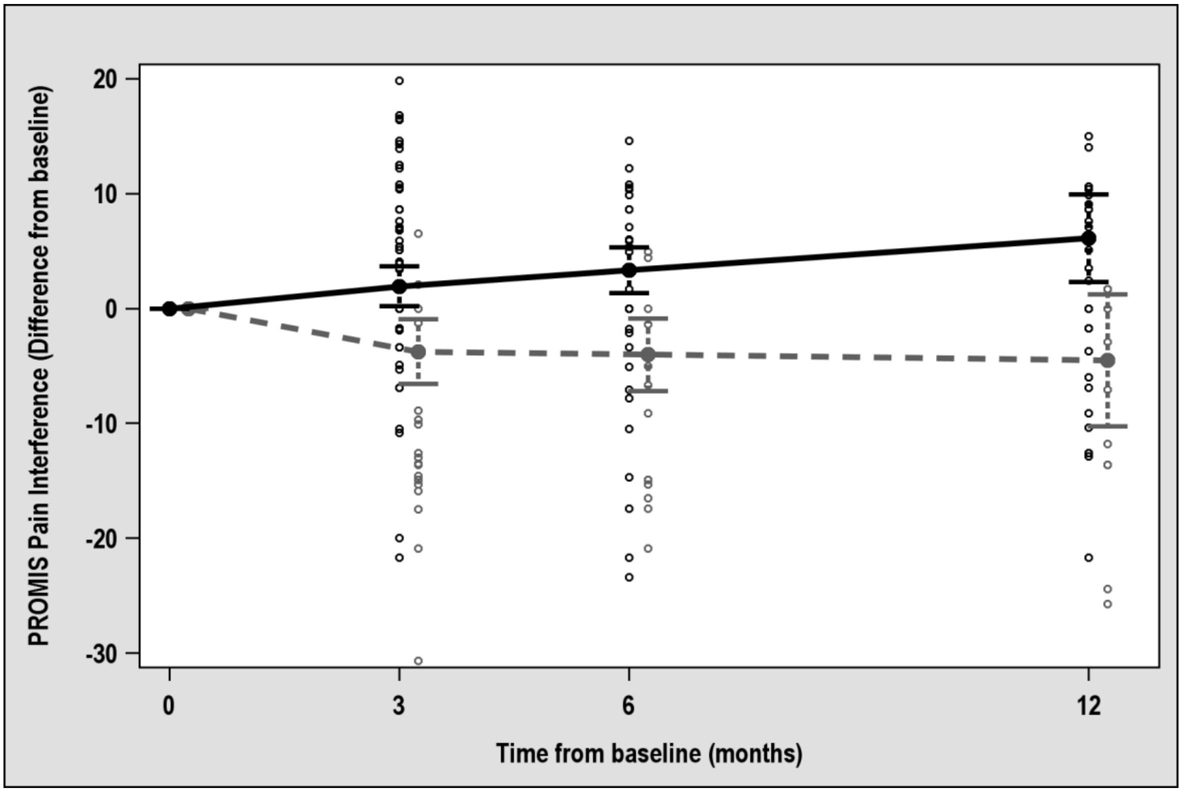
Legend: *p<0.05, **p<0.01, *** p<0.001.

NOTE: Adjusted for pain score at baseline (mean=5.34), sex, and average pain score.

The visual representations of the progression of adjusted means over time for these outcomes are illustrated in Figures 2, 3, and 4

Figure 2.

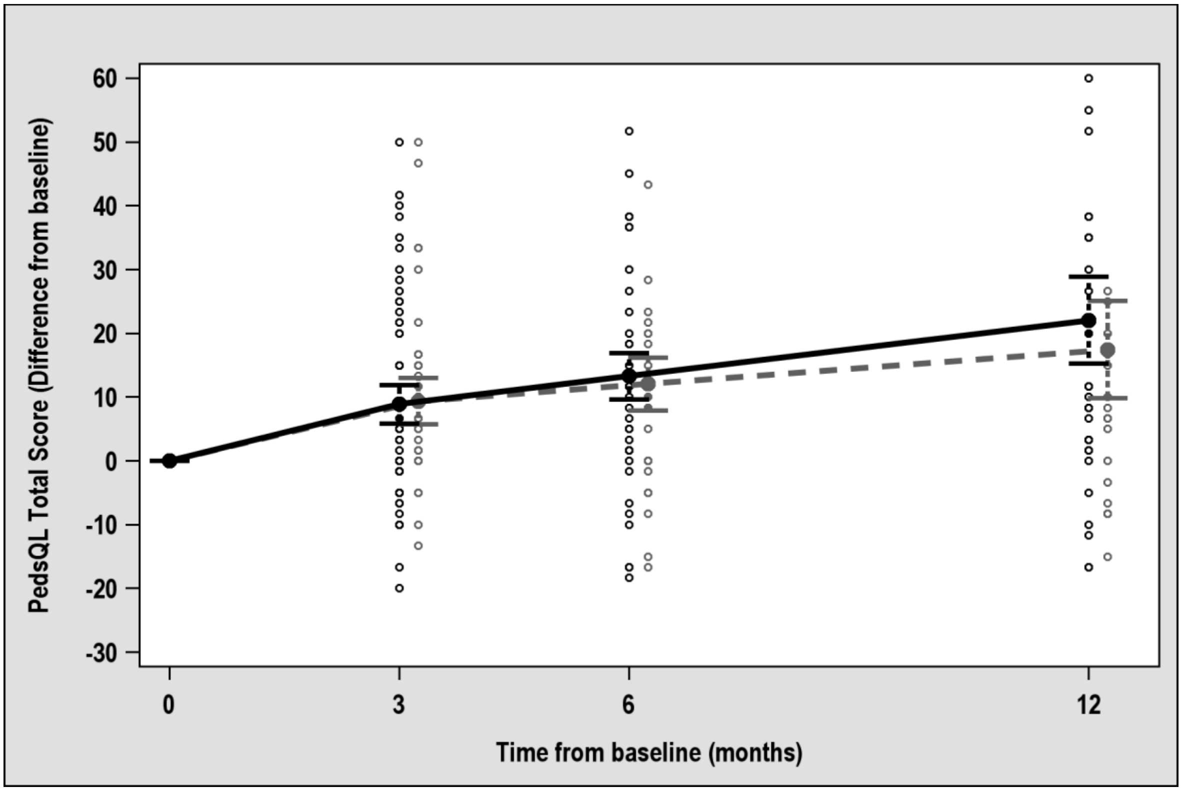
PROMIS Pain Interference for Child and Youth



Legend: ——— = MMT; – – – = IIPT

Figure 3.

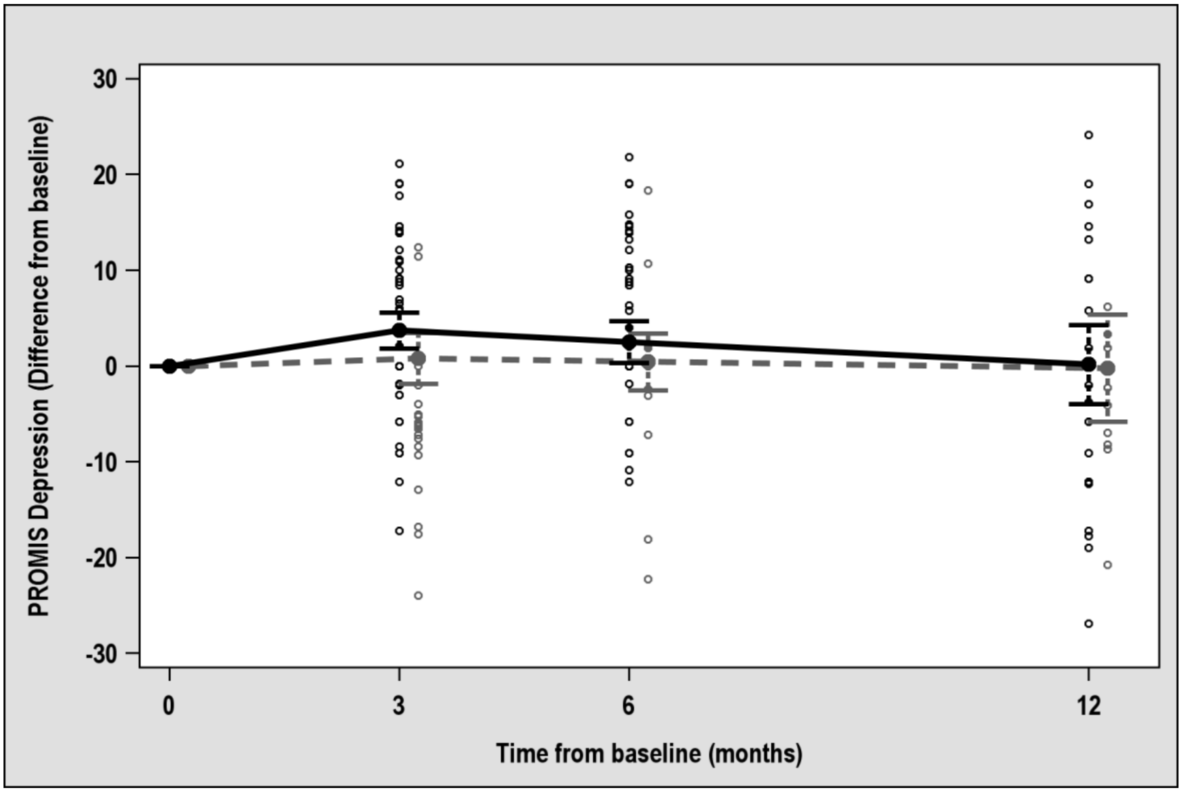
PedsQL 4.0 Generic Core Scale



Legend: ▬▬ = MMT; ‒ ‒ ‒ = IIPT

Figure 4.

PROMIS Depressive Symptoms



Legend: ——— = MMT; – – – = IIPT

Outcomes

Pediatric Pain Interference

As per Table 3, LMM findings revealed a deterioration in PPI scores over time for the MMT group. The IIPT group had a protective effect on progression rate and showed an improvement over time, when corrected for other covariates (see Figure 2). A higher baseline pain interference score was associated with a greater rate of improvement in the pain interference score difference (β = -0.04 points per month for each unit increase of the baseline score; ρ <0.001). The average pain score at baseline was also associated with an increase in pain interference mean difference from baseline. Sex was not a significant predictor of the mean or of the progression rate from baseline.

As noted in Table 4, the IIPT group demonstrated statistically significant improvement at the 3-month timepoint (pain interference difference from baseline = -3.77) as compared to the MMT group (pain interference difference from baseline= 1.94) with a difference of -5.71 in favor of the IIPT group. This improvement in the IIPT group remained at the 12-month timepoint (pain interference change score difference from baseline = -4.52), while the MMT group demonstrated further worsening (pain interference change score difference from baseline = 6.13), for a difference of -10.65 in favor of the IIPT group, which was statistically significant ($\rho<0.001$).

For the PPI scores, our sample MCID threshold was noted to be between 2.63 (effect size calculation) and 6.59 (0.5 standard deviation). The IIPT post- treatment change in PPI scores did achieve the MCID at 3-months as compared to the MMT group and was noted to be well beyond it at 12-months.

PedsQL

As highlighted in Table 3, the total sample (i.e., both groups) demonstrated improvements in their PedsQL total change in scores differences over time with a greater increase in progression rate noted during the initial 3-months. When compared, the IIPT group tended to have a slightly slower progression rate (i.e., slope <3-months = 3.10 points per month; slope >3-months = 1.17 points per month) as compared to the MMT group (i.e., slope <3-months = 3.67 points/month; slope >3-months = 1.74 points per month); however this difference was not statistically significant. The PedsQL overall score mean difference was influenced by the average pain score at baseline, with each increase in pain intensity score unit, reducing the mean PedsQL score by 2.19 points ($\rho<0.001$). Furthermore, male participants demonstrated a trend toward higher PedsQL scores and faster improvement (4.48 points per month) as compared to females (3.67 points per months) initially post program ($\rho=0.05$), yet this difference was not statistically significant.

Adjusted means, corrected for the average pain intensity scores and baseline PedsQL overall score, were calculated at 3 and 12-months. As illustrated in Table 4, both IIPT and MMT groups demonstrated statistically significant improvements at 3 months (9.36 and 8.88 points respectively), with a minimal difference (0.48 points) in favor of the IIPT group, which was not statistically significant. The improvement remained important for both groups at 12-months, with an increase from baseline of 17.43 (IIPT group) and 22.06 (MMT group) with a difference between groups of 4.63, in favor of the MMT group, yet did not reach statistical significance.

For our sample, the MCID threshold fell between 4.29 (effect size calculation) and 10.71 (0.5 standard deviation). The change in PedsQL score did achieve MCID threshold at 3 months for both groups, and well surpassed it at 12 months, with further gains noted in the MMT group.

Depression score

Like the PPI, the depression score worsened in the overall study population initially, as highlighted in Table 3, but returned to baseline levels at 12 months. The IIPT group had a favorable progression (i.e., slope < 3-months = 3.16 units per month; slope > 3-months = 1.58 units per month), compared to the MMT (i.e., slope < 3-months = 2.88 units per month; slope > 3-months = 1.30 units per month), although it did not reach statistical significance. Patients with a higher baseline depression score (i.e., more negative mood) tended to improve more ($\beta = -0.04$ units/months from each unit increase of the baseline score; $\rho < 0.001$), as did the males ($\beta = -0.66$ units/months; $\rho < 0.05$). Male sex was also associated with a decreased mean depression score ($\beta = -1.09$ points) as compared to being female, yet statistical significance was not achieved. In contrast to the other outcomes, the baseline average pain score was not associated with depression score progression over time.

The adjusted means for both the IIPT and the MMT group showed a slight improvement at 3 months (depression change score difference = 0.77 for the IIPT and 3.69 in MMT group) with a -2.29 difference in favor of the IIPT, which achieved statistical significance. However, this improvement did not persist in either group, with a regression towards baseline values observed at 12-months (depression score change difference = -0.24 for IIPT group and 0.15 in MMT group), and no statistical difference between groups (see Table 4). Similarly, the change in depression scores achieved the MCID threshold for the IIPT group as established for our sample (i.e. 2.14 to 5.36) at 3 months but was not sustained.

DISCUSSION

This study evaluated changes in patient-oriented outcomes across the first year following treatment among youth with pain-related disability participating in IIPT program as compared to those participating in an MMT. The benefits of IIPT in the treatment of pain-related disability have been the focus of many recent studies, leaving the effects of MMT understudied, and comparison of the two interventions scarcely explored. Our study used an approach where outcome domains prioritized by stakeholders, which included youth with pain-related disability and their parents, were used as the foundation for this evaluation.[37] Additionally, outcome measures, recognized as patient-oriented, were matched to these domains. Moreover, the chosen measures are not those typically seen in IIPT evaluation studies, and instead are those typically used in examining other pain population (i.e., PedsQL) and are those recently introduced to the field (i.e., PROMIS). As patient-reported outcomes could only be matched for 4 out the 6 stakeholder-prioritised outcome domains, development of additional patient-reported outcomes in this population is needed.

Our finding revealed that youth participating in a specialized treatment program, regardless of the treatment option (i.e., IIPT or MMT), reported initial and longer-term improvements in outcomes (PedsQL and PPI). When comparing IIPT and MMT, the IIPT demonstrated particular effectiveness, both statistically and clinically, in improving pain interference initially at 3-month post-treatment, with ongoing improvement noted at 12-months. This result is consistent with other similar IIPT studies.[29,39] Youth with higher baseline average pain intensity and pain interferences scores had greater improvement. Depressive symptoms also demonstrated greater initial improvement in the IIPT group, which were noted to be statistically and clinically significant. Hhowever, this benefit was not maintained long-term, either statistically or clinically. Both groups demonstrated clinically significant improvement on change in health quality of life scores, yet the MMT group demonstrated greater improvement long term. Differences between sex were also noted with boys improving more than girls in pain interference and depressive symptoms scores, while baseline pain intensity scores impacted pain interference and health quality of life scores, but not depressive symptoms scores.

Furthermore, in both programs studied, improvements in health-related quality of life were also reported initially. These improvements in youth's perception of their functioning[48] were still evident at 12 months, yet counter to our hypotheses, more improvements were found in those participating in MMT as compared to the IIPT. These finding may be somewhat explained by the differences in treatment approaches between IIPT and MMT. IIPT aim for functional restoration, whereas the goals of MMT may be more diffuse.[27] More specifically, IIPT employ a more standardized protocol focused on improving physical functioning and addressing fear-based co-morbidities over a short period of time, with limited treatment continuation following program completion. On the other hand, MMT tend to be more individualized and continue over a prolonged period of time, which may better suit certain patient profiles. Previous authors have suggested that subgroups of youth with varying profiles of chronic pain (e.g., more or less function disability, school absenteeism, anxiety or depressive symptoms) may require specific treatment protocols.[26,27,61] For example, youth with high emotional impairment and pain-related affect may require more individualized treatment focused on affect and emotion over a longer period of time, for which MMT may be a better treatment option.[27] Alternatively, some authors have also suggested that this subgroup of youth may require ongoing psychological support following IIPT. [28,62,63] Furthermore, as indicated in this study and others[26,27], youth with higher levels of pain interference may be those better suited for IIPT. Health related quality of life has only been used in a handful of studies in this population. As other factors (e.g., social difficulties) begin to emerge as treatment outcome predictors,[26] multidimensional constructs, such as health quality of life, should be incorporated into research. This will ensure the development of clinical treatment protocols, with explicit dosing parameters, that best meet the global needs of these youth.

Our findings also draw further attention to the potential importance of the baseline depressive symptom score on treatment outcomes, a finding reported in other studies.[64] Like these studies[38,65], the IIPT did demonstrate short-term effectiveness, yet failed to show sustained long-term change in depressive symptoms scores in the IIPT group, which conflicted with our hypothesis. Generally, evidence-based psychological interventions in specialized rehabilitation programs have targeted fear-based constructs and co-morbidities such anxiety and trauma in these youth.[66] Depression is not a fear-based disorder and unique factors (e.g. suicidality, behavioral inactivation, specific parenting responses) underlie its association with chronic pain,[66] in particular its negative association with function.[64] Existing evidence-based interventions shown to be effective in the treatment of depression should be more consistently integrated into these treatments for those demonstrating the co-occurrence of depressive symptoms and chronic pain.[66]

Individual socio-demographic and pain characteristics were also found to influence certain outcomes, similar to other studies.[61,67] Specifically, the boys reported higher quality of life at baseline, and faster improvement rates, as compared to the girls. Along with previous research results, this study further underscored baseline pain intensity score as important predictors of treatment outcomes.[26,27,33] Pain intensity scores at baseline were associated with a greater improvement in pain interference and health quality of life scores. However, it did not influence the depressive symptoms score.

This study is one of the first to our knowledge to use stakeholder prioritized outcome domains in the effect analysis of interventions targeting youth with chronic pain. In addition to representing the outcome upon which the value of intervention sould be judged, the measures were also chosen because of their relevance to the stated goal of specialized rehabilitation programs, i.e., youth with chronic pain will assume an active self-management approach in coping with pain and to enable age-appropriate daily activities despite pain. For example, a health-related quality of life (i.e., the PedsQL) representative not only as youth's subjective sense of well-being, but their ability to self-manage,[68] was included.[47] The PROMIS pain interference, a measure of the extent to which pain hinders engagement with social, cognitive, emotional, physical, and recreational activities focused on youth's perception of their engagement in age-appropriate activities.[69] Due to its association with mood, PROMIS depressive symptoms scale was also included. Matching outcomes to the aim of the intervention has been advocated for self-management programs for youth with chronic conditions, like chronic pain.[70] Losing such focus hampers conclusions about their effectiveness.[70] Furthermore, this study compared PROMIS outcome measures across specialized rehabilitation pain treatment options enabling the examination of statistical and clinical relevance. The MCID thresholds calculated in our sample were similar to those identified in the literature.[46,71,72] Future research should consider establishing anchor-based MCID specific values, where youth with pediatric pain are involved in determining them.

The findings of the current study should be interpreted within the context of several limitations. Firstly, this type of non-randomized study design has been recognized as challenging to implement in the real-world clinical setting.[53] Our small sample size precluded our ability to examine some outcome subscale due to participant attrition experienced in both our IIPT and MMT groups. Other longitudinal studies have experienced similar issues rendering monitoring outcomes over time challenging in this population.[18,34,53] Although reasons for attrition are unclear, as suggested by other authors,[34] and future research should investigate ways to increase participation adherence in these types of studies. Secondly, due to the volunteer aspect of our recruitment strategy and the non-randomized nature of our study, an unintended referral and selection bias may have occurred. These biases may be due to the specific criteria for admission to IIPT program (e.g. did not respond to outpatient physiotherapy treatment).[28] Also, IIPT programs were originally designed for those suffering from higher levels of functional disability and neuropathic pain.[8,18,41] Our sample characteristics at admission are consistent with other pediatric day-hospital IIPT programs.[18,19,20] However, IIPT are often recommended for youth exhibiting higher levels of functioning, low levels of depression, with varying levels of coping, where the long-term outpatient MMT may be more appropriate.[27] Although, it is recognized that treatment decisions can be influenced by many family contextual factors (e.g., distance, parent preference),[28,73,74] our data suggests that the child's clinic team's recommendations and the child's condition play the largest role. Due to the time-intensive, financial investment, and significant commitment and engagement required in participating in specialized rehabilitation treatment, conducting such studies and sharing the information is imperative in assisting families in care decision-making.[27] Comparison studies are necessary to conduct much needed economic evaluations of these two treatment approaches.[28] Thirdly, shared method variance is also a limitation that should be underscored, as only youth self-reported measures were use in our analyses. As significant discrepancies have repeatedly been shown between parents and youth with chronic pain report of physical and emotional functioning [75,76,77,78], caregiver-report was not included. However, to address this limitation, and to provide a more global picture of gains, the addition of objective measures to specifically assess functional gain in daily tasks and basic physical activity is recommended in future evaluations.[79] Finally, neither the IIPT nor the MMT have fully standardized protocols. Whereas the average length of stay (5 weeks) is comparable to other IIPT programs, the current program adopted a more individualized goal-based approach. More specifically, although the average number of intervention hours with key disciplines was similar in both the treatment as usual (MMT) and the intervention groups (IIPT), some variability in length of stay and professionals consulted was noted. In other words, some youth may have received a greater dose of a specific treatment than others, which influenced their outcomes. Furthermore, specifics associated with follow-up treatment recommendations, adherence to these recommendations, and other factors interfering with treatment (e.g., geographical distance, appointment cancellations) were not tracked. These factors and their impact on youth outcomes should be considered in future studies with larger samples sizes.

CLINICAL IMPLICATIONS AND CONCLUDING REMARKS

Specialized rehabilitation pain programs do appear to contribute to improving health-related quality of life for youth with pain-related disability. However, based on the findings of this study, IIPT seems particularly effective for youth for which pain interferes significantly on all relevant aspects of their lives, and impacts their engagement with social, cognitive, emotional, physical, and recreational activities. Regular use of pain interference questionnaires may be helpful in the early identification of these youth and assist a prompt recommendation of this treatment to families. Unfortunately, neither the IIPT, nor the MMT protocol seemed effective long-term in treating depressive symptoms in these youth. A focus on incorporating evidence-based interventions specifically designed to target these symptoms in this population into these interventions is required. Again, the identification of youth with chronic pain and co-occurring depression, in contrast to fear-based psychological conditions such as anxiety, may be useful in guiding parents and youth with pain-related disability to the most effective treatment. However, it also provides important comparative information to clinicians on the effects of another specialized rehabilitation pain treatment option (i.e. MMT). It is our hope that through presenting not only the statistical differences, but also the relevant clinical impact of these treatment options, that our study findings will provide clinicians with much needed evidence to help guide parents in selecting the best option for their child’s condition. Furthermore, such effectiveness studies create a foundation for much needed economic evaluations of the interventions. Based on the findings of this study, clinicians can feel confident in recommending IIPT for youth struggling with moderate to severe pain associated disability, with minimal depressive symptoms. For those not fitting this description, further research is required to identify the treatment that best meets their particular needs.

SUPPLEMENTAL MATERIALS

LMM specifications

We used a hierarchical inclusion method for independent variables and therefore, included variables one-by-one in the model, starting with variables with highest effect-size in univariate analysis. Variables were kept in the model in they showed significant association with mean score difference or progression rate. The restricted maximum-likelihood estimation method was used.

Random effects were included in the model to correct for autocorrelation of repeated measures throughout time in same patients and heterogeneous time lapse between follow-up visits (0-3-6-12 months). A spatial power (SP(POW)) covariance structure was used for repeated measures with an unstructured covariance term for random intercept and slope (when adequate). Random effects covariance structure choice was based on model fit statistics (Akaike Information Criteria and Bayesian Information Criteria).

LMM Validation

Models were validated for normal distribution of residuals and absence of heteroscedasticity. Evaluation of the normality of residual was undertaken with an integrative point of view, basing assessment on visual evaluation of diagram of distribution, Q-Q plot, normality testing (Shapiro-Wilk, Kolmogorov-Smirnov, Cramer-Von Mises and Anderson-Darling), Kurtosis and Skewness indexes and correspondence of median to mean.

In all cases, models residuals showed significant departure from normal distribution that was thought to be caused by non-linear relationship of score-difference change to time, particularly in the IIPT group. At visual assessment of the outcome progression through time, the IIPT group systematically showed an inflexion point at 3 months, separating two linear functions. To correct this situation, we included a dummy variable to identify time periods before and after 3 months, and computed two different slopes, resulting in a significant improvement of normality of residuals (final models). However, a slight departure from normal distribution still persisted for the majority of model residuals after this adjustment. As mentioned by Warrington and colleagues,[80] LMM's offer robust computation of independent variables estimated effects, even in the presence of departure from normal distribution, but confidence intervals of adjusted means can be unreliable in this situation, which could cause a type-1 error inflation.[80] Authors suggested that bootstrapping adjusted means and 95% confidence interval may improve reliability of adjusted means dispersion estimation and therefore reduce the chance of type-1 error when comparing adjusted means for both groups at specific timepoints.[81-83]

Results

Evaluation of the normality of residual was undertaken with an integrative point of view, basing assessment on visual evaluation of diagram of distribution, Q-Q plot, normality testing (Shapiro-Wilk, Kolmogorov-Smirnov, Cramer-Von Mises and Anderson-Darling), Kurtosis and Skewness indexes and correspondence of median to mean. Results from initial and final models are presented in table S1.

Bootstrap estimation

A random sampling of patients (with their associated follow-up assessments) was undertaken to create 1000 databases with the same number of subjects as the initial database. LMM analysis was performed on each individual random-sampled database and pooled confidence intervals were computed for adjusted means of groups differences at 3 and 12 months.

Results of bootstrapped adjusted means according to groups at 3 and 12 months are presented in Table S1. Both means and confidence intervals are very similar to the results obtained from standard final models and conclusions remained the same regarding differences between groups. This highlights the strong resilience of LMM’s in presence of slight deviation of residuals from normal distribution.

Table S1.

Bootstrap Adjusted Means and 95% Confidence Intervals According to Treatment Groups at 3- and 12-month Follow-up

Outcome	Treatment group	3 months Adjusted mean (95%CI)	12 months Adjusted mean (95%CI)
PedsQL Total	IIPT	9.26 (5.63,12.88)	17.39 (9.67,25.10)
	MMT	8.90 (5.90,11.90)	22.28 (15.42,29.14)
PROMIS Pain Interference	IIPT	-3.59 (-6.39,-0.78) *	-4.51 (-10.37,1.35) *
	MMT	1.96 (0.24,3.69) *	6.07 (2.34,9.79) *
PROMIS Depression	IIPT	0.72 (-1.93,3.37) *	-0.83 (-6.60,4.93)
	MMT	3.68 (1.79,5.56) *	0.08 (-4.04,4.21)

Legend: *=Group difference IIPT - MMT reaching significance level of $p<0.0$

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Article 4

Preamble

Youth and parent perceptions of participating in specialized multidisciplinary pain rehabilitation: A qualitative timeline effect analysis

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Karen Hurtubise's contribution to this manuscript included its conceptualization, the literature review, the data collection, analysis and presentation of the findings and the writing of this manuscript. The other co-authors (Astrid Brousselle, Melanie Noel, Abbie Jordan, Nivez Rasic, and Chantal Camden) supervised and provided guidance during the various stages of the study, reviewed the results and provided feedback on the manuscript prior to submission. Jo White provided expertise on the use of timelines as a data collection tool, and in their analysis.

The six authors authorized the integration of this article into Karen Hurtubise's dissertation.

Résumé

Objectif: Le devenir des jeunes ayant une incapacité lié à la douleur suite à leur participation à divers modèles multidisciplinaires de réadaptation spécialisée est peu connu. Notre étude vise à décrire et comparer, à l'aide de ligne du temps, les effets perçus de ces interventions par les jeunes et leurs parents inscrits soit à un traitement interdisciplinaire intensif (TII) ou un traitement multimodal (TMM).

Méthodes: Onze jeunes et 5 parents ayant participé à un TTI, et 3 jeunes et 5 parents ayant participé à un TMM ont été recrutés. Avant l'entrevue, ils ont complété une ligne du temps portant sur leur parcours en lien avec leurs douleurs et leurs traitements. Avec leur ligne du temps comme référence, les parents et les jeunes ont ensuite participés séparément à une entrevue semi-structurée. Les entrevues ont été transcrites et analysées en utilisant une analyse thématique approfondie.

Résultats : Quatre thèmes ont été identifiés: 1) Lutte pour trouver une cause, un traitement, et pour poursuivre la vie, représente principalement comme les défis vécus entre l'événement causant la douleur jusqu'au début du traitement spécialisé; 2) les acquisitions et les perturbations, en lien avec les effets positifs et négatifs généraux de la participation au traitement, ressentis pendant la durée du programme; 3) le soutient et les nouvelles réalités, décrivant les évènements après la fin du programme qui étaient associés à l'intervention; et 4) la douleur et la vie, associées aux impacts finaux attribués au programme, et influençant où les jeunes se trouvent dans leur vie actuelle au moment de l'entrevue. Pour chaque thème, des sous-thèmes liés aux avantages et aux inconvénients des traitements ont émergés.

Discussion : Une description des trajectoires de traitements et des expériences des jeunes ayant une incapacité liée à la douleur ayant participé à un programme de réadaptation multidisciplinaire spécialisé a été détaillée. De plus, l'impact sur leur vie quotidienne et celle de leur famille a été mis en évidence. Non seulement les avantages de ces traitements ont été soulignés, mais les effets négatifs, jusque maintenant inexplorés, ont également été identifiés.

Abstract

Background: Little is known about pain and treatment journeys of youth with chronic pain-related disability and those of their parents in Canada.

Aims: To describe and compare the longitudinal treatment effects as perceived by youth and their parents enrolled in either intensive interdisciplinary pain treatment (IIPT) or multimodal treatment (MMT).

Methods: Eleven youth and five parents from the IIPT, and three youth and five parents enrolled in MMT were recruited. All were asked to complete a timeline of their pain and treatment journey, followed by separately conducted semi-structured interviews. Transcribed interviews were analyzed using reflexive thematic analysis, with their timelines used as a reference.

Results: The main themes generated included: 1) Struggling to find a cause, a cure, and to keep up, which spanned the period between the initial pain event to the beginning of specialized treatment; 2) Acquisitions and Disturbances portrayed the general effects experienced during the program; 3) Reality and Support depicted the events following discharge, which were associated with the intervention outcomes; and 4) Pain and Life represented the end results of the treatment as perceived at their current life juncture. Subthemes reflecting the benefits and detriments of each treatment also emerged.

Conclusions: Detailed description of the treatment and post-treatment trajectories of youth with chronic pain-related disability enrolled in specialized multidisciplinary pain rehabilitation, and the impacts on their lives were uncovered. Not only were the benefits of these treatments highlighted, but the detrimental effects were also unveiled, which had previously been unexplored.

Key words: Pediatric pain-related disability; specialized pain rehabilitation; treatment experience; timelines, qualitative method.

Introduction

Pediatric chronic pain is a complex medical issue. For a clinically important subset of youth, it results in severe dysfunction and worsening disability affecting their physical, emotional, and social well-being.[1-5] These youth experience challenges related to social development, peer interactions, and family functioning.[6-9] Parents of these youth report emotional distress, helplessness, and altered parenting experiences.[5,10-12] Due to the complexity of the pediatric chronic pain experience, comprehensive treatments, grounded in a biopsychosocial model and involving the expertise of an array of healthcare disciplines are required.[13] Specialized multidisciplinary pain rehabilitation, including outpatient multimodal treatment (MMT), inpatient or day-hospital intensive interdisciplinary pain treatment (IIPT), is supported as the treatment choice.[14,15] MMT consists of an amalgamation of medical (e.g., medications), physical (e.g., physical therapy, occupational therapy) and psychological interventions (e.g., cognitive behavioural therapy).[15,16] IIPT consists of a defined period (e.g. 4 weeks) of intensive daily physical, occupational, and psychological therapies, along with medical support focused on functional restoration and self-management.[13, 17]

The literature surrounding the evaluation of these treatments is expanding. To date, findings have relied on quantitative focused effectiveness studies, using quasi-experimental, non-randomized cohort designs without a control group. These studies entail administering a battery of self-report questionnaires to program participants at various time points, typically at baseline, discharge and shortly thereafter (e.g. three months).[13] Recently, the relevance of some of these outcome domains (e.g. pain intensity) has been called into question by stakeholders, and the lack of appropriate tools to measure some of these domains has been underscored.[18] Despite promising results demonstrated by these quantitative studies, many questions remain unanswered. For example, little is known about why some youth benefit more than others, why adherence to therapeutic recommendations can be problematic, and what aspects of treatment promote long-term benefits. More importantly, the negative effects, outcomes, and impacts of specialized multidisciplinary pain rehabilitation contexts have yet to be explored in the current literature, limiting the knowledge associated with iatrogenic effects.

To address these unanswered questions and to increase our understanding of specialized multidisciplinary pain rehabilitation, rigorous qualitative studies are required.[19,20] Few studies have targeted the treatment experiences of youth with pain-related disability[7,19,20] or those of their parents[21]. No studies have yet, to our knowledge, applied qualitative methods longitudinally, and explored the post-discharge outcomes and the longer-term impacts of specialized multidisciplinary pain rehabilitation, or compared the differences between the various treatment options.

In clinical research, the collection of patient narratives can generate open-ended and inclusive stories, which may underscore unanticipated ideas, and highlight previous unconsidered relationships, explanations, and solutions to clinical issues.[7] These stories include a plotline and characters and are reflective of ongoing meaning-making associated to a certain condition, depending on the chosen events and their chronological order.[7] Time is an important feature of the participants' story, defining and intrinsically weaving together an individual’s narratives, and helping create meaning from experience.[22] Timelines are visual depictions of life history and events which can provide context and structure to narrative interviews and allow interviewees to reflect longitudinally on their condition, their journey and experiences, using the temporal dimensions of the past, present and future. They have also been reported as useful for data comparison by placing a clinical problem in the context of other salient life events. [23,24] Using narrative timelines, this effect analysis study aimed to explore, in a longitudinal manner, chronic pain experiences and program effects, outcomes and impacts as perceived and experienced by youth with chronic pain and their parents, at least one year following participation in a specialized multidisciplinary pain rehabilitation treatment option (i.e., either IIPT or MMT) and compare them.

Materials and Methods

This study was part of a larger participatory program evaluation, which purpose was to assist decision makers in determining the future of an IIPT at a pediatric facility in Western Canada. This effect analysis study aimed to highlight all the program effects, whether positive or negative and whether they were attributable to the program. Effect analysis is a type of evaluation design, which aspires to uncover all the effects associated with an intervention, including those less explored and/or perceived as negative.[26]

Study Context

The program context involved three organizations: a) a tertiary care pediatric health and rehabilitation facility; b) a province-wide publicly funded healthcare organization; and c) a specialized school, located within the walls of the tertiary care facility, and part of the regional board of education. The family-centred care philosophy unified the organizations. The program included comprehensive clinics, associated outpatient multimodal treatment (MMT) services (e.g., psychology, physiotherapy, medical interventions, psychoeducation), and a day-hospital IIPT. In addition to coordination and administrative staff, the program incorporated a comprehensive interdisciplinary team (i.e., physicians, nurses, psychologists, physiotherapists, and a family counsellor), trained in pediatric pain. Furthermore, it shared a staff complement with the rehabilitation day-hospital services (e.g., recreation, occupational therapist, and program coordinator), and facility-wide services (i.e., teachers, spiritual care, art- and music therapy). The assistant manager of rehabilitation services oversaw the allied health and nursing human resources compliment. Stakeholders within a program context can challenge the establishment of common evaluation goals.[25] As part of the overall participatory approach, a 13-member advisory committee, composed of youth with chronic pain-related disability, their parents and other important stakeholders (e.g. clinicians, healthcare managers, and teachers) were involved in key decisions throughout the study cycle. As part of these decisions, the advisory committee completed a consensus-building exercise where six outcome domains were prioritized for measurement in this evaluation.[18]. These outcome domains included participation in meaningful activities, activities of daily living, mood and affect, roles and relationships, school engagement and self-efficacy. Validated measures were available for 4 of the 6 prioritized domains. To further investigate the remaining domains and explore additional impacts of the aforementioned specialized rehabilitation pain options, this qualitative effect analysis was conducted.

Study Design

An interpretive descriptive methodology, a qualitative design which aims to generate knowledge relevant to an applied health context,[27] was adopted. This methodology was chosen to gain insight into the way youth with pain-related disability and their parents described the effects (short-term), outcomes (medium-term) and impacts (long-term) of participating in one of two specialized multidisciplinary rehabilitation pain treatment options, and whether differences between the programs acknowledged.

Participants

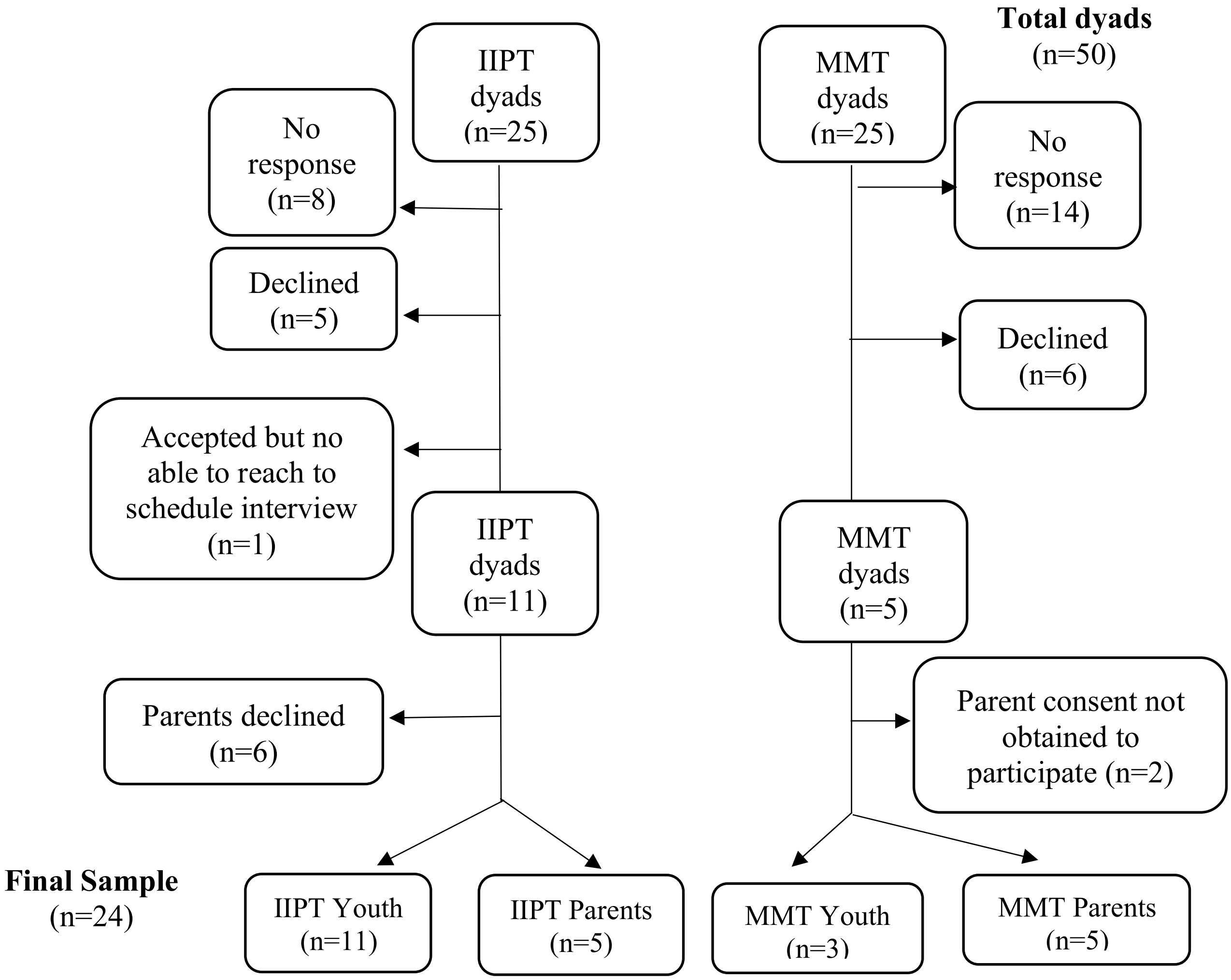
A purposive sample of youth with chronic pain and their parents was recruited through a tertiary-level pediatric complex chronic pain service. Purposeful sampling is widely used in qualitative research for the identification of information-rich cases and is an effective use of limited resources.[28] Fifty eligible parent and youth dyads (25 parents and 25 youth who participated in the IIPT, and 25 parents and 25 youth involved in the MMT) who acknowledged an interest in participating in research during their initial clinic team visit, were contacted (up to 3 times) by the research team using parent provided contact information.

Potential participants were excluded if they were in an acute diagnostic stage where all "organic" or disease-related causes for their pain had not been reasonably ruled out (e.g., cancer) or if they presented with a psychological condition (e.g., psychosis) for which admission to a specialized psychiatric program was recommended. Youth were eligible to participate if they were 12-18 years of age (i.e., the age range of the IIPT program), could follow verbal instructions in English, had no underlying disease that could explain their pain, had reported pain for at least 3 months (in accordance with the definition of chronic pain endorsed by the International Association for the Study of Pain),[29] and, when screened by the clinical team, met the established pain-related disability criteria (i.e., repeated school absenteeism, withdrawal from leisure and sporting activities, and/or difficulty with mobility, daily hygiene, or other activities of daily living). Youth were required to have participated in one of two specialized multidisciplinary treatment options at the facility, i.e., either the IIPT or MMT. Parents were eligible to participate if they were the youth's legal guardian.

As depicted in Figure 1, 68% of the potential sample did participate. Of those who did not, many did not respond (n= 22 dyads), others declined (n=11 dyads), while another agreed to participate, yet could not be reached to schedule the interview (n= 1 dyads). Although limited time was a cited reason for declining to participate in both groups (IIPT n=4; MMT n=1), other reasons varied slightly between the treatment options. For the IIPT, one youth was hospitalisation for mental health crisis (n=1), while for the MMT, frustrating experience with the program (n=2); pursuit of other interventions (n=1); parent hospitalised (n=1); and participation in too many studies already (n=1) were other cited reasons. Of the 19 remaining, six IIPT parents declined participation themselves, yet their child agreed to participate. Although most of these parents initially cited a lack of time to participant, many also acknowledged limited knowledge of their child's pain status since the program, as it was rarely a discussion topic. Conversely, in the MMT, two parents elected to participate instead of their child suggesting that youth involvement in the study would remind the child of their pain, the negative experiences surrounding it, or that the child was seeking other treatment to "resolve" their pain.

Figure 1.

Participant Recruitment



Twenty-four youth and parents were interested, deemed eligible, and completed, signed, and returned the online consent forms to the research team; 14 were youth (11 from the IIPT and three from the MMT), while 10 were parents (five from IIPT and five for the

MMT). Eight parent-youth dyads were present across our sample; five dyads representing the IIPT, and three in the MMT. Youth (M_{age}=16 years) predominant complaints were musculoskeletal and neuropathic pain (79%), yet some also presented with generalized pain (21%), and headache (14%). For most youth, symptoms had been present for over 12 months (93%). Table 1 provides additional socioeconomic (e.g. household income, marital status) and medical characteristics. When contextualizing our study, the sample characteristics present many similarities (e.g., sex, socioeconomic and marital status) to other qualitative studies in the field.[16, 30-34]

Table 1.

Participants Demographic and Pain Characteristics

	IIPT	MMT
Youth Participants	N=11	N=3
Youth's Age, years (mean)	16	16
Youth's Sex		
Female	10	3
Male	1	0
Youth's School Status		
Full Time	9	1
Part-time	1	1
On-line	1	1
Pain Location		
MSK	6	1
CRPS	2	0
Headache	1	1
Generalized	2	1
Pain Chronicity		
6-12 months	1	0
> 12-months	10	3
Time since participation in program, months (mean)	24	19
Parent Martial Status		
Married	9	2
Separated/Divorced	2	1
Parent Participants	N=5	N=5
Relationship to youth		

Biological mother	4	4
Biological father	1	0
Caregiver	0	1
Household income		
$30,000 – $59,999	0	0
$60,000 - $89,999	0	0
> $90,000	3	4
Do not want to answer	2	1
Marital Status		
Married	5	3
Divorce/Separated	0	2

Our sample size is also consistent with previous studies using reflexive thematic analysis.[8,35-36], those using qualitative methodology in the field of pediatric chronic pain,[8-10,21,37-40] and those using timelines as the data collection approach.[41-45] In addition to representability, our sample was also examined for extreme cases, in particular for characteristics related parent marital status and pain complaints to ensure similarities.[28] As recommended by current qualitative research standards, no attempt was made to achieve data saturation, as it is now recognized as an unhelpful concept.[46,47] As transparency in qualitative research is acknowledged as a marker of quality compared,[48] the focus was instead placed on sufficiently detailing the data collection process.[49]

Treatment programs

The IIPT comprised a 3-6-week day-hospital program (mean duration = 5 weeks). For six hours daily, five days per week, youth and their families participated in goal-oriented rehabilitation therapies (i.e., physiotherapy, occupation therapy, psychology, recreation therapy, art and music therapy, academic support), aimed at enhancing their pain management skills, facilitating their emotional adjustment and coping, and improving their physical functioning. Alternatively, the outpatient multimodal treatment (MMT) program combined self-management pain education sessions for parents and youth, as well as individual physiotherapy (e.g., functional stretching, strengthen, postural re-education and endurance training through physical activity), psychology sessions (i.e., using a cognitive behavioural approach), medical treatment (e.g. medication regimen), and return to participation in regular activity (i.e. return to school) for youth as clinically indicated. The aim of the MMT program was to also improve youth's self-management abilities, emotional coping and physical functioning, with minimal disruption to their regular lives.

Procedures

Approvals from the institutional research ethics board were obtained for all study procedures prior to initiating participant recruitment and data collection (Ethics #REB16-0916; 2017-1543). Once the consent process complete, youth and parents were contacted to schedule an interview and provided with instructions on how to create a timeline of the pediatric chronic pain journey, including their treatment experiences. A sample of the timeline development instruction and semi-structured interview schedule are provided in Table 2. As per protocols used in previous research,[22,23] the timeframe used was determined by the participant themselves, with encouragement to focus on the period in their lives when pain (or their child's pain) was a concern. The timelines ranged from 3 years to whole lifetimes (see example in Appendix 1) and most were completed prior to the scheduled interview.

Table 2.

Semi-structured Interview Schedule

Timeline Semi-Structured Interview Script	
General Instructions	Please draw a timeline of your life up until now and mark the most important events and the changes that have happened (asked about X days/weeks before interview?).
Prompts	Please tell me what was happening in your life at this time *(interviewer points to an area on the timeline).* Please tell me what was happening in your life immediately after the program (*interviewer points to an area on the timeline*). Please tell me what was happening in your life a few months after the program (*interviewer points to an area on the timeline*).
Potential Sub-Prompts	What was happening at home/in your family? What was happening at school? What was happening with your friends/relationships? How were you feeling? How do you think your pain condition and its treatment impacted this?

To ensure appropriate interpretation of participants' timelines, individual semi-structured interviews were conducted with each participant by one co-author (KH), who is experienced in qualitative interviewing in this setting.[18,50] Parents and youth were informed that KH was not involved in the delivery of services with either program, and that this work was associated with a doctoral dissertation. Using the timeline as an aide-memoire, to facilitating the recollection, sequencing and reflection of personal events, participants lead the interview process, with KH simply identifying events on the graph by asking, 'What happened here?' or 'Tell me more about this'.[22] Specific inquiries about school, family, peer relationships and other meaningful activities (e.g. sports, recreation, work) were used occasionally (as per the interview schedule sub-prompts listed in Table 2). In-depth interviews are designed to elicit a vivid picture of the participant's perspective on the research topic, and an effective method for stimulating 'talk' concerning a variety of topics, experiences, perspectives, personal feelings, and opinions, allowing insight into how people interpret, order and create meaning in their own worlds.[51]

The data collection procedures, including the timeline creation and the interview process, were pilot tested with the youth and parent members of the advisory committee guiding the larger study. The purposes of these pilots were to generate feedback on the technique and its relevance, identify the optimal time required for the interview, and test the appropriateness of the interview prompts. The feedback received was used to refine the procedure in the following ways. To facilitate inclusion, participants were provided with a range of locations where the interview could be conducted (i.e., their homes or a quiet room at the hospital) and methods were expanded from predominantly face-to-face interviews to include other media for information-sharing and communication technologies (i.e., telephone, FaceTime, Skype, and email). Participants were provided with the timeline instructions and the interview schedule prior to the interview to allow preparation, reflection, and to decrease anxiety. At the time of the interview, youth and parents were reminded that the interviews would be audio-recorded and transcribed verbatim, and permission was obtained for interview quotes to be published. Interviews ranged in length from 25- to 150 minutes. Reflective memos and field notes were maintained by the interviewer (KH) throughout the interview process, with a particular focus on situational, relational and performance reflexivity.[51]

The timeline approach has been reported as empowering for participants, allowing them to take charge of framing their own realities.[22,52] Moreover, it discourages researcher-research participant power imbalances that can exist and must be carefully managed, in particular when youth are involved.[22,52] This data collection procedure was chosen in response to guidance provided by the study's advisory committee. All committee members, including youth with chronic pain and their parents, agreed that understanding the relativity of the pain experience across time was important and timelines was a methodological tool which could be achieved this.

Data Analysis

Similar to previous studies using timelines, reflexive thematic analysis guided our data analysis.[41-44,53] More specifically, the six phases described by Braun and Clarke [54,55] were used. Firstly, familiarization with the data was achieved through listening to the interviews and active reading and re-reading of the transcripts. During this iterative familiarization process, initial codes were generated from data segments relating to the pain trajectory, the effects and impacts of either treatment program, and were then organized into potential themes. All IIPT youth interviews were initially analyzed, followed by parental interviews for this same intervention. Subsequently the same analysis process was conducted for the MMT group. Codes were grouped into time periods and then organized into negative or positive categories for each intervention. The time periods and categories were then contrasted across treatment groups to identify intervention group patterns. These initial coding and theming steps were completed by the first author, KH. Throughout this process, debriefing was conducted with CC, AB and MN to discuss the development and interpretation of themes. Themes were named, reviewed, refined, and concise definitions were generated for each theme with the assistance of CC, AB, MN, AJ, and JW. Agreement was achieved on the analysis and interpretations among co-authors, providing credibility and trustworthiness to the analytical data and interpretations processes. Themes were transformed into draft visual graphics, using a timeline as its foundation for reporting purposes. These graphic representations were reviewed by three interviewees in a member checking process to seek clarification, further explanation, and ensure accurate representation of their experience.[52] In keeping with the inductive thematic processes, frequencies of themes were not counted, as the importance or meaningfulness of a theme does not necessarily equate with frequency or quantifiable measures.[54] Throughout the analytical process, data were managed using QSR International's NVivo 12,[56] a computer-assisted qualitative analysis software package, to check the validity of the translation into graphic representation for the whole sample. Attention was paid to include quotations from various participants for representation of all perspectives within each theme.

Numerous steps were taken to address the quality of this research study and its qualitative analyses. Aligned with good qualitative research methodological practice and to enhance the credibility of this study, the authors' backgrounds, experience, and expertise in the field associated with this study, and their specific role in the analysis are of relevance and are therefore briefly described.[52,57,58] At the time of conducting the interviews and analysis, KH was completing a doctoral degree in health services research, and had many years of rehabilitation experience with youth with disabilities, including those with pediatric chronic pain. AB, an established researcher, with expertise in program evaluation and qualitative methodology, oversaw the effect analysis including the formulation of the evaluation question and review of the final analysis. JW, a researcher with qualitative methodology expertise, in particular timelines, provided guidance on the implementation of the timeline data collection tool, advised on the analysis, and reviewed the final analysis. AJ and MN, both clinicians and researchers with many years' experience of working in the field of pediatric chronic pain, and with expertise in the use of qualitative and narrative methods, provided advice on the data collection phase, reviewed initial timelines for content, helped guide the data analysis, and offered content expertise on the themes and final analysis. NR, with clinical expertise with the pediatric chronic pain and specialized multidisciplinary pain rehabilitation, also provided content expertise and reviewed the final themes. Finally, CC, who has many years of experience in pediatric rehabilitation, as a clinician, evaluator and researcher, using participatory and qualitative methodological and a variety of analytical approaches, provided advice during the data collection, reviewed the initial coding of the interviews and thematic scheme, and offered guidance on the final themes and their presentation. The use of both drawings and narratives further enhanced the credibility as the researchers triangulated the results across both mediums.[59] A member reflection process, a technique of discussing findings with participants, providing opportunities to question, critique and provide feedback was also used.[60] Finally, trustworthiness was ensured through the presentation of quotations from across a varied range of our participants accounts.[58]

Results

Participants' narratives were extremely complex, spanning many years, describing linkages between short, medium and long-term treatment outcomes. The themes generated for each of the interventions included the following: 1) Struggling to find a cause, a cure and to keep up; 2) Acquisitions and Disruptions; 3) Supports and Realities; and 4) Pain and Life*,* and were associated to a specific time period, relative to the youth's pain and treatment journey.

Struggling to find a cause, cure and to keep up related to the period before accessing specialized multidisciplinary pain rehabilitation services and was reflective of the consequences of the pain experience itself; *Acquisitions and Disruptions* occurred during treatment, ending at discharge and were associated with the immediate effects of participating in the treatment; *Supports and Realties* were linked to medium term outcomes surfacing months and even years following the intervention; and the *Pain and Life* related to participants current status, reflecting the longer-term impacts perceived to be a result of participating in one of the two programs. Figures 2 and 3 provide a graphic representation of all four themes, when the treatment occurred, and the relationship with the timing of the intervention.

As illustrated, the aforementioned four main themes crossed both treatment options, while the generated subthemes were specific to each program. The themes and subtheme for each intervention are further compared in Table 3, where the themes and their subthemes, along with a list of the codes are presented. A detailed description of each of the themes and subthemes, accompanied by representative youth and parent quotations illustrating their perspectives and experiences, is provided below. Additionally, the subthemes identifying positive and negative effects of treatment options and the final perceived program outcomes will be further contrasted. To protect participants confidentiality, pseudonyms have been used.

Figure 2.

Youth and Parent Participant Perceptions of IIPT Involvement

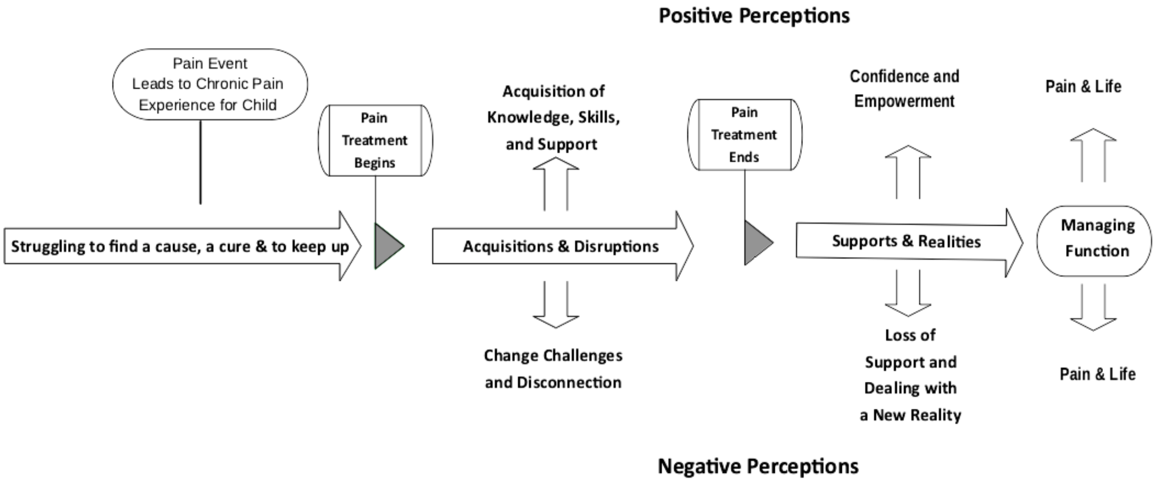


Figure 3.

Youth and Parent Participant Perceptions of MMT Involvement

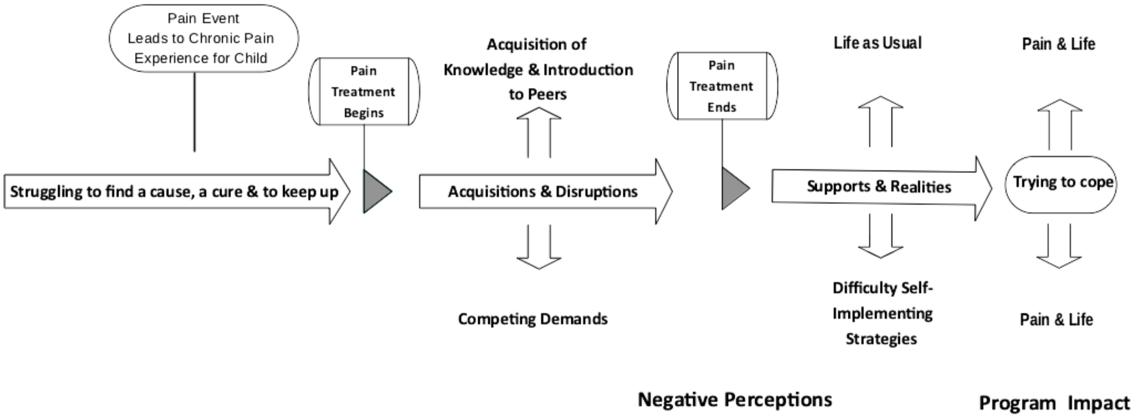


Table 3 presents a comparison of the themes and their subthemes, along with a list of the codes generated for each. A detailed description of each of the themes and subthemes, accompanied by representative youth and parent quotations illustrating their perspectives and experiences, is provided below. Additionally, the negative and positive outcome subthemes and the final program outcomes will then be further contrasted. To protect participants' confidentiality, pseudonyms have been used.

Table 3.

Comparison between the IIPT and MMT Themes and Subthemes

Time	**Theme**	**IIPT & MMT**	
Before specialized services	**Struggling to find a cause, a cure & to keep up**	***Codes*** 1. Increasing school and work absences 2. Loss of interest in hobbies and cessation of sports and physical activities 3. Loss of friends 4. Missing family outings and vacations 5. Lack of enthusiasm for school and social activities 6. Social isolation 7. Increased dependency on parents 8. Parental and family stress 9. Emergence of depressive symptoms 10. Initiation of negative pain coping cycle 11. Constant advocacy for specialized pain services	
During treatment	**Acquisitions & Disruptions**	**IIPT**	**MMT**
		Positive-Acquisition of Knowledge, Skills, and Support ***Codes*** 1. Meeting and daily interactions with peers and parents with similar challenges and experiences 2. Acquisition and integration of pain knowledge 3. Repeated practice of various pain strategies in a supportive milieu with readily available coaching 4. Parental training in coaching and support	**Positive-Acquisition of Knowledge and Introduction to Peers** ***Codes*** 1. Acquisition of preliminary pain knowledge and introduction to management strategies 2. Introduction to other peers and parents with similar challenges and experiences 3. Readjustment of parental expectations of their child.
		Negative- Change Challenges & Disconnection ***Codes*** 1. Physical and emotional exhaustion 2. Challenging to parent-child relationship 3. Absence from school and work 4. Distance and accommodation away from home 5. Missing family events 6. Communication challenges between parent unit 7. Challenges in meeting sibling needs	**Negative-Competing Demands** ***Codes*** 1. Frequency of school and work absence 2. Strain on youth and parent due to travel associated to and from multiple appointment 3. Difficulty access recommended services in some communities 4. Difficulty problem-solving and practicing pain strategies between appointments without close supervision
		IIPT	**MMT**
		Positive- Confidence and Empowerment ***Codes*** 1. Confidence in knowledge, skills and confidence to self-manage	**Positive-Life as Usual** ***Codes*** 1. Application of knowledge 2. Minimal disruptions to routine

Following Program	**Supports & Realities**	2. Empowerment in ability to attend school consistently and improved school attendance 4.Program legitimizes pain to school personnel and facilitates negotiation of accommodations 5.Emotional support from, and friendships with program peers 6.New peer networks & improved socialization skills 7. Discovery of new leisure and sport interests	3. Remain with peers and in community school
		Negative – Loss of Support and Facing a New Reality ***Codes*** 1.Lack of clinical support follow program 2.Back to school stress and impact on post-secondary plans 3.Loss of previous support networks and daily connections with pain peers. 4.Consequences of uncovering family issues and being unable to resolve them within the program timeframe 5.Loss of rigorous structure post-program and therefore some associated gains 6. Recognition of the lifelong permanence of pain condition.	**Negative- Difficulty Self-Implementing Strategies** ***Codes*** 1.Youth require parental support to self-manage 2.Frequent school absences due to pain 3. Continuation of negative pain coping strategies 4. Socialization only when pain and fatigue allow
Impact	**Pain & Life**	**IIPT**	**MMT**
		Managing function ***Codes*** 1. Living with pain, focus on function, and gaining control 2. Doing what ones wants to do, needs to do, is expected to do 3. Resumption of age-appropriate roles and responsibilities 4. Prioritization and choices 5.Pain in the background	**Trying to cope** ***Codes*** 1. Focus still on pain and symptom reduction 2. Pain remains a limiting factor to function, roles and responsibilities 3. Pain in the forefront and a heavy burden.

Theme 1. Struggling to find a cause, a cure, and to keep up

Struggling to find a cause, a cure, and to keep up captures the crux of participants' narratives related to their initial negative consequences of youth's pain journey. For most, the pain journey began with a specific event when the pain first emerged (e.g., injury, illness), while for others, it was marked by the time point (e.g., a specific date, association a life events (e.g., child's school grade, parent employment). For both intervention groups, participants described an increase in pain intensity, locations, or frequency between the emergence of the pain sensation and gaining access to specialized multidisciplinary pain teams. This pain amplification process was paralleled by intensified parent advocacy efforts for referrals, increased utilization of healthcare speciality services, and a multitude of investigations, prescriptions, conventional and complementary treatments. This participant recounts her experience:

> "*I was on a hike and I started to feel this pain. [The pain] kept increasing throughout the summer. We started to get it checked. I received physio, chiro, active release, and then I started getting X-rays and CAT scan. They could not find anything. I was getting super frustrated, but we kept going*" [Adelaide, 17 years, MMT youth].

The lack of answers received from healthcare professionals increased youth and parental frustrations, which was further exacerbated by youth's deteriorating function. Furthermore, youth and parent battled to continue with the demands and expectation of everyday life, while being progressively burdened by the deteriorations caused by the pain. Both parent and youth participants' narratives expressed a spiralling loss of function in all areas of life as time progressed. In addition to declines in physical function, they also portrayed deteriorations in youth's psychological well-being, family roles, and school and social engagement. This participant described her experiences:

> "*We went on a [family] trip to Florida. I could barely stand in line for the rides and I didn't have any fun. When I started a new school, I had a really hard time socializing and missed a lot of classes, because I was having pain. I started to be afraid to go to school. I wasn't sleeping at all*" [Olivia, 17 years, IIPT youth].

Parents also provided vivid accounts of their desperation during this time, which spoke not only to the impact on their child, but how the pain affected the immediate household, the broader family, and their community. This participant provided this powerful exemplar of the consequences of her daughter's chronic pain:

> *"[My daughter] was having three-hour panic attacks every night. I was massaging her, talking to her and really not having any skills and not knowing what I was doing. My anxiety was through the roof for two years. Of course, that affected the others [family members]*" [Rose, IIPT parent].

The theme Struggling to find a cause, a cure, and to keep up provides the contextual background for the other themes and their associated subthemes. In describing their perceptions of participating in treatment and the effects and outcomes overtime, youth and parents often made reference to this pre-treatment time period, as a means of underscoring the relevance of their statements, of giving meaning to the pain journey, and of justifying their treatment choices.

Theme 2. Acquisitions and Disruptions

The theme Acquisitions and Disruptions depicts the effects of participating in specialized multidisciplinary pain rehabilitation interventions. These immediate effects were related to many implicit and explicit factors including youth and family intervention expectations, the intervention itself, and family circumstances. Many youths, although cautiously optimistic, also expressed self-protective attitudes and a reluctance toward pursuing specialized multidisciplinary pain interventions due to previous disappointing or failed treatment experiences. One participant described her situation in this way:

> "*It was mostly my dad because I had missed so much school and he was very concerned. I wasn't really sure about it, because at this point, it had been years trying different things, and I was not 100% convinced that this would help*" [Danielle, 18 years, IIPT youth].

The reasons for choosing the specialized pain treatment, youth attitudes towards the intervention, their involvement in the decision-making process, and parental and youth treatment expectations influenced participants' perceptions of treatment participation and its effects, in particular whether a positive or negative label was assigned. Positive and negative effects were identified in both treatment options (i.e., IIPT and MMT); some similar, others quite different. In presenting these effects, comparative details will be added as appropriate.

IIPT Positive Effects: Acquisition of Knowledge Skills and Support

The Acquisition of Knowledge, Skills and Support subtheme was the most commonly cited positive effects and valued attribute of participating in the IIPT program. Gaining knowledge and having the opportunity to apply it to daily life situations were perceived as the cornerstones, and most valued activities of the IIPT. More specifically, participants articulated the importance of pain education during the program in improving their understanding of pain (e.g., mechanisms, triggers), and the strategies to manage it. This participant explained her feelings as follows:

> "*At the end of it, it felt like so much had changed. I didn't have a lot of pain anymore and when I had it, I had an understanding of why and how I could manage it*" [Dominque, 19 years, IIPT youth].

Parents also acknowledged the benefits of sharing the same knowledge as their child, which included a better understanding of the pain, and the strategies their child had been taught. Additionally, many parents reported gaining an awareness of the negative contribution of their own behaviour on their child's pain and recognized a need to change their parenting style and acquire different skills. The positive effect of the simultaneous acquisition of pain knowledge and skills by parents and youth was vital in changing parenting behaviours and enhancing youth's ability to cope with their pain. As this participant explains it, the knowledge and skills acquired changed the communication within the parents-youth relationship, creating a new language to use between them, as well as with the families and their community members:

> "*Understanding chronic pain, learning about the beast that it actually is, and what it looks like in the future, learning how to communicate about it, was a huge help. What it offered our family was the language to understand, communicate, about, to learn to accept the pain*" [Rose, IIPT parent].

The supportive milieu created during the IIPT was also viewed as a positive effect of participating in this intervention. Peers and staff support were both acknowledged as beneficial. Parents and youth recalled meeting others facing similar challenges with much fondness. Phrases such as "no longer feeling alone or isolated,", and "feeling understood" permeated their discourse. As captured in this participant quotation, many also highlighted a sense of feeling accepted and not judged in this peer context:

> "*[Meeting other parents] is very therapeutic. You talk about certain things you can't talk about with other parents. Even though we have group therapy, where the parents get together, it was based on clinically scripted questions. When you are just chatting, there are things we are able to say that we might not in a group setting. There is a comfort with what we can say, being able say it, and not be judged*" [Alice, IIPT parent participant].

Policies limiting contact and communication between IIPT program participants during treatment were criticized by most youth and parents. Other benefits attributed to participating in the IIPT peer-group programming included increased motivation to challenge oneself through friendly competition, the development of empathy, the recognition of others' struggles and a focusing on successes, as well as shared learning.

In addition to peers, the support of IIPT staff was also identified as a positive effect of participating in this program. Many participants described vivid examples of clinicians providing youth with support during challenging times, (e.g., pain flares), modelling appropriate coaching behaviours for parents, which were perceived as vital to both parent and youth skill acquisition. One mother described how it transpired for her:

> "*I was hesitant because I didn't know medically how far to push [my daughter]. [The physio] knew how to push [my daughter] through. I didn't know if I could do it without hurting her. Witnessing [the staff] being able to push [my daughter] through things was helpful. The next pain flare that she had, I just talked her through it. And she was able to use some of the strategies she had learned, and she got herself through a really bad spell. She just needed a little bit of coaching from me*" [Alice, IIPT parent].

IIPT Negative Effects: Change Challenges and Disconnection

The IIPT negative effects subtheme, Change Challenges and Disconnection, depicted the extreme effort required to change behaviours and the sacrifices required to do so. The program intensity and the physical and emotional effects required to participate were the most commonly cited challenges. Youth and parents both recognized the intense, time-limited structure, and the very high participation expectations as incredibly difficult to manage. This participant provided this vivid memory of completing the IIPT program:

> "*I was completely exhausted at the end of IIPT. They just worked us so hard. It was mentally exhausting, physically exhausting, emotionally exhausting. So, I would say that the immediate effects were kind of negative*" [Mila, 19 years, IIPT youth].

Some IIPT parent and youth participants also ascribed negative effects related to the disconnection from their regular lives and daily routines required to participate in the IIPT option. For those who lived at a significant geographical distance, the burden of being away from their partners, siblings, other family members, in addition to missing important events and celebrations were perceived by most parents and some youth as negative effects associated with treatment participation. More commonly, however, youth highlighted the loss of regular connection with their peers and academic milieu. As expressed by this participant:

> "*I found so much support at school with my friends, so it was really hard to be away from my support system during the program*" [Luisa, 18 years, IIPT youth].

Some reported making efforts to maintain contact with friends on weekends, while others used technology (e.g., group video games), social media and text messaging in an attempt to remain socially connected.

In addition to family and social disconnection, being away from school for the intensive period of time required for IIPT engagement was perceived as one of the most significant burdens recognized by participants. Factors such as the timing of the program, participant's grade level, prior school attendance, and youth's involvement in making the decision to participate in the IIPT influenced youth perceptions. This participant described her struggles:

> "*I definitely found it difficult. I was taking a full course load during the program and, because I was taking grade 12 courses, if you're not there, you have to do double or triple the work to catch up*" [Danielle, 18 years, IIPT youth].

The negative effects of treatment participation on parents' lives were also underscored. Loss of productivity associated with work, communication challenges, and compromises associated with parental duties were commonly cited challenges. All IIPT parent study participants highlighted that at least one parent within their household worked part-time, as the full-time family caregiver, or owned their own business. This flexibility was acknowledged as being key in affording these parents and their child the opportunity to participate in the IIPT. Furthermore, empathy was shown towards families for whom these arrangements were impossible, and for whom a significant participation burden was perceived (e.g., distance from home, single parent).

MMT Positive Effects Subtheme: Acquisition of Knowledge and Introduction to Peers

Acquisition of Knowledge and Introduction to Peers was selected as the subtheme title for the positive effects of participating in the MMT interventions as it encompassed the most valued activities identified by these participants. In contrast, to the IIPT, most MMT parents described a full one-day session, where pain education was provided to parents and youth separately. Meeting peers was important in validating youth's pain experience and in fostering resilience. Furthermore, parent participants identified key messages that they integrated during the sessions, as well as those that resonated with their child. This participant provided this example:

> "*[My daughter] met a whole bunch of other kids who are living with pain the way she is. I think it helped to know she's not alone. But she also realized living with pain is tough. and that sometimes you just have to suck it up and push through*" [Elena, MMT parent].

Similar to the IIPT, parent participants also acknowledged the benefit of meeting other parents "that are trying to navigate what it looks like to be a parent of someone in pain" [Elena, MMT parent]. Furthermore, they underscored the benefits of learning the same information and strategies as their child and gaining an awareness of realistic developmentally appropriate expectations to have of their child, despite their pain condition. Their queries ranged from school attendance, to social engagement, to family roles. One parent expressed her learning this way:

> "*Still expecting him to do chores. They said, you need to stop doing everything for him and make him do some of it himself. Because he has to learn to live with this pain. That I didn't think of and I know the other parents there didn't think of it either. So then, [my son] and I had a talk and I said, OK, I learned this; you learned that, this is what we are going to do. That helped me a lot*" [Delphine, MMT parent].

As exemplified in this quote, much like the IIPT participants, the MMT parents also underscored an awareness of the need to change overprotective and solicitous parenting responses to child pain. Furthermore, they also acknowledged the pivotal role of the pain education in creating a common language and understanding between them and their child.

MMT Negative Effects: Competing Demands

The subtheme Competing Demands designates the MMT negative effects, portraying the clear tensions reported by youth and parents in their attempt to adhere to treatment recommendations while trying to meet regular life expectations. Unlike the IIPT where youth and parents who were disconnected from their lives for a specific period, for MMT youth and parents, treatment recommendations required inclusion into their already very full and busy lives. Weekly appointments and the associated travel as well as difficulty accessing suggested services when and where they were most needed were commonly reported challenges. Appointments and the travel were identified as important impediments to school and work attendances and productivity. For some families, the distance they lived from pediatric specialized pain rehabilitation services, and the lack of specialized services (e.g., pediatric psychology) in their communities created additional challenges and dilemmas, as explained by this participant:

> "*There were recommendations from [the team] and I just didn't know where to go and who were experts in dealing with children. I didn't want an adult specialist counseling my 13-year-old. I want someone who specializes in adolescents and that doesn't exist in [my community]. Due to work, there were times when I had to rely on my parents to make a few trips up to [the city] for me. It was difficult but we did our best*" [Fleur, MMT parent].

Although distance was also mentioned as a challenge for IIPT participants, in their case it was related to being separated from loved ones for a period of time, and less about the repeated distance travelled and associated productivity loss (i.e., work and school time). Furthermore, lack of access and availability of local specialized multidisciplinary pain rehabilitation services was identified by some families as a precipitating factor in opting for the IIPT. Finally, many youth participants in the MMT also discussed the difficulties encountered in applying pain management knowledge and strategies into their everyday lives, as this participant expressed:

> "*They offered me physio and then psychology, every 2 weeks, back to back. I would see them both in the same day so I wouldn't have to travel multiple times. However, I would learn a strategy and then would often have to wait until I did something active to practise the strategy again and then, say if I had pain or if I was struggling, I would have to wait for two weeks before having help again*" [Sabrina, 17 years, MMT youth].

In contrast, this lack of practice and access to clinical support when needed was not raised in the IIPT. Instead, the ability to practise learned skills and receive timely feedback was underscored as a positive effect of participating in the IIPT option. These positive and negative effects of treatment participation often influenced youth and parent participant perceptions of the longer-term outcomes and impacts.

Theme 3. Supports and Realities

The theme *Supports and Realities* defined the more sustained, profound, and longer-term program outcomes as perceived by those enrolled in pediatric specialized multidisciplinary pain rehabilitation. Their description often referred to the consequences of youth's chronic pain prior to accessing specialized multidisciplinary pain rehabilitation, and often built on the effects of their participation in treatment. This is evidenced in the description by this participant:

> "*I can completely handle my pain now even though I still have it sometimes. I have a physical and mental strategy to completely get [my pain] under control. Up until the program, [my pain] was essentially like a massive obstacle that was unscalable. Now, if pain is becoming an issue, I can deal with it. I also had social support coming from the IIPT. It gave me like my first toolkit of adult social skills*" [Adrian, 19 years, IIPT youth].

These sustained program effects were often explained in relation to intervention goals, participants' interpretations of their life situations, including their pain status, and the milestones they had attained and still hoped to achieve. These relationships influenced whether a positive or negative identifier was assigned to the program outcomes.

IIPT Positive Outcomes: Confidence and Empowerment

The subtheme of Confidence and Empowerment describes the positive impact highlighted by youth and parent IIPT participants of the belief in themselves and their ability to manage the pain, and subsequently control their life. Gaining confidence in self-management was described as the most positive outcome of the IIPT program. Participants described achieving this confidence by acquiring the required knowledge and skills (a recognized positive effect of treatment participation), and essential in achieving self-management. One participant explained the changes he observed in his son:

> "*I think the program changed him because he got a lot of confidence and I think he understood the nature of his pain and how it was affecting him. And above all, what he could do about it*" [Jean, IIPT parent].

For youth, experiencing success in the application of the knowledge and skills to various contexts, and problem-solving through challenging, atypical, complex or unplanned situations was particularly helpful in creating this self-reliant belief. Some IIPT participants reported having setbacks in pain management and relapses since their discharge from the program. Setbacks were associated with a series of emotionally difficult life events (e.g. death of a family member, repeat injury). However, despite these, youth, along with their parents, expressed self-confidence in their abilities to return to self-management. This self-confidence was also fuelled by their sustained peer support network created during the IIPT. Many youths and some parents reported having maintained contact with other IIPT participants. They highlighted that these relationships were not only founded on shared pain experiences, but also interests outside of pain (e.g., shared education goals). Moreover, youth credited their IIPT peer-support network with enhancing their coping skills and helping maintain their physical and psychological well-being following the program, even months and years later. As one participant shared:

> "*Following the program, I had a depressive relapse. But I had two close friends from the program, and so I had social support. It was a really small episode, much shorter than others I had had. It gave me confidence to know if something goes wrong, I still have people that I can fall back on, besides my family*" [Adrian, 19 years, IIPT youth].

The enhanced social skills and re-establishment of meaningful peer relationships following the program were also attributed to the development of social networks during the IIPT program. In addition to social skills, many participants associated their new peer networks with new interests, hobbies, and meaningful activities, first experienced during the IIPT. This participant provided this explanation:

> "*A huge challenge for [my daughter] was to connect with people who weren't active and involved in [her sport], but she still found interesting. One thing she discovered through the program was music. She was able to connect with other friends in her love of music that allowed her to find other ways to connect*" [Sophia, IIPT parent].

Youth's enriched social capacities empowered them to develop and refine their self-advocacy skills. More particularly this applied to advocating for modifications and accommodations in school, drawing on supports they had received during treatment. As one participant described it:

> "*Accommodations have allowed me to, if it's a written exam, I can type it. Writing that much by hand is too much for me. I requested [the accommodation] actually coming into university because I knew that I relied heavily on them in high school. One of the doctors in the program filled out the form for me that the university required, and the people at the accessibility centre were very understanding*" [Luisa, 18 years, IIPT youth].

As highlighted in Luisa's narrative, support was required to accompany these requests. Many youth acknowledged that past participation in the IIPT was enough to validate their condition to the outside world, specifically to people who were skeptical about the existence of their pain.

IIPT Negative Outcomes: Loss of Support and Facing a New Reality

The subtheme Loss of Support and Facing a New Reality described participants' negative experiences and feelings associated with leaving the protective nurturing IIPT milieu and transitioning back to the expectations of everyday life. Many parents and youth acknowledged the significant struggles of this transition. As expressed by this participant:

> "*[My daughter] came from alienation, having an invisible condition and people not believing her, to this beautiful little bubble, the program, where she was validated, supported, and encouraged, and then dropped back into the real world. I know that part of the program is to teach you to live life independently with your pain. But she was riding a high, ready to conquer the world, with nothing but supportive people around her. And then there was nothing. It was a bit tough*" [Alice, IIPT parent].

Moreover, some participants expressed anger towards the program staff, sharing feelings of perceived abandonment, in particular if issues identified during IIPT (e.g., family conflict) were perceived to be unresolved prior to discharge.

Negative program impacts were also associated with the repercussions on school and social engagement. IIPT youth narratives were peppered with multiple examples of the challenges they faced when returning to school following treatment. These included feeling forgotten by classmates, undervalued by teachers, and the excessive and overwhelming academic "catch-up" required to meet academic performance targets (e.g., course credits). Some of these impacts had long-term negative impacts on youth post-secondary academic path and the achievement of their ideal career, as exemplified by one participant:

> "*I had a really good average coming out of high school, but those 2 classes I took during the IIPT set my average back. I really wanted to get into the Neuroscience Program. But when I got the marks back from 2 exams, I wrote during the IIPT, they were quite a bit lower. I didn't get into Neuroscience and instead settled for Biology*" [Luisa, 18 years, IIPT youth].

In addition to highlighting the negative outcomes on school engagement, some participants also discussed the negative impact on peer relationships. For example, some youth noted that new peer groups had formed during their absence and "private jokes" had emerged, which they failed to understand. These situations were often referenced in relation to the amount of time spent away from their peer group and had the unfortunate effect of leaving many with feelings of exclusion, isolation and alienation all over again. These negative program impacts were often not as evident in the discourse of MMT participants, and the positive program impacts also varied widely.

MMT Positive Outcomes: Life as Usual

The subtheme Life as Usual highlighted in MMT participants' reflections on the minimal perceived disruptions caused by the treatment to their regular lives. Most participants normalized the daily accommodations made for pain in their descriptions of their routines. Their descriptions focused on their lives within their communities, including school, peers, and family life, and were less focused on the hospital services received as compared to those in the IIPT. Parents and youth provided examples of the positive outcomes of MMT. More specifically, it allowed them to create partnerships and alliances with local school personnel which were perceived as vital in creating an academic plan that worked for the youth. Most parent narratives identified a member of the school personnel (e.g., principal, guidance counsellor, teacher) who was instrumental in facilitating an individualized plan, and who had taken the time to get to know the child, their condition, and their academic capabilities. Furthermore, the use of a combination of learning methods, in particular online courses, was more common in this intervention group than the IIPT. This participant provides this description:

> "*I just really can't get up at the correct time to get to school in the morning because of my pain. I have to sleep a little bit longer and I can because I do some online*

> *schooling instead of morning classes. So, I do a lot of my schooling at night instead. I have one class at [the community school]. It's the last class of the day and is my option class*" [Brittany, 17 years, MMT youth].

Similar to the IIPT, the knowledge acquired in participating in the MMT program was applied by youth in identifying and proposing accommodations that may help them in school. However, unlike the self-advocacy noted in the IIPT youth participants, parents often negotiated the accommodation with the school administration instead of the youth themselves.

The maintenance of peer relationships in and outside of school was also noted to be a positive impact of the MMT program. Despite highlighting similar peer-related issues (e.g., bullying, teasing) as those enrolled in IIPT, most MMT youth participants made references to long-standing friendships, with either one individual or a small group of peers. This participant described her friends like this:

> "*I always had close friends, actually a close group of friends. The therapists I saw suggested that strengthen my entire leg would help my foot. My friend also just wanted to start something new, so she came with me. We often go to a gym together and have a personal trainer now*" [Adelaide, 17 years, MMT youth].

MMT Negative Outcomes: Difficulty self-implementing strategies

Difficulty self-implementing strategies describes youth limited ability to integrate pain strategies into their daily lives and was perceived as a negative outcome of the MMT intervention. In comparison to the IIPT program, MMT participants' pain knowledge was more superficial and more parental input was required to assist youth in managing their pain. For example, some participants had difficulty naming triggers that exacerbated their pain, as demonstrated in this example:

> "*My mom thinks that when I'm stressed [my pain] gets worse. I have no idea*" [Adelaide, 17 years, MMT youth].

Furthermore, examples of effective implementation of management strategies to foster function were rare and evidence of negative pain coping strategies remained. The lack of effective pain coping strategies negatively impacted school attendance and social engagement as highlighted in this participant's description:

> "*Last semester [my daughter] missed 51 days of school. When she has a pain spike, she cannot get out of bed. She had to drop out of a bunch of classes last semester. She's now taking online courses*" [Elena, MMT parent].

This participant provides insight into her limited ability to socially engage due to ineffective implementation of pain strategies.

> "[*My pain] can keep me away from people sometimes, and sometimes I have to bail on plans. I've got some very close friends that I keep very dear to me and they understand that sometimes I can't keep a plan and they get that. So, we just reschedule*" [Brittany, 17 years, MMT youth].

Many other examples of youth's reliance on their parents for pain management were reported.

Theme 4. Pain and Life

The theme Pain and Life represented the perceived final impact of pediatric specialized multidisciplinary program options. These end results were quite different between treatment options, and therefore will be presented separately.

IIPT Final Impact: Managing Function

Managing Function was identified as the final impact subtheme for the IIPT within the Pain and Life theme. It depicted the focus of IIPT youth and parent participants on their perceived function as a result of the program, and their shift away from symptom reduction to one of living well despite their pain. When making reference to their function, participants included examples of what youth needed to do, were expected to do, and wanted to do. For many youths, although some level of pain, or intermittent discomfort remained, it was rarely mentioned in describing their current daily lives. Youth self-management skills went beyond managing their pain, to include gaining control over their pain and enabling them to fulfill most age-appropriate expectations, roles and responsibilities. These expectations included family responsibilities, academic obligations, and for some social commitments. Compromises and prioritization of demands were often required to achieve the level of function needed. Despite their ability to manage their function, the complexities of chronic pain and its consequences remained in the background. This participant stated:

> "*Because of the IIPT, [my daughter] is a high-functioning kid with pain. But every day she doesn't feel good and every day is hard, and it's a slog*" [Rose, IIPT parent].

MMT Final Impact: Trying to cope

The subtheme Trying to Cope was selected as the final impact for the MMT option as it represented the influence pain still had on participants' day-to-day lives, their ongoing focus on symptom reduction, and the acknowledgement of attempting to regain control. In providing descriptions of their day, many participants emphasized pain as the limiting factor in fulfilling family, academic and social expectations, roles and responsibilities. Furthermore, pain was more often referenced in their narratives. For most MMT participants, pain took centre stage in their lives, still exerting much control over how they navigated through each day. This participant stated it this way:

> "*I just have to push through and get done what I want to get done*" [Adelaide, 17 years, MMT youth].

As evident in this quote, pain remained a heavy burden, an obstacle to overcome to live life.

Discussion

The purpose of this study was to longitudinally explore and compare the effects of specialized multidisciplinary rehabilitation programs as perceived by youth participants and their parents. Some of our study results confirm existing research findings. These include recognizing that pain consequences reach far beyond the youth themselves, affecting parents, families, and peers.[9,10,20,21,39] Furthermore, the functional losses in all life domains, including worsening physical and emotional well-being, and health-related quality of life were also evident.[9,10,11,20,21,37-40,61] However, novel to this study is the recognition of how the consequences of chronic pain prior to accessing specialized multidisciplinary pain rehabilitation established the relevance for treatment participation effects, program outcomes and final impacts. More specifically, each individual's journey integrally influenced how and whether they benefitted from their chosen treatment option.

Our study underscores shared as well as unique positive and negative perceptions attributed to the two treatment options, as well as valued treatment components within each option. Acquiring knowledge and interacting with peers facing similar chronic pain consequences were the primary benefits common to participants in both programs.

Moreover, they were viewed as instrumental in gaining the ability to managing the pain, for youth and their parents. Although pain education has been acknowledged in the adult pain literature,[61] evidence in pediatrics is nascent.[15,19,63,64] The effects of pain education may be better understood when associated with peer interactions,[19] also recognized as a positive treatment effect in our study. Group treatment reportedly creates an environment for normalization, for sharing experiences, and for reflection on one's own circumstances in contrast to others.[19] However, concerns have previously been raised about the potential of peer interactions contributing to youth further identifying with the sick role, fostering relationships founded solely on health and pain issues, and lead to peer contagion.[65,66] Along with Forgeron and colleagues,[67] our findings refute these claims, demonstrating that peer relationships are not necessarily sustained on the common pain experiences alone, but also resulted in the sharing of common interests outside of it. Researchers have described a curative aspect of connecting with peers, feeling understood for the first time and a sense of belonging.[20] Our study participants credited the treatment milieu and its effects as enabling knowledge acquisition and skill mastery, reducing feelings of isolation, and enhancing coping and self-management skills.

Parents and youth participants also reported confidence and empowerment, pivotal to self-management, as the most common program outcome of IIPT. In addition to restoring parent's confidence in their parenting, even in the presence of persistent youth pain, IIPT parents felt empowered as a result of gaining the knowledge and skills to help their child cope with the pain, a finding supported by previous evidence.[21] The IIPT youth participants in our study also reported enhanced belief in their capacity to better manage their pain and to have more control over their lives, and expressed a renewed sense of hope and confidence associated with greater self-efficacy at discharge. Similar findings have recently been reported in a previous qualitative study, where six youth expressed a renewed sense of hope, improved confidence, and self-efficacy at IIPT discharge.[20] The IIPT participants in our study provided vivid examples of how this had been accomplished since the program's end. On the other hand, MMT participants did not express the same level of commitment to self-management or degree of self-efficacy. This may be linked to adherence difficulties associated with MMT programs, as expressed by our MMT participants and reported by other authors.[68]

Perhaps even more important than the positive perspectives, our study findings also underscored some negative effects, outcomes and impacts specific to each treatment option, which have previously been underexplored. IIPT participants felt disconnected from their social and academic communities due to the lack of daily interactions with their networks and communities during treatment. This is an important consideration when recommending IIPT to potential participants, as these networks are deemed critical to this age group.[69,70] Moreover, maintaining friendships may be a way for youth with chronic pain to preserve their identity.[9] In addition, disconnection from school, an important context for academic, cognitive identity, independence, and social relationships development,[71] was often reported as a negative treatment effect by our IIPT participants, but also by MMT participants experiencing frequent pain episodes and treatment appointments. Youth in both interventions raise education concerns and academic achievement worries. As a result, and as recognized by other researchers, these concerns place this group at risk of dropping out of school, exacerbating self-esteem issues, and possible role loss within society.[9] However, our study confirmed that sharing relevant information to receive the necessary modifications from the school administrators and teachers can result in better reintegration of these youth, in their roles as students, peers, and members of society.[9] In highlighting their academic disconnection, some IIPT youth participants in our study also underscored the perceived negative outcomes of treatment on their school performance, and its contribution to altering post-secondary and career paths. This finding is consistent with the reported perception that youth with chronic pain feel they lag behind their peers in school progress and employment,[72] even if some studies suggest that they are on track developmentally on milestones such as school graduation, college attendance, and independent living.[73] Further research with older adolescents and young adults is needed to examine educational and vocational outcome related to intervention options, due to its significant impact on the future socioeconomic status and financial independence of these youth.

Finally, many youth IIPT participants and their parents experienced a challenging transition back to daily life after the program. The loss of support from program peers and staff was described as abandonment, and detrimental as youth and parents struggled to adapt their knowledge and skills to real life situations without support. Additional discharge planning and preparation for the transition back to their community have been suggested in the literature and underscored as a need for focus post program.[74] Exploring the role of comorbidities such as anxiety, depression, or post-traumatic stress disorder following program completion also deserves more attention, for which potentially additional or even different interventions may be required.[75] Booster sessions three months after IIPT admission may also be important to sustain improvements.[76]

A strength of this study lies in the exploration of both youth and their parents' perspectives, given their unique needs. The qualitative nature not only provided an opportunity to better understand the processes of change and effects of two specialized multidisciplinary pain rehabilitation options, it also offered important clinical information on how to improve care and the program components most valued by participants. The innovative methodology, using timelining followed by in-depth interviews, created rich longitudinal narratives due to the reflective preparation (i.e. producing the timeline) that preceded questioning. Moreover, it placed the participant in control of the process, allowing their story to be told, and anxiety to be decreased through the pre-circulation of interview questions. It also provided the opportunity to document both positive and negative intervention perceptions, across a time continuum, assisting in creating a trajectory of effects unexplored in previous effect analysis studies.

The small sample size limited the depth of the data collected in particular associated with MMT youth participants. A conscience effort was made to examine deviant cases as a means of capturing broader variations of perceptions on pain and treatment journeys, expanding the breadth of the sample, despite its size. Furthermore, the small sample did allow for a more in-depth interviews (i.e., 45-75 minutes in lengths). However, it remains that the phenomenon of participating multidisciplinary rehabilitation interventions may not yet have been fully explored[46].

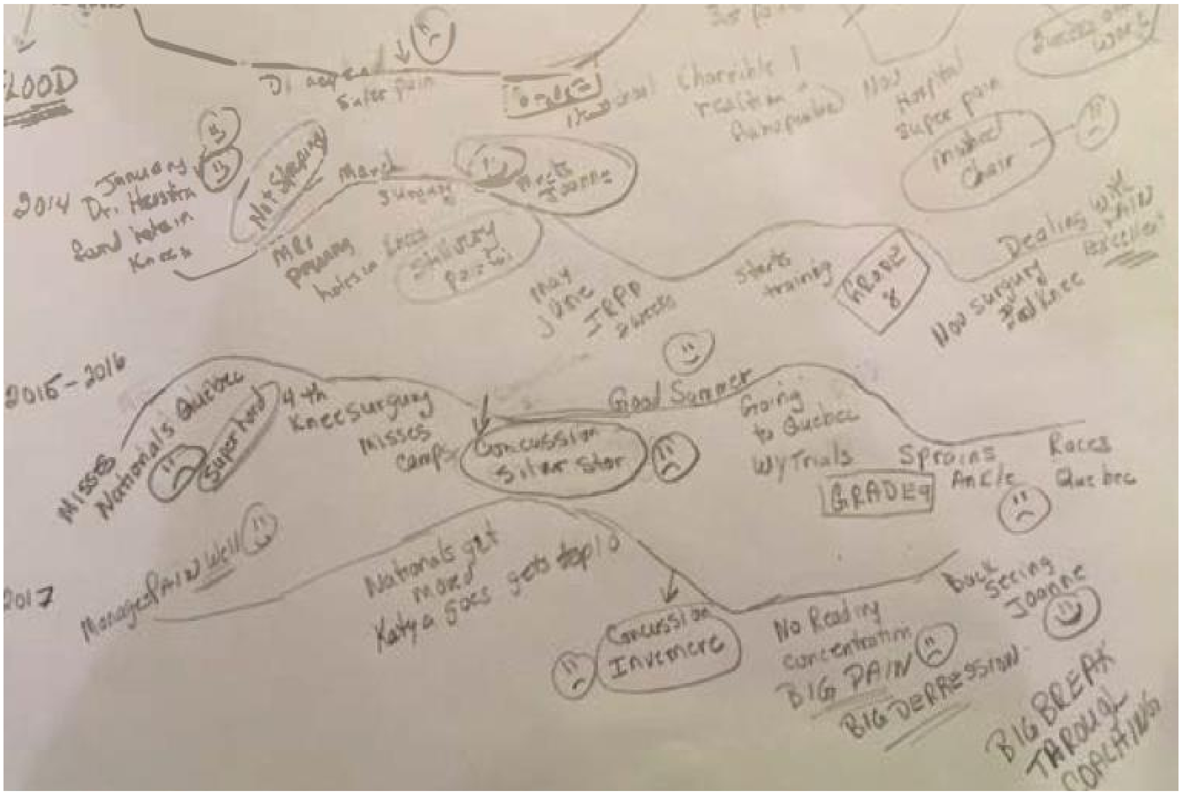
In light of our findings, several clinical implications and recommendations emerged. The program differences identified in this study, along with their unique strengths and weaknesses will be helpful in supporting clinicians in their discussions with families about treatment options and are crucial in facilitating collaborative care decisions and establishing realistic treatment expectations. Recommendations for IIPT should carefully consider youth's community peer support network, their school attendance record, and future academic and career goals to minimize potential negative impacts. Our study also provides some insight into essential program components which contributed to achieving pain self-management. Interventions to support the development of better social functioning and peer relationships despite absences and limitations caused by chronic pain are required. Clear transition pathways should be developed and studied. Supportive mechanisms such as recommendations for ongoing intervention closer to home, collaboration with teachers, coaches and other instructors, and cohort booster sessions should also be trialed. MMT programs could consider ways to further promote peer support and skill practice for both parents and youth with timely coaching support from clinicians. Attempting to minimize travel and time away from school and work for these families should be carefully considered. Finally, adherence to MMT recommendations, with focus on living well with pain, instead of symptom reduction could be further explore with youth and their families.

In conclusion, our findings help identify which youth and parents are most likely to benefit from two different chronic pain treatments, and the perceived detriments of each. This information is imperative in supporting families in making care decision and in improving clinical care pathways. Future research should focus on increasing access to these interventions, while addressing the perceived negative effects and impacts associated with them.

SUPPLEMENTARY MATERIALS

Appendix 1.

Example of participant timeline



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CHAPTER 3. DISCUSSION

This thesis is a multifaceted evaluation of an intensive interdisciplinary pain treatment program. The purpose was two-fold, resulting in the use two primary evaluation types. First, to evaluate the plausibility of the IIPT program, a logic analysis was employed. Secondly, to assess and uncover all the effects of a donor-funded IIPT day-hospital program as compared to an MMT offered at the same facility, an effect analysis was conducted. These evaluations were requested and authorized by the local and provincial decision makers to assist in determining the impact and future of the IIPT program. To our knowledge, this is the first study to evaluate and compare a day-hospital IIPT and an outpatient MMT provided at the same facility.

This thesis was focused on four main objectives and constituted four articles that together addressed each objective as follows:

1. To model the IIPT program theory and test its plausibility in achieving the anticipated outcomes (Article 1);
2. To identify and prioritize a set of outcomes meaningful to all program stakeholders upon which the evaluation will be based (Article 2);
3. To longitudinally compare the change in outcomes of youth with pain-related disability participating in the IIPT to those enrolled in the MMT based on the stakeholder-prioritized outcomes at 3 and 12 months (Article 3);
4. To explore and compare the impacts of both specialized multidisciplinary pain rehabilitation programs (i.e., IIPT and MMT) based on the experiences and perceptions of youth and parent program participants (Article 4).

3.1. Study Contributions

The multifaceted strategies use in this thesis to evaluate the IIPT intervention helped deepen both our understanding of the intervention and the experiences of youth and parents program participants. The first article, a logic analysis of the IIPT, assessed the plausibility the IIPT. It resulted first in an updated conceptualization of the IIPT program theory, unveiled clinicians' assumptions about the program mechanisms, and in raising awareness of the components most valued by parents and youth program participants. The established IIPT program theory was evaluated against relevant empirical evidence and following discussions with the expert advisory group panel, the IIPT was deemed well designed, and deemed ready for an effectiveness evaluation. Strong evidence supporting group interventions, peer learning, pain education, and integration of activities allowing knowledge and skills, with appropriate levels of coaching surfaced. An awareness of the complexity of the intervention, its scientific evidence foundation, and the patient values was created amongst stakeholders and provided a new lens through which to examine the program and clinical practice. A parallel process was also conducted for the MMT. The agreed upon logic model resulting from a logic analysis involving expert consultation and influenced by the created conceptual framework is presented in Appendix 7.

The second article, six outcome domains upon which the subsequent evaluation would be based, were identified and prioritized by a 13-member stakeholder advisory committee using a modified nominal group technique: 1) Activities of Daily Living; 2) Participation in Meaningful Activity; 3) Social Roles and Relationships; 4) Engagement in School; 5) Mood and Affect; and 6) Self-Efficacy. Although the majority of published program evaluations have considered pain intensity as an outcome variable, youth with pain-related disability most commonly consult specialized services due to resulting disability, reported as disruption of day-to-day functioning and overall poorer quality of life (QoL) (Forgeron *et al.*, 2013; Gold *et al.*, 2009; McKillop & Banez, 2016; Sorensen & Christiansen, 2017). The omission of pain intensity is perhaps the most noteworthy difference between our stakeholder-prioritized outcome domains and the existing dataset. (McGrath *et al.*, 2008). Also, the lack of patient-reported outcomes available to measure many of our prioritized domains should be underscored; only three domains (i.e. 50%) could be matched to available measures.

In article 3, we evaluated the effectiveness of the IIPT, by comparing the IIPT participant changes in scores on the three identified patient-reported outcome measures to those of youth enrolled in the MMT. The evaluation demonstrated improvements in both the IIPT and the MMT groups in association with the outcome change in score. More specifically, greater improvements were noted with some patient-reported outcomes associated with one program over the other, yet these varied depending on the program and the outcome. PROMIS Pain Interference (Varni *et al.*, 2010), selected to measure Activities of Daily Living, demonstrated significant improvement in the IIPT at 3- and 12-months. At 3-months, the IIPT also demonstrated statistically significant improvements in PROMIS Depressive Symptoms (Irwin *et al.*, 2010), the measure chosen for Mood and Affect; yet these improvements were not maintained at 12-months. The PedsQL 4.0 Core Generic Scale (Varni *et al.*, 2007) a health quality of life measure, which includes school and social subscales, improved in both groups at 3-months, while a surprising pattern of improvement was noted in the MMT at 12-months. However, between group difference did not achieve statistical significance.

Finally, in article 4, four themes related to youth's pain and treatment trajectory (i.e., either the IIPT or the MMT) were generated from in-depth timeline facilitated youth and parent n interviews. Positive and negative intervention effects, outcomes and impacts as perceived by youth and parent participants were identified and compared. Youth and parents participating in the IIPT underscored confidence and empowerment in self-managing the pain, as the greatest positive impact of the program, while negative school and social repercussions were also underscored. Those enrolled in MMT did not achieve the same level of self-efficacy in pain management and highlighted issues of adherence to prescribed treatment recommendations but did acknowledged receiving the required school and peer supports. IIPT youth felt the program provided them with the ability to manage their daily function, while those in the MMT focused on trying to cope with the pain consequences.

Beyond each one of the articles, merging and comparing the results across the studies provided an opportunity to gain a deeper understanding of 1) what treatments work, 2) for whom, and 3) provided some insight into why they work, questions commonly cited as unanswered in previously published specialized multidisciplinary pain rehabilitation literature (Harrison *et al.*, 2019; Lossi *et al.*, 2019; Odell and Logan, 2013). In defining what treatments work, participants in both programs demonstrated significant improvements. The patient-reported outcomes used in this study did not conclusively recognize one program as more effective than the other. When examined with the themes generated from the timeline interview analysis, participants program perceptions supported some of the anticipated change in scores, while providing possible explanations for some of the unanticipated patterns observed.

More specifically, youth and parents in the IIPT expressed confidence and empowerment in self-management which provides support for the significant decrease in change scores reported by this group on the pain interference measure. Those enrolled in the MMT described the prominence of pain in their lives and as a consequence exhibited higher pain interference scores. Somewhat surprisingly and not previously reported, these improvements in pain interference scores did not reflect similar improvements in health-related quality of life scores as indicated on the PedsQL measure. Initial improvements in the PedsQL measure were noted in both treatment groups, yet a pattern of greater improvement in the MMT group at 12 months was noted. In the interviews, IIPT youth described making meaningful peer connections with others in the program and developing a sense of belonging in the IIPT milieu. On the other hand, those in the MMT detailed how they maintained their regular friendships and peer interactions. As sense of belonging and friendship are closely linked to QoL and well-being in healthy adolescents (Helseth & Misvaer, 2010), these qualitative findings could explain the improvements on the PedQL reported in both groups at 3 months. However, IIPT youth also described decreased connection with IIPT program peers, loss of friendships in their community, and emerging social issues upon program discharge and return to regular life. These post-program challenge could provide an explanation for the levelling off of PedsQL after 3 months in the IIPT as compared to the ongoing improvements reported in the MMT participants. The sense of belonging derived from the IIPT milieu may also have created a greater awareness of this missing component in the lives of these youth and may have caused an even greater void at discharge, when these connections were severed, negatively impacting IIPT youth PedsQL scores at 12 months.

These noted differences between pain interference and health quality of life can be further explained by how adolescents view of these two concepts. More specifically, evidence suggests that health is more representative of one's perception of their physical health and functioning, and thus more representative of the pain interference, and QoL is more closely associated with one’s psychological well-being and acceptance by a peer group (Corsano *et al.*, 2006; Helseth & Misvaer, 2010). While this conceptualizations of the health and quality of life concepts support the lack of association between the improvement in pain interference and health-related quality of life scores, further research is required to determine whether these findings are consistent across specialized multidisciplinary pain rehabilitation programs, orunique to our study context. Furthermore, studies spanning greater than 12 months are required to fully grasp the longitudinal social and school-based impacts of these interventions.

Secondly, combining quantitative and qualitative studies also helped reveal important information about which intervention option worked for whom. To date, current practice suggests recommending IIPT as the last resort for youth and their families who have not experienced success in outpatient MMT (Simons *et al.*, 2018). However, our findings call into question this practice. More specifically, our study recognized important distinguishing characteristics of youth with pain-related disability who are best suited for IIPT, and those for whom MMT would be more effective. Recent findings from a systematic review of interdisciplinary pediatric chronic pain management intervention have also suggest equal effectiveness of MMT and IIPT programs (Liossi *et al.*, 2019). Efforts to better match the characteristic of each youth with pain-related disability and their family with the most appropriate treatment, intensity and subsequent service model could reduce “treatment failure” experienced by some youth when engaged in an intervention that does not address their needs, symptoms, or level of functioning (Harrison *et al.*, 2019; Simons *et al.*, 2018).

Our results suggest that the IIPT was be more beneficial for youth in middle school or beginning of high school, demonstrating high levels of pain interference at admission, with significant school absenteeism, who lack school engagement and have important social issues. It should, therefore, be recommended to this specific target popualtion. On the other hand, youth nearing high school graduation, who are still engaged academically and socially, and for those whom pain is not interfering with participating in these and other daily activities, the MMT represented a more appropriate recommendation. Although some awareness exists in the literature related to these patient characteristics (Simons *et al.,* 2018;

Wager *et al.*, 2014), they have yet to be applied consistently as clinical eligibility criteria and/or used to guide treatment recommendations in practice (Harrison *et al.*, 2019; Lossi *et al.*, 2019; Stahlschmidt *et al.*, 2016). A multifaceted clinical decision aid that incorporates the characteristics identified in our study should be developed, used, and assessed in relation to its impact on clinical outcomes reported treatment failure rates.

Thirdly, our findings also shed light on why youth experience the aforementioned changes in outcomes, and what program components are perceived to be change facilitators. Our study underscored the IIPT components most valued by youth and parents initially through the logic analysis and were later confirmed through our qualitative findings. More specifically, positive IIPT treatment effects were associated with chronic pain education, peer support, and the application of knowledge and skills to real-life situations, where coaching was available. These components have been acknowledged as key facilitators in the field of adult chronic pain self-management, and in studies of self-management in youth with other chronic conditions (Bal *et al.*, 2016; Blair *et al.*, 2009; Sattoe *et al.*, 2015). When considering these key components alongside the perceived negative treatment effects, important avenues for further program improvements and tailoring emerge. Research on the use of web- and application-based technology in delivering different components (e.g., pain education and peer networks) has shown promise, and their routine addition to specialized multidisciplinary pain rehabilitation warrants exploration and assessment (Kohut *et al.*, 2016; Palermo *et al.*, 2018). Furthermore, hybrid models should be considered, where the aforementioned key components are conserved as per the current models, while youth sustain social and academic routines and connections. For example, youth could attend intense interdisciplinary programming on a part-time basis (e.g., 3 days per week; or daily only in the morning) with the same cohort over a predetermined length of time, while attending their community school, regular activities, and socializing with their regular peers at the alternate times. Models similar to those suggested here are under investigation, however results have yet to be published (Dekker *et al.*, 2016; Harrison *et al.*, 2019).

The effect analysis also provided further evidence supporting the importance of considering IIPT treatments from a wholistic interdisciplinary perspective. More specifically, when describing the effects, outcome and impacts, parents and youth rarely separated or recognized specific physical, psychological, and social program activities and components as most helpful in their progress. Instead, youth and parent narratives described the program experience as a whole, including the components, structure, staff and milieu, and described the effects, outcomes and impacts resulting from the program in its entirety. Previous pediatric and adult specialized multidisciplinary pain rehabilitation programs studies have reported similar observations (Scascighini *et al.*, 2008; Risko, 2018). Interestingly, these findings contrast those in our logic analysis study, where clinician members of the expert panel, when asked to identify the program mechanisms, associated them with specific program activity provided by a designated discipline. This discipline-specific activity reporting approach mirrors that of previous studies and aligns with the current program recommended guidelines for program descriptions (Benore *et al.*, 2018; Hechler *et al.*, 2015; Stahlschmidt *et al.*, 2016).

Our study findings suggest new program reporting guidelines incorporating program theory is warranted. These guidelines could assist in uncovering previously unexplored processes and mechanisms and provide a sounder basis upon which to interpret the effects and outcomes of these interventions. More specifically, future evaluations of specialized multidisciplinary pain rehabilitation programs should decrease their focus on the clinical outcome associated to a specific program component (e.g., cognitive behavioural therapy), and instead, focus on the dynamic, interrelated, and interactivity between the program components and its impacts. Previous evidence has suggested that if a theory is not well specified, components can be misunderstood and their function obscured (Hawe, 2015). Furthermore, interventions do not take place in a vacuum and instead have meaning when contextualized (King *et al.*, 2017*)*. Therefore, the interaction between the multiple components and the context in which they occur should also be carefully considered (Hawe, 2015). As part of the context, the role of those delivering interventions should also be considered (King, 2017). Existing evidence suggested that clinical outcomes and the therapeutic change process can be influenced by the interactions among the team members, the youth, their parent and the intervention (King *et al.*, 2017; Stahlschmidt *et al.*, 2016). These transactional mechanisms between the therapist, the client and the intervention, although complex and often dynamic, are viewed as the foundational in the process of therapeutic change inherent to rehabilitation (Kazdin, 2007). Unfortunately, to date, rarely have these mechanisms been explored in the field of specialized multidisciplinary pain rehabilitation for youth with pain related disability.

3.2. Study Limitations and Strengths

All research has limitations regardless of the study design and methods employed. These limitations are examined here through the criteria for methodological rigour of both quantitative and qualitative studies (Polit & Beck, 2010). Due to the size and composition of our participant sample, some threats to the generalizability of the quantitative study findings should be mentioned. The relatively small unequal sample size of local volunteer participants at a single site resulted in a fairly homogenous sample, with limited sex and socioeconomical diversity impacted our ability to generalize our findings. The volunteer nature of our recruitment strategy may have also caused an unintended referral and selection bias. Despite its size and homogeneity, the participant sample did share many characteristics of those in other internationally published studies (e.g., Sherry *et al.*, 1999; Lee *et al.*, 2002; Logan *et al.*, 2012, Simons *et al.*, 2013, Banez *et al.*, 2014; Revivo *et al.*, 2019). Furthermore, our sample was also deemed representative of the youth and parents receiving treatment in our real-world clinical context. However, generalizing our findings to other programs and pain centers with more gender and socioeconomical diversity should done with cautioned.

Active efforts to minimize attrition were incorporated to minimize dropouts in the dataset for the quantitative analysis, yet missing data were still an issue. The statistical analysis carefully considered this issue. However, impact on the internal validity of our study may still have occurred. The amount of missing data and smaller sample hindered our ability to complete some analyses (e.g., the stratification of the sample by pain conditions) and comparison of some of the subscales (e.g., PedsQL social and school subscales), which could have provided even further understanding of the effects of the studied intervention. Furthermore, weighting of our results was used in some instances (e.g., to adjust for unequal participants numbers per stakeholder groups). Although weighting limited the underestimation of a value variable related to a more marginal group (e.g. healthcare manager), it did make the assumption that the opinion expressed was representative of that stakeholder group.

The use of self-reported measures in pediatric pain research and clinical practice is common, given that pain is defined as a subjective experience (Birnie *et al.*, 2019). However, self-reported measures are subject to response shift, reporter bias, and recall. Changes in subjective reports from pre- to post-treatment may reflect internal changes in participants' perception of their pain, rather than an observable change in functioning (Brossart *et al.*, 2002). In addition, self-reported measures are subject to recall bias when asking youth to remember their pain, physical function or emotional state over prolonged periods (e.g., weeks) (Stinson *et al.,* 2006). Although individualized objective measures (e.g., 6-minute walk-test) are collected clinically as part of standardized care, available data could not easily be collated, which rendered analysis impossible to incorporate in this study. Future research should assess correlations of self-reported outcomes with objective therapy measures.

From an intervention standpoint, neither the IIPT nor the MMT were fully standardized. Whereas the average length of stay (5 weeks) is comparable to other IIPT programs, like many other IIPT, the studied program adopted a more individualized functional and goal-based approach (Logan *et al.,* 2010, Logan *et al.*, 2012a; Simons *et al.*, 2013). More specifically, both the treatment as usual (MMT) and the intervention groups (IIPT) demonstrated some variability in dose (i.e., treatment frequency and duration), and professionals consulted. In other words, some youth may have received a greater dose of a specific treatment than others, which influenced their functioning. That being said, it should be noted that the key structural and therapeutic components of our IIPT and MMT were comparable to those previously published (Stahschmidt *et al.*, 2016; Liossi *et al.*, 2019).

In clinical settings, some variations in standardization should be expected as a function of the different pain populations recruited, along with such factors as health human resource availability, and clinician preferences (Liossi *et al.*, 2019). Moreover, many argue that standardization of complex interventions should be considered differently, where key program components may assume a different form from site to site, but the function they perform within the local context always remains the same (Hawe, 2004; 2015).

This study is context-specific, where social, political, economic factors, and time, despite being outside the program boundaries, can influence the program and its evaluation. Therefore, to facilitate transferability of our findings, thick rich descriptions of the interventions, study participants, the contextual factors, and their connections were elaborated in an effort to provide adequate information upon which research consumers can judge the application of the finding to their context (Polit and Beck, 2010). In this thesis, although the smaller participant sample involved in the interview did allow for a more detailed description of our sample, it hindered our ability to complete additional comparisons in our analyses (e.g. parent-youth dyads) and to qualitatively explore specific outcomes of interest (e.g., PedsQL school and social subscales). Furthermore, it may also have impacted our ability to fully explore the phenomenon of participating in multidisciplinary rehabilitation interventions, in particular by those involved in the MMT. Although caution is required when interpreting and comparing of these results, important avenues for future research were revealed.

From the qualitative data collection perspective, the retrospective nature of the interviews may have affected the precision by which they recount the program components and expectations. Furthermore, interviews were conducted at a single point in time and provide only a snapshot of parent and youth perceptions at a particular moment during their pain experience. To counteract the impact of these data collection limitations, a timeline activity, a longitudinal qualitative data collection technique, was used. Timelines have been reported to improve the accuracy of participants' recall of the chronology and detail of events (Glasner & van der Waart, 2009; Hope *et al.*, 2013), to be effective in encouraging participant reflection on the course of their lives (Sheridan *et al.*, 2011), and to identify specific turning points and epiphanies (Hanks & Carr, 2008; Nico &Van der Vaart, 2012). In our study, timelines were provided participants with an opportunity to reflect and prepare prior to the interview as a means to improve their recall of events and feelings. To further improve the credibility and trustworthiness of our study findings, participants were offered the opportunity to review their interview transcripts, known as member checking. Unfortunately, all parents and youth participants declined to do so. Tracy and Hinrichs (2017) suggest that member checking is not the only method that can be used to ensure that findings are accurate reflections of study participants experience, and that member reflections may be a preferred method.

Member reflections involves sharing and dialoguing with participants about the study's findings, and providing opportunities for questions, critique, feedback, affirmation, and collaboration on the data analysis (Tracy & Hinrichs, 2017). In our study, two youths and one parent participant reviewed the themes, sub-themes, their definitions, and the visual maps for each intervention and provided feedback. The feedback was used to refine the definitions of both the themes, subthemes, and maps. More active approaches (e.g., telephone conversation) or an interactive intervention (e.g., organization of a focus group) could be incorporated in future studies, to further enhance participant engagement in findning validation (Birt *et al.,* 2016).

This thesis also presents many strengths rooted in the methodological approaches included in this study. It incorporated and responded to two complementarity evaluation questions, one associated with the theoretical foundation of the IIPT program, while the other aimed to explore its effects. To answer these questions, two types of evaluation research analysis were required: logic analysis and effect analysis. Rarely have these evaluation research approaches been applied to pediatric pain intervention research. Although many authors have underscored the importance of evaluating a program's soundness prior to proceeding to a more in-depth evaluation, such as effect analysis (e.g., Bonell, 2012; Brousselle & Champagne, 2011; Moores, 2014), few studies have done so. The logic analysis process allowed us to theorize the complexity of the IIPT, empirically test the theory against available scientific and experiential knowledge, providing a sound foundation upon which to interpret the intervention effects. The logic analysis methodology used also filled some previously identified methodological gaps. Once reproduced and tested in other pediatric pain rehabilitation settings, it is hoped that the step-by-step description of this methodology will be helpful in improving program theory descriptions for future trials.

Although effectiveness evaluation has been dominated pediatric rehabilitation (Moreau & Cousins, 2011), including pediatric pain rehabilitation (Hechler *et al.*, 2015; Lossi *et al.*, 2019), effect analysis is uncommon. Effect analysis aims to identify all the observed effects, whether positive or negative, and recognise those that are attributable to the intervention and those related to other factors. The effect analysis in this thesis was also designed in a novel way, incorporating many methodological approaches, multiple, yet complementary data collection and analytical processes, and numerous strategies to engage intervention stakeholders and beneficiaries throughout the research process. First, we used a nominal group technique to establish the core set of stakeholder-prioritised outcome domains, upon the effect analysis would be based. Second, we incorporated a treatment as usual group for comparison in the study design, patient-reported outcomes in the data collection and longitudinal mixed modelling in the statistical analysis. Thirdly, a longitudinal qualitative data collection strategy (i.e., timeline) was employed. Finally, we combined the quantitative and qualitative analyzed findings in a convergent mixed method design to draw our final conclusions. The use of the multiple research approaches, methods and strategies, allowed triangulation of the different data sources, increasing our confidence and the credibility of the conclusions reached (Tracy & Hinrichs, 2017).

As a consequence of the study design and approach features, many findings previously unreported in the literature were uncovered. First, outcome domains unexplored in previous IIPT effectiveness studies (e.g., self-efficacy, and participation in meaningful activities) were prioritized by the stakeholder advisory group. This raises interesting questions surrounding the relevance of previously evaluated outcomes study findings to stakeholders, including program users. Secondly, the inclusion of a treatment as usual group in the effect analysis highlighted beneficial effects unique to both programs over the first 12-months. Many previous studies supported IIPT as the treatment of choice for youth with pain-related disability (Hechler *et al.*, 2015). However, our findings recommend this treatment for a specific subset of this population. Although one comparison study does exist, only short-term outcomes were compared. The monitoring and comparison of the stakeholder-prioritized outcomes over a 12-month period provided new insight into the sustainable effects of these treatments over the longer term and to identify other impacts for which time was required for their emergence to occur. Thirdly, a deliberate choice was made to involve children and their parents beyond their responses to self-report questionnaires, striving to gain a true sense of their experience through qualitative methods.

The use of timeline facilitated interviews as a qualitative data collection method was also quite novel in this field. This method provided a glimpse into the IIPT treatment perceptions of both youth with pain related disability and their parents, their expectations, and the associated burden as well as the benefits. We are aware of the existence of one study that explored the IIPT experiences of youth (Risko, 2018), and another that examined that of parents (Gaughan *et al.*, 2014). Even fewer studies are available related to participants' perspectives of MMT (Gorodzinsky *et al.*, 2012). The available IIPT studies limited the exploration of these perspectives and experiences to the duration of the IIPT program, conducting interviews at the time of discharge (Gaughan *et al.*, 2014; Risko, 2018). The MMT study, used an even shorter period, interviewing their participants at the end of their initial clinic visit (Gorodzinsky *et al.*, 2012). Although useful in gaining participants initial experiences, this approach limits the insight into the long-term impacts of these treatments. The use of timelining and the timing of the interview at least 12-months post discharge provided a longitudinal perspective of youth and parents' treatment and evolving pain journey since program completion. The treatment trajectory narratives, in particular post program discharge, have been lacking in the literature until now.

Finally, the mixed methods approach allowed us not only to highlight the positive impacts, but also uncover those that were negatively perceived and attributed to the interventions. Until now, negative treatment impacts of IIPT had rarely been underscored, and reflected a transparency reporting gap previously highlighted (Caes *et al.*, 2018). The interest in these mixed method designs is growing overall in health and social research, in particular as different outcomes (i.e., measures vs. stories) and varying methodological approaches have demonstrated appeal to various stakeholders (Farquhar *et al.*, 2011). In this thesis, as typically recognized in the mixed methods research (O'Cathain *et al.*, 2007), combining quantitative and qualitative data yielded greater knowledge and comprehensiveness of specialized multidisciplinary pain rehabilitation interventions and deepened our understanding of the effects, outcomes, and impact of these programs on youth and their parents. It helped in recognizing important youth characteristics upon which treatment recommendations should be based, eligibility criteria should be defined, and screening processes should be developed. Although the number of longitudinal IIPT intervention studies are increasing, studies examining the outcomes of MMT and those comparing interventions are still desperately needed. Without combining and comparing the quantitative outcomes and the qualitative participant reported treatment perceptions, the benefits and detrimental effects of one treatment option over another will remain under-recognized, and clinical recommendations will rest upon incomplete evidence.

3.3. Stakeholder Engagement Processes and Strategies

Our commitment to engaging youth with pain-related disability and their parents went beyond including their treatment perspectives and experiences in the effect analysis. This study is among the first in the field of pediatric chronic pain intervention to engage a wide variety of vested stakeholders. Along with youth with pain-related disability and their parents, other stakeholders deemed important in their daily lives (i.e., teachers) were also included. Their engagement went beyond the data collection stage and instead stakeholders were considered as key contributors throughout the research process. To our knowledge, only one other published study in the field of pediatric chronic pain has included stakeholder engagement principles (Birnie *et al.*, 2019). This study engaged youth with chronic pain, their parents and clinicians in prioritizing the top10 patient-oriented research priorities in pediatric chronic pain in Canada. These researchers noted that involvement of youth and family members led to different identified priorities than previous efforts, where public or youth involvement was limited. It is interesting to note that the findings of this thesis addressed some of the priorities identified by Birnie and colleagues (2019) and acknowledge similar knowledge gaps. The parallels between our study findings and those of Birnie and colleagues (2019) help to underscore the value of stakeholder engagement in this field of research and provide further evidence of the shared concerns and challenges faced by theses youth and their families.

Unfortunately, the uptake of patient engagement in research in chronic pain, like many others, has been slow. This may be a reflection of perceived barriers to engaging stakeholders in research and the lack of guidance on effective engagement approaches (Birnie *et al.*, 2019; Camden *et al.*, 2105; Mafano *et al.*, 2018). With the interest in bridging this knowledge-to-practice gap, a detailed description of the terms and concepts adopted, and activities and processes utilized in this thesis, along with the supporting evidence, are summarized below.

Stakeholders were engaged in this study in the first place due to the purpose and aim of the study, which is perceived by most researchers as a feat (Camden *et al.*, 2015). More specifically, the evaluative study reported in this thesis had two core purposes: 1) to assist decision makers at the organizational level in assessing the accountability of the IIPT and determining its future; and 2) to foster mutual learning for the purpose of program improvement. Evaluations conducted for accountability purpose involves rendering judgment about the value and worth of a program, the acceptability and effectiveness of a program, or to inform decisions about future programming (Clarke, 2006). This judgment is contingent on the relevance of the evaluation outcomes to stakeholders (Moreau & Cousins, 2011). Although managing the perspectives of a diverse group of stakeholders can be challenging, their inclusion can increase the project feasibility, outcomes relevance, ownership of the evaluation, and the usability of the research findings (Camden *et al.*, 2015; Cousins & Whitemore, 1998; Patton, 2008). The evaluations presented in this thesis was also conceptualized as a learning process and engagement of clinicians, physicians, and management was deemed crucial in the acceptance of evaluation findings and the adoption of its recommendations.

Using a direct recruitment strategy, a variety of stakeholders were targeted. Partner organizations were contacted, and asked to nominate members for the advisory committee General agreement exists in the evaluation literature that the patients and/or caregivers receiving or who have been involved in the program under study provide more meaningful input than the general public or those who have used other services at the same facility (Gagliardi *et al.*, 2008; Moreau, 2012). However, direct recruitment can also create bias, those selecting participants may only approach individuals who share a similar perspective about what should be valued in the evaluation, thus limiting the variability in viewpoints. In an attempt to counteract this potential bias, several criteria were carefully considered in stakeholder selection. These included: 1) characteristics representative of the stakeholder group; 2) willingness to speak on behalf of the group; 3) communication abilities; 4) ensuring diversity of perspectives in the group (Johns *et al.,* 2004; McGrath *et al.*, 2009; Sax, 2007). In our study, a specific consideration was the inclusion of male representation, which traditionally has been difficult to achieve due to the dominance of females in health and education workforce and in this patient population.

To evaluate communication skills and willingness to speak on behalf of a group, an interview process was employed. Participant was interviewed about their reasons for wanting to be involved in the study. To maximize comfort and authenticity, youth were offered the choice of being interviewed with or without their parents. Conversations were monitored for inclusive language, consciousness of difference between own opinion and that of others, and awareness of those not present (e.g., other health disciplines, other youth and parents in treatment cohorts). To date, details related to interview or selection process have been limited in the literature (Camden *et al.*, 2015). Inquiring about when, how, and why stakeholders want to engage is a useful strategy in determining, their motivation and willingness to be involved in the study and can provide valuable insight into needs that may require accommodation during the process (Nguyen *et al.*, 2019). To foster inclusion, maintain stakeholder engagement, and provide access to regular and sustained interactions, a variety of methods were included: multiple meetings options, a progressive work strategy, a variety of communication technologies (e.g., FaceTime, Skype, teleconferences), and a regular communication strategy (e.g., predictable meeting schedule, e-newsletter). Over the 3-year study period, all stakeholders remained engaged, with only one advisory member dropping out, due to a change in employment, and was quickly replaced.

Power sharing, acknowledged as essential in negotiating the study agenda, resolving conflict, and supporting meaningful engagement, teamwork and collaboration (Buettgen *et al.*, 2012; Ottman *et al.*, 2009; Sax, 2007), was also carefully addressed. The importance of clarifying roles and agreeing on realistic expectations among stakeholders and the research team prior to the beginning of the research process has been emphasized in the literature (Mafano *et al.*, 2018). However, it has also been suggested that predetermining roles and expectations can also limit the possibility for stakeholders to determine how they would like to engage (Buettgen *et al.*, 2012; Nguyen *et al.*, 2019; Walmsley *et al.*, 2009). This study chose to complete the NGT process prior to negotiating roles and expectations. The active dialogue inherent to the NGT technique has been found to nurture a comfort level among the stakeholder that promotes engagement and empowerment, fosters respect for all ways of knowing, helps partnership development, and promotes mutual learning across stakeholder groups (Potter *et al.*, 2004; Domecq *et al.*, 2014). A team charter was then co-created with the advisory committee and once completed, was signed by all members (please see Appendix 6 for anonymized, unsigned version). This charter included the project's mandate and objectives, the composition of the committee, including available skills, expertise, and learning needs, a description of the roles and participation expectations, meeting rules and operating procedures, communication strategy, shared decision-making and a conflict resolution process. Not only was the charter important in preventing and managing challenges and issues, it also provided the structure and support required to fostering a trusting and positive meeting environment.

Formal (e.g., research design; literature review) and informal (e.g., meeting discussions) training have been recognized as helpful in developing research skills or to increase knowledge on certain topics that foster stakeholder engagement (Camden *et al.*, 2015; Mafano *et al.*, 2018). In this study, several short presentations were provided to both the research team and stakeholder group. These included an introduction to participatory research, to logic model development, and to scoping review methodology. Furthermore, a common language (i.e., the "F"words of Childhood Disability) was adopted to minimize jargon and ensure understanding, comfort, and confidence for meaningful dialogue (Amtmann *et al.*, 2011; Buettgen *et al.*, 2012). An adapted version of a parent co-designed and validated video (Cross *et al.,* 2015) was used to explicate the "F" words and their application to pediatric rehabilitation. These and other online references (e.g., mixed methods, scoping review) aligned with the groups identified learning needs were shared with the stakeholders for their reference throughout the research process. As part of the team charter, regular temperature checks were conducted with each stakeholder to identify addition support and training needs.

Stakeholder engagement is critical in building and strengthening the relevance, and quality of a study (Alberta SPOR Support Unit, 2018). In our study, engagement of our stakeholder advisory group proved crucial to the data collection phase and resulted in the identification and use of the patient-reported outcome measures into the data collection phase, the elimination of a time point minimizing participant burden, and the implementation of an individual in-depth timeline facilitated interview process. Although data analysis requires specialized skills, which may not be possessed by stakeholders (Camden *et al.*, 2015), stakeholders in this study did engage in the interpretation of the findings and provided vital insight in the qualitative analysis, and when analyzed qualitative and quantitative results were merged. As previously reported, this helped foster meaningful contextualization of the results for the research team, a recognized step toward the generation of useful and relevant knowledge (Graham *et al.*, 2006).

The dissemination phase of research is a critical one, for both researchers and stakeholders, where few concrete strategies have been reported and little evidence of its effectiveness exists (Camden *et al.*, 2015; Gagliardi *et al.*, 2016). In our study, knowledge dissemination to another sector (i.e., public education) occurred through stakeholder engagement. As argued by some authors, stakeholders in our study acted as early ambassadors of the research findings and facilitated their uptake into the broader community (Shippee *et al.*, 2013; Esmail *et al.*, 2015). Stakeholder engagement represents a revolution of the traditional research paradigm to one that empowers stakeholders as a research partner. The interest in collaborative forms of inquiry in health science research has increased dramatically and has recognized stakeholders as essential in the evaluation of health services. It is hoped that the details of the extensive stakeholder engagement structures, strategies, methods and approaches documented in this thesis, along with the evidence supporting them, provides inspiration and the guidance for others in the field of pediatric pain interested to engage stakeholders in their research.

CONCLUSION

Pediatric chronic pain is a significant problem among children and adolescents worldwide, which has devastating physical, psychological, academic, social impacts on both youth and their families. It is recognized as a complex health condition, for which complex multidisciplinary interventions are needed. Programs addressing the interplay of biological, psychological, social, and environmental factors contributing to and maintaining pain and related disability are needed. However, previous evaluations of these programs have been limited in type and design. As a result, the evidence has been missing for healthcare managers to reproduce these programs in other contexts, for clinicians to be able to effectively interpret clinical outcomes, and for families to make informed treatment choices.

Recognized as complex interventions, specialized multidisciplinary pain rehabilitation programs may be best assessed using multifaceted evaluation, varied data collection methods and analytical strategies. Using these methods alongside a participatory approach, allowed us to fill some important knowledge gaps. From a research point of view, this study identified outcomes relevant to its stakeholders, and recognized the valued components of these programs and their scientific foundations. These findings contributed to establishing the effectiveness of both available treatment options, while helping to explain divergences in our findings and uncovering previously unreported program effects. From a clinical standpoint, the valuable comparison between treatments assisted in recognizing key characteristic of youth most likely to benefit from one program or the other, generated the knowledge needed for clinicians to make targeted treatment recommendations and for youth and parents to make informed care decisions, and underscore future program improvements. Finally, it allowed for the testing and refinement of a variety methodological approaches, data collection strategies and analytical processes emerging in other research fields, yet rarely used with this population.

The study also proposed a number of recommendations to improve the care for this population and their families. The study findings highlighted key participant characteristics which, if incorporated into the clinical decision-making process, could better match individual youth with the appropriate treatment modality and level of intensity and potentially lead to reduced treatment failure rates. However, careful monitoring of participant outcomes would need to occur if such changes were implemented. Furthermore, longitudinal studies following these youth into adulthood are needed. Recommendations for program refinements were also highlighted. Modifications that use new modalities or models to increase participants access to evidence-based and valued program components, while minimizing those attributed to negative effects should be a focus of future evaluations.

Several promising directions for research were also underscored. More specifically our findings highlighted measurement gaps in stakeholder prioritized outcomes and in minimal clinical important difference cut-off scores for this population. There is also a need to identify and explore additional mechanisms of change within specialized pain rehabilitation programs from an integrated multidisciplinary perspective. Relationship between intervention, its context, and the interpersonal transactions between the team members themselves, and with those participating in treatment should be explored. The investigation of these mechanisms may help to further tailor these interventions, improving their efficiency and cost effectiveness.

In summary, to respond to our overall research question, IIPT programs are worth it, as long as they are carefully designed and implemented, and offered to the right patients. Given the complexity of factors involved and functional consequences of pediatric pain-related disability, treatment design and matching of youth with pain-related disability and families to most effective specialized multidisciplinary treatment approaches is imperative. The findings from this study provide comprehensive and transparent evidence of the various impacts of the IIPT and generate important new knowledge upon which youth, parents, clinicians, healthcare managers and local and provincial policy makers can base treatment decisions. Furthermore, recommendations to improve interventions also surfaced. Not only is the knowledge generated by this thesis pivotal in alleviating the suffering and improving the lives of youth with chronic pain and their families in Alberta, in Canada, and beyond, it sets a new standard for evaluation research of complex rehabilitation interventions.

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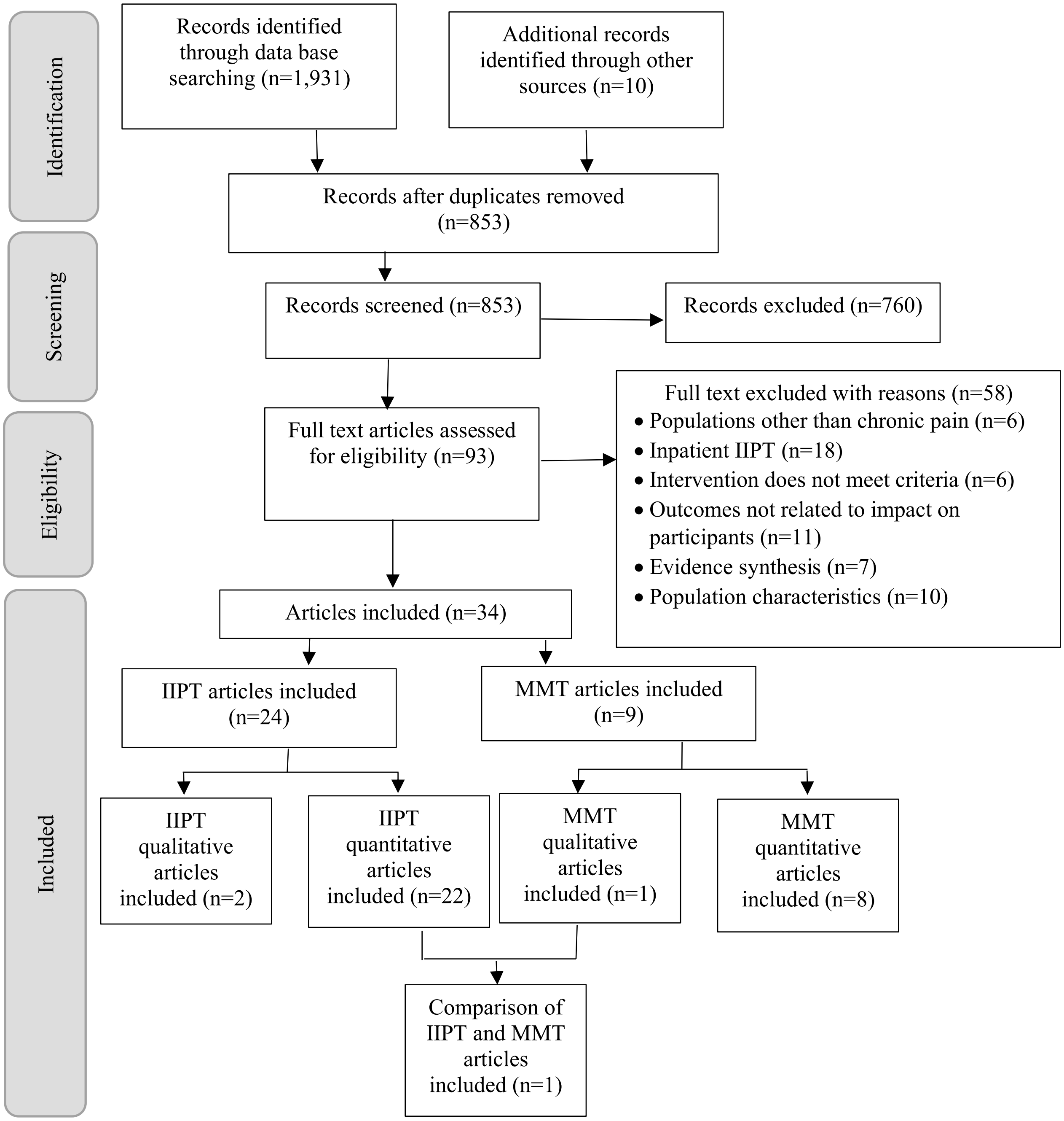
APPENDICES

Appendix 1.

Flow Diagram of Selected Studies



Appendix 2.

Characteristics of the IIPT and MMT Studies, their Measures, and PedIMMPACT Comparison

Study	Country	Design	Participant demographics	Intervention	Data collection time points (*n* if attrition)	Outcome domains and measures	PedIMMPACT recommendations comments
Sherry *et al*., 1999	USA	Pre-/post-intervention single group	103 patients diagnosed with Complex Regional Pain Syndrome (CRPS) who participated in an exercise therapy program. Mean age: 13 years (range 7-21 years) Sex: 87% female. Type of Pain: lower extremity (83%); upper extremity (11%); upper and lower extremity (9%); bilateral involvement (17%)	Inpatient (*n*=60) and outpatient (*n*=43) treatment provided	Pre-treatment (*n*=74), 6-8 months post-treatment (*n*= 84), long-term data (*n*=49)	Pain intensity: Visual Analogue Scale (VAS) Physical functioning: Participation in age- and limb-appropriate activities (e.g., physical education, dressing tasks, walking endurance and ability to open the car door. Condition reoccurrence: whether symptoms reoccurred (yes/no)	Pain intensity and physical functioning domains were included. No other domains (i.e., emotional and role functioning, global satisfaction with treatment, adverse events, sleep and economic factors) were assessed. No measurement tools aligned. Note: Predominantly objective assessment tools were used for physical performance.
Lee *et al.*, 2002	USA	Randomized-control trial (RCT)	28 youths Mean age =18.8 years (SD =2.2) Sex: 93% female. Type of pain: Complex Regional	Multidisciplinary treatment	Pre-treatment (*n*=28); short-term follow-up (*n*=25); long-term follow-up (6-12 months) (*n*=24)	Pain Intensity: VAS Allyodynia:6-point scale (1=extreme	Pain intensity, physical and emotion functioning domains included. Pain intensity measurement tool aligned with recommendation, yet

			Pain Syndrome (64%); CRPS2 (36%) 15 randomized to 1 physiotherapy (PT) session/week, 1 cognitive-behaviour session/week + education program for 6 weeks 13 randomized to 3 PT sessions/week, 1 cognitive-behaviour session/week + education program for 6 weeks			allodynia; 7=no allodynia) Health Related Quality of Life (HRQoL): The Child Health Questionnaire (CHQ-CF87); Physical functioning: Untimed 30 metre gait analysis and support requirement; unassisted stair climbing and descending Emotional functioning: Child Depression Inventory (CDI); Revised Children's Manifest Anxiety Scale (RCMAS). Condition reoccurrence: whether symptoms reoccurred (yes/no)	assessment over a longer period of time (e.g., over the past 7-days), number of pain-free days or using pain diaries over multiple days is advised by the recommendation. Emotional functioning (depression and depression) measurement tool aligns with recommendations, while physical functioning did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Other domains explored: Allodynia, condition reoccurrence Note: Objective assessment tools were used for physical performance.
Eccleston *et al.,* 2003	UK	Pre-/post-intervention single group	78 patients referred from tertiary clinics assessed.	Residential interdisciplinary programme	Pre-treatment (*n* = 78), immediate post-treatment (n = 56),	Pain intensity: VAS	Pain intensity, physical, emotional and role functioning domains considered.

	57 entered treatment and 56 completed. Mean age = 14.28 years (SD (range) = 1.60 (11–17)). Sex: 71% female. Type of Pain: Fibromyalgia 9 (15.8%), CRPS 15 (26.4%), Diffuse idiopathic pain 14 (24.6%), Localized idiopathic pain 7 (12.3%), Disease related 8 (14.0%), Headache 1 (1.7%), RAP 2 (3.5%), Renal pain 1 (1.7%)		and 3-month follow-up (*n* = 43)	Emotional functioning: The Spence Children's Anxiety Scale (SCAS); Pain Coping Questionnaire (PCQ); CDI (depression); Modified Somatic Perception Questionnaire (MSPQ). Physical functioning The Functional Disability Inventory (FDI); timed 10 metre walk and sit-to-stand task (observational measures). Role functioning: School attendance: Number of half-day sessions attended in the previous week, seven categories of school attendance were also recorded (graduated, full time, part-time (1–4 sessions), part-time (5–9	Pain intensity measurement tool aligns with the recommended measure, yet assessment over a longer period of time (as above) is recommended. Physical and emotional (depression) functioning measures align with recommendations Emotional (anxiety) and school functioning measure did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Other domains explored: Parent perception of child's pain, disability, and parent's anxiety, depression and stress.

						sessions), home tutoring, none, left having completed schooling). Parent measures Perceptions of their child's pain: Adult version of two VAS. Perceptions of their child's disability: Adult version of FDI. Anxiety and Depression: Hospital Anxiety and Depression Scale (HADS) Stress: Parenting Stress Index (Short Form)	
Wicksell *et al.,* 2009	Sweden	Randomized control trial	34 referrals to pain treatment service eligible. 32 randomized Mean age = 14.8 years (SD = 2.4) Sex: 78% female Type of Pain: headache (25%), back/neck pain (22%), widespread musculoskeletal pain	Multidisciplinary treatment approach	2 weeks pretreatment, immediately post-treatment (mean 5.3 (*SD* = 1.6) months after pretreatment assessments, *n* = 32), follow up 1 (F-u 1) (mean 3.5 (*SD* = 0.8) months after post-treatment assessments, *n* = 25), follow-up 2 (F-u 2)	Pain intensity: VAS (0 = no pain; 10=worse pain imaginable) Pain: The Multidimensional Pain Inventory, Interference scale (MPI); the Brief Pain Inventory, pain interference items (BPI); The Pain and	Pain intensity, physical and emotional functioning domains considered. Pain intensity measurement tool aligns with the recommended measure, yet assessment over a longer period of time as recommended, was not included. Physical functioning measures align with recommendations.

			(22%), CRPS (22%), visceral pain (6%), lower extremity pain (6%), postherpetic cheek pain (3%) 16 randomized to MDT (multidisciplinary treatment approach including amitriptyline) group 16 randomized to ACT (Acceptance and Commitment Therapy) group		(mean 6.8 (*SD* = 1.1) months after post-treatment assessments, *n* = 24) MDT: 15 completed assessments, 14 completed first follow-up, 13 completed second follow-up ACT: 14 completed assessments, 11 completed first follow-up, 11 completed second follow-up	Impairment Relationship Scale (PAIRS) Physical functioning: FDI Emotional functioning: Center for Epidemiological Studies Depression Scale for Children (CES-DC); Fear of pain: Tampa Scale of Kinesiophobia (TSK-11); The Pain Coping Questionnaire (PCQ) HRQoL: The Short Form-36 Health Survey (SF-36)	However, emotional functioning measures (depression and anxiety) did not. Role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Other domains explored: Pain interference, beliefs and attitudes, HRQoL, fear of injury, beliefs and attitudes about pain, pain catastrophizing
Simons *et al.*, 2010a	USA	Pre-/post intervention single group	120 participants 57 children and 70 parents Youth: Mean age = 14 years (SD=1.9) Sex: 79% female Type of pain: 29% musculoskeletal, 22%	Outpatient multidisciplinary intervention	Pre and 3 months post	Pain: current, average, worst and least pain on 11-point NRS (0=no and 10=worst pain imaginable). Emotional functioning: Somatic symptoms: Child	Pain, physical, emotional, role functioning and global judgement of satisfaction were measured. Physical and role functioning measures aligned with the recommendations. Pain and emotional functioning did not.

			neuropathic, 22% headache, 7% abdominal, 4% other			Somatization Inventory (CSI) Physical functioning: FDI Role functioning: number of days of school missed in the past 3 months Parent measures: Treatment expectations and familiarity questionnaires (developed for the study); Healthcare utilization: number of physician, emergency visits and hospital stays Adherence and Barriers: Telephone interview form Clinic Satisfaction Questionnaire	Role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Emotional functioning (anxiety and depression), global judgment of satisfaction, sleep, and economic factors not assessed. Other domains explored: treatment expectations, healthcare utilisation, barrier and adherence to treatment.
Gauntlett-Gilbert *et al.,* 2012	UK	Pre-/post-intervention single group	98 referrals from secondary or tertiary centres, Mean age = 15.6 years (SD = 1.7, range = 10.8–19.0) Sex: 75% female Types of Pain: Diffuse/localized	ACT-based, group residential pain management program	Pre-treatment (*n* = 98), post-treatment (*n* = 94), and 3-month follow-up (*n* = 73).	Pain intensity: VAS (0=no pain; 10=worse pain imaginable). Physical functioning: physical functioning	Pain intensity, physical and emotional and role functioning domains were considered. Pain intensity measurement tool aligns with the recommended measure, yet assessment over a longer period of time as recommended, was not included.

	idiopathic pain = 27%, Complex regional pain syndrome = 20%, Back pain = 19%, Abdominal pain = 13%, Pain associated with hypermobility = 11%, Other (e.g., headache) = 10%			subscales of Bath Adolescent Pain Questionnaire (BAPQ); Timed walk test, sit-to-stand task (observational measure). Emotional functioning: Depression, general anxiety and pain-specific anxiety subscales of the BAPQ catastrophizing scale; Chronic Pain Acceptance Questionnaire-Adolescent version (CPAQ-A) Role functioning: Social functioning subscales of Bath Adolescent Pain Questionnaire (BAPQ); school attendance (total weeks missed, current attendance status) self-reported by adolescents. Health Care Use: Previous 3 months	Physical, emotional (depression and anxiety), and role functioning measures do not align with recommendations. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Other domains explored: Acceptance of pain, healthcare utilization Note: Objective assessment tools were used for physical performance.

						healthcare use (number of nights admitted, number of outpatient appointments, number of primary care physician attendances, number of emergency room visits as reported by parents	
Gaughan *et al.*, 2012	USA	Qualitative descriptive design	9 parents of youth with chronic pain Youth: Age range: 11-17 Sex: 89% female Type of pain: 88% CRPS, 22% neuropathic	Day-hospital interdisciplinary paediatric pain rehabilitation	At discharge	Parental empowerment: semi-structured interviews	None of the outcome domains were used, and the tool did not align with the recommendations. Other domains: Parental empowerment
Logan *et al.,* 2012a	USA	Pre-/post-intervention single group	56 participants with CRPS Mean age = 14.1 years (SD = 2.5; range = 8–18); Sex: 89.3% female	Day-hospital interdisciplinary paediatric pain rehabilitation	Follow-up median 10 months, range 2-24 months, *n* = 30; 11% attrition	Pain intensity: 11-point numeric rating scale (NRS) (0=no pain; 10=worse pain imaginable) Physical functioning: FDI; Lower extremity functional scale (LEFS); Canadian Occupational Performance Measure (COPM); Physical functioning: The	Pain intensity, physical emotional, and role functioning domains were considered. Physical, emotional (depression), role (school) functioning measures aligned with recommendations. Pain intensity and emotional functioning (anxiety) did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed.

						Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition (BOT-2) (objective measures) Emotional functioning: Anxiety: The Multidimensional Anxiety Scale for Children (MAS-C) Depression: CDI Role functioning: Paediatric Quality of Life Inventory (PedsQL) school functioning subscale.	Note: Objective assessment tools were used for physical performance.
Simons *et al.,* 2012	USA	Pre-/post-intervention with single group	154 patients enrolled in the program, 145 eligible completers Mean age =14.5 years, SD = 2.2, range = 8.4–18.3) Sex: 86% female. Types of pain: Neuropathic pain (93%), musculoskeletal (7%)	Intensive interdisciplinary paediatric pain rehabilitation day program	Pre-treatment, post-treatment, approximate 1-2 months follow-up. 125 provided some or all data at all three-time points. 16 did not report follow-up (13.7% attrition.)	Pain intensity: 11-point NRS (0=no pain; 10=worst pain imaginable) Emotional functioning: The Fear of Pain Questionnaire (FOPQ-C); CDI Physical functioning: FDI	Pain intensity, physical and emotional functioning (depression) domains were considered. Physical and emotional functioning (depression) measures aligned with recommendations. Pain intensity did not. Emotional (general anxiety) functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed.

							Other domains: Pain specific anxiety.
Logan *et al.*, 2012d	USA	Pre-/post intervention single group	157 youth consented Mean age =14.1 (SD=	Intensive interdisciplinary paediatric pain rehabilitation day program	Admission, discharge, 144 youth (92%) at admission 148 parents (94%) at admission 135 youth at discharge 139 parents at discharge	Readiness to change: Pain Stages of Change Questionnaire adolescent (PSOCQ-A) and parent versions (PSOCQ-P) Pain intensity: 11-point NRS (0=no pain; 10=worst pain imaginable); Pain response inventory (PRI) Physical functioning: FDI Emotional functioning: CDI; FOPQ-C and parent version (FOPQ-P)	Pain intensity, physical and emotional (depression) functioning were considered. Recommended measures aligned only with physical functioning Emotional (general anxiety and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain-specific anxiety (parent and youth) and readiness to change (parent and youth)
Simons *et al.,* 2013	USA	Retrospective pre-/post-intervention with control group	296 patients enrolled in the program, 100 were included following matching process. 50 participants included in the IIPT group and 50	Intensive interdisciplinary paediatric pain rehabilitation day program and outpatient multimodal treatment	Pre-treatment, post-treatment, 3- months.	Pain intensity: 11-point NRS (0=no pain; 10=worst pain imaginable) Physical functioning: FDI	Pain intensity and physical functioning domains were considered. Physical functioning measure aligned with recommendations. Pain intensity did not.

			participants in outpatient treatment. Mean age =13.8 years (SD=2.36) Sex: 85% female. Types of pain: Neuropathic pain (63%), musculoskeletal (33%), abdominal (4%)			Readiness for change: PSOCQ-A, PSOCQ-P	Emotional (depression and anxiety) functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed. Other domains: Readiness for change
Soee *et al.,* 2013	Denmark	Prospective cohort (Pre-/post-intervention single group)	169 children with headache (39% chronic headache). Mean age = 11.7 years (SD = 3.04) Sex: 54% female	Outpatient multidisciplinary headache treatment	Baseline, 6 and 12-month follow-up. 156 (92%) at 6-month follow-up 130 (77%) at 12-month follow-up	Pain: Headache frequency parent and child-report HRQoL: PedsQL child-reported and parent proxy	Pain intensity outcome domain considered, and measure aligned with recommendations Emotional functioning (general anxiety), global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not assessed Other domains: Pain frequency; and HRQoL
Weiss *et al.*, 2013	USA	Pre-/post-intervention single group	112 referrals from speciality clinics. Mean age = 15.47 years (SD = 1.83, range 11–18) Sex:76% female Type of Pain: abdominal (30%), headache (26%), generalized (25%), back/neck (8%), extremity/extremities	Outpatient rehabilitative program (group format)	Pre-and post-treatment	Pain intensity: 11-point NRS (0=no pain; 10=worst pain imaginable) Emotional functioning: CES-DC; The Pain Catastrophizing Scale for Children (PCS-C)	Pain intensity, physical and emotional functioning (depression) domains were considered. Physical functioning measure aligned with recommendations. Pain intensity and depressive symptoms did not. Emotional (general anxiety) and role functioning, global judgment of satisfaction with treatment,

			(7%), chest (2%), and pelvic area (2%).			Physical functioning: FDI	symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain acceptance; pain catastrophizing
Banez *et al.*, 2014	USA	Retrospective chart review	173 youth Mean age = 15.1. years (SD=2.5) Sex: 73% female Type of pain: CRPS (35%); headache (22%); abdominal pain (13%); fibromyalgia (12%); other pain (18%)	Pediatric pain rehabilitation program (inpatient and day hospital)	Pre-treatment, discharge, 1, 12, 24-48 months 166 (96%) at admission 151 (87%) at discharge 49 (28%) at 1-month 59 (34%) at 12-months 79 (46%) at 24-48 months	Pain intensity: Measure not specified Role functioning: number of school days missed in the past week Parent productivity: Number of days of work missed by parent due to child's pain in the past week Health care utilization: number of days youth hospitalized for pain in the past month	Pain intensity and role functioning domains were considered. Role functioning aligned with recommendations Pain intensity measure not specified. Physical and emotional (depression and anxiety) functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other measures: health care utilization, parent productivity
Benore *et al.*, 2015	USA	Retrospective cohort study (Pre-/post-intervention single group)	461 patients in the initial sample Mean age = 15.4 years (SD = 2.7) Sex: 74.6% female Final sample of 119 patients included in the study	Paediatric pain rehabilitation program (inpatient and day hospital)	Pre-treatment, 1-month post-treatment	Emotional functioning: BAPQ General anxiety, pain-specific anxiety, and depression subscale; PCS-C	Physical, emotional (depression and anxiety) and role functioning domains were considered. None of the recommended measures were used. Pain intensity, role functioning, global judgment of satisfaction

			Mean age = 15.1 years (SD = 2.6) Sex: 76.5% female. Types of pain: CRPS (34.5%), headache (16.8%), abdominal pain (21%), back pain (6.7%), other (21%)			Role functioning: Social functioning, subscales of BAPQ Physical functioning: physical functioning and development subscales of BAPQ Family functioning subscales of BAPQ HRQoL: PedsQL	with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain catastrophizing; family functioning; development, HRQoL
Logan *et al.*, 2015	USA	Pre/Post intervention single group	274 youth enrolled Mean age=14.6 years (SD=2.3) Sex: 84%female Types of pain: Neuropathic or musculoskeletal pain (100%)	Pediatric pain program (day hospital)	Pre-treatment, post-treatment, first follow-up (1-3 months)	Pain intensity: 11-point NRS (0=no pain; 10=worst pain imaginable) Physical functioning: FDI Emotional functioning: CDI Role functioning: PedsQL school subscale Sleep Habits: Children's Sleep Habit Questionnaire (Parent report);	Pain intensity, sleep, physical, emotional (depression) and role functioning were considered. Measures aligned with recommendations except for pain intensity and school (a number of days missed). Emotional functioning (anxiety), global judgment of satisfaction with treatment, symptoms and adverse events, and economic factors not considered or assessed. Note: No recommendations for sleep measures existed in PedIMMPACT

						child self-report (pain interference with sleep, length of time needed to fall asleep, average number of wakings per night. Medication use: Number of medications being used.	
Sherry *et al.*, 2015	USA	Pre/post intervention single group	81 youth eligible 64 youth participated Median age = 16 years (IQR=15-17) Sex: 90% female Type of pain: Fibromyalgia (100%)	Intensive pain program (inpatient and day hospital)	Pre-program, post-program, 1-year	Pain intensity: VAS Physical functioning: Bruce treadmill protocol (observational measure); BOT-2 (observational measure); FDI Emotional functioning: PSOCQ, adolescent version HRQL: PedsQL	Pain and physical function domains considered, and recommended measures used. Emotional (anxiety and depression) and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Readiness for change, HRQoL Note: Objective assessment tools were used for physical performance
Kanstrup *et al.*, 2016	Sweden	RCT	48 adolescents with chronic debilitating pain and their parents 24 in group treatment (2 hours each)	18 adolescents outpatient sessions, 3 parent-sessions, and 1 joint session conducted by psychologist, PT and physician.	Pre-, mid- and post-intervention	Pain: Pain interference index (PII); Pain reactivity scale (PRS) Physical functioning: FDI	Pain, physical and emotional functioning (depression) were considered. Recommended measures were only used for physical functioning

			24 in individual treatment (45 mins each) Mean age=16 years (SD=1.5) Sex: 80% female Type of pain: 90% headache, 40% abdominal, 43% back, 17% joint, 1% CPRS, 20% widespread			Emotional functioning: CES-DC, Psychological Inflexibility in Pain Scale (PIPS) Parent Measures: HADS, Pain Psychological Flexibility Questionnaire (PPFQ); PRS-parent	Emotional (anxiety) and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain interference, parent anxiety and depression, pain reactivity (parent and youth), psychological flexibility (parent and child)
Kashikar-Zuck *et al,*2016	USA	Qualitative design (not specified)	17 youth Mean age=16 years (SD=2.15) Sex: 100% Female Type of pain: JFM	Outpatient multimodal treatment 16, 1.5 hours, group-based sessions, 45 mins neuromuscular training, 45 mins CBT skills)	Post-intervention	Tolerability/acceptability, safety, content, format, perceived efficacy, continued physical activity participation; semi-structerd interviews	Only the adverse events domain was considered. Tool did not align with the recommendations. Other domains: perceived efficacy, continued participation, tolerability/acceptability of the intervention
Pielech *et al.*, 2016	USA	Pre/post with single control group	114 youth and their parents Youth: Sex: 83% female Type of pain: 80% widespread pain, 20% localized pain	Intensive day-hospital rehabilitation program (40 hours/week)	Admission, discharge, 1, 6, and 12 months post	Pain intensity: VAS Pain acceptance: Chronic Pain Acceptance Questionnaire-adolescent version (CPAQ-A) Emotional functioning: PROMIS Depressive Symptoms,	Pain, physical, and emotional functioning were measured. However, only the recommended measure for pain was used. Role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed.

						PROMIS Anxiety Scale, Pain-related cognition questionnaire for children (PRCQ-C) Parent measures: Parent pain-related responses: Adult responses to children's symptoms (ARCS) Child's physical functioning: FDI parent version	Other domains: Pain acceptance, pain catastrophizing, and parent pain-related response.
Tran *et al.*, 2017	USA	Prospective cohort (Pre/post-intervention, single group)	24 adolescents with juvenile fibromyalgia (JFM) Mean age= 16 years (SD=1.59) Sex: 100% female	16 combined 60-minute CBT and neuromuscular training over 8-weeks in an outpatient setting	Pre-and post-intervention	Pain intensity: VAS (0=no pain; 10 = worst pain imaginable); Physical functioning: FDI Emotional functioning: CDI; TSK-11; PSC-C; PSOCQ, adolescent version	Pain intensity, physical and emotional function domains were considered Pain, physical, and emotional (depression) functioning measures aligned with recommendations Emotional (anxiety) functioning did not Role functioning, global judgment of satisfaction with treatment, sleep, and economic factors were not assessed Other domains: Pain catastrophizing, readiness for change, fear of movement
Bruce *et al.*, 2017a	USA	Retrospective cohort	192 eligible adolescents	Interdisciplinary chronic pain	Baseline, discharge, 3-months following discharge	Pain intensity: 11-point NRS (0=no	Pain intensity, physical, emotional (depression) and role

			171 patients Mean age = 15.3 years (SD = 1.7) Sex: 72% female Type of Pain: Headache (41%); musculoskeletal (36%); abdominal (27%) generalized (10%).	rehabilitation program	171 (89%) at discharge 79 (41%) at 3-months	pain; 10=worse pain) Physical functioning: FDI, Role functioning: type of schooling (i.e., full-time, part-time, home-schooled, tutored, no schooling or completed high school), school days missed (in the past month). Emotional Functioning: CES-DC; PCS-C Medication Use: self-report past and current opioid use	functioning domains were considered. Physical and role functioning measures align with recommendations. Emotional functioning (depression) did not. Emotional (anxiety) functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain catastrophizing, medication use
Bruce *et al.*, 2017b	USA	Retrospective study	960 participants eligible, data available on 765 participants Mean age=15.9 (SD 2.20) Sex: 74.5% female Types of Pain: headache (31.8%); abdominal pain (22.0%); generalized pain (13.0%), back pain (7.6%), and	Interdisciplinary chronic pain rehabilitation program	Pre-treatment and at discharge	Pain Intensity: 11-point NRS (0=no pain; 10=worst pain imaginable) Physical Functioning: FDI Emotional Functioning: CES-DC; PCS-C	Physical and emotional (depression) domains were considered. Physical functioning measures align with recommendations. Emotional functioning (depression) did not. Emotional (general anxiety) functioning, global judgment of satisfaction with treatment, symptoms and adverse events,

			lower extremity pain (6.4%).			Opioid use: self-report pre-treatment and at discharge	sleep, and economic factors not considered or assessed. Other domains: Pain catastrophizing, opioid use
Kempert *et al.*, 2017a	USA	Retrospective database (Pre-/post-intervention single group)	109 patients (21 excluded) 88 patients included Mean age = 15.2 years (SD = 2.28, range = 8–19) Sex: 83% female. Type of Pain: headache (25%), back pain (9.1%), abdominal pain (11.3%), amplified musculoskeletal pain syndrome (21.6%), CRPS (19.3%).	Interdisciplinary chronic pain rehabilitation program	Pre-treatment (day of admission) and post-treatment (day of discharge)	Pain Intensity: 11-point NRS (0=no pain; 10 =worst imaginable pain) in 24 hours Physical functioning: Lower Extremity Functioning Scale (LEFS) and Upper Extremity Functioning Index (UEFI)	Pain intensity and physical, functioning domains were considered. No measures aligned with recommendations. Physical, emotional (depression and anxiety), and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed.
Kempert *et al.*, 2017b	USA	Retrospective cohort study	116 patients (38 excluded) 78 patients included Mean age = 15.1 years (SD = 1.83, Range = 8-19) Sex: 77% female Types of Pain: headaches/migraine (29.1%), abdominal pain (10.4%), AMPS (11.4%), CRPS (26%)	Interdisciplinary chronic pain rehabilitation program	Pre-treatment (day of admission) and post-treatment (day of discharge	Pain Intensity: 11-point NRS (0=no pain; 10 =worst pain imaginable) in 24 hours Physical functioning: LEFS and UEFI, self and therapist-tracked objective measures (push-ups, crunches, prone extension, step ups, and jump rope	Pain intensity and physical, functioning domains were considered. No measures aligned with recommendations. Physical, emotional (depression and anxiety), and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed.

						repetitions in 60 seconds).	Note: Objective assessment tools were used for physical performance.
Sieberg *et al.*, 2017	USA	Retrospective cohort study	104 consecutive father/mother dyads and their child Mean age: 14 years (SD=2.8) Sex: Female 82% Types of Pain: Headache (15%); musculoskeletal (58%); generalized (4%)	Interdisciplinary pediatric pain rehabilitation program	Admission, discharge	Child Measures: Pain Intensity: 11-point NRS (0=no pain; 10=worse pain imaginable) at rest and with activity Physical Functioning: FDI Parent Measures: Parent Pain-Related Attitudes-Readiness to Change: PSOCQ, parent version Pain-related anxiety: PCS-Parent version Parent Behaviour-Parent responses: (ARCS) Parent perceptions of child pain-pain related fear: FOPQ-P; CPAQ-P	Pain intensity and physical, functioning domains were considered. Physical functioning measure aligned with recommendations; pain intensity measure did not. Emotional (depression and anxiety), and role functioning, global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Parent readiness for change, pain-related anxiety, pain catastrophizing, response to their child's symptoms, measures; parent-proxy measures of pain acceptance, parent perception of

Benore *et al.*, 2018a	USA	Retrospective cohort study	135 youth 135 patients included with confirmed diagnosis of pain Mean age=15.2 years (SD =2.2) Sex: 74% female	Intensive interdisciplinary pain treatment program	Admission (*n*=135), discharge (*n*=120), 1-month (*n*=29), 6-months (*n*=24), and 12-month (*n*=17) follow-up.	Pain intensity: 11-point NRS (0=no pain; 10=worse pain imaginable) Physical functioning: Physical functioning and developmental subscales of Bath Adolescent Pain (BAPQ) Emotional functioning: Depression general anxiety, pain-specific anxiety subscales of BAPQ Role functioning: Social functioning subscale of the BAPQ; Number of school days missed in the past month Family functioning: Family functioning subscale of the BAPQ	Pain intensity, physical, emotional (depression and anxiety) and role functioning domains were considered. Role functioning measure aligned with recommendations; pain intensity, physical and emotional (depression and anxiety) measures did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: HRQoL, family functioning, development

						HRQoL: Pediatric Quality of Life Inventory (PedsQL) Generic Core Scales 4.0	
Benore *et al.*, 2018b	USA	Retrospective pre/post single group	670 youth and parent participants Youth: Mean age = 15.4 (SD =2.8) Sex: 77% female Types of pain: 31% CRPS, 20% headache, 15% abdominal, 7% back, 28% other	Intensive interdisciplinary pain rehabilitation program	Admission (n=670), discharge(n=504), 6-months post (n=110)	Pain intensity: 11-point NRS (0=no pain; 10=worse pain imaginable) Physical functioning: Physical functioning and developmental subscales of Bath Adolescent Pain (BAPQ) Emotional functioning: Depression general anxiety, pain-specific anxiety subscales of BAPQ Role functioning: Social functioning subscale of the BAPQ; Number of school days missed in the past month Family functioning: Family	Pain, physical, emotional, and role functioning domains assessed. Role functioning measure aligned with recommendations; pain intensity, physical and emotional (depression and anxiety) measures did not. . Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: HRQL, youth development and family functioning, parent depression, anxiety, child-related catastrophizing, self-blame, helpless, partner relationship, leisure functioning, parental behaviour and strain

						functioning subscale of the BAPQ HRQoL: Pediatric Quality of Life Inventory (PedsQL) Generic Core Scales 4.0 Parent measures: Parent psychosocial functioning : Bath Adolescent Pain-parental Impact Questionnaire (BAP-PI) (depression, anxiety, child-related catastrophizing, self-blame, helpless, partner relationship, leisure functioning, parental behaviour, parental strain subscales.	
Buchannan *et al.*, 2018	USA	Retrospective cohort post-test single group	125 parents eligible. 41 (33%) parents participated Mean age = 15-16 years Sex: 75% female	Intensive interdisciplinary pain treatment program	Post-treatment	Global overall judgment of satisfaction: Satisfaction survey (based on hospital-based criteria)	Global overall judgment of satisfaction

			Types of pain: CRPS, headache, abdominal pain, fibromyalgia				
Randall *et al.*, 2018	USA	Retrospective cohort study	131 patients recruited 95 (73%) participated Mean age = 20 years	Intensive interdisciplinary pediatric pain rehabilitation day program	Admission, 5-years	Pain Intensity: 11-Point NRS (0=no pain; 10=worse pain imaginable) since program discharge Physical functioning: FDI HRQoL: PedsQL Role functioning: Developmental and Psychological Factors: Survey on school, work, living accommodations Health care utilization: medications prescribed and taken, health service use (number of emergency room visits, specialist and therapists' appointments, hospitalizations, diagnostic tests)	Pain intensity, physical, emotional (depression and anxiety) and role functioning domains were considered. Physical functioning measure aligned with recommendations; pain intensity, emotional (depression and anxiety) and role functioning measures did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: HRQoL, healthcare utilization, developmental factors
Risko, 2018	USA	Interpretive phenomenology	6 youth with chronic pain	Intensive interdisciplinary	At discharge	Return to meaningful activity (getting	None of the outcome domains were used, and the tool did not align with the recommendations.

			Mean age = 16 years (SD =1.0) Sex: 83% female Type of pain: abdominal, generalised, headache, neck pain, post-concussion syndrome, CRPS and other	pain treatment program		their lives back), Development of meaningful friendships, self-awareness, self-confidence, self-efficacy: semi-structutred interview	
Simons *et al.,* 2018	USA	Pre-post, single group	301 patients enrolled in the program. 280 patients consented to participate, 253 of whom had at least 3 pain data points to be included in trajectory analysis, and 194 patients had at least 3 functional disability data points to be included in trajectory analysis Mean age of the 253 patients was 14.5 (SD = 2.7, range 8–22). Sex: 84% female Neuropathic pain (61%) musculoskeletal pain (27%), headache (6%), and functional abdominal pain (6%)	Intensive interdisciplinary pediatric pain rehabilitation day program	Admission, discharge, 1-month, 4-month, and 12-month follow-ups	Pain intensity: 11-point NRS (0=no pain; 10=worse pain imaginable) Physical Functioning: FDI Emotional Functioning: CDI; MASC; PCS-C; FOPQ-C Role functioning: PedsQL 4.0 School and Social Subscales Readiness for change: PSOCQ-A	Pain intensity, physical, emotional (depression and anxiety) and role functioning domains were considered. Physical, emotional (depression) and role functioning measure aligned with recommendations. Pain intensity, emotional (anxiety) measures did not. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Readiness for change
Kashikar-Zuck *et al.*, 2018	USA	RCT	40 youth Mean age = 15 years (SD=1.5) Sex: 90% female	Outpatient multimodal treatment	Pre- (n=40), post (n=36), 3-months	Pain intensity: VAS Physical functioning: FDI	Pain intensity, physical, emotional (depression), and adverse events domains were considered.

			Type of pain: JFM	20 16 CBT sessions 20 FIT (16, 1.5 hours, group-based sessions, 45 mins neuromuscular training, 45 mins CBT skills)		Emotional functioning: CDI, TSK-11, PSC-C Adverse Events: any new or worsening symptoms	Pain intensity, physical and emotional functioning (depression), and adverse events aligned with recommendations. Emotional (anxiety) and role functioning, global judgment of satisfaction with treatment, , sleep, and economic factors not considered or assessed. Other domains: pain specific anxiety, pain catastrophizing, and kinesiophobia
Revivo *et al.,* 2019	USA	Retrospective cohort study	30 patients with joint hypermobility who participated in an intensive pain management program Mean age was 14 years (SD=2.84, range 9-18 years); 90% female.	Outpatient multimodal treatment associated with an interdisciplinary clinic	Pre- and post-treatment	Functioning: Social and physical functioning, depression general anxiety, pain-specific anxiety, family functioning, and development subscales BAPQ Pain Intensity: 11-point NRS ((0=no pain; 10=worse pain imaginable) Parent measures: Functioning: emotional functioning, catastrophic	Pain intensity, physical, emotional (depression and anxiety), and role functioning domains were considered. No measures aligned with the recommendations. Global judgment of satisfaction with treatment, symptoms and adverse events, sleep, and economic factors not considered or assessed. Other domains: Pain catastrophizing, parent measures

thinking about one’s child self-blame and hopelessness, relationship with one’s partner, leisure functioning, parental behaviour and parental strain subscales BAP-PIQ

Appendix 3.

Advisory Group Demographic Information Form

INSTRUCTIONS: This questionnaire includes 4 multiple-choice questions that explore your experience with youth with complex and chronic pain. Please check the box that provides the most appropriate answer option.

Q1. How many years have you been involved with children and adolescents with pain?

☐ 1 to 2 years
☐ 3 to 5 years
☐ 6 to 10 years
☐ More than 10 years

Q2. How many years have you been involved with services specific for children and adolescents with complex/chronic pain?

☐ 1 to 2 years
☐ 3 to 5 years
☐ 6 to 10 years
☐ More than 10 years

Q3. How many years have you been involved with children and adolescents?

☐ 1 to 2 years
☐ 3 to 5 years
☐ 6 to 10 years
☐ More than 10 years

Q4. How old are you?

☐ Less than 30 years
☐ 31 to 40 years
☐ 40 to 50 years
☐ More than 50 years

Sincerest thank you for your assistance!

Appendix 4.

Advisory Group Domains and Measures Questionnaire

Purpose: This questionnaire includes 2 short answer questions that will assist in providing us with some information about your thoughts on the current intensive rehabilitation program to help us better understand what information you require to decide on the value of a pain intervention program.

Instructions: Carefully read the question and fill in the space below. Please note there are no wrong answers and all information is welcomed.

Q1. What are the effects that you consider most important to measure when evaluating the effects of an intervention for yourself/your child/your patient/your student with chronic pain?

Q2. What information do you need to judge whether a treatment was successful or not? Please provide examples.

Sincerest thank you for your thoughts and collaboration!

Appendix 5.

Parent Program Selection Information Form

INSTRUCTIONS: This questionnaire includes 2 questions that will help us better understand the reason you selected to participate in this treatment and not another. Please check all the boxes that apply.

Q1. I decided to participate in this treatment program because…

- ☐ My child's condition
- ☐ My child's academic requirements
- ☐ My child's social needs
- ☐ The clinic team's assessment and recommendations
- ☐ My work situation
- ☐ The needs of the other children in the family

- ☐ Other family commitments
- ☐ Our financial situation
- ☐ The time commitment
- ☐ The amount of treatment

- ☐ The distance we live from the hospital
- ☐ Other

Q2. Can you please expand in more details on your answer in Q1.

Sincerest thank you for your help!

Appendix 6.

"Is it Worth It?" Advisory Committee Team Charter

1. Purpose Statement and Team Objectives: The team has been formed to guide and supervise until completion a research study at Alberta Children's Hospital (ACH).

1.1 **Team Objective**: The core objective of our team is to provide recommendations to ACH leadership team for their consideration in the future programming and clinical service decisions as they relate to youth with pain-related disability. To do so, the team commits to working effectively as a team, by monitoring our process effectiveness, following through on commitments, and helping each other learn.

1.2 **Research Project Objective**: The objective of the study is to evaluate the effectiveness of the Intensive Pain Rehabilitation Program (IPRP) in treating adolescents suffering from chronic pain compared to a standard multimodal outpatient program.

2. Analysis of Strength and Developmental Needs: In considering the team's purpose and objectives, we analysed the skill sets of our team members in relation to the nature of the work we have to complete. We identified background, experience and complementary skills of each team member and defined individual levels of accountability.

2.1 Strengths:

2.1.1 ***Knowledge and Skills****:* Members include one manager, a program coordinator, five clinicians with experience treating youth with chronic and complex pain (i.e. a physician, a psychologist, a physiotherapist, occupational therapist, a clinical nurse specialist/IPRP program coordinator), two teachers from the largest school board in the province, two parents and two youth who have a history of chronic pain treated for at least a year at ACH. Many members of the team have training and experience in research and/or program evaluation. Others have a good understanding and working knowledge of the pain and disability theoretical models, outcome measures and questionnaires validated for youth with pain-related disability and disability related to other conditions. Two members have experience at an organizational and governmental policy level, which will serve the group well in devising the recommendations related to the program and the knowledge transfer strategy. A broad network of colleagues within the paediatric pain research community is also accessible thanks to group members with previously established collaboration, which will aid in the broader dissemination of the results of this project.

2.1.2 ***Background and Experience****:* The clinicians, teachers, manager, youth and parents have experience with a wide variety of pain and rehabilitation intervention models, including standard multimodal outpatient interventions, rehabilitation day-hospital, and intensive pain rehabilitation programs. Furthermore, most committee members have over 10 years experience with children and adolescents, at least 6 years of involvement with chronic pain, and a minimum of 3 years of association with services addressing the specific needs of youth with chronic pain and their families (see Appendix 1 for stakeholder specific experience). The team has also advanced expertise in the paediatric rehabilitation of youth with other chronic physical and mental health conditions, and in developing and sustaining partnerships with important associated community agencies and partners. The group's background and experiences are well rounded and is representative of the various spheres of a child's life that includes home, school, family, and community and leisure activities. Committee member experience and expertise also links with other spheres of knowledge (e.g. sports psychology, physical fitness training, mental health models, coaching, pedagogy, change management), useful in broadening the perspectives considered relevant throughout the various project phases.

2.1.3 ***Interpersonal Skills****:* All members have extensive experiences working collaboratively and respectfully in team environments. This has been evidenced in the activities of Phase 1 by strong communication skills, flexibility in their opinions and thought processes, and a commitment to engagement and participation by all members. Domination by one individual or group is absent, while a strong interest to building consensus and shared understanding, and a genuine attention of learning from each other has been visibly apparent. A shared hope that involvement in this project will make a difference in the lives of youth living with chronic/complex pain and their families has also been voiced by members.

2.2 Developmental Needs:

2.2.1 ***Meeting Scheduling*****:** As many members identify busy schedules as a potential barrier to their active participation, the meetings will be scheduled with at least 3 weeks notice. Furthermore, various meeting methods will be adopted such as multiple small-group meetings of the committee on the same topic, and the possibility of providing feedback via email, telephone or teleconferencing as alternate methods of gathering the members' perspectives.

2.2.2 ***Limited Experience with the Research and Evaluation Processes*****:** Several members identified a lack of experience with the research process used in this study and in evaluation methodology. In order to address this, formal presentations have been conducted related to participatory research and program evaluation and distributed via email to members as a reference. Data collection, measurement tools and data analysis processes will be reviewed in upcoming meetings in order to further build capacity within the group. Finally, team members well versed in various research processes (including qualitative and quantitative methods) have agreed to act as resources for other members.

2.2.3 ***Limited experience with the study population***: Several members acknowledged limitations in their exposure and work experience with this population. A depth of work and lived experience in other team members will be used provides to counterbalance this limitation.

2.2.4 ***Managing relationships amongst the various stakeholder groups***: It has been acknowledged by all members that the interest of the various stakeholder groups may vary significantly, posing potential risk for frictions and conflict amongst stakeholder groups or members. Regular reminders of the realistic expectations of the interventions studied throughout the research process and the outcomes of the project will be provided by the research. However, should, at any time, a stakeholder group or individual committee member feel uncomfortable with the discussion, if communication breakdowns occur between members outside of meetings or if one fear potential repercussions as a result of voicing opinions, they are encouraged to follow the conflict resolution process outlined below.

3. Functional Roles:

- Researcher/Facilitator –
- Research Assistant/Recorder –
- Clinicians
  - Physician -

        - Clinical Nurse Specialist -
        - Psychologist –
        - Physiotherapist –
        - Occupational Therapist –
    - School/Education
        - Teacher –
    - Management
        - Unit Manager and Day Hospital Program Facilitator –
    - Patient Perspective
        - Parents –
        - Youth -

4. Team Process Management:

4.1 Team Meetings: Meeting will be scheduled using a Doodle Poll to verify team member's availability 3 weeks prior to meetings scheduling. Every effort will be made to incorporate the perspective of each team member into a face-to-face meeting time. Email or telephone follow-up members who are unable to attend will be used a secondary method if all alternative meeting methods have been exhausted.

The purpose of the meeting will be clearly identified in the meeting call at the time the Doodle Poll is circulated.
The agenda topic(s) will be fixed with the meeting confirmation.

4.2 Member Responsibilities

Every meeting will have a facilitator (i.e. the researcher).
4.2.1 ***The facilitator*** (i.e. researcher) will be responsible for:

- Meeting planning, including calling meeting, and agenda preparation & distribution,
- Clearly outline meeting preparation and discussion expectations,
- Keeping the meeting on topic,
- Guide the team through discussion topics,
- Encouraging the participation of all members,
- Promoting an atmosphere of shared learning, open and honest discussion, and respect,
- Facilitating resolution of differences of opinion (if required),
- Ensuring that members adhere to described meeting guidelines,
- Summarizing previous group discussions on the same topic (if applicable),
- Outlining next steps and delegate tasks (if appropriate),
- Distributing a synthesis of the data gathered on a specific topic and any key outcomes and decision-made by the group,
- Update and report on the critical path the project progres

Every meeting will be audio-recorded.
4.2.2 ***The recorder*** (i.e. research assistant) will be responsible for:

- Documenting meeting discussion and observations,
- Recordings any of the key meeting outcomes, activities, and decision
- Keeping track of time in relations to meeting topics

The audio-recordings and the recorder's notes will be used to prepare group documentation and reports that will be shared with the committee.

4.2.3 ***Code of conduct***: All team members will be expected to:

- Arrive on time, so that meetings may be completed within designate timeframe;
- Be prepared to the meeting, having reviewed any pre-circulated materials if circulated prior to the meeting;
- Adhere to the agenda topics
- Follow meeting ground rules (see Appendix 2)
- Respect one person speaking at a time, and limit/avoid sidebar conversations.
- Actively listen and contribute to the discussion

4.3 Expectations of Team Members:

Meeting attendance: Attendance is expected for every meeting. If a member is absent, a secondary method (such as telephone or teleconferencing) may be considered. On-time arrival for the meeting is also expected.

4.3.1 ***Participation****:* Full participation is expected of all team members. If someone feels that others are not doing their job, the concerned member will be encouraged to discuss it one-on-one, off-line. If that doesn't work, then the member will bring it up with either the facilitator, or the manager/designate.

4.3.2. ***General Courtesy and Respect****:* Members will respect the different role of each member designate. Discussion that deviates from the current topic and objectives should be avoided. The facilitator will bring the focus back to the topic at hand.

Should a member be deemed not to be meeting team member or discussion expectations, the facilitator will privately meet with them and explore barriers, facilitators and strategies that will assist the member in meeting expectations. For clinicians, information will be shared with manager as appropriate.

4.4 Decision Making Procedure:

An open discussion will occur on all topics with the aim of soliciting opinions from all members. Facts and experience will be used to generate resolutions. At conclusion of discussion, feedback will be sought for any unresolved issues related to the topic. Opportunities will be provided to all team members for final comments prior to the closure of any discussion.

Group consensus will be prioritized as the main means of decision-making. As a group, we defined consensus as all members being able to say: "This may not be my ideal decision outcome, but I can live with it and support it."

4.1 Consensus Procedure

1. A proposal for resolution is put forward.
2. The proposal can be amended and modified through discussion
3. Those participants that disagree with the proposal have the responsibility to put forward alternative suggestions or rewording.
4. When a proposal seems to be well understood by everyone, and there are no new changes are asked for, the facilitator will ask if there are any objections to it.
6. If there are no objections, the facilitator will call for consensus. Silence by committee members will indicate that consensus has been achieved.

8. If consensus does appear to have been reached, the facilitator will repeat the decision to the group, so everyone is clear on what has been decided.

If consensus can still not be achieved, the impact of the decision on other future decisions will be discussed by the group and an alternate decision-making process may be used.

4.2. Alternative Decision-Making Procedure

1. The facilitator will review the outcome attempting to be achieved with the group.
2. The group will create a decision matrix with the different decision options listed as the rows, and the relevant factors affecting the decisions listed as the columns.
3. A rating scale will then be established for each option/factor combination.
4. The original ratings will be multiplied by the weighted rankings to get a score. All of the factors under each option should then be added up. The option that scores the highest is the decision that should be made, or the first item addressed.

Table 1 presents an example of such a decision-making matrix:

Table 1. Decision Matrix

	Rating Scale	30%	45%%	10%	15%
		Factor 1: Feasibility	Factors 2: Cost	Factor 3: Alignment	Factor 4: Desirability
Option A	40				
Option B	30				
Option C	20				

4.3 Conflict Resolution

Each committee member will have a unique way of viewing the world, so conflict is not necessarily a bad thing. In fact, differences of opinion often foster open communication, promote change, and lead to better decision-making.

However, should conflict arise during meeting discussions between stakeholder groups or individuals that are judged by the facilitator to be detrimental to future team functioning, she will attempt to resolve it by:

- Determining the cause of the conflict and clarifying the problem;
- Rephrasing the issue to depersonalize it, focusing on the topic not the person;
- Promoting the presentation of all the various facts and information related to the issue;
- Clarifying, questioning, and facilitating other committee members to do the same;
- Finding common ground upon which to begin to explore solutions;
- Proposing possible solutions to resolve the conflict and exploring their consequences; and
- Identifying an action plan that can be agreed upon.

Should an individual committee member feel uncomfortable with the discussion, if communication breakdowns occur between members outside of meetings or if one fear potential repercussions as a result of voicing opinions, the member will be encouraged to contact the researcher, their manager or the manager designate. The manager or her designate and the researcher will meet to determine how and who will handle such situations, respecting the broader organizational policies, guidelines, and leadership structure. Members will initially be encouraged to address the issue with the other member in question. Coaching, and if necessary, mediation will be provided. Regular check-in with group members will also be conducted to verify their ongoing comfort with the process and to investigate any potential conflicts or discomforts.

4.4 Team Communication

We will communicate with one another using various means, including face-to-face meeting, email, telephone or teleconferencing. Face-to-face and teleconferencing are the preferred methods of communication to promote exchange of ideas, opinions and learning. Prompt responses to messages/inquires to one another are expected and may include a targeted deadline for response.

5. Assessment of Team Effectiveness:

At the conclusion of twice yearly, a review will be conducted to assess the team's effectiveness in relation to its stated team objectives (i.e. Are we meeting the goals of each phase of our project? Are we where we need to be in order to complete our project on time?) and the effectiveness of team collaboration (i.e. Are we living up to our process management guidelines?). A critical path of the project will be distributed to the committee quarterly in order to assist in the assessment team effectiveness related to its objectives and project progress.

The following Team Feedback Survey will be administered at the end of each meeting to assess members perception of collaborative effectiveness. Adjustments to the team processes and procedures will be based upon these reviews.

Team Feedback Survey

1. Overall, how effectively has your team been working together on this project?

1	2	3	4	5
not at all	poorly	adequately	well	extremely well

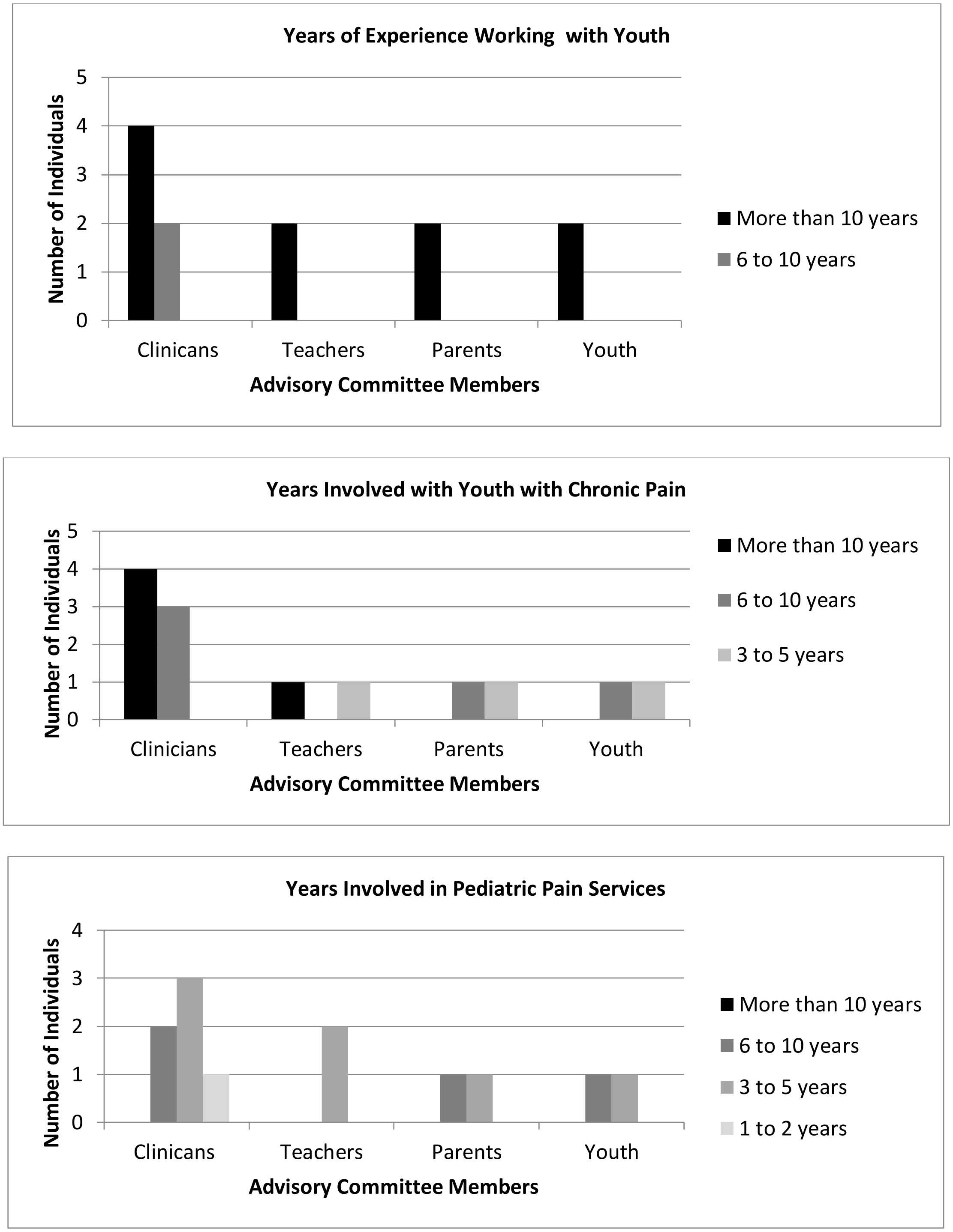
2. How many of the team members participated actively most of the time?
3. How many seemed fully prepared for the teamwork most of the time?
4. Give one specific example of something you learned from the team that you probably would not have learned on your own.
5. Give one specific example of something other team members learned from you that they probably would not have learned without you.
6. Suggest one specific, practical change the team could make that would help improve everyone's learning.

6. Review of Team Charter

This charter will be distributed electronically every six months for feedback in order to maintain its currency and to ensure it continues to reflect the teams functioning and process. Feedback will be discussed at the next team meeting, changes will be made as needed following the discussion, and member will be asked to re-endorse the charter following any significant change.

Figure. 1

Stakeholder Groups Specific Experience at Recruitment (September 2016)



Team Charter – Agreed Upon Meeting Ground Rules

1. Tough on topic, easy on people
2. If the horse is dead, get off it
3. Generally correct, not specifically wrong.
4. Aim to see things from more than one perspective
5. If the sale is made, stop selling.
6. Influence the future, don't score keep the past.
7. One conversation at a time
8. No sacred cows
9. Everyone contributes, no one dominates.
10. Think outside the box.

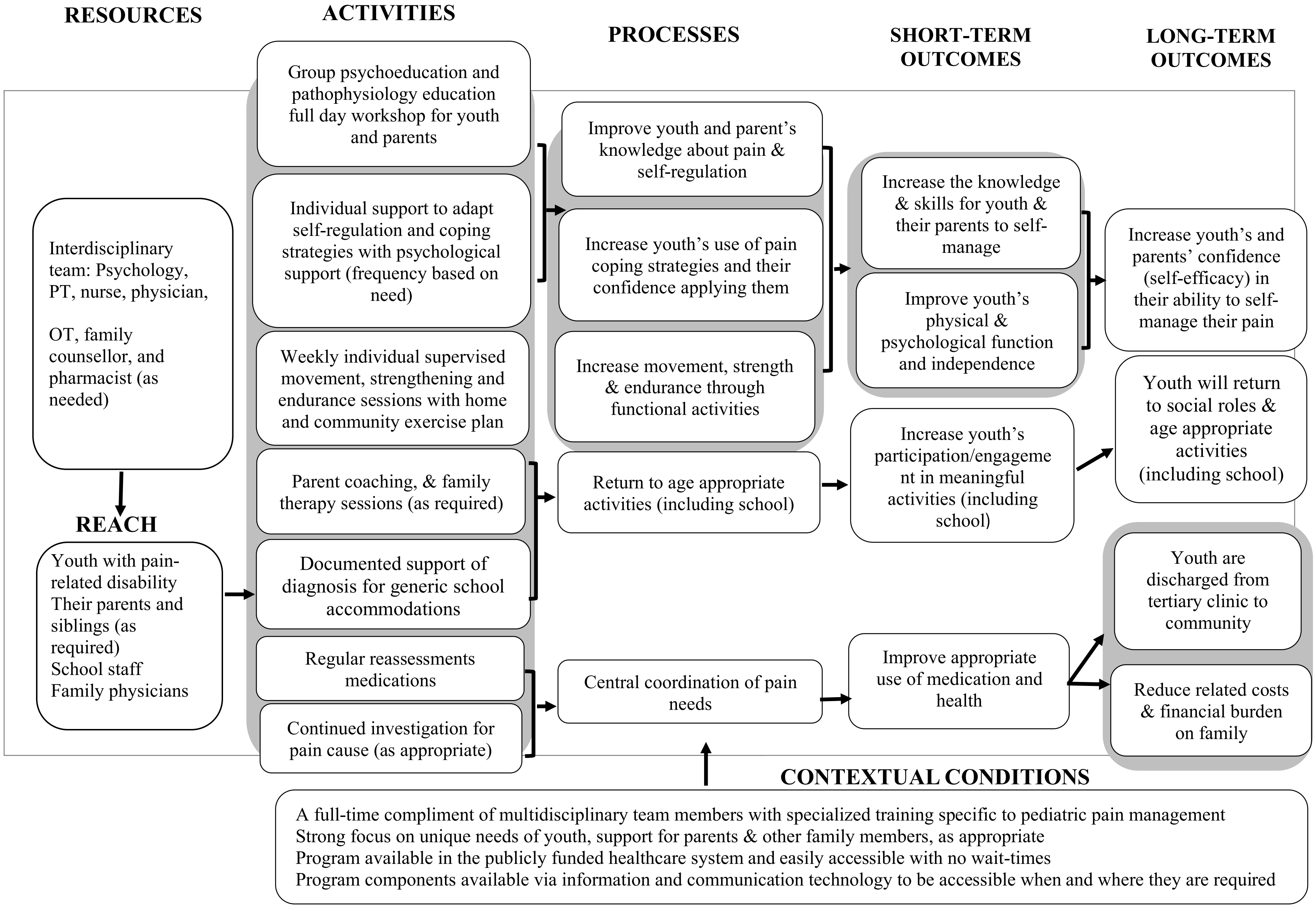
Developed by Shauna Lagenberger, MN, Quality Improvement Consultant, Alberta Health Services

Team Members' Endorsement

By my signature, I endorse this charter, commit, to the best of my ability, to upholding the expectations of a team member and to following the outlined processes and procedures.

Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date
Signature	Date

Appendix 7.
MMT Logic Model Resulting from Logic Analysis



RESOURCES

Interdisciplinary team: Psychology, PT, nurse, physician,

OT, family counsellor, and pharmacist (as needed)

REACH

Youth with pain-related disability
Their parents and siblings (as required)
School staff
Family physicians

ACTIVITIES

Group psychoeducation and pathophysiology education full day workshop for youth and parents

Individual support to adapt self-regulation and coping strategies with psychological support (frequency based on need)

Weekly individual supervised movement, strengthening and endurance sessions with home and community exercise plan

Parent coaching, & family therapy sessions (as required)

Documented support of diagnosis for generic school accommodations

Regular reassessments medications

Continued investigation for pain cause (as appropriate)

PROCESSES

Improve youth and parent's knowledge about pain & self-regulation

Increase youth's use of pain coping strategies and their confidence applying them

Increase movement, strength & endurance through functional activities

Return to age appropriate activities (including school)

Central coordination of pain needs

SHORT-TERM OUTCOMES

Increase the knowledge & skills for youth & their parents to self-manage

Improve youth's physical & psychological function and independence

Increase youth's participation/engagement in meaningful activities (including school)

Improve appropriate use of medication and health

LONG-TERM OUTCOMES

Increase youth's and parents' confidence (self-efficacy) in their ability to self-manage their pain

Youth will return to social roles & age appropriate activities (including school)

Youth are discharged from tertiary clinic to community

Reduce related costs & financial burden on family

CONTEXTUAL CONDITIONS

A full-time compliment of multidisciplinary team members with specialized training specific to pediatric pain management
Strong focus on unique needs of youth, support for parents & other family members, as appropriate
Program available in the publicly funded healthcare system and easily accessible with no wait-times
Program components available via information and communication technology to be accessible when and where they are required