# RISK AND RESILIENCE IN RECOVERY FROM SURGERY FOR ADOLESCENT IDIOPATHIC SCOLIOSIS

by

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“Although the world is full of suffering, it is also full of the overcoming of it”
*– Helen Keller*

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## ABSTRACT

The majority of children who undergo major surgery for scoliosis tend to recover well, but there is variability in recovery outcomes with retrospective studies demonstrating that some patients report impairments in certain aspects of their quality of life as well as pain that can impact functioning. A few prospective studies of pediatric postsurgical recovery have been conducted more recently, but most of what is known about postsurgical pain and functioning comes from the adult literature. Additionally, most studies to date are focused on identifying risk factors for the development of chronic postsurgical pain with little attention paid to protective factors related to postsurgical pain and functioning. This dissertation sought to further our understanding of pediatric postsurgical pain and recovery following major surgery and to examine recovery from a resilience perspective. Paper one used a prospective longitudinal cohort design to identify postsurgical pain trajectories over the first postsurgical year in adolescents ($n$ = 202) who underwent surgery for scoliosis. Differences in functional outcomes at one year were examined across identified trajectories and baseline parent and child predictors of trajectory membership were examined. Three distinct trajectories were identified. The majority of adolescents followed a trajectory with low pain and good functional outcomes. The other two trajectories were characterized by high immediate postsurgical pain but only one was characterized by pain that persisted over the first year. Patients in the latter trajectory reported worse functional outcomes at one year and membership in the trajectory was predicted by greater baseline pain and child anxiety. Paper two was a topical review that provided a rationale for identifying and examining protective factors in pediatric postsurgical recovery, reviewed the pediatric chronic pain and adult postsurgical literature to identify potentially relevant protective factors at the individual, parent and peer levels and provided specific recommendations for future research. Paper three examined the role of optimism, a protective factor, in pediatric postsurgical recovery outcomes at two time points in the postsurgical recovery period using a sample of adolescents ($n$ = 151) who had undergone surgery for scoliosis. Optimism was predictive of quality of life at one year after surgery and was found to moderate the relation between pain and functional disability one year after surgery. Optimism was not related to either quality of life or functional disability during a more acute phase of recovery. Taken together, the findings of this dissertation demonstrate that a small subset of adolescents are at risk of developing chronic postsurgical pain and poor functioning but that the majority recover well from major spinal surgery and that optimism may be one of several protective factors that could be used to promote good postsurgical outcomes.

## LIST OF ABBREVIATIONS AND SYMBOLS USED

| | |
|---|---|
| ACT | Acceptance and Commitment Therapy |
| AIC | Akaike Information Criterion |
| AIS | Adolescent Idiopathic Scoliosis |
| ANOVA | Analysis of Variance |
| *B* | Unstandardized Coefficient |
| BIC | Bayesian Information Criterion |
| BLRT | Bootstrapped Lo-Mendell-Rubin Test |
| CASQ | Children's Attributional Style Questionnaire |
| CIHR | Canadian Institutes of Health Research |
| $\alpha$ | Cronbach's alpha coefficient |
| *d* | Cohen's d |
| *F* | *F* statistic for ANOVA |
| FDI | Functional Disability Inventory |
| FIML | Full Information Maximum Likelihood |
| GMM | General Mixed Modelling |
| HRQOL | Health-related Quality of Life |
| LGBTQ | Lesbian Gay Bisexual Transgender Queer |
| LGMM | Latent Growth Mixed Modelling |
| LMRT | Lo-Mendell-Rubin likelihood ratio test |
| *M* | Mean |
| M*plus* | M*plus* statistical software |
| *n* | Sample size |
| NRS | Numerical Rating Scale |
| *p* | *p*-value for Testing of significance |
| PCS | Pain Catastrophizing Scale |
| PedsQL | Pediatric Quality of Life Inventory |
| PORSCHE | Post-Operative Recovery following Spinal Correction – Home Experiences |
| PROCESS | macro used to perform moderation in SPSS software |
| *r* | Pearson Correlation Coefficient |
| $R^2$ | Squared Multiple Correlation Coefficient |
| RCT | Randomized Control Trial |
| *SD* | Standard Deviation |
| *SE* | Standard Error |
| SPOR | Strategy for Patient Oriented Research |
| SRS | Scoliosis Research Society |
| *STAI* | State-Trait Anxiety Inventory |
| *t* | *t*-test Statistic |
| $\chi^2$ | Chi-squared test statistic |

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# CHAPTER 1. INTRODUCTION

This dissertation focuses on understanding pediatric postsurgical pain and recovery following major surgery and examining pediatric postsurgical pain and recovery from a resilience perspective. This dissertation addresses three main objectives: 1) Examine postsurgical pain trajectories and functioning following major pediatric surgery, 2) Review protective factors relevant to pediatric postsurgical recovery, 3) Examine one protective factor for pediatric postsurgical recovery in greater depth. These objectives were addressed through three publication-style manuscripts. Before presenting the three individual papers, the current chapter will provide an introduction to the relevant literature that informs this research, as well as rationale for the current research.

## 1.1 Scoliosis and Surgical Treatment

Scoliosis is the most common spinal deformity, and is defined as, "a lateral curvature of the spine that is 10 degrees or greater on a coronal radiographic image while the patient is in a standing position" (Hresko, 2013, p. 834). There are several types of scoliosis that are categorized according to cause. Congenital scoliosis is due to improper formation of the spinal column which may lead to progressive deformity with growth. Neuromuscular scoliosis is deformity of the spine due to dysfunction of the central nervous system, dysfunction of the peripheral neuromuscular system or a combination of sensory and motor dysfunction. The majority of patients do not have a known cause for their scoliosis, which is thus deemed idiopathic scoliosis. Idiopathic scoliosis is subclassified as infantile (birth to 3 years), juvenile (3 to 10 years), adolescent (more than 10 years) and adult.

As children grow, disfigurement of the torso, shoulder and waist asymmetry, trunk imbalance and rib rotation are the primary complication of scoliosis (Miller, 1999; Roach, 1999).

Mild pain can be common with significant curvature but severe pain that interferes with functioning is rare (Hresko, 2013). Increasing amounts of curvature can interfere with lung function as growth of the thoracic spine is necessary to achieve adult chest volume. Curves greater than 50 degrees can result in shortness of breath later in life (Weinstein et al., 2003), curves greater than 50 degrees can result in reduced lung volume and curves greater than 100 degrees can result in restrictive pulmonary disease (Johnston et al., 2011).

Idiopathic scoliosis is not progressive in most patients, but the main determinants of progression are female gender, maturity (age at diagnosis, menarchal status, amount of skeletal growth remaining), curve magnitude and position of the curve apex (Miller, 1999; Weinstein, Dolan, Cheng, Danielsson, & Morcuende, 2008). Curves of small magnitude are equally prevalent in females and males, but as curve size increases, females predominate over males by as much as ten to one (Miller, 1999; Reamy & Slakey, 2001; Weinstein et al., 2003). Curves with a thoracic apex are more likely to progress (Brunnell, 1986; Picault, DeMauroy, & Mouilleseaux, 1981). Curve magnitude of less than 30 degrees in late adolescence rarely worsens into adulthood whereas curves greater than 50 degrees tend to worsen at a rate of .75 to 1.00 degrees per year and are associated with more health problems in adulthood, reduced quality of life, disability, and progressive functional limitations (Lonstein, 2006; Negrini et al., 2006; Weinstein et al., 2003; Weinstein, Zavala, & Ponseti, 1981).

### 1.1.1 Adolescent Idiopathic Scoliosis

Adolescent Idiopathic Scoliosis (AIS) is the most common form of scoliosis and occurs in approximately two to four percent of children between the ages of 10 to 16 years (Roach, 1999). As with all types of Idiopathic scoliosis, more females than males tend to have significant curve progression that requires treatment. No single cause of AIS has been identified, but the

most widely investigated causes include hormonal imbalance, asymmetric growth and muscle imbalance (Scoliosis Research Society, 2020). Studies have also found that approximately 30 percent of AIS patients have a family history of scoliosis, suggesting a genetic component (Reamy & Slakey, 2001). The majority of individuals with AIS are generally healthy and do not have any other significant medical history (Scoliosis Research Society, 2020).

Treatment for AIS can involve both non-operative and operative techniques. The goal of non-operative treatment is to prevent further curve progression whereas the goal of operative treatment is to more aggressively prevent further curve progression, achieve maximal and permanent correction, improve appearance and minimize short-term and long-term complications (Weinstein et al., 2008). Surgical treatment in adolescents is generally indicated when the individual has a primary curve greater than 45 degrees (Weinstein et al., 2008). Several types of surgical procedures can be used to treat AIS, but the type of procedure chosen depends upon the age of the child, their prognosis, and the shape of the curve. Posterior Spinal Fusion with Instrumentation tends to be the most common surgical treatment for idiopathic scoliosis (Lonner, Kondrachov, Siddiqi, Hayes, & Scharf, 2006; Maruyama & Takeshita, 2008). The procedure provides permanent stabilization in the corrected spinal position by removing joints between vertebrae and placement of metal rods to hold the spine in place while the vertebrae fuse and heal. Recovery from this procedure occurs in stages with most adolescents staying in hospital for several days, returning to school within two to four weeks and returning to regular activities within two to twelve months (American Academy of Orthopaedic Surgeons, 2020)

### 1.1.2 Recovery from Surgery for AIS

Overall, adolescents appear to recover well from surgery to treat AIS, but there is variability in recovery outcomes. Long-term, retrospective studies of patients who have received

surgical treatment for AIS are generally favorable and describe either comparable or mildly lower activity level and mental health than healthy controls (Andersen, Christensen, & Thomsen, 2006; Bjerkreim, Steen, & Brox, 2007; Danielsson & Nachemson, 2003; Mariconda, Andolfi, Cerbasi, & Servodidio, 2016; Takayama, Nakamura, & Matsuda, 2009). However, there are also several longitudinal studies that suggest patients experience persistent concerns about other aspects of quality of life such as their appearance and social interactions as well as persistent pain following surgery (Bartie, Lonstein, & Winter, 2009; Danielsson, Wiklund, Pehrsson, & Nachemson, 2001; Goldberg, Mayo, Poitras, Scott, & Hanley, 1994; Rushton & Grevitt, 2013; Weigert, Nygaard, Christensen, Hansen, & Bünger, 2006). Due to the retrospective design of most of these studies, it is unclear whether the persistent pain reported by participants is unresolved presurgical pain or new onset postsurgical pain. Qualitative research suggests that the pain is intense during the first few months of the postsurgical period and that it negatively impacts mobility, social interactions and daily activities (Rullander, Jonsson, Lundström, & Lindh, 2013). Compared to the general population, AIS surgical patients report greater levels of pain and back-related disability decades after their spinal surgery (Danielsson & Nachemson, 2003; Goldberg et al., 1994; Wong, Yuen, Chow, & Irwin, 2007) but it appears that only a small percentage of patients report that the pain is severely disabling (Danielsson et al., 2001). In conclusion, the majority of patients who undergo spinal surgery to treat scoliosis tend to recover well but some patients continue to experience impairments in certain aspects of their quality of life as well as persistent pain that can impact their functioning.

### 1.2 Postsurgical Pain Trajectories and Functioning

While several longitudinal, retrospective studies have been conducted to examine long-term postsurgical recovery outcomes in scoliosis surgery patients, the course of postsurgical pain

and the factors that contribute to postsurgical pain and functioning in children after major surgery is not well understood. It has been argued that understanding pain resolution over time is key to understanding the development of chronic postsurgical pain and the factors that predict chronic postsurgical pain (Chapman, Donaldson, Davis, & Bradshaw, 2011).

In adult populations, the persistence of postsurgical pain has been extensively studied (Glare, Aubrey, & Myles, 2019; Katz & Seltzer, 2009a; Kehlet, Jensen, & Woolf, 2006) and researchers have begun to identify unique trajectories of pain resolution and non-resolution over time (Althaus et al., 2018; Pagé et al., 2015; Thomazeau et al., 2016). Several important conclusions can be drawn from these studies. First, there is substantial individual variation in postsurgical pain intensity. Second, while many individuals demonstrate pain trajectories characterized by diminishing pain over time there appears to be a subset of individuals who report high levels of pain that is slow to resolve. Lastly, these studies suggest that levels of preoperative pain intensity and the rate of pain resolution in the immediate postsurgical period are important factors for predicting chronic postsurgical pain. Only one of the above mentioned studies also examined functional outcomes in the postsurgical period and found that membership in a postsurgical pain trajectory characterized by persistent pain was associated with more functional impairment 12 months after surgery (Pagé et al., 2015). This is consistent with many other adult studies showing the chronic postsurgical pain is associated with delayed return to normal activity (Macrae & Davies, 1999) and impaired quality of life (Courtney, Duffy, Serpell, & O'Dwyer, 2002; Gottschalk & Ochroch, 2008; Poobalan et al., 2003; van Hanswijck de Jonge, Lloyd, Horsfall, Tan, & O'Dwyer, 2008).

It is important to understand the course of postsurgical pain in children who have undergone major spinal surgery to determine if similar patterns of pain resolution exist. It is also

important to determine how postsurgical pain in children relates to functional outcomes in the postsurgical period. Data about pain trajectories and functional outcomes is also essential to identify children who may be slower to recover and to determine how and when to intervene to prevent the development of chronic postsurgical pain and poor functioning.

A few studies have investigated postsurgical pain trajectories and functional outcomes in children. Sieberg and colleagues (2013) examined postsurgical pain in children who had undergone spinal fusion to treat scoliosis at one, two ($N = 190$) and five ($N = 77$) years after surgery. They identified five distinct trajectories with the majority of the sample being classified into either a no pain (11.6%), short-term pain (18.4%) or improved pain (53.7%) trajectory. A smaller proportion of children were classified into a delayed pain (10.5%) or high pain (5.8%) trajectory. The high pain trajectory was characterized by persistent pain five years after surgery, and participants classified into this trajectory had greater baseline pain and more missed days of school compared to members of other trajectories. The findings presented by Sieberg and colleagues provide valuable information about the individual variation in children's postsurgical pain course but given the limited measurements of pain during the first postsurgical year, when pain is thought to transition from acute to chronic, studies with more frequent pain measurements are warranted.

This limitation was addressed in part by Connelly and colleagues (2014) who sought to describe a typical pain trajectory for children ($N = 50$) in the first six months after spinal fusion and to examine predictors of change in pain scores in this time period. They measured pain at baseline and five time points after surgery (in-patient stay, two weeks, six weeks, three months and six months). Results indicated that pain levels declined in the days after surgery but that 22% of the sample exhibited pain that was the same as or above baseline levels for six months after

surgery. They also found that greater baseline pain and anxiety predicted slower improvement in pain whereas greater pain self-efficacy predicted more rapid improvement in pain. Frequent pain assessments were also used by Rabbitts and colleagues (2015) who measured postsurgical pain at four time points (in-hospital, two weeks, four months and 12 months) during the first postsurgical year in a sample of children ($N = 60$) who underwent spine or chest wall surgery. They identified two trajectories which they labelled as, 'early recovery' and 'late recovery'. The majority of their sample ($N = 49$) was classified into the early recovery trajectory which was characterized by increased pain in the immediate postsurgical period that rapidly declined in the first few months after surgery and remained low until one year. A smaller proportion of their sample ($N = 11$) was classified into the late recovery trajectory which was characterized by increased pain in the first few weeks after surgery that then declined slowly over the postsurgical year. Individuals in this trajectory had significantly worse quality of life and activity limitations at twelve months and membership in this trajectory was predicted by greater baseline pain and parent pain catastrophizing. Although this study is a valuable contribution to the literature, its limited sample size may have prevented the authors from identifying a, 'true', persistent pain trajectory.

One more recent study that included both frequent pain measurements during the first postsurgical year and a larger sample size ($N = 265$), also identified two trajectories (Rosenbloom et al., 2019). One trajectory ($N = 136$) was characterized by mild baseline pain which remained mild immediately after surgery and throughout the first postsurgical year. An almost equal number of children were classified into a second trajectory ($N = 123$) which was characterized by moderate to severe baseline pain which remained moderate to severe throughout the postsurgical year. Membership in this second trajectory was predicted by baseline functional

disability (Rosenbloom et al., 2019). The findings presented by Rosenbloom and colleagues address several limitations of previous studies, however, it should be noted that this study was conducted with a mixed surgical sample that included spinal surgery for scoliosis as well as several other major orthopedic procedures. This mixed sample makes the results difficult to generalize to a purely scoliosis surgical sample. More studies are needed to prospectively measure the course of pain following pediatric major spinal surgery and examine how different trajectories of pain resolution relate to functional outcomes in this population.

### 1.3 Predicting Postsurgical Recovery

A model describing the development of chronic postsurgical pain in adults was proposed by Katz and Seltzer (2009). The model divides the perioperative period into distinct phases: preoperative, intraoperative, acute postoperative and chronic postoperative. The authors use a biopsychosocial framework to suggest factors within each perioperative phase that may contribute to the development of acute and chronic postsurgical pain. Psychosocial factors are present in all of the perioperative phases with the exception of the intraoperative phase. In the preoperative phase, state anxiety and pain catastrophizing are proposed to be associated with acute and chronic postsurgical pain and mental health in the chronic postoperative phase. In the acute postoperative phase, pain catastrophizing, social support and solicitous responding are proposed to be associated with acute postoperative pain and with chronic postoperative pain and disability. In the chronic postoperative phase, mental health symptoms are proposed to be associated with both chronic postsurgical pain and disability.

Katz and Seltzer's model has been widely cited in the adult literature and used to guide empirical research to further our understanding of factors related to the transition from acute to chronic postsurgical pain (Bayman, Parekh, Keech, Selte, & Brennan, 2017; Bruce et al., 2014;

Castillo et al., 2013; Chaparro, Smith, Moore, Wiffen, & Gilron, 2013; Clarke, Soneji, Ko, Yun, & Wijeysundera, 2014; Gerbershagen et al., 2014). However, one criticism of the model is that it focuses mainly on factors that predict pain with a lesser emphasis on factors that predict functional outcomes such as disability and quality of life. According to the fear-avoidance model of chronic pain (Vlaeyen & Linton, 2000) and its pediatric adaptations (Asmundson, Noel, Petter, & Parkerson, 2012; Simons & Kaczynski, 2012), functional outcomes are central to understanding both the development and maintenance of chronic pain, as well as for understanding the impact of pain on an individual. The fear-avoidance model of chronic pain describes a cyclical pattern of pain-related cognitions (e.g., pain catastrophizing, pain-related fear) leading to avoidance and hypervigilance which contribute to depression and disability which continue to influence an individual's experience with pain. Therefore, any model designed to further research in the area of pediatric postsurgical recovery should emphasize factors related to postsurgical functioning as well as postsurgical pain.

Katz and Seltzer's model is also limited in its application to a pediatric surgical sample. Pediatric pain occurs in a specific social context in which parents are influential agents. Children's beliefs and cognitions about pain are shaped to some extent through observational learning of parental reactions to and coping with pain (Craig, 1986; Goodman & McGrath, 2003; Goubert, Vlaeyen, Crombez, & Craig, 2011). Parent and child interactions within the context of the child's pain also play a role in shaping children's pain-related cognitions and behavior. The interpersonal fear avoidance model of pain, a pediatric adaptation of the original model, describes the development of chronic pain within an interpersonal context (Goubert & Simons, 2014). The model describes how children express pain in an overt way, with pain-related cognitions such as pain catastrophizing contributing to more intense overt displays. Parents then

decode the child's overt display of pain leading to their own emotional and behavioral responses which are influenced by their own pain-related beliefs and cognitions. Specific parental emotional responses, such as catastrophizing, have been found to be related to parent behavioral responses to their child's pain, such as protectiveness, which in turn leads to maladaptive child coping responses and increased pain-related cognitions (Birnie, Chambers, Chorney, Fernandez, & McGrath, 2016; Guite, McCue, Sherker, Sherry, & Rose, 2011; Wilson, Moss, Palermo, & Fales, 2014). The body of work used to build and subsequently support the interpersonal fear avoidance model of pain highlights the importance of the social context in which children experience pain. This research has predominantly been conducted with children and adolescents who have established chronic pain; thus, less is known about how parent factors may influence a child's pain experience in an acute stage of pain and functioning such as that experienced after major surgery. Nevertheless, there is ample evidence to suggest that factors outside the individual, such as parent and peer behavior and cognitions, that may influence different stages of the postsurgical recovery process are important for understanding pediatric postsurgical recovery.

A final limitation of Katz and Seltzer's model and of the fear-avoidance models of chronic pain discussed thus far, is that they almost exclusively focus on risk factors related to pain and functioning. Longitudinal studies and epidemiological studies have shown that the majority of surgical patients in both adult and pediatric samples fare well, while only a subset tends to experience persistent pain and disability (see Connelly et al., 2014; Fortier, Chou, Maurer, & Kain, 2011; Katz & Seltzer, 2009b; Kehlet et al., 2006; Pagé, Stinson, Campbell, Isaac, & Katz, 2013). In an effort to identify which patients are going to develop pain and disability research has focused on identifying factors that put them at risk so that the risk can be

mitigated. However, this work has neglected factors that protect most individuals from developing chronic pain and disability that could also be used to mitigate risk. While Katz and Seltzer (2009) include 'protective factors' in the title of their paper, discussion of protection factors in their model is mainly confined to the use of multimodal preventative analgesia for the acute stage of pain. Potentially protective psychosocial factors are not reviewed or included in the model. An updated version of Katz and Seltzer's model included in a recent paper includes some potentially protective psychosocial variables (e.g., optimism) but the paper does not review the evidence for their inclusion (Katz et al., 2015). Similarly, the predominant pediatric chronic pain models are also risk focused and do not include potentially protective cognitions, personality traits or behaviors that may help to interrupt the cycle of pain, fear and disability.

## 1.4 Resilience– A New Perspective for Postsurgical Pain and Functioning

### 1.4.1 Defining Resilience

Themes of resilience emerged in the field of child development research in the 1970's when it was observed that some children developed quite well and sustained positive functioning despite being exposed to many risks such as abuse and low socioeconomic status (see Masten & Cicchetti, 2016; Nichols, 2013; Walsh, 2016). As researchers began to study the processes that could account for these good outcomes, their findings started to shift many assumptions away from a deficit-based model of development to a recognition of the robustness of human adaptation systems.

There is considerable variability in how resilience has been conceptualized and operationalized within academic literature (Kaplan, 1999; Luthar, 2006; Masten, 2007). In order to clarify the concept of resilience and develop an operational definition that could be meaningful across disciplines, Windle (2011) used a concept analysis and a systematic review to

examine 271 studies across various disciplines that investigated resilience. The paper identified three necessary requirements for resilience: 1) that the individual has experienced a significant source of adversity or risk, 2) there are protective factors present to offset the negative effects of the risk, 3) there is evidence of positive adaptation or the avoidance of a negative outcome. With these requirements in mind, resilience can be defined as, effectively negotiating, adapting to, or managing significant sources of stress or trauma by utilizing protective factors within the individual or their environment (Masten, 2001; Masten, 2018; Windle, 2011). This definition of resilience acknowledges the contribution of factors outside the individual (e.g., safe neighborhoods, access to high quality education, change in SES) that likely play a large role in an individual's ability to be resilient.

### 1.4.2 Studying Resilience

One way that researchers have approached the study of resilience is through what is referred to as a variable-focused approach (Masten, 2014, 2018; Windle, 2011). Variable-focused approaches use multivariate statistics to examine the relations between risk factors, protective factors and outcomes. Within variable-focused approaches there are different models to explain how protective factors may alter the effects of risk factors on outcomes. These models have been described with different terminology in various publications but for the purposes of this dissertation they will be described using the statistical analysis terms that correspond to the model. The first type of variable-focused approach is a main effects model that reflects the independent contribution of risks and protective factors to the outcome. Protective factors in these models can be beneficial at both high and low levels of risk. For example, family support (protective factor) has been shown to be beneficial for reducing the odds of non-suicidal self injury and suicidality (outcomes) in both non-LGBTQ youth as well as in LGBTQ youth (risk

factor for NSSI and suicidality) (Reisner, Biello, Perry, Gamarel, & Mimiaga, 2014). Mediator models are another type of variable-focused approach to studying resilience. These models describe how risk or protective factors may indirectly impact an outcome. For example, the impact of having a child with a disability (risk factor) on a parent's quality of life (outcome) was found to be fully mediated by the quantity of the parent's social support (risk/protective factor gradient) (Migerode, Maes, Busse, & Brondeel, 2012). Finally, moderator models are another type of variable-focused approach to studying resilience. These models describe how protective factors may buffer or lessen the full effects of a risk factor on an outcome. Individual differences in personality are often examined using moderator models in studies of resilience. For example, one study of middle school students reported a negative association between peer victimization (risk factor) and academic performance (outcome) which was moderated by self-esteem and self-efficacy (protective factors) such that higher levels of these protective factors were associated with better academic performance even in the face of peer victimization (Raskauskas, Rubiano, Offen, & Kathleen, 2015).

One aspect of resilience that is not explicitly addressed by the approaches described above concerns adaptation. By definition, resilience involves adaptation to stress or trauma and the nature of adaptation implies flexibility, such that different approaches must be taken for different problems that arise in different contexts. For example, a study comparing the coping strategies used by individuals with either acute or chronic illnesses found that individuals with chronic illnesses were more likely to use a combination of coping strategies as compared to individuals with acute illnesses (Endler, Kocovski, & Macrodimitris, 2001). The types of coping strategies used by individuals with the chronic illnesses likely evolved over time and the authors of this study concluded that using a combination of strategies was likely adaptive for individuals

with chronic illness as they had to make adjustments to various aspects of their life in order to manage their illness. The importance of considering time and context when examining the relation between protective factors and resilient outcomes is also illustrated by Waugh's (2014) temporal-functional model for understanding the role of positive and negative emotions in response to a stressor. In his model, Waugh describes how negative and positive emotions can co-occur but that they serve different functions at different times during a stressor (anticipatory versus recovery from stressor) and their relations with resilient outcomes are dependent on whether the stressor is controllable or uncontrollable. In summary, adaptation, which is central to the definition of resilience, implies that protective factors may serve different functions at different times. Consequently, any study of protective factors as they relate to resilient outcomes should consider the specific context or phase of the stressor in which they occur.

### 1.4.3 Resilience and Pain

Sturgeon and Zautra (2013) developed a risk-resilience model for adults with chronic pain. Within this model, the authors describe three aspects of resilience: an outcome of adaptation efforts, differential resilience in response to different stressors and a set of resources that influence an individual's response to stressors. The authors further break down resilience outcomes in terms of recovery, sustainability and growth. Recovery is defined as the extent to which the individual regains equilibrium after a stressful event. Sustainability is defined as the extent to which an individual can persevere towards their goals in the face of a stressor. Growth is defined as new learning or a better understanding of one's capacity in the face of a stressor and following one's coping efforts. The crux of Sturgeon and Zautra's model uses a variable-focused approach to propose relations between risk and protective factors and resilient outcomes. Protective factors are classified as either, resources or resilience mechanisms. Resources are

defined as stable individual trait characteristics (e.g., optimism) or environmental characteristics (e.g., supportive family) that promote effective adaptation to stressors. Resources are proposed to function through resilience mechanisms defined as modifiable processes (e.g., cognition, affect, behaviors) that can facilitate effective coping and promote positive resilience outcomes.

Sturgeon and Zautra's (2013) model was adapted for pediatric pain by Cousins and colleagues (Cousins, Kalapurakkel, Cohen, & Simons, 2014). Cousins' (2015) model maintained the same model pathways (i.e., resources and mechanisms) but adapted the model for a pediatric population by integrating variables empirically supported by the pediatric pain literature as well as providing an ecological context for the model based upon part of Bronfenbrenner's System's Theory (Bronfenbrenner, 1979). Bronfenbrenner's System's Theory was developed to explain how children's inherent qualities and their environment interact to influence their growth and development. The theory describes several ecosystems that begin at the level of the individual and broaden out to family and peers, community, school, social services and then to broader culture and sociohistorical time context. According to the theory, each of these ecological systems interact with and influence each other in the child's life.

### 1.4.4 Studying Resilience in Pediatric Postsurgical Recovery

The risk-resilience models of chronic pain described above provide a useful starting point to begin examining pediatric postsurgical recovery from a resilience perspective. However, given the novelty of this perspective in postsurgical recovery an important first step is to begin identifying and examining protective factors that may lead to resilient outcomes in pediatric postsurgical recovery. Protective factors identified by the chronic pain risk-resilience models provide a starting point, particularly for the chronic postsurgical period, but they provide less insight into the role of protective factors for acute postsurgical outcomes. Katz and Seltzer's

model of postsurgical pain development highlights the importance of considering the impact of psychosocial factors at different time points in the perioperative period and their relation to postsurgical outcomes. Thus, it is important that the relation between protective factors and resilient outcomes be examined not only in the chronic postsurgical period but also the acute postsurgical period.

### 1.5 Introduction to Dissertation Papers

The current dissertation sought to extend research findings on postsurgical pain and functioning following major pediatric surgery and examine recovery from a resilience perspective. The overall objectives of this dissertation were to: 1) Examine postsurgical pain trajectories and functioning following major pediatric surgery, 2) Review protective factors relevant to pediatric postsurgical recovery, 3) Examine one protective factor for pediatric postsurgical recovery in greater depth. To accomplish these objectives, three separate manuscript-style papers were presented as part of this dissertation. Paper one (Chapter two) addresses objective one by examining pain trajectories and functional outcomes in first year following major pediatric surgery. Paper two (Chapter three) addresses objective two in the form of a topical review. Paper three (Chapter four) addresses objective three by examining optimism as a protective factor for postsurgical functioning.

This dissertation utilizes data from a multi-centre longitudinal study. Children and their parents who took part in the longitudinal study completed a series of demographics and self-report measures regarding their pain and psychosocial factors prior to undergoing major spinal surgery for scoliosis. Children and their parents were then followed for up to one year and during that time completed additional measures regarding their pain and psychosocial factors while in hospital, during their first week home, and then at four to six weeks, three months, six

months and 12 months after their surgery. Results of children's satisfaction with pain management are reported elsewhere (Khadra et al., 2017) as are a subset of children's memories of their postsurgical pain (Noel, Rabbitts, Fales, Chorney, & Palermo, 2017), and actor-partner effects between parent and child pain-castrophizing from baseline to the first six weeks post-surgery (Birnie, Chorney, & El-Hawary, 2017). The findings presented as part of this dissertation are unique in that they describe the entire course of postsurgical pain trajectories over the postsurgical year and how they relate to functional outcomes. The findings presented as part of this dissertation are also the first to examine the role of a protective factor, optimism, on functional outcomes at different points in the postsurgical period.

The first paper (Chapter 2) describes trajectories of pain over the first postsurgical year after major spinal surgery using assessments of pain completed prior to surgery as well as at six points after surgery. Latent class growth mixture modeling was used to estimate the number of trajectories that best fit the data. Baseline parent and child factors were examined as predictors of trajectory membership and 12 month functional outcomes were compared across trajectories. It was hypothesized that the majority of the sample would fall into a trajectory characterized by significant pain in the immediate postsurgical period that would then decline over the first several weeks and months until near resolution by one year after surgery while a subset of the sample would fall into a trajectory characterized by pain that is significant and persisting throughout the postsurgical year. It was also hypothesized that baseline pain, younger age, child and parent baseline anxiety and pain catastrophizing would be predictive of membership in the persistent pain trajectory and that membership in this trajectory would be associated with worse functional outcomes at one year as compared to membership in other trajectories.

The second paper (Chapter 3), is a topical review of protective factors relevant to a pediatric postsurgical population. The paper reviews the rationale for identifying and examining the role of protective factors in a pediatric postsurgical population. Potentially relevant protective factors from the pediatric chronic pain, and adult postsurgical pain literature are reviewed and recommendations for future research directions are provided.

The third paper (Chapter 4) examines the role of one protective factor, optimism, in the relation between pain and functional outcomes (i.e., functional disability, quality of life) during two time points in the postsurgical period. It was hypothesized that optimism would moderate the relation between pain and functional outcomes during the chronic postsurgical period (12 months) such that the relation between pain and functional outcomes would be more favorable for adolescents with higher levels of optimism. The same moderation was performed during the acute postsurgical period (four to six weeks) but was treated as an exploratory analysis as scant empirical findings are available to support a specific hypothesis.

Following the presentation of each paper, a discussion of the overall results is provided (Chapter 5), including theoretical and clinical implications, strengths and limitations, and future research directions derived from this dissertation.

# CHAPTER 2. PAIN AND FUNCTIONAL OUTCOMES FOLLOWING MAJOR SCOLIOSIS SURGERY IN ADOLESCENTS: A LATENT GROWTH MIXTURE MODELLING ANALYSIS

The paper prepared for this empirical study is presented below. The data used for this study was derived from the Post-Operative Recovery following Spinal Correction – Home Experiences (PORSCHE) dataset which was a multi-centre, longitudinal cohort study that collected data from 2010 to 2015. Data was collected by numerous research assistants and nurses at eight different research sites. Kristen Bailey, under the supervision of Dr. Jill Chorney, contributed to the development of the research questions and hypotheses and planned the analytic approach. Kristen consulted with her dissertation committee (Drs. Simon Sherry and Allen Finley), a statistical consultant (Dr. Sean MacKinnon) and pediatric orthopedic surgeons (Drs. Jason Howard and Ron El-Hawary) during the conceptualization and analytic approach phase and the their feedback was incorporated into the study. Kristen was responsible for preparing the dataset for analysis, conducting the analysis, and writing the current manuscript. The manuscript was reviewed by all of the above-mentioned individuals and feedback was incorporated accordingly. Kristen applied for and was successful in obtaining funding from the Maritime Strategy for Patient Oriented Research Support Unit to support this research.

## 2.1 Abstract

Significant postsurgical pain is reported in upwards of 20% of children after major surgery. However, few studies have examined changes in children's pain over time. This study describes children's postsurgical pain trajectories using repeated assessments over 12 months, examines psychosocial predictors and compares functional outcomes across trajectories. A cohort of two hundred and twenty children aged 10 to 18 years who underwent surgery for idiopathic scoliosis were included. Participants completed assessments prior to surgery and at six postsurgical time points. Latent class growth mixture modeling was used to produce a three-trajectory model of the data. Nearly half the sample followed a, 'moderate-severe pain with good resolution' trajectory. More than a third followed a 'mild-moderate pain with good resolution' trajectory, and less than 15% followed a, 'moderate-severe pain with incomplete resolution' trajectory. Membership in the 'Moderate-severe pain with incomplete resolution' trajectory was predicted by greater baseline anxiety. Individuals in this trajectory also had the poorest 12-month functional outcomes. Our findings suggest that most individuals with moderate immediate postoperative pain can experience good recovery. A small subset of children demonstrated incomplete recovery and lower functioning which may be predicted by greater baseline anxiety; the clinical implications of these findings should be evaluated.

## 2.2 Introduction

Approximately 20% of children who have major surgery report significant postsurgical pain long after tissue healing should be complete (Connelly et al., 2014; Fortier et al., 2011; Pagé, Stinson, et al., 2013). Use of validated tools to measure the patient's postsurgical pain has increased, but it is uncommon for pain to be serially assessed after the patient leaves the hospital. There is growing recognition that measuring pain at a single time point, or using averages of pain scores over time, limits our understanding of the natural course (i.e. trajectory) of an individual's postsurgical pain. As such, recent studies have measured pain at multiple time points and used trajectory modeling to describe the change in pain. When used in groups of individuals with the same diagnosis or surgery, trajectory modeling can aid in classifying individuals into different trajectories of pain resolution (or non-resolution)(Chapman et al., 2011).

Sieberg and colleagues (Sieberg et al., 2013) identified five pain trajectories among a group of 260 adolescents who underwent surgery for idiopathic scoliosis; one trajectory (5.8% of the sample) was characterized by persistent pain. This study provides valuable information regarding long-term pain outcomes for major pediatric surgery, but the authors only assessed pain scores 1-3 weeks before surgery and 1, 2 and 5 years after surgery. Given that the transition from acute to chronic postsurgical pain is understood to occur between two and three months after surgery (Macrae & Davies, 1999), more frequent measurement is needed. This limitation was addressed in part by Connelly and colleagues (Connelly et al., 2014) who sought to describe a typical pain trajectory for children ($N = 50$) in the first six months after spinal fusion by measuring pain at baseline and five time points after surgery. Results indicated that pain levels declined in the days after surgery but that 22% of the sample exhibited pain that was the same as or above baseline levels for six months after surgery. Rabbitts and colleagues (Rabbitts et al.,

2015) also measured postsurgical pain at several time points over the first postsurgical year in 60 children who underwent major spine or chest wall surgery. Rabbitts identified two trajectories. One trajectory (18% of the sample), described as "late recovery", demonstrated slower postsurgical pain resolution. Although this study was a valuable contribution to the literature, it did not comment on the role that baseline pain may have played in the trajectories, and was limited by a small sample size. A more recent study with a larger sample ($N = 265$) and several measurements of postsurgical pain, also identified two trajectories but reported that nearly half of their sample ($N = 123$) followed a trajectory characterized by moderate to severe baseline pain that remained throughout the postsurgical year (Rosenbloom et al., 2019). This study also utilized a mixed sample of orthopedic surgical procedures which makes it difficult to generalize to a purely scoliosis surgical sample.

It is also important to examine potentially modifiable baseline risk factors that could be incorporated into a presurgical intervention. Connelly and colleagues (Connelly et al., 2014) found that greater baseline pain and anxiety predicted slower improvement in pain. Rabbits and colleagues (Rabbitts et al., 2015) found that greater parent but not child baseline catastrophizing, was predictive of membership in the 'late recovery' pain trajectory. They attributed the lack of child findings to their small sample size and suggested that future studies with a larger sample may be more likely to find an association between child catastrophizing and postsurgical pain trajectories. Sieberg and colleagues (Sieberg et al., 2013) did not examine parent factors, but found that greater baseline child anxiety was associated with slower daily rate of pain improvement whereas greater baseline pain coping efficacy was associated with faster daily rate of pain improvement. Rosenbloom and colleagues (Rosenbloom et al., 2019) found that baseline functional disability predicted membership in their persistent pain trajectory. Findings from the

available literature suggest that both parent and child baseline factors may be important predictors of postoperative pain trajectories.

Despite the potential impact of pain on function, few studies have examined functional outcomes associated with prolonged postsurgical pain in children. Rabbitts and colleagues (Rabbitts et al., 2015) found that membership in the "late recovery" trajectory was associated with greater activity limitations 12 months after surgery. Rosenbloom and colleagues (Rosenbloom et al., 2019) found that pain unpleasantness trajectories predicted greater functional disability one year after surgery. In adults, postsurgical pain has been associated with both short term and long term declines in quality of life (Caffo et al., 2003; Wu et al., 2003), greater impairment in activities of daily living (Rosseland, Solheim, & Stubhaug, 2008) and greater pain-related disability (Staal, Nienhuijs, Keemers-Gels, Rosman, & Strobbe, 2008).

## 2.3 Objectives

Though they have merit, the available studies examining postsurgical pain trajectories for children undergoing major surgery are limited by relatively small sample sizes, infrequent measurements in the postsurgical period or mixed surgical samples. To address these limitations, this study has four aims: [1] to identify pain trajectories of a sizeable sample of adolescents following major scoliosis surgery, [2] to examine differences in baseline pain across identified trajectories, [3] to examine parent and child predictors of trajectory membership and [4] to compare the functional outcomes across identified trajectories.

## 2.4 Methods

### 2.4.1 Participants

Eligible participants included children and adolescents aged 10 to 20 years old who were scheduled to undergo posterior spinal fusion and instrumentation for adolescent idiopathic

scoliosis. The indication for surgery was a progressive scoliosis greater than 40 to 45 degrees in skeletally immature patients, and greater than 50 to 55 degrees in skeletally mature patients. Exclusion criteria were children who did not speak English, children with diagnosed developmental delay that would interfere with completing study measures, or children with major chronic medical conditions (ASA status III or higher).

The data presented in this manuscript is part of the *Post-Operative Recovery following Spinal Correction: Home Experience (PORSCHE)* study, a larger project examining prevalence, predictors, and consequences of children's pain following scoliosis surgery. Participants were recruited at eight children's hospitals across Canada, including the IWK Health Centre (Halifax, Nova Scotia), Saint John Regional Hospital (Saint John, New Brunswick), Montreal Children's Hospital (Montreal, Quebec), CHU Sainte-Justine (Montreal, Quebec), Children's Hospital of Eastern Ontario, (Ottawa, Ontario), McMaster Children's Hospital (Hamilton, Ontario), Stollery Children's Hospital (Edmonton, Alberta), and Alberta Children's Hospital (Calgary, Alberta). Results of children's satisfaction with pain management are reported elsewhere (Khadra et al., 2017) as are a subset of children's memories of their postsurgical pain (Noel et al., 2017), and actor-partner effects between parent and child pain-catastrophizing from baseline to the first six weeks post-surgery (Birnie, Chorney, El-hawary, & Group, 2017).

### 2.4.2 Child Measures

***Demographics***. Baseline demographics were collected that included sex, parents' education, age, length of hospital stay, and history of previous surgeries, hospitalizations and prematurity at birth.

***Numerical Rating Scale of Pain***. The child was verbally asked to rate their worst and average pain over the past 24 hours and current pain on a numerical rating scale (NRS) of 1 to

10. Average pain ratings were used for this manuscript. Average pain was assessed at seven time points: baseline (up to three weeks prior to the day of surgery), in hospital (averaged over all inpatient days), during the first week at home (averaged over 1 to 3 assessments in the first week), four to six weeks, three months, six months and 12 months after surgery. There are few studies determining cut-offs for pain scores on the NRS in pediatric populations, consequently, for the purposes of labeling in this paper, adult cutoffs will be used. Pain ratings from zero to three are considered, 'mild pain', ratings of four to six are considered, 'moderate pain', and ratings of seven or higher are considered, 'severe pain' (Boonstra et al., 2016; Gerbershagen, Rothaug, Kalkman, & Meissner, 2011). The NRS is considered as adequate, reliable and valid a measure of pain in children greater than eight years old as compared to other pain measures used in pediatric populations (i.e., Faces Pain Scale Revised) (von Baeyer et al., 2009).

***State-Trait Anxiety Inventory – Child Form*** (STAIC) (Spielberger & Edwards, 1973b). This measure was administered at baseline, and at four to six weeks, three, six and 12 months postoperatively. This study uses only the baseline outcomes. This questionnaire is designed to measure both the state and trait level of anxiety of the child. Children are presented with statements of how people describe themselves and are asked to evaluate on a scale of 1 to 3, if they agree with the statement at the current moment as well as in general. Some items are reverse scored so that a greater score indicates greater anxiety with scores ranging from 20 to 60 for both state and trait anxiety.

***Pain Catastrophizing Scale – Child Form*** (PCS-C) (Crombez et al., 2003). This questionnaire was administered at baseline, and at four to six weeks, three, six and 12 months. Only the baseline outcomes were used in this study. This questionnaire is designed to measure children's thoughts and feelings in response to pain. Children rate their anxiety responses to pain

on a 5-point, 13 item, Likert-type scale ranging from, “Not at all” to “Extremely”. Items are summed to yield total scores ranging from 0 to 52 with higher scores indicating a greater amount of catastrophizing in response to pain.

***Pediatric Quality of Life Inventory – version 4*** (PedsQL-4) (Varni, Seid, & Rode, 1999). This questionnaire was administered at baseline, and at four to six weeks, three, six and 12 months postoperatively. This study uses only the 12-month outcomes. This questionnaire is designed to measure the child’s general quality of life in four categories: physical, emotional, social and school. Children are required to indicate on a scale of 0 to 4, the degree of problems the child experiences with each item. Items are then reverse scored and linearly transformed to a 0-100 scale such that higher scores indicate greater quality of life. Age appropriate versions were used (validated versions are for ages 8-12 and 13-18).

***Functional Disability Inventory (FDI)*** (Walker & Greene, 1991). This questionnaire was administered at four to six weeks, and three, six and 12 months postoperatively. This study uses only the 12-month outcomes. The FDI is a 15-item scale that assesses the extent to which children experience difficulties in completing specific tasks (e.g., walking to the bathroom, being at school all day). Items on the FDI are rated on a 5-point Likert scale with total scores ranging from 0 to 60 with higher scores indicating greater functional disability. The FDI has been used with a variety of pediatric populations including children with chronic pain (Kashikar-Zuck, Vaught, Goldschneider, Graham, & Miller, 2002; Lynch, Kashikar-Zuck, Goldschneider, & Jones, 2006; Reid, McGrath, & Lang, 2005) and postsurgical pain (Gidron, McGrath, 1995).

***Scoliosis Research Society Health-Related Quality of Life Tool (SRS-30)***. This questionnaire was administered at baseline, and four to six weeks, three, six, and 12 months postoperatively. This study uses only the 12-month outcomes. The SRS-30 is comprised of 30

items that measures health-related quality of life (*HRQOL*) specific to scoliosis. It is organized into domains (pain, self-image/appearance, function/activity, mental health and satisfaction with disorder management) and also provides a total score. The score for each domain ranges from one to five with higher scores indicating a better outcome. This study reports the total score. This measure has been extensively validated and has become a widely accepted measure to study *HRQOL* in populations with scoliosis (Adobor, Rimeslåtten, Keller, & Brox, 2010; Asher, Min Lai, Burton, & Manna, 2003; Bastrom, Marks, Yaszay, & Newton, 2013; Bridwell et al., 2007; Carreon et al., 2013; Parent et al., 2010; Verma et al., 2010).

### 2.4.3 Parent Measures

***State-Trait Anxiety Inventory – Parent Form*** *(*STAIP) (Spielberger, Gorsuch, Lushene, Vagg, & Jacobs, 1970). This questionnaire was administered at baseline, and at four to six weeks, three, six and 12 months. Only the baseline outcomes are used for this study. This questionnaire is designed to measure both the state and trait level of anxiety of the parent. Parents are presented with statements of how people describe themselves and are asked to evaluate on a scale of 1 to 4, if they agree with the statement at the current moment as well as in general. Some items are reverse scored so that a greater score indicates greater anxiety with scores ranging from 20 to 80 for both state and trait anxiety.

***Pain Catastrophizing Scale – Parent Form*** (PCS-P) (Goubert, Eccleston, Vervoort, Jordan, & Crombez, 2006). This questionnaire was administered at baseline and at four to six weeks, three, six and 12 months. Only the baseline outcomes were used for this study. This questionnaire is designed to measure parent's thoughts and feelings in response to their child's pain. Parent's rate their responses to their child's pain on a 5-point, 13 item, Likert-type scale ranging from, "Not at all" to "Extremely". Items are summed to yield total scores ranging from

0 to 52 with higher scores indicating a greater amount of catastrophizing in response to their child's pain.

### 2.4.5 Procedure

Participants were identified and informed of the study by their attending surgeon at the time they decided to undergo the surgical procedure or at the pre-surgical visit. Families that expressed interest in the study were provided study information by a research assistant and were given the option to either consent in person or to take the materials home to review and then consent later via phone prior to the surgery. Depending on the method of consent, families received baseline questionnaires either in person or via mail and all questionnaires were returned via mail. Follow-up questionnaires were either mailed to families or completed at clinic visits that occurred at four to six weeks, three months, six months and 12 months after surgery. Data in the first week after participants were discharged from the hospital were collected via phone. This study was approved by the respective Research Ethics Boards at all study sites

### 2.4.6 Data Analysis

To identify trajectories of postsurgical pain, fairly recent advances in growth mixture modeling (GMM) called latent class growth analysis and latent growth mixture modeling (LGMM; (Jung & Wickrama, 2008; Muthén & Muthén, 2000)) were applied to the data. These statistical techniques are used to identify multiple homogeneous subpopulations in a heterogenous sample in order to determine meaningful groups or classes of individual variation over time called trajectories (Jung & Wickrama, 2008; Muthen, 2004). Because LGMM relaxes the assumption of a single homogenous distribution, they can tailor growth parameters (e.g., intercept, slope) and other model specifications to fit the data with precision. This method of modeling allows trajectories to be identified more empirically based on the data rather than

through *a priori* conceptual models. LGMM is also ideally suited to longitudinal data as it is robust to the effects of missing data. In this study, LGMM using Mplus v.8 software was employed to identify discrete growth trajectories.

LGMM modeling involves a number of iterative steps; the final model is determined by a variety of fit indices as well as by interpretability and conceptual rationale (Jung & Wickrama, 2008). The first step involves identifying a univariate single-class growth model without any covariates. Next, more complex models with an increasing number of unconditional trajectories (i.e., trajectories without covariates) are then tested and compared for fit against previously tested models with fewer trajectories. After settling on a final number of latent classes, the model is then tested with covariates (i.e., conditional model) to further enhance model fit.

Consistent with recommendations for model testing, this study compared models with one to four trajectories for NRS ratings over time. Data were first examined using line plots grouped by participant ID and visually examined to determine possible relations between the data (i.e., linear, cubic, quadratic) that could be applied in the univariate growth curve (see Figure 2.1). Visual inspection of the data did not reveal any naturally occurring relations among the data. Ordinarily, past research can be used to *a priori* determine relations among data that can be applied in a univariate growth curve. However, due to the sparsity of previous studies examining pediatric postsurgical pain following major scoliosis (or other) surgery, it was decided that a non-linear model would be appropriate for these data. The initial NRS data point (i.e., in-hospital NRS) was coded as '0' and the final NRS data point (i.e., 12 month NRS) was coded '1' and all other data were allowed to vary freely so as not to be constrained into any specific relation. This allowed for the modelling of non-linear patterns of change over time.

There are several goodness of fit statistics to consider when examining and deciding upon the best fitting model for the data. One must use measures of classification certainty/accuracy (i.e., a measure of confidence that the correct individuals were classified into the correct trajectory) and measures of overall fit. A final consideration is the size of the smallest class. If an additional class contains a small

number (e.g., <1% or $n$ <25), then the researcher must be able to justify the additional class as it likely has low power and precision relative to the other, larger classes (Lubke & Neale, 2006). This study utilized the Bayesian Information Criterion (BIC), adjusted BIC and Akaike Information Criterion (AIC) as well as entropy values to assess classification accuracy. Overall fit was assessed with the Lo-Mendell-Rubin likelihood ratio test (LMRT) and Bootstrapped Lo-Mendell-Rubin Test (BLRT). Given the number of parameters to consider when determining best model fit, it can be difficult to come to a final conclusion if there is a great deal of variation between indices for different class solutions. In cases such as these, simulation studies have demonstrated that a significant BLRT in combination with a low BIC or low adjusted BIC are the most robust indicators of a good fitting model (Nylund, Asparouhov, & Muthén, 2007).

Regarding sample size calculations, simulation research has suggested reliable results in trajectory estimation can be expected if sample size is greater than 50 (Tolvanen, 2007). Additionally, when choosing goodness of fit statistics, the BIC is most useful in smaller sample sizes like the present study ($n$ <500) while the BLRT test is useful in both small and larger sample sizes.

Baseline pain ratings were not included in the univariate growth curve nor the unconditional model due to variability in the baseline data collection time points. However, given the potential importance of baseline pain on postsurgical pain, SPSS version 24 was used

to conduct a one-way Analysis of Variance (ANOVA) in order to examine baseline pain scores across trajectory group from the unconditional model.

To identify risk factors for a persistent pain trajectory, a multinomial logistic regression analysis was conducted examining baseline psychosocial factors (child and parent pain catastrophizing, child anxiety), controlling for age and sex. Internal consistency for the risk factors examined in this study ranged from (0.86 – 0.91) which is considered to be good to excellent. In order to examine functional outcomes associated with each trajectory, a one-way Analysis of Variance (ANOVA) was conducted in order to examine the relation between trajectory membership (independent variable) and participants' scores on the three functional outcomes (i.e., FDI, PedsQL & SRS-30; dependent variables). Internal consistency on the functional outcomes for this study ranged from 0.80-0.90 which is considered good to excellent. Post-hoc tests were then used to compare each trajectory on the functional outcome scores measured at 12 months and determine trajectory differences. SPSS version 25 was used to perform both of these analyses.

## 2.5 Results

### 2.5.1 Participants

Across sites, 267 potential participants were approached by a research assistant and 246 provided consent. The final sample included 220 adolescents who completed at least some part of the baseline (T0) assessment (see Figure 2.2). The retention rates for each time point are as follows: First week home, 89%; 4-6 week follow-up, 89%; 3 month follow-up, 67%; 6 month follow-up, 70%; and 12 month follow-up, 63%. See Table 1 for demographic information including child age, child sex, baseline time point, length of postsurgical hospital stay, self-reported average baseline pain, history of previous surgery, hospitalization, chronic illness and/or

premature birth. In order to examine differences between participants who completed versus those who did not, a series of independent *t* tests and $\chi^2$ tests were conducted. Completers and non-completers were compared on child age, sex, length of stay in the hospital, parent years of school, baseline pain, immediate postoperative pain, as well as history of previous surgery, previous hospitalization, chronic illness, prematurity, whether the child had a preadmission visit, and whether the child or parent had experienced a major life change in the last six months. Completers vs. non-completers did not differ on any of the analyzed variables except for length of stay in hospital. The average hospital stay for the completers was 5.6 days (*S.D.* = 1.3) which was lower than non-completers average stay of 6.2 days (*S.D.* = 1.6).

### 2.5.2 Descriptives, Correlations and Possible Covariates

See Table 2.1 for descriptives and correlations between baseline variables and NRS ratings of child average pain at each time point. With regards to potential covariates, child sex was correlated with both in-hospital postsurgical NRS ($r$=0.21, $p$=.002) and first week at home postsurgical NRS ($r$=0.18, $p$=.01). Parent years of education was correlated with in-hospital postsurgical NRS ($r$=-0.15, $p$=.03) and 3 month postsurgical NRS ($r$=0.18, $p$=.04). History of previous hospitalization was correlated with baseline NRS ($r$=0.14, $p$=.05), 4-6 weeks postsurgical NRS (r=0.17, $p$=.02) and 3 month postsurgical NRS ($r$=0.20, $p$=.02). Prematurity was correlated with 3 month postsurgical NRS (r=0.17, p=.04). Lastly, baseline NRS was positively correlated with each of the postsurgical NRS variables. Given these relations, child sex, parent years of education, history of hospitalization, prematurity and baseline pain were all examined as potential covariates first in the univariate growth model of the NRS data.

### 2.5.3 Univariate Growth Curve

The univariate growth curve for NRS was a fit to the data, [$Chi^2$ ($N$ = 220) = 22.5, $p$ = .05; $Chi^2/df$ = 13; CFI = .95; TLI = .94; RMSEA = .06 (90% CI: .005, .097)]. Figure 2.3 shows a plot of the mean NRS scores at each time point for the univariate model. The data demonstrated a non-linear pattern of change over time which can be seen in Figure 2.3 as well as by the slopes and standard errors for each NRS time point: in-hospital (slope = 0, *SE* = 0), 1st week at home (slope = .395, *SE*=.024), 4-6 weeks (slope = .874, *SE* = .026), 3 months (slope = .972, *SE* = .027), 6 months (slope = .988, *SE* = .025) & 12 months (slope = 1, *SE* = 0). Child age, baseline time, length of hospital stay, history of previous surgery, history of previous hospitalization and history of prematurity did not emerge as having a significant slope and/or intercept when added to the univariate model. However, baseline pain, child sex and parent years of education emerged as having a significant slope and/or intercept. The contribution of parent years of education to the univariate growth curve was quite small and – in light of the fact that the majority of parents in this sample had at least some post-secondary education – it was decided that parent years of education would not be considered as a possible covariate in the LGMM model. However, child sex and baseline pain were retained as potential covariates to consider in the LGMM model.

### 2.5.4 Conditional Model

As shown in Table 2.2, each of the information criterion indices (AIC, $BIC_a$) demonstrated lower values (improved fit) for each additional class from the two-trajectory to the four-trajectory solutions. Additionally, the LMRT demonstrated significantly lower values with each successive model from the two-trajectory to the four-trajectory solutions. There was some variability in entropy values and the latent class probabilities worsened slightly with each successive model. The BLRT, which simulations have demonstrated to be the most robust

indicator (Nylund et al., 2007), consistently improved fit up to the four-trajectory solution. However, when the trajectory counts and proportions were examined for each trajectory model and discussed for clinical relevance with the pediatric spine surgeons involved in the study (JH, RE) – it was decided that the three-trajectory model solution was the best fit for the data based upon fit indices, clinical relevance and conceptual rationale. As can be seen in Figure 2.4a, the three-trajectory solution identified three distinct trajectories of NRS ratings across the first postsurgical year. The most common trajectory, capturing 50.9% of the sample, describes individuals with a mean NRS rating of 5.8 (*S.D.* = 1.5) in-hospital that sharply declines over the next four to six weeks to a mean NRS rating of less than two and further declines and stabilizes until the 12-month postsurgical time point. This trajectory will hence be referred to as 'moderate-severe pain with good resolution'. Another trajectory (35.9% of the sample) describes individuals with a mean NRS rating 4.25 (*S.D.* = 1.5) while in-hospital that sharply declines over the next four to six weeks to a mean NRS rating less than one and remains stable and low until the 12-month postsurgical time point. This trajectory will hence be referred to as, 'mild-moderate pain with good resolution'. The third trajectory (13.2% of the sample) describes individuals who report a mean NRS rating of 6.0 (*S.D.* = 1.6) in hospital that then sharply declines over the next four to six weeks to a mean NRS rating of 2.4 (*S.D.* = 1.8) that then stabilizes and remains at a rating above two until the 12-month postsurgical period. This trajectory will hence be referred to as, 'moderate-severe pain with incomplete resolution'.

### 2.5.5 Baseline Pain

Results of the one-way ANOVA showed a significant difference in baseline pain scores between the three trajectories ($F = 6.04$, $p < .05$, $d = .88$). Participants in the moderate-severe pain with incomplete resolution trajectory had greater baseline pain ($M = 3.8$, *S.D.* = 2.2) as

compared to participants in the 'Moderate-severe pain with good resolution' trajectory, ($M$ = 3.0, $S.D.$= 2.4) and the 'Mild-moderate pain with good resolution' trajectory, ($M$ = 2.1, $S.D.$ = 2.0). There were no statistically significant differences between the trajectories labelled 'moderate-severe pain with good resolution' and 'mild-moderate pain with good resolution'.

### 2.5.6 Conditional Model with Baseline Pain as Covariate

Based on the findings from adding covariates to the univariate growth curve model, potential covariates (sex and baseline pain) were added one at a time to the three-trajectory model and fit indices were again examined for improved fit. As compared to the 3-trajectory model without covariates, sex demonstrated improved fit via lower information criterion indices (i.e., AIC & $BIC_a$), lower LMRT and BLRT values but the latent class probabilities worsened as did the entropy value. Additionally, the logistic regression odds ratios for sex as a covariate were not significant. Due to the variability in improvement among the various fit indices for the 3-trajectory model with sex as a covariate as compared to the 3-trajectory model without covariates, it was determined that the addition of sex to the 3-trajectory model did not improve the model fit. Baseline pain was then added and fit indices were compared against fit indices for the 3-trajectory model without covariates. As can be seen in Table 2, this addition resulted in lower values (improved fit) for each of the information criterion values (i.e., AIC, $BIC_a$). Similarly, the LMRT and BLRT values were lowered and the latent class probabilities and entropy value were increased. Additionally, the logistic regression odds ratios for baseline pain as a covariate was significant and indicated that individuals with higher baseline pain were approximately 1.4 times more likely to be classified into the moderate-severe pain with incomplete resolution trajectory. Due to the improvements observed in the fit indices when compared against the 3-trajectory model without covariates, baseline pain was deemed to have

improved model fit and was retained in the final model as a covariate. While, the structure of the three-trajectory conditional model (i.e., covariate) model remained similar to the three-trajectory unconditional (i.e., no covariates) model there were some slight changes in the shape and proportions of each trajectory. The most noteworthy changes in the conditional model as can be seen in Figure 2.4b, was that the shape of the smallest trajectory changed. Like in the unconditional model it describes individuals who reported a mean NRS in-hospital in the moderate to severe range ($M$ = 6.0, $SD$ = 1.3) that then declines over the next four to six weeks, but the decline slope is shallower and the mean NRS ratings stabilize at four to six weeks and remain elevated until 12 months with a mean NRS of 3.3. Individuals in the moderate-severe pain with complete resolution trajectory continued to report a mean NRS in-hospital in the moderate range (M = 6.2, $SD$ = 1.1) but individuals in the mild-moderate pain with complete resolution trajectory reported a slightly lower mean NRS in-hospital ($M$ = 3.8, $SD$ = 1.3). In the conditional model, it is also noteworthy that the proportion of the largest trajectory, labelled moderate-severe pain with good resolution, was reduced to 48.5%, the proportion of the second largest trajectory, labelled mild-moderate pain with good resolution, was increased to 38.6% and the proportion of the smallest trajectory labelled, moderate-severe pain with incomplete resolution, was decreased to 12.9%.

### 2.5.7 Risk Factors

Results of the multinomial regression analysis predicting group membership are shown in Table 2.3. Using the, 'mild-moderate pain with good resolution', trajectory as the reference class revealed the most robust differences between the trajectories. Compared to the, 'mild-moderate pain with good resolution' trajectory, the 'moderate-severe pain with incomplete resolution' trajectory had higher child trait anxiety at baseline. When compared with the, 'mild-moderate

pain with good resolution' trajectory, the 'moderate-severe pain with good resolution' trajectory was also more likely to have females.

### 2.5.8 Functional Outcomes

Results of the one-way ANOVA are shown in Table 2.4. Participants in the moderate-severe pain with incomplete resolution trajectory had greater functional disability, worse overall quality of life, and worse scoliosis-specific quality of life, 12 months after surgery as compared to participants in the other two trajectories. There were no statistically significant differences between the trajectories labelled moderate-severe pain with good resolution and mild-moderate pain with good resolution[1].

## 2.6 Discussion

Using LGMM (Muthén & Muthén, 2000), we identified three distinct trajectories of postsurgical pain in adolescents. For one trajectory, the peak pain experienced in the immediate postsurgical period (first week after surgery) was 6.2, which we termed moderate-severe. This pain declined sharply by more than 80% over the next 4-6 weeks, and gradually reduced to, or lower than, preoperative values by 12 months. For another trajectory, the peak pain experienced in the immediate postsurgical period was mild-moderate, declining even further over the next 4-6 weeks; gradually reducing to or lower than preoperative values by 12 months. But, as expected,

[1] In order to account for the missing data observed in the 12 month functional outcome variables, multiple imputation modelling with 40 imputations using the Markov Chain Monte Carlo algorithm available in SPSS 25.0 with auxiliary variables identified using the Missing Data Analysis function was conducted. One-way ANOVAs using Bonferonni corrected p value were conducted with the imputed data to examine functional outcomes across trajectory membership. SPSS 25.0 does not provide pooled statistics for tests of between subject effects but results across all 40 imputations were consistent with what is reported in the manuscript. Statistics across analyses using the FDI imputations range from (F = 7.59 – 16.62, d = .89 – 1.0, adusted R squared = .062 - .122); PedsQL imputations ranged from (F = 7.17 – 16.04, d = .87-1.0, adjusted R squared = .06 -.13); SRS30 imputations ranged from (F = 7.93 – 15.80, d = .95 – 1.0, adjusted R squared = .07 - .13).

a subset of adolescents – with more moderate-severe immediate postsurgical pain scores – continued to experience mild to moderate persistent pain at 12 months.

These findings extend those obtained by Rabbitts and colleagues (Rabbitts et al., 2015) who observed two distinct trajectories labelled as 'early recovery' and 'late recovery'. Our findings suggest the importance of having adequate sample size and thus sufficient power to detect nuances among individuals whose pain improves within the first four to six weeks. Our study demonstrates that even individuals with moderate-severe immediate postoperative pain can experience rapid improvement over the first 4-6 weeks and little to no pain by 12 months after surgery.

Pain scores for individuals in the 'moderate-severe pain with incomplete resolution' trajectory was 3.8 at baseline and 3.3 at 12-months. This finding raises questions about whether the pain experienced in this group should be thought of as chronic postsurgical pain, or unresolved baseline pain. According to Macrae & Davies' (Macrae & Davies, 1999) definition of chronic postsurgical pain, the pain must develop after surgery and any pre-existing problems possibly causing the pain should be excluded. This definition suggests that the pain observed in this trajectory may better represent an exacerbation and continuation of pre-existing pain rather than chronic postsurgical pain caused by the surgery itself. Identifying patients with elevated baseline pain may allow for the application of mitigating factors pre- and peri-operatively – including pharmacologic and psychologic interventions – to help influence a more favourable pain trajectory.

Several adult studies have identified moderate to severe immediate postsurgical pain as a robust risk factor for chronic postsurgical pain (Callesen, Bech, & Kehlet, 1999; Gottschalk & Ochroch, 2008; Hanley et al., 2007; Hayes, Browne, Lantry, & Burstal, 2002; Katz, Jackson,

Kavanagh, & Sandler, 1996; Lau, Patil, Yuen, & Lee, 2003; Poleshuck et al., 2006; Romundstad et al., 2006; Senturk et al., 2002; Tasmuth, Kataja, Blomqvist, Von Smitten, & Kalso, 1997). However, findings from this study suggest that moderate immediate postsurgical pain may not be the best predictor of persistent postsurgical pain in this adolescent population, as nearly half of our sample demonstrated moderate immediate postsurgical pain which then declined sharply over the next four to six weeks. The shape of the two trajectories with moderate immediate postsurgical pain suggests that other risk factors may be important to consider when predicting persistent postsurgical pain. The pain course of these two trajectories also suggests that it may be important to measure risk factors beyond baseline. In particular, it may be important to examine potential risk factors (i.e., return to sports, pain catastrophizing, emotion regulation, anxiety) between 4-6 weeks and 3 months after surgery, when these trajectories begin to diverge. Katz (Katz, 2012) has suggested that different factors may contribute to the transition from acute to chronic postsurgical pain than contribute to the maintenance of chronic pain. For example, factors that relate to adolescent development (e.g., emotional regulation) or are associated with spinal deformity (e.g., concerns with body image) may be especially important to consider when determining predictors of postsurgical pain trajectories in this population. Sex and gender are likely also important factors to consider as possible predictors of postsurgical pain trajectories, as sex differences in pain appear to emerge during adolescence (King et al., 2011).

The current study also investigated parent and child baseline predictors of trajectory membership and found that child anxiety and sex were predictive of trajectory membership. Specifically, being female was predictive of membership in the 'moderate-severe pain with good resolution' trajectory and child trait anxiety in the 'moderate-severe pain with incomplete resolution' trajectory. The latter finding is consistent with previous literature (Sieberg et al.,

2013) that showed child anxiety was predictive of a slower rate of pain improvement. The present study did not find either parent or child catastrophizing to be predictive of trajectory membership. Eccleston (Eccleston, Fisher, Vervoort, & Crombez, 2012) has proposed that pain catastrophizing as is currently measured by the *Pain Catastrophizing Scale – Child version* does not incorporate social and/or developmental context of cognition nor threat appraisal. More recent research, has shown that the relationship between catastrophizing and pain intensity is moderated by age, being stronger in adults than in children/adolescents (Feinstein et al., 2017).

The current study also investigated the functional outcomes associated with each trajectory. Consistent with our expectations, the 'moderate-severe pain with incomplete resolution' trajectory reported lower *HRQOL* and more functional disability at 12 months after surgery. These results are also consistent with Rabbitts and colleagues' findings that their 'late recovery' pain trajectory was associated with greater activity limitations and lower HRQOL at 12 months. As in their study, our results also support the notion that moderate immediate postsurgical pain alone does not necessarily lead to poor functional outcomes. Rather, a pain trajectory characterized by both moderate immediate postsurgical pain and pain that persists many months later appears to be associated with poorer functional outcomes. Consequently, pain control is important both in the immediate postoperative period as well as over the longer term.

While the 'moderate-severe pain with incomplete resolution' trajectory reported worse functional outcomes as compared to the other two trajectories, their respective scores seemed more favorable overall than those for children and adolescents with chronic pain conditions (i.e., Arthritis, Fibromyalgia). For example, FDI mean scores in pediatric chronic pain samples tend to be around 23 whereas in this study, mean scores ranged from 2.8 in the 'moderate-severe pain

with good resolution' trajectory to 7.7 (*S.D.* = 7.5) in the 'moderate-severe pain with incomplete resolution' trajectory. Similarly, mean scores on the PedsQL in a study of children attending a chronic pain clinic were 51 (Cousins, Cohen, & Venable, 2014) whereas adolescents in this study reported more favorable mean scores in the 'moderate-severe pain with incomplete resolution' trajectory (72.5) and the 'mild-moderate pain with good resolution' trajectory (86.1).

### 2.6.1 Limitations & Future Research

Our findings should be considered in light of several limitations. First, this study only measured pain up to 12 months after surgery. Whereas our study demonstrated important changes in pain trajectories over the course of 12 months, studies that examined pain scores over the course of several years (Chidambaran et al., 2017; Sieberg et al., 2013) suggest that pain trajectories continue to evolve in important ways beyond the first postsurgical year. As such, the conclusions drawn in this study can only be applied to pain in the first postsurgical year and may not apply to pain or functional outcomes in the longer term. Another limitation concerns the labelling of the pain trajectories. Adult NRS cutoffs were used in this study, as there are no agreed upon pediatric NRS cutoffs. Furthermore, there is considerable variability and disagreement regarding cutoff values used to determine mild vs. moderate vs. severe pain (Boonstra et al., 2016; Gerbershagen et al., 2011; Jensen, Tomé-Pires, Vega, Galán, Solé & Miró, 2017; Rothaug, Weiss & Winfried, 2013; van Dijk, van Wijck, Kappen, Peelan, Kalkman & Schuurmans, 2012). More research is needed in pediatric populations to determine the most appropriate cutoffs in a pediatric postsurgical population. Another important limitation in this study concerns participants who did not complete the 12-month time point. Participants who were no longer experiencing significant pain or who felt that they were recovering favorably may have dropped out of the study before the final data collection. Consequently, the final sample

may be subject to selection bias. The percentage lost to follow-up was 37% from baseline NRS measurements to 12-month NRS measurements. In order to counteract the effects of missing data, the LGMM data analysis method used employs a robust full-information maximum-likelihood (FIML) estimation procedure for handling missing data, and assumes this data can be predicted from other variables in the model(McArdle & Nesselroade, 2003). LGMM is very well suited to datasets that have missing data because FIML is considered one of the most robust methods for dealing with it. Lastly, given that this study involved a sample of adolescents undergoing spinal surgery to treat idiopathic scoliosis, the results may not be as generalizable to other major pediatric surgery populations.

### 2.6.2 Conclusion

In conclusion, pain recovery following scoliosis surgery in adolescents follows three main trajectories, two of which have a very favourable outcomes and one with persistent pain at 12-months post-operatively. The risk factors most associated with the latter trajectory included increased baseline pain and child anxiety. This trajectory also had worse functional outcomes. Future research should examine additional risk and protective factors associated with these pain trajectories as well as the functional outcomes for this population, with a view to developing mitigating strategies.

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## 2.9 Figures

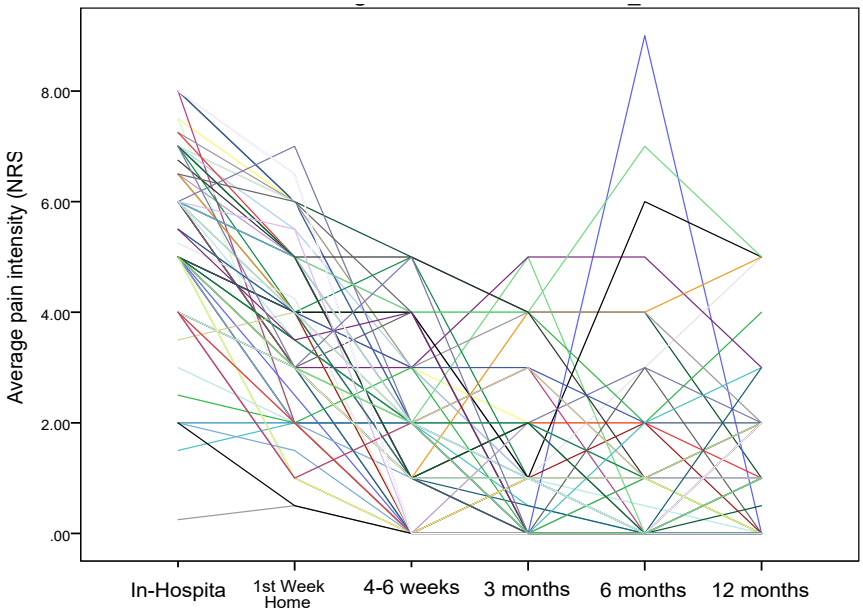


*Figure 2.1 .Spaghetti plot of NRS average pain data. Visual inspection of the data did not reveal any naturally occurring relations among the data (n = 220).*

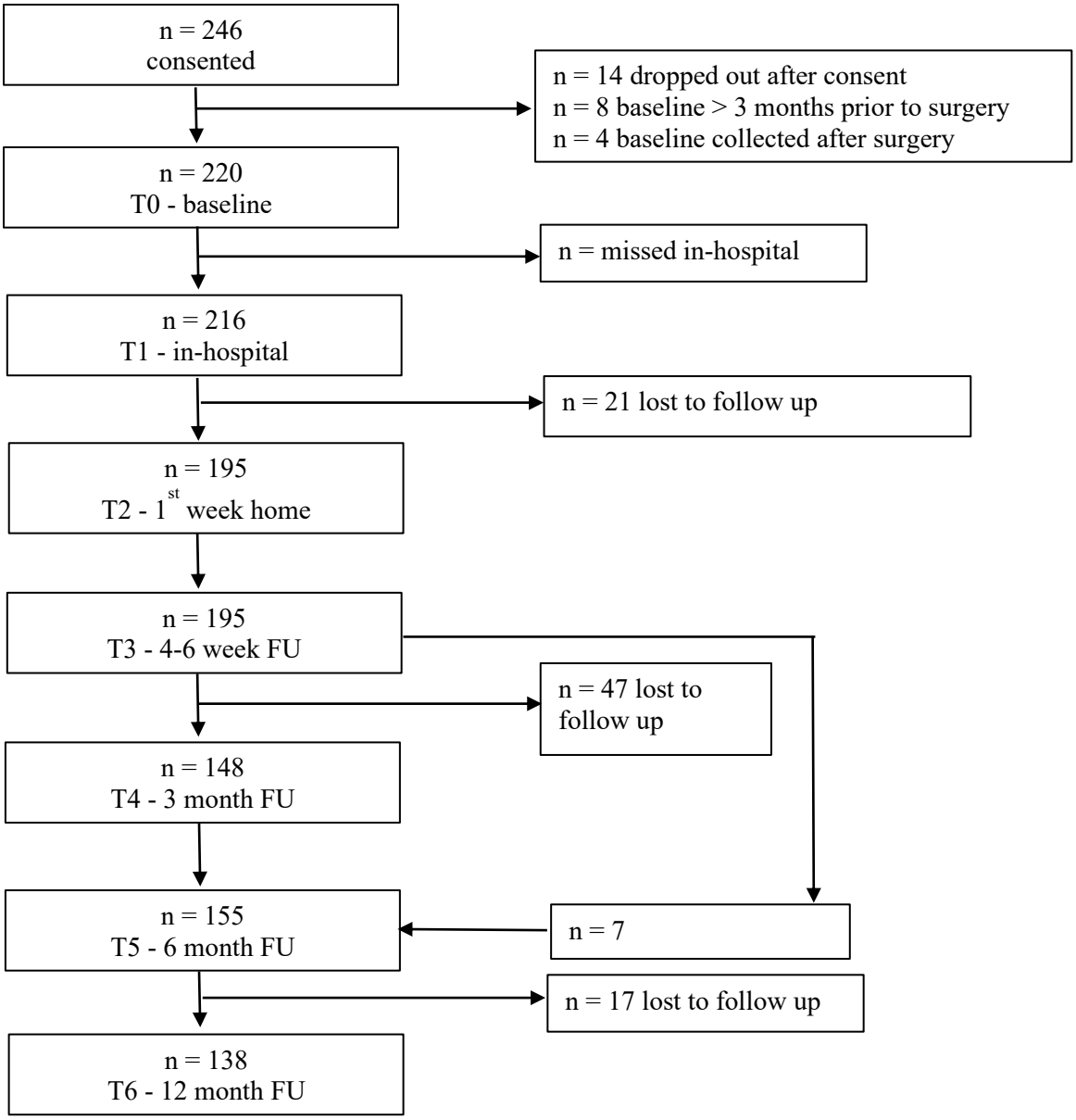


*Figure 2.2 Participant flow chart*

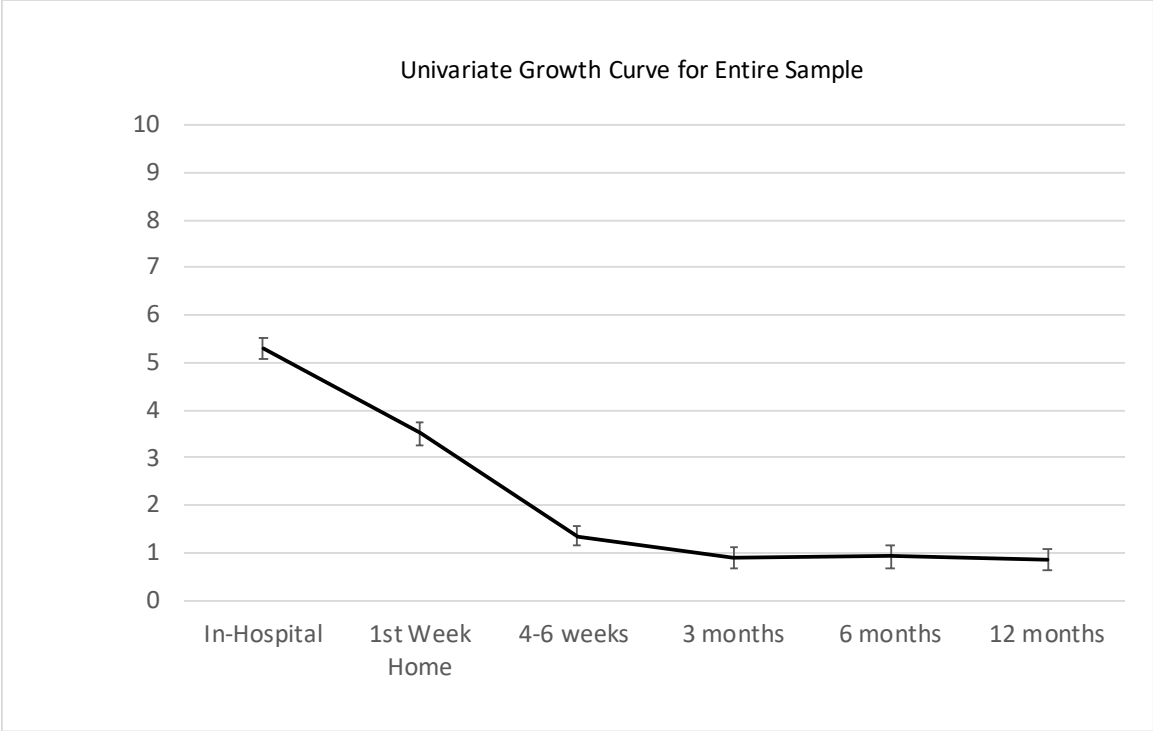


*Figure 2.3. Univariate growth curve of NRS average pain ratings for the entire sample showing a moderate amount of acute postsurgical pain that sharply declines over the first four to six weeks, stabilizing by the end of the first postsurgical year (n = 220).*

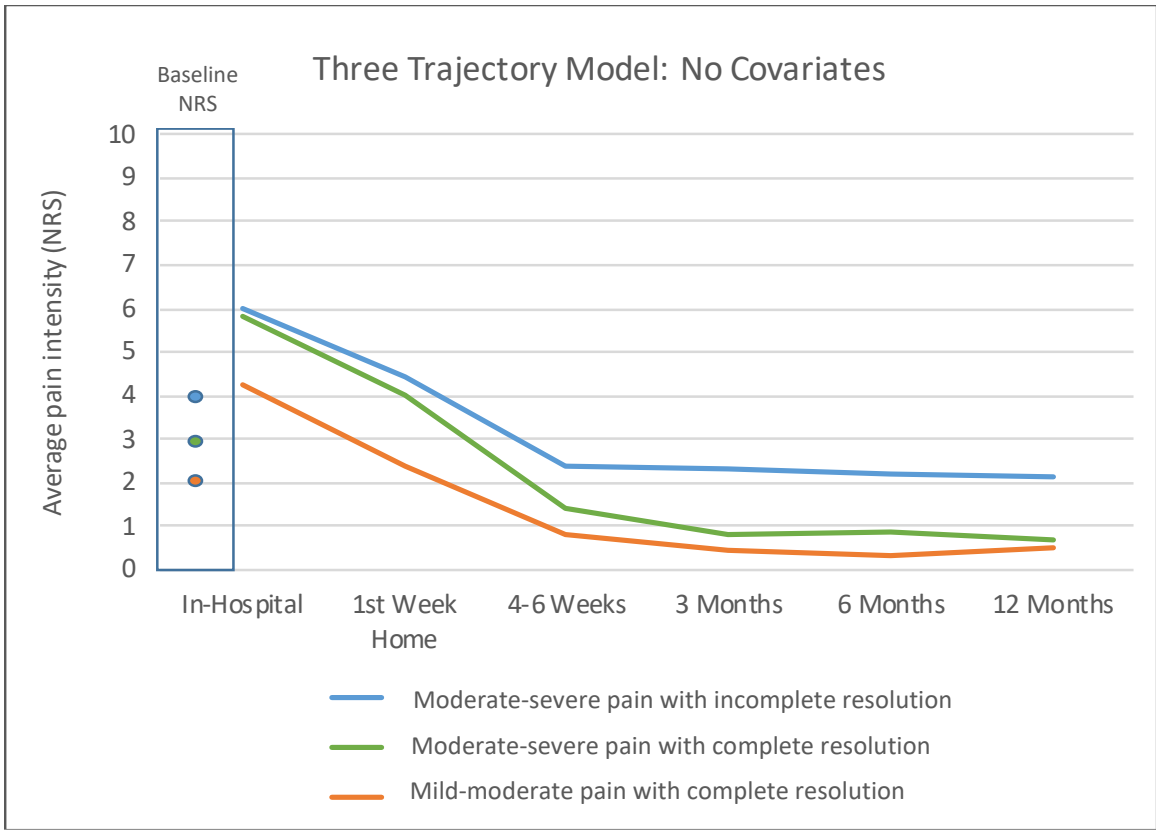


*Figure 2.4a. Conditional model of NRS average pain data for the sample showing three trajectories: [1] moderate-severe pain with incomplete resolution (n = 29; blue line), [2] moderate-severe pain with good resolution (n = 112; green line), and [3] mild-moderate pain with good resolution (n = 79; orange line).*

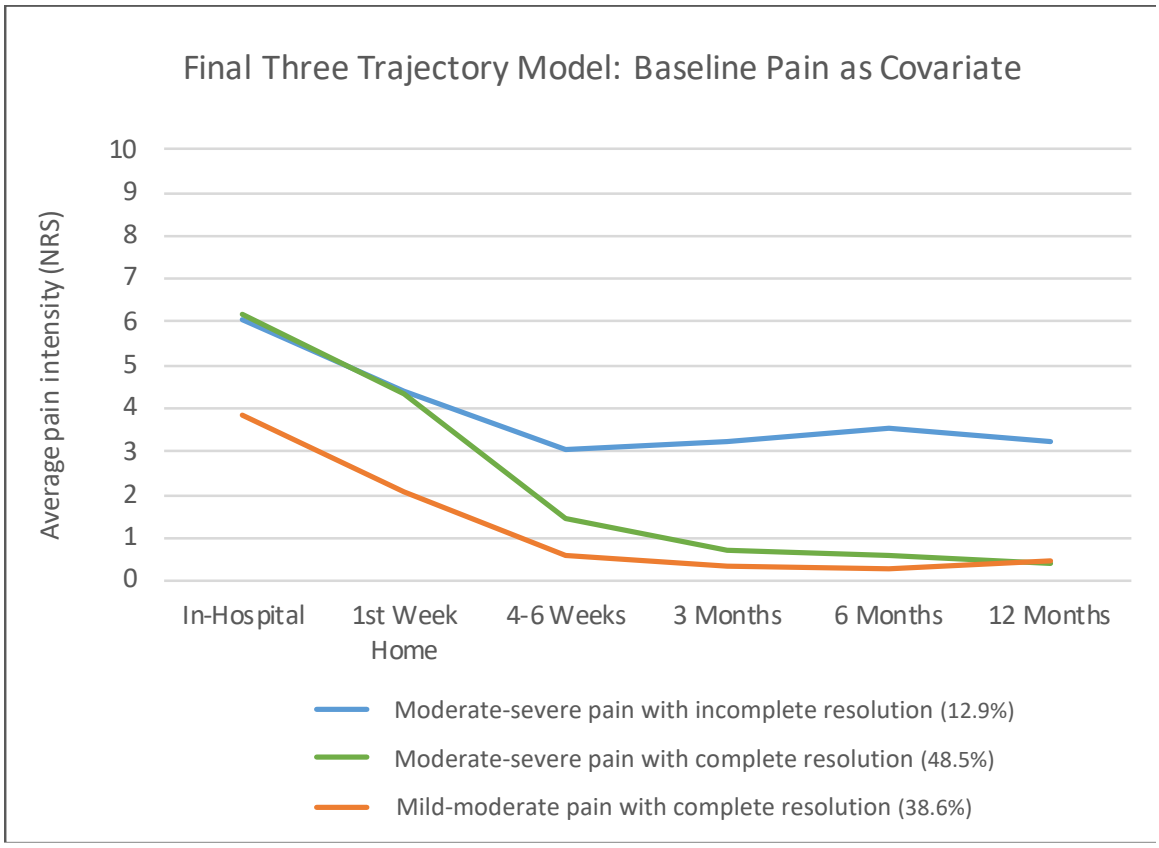


*Figure 2.4b. Conditional model of NRS average pain data for the sample with baseline pain included as a covariate showing three trajectories: [1] moderate-severe pain with incomplete resolution (n = 26; blue line), [2] moderate-severe pain with good resolution (n = 98; green line), and [3] mild-moderate pain with good resolution (n = 78; orange line).*

## 2.10 Tables

Table 2.1. Descriptives and correlations between baseline variables and average pain ratings in the postsurgical time period

| | Mean | SD | Range | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|---|---|---|---|
| **NRS average pain ratings** | | | | | | | | | | |
| 1. Baseline NRS | 2.79 | 2.29 | 0-9 | | .34** | .26** | .28** | .38** | .34** | .38** |
| 2. In-hospital | 5.27 | 1.69 | 0-9.3 | | | .68** | .37** | .27** | .16* | .15 |
| 3. 1 week post | 3.50 | 1.75 | 0-9.5 | | | | .36** | .30** | .17* | .14 |
| 4. 4-6 weeks post | 1.35 | 1.43 | 0-7 | | | | | .52** | .35** | .36** |
| 5. 3 months post | .93 | 1.34 | 0-7 | | | | | | .48** | .39** |
| 6. 6 months post | .89 | 1.66 | 0-9 | | | | | | | .63** |
| 7. 12 months post | .84 | 1.37 | 0-6 | | | | | | | |
| **Baseline Variables** | | | | | | | | | | |
| 8. Age (years) | 14.6 | 1.9 | 10-20 | .05 | .02 | .01 | .03 | .17 | .09 | .01 |
| 9. Sex (%F) | 86 | | | .10 | .21** | .18* | .10 | .01 | .07 | .14 |
| 10. Baseline time (# of days prior to surgery) | 7.8 | 13.8 | 0-85 | .04 | -.01 | .001 | .03 | -.001 | .07 | .10 |
| | 14.3 | 3.0 | 6-20 | .03 | -.15* | -.11 | .10 | .18* | -.08 | .03 |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| 11. Parents years of education | | | | | | | | | | |
| 12. Length of hospital stay (days) | 5.7 | 1.4 | 3-12 | .04 | .10 | -.001 | .11 | .05 | .13 | -.10 |
| 13. Previous surgery (%) | 31.1 | | | -.04 | -.02 | -.10 | .00 | .04 | -.03 | -.11 |
| 14. Previous hospitalization (%) | 37.7 | | | .14* | .08 | -.07 | .17* | .20* | .13 | .05 |
| 15. Prematurity (%) | 6.8 | | | .05 | .01 | -.12 | -.01 | .17* | -.03 | -.12 |

Table 2.2. Fit Indices values used to decide upon the number of trajectories that most appropriately fit the NRS average pain data.

| **Model** | **Loglikelihood** | **LMRT** | **LMRT (p)** | **BLRT** | **BLRT (p)** | **AIC** | **BICa** | **BIC difference** | **Latent class Probabilities [range]** | **Entropy** |
|---|---|---|---|---|---|---|---|---|---|---|
| 1 trajectory | -1797.47 | - | - | - | - | 3622.94 | 3626.08 | - | [1.0] | - |
| 2 trajectories | -1828.59 | -1939.28 | $p = 0.20$ | -1931.28 | $p < 0.001$ | 3687.19 | 3690.56 | (1-2)= -64 | [0.92-0.98] | 0.902 |
| 3 trajectories | -1796.30 | -1828.59 | $p = 0.56$ | -1828.30 | $p < 0.001$ | 3628.61 | 3632.65 | (2-3) = 57.91 | [0.84-0.90] | 0.700 |
| 4 trajectories | -1776.46 | -1796.30 | $p = 0.15$ | -1796.30 | $p < 0.001$ | 3594.92 | 3599.64 | (3-4) = 33.01 | [0.84-0.99] | 0.769 |
| 3 trajectories with Sex | -1768.94 | -1799.09 | $P = 0.83$ | -1799.09 | $p < 0.001$ | 3581.87 | 3586.61 | (3-3a) = 46.04 | [0.82 – 0.89] | 0.682 |
| 3 trajectories with BP | -1624.75 | -1658.32 | $P = 0.15$ | -1658.32 | $P < 0.001$ | 3293.51 | 3296.59 | (3-3b) = 336.06 | [0.87-0.93] | 0.740 |

BP – Baseline Pain

Table 2.3. Multinomial Logistic Regression for Predictors of Pain Trajectory Membership ($n$ = 156)

| | **'Mild-moderate pain with good resolution' vs.** | | | |
|---|---|---|---|---|
| | **'Moderate-Severe Pain with Incomplete Resolution'** | | **'Moderate-Severe Pain with Good Resolution'** | |
| **Predictor** | **Est.** | ***SE*** | **Est.** | ***SE*** |
| Child Sex | .95 | .87 | 1.1* | .54 |
| Child Age | .18 | .15 | .12 | .11 |
| Child PCS | -.01 | .03 | .02 | .02 |
| Child State Anxiety | .04 | .04 | -.05 | .03 |
| Child Trait Anxiety | .12* | .05 | .06 | .03 |
| Parent PCS | -.06 | .03 | -.04 | .02 |

*Note.* All variables were measured at baseline; PCS = Pain Catastrophizing Scale
*$p < .05$

Table 2.4. Means, standard deviations, and *ANOVA* statistics comparing 12 month functional outcomes across pain trajectories.

| **12-month Outcome** ***Mean (SD)*** | **#1 (Moderate-Severe Pain, Incomplete Resolution)** | **#2 (Moderate-Severe Pain, Good Resolution)** | **#3 (Mild-Moderate Pain, Good Resolution)** | **n** | **F** | **d** |
|---|---|---|---|---|---|---|
| FDI | 7.7 (7.5) | 2.8 (3.4) | 3.2 (3.2) | 128 | 9.88** | .99 |
| PedsQL | 72.5 (17.2) | 83.3 (13.4) | 86.1 (11.7) | 128 | 6.83** | .98 |
| SRS-30 Total | 3.8 (.54) | 4.1 (.37) | 4.1 (.37) | 128 | 6.32** | .96 |

Trajectory values given as mean (*S.D.*).
*$p < .05$
** $p < .01$

# CHAPTER 3. TOPICAL REVIEW: PROTECTIVE PSYCHOSOCIAL FACTORS IN PEDIATRIC POSTSURGICAL RECOVERY

The manuscript prepared for this paper is presented below. Kristen Bailey, under the supervision of Dr. Jill Chorney, was responsible for conceptualizing the paper, the review methodology, analysis of the included papers and all aspects of the writing process. She received critical feedback from her dissertation committee members (Dr. Simon Sherry and Dr. Allen Finley) on the initial proposal of the review and their feedback was incorporated accordingly. The manuscript was reviewed by Dr. Chorney and Kristen's dissertation committee members, and feedback was incorporated accordingly.

## 3.1 Abstract

**Objective**: To provide a rationale for studying protective factors as they relate to pediatric postsurgical recovery, review literature on protective factors that may be relevant and to provide directions for future research in the area. **Method**: Literature review of the postsurgical recovery and pediatric pain literature to identify protective factors relevant to pediatric postsurgical recovery. **Results**: Very little research has been conducted on protective factors in pediatric postsurgical recovery but findings form the pediatric chronic pain literature and adult postsurgical literature highlight several protective factors at the individual, parent and peer level that may be relevant for pediatric postsurgical recovery. The context in which individual factors are measured may be different for a pediatric population as compared to an adult population and parent protective factors may be secondary to individual factors. Future studies need to include both risk and protective factors to provide a larger evidence base that can be more systematically reviewed. **Conclusions**: Identifying and examining the role of protective factors in pediatric postsurgical recovery is important for furthering our understanding of postsurgical pain and functioning as well as for developing more effective and targeted interventions for individuals deemed to be at high risk of poor outcomes.

*Keywords*: postsurgical pain; postsurgical recovery; pediatric pain; resilience; protective factors

## 3.2 Introduction

The majority of youth that undergo major surgery, experience significant pain and disruption to their lives in the short term, but then go on to recover uneventfully. A subset of these patients go on to experience persistent pain but there is variability in the longitudinal findings about dysfunction associated with persistent pain; some individuals experience severe disability while others do not (Danielsson & Nachemson, 2003). These findings lead to a question regarding *why* some adolescents do not experience as much dysfunction as a result of acute or persistent postsurgical pain. Most models of postsurgical pain and chronic pain have focused on identifying those at risk of developing pain and dysfunction, with the aim of tailoring strategies to mitigate that risk. In doing so, very little attention has been paid to identifying protective factors which could be used to better understand which individuals are likely to recover well and to tailor strategies to promote resilience in those that are identified to be at risk of poor recovery. The purpose of this topical review is to advocate for the inclusion of protective factors in future research on pediatric postoperative pain and recovery following major surgery. This is accomplished by presenting a rationale for examining protective factors, reviewing available literature for protective factors, and suggesting future directions for research in this area.

## 3.3 Why Protective Factors Matter

Most of what is known about postsurgical pain and functioning comes from the adult literature (Glare et al., 2019; Katz & Seltzer, 2009b; Kehlet et al., 2006). While these seminal papers have titles that use the words, 'preventative' and 'protective factors', their models are generally risk focused and their discussions of protective factors

is generally limited to preventative analgesia. Only a few studies in the adult postsurgical literature have begun to examine factors that may serve a protective function for postsurgical pain(Mahler & Kulik, 2000b; Pinto, McIntyre, Araújo-Soares, Costa, & Almeida, 2015; Pinto, McIntyre, Ferrero, Almeida, & Araújo-Soares, 2013; Ronaldson et al., 2014). The importance of protective factors is also implied by a recently published paper that details the development of a multidisciplinary transitional pain service for adult postsurgical patients uses an Acceptance and Commitment Therapy intervention to increase psychological flexibility and mindfulness in patients identified to be at the highest risk of developing chronic postsurgical pain(Katz, Weinrib, & Clarke, 2019). Preliminary evaluation of the described transitional pain service has shown promising results regarding postsurgical pain and functioning (Azam et al., 2017; Clarke et al., 2018; Weinrib, Burns, et al., 2017) but results are still pending from a larger multi-center RCT which is still underway.

While there has been some interest in protective factors in adult postsurgical pain, the potential importance of resilience and protective factors has rapidly been growing in the area of chronic pain. A risk-resilience model of adult chronic pain has been described(Sturgeon & Zautra, 2010) and has since been adapted for pediatric chronic pain (Cousins, Kalapurakkel, et al., 2014). These models describe psychosocial risk and protective factors that are important for chronic pain and have since been cited by several studies. In terms of evidence for the potential importance of protective factors, a recent systematic review child and parent protective factors in juvenile idiopathic arthritis identified several protective factors including child and parent psychological flexilibity, child self-efficacy, adherence, pain acceptance and perceived social support (Hynes,

Saetes, McGuire, & Caes, 2019). Other studies have begun to test aspects of the pediatric risk-resilience model (including proposed mechanisms) in chronic pain (Beeckman, Hughes, et al., 2019; Cousins, Cohen, & Venable, 2015; S. Lee, Mcmurtry, et al., 2020) and found evidence of associations between risk and protective factors and that protective factors are predictive of resilient outcomes such as better psychosocial functioning and quality of life and less disability. While it is likely that some of the protective factors identified in the chronic pain resilience models could apply to pediatric postsurgical pain and functioning, differences may exist in the relation between protective factors and pain/functioning during the acute postsurgical period as compared to the chronic postsurgical period. For example, findings from the general pain literature have demonstrated that some behaviors that contribute to the maintenance of chronic pain (e.g., coping with distraction/disengagement; (Goubert, Crombez, Eccleston, & Devulder, 2004) may actually help reduce pain and distress in an acute pain context (Kohl, Rief, & Glombiewski, 2013). Therefore, it seems plausible to assume that protective cognitions and behaviors may also have different relations to pain and functioning in an acute vs. chronic pain context as would be experienced in the postsurgical recovery period. Thus, protective factors that are specific to pediatric postsurgical pain and functioning need to be identified and examined.

Studying both risk and protective factors in postsurgical pain and recovery can also contribute to the development of more effective interventions. By understanding how and why some individuals experience less pain or function well despite pain, interventions can be tailored and timed appropriately in order to bolster the resilience of individuals who are identified as being at risk of high postsurgical pain and poor

functioning. Although not yet applied in postoperative pain, the field of positive psychology provides numerous examples of how interventions based on protective factors can be used to prevent deleterious health outcomes and promote good outcomes. For example, several reviews have found that strength-based interventions can increase psychological wellbeing and reduce depressive symptoms in diverse samples (see Bolier et al., 2013; Sin & Lyubomirsky, 2009). Strength-based interventions have also been utilized in specific patient populations. For example, interventions that aimed to increase positive factors such as mindfulness or the expression of positive emotions have been found to result in enhanced quality of life and wellbeing in breast cancer patients (Casellas-Grau, Font, & Vives, 2014).

In summary, most of what is known about postsurgical pain and functioning is based upon the adult literature which does not adequately explain why most individuals recover well and why some individuals have good functioning despite experiencing pain. It is important to examine protective factors related to pain and functioning in postsurgical recovery not only to further our understanding of pain and functioning in this context but also to develop more effective interventions for those at risk of poor postsurgical outcomes. Protective factors that contribute to resilient outcomes have begun to be investigated in the chronic pain literature but little examination of similar protective factors have been done in the postsurgical literature. The following section reviews protective factors that may be relevant in pediatric postsurgical pain and functioning.

### 3.4 Protective Factors for Pediatric Postsurgical Pain and Functioning

The following section reviews protective factors that may be relevant to pediatric postsurgical pain and functioning. The search strategy and results summary were

informed and organized according to Bronfenbrenner's Ecological Systems Theory as an apriori model (Bronfenbrenner, 1979). Bronfenbrenner's Systems Theory was developed to explain how children's inherent qualities and their environment interact to influence their growth and development. The theory describes several ecosystems that begin at the level of the individual and broaden out to family and peers, community, school, social services and then to broader culture and sociohistorical time context.

Factors described in the following section were identified by conducting a literature search using five major concepts: 1) Children/Adolescents/Young Adults; 2) Surgery; 3) Pain; 4) Resilience Outcomes (i.e., quality of life, function); 5) Protective Factors (i.e., positive psychology, optimism). Most of the protective factors discussed come from the pediatric chronic pain literature as there is a paucity of research on protective factors and resilience in the pediatric postsurgical literature. Given the paucity of studies in the pediatric postsurgical literature, findings from the adult literature are also included. Consequently, the following review contains some studies from the adult acute and postsurgical pain literature and the role of the protective factors in an acute postsurgical vs chronic postsurgical context is differentiated whenever empirical evidence is available to support it. Details regarding the search strategy (i.e., Databases, search terms) can be found in Appendix A. A summary of protective factors to be considered in pediatric postsurgical recovery research is included in Table 3.1.

### 3.4.1 Individual Level Protective Factors

*Optimism*. One of the most investigated protective factors within both acute and chronic pain contexts is optimism (see Rasmussen, Scheier, & Greenhouse, 2009; Scheier & Carver, 2018). Optimism has most often been defined as the extent to which an

individual holds favorable expectancies for the future (M. E. Scheier & Carver, 1987). Optimism has been shown to be related to lower pain intensity and sensitivity following an acute pain stimulus (Geers, Wellman, Helfer, Fowler, & France, 2008; Hanssen, Peters, Vlaeyen, Meevissen, & Vancleef, 2013; Hanssen, Vancleef, Vlaeyen, & Peters, 2014) and during the acute postsurgical recovery period (Mahler & Kulik, 2000a; Pinto et al., 2015, 2013; Ronaldson et al., 2014). The relation between optimism and acute pain may be explained by more adaptive coping (Geers et al., 2008; Hanssen et al., 2013) and enhanced conditioned pain modulation (Goodin et al., 2013).

There has been growing interest in examining the role of optimism in pediatric chronic pain (Ramírez-Maestre & Esteve, 2013). Findings from some studies suggest that optimism may be related to adaptive coping. For example, a study of children with sickle cell disease found that optimism moderated the relation between pain and medication use such that children with greater optimism were better able to match their medication use with their pain ratings (Pence, Valrie, Gil, Redding-Lallinger, & Daeschner, 2007). In a study of children with various chronic pain conditions, optimism was found to be directly related to better quality of life, and this relation was mediated by reduced pain catastrophizing and reduced fear of pain (Cousins et al., 2015). In a postsurgical context, optimism was found to be positively associated with physical functioning in adolescents one year after major hip surgery (Richard, Nguyen, Podeszwa, De La Rocha, & Sucato, 2018). Several adult studies have also demonstrated a relation between optimism and quality of life (Hoofwijk et al., 2015; Jowsey et al., 2012; J. Lee, Kim, Shim, & Park, 2017) and with physical functioning six months or more after major surgery (Peters et al.,

2007; J. A. Singh, O'Byrne, Colligan, & Lewallen, 2010; Jasvinder A. Singh, Colligan, O'Byrne, & Lewallen, 2016).

Overall, optimism appears to be related to better overall functioning and more adaptive coping in a chronic pain context and in the later stages of postsurgical recovery. Much less is known about the role of optimism in an acute postsurgical context, but findings from adult experimental pain and acute postsurgical pain research suggest that optimism may be related to lower pain through adaptive coping.

*Self-efficacy***.** Self-efficacy is another promising individual protective factor that has received attention in the chronic pain literature (Tomlinson, Cousins, Mcmurtry, & Cohen, 2017). Self-efficacy can be defined as an individual's belief about their ability to function effectively even while experiencing pain (Bandura, 1977; Bursch, Tsao, Meldrum, & Zeltzer, 2006). A recent systematic review of resilience factors related to child and parent adaptation to Juvenile Idiopathic Arthritis found that at least two studies demonstrated self-efficacy was related to better quality of life and less functional disability in adolescents with Juvenile Idiopathic Arthritis (Hynes et al., 2019). Another recent study found that youth pain self-efficacy was strongly positively related to pain acceptance and quality of life in a sample of youth with various chronic pain conditions, but did not buffer the relation between pain intensity and quality of life (Lee, Mcmurtry, et al., 2020).

Little research has been conducted to examine the role of self-efficacy in a pediatric postsurgical context. One study that examined psychological and functional outcomes in adolescents who underwent hip preservation surgery reported that return to activity one year after surgery was positively associated with self-efficacy (Richard et al.,

2018). However, it is difficult to draw many conclusions about the role of self-efficacy in pediatric postsurgical functioning from this study because the authors do not describe how or when self-efficacy was measured, nor do they describe any hypotheses related to self-efficacy to justify the analyses reported. Slightly more can be gleaned from the adult postsurgical literature which has predominantly examined the role of self-efficacy in orthopedic surgery outcomes. A systematic review of eight studies that investigated the role of self-efficacy in functional recovery and wellbeing at various postsurgical timepoints after joint replacement surgery concluded that self-efficacy measured in the postsurgical period was a fairly consistent predictor of several recovery outcomes such as greater walking distance, exercise repetition/frequency, walking speed and less disability but that self-efficacy measured at baseline was a less consistent predictor of these outcomes (Magklara, Burton, & Morrison, 2014). Findings from the pediatric chronic pain literature and the adult postsurgical literature suggest that self-efficacy may be a protective factor that leads to better functional outcomes during the postsurgical period, but that it needs to be assessed during the postsurgical period once the individual has begun working towards recovery.

*Psychological Flexibility*. Psychological flexibility is defined as the capacity to be present-moment focused and behave in accordance with personal values even in the midst of experiencing interfering thoughts, emotions and bodily sensations (McCracken & Morley, 2014). A daily diary study in adolescents with various chronic pain conditions found that psychological flexibility was associated with less avoidance of activities and that it buffered the relation between pain and activity engagement such that adolescents with greater psychological flexibility tended to persist in their valued activities even

when experiencing pain (Beeckman, Simons, Hughes, Loeys, & Goubert, 2019). Psychological flexibility was also found to predict better quality of life and less negative affect among children with juvenile idiopathic arthritis (Beeckman, Simons, et al., 2019). Another study examined various mediators of change in an ACT intervention delivered to pediatric chronic pain patients reported that of several cognitive predictors, *only* psychological flexibility variables significantly mediated the relation between ACT treatment and depression scores and pain interference during the follow up period (Wicksell, Olsson, & Hayes, 2011). However, findings of this study are difficult to interpret because the authors actually measured psychological *inflexibility* (pain impairment beliefs & pain reactivity) which could be considered as more of a risk factor, but then the authors presented their findings as if they had measured a protective factor.

*Pain Acceptance***.** Pain acceptance is closely related to psychological flexibility and is another individual protective factor that has increasingly received attention as a stand-alone construct in the pediatric chronic pain literature. Pain acceptance refers to a person's willingness to acknowledge pain along with continued engagement in activities and a commitment towards living a valued life even in the presence of pain (McCracken, 1998). Pain acceptance has been associated with less functional disability amongst youth with chronic headache pain (Smith, Sieberg, Odell, Randall, & Simons, 2015b), Juvenile Idiopathic Arthritis (Beeckman, Hughes, et al., 2019) as well as amongst mixed samples of youth with various chronic pain conditions (Connolly, Ferreira, McGarrigle, & DeAmicis, 2019; Feinstein et al., 2018). Pain acceptance has also been associated with fewer depressive symptoms among youth with Juvenile Idiopathic Arthritis (Beeckman,

Hughes, et al., 2019) and better health-related quality of life among youth with various chronic pain conditions (Connolly et al., 2019).

*Mindfulness***.** Mindfulness is a core component of psychological flexibility and has been defined as paying attention on purpose, in the present moment, non-judgmentally (Kabat-Zinn et al., 1992). Much of the research on mindfulness and pain has been conducted in adults, but a few studies have examined the role of mindfulness in pediatric pain. In a community sample of adolescents, dispositional mindfulness was found to predict decreased pain interference from daily pain and decreased pain intensity and higher pain tolerance during an experimental pain task (Petter, Chambers, McGrath, & Dick, 2013). Dispositional mindfulness has also been shown to be associated with reduced anxiety and depression in youth with and without chronic pain even after controlling for pain intensity, pain catastrophizing and pain acceptance(Waldron, Gauntlett-Gilbert, Marks, Loades, & Jacobs, 2018). The study by Waldron and colleagues also found that dispositional mindfulness was positively associated with social functioning in youth with chronic pain. Studies that have examined the effect of mindfulness-based interventions in youth with pain also provide support for mindfulness as a protective factor. One study implemented a mindfulness-based intervention for adolescents with recurrent headaches through a pilot nonrandomized clinical trial and found that participants reported improved depression symptoms and improved ability to accept their pain (Hesse, Holmes, Kennedy-Overfelt, Kerr, & Giles, 2015). Parents of youth in the same study reported that their children experienced improved quality of life and improved physical functioning as well. Another study that tested a mindfulness-based intervention for youth with various chronic pain conditions using a prospective pre-

post design found that participants reported improvements in pain willingness, activity engagement, body awareness and ability to cope with stress from baseline to the three month follow up (Ruskin, Gagnon, Kohut, Stinson, & Walker, 2017). A similarly designed study that tested an eight-week mindfulness intervention found that participants reported reduced pain intensity which was maintained at three month follow up and improvements in functional disability and somatic symptoms at three month follow up (Lovas et al., 2017).

*Psychological Flexibility and Related Constructs in Postsurgical Contexts.* There has been some recognition that psychological flexibility and factors related to this construct (i.e., acceptance, mindfulness) are likely important for understanding and preventing the transition from acute to chronic postsurgical pain (Wicksell & Olsson, 2010). However, nearly a decade on, a systematic review of RCTs investigating the efficacy of psychological approaches for pain-related surgical outcomes did not find any studies that used psychological approaches designed to improve psychological flexibility, acceptance or mindfulness (Nicholls et al., 2018). Since the review was published there has been little additional research in this area and the findings from studies that have been conducted are somewhat mixed. For example, a pilot study examining the efficacy of a one day ACT workshop to prevent chronic pain and opioid use after orthopedic surgery in adult veterans deemed to be at greater risk found that increased pain acceptance as a result of the workshop was related to better postsurgical outcomes assessed three months after surgery (Dindo et al., 2018). Somewhat conversely, baseline pain acceptance was not found to be predictive of pediatric postsurgical pain trajectories or functional

disability after accounting for the effect of several other baseline predictor variables (Rosenbloom et al., 2019).

Overall, findings from the pediatric chronic pain literature suggest that psychological flexibility, pain acceptance and mindfulness are associated with better mental health, quality of life and activity engagement which may be generalizable to the chronic postsurgical period. There is very little research that has specifically examined these factors in a postsurgical context, but findings from the few studies that have been conducted suggest that the timepoint in which these factors are measured (i.e., baseline vs. postsurgical) may be important for their predictive value as protective factors.

### 3.4.2 Family Level Protective Factors.

Parents generally play a large role in a child's experience with, expression of, and management of pain (Asmundson, Noel, Petter, & Parkerson, 2012; Palermo, Valrie, & Karlson, 2014). In a postsurgical context, parents also play a large role in recovery as parent cognitions and behaviors regarding their child's pain have been found to influence their child's pain and cognitions (Noel et al., 2017; Page, Campbell, Isaac, Stinson, & Katz, 2013). Parent cognitions and behaviors may also act as risk factors for the development of chronic postsurgical pain (Connelly et al., 2014; Pagé, Campbell, et al., 2013; Rabbitts et al., 2015).

Whereas most research on parent and family factors in pediatric pain has focused on identifying risk factors, there is a growing body of research that has begun to identify protective factors (Beeckman, Hughes, et al., 2019; Feinstein et al., 2018; Hynes et al., 2019; Lee, McMurtry, et al., 2020). Parent support and good family functioning may help youth with pain to better cope with the pain and function adaptively. In general,

clear and open communication, clearly defined roles and structure, cohesion, good problem solving and emotion regulation characterize good family functioning (Palermo et al., 2014). Parent responses to children's pain are also important as parental responding to youth pain with distraction (taking attention away from the pain) has been found to reduce pain and distress in the context of acute pain (Birnie et al., 2014).

The way that parents respond to their child's pain is in part influenced by their own cognitions and beliefs regarding pain (Asmundson et al., 2012; Jaaniste et al., 2016). One of the most studied protective parental cognitions is psychological flexibility, which is defined as the capacity of a parent to be present-moment focused and behave in accordance with personal values even in the midst of experiencing interfering thoughts, emotions and bodily sensations (McCracken & Morley, 2014). Parent psychological flexibility has been related to improved child social and emotional functioning, lower functional disability and greater child pain acceptance in the context of chronic pain (Beeckman, Hughes, et al., 2019; McCracken & Gauntlett-Gilbert, 2011; Timmers, Simons, Hernandez, McCracken, & Wallace, 2019; Wallace, McCracken, Weiss, & Harbeck-Weber, 2015). One study has demonstrated that parent psychological flexibility may indirectly impact these child outcomes through both child psychological flexibility and child pain acceptance (Beeckman, Hughes, et al., 2019).

Pain-related acceptance is another parent protective factor that has received some support in the pediatric pain literature. When measured in parents, pain acceptance has been defined as a parent's willingness to allow their child to experience pain without actively trying to control or reduce it (McCracken, 1998). Similarly to parent psychological flexibility, parent pain-related acceptance has been associated with better

mental health, less catastrophizing and disability among children with chronic pain (Weiss et al., 2013). Parent pain acceptance appears to relate to less functional disability in children with chronic pain via increased child pain acceptance (Feinstein et al., 2018) and reduced parental protective responses to children's pain (Smith, Sieberg, Odell, Randall, & Simons, 2015a).

All of the research investigating the above mentioned parent-level protective factors has been conducted in youth with chronic pain. Little is known about how parent or family factors may serve a protective function in pediatric postsurgical pain but the findings of one recent study that investigated the role of numerous baseline psychosocial factors on pediatric postsurgical outcomes suggest that parent factors may play a secondary role to individual factors. In their study, Rosenbloom and colleagues (Rosenbloom et al., 2019) found that parent psychological flexibility was predictive of pain intensity and unpleasantness trajectories when examined using univariate analyses but that the effect disappeared in the multivariate analyses when several other individual variables were included as predictors. Taken together with the findings from the pediatric chronic pain literature, parent protective factors may play a role in pediatric postsurgical recovery but their influence may be more distal or indirect on postsurgical outcomes.

### 3.4.3 Peer Level Protective Factors.

Findings from a qualitative study of youth who underwent major surgery suggest that losing contact with friends as a result of the recovery process and their inability to continue the same sporting activities negatively impacted the youths' emotional wellbeing and forced them to change social networks (Rullander, Isberg, Karling,

Jonsson, & Lindh, 2013). These findings suggest that being able to continue receiving social support from peers may serve as a protective factor for adolescents wellbeing in the postsurgical recovery period. Indeed, strong peer relationships have been associated with greater independence and emotional wellbeing (Eccleston, Wastell, Crombez, & Jordan, 2008) and can strengthen perceived social competence and development (Forgeron et al., 2011) in adolescents with chronic pain. Social connection has also been found to buffer the negative impact of chronic pain on school functioning (Simons, Logan, Chastain, & Stein, 2010) and physical functioning (La Buissonniere-Ariza et al., 2018). The positive effect of peer connections is not ubiquitous, however; findings from one recent study suggests that strong peer relationships may not be associated with good functioning in the context of problematic parent pain-related cognitions (Ross, Simons, Feinstein, Yoon, & Bhandari, 2018). While it has not been investigated in in adolescents who are recovering from surgery, findings from the chronic pain literature showing that adolescents with chronic pain can be challenged to maintain their social relationships due to school absences, missed activities, not feeling understood and friends not knowing how to help when they are experiencing pain (Forgeron, Evans, McGrath, Stevens, & Finley, 2013; Forgeron et al., 2010; Sällfors, Fasth, & Hallberg, 2002) likely also apply in a postsurgical recovery context. Because social functioning can suffer in adolescents with chronic pain, peer-to-peer support programs have received some attention as potentially protective factors for youth with chronic pain as they may not only provide an adolescent with some social support but may also increase the likelihood of engagement in pain management strategies and help the adolescent to develop a more positive self-identity. One study that looked at peer-to-peer relationships that were informally

established found that such relationships provided them with emotional support that was desired but that these types of relationships tended not to last and were not often integrated into other friendship groups (Forgeron, MacLaren Chorney, Carlson, Dick, & Plante, 2015).

### 3.4.4 Community Protective Factors.

There is very little empirical research on factors more distal from the individual that could protect against pain and contribute to resilient outcomes in pediatric recovery from surgery. However, some research that examined the role of community level factors in pediatric chronic pain suggest that factors distal to the individual and family may also be important to consider for pediatric postsurgical recovery. One study that examined the relation between neighborhood characteristics, pain and pain-related disability among adolescents while controlling for familial history of chronic pain found that neighborhood features such as walkability and proximity to parks were related to lower pain, pain-related disability and higher performance on physical functioning tests (Schild, Reed, Hingston, Dennis, & Wilson, 2016). The authors concluded that features of adolescents' built environment could contribute to pain outcomes over and above family history of pain. These findings may be applicable to a pediatric postsurgical recovery context as they are likely relevant to an adolescent's physical rehab and mental health during the recovery period.

### 3.4.5 Summary of Protective Factors

In summary, there is increasing interest in identifying and understanding the role of protective factors in pain and functioning in both the pediatric chronic pain literature as well as the adult postsurgical literature. At the individual level, optimism,

psychological flexibility, pain acceptance, mindfulness and self-efficacy have all received strong support in the pediatric chronic pain literature. The protective potential of these factors in a postsurgical context is less clear due to a paucity of research but findings from studies in the adult postsurgical literature suggest that: optimism is related to less acute postsurgical pain and good functioning in the chronic postsurgical period; self-efficacy measured post-surgically is related to better physical functioning across the postsurgical period; and that pain acceptance may protect against the development of chronic postsurgical pain. At the family and peer level, good family functioning, parent psychological flexibility, parent responses to pain, parent pain acceptance and strong peer relationships have also received support as protective factors in the pediatric chronic pain literature and may be generalizable to the chronic phase of postsurgical recovery. However, the minimal research that has examined some of these factors in a pediatric postsurgical context suggests that parent protective factors may play a secondary role to individual protective factors. While there is scant research about more distal factors that relate to pain and functioning, there is some research that suggests certain features of an adolescents' neighborhood could also act as a protective factors for chronic pain and functioning that may also apply in a postsurgical recovery context.

### 3.5 Recommended Future Research Directions

- Include of both risk and protective factors studies designed to predict pediatric postsurgical pain and functioning. These factors should be measured using standardized measures so that data can be compiled and compared across studies.

- Risk and protective factors should be measured and relations between them and pain/functioning should be examined at various points in the perioperative period (i.e., baseline, acute postsurgical, chronic postsurgical).
- In addition to measuring the direct relation between protective factors and postsurgical outcomes, researchers should begin to examine the mediating and moderating effects of protective factors.
- Once a sufficient number of studies have begun to examine both risk and protective factors in pediatric postsurgical pain and functioning, a scoping review should be conducted to further elucidate the role of these factors so that a pediatric-specific model of postsurgical pain and functioning can be developed.

## 3.6 Conclusion

The vast majority of previous research regarding postsurgical recovery has focused on identifying and mitigating risk factors. This perspective does not explain why most individuals recover well from surgery nor does it explain why some individuals experience good functioning despite also experiencing pain. Identification and understanding of protective factors for postsurgical recovery could aid in the development of more effective and timely interventions for individuals deemed at risk of developing poor postsurgical outcomes. Risk-resilience models of chronic pain have begun to generate some research investigating protective factors in the chronic pain literature, some of which may apply in a postsurgical context. However, the postsurgical context is unique in that it involves an acute recovery phase which is key to understanding outcomes in the chronic recovery phase and consequently much more

research is needed to examine how protective factors identified in the chronic pain literature and elsewhere may function across the postsurgical recovery period.

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## 3.8 Tables

Table 3.1. Potential Protective Factors for Pediatric Postsurgical Recovery

| Level | Protective Factors |
|---|---|
| Individual | Psychological flexibility<br>Pain acceptance<br>Mindfulness[A]<br>Self-efficacy[A]<br>Optimism[A] |
| Family/Peer | Adaptive Family Functioning<br>Supportive school/teacher<br>Parent psychological flexibility<br>Parent pain acceptance<br>Parent promotion of adaptive coping<br>Strong peer relationships<br>Parent responses to pain – distraction[A]<br>Peer-to-peer support |
| Community | Neighbourhood – proximity to parks, walkability |

[A]Investigated as a protective factor in an acute pain context.

# CHAPTER 4. RESILIENCE IN PEDIATRIC MAJOR SURGERY: EXPLORING THE PROTECTIVE ROLE OF OPTIMISM

The manuscript prepared for this empirical study is presented below. The data used for this study was derived from the Post-Operative Recovery following Spinal Correction – Home Experiences (PORSCHE) dataset which was a multi-centre, longitudinal cohort study that collected data from 2010 to 2015. Data was collected by numerous research assistants and nurses at eight different research sites. Kristen Bailey, under the supervision of Dr. Jill Chorney, was responsible for developing the research questions and hypotheses and planning the methodology and analytic approach. Kristen consulted with her dissertation committee (Dr. Simon Sherry and Dr. Allen Finley) during the development and planning phase, and their feedback was incorporated into the conceptualization and design of the study. Kristen was responsible for preparing the dataset for analysis, conducting the analyses and writing the manuscript. The manuscript was reviewed by Dr. Chorney and Kristen's dissertation committee members, and feedback was incorporated accordingly. The authors (Bailey, K. & Chorney, J.) plan to submit the manuscript for peer-review and publication at a later date.

## 4.1 Abstract

**Objective**: Most of the research on pediatric postsurgical pain and functioning has focused on identifying and examining risk factors. There is growing interest in the pediatric chronic pain literature to identify protective factors and examine outcomes from a resilience perspective but little of this work has been done in a postsurgical context. The purpose of this study was to examine the role of optimism as a potentially protective factor for pediatric postsurgical functioning. It was hypothesized that optimism would moderate the relation between pain and functional outcomes one year after surgery. **Methods**: Youth aged 10 to 20 years old who underwent major spinal surgery for scoliosis ($N = 151$) participated in the study by completing self-reported measures of average pain, pain-catastrophizing, trait anxiety, optimism, quality of life and functional disability. Optimism and anxiety were measured at baseline, pain-catastrophizing, quality of life and functional disability were measured at four to six weeks and one year after surgery and pain was measured at all three time points. **Results**: Optimism was found to be an independent predictor of quality of life at one year and it moderated the relation between pain and functional disability at one year. Optimism was not found to moderate the relation between the same functional outcomes measured at four to six weeks. **Conclusion**: Optimism may be a protective factor for the later stages of pediatric postsurgical recovery. More research is needed to determine when in the postsurgical period optimism may be most adaptive as well as to identify and examine relations between other psychosocial protective factors.

## 4.2 Introduction

Adolescents who undergo major surgery are at risk of developing persistent and chronic pain (Nikolajsen & Brix, 2014; Page, Campbell, et al., 2013; Rabbitts, Fisher, Rosenbloom, & Palermo, 2017a). Undergoing major surgery as an adolescent can have a major impact upon normal developmental activities such as school and social interactions. Deviation from such normal developmental activities as well as the experience of ongoing pain and physical limitations can impact quality of life (Rabbitts, Aaron, et al., 2017). Most research to date has focused on identifying risk factors related to development of persistent pain and disability following surgery (Rabbitts, Fisher, Rosenbloom, & Palermo, 2017b; Rosenbloom et al., 2019; Williams, Howard, & Liossi, 2017). Identification of risk factors is important as they may be used to identify targets for intervention (Katz et al., 2015). However, it is equally important to identify protective factors in order to broaden our understanding of why many individuals have good postsurgical outcomes and to identify additional targets for intervention.

There is a small but growing body of research examining the role of positive psychology constructs in chronic and postsurgical pain populations (Cousins et al., 2015; Weinrib, Azam, et al., 2017). One of the most studied positive psychology constructs in the wider health literature is optimism (Christopher Peterson, 2000; Rasmussen et al., 2009; Scheier & Carver, 2018). Optimism has been defined as both a general tendency to expect favorable life outcomes (Scheier & Carver, 1993) and a characteristic explanatory style (Peterson & Semmel, 1982). Individuals with an optimistic explanatory style tend to attribute bad events to external factors and view bad events as temporary and specific. Individuals with an optimistic explanatory style also tend to attribute good events to internal factors and view them as enduring and global (Gillham, Shatte, Reivich, & Seligman, 2002; Peterson & Buchanan, 1995). Therefore, an optimistic explanatory

style tends to minimize and externalize negative experiences and emphasize an individual's agency in positive experiences.

Optimism is associated generally with greater physical and mental wellbeing (Scheier & Carver, 2018; Wise & Rosqvist, 2006), and is considered to promote resilience by enhancing an individual's ability to withstand and rise to challenges (Sweetman, Munz, & Wheeler, 1993). Optimists are more likely to persevere when confronted with difficulties in academic and work-related challenges, to put more effort into their personal relationships, have greater social support and engage in more health-promoting behaviours (Carver & Scheier, 2014). In contrast, pessimism has been related to poorer academic and work performance, a tendency to give up more easily when challenged, the experience of more negative emotions, and poorer health status (Wise & Rosqvist, 2006). One of the ways in which optimism is believed to impact physical health is through the way that an individual copes with difficulties. In a meta-analysis of optimism and coping, optimistic individuals were found to use more adaptive and flexible coping which are both thought to contribute to better emotional and physical health (Nes & Segerstrom, 2006).

Regarding the role of optimism in the context of pain, optimism has been associated with reduced pain perception and increased pain tolerance in both healthy adults (Geers et al., 2008; Hanssen et al., 2013; Muller, 2013) and in clinical samples (Costello et al., 2002; Shnek, Irvine, Stewart, & Abbey, 2001). Studies of adults with chronic pain suggest that optimism may be a protective factor (Goodin & Bulls, 2013). For example, in a study of adults undergoing treatment for shoulder pain, optimism was found to moderate the relation between pain catastrophizing and physical functioning such that the relation was weaker among those with high optimism as compared to those with low optimism (Coronado et al., 2017). Fewer studies

have been conducted in youth with chronic pain, but a growing body of evidence suggests that optimism may also serve a protective function. In a sample of youth with sickle cell disease, optimism moderated the relation between pain and opioid medication use suggesting that youth with greater optimism were able to better match their medication use to their pain severity (Pence et al., 2007). In a sample of youth with various chronic pain conditions, optimism was related to greater overall quality of life and to better physical functioning through lowered pain catastrophizing and pain-related fear (Cousins et al., 2015). Thus, optimism appears to buffer the negative impact of pain and pain-related risk factors on functional outcomes when pain is chronic.

Optimism has also increasingly been examined in surgical populations. In adults, optimism has been found to be related to acute postsurgical pain (Mahler & Kulik, 2000a; Pinto et al., 2013) but findings on the relation between optimism and persistent or chronic postsurgical pain are more inconsistent ( Peters et al., 2007; Pinto et al., 2015; Singh et al., 2010). Regarding functional postsurgical outcomes, optimism has been positively associated with postsurgical quality of life and overall recovery in adults undergoing elective surgeries (Hoofwijk et al., 2015; Peters et al., 2007). However, the findings regarding physical function are more mixed. Using a continuous optimism-pessimism measure, Singh and colleagues (2010) found that higher pessimism resulted in less improvement in knee function in adults two years after knee replacement surgery but not five years after surgery. Singh also found that the optimism-pessimism variable was not predictive of overall activity limitations. In a sample of adults who had undergone hip replacement, the same authors found that the optimism-pessimism measure was predictive of general activity limitation but not hip function two years after surgery ( Singh et al., 2016). Thus, optimism appears to be related to functional postsurgical outcomes, but

given the variability in findings the relation may not always be direct. It is also unclear at which point in the recovery time period the relation between optimism and functional outcomes may be strongest. Furthermore, optimism has not been examined as a protective factor in any pediatric surgical populations.

## 4.3 Objectives & Hypotheses

The overall objective of the present study is to explore the potentially protective role of optimism in a pediatric surgical sample. There are two specific aims of this study. The first aim is to examine the moderating effect of optimism on functional outcomes (i.e., quality of life, functional disability) during the chronic phase (12 months) of postsurgical recovery. It is expected that optimism will moderate the relation between pain and both functional outcomes (i.e., quality of life, functional disability) in a chronic stage of recovery (12 months) such that the relation between pain and functional outcomes at 12 months will be more favorable for those with high levels of optimism. The second aim is to examine the moderating effect of optimism on functional outcomes during the acute phase (4-6 weeks) of postsurgical recovery. Given the lack of research in pediatric populations and the mixed findings in adult populations, the analyses during the acute phase of recovery will be treated as exploratory and no hypotheses are proposed.

## 4.5 Methods

### 4.5.1 Participants

Eligible participants included children and adolescents aged 10 to 20 years old who were scheduled to undergo posterior spinal fusion and instrumentation for adolescent idiopathic scoliosis. The indication for surgery was a progressive scoliosis greater than 40 to 45 degrees in skeletally immature patients, and greater than 50 to 55 degrees in skeletally mature patients.

Exclusion criteria were children who did not speak English, children with diagnosed developmental delay that would interfere with completing study measures, or children with major chronic medical conditions (ASA status III or higher).

The data presented in this manuscript is part of the *Post-Operative Recovery following Spinal Correction: Home Experience (PORSCHE)* study, a larger project examining prevalence, predictors, and consequences of children's pain following scoliosis surgery. Participants were recruited at eight children's hospitals across Canada, including the IWK Health Centre (Halifax, Nova Scotia), Saint John Regional Hospital (Saint John, New Brunswick), Montreal Children's Hospital (Montreal, Quebec), CHU Sainte-Justine (Montreal, Quebec), Children's Hospital of Eastern Ontario, (Ottawa, Ontario), McMaster Children's Hospital (Hamilton, Ontario), Stollery Children's Hospital (Edmonton, Alberta), and Alberta Children's Hospital (Calgary, Alberta). Results of children's satisfaction with pain management are reported elsewhere (Khadra et al., 2017) as are a subset of children's memories of their postsurgical pain (Noel et al., 2017), and actor-partner effects between parent and child pain-catastrophizing from baseline to the first six weeks post-surgery (Birnie, Chorney, El-hawary, et al., 2017).

### 4.5.2 Measures

***Numerical Rating Scale of Pain***. This measure was administered at baseline, in-hospital, during the first week at home, at four to six weeks, three, six and 12 months. Only the baseline, four to six weeks and 12 month ratings were used in this study. The child was verbally asked to rate their average pain over the past 24 hours on a numerical rating scale (NRS) of 1 to 10. Average pain was assessed at three time points: baseline (up to three weeks prior to the day of surgery), four to six weeks and 12 months after surgery. The NRS is considered as adequate, reliable and valid a measure of pain in children greater than eight years old as compared to other

pain measures used in pediatric populations (i.e., Faces Pain Scale Revised) (von Baeyer et al., 2009).

***Children's Attribution Style Questionnaire (CASQ***; Seligman & et al, 1984). This measure was administered at baseline. The CASQ presents 48 hypothetical events (24 positive and 24 negative events) in a forced-choice format. Participants are asked to imagine themselves in the situation and choose one of two possible causes of the event. The two causes provided hold constant two of three attributional dimensions while varying the third. An equal number of questions pertain to each of the three dimensions (internality, stability and globality). The CASQ is scored by assigning a 1 to each internal, stable or global response and a 0 to each external, unstable or specific response. Subscale scores are summed for responses to good and bad events and a total composite score is obtained by subtracting the subscale scores for bad events from the subscale score for good events. Higher scores are indicative of more positive attribution style (i.e., more optimistic) while lower scores are indicative of a more negative attribution style (i.e., more pessimistic).

***Pain Catastrophizing Scale – Children's version (PCS-C****;* Crombez et al., 2003). This questionnaire was administered at baseline, and at four to six weeks, three, six and 12 months. Only the four to six weeks and 12 month outcomes were used in this study. This questionnaire is designed to measure children's thoughts and feelings in response to pain. Children rate their responses to pain on a 5-point, 13 item, Likert-type scale ranging from, "Not at all" to "Extremely". Items are summed to yield total scores ranging from 0 to 52 with higher scores indicating a greater amount of catastrophizing in response to pain.

***State-Trait Anxiety Inventory – Children's version (STAI-C****;* Spielberger & Edwards, 1973). This measure was administered at baseline, and at four to six weeks, three, six and 12

months postoperatively. This study uses only the baseline outcomes. This questionnaire is designed to measure both the state and trait level of anxiety of the child. Children are presented with statements of how people describe themselves and are asked to evaluate on a scale of 1 to 3, if they agree with the statement at the current moment as well as in general. Some items are reverse scored so that a greater score indicates greater anxiety with scores ranging from 20 to 60 for both state and trait anxiety.

***Pediatric Quality of Life Inventory – version 4*** (PedsQL-4; Varni, Seid, & Rode, 1999). This questionnaire was administered at baseline, and at four to six weeks, three, six and 12 months postoperatively. This study uses only the 12-month outcomes. This questionnaire is designed to measure the child's general quality of life in four categories: physical, emotional, social and school. Children are required to indicate on a scale of 0 to 4, the degree of problems the child experiences with each item. Items are then reverse scored and linearly transformed to a 0-100 scale such that higher scores indicate greater quality of life. Age appropriate versions were used (validated versions are for ages 8-12 and 13-18).

***Functional Disability Inventory (FDI***; Walker & Greene, 1991). This questionnaire was administered at four to six weeks, and three, six and 12 months postoperatively. This study uses only the 12-month outcomes. The FDI is a 15-item scale that assesses the extent to which children experience difficulties in completing specific tasks (e.g., walking to the bathroom, being at school all day). Items on the FDI are rated on a 5-point Likert scale with total scores ranging from 0 to 60 with higher scores indicating greater functional disability. The FDI has been used with a variety of pediatric populations including children with chronic pain (Kashikar-Zuck et al., 2002; Lynch et al., 2006; Reid et al., 2005) and postsurgical pain (Gidron & McGrath, 1995).

### 4.5.3 Procedure

Participants were identified and informed of the study by their attending surgeon at the time they decided to undergo the surgical procedure or at the pre-surgical visit. Families that expressed interest in the study were provided study information by a research assistant and were given the option to either consent in person or to take the materials home to review and then consent later via phone prior to the surgery. Depending on the method of consent, families received baseline questionnaires either in person or via mail and all questionnaires were returned via mail. Follow-up questionnaires were either mailed to families or completed at clinic visits that occurred at four to six weeks and 12 months after surgery. This study was approved by the respective Research Ethics Boards at all study sites.

### 4.5.4 Data Analysis

*Preliminary Analyses.* Data were analyzed with IBM SPSS Statistics for Windows, Version 25.0. Descriptive statistics were performed to characterize the sample (Table 1). Data were tested for normality and statistical assumptions were inspected. Kendall's tau-b bivariate correlation analyses were conducted to examine associations between demographic and baseline variables and variables in the primary analysis in order to identify potential covariates. Kendall's tau-b bivariate correlation analyses were also conducted to examine associations between variables in the primary analysis and baseline anxiety and state pain catastrophizing as both of these variables have been well established as psychological risk factors related to poor outcomes in pediatric chronic pain (Cohen, Vowles, & Eccleston, 2010; Karibe et al., 2015; Simons & Kaczynski, 2012; Tine Vervoort, Goubert, Eccleston, Bijttebier, & Crombez, 2006) and as risk factors for the development of persistent postsurgical pain and disability (Burns et al., 2015; Katz & Seltzer, 2009b; Khan et al., 2011; Theunissen, Peters, Bruce, Gramke, & Marcus, 2012). Findings regarding baseline pain catastrophizing as a risk factor in youth samples have

been mixed (Birnie, Chorney, & El-Hawary, 2017; Rabbitts, Fisher, et al., 2017a; Rabbitts et al., 2015; Rosenbloom et al., 2019) and it has been suggested that a child's level of pain catastrophizing measured at baseline may no longer be accurate after exposure to the pain of a major surgery (Katz, 2015). Thus, *state* pain catastrophizing at 4-6 weeks and 12 months was examined in the present study.

*Primary Analyses*. The primary analyses involved separate modeling to account for covariates, main effects and interaction terms. The PROCESS macro for SPSS was used to conduct this analysis (A. Hayes, 2018). The PROCESS macro generates hierarchical models for quality of life and functional disability to examine the comparative interaction of pain with optimism after controlling for main effects. PROCESS was also used to generate final multivariate regression models examining the interactive effect of pain and optimism on each functional outcome while accounting for main effects and covariates identified in the preliminary analyses. For all regression models, centering of variables and product terms was used to avoid multicollinearity. Significant interactions were further examined using the Johnson-Neyman technique. This technique aids in identifying the region on the optimism continuum (i.e., low versus high) where the influence of the predictor variable (e.g., average pain) on the outcome transitions from statistically significant to non-significant. The PROCESS macro default is to probe interactions at a *p* level of .10 or less. Additionally, a heteroscedasticity-consistent standard error estimator was applied to all moderation analyses in order to limit the biased and inconsistent results that can arise when conducting ordinary least squares regression when the assumption of homoscedasticity is violated (Hayes & Cai, 2007).

## 4.6 Results

Data were inspected for normality and statistical assumptions of the proposed analyses. Two outliers were identified in the 4-6 week FDI variable and four outliers were identified in both the 12 month FDI variable and the 12 month average pain variable. All identified outliers were examined and determined to be deliberate and were thus winsorized rather than deleted (Reifman & Keyton, 2010). A normal distribution was not observed for any of the 12 month variables nor for the 4-6 week average pain or FDI variables. Rather than applying a data transformation to obtain a more normal distribution, a bootstrapping procedure was applied to the moderation analysis. Bootstrapping methods are robust against violations of parametric assumptions and are recommended over data transformations when performing moderation analyses (Russell & Dean, 2000). The final sample included 151 participants. Demographic, psychological and clinical characteristics of the included sample are listed in Table 1. A summary of correlations between covariates and primary study variables is also listed in Table 1. As a result of the correlation analysis, baseline pain, trait anxiety and state catastrophizing (4-6 weeks and 12 months) were included in subsequent analyses as covariates. It should be noted that by the 12-month time point, data were only available for 106 participants. In order to examine differences for participants who completed versus those who did not, a series of independent $t$ tests and $\chi^2$ tests were conducted. Completers and non-completers were compared on age, sex, length of stay in the hospital, baseline pain, as well as history of previous surgery, previous hospitalization, chronic illness, prematurity, and whether the child had a

preadmission visit. Completers versus non-completers did not differ on any of the analyzed variables.[2]

### 4.6.1 Regression Analysis with Moderation Predicting Quality of Life

**Acute Recovery Phase (4-6 weeks)**. The overall multivariate regression model was significant, $F$ (*6, 139*) = 17.5, $p < .001$, $R^2 = .42$. There was a significant main effect of 4-6 week pain ($t = -2.90$, $p < .01$) after accounting for significant covariates. There were no other significant main effects and the interaction between 4-6 week pain and optimism was also not significant ($t = .04$, $p = .97$) (see Table 2).

**Chronic Recovery Phase (12 months)**. The overall multivariate regression model was significant, *F(6, 99)* = 8.55, $p < .001$, $R^2 = .40$. There were significant main effects of 12 month pain ($t = -2.29$, $p < .05$) and optimism ($t = 2.54$, $p < .05$) after accounting for significant covariates. There were no other significant main effects and the interaction between optimism and 12 month pain was not significant ($t = -.11$, $p = .91$) (see Table 2).

### 4.6.2 Regression Analysis with Moderation predicting Functional Disability

**Acute Recovery Phase (4-6 weeks).** The overall multivariate regression model was significant, *F (6, 144)* = 11.2, $p < .001$, $R^2 = .30$. There was a significant main effect of 4-6 week pain ($t = 5.4$, $p < .001$) but no other significant main effects or interaction effects after accounting for significant covariates (see Table 3).

[2] In order to additionally account for the missing data observed in the proposed analyses, multiple imputation modelling with 40 imputations using the Markov chain Monte Carlo algorithm available in SPSS 25.0 with auxiliary variables identified using the Missing Data Analysis function were conducted. A series of linear regressions examining the proposed main effects as well as the NRS by Optimism interaction term after accounting for relevant covariates were conducted to predict variance in the functional outcomes during both the acute and chronic recovery periods. SPSS 25.0 does not provide pooled statistics for model summary statistics but results across all 40 imputations were consistent with what is reported in the manuscript.

**Chronic Recovery Phase (12 months).** The overall multivariate regression model was significant, $F\ (6,\ 103) = 7.10$, $p < .001$, $R^2 = .31$. There was a significant main effect of 12 month pain ($t = 3.56$, $p < .001$). There was also a significant interaction effect of optimism by 12 month pain ($t = -2.62$, $p < .05$) (see Table 3). The Johnson-Neyman technique was used to determine the range of values of the moderator (Optimism) for which the relation between the predictor (NRS pain) was significantly related to the outcome (FDI). The positive relation between 12 month pain on functional disability in the chronic recovery phase was significant in participants with lower optimism (CASQ values ranging from -12.8 to 2.97, $p < .05$), while this relationship was not significant in participants with greater optimism (CASQ values ranging from 3.59 to 7.17, $p > .05$)(see Figure 1).

## 4.7 Discussion

This study sought to explore the potentially protective role of optimism in a pediatric surgical sample by investigating the relation between optimism and functional outcomes and examining optimism as a moderator of the relation between pain and functional outcomes at different time points in the postsurgical recovery period. As hypothesized, a main effect of optimism on quality of life was found in the chronic recovery phase (12 months). This finding is consistent with findings from the pediatric chronic pain literature (Cousins et al. 2015) as well as from the adult surgical literature (Hoofwijk et al., 2015;Peters, Sommer, Van Kleef, & Marcus, 2010;Peters et al., 2007) and it provides further support to the extant literature suggesting that optimism is related to greater physical, social and mental well-being (Alarcon, Bowling, & Khazon, 2013; Goodin & Bulls, 2013; Ramírez-Maestre & Esteve, 2013).

The first aim of the study was to examine optimism as a moderator of the relation between pain and functional outcomes in a chronic phase of postsurgical recovery. Results of

the present study provided partial support for the hypothesis associated with this aim. Optimism was found to moderate the relation between pain and functional disability at a later stage of recovery (12 months) such that the relation between pain and disability was weaker amongst individuals with higher levels of optimism and was stronger amongst individuals with higher levels of pessimism. This finding is in line with previous studies that have shown optimism may protect by lessening the negative impacts of pain and pain-related constructs (Coronado et al., 2017; Cousins et al., 2015). Individuals who are more optimistic may be more likely to continue engaging in activities that contribute to rehabilitation progress or be able to match their activity levels to their pain levels. Individuals in the present study who reported greater optimism did not seem to be as negatively impacted by the experience of pain at 12 months as individuals who reported greater pessimism. Thus, optimism did appear buffer the negative impact of pain on function for those adolescents experiencing more significant pain in the chronic recovery period after major spinal surgery.

Contrary to hypotheses, optimism was not found to moderate the relation between pain and quality of life. Rather, main effects of optimism and 12 month pain were observed in addition to the variance accounted for by covariates (trait anxiety and pain catastrophizing) included in the model. This finding is consistent with what has been reported in both the pediatric chronic pain literature (Cousins et al., 2015) and the adult postsurgical literature (Hoofwijk et al., 2015; Peters et al., 2007). Therefore, optimism does appear to serve a protective function but may not buffer the negative experience of pain in and of itself. Cousins and colleagues found that the relation between optimism and quality of life in a sample of youth with chronic pain was mediated fear of pain and pain catastrophizing such that youth with greater optimism reported less of these pain-related cognitions. It is possible that optimism may

better function as a moderator or buffer of the relation between pain related cognitions such as fear of pain and quality of life rather than the relation between pain and quality of life in the chronic postsurgical period. It is also possible that there was insufficient power to detect a moderation effect with these variables as these analyses were secondary to the primary intent of the cohort study design.

Regarding the second aim of this study, no hypotheses were made in regards to the analyses for the acute recovery period (4-6 weeks). Optimism was not found to moderate the relation between pain and functional outcomes in the acute phase of recovery. Pain, pain catastrophizing and trait anxiety were the only significant predictors of quality of life during the acute recovery period. Similarly, pain and pain catastrophizing were the only significant predictors of functional disability during the acute recovery period. This suggests that during a more acute phase of recovery, pain and pain-related cognitions (i.e., pain-catastrophizing) may have a more salient impact on physical function and quality of life than a broader dispositional construct such as optimism. After a major surgery such as the one experienced by participants in the present study, it is expected that patients would still be experiencing pain as healing is still occurring and thus pain may still be serving a useful function to guide return to activity. More research is needed to determine the mechanisms by which optimism may serve to protect against the negative impact of pain and to determine the optimal point during the postsurgical period at which optimism may begin to serve as a protective factor for adolescents recovering from a major surgery.

The present study used a measure of optimism with theoretical underpinnings in attribution theory and learned helplessness (Abramson, Metalsky, & Alloy, 1989; Abramson, Seligman, & Teasdale, 1978). The other major measure of optimism referred to as dispositional

optimism, has different theoretical underpinnings in expectancy theory (Scheier & Carver, 1985). While dispositional optimism tends to be used with greater frequency, a systematic review examining the relation between optimism and physical health outcomes found no significant difference in effect size as a function of the type of optimism measure used suggesting that both types of optimism relate to health outcomes (Rasmussen et al., 2009). Nevertheless, it is possible that some of the findings observed in the present study may be different from that of previous research due to the measure of optimism that was used. Future research should examine the utility of both measures of optimism in a pediatric postsurgical context.

### 4.7.1 Limitations & Future Directions

When interpreting the results of this study, some caveats should be considered. It is difficult to determine causal relations given the cross-sectional design and analyses employed in the present study. While optimism is thought of as a trait-like variable, longitudinal studies that examine the stability of the construct in youth samples are needed to lend support to that notion. The present study is novel in that it examines the relations between these constructs at two different points in the postsurgical period but the analyses cannot be used to determine causality between the variables.

While the most prominent risk factors studied in postsurgical recovery are baseline pain, pain catastrophizing and anxiety, this study did not account for other factors that might influence these constructs such as anxiety sensitivity, depression and baseline functioning. Similarly, while optimism is one of the most prominent positive psychology variables explored in the pain literature and wider health literature, future research should examine other potential protective factors in pediatric postsurgical recovery such as pain acceptance, psychological flexibility, parenting style and cultural female ideal body image. Understanding more about other protective

factors will provide a broader foundation upon which to explore how protective factors relate to coping strategies, cognitions, and recovery from major surgery. Furthermore, a better understanding of potential protective factors could be used to develop more comprehensive preparation and rehabilitation programs that could be incorporated into existing transitional pain services for children and adolescents. Incorporation of such protective factors has already begun in some adult transitional pain services (Katz et al., 2015).

### 4.7.2 Conclusion

In conclusion, optimism was independently predictive of quality of life and buffered the negative impact of pain on functional disability in adolescents one year after major spinal surgery but these same relations were not observed four to six weeks after surgery. Future studies should examine the relation between optimism, pain and functional outcomes at different points in the postsurgical period to determine when optimism may begin to be beneficial to the recovery process. Future studies should also explore other potentially protective factors at the individual, family, and community levels in order to develop more comprehensive rehabilitation programs for children and adolescents undergoing major surgery.

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## 4.10 Figure

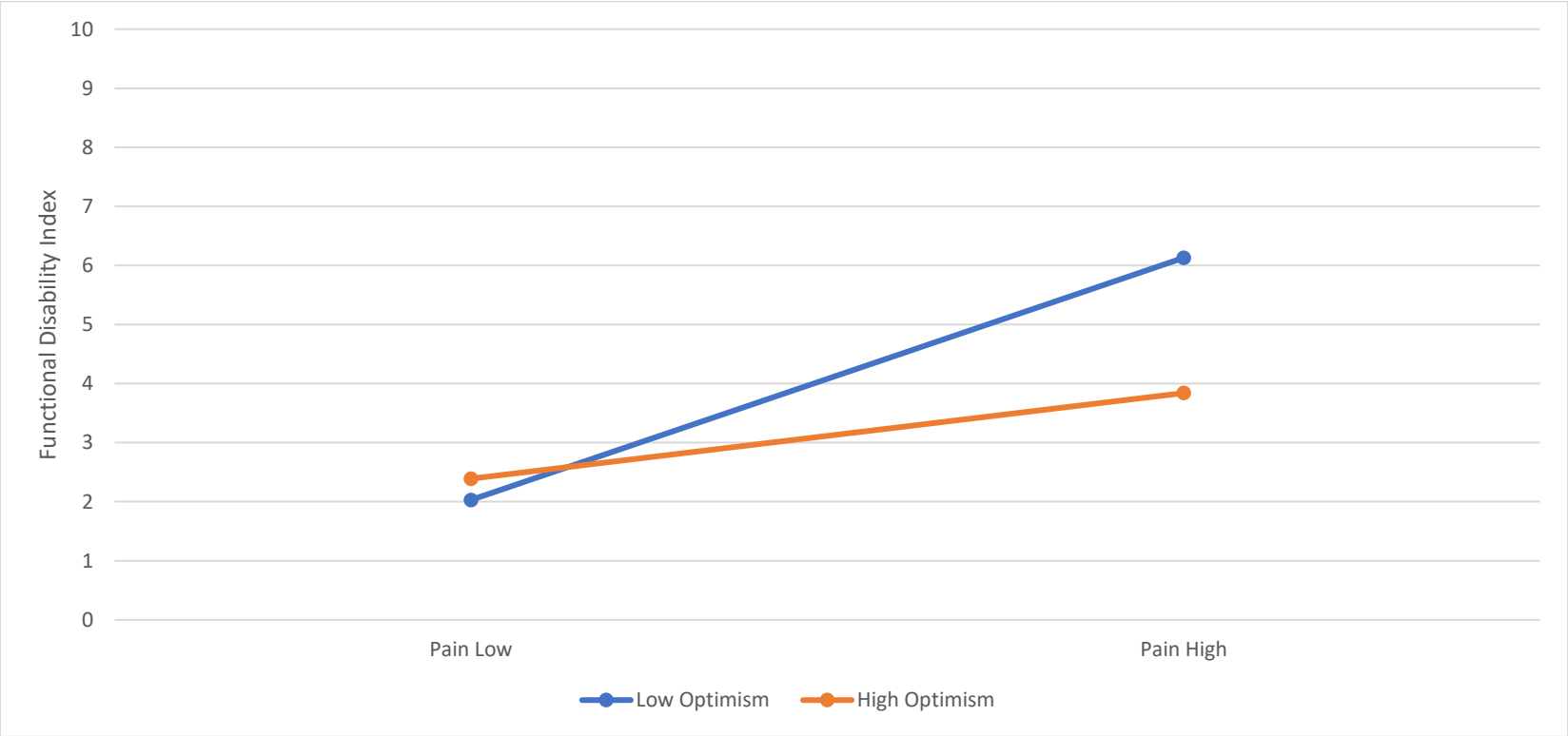


*Figure 4.1 Moderating effect of optimism on the relation between 12-month pain and functional disability.*

## 4.11 Tables

Table 4.1. Descriptives and correlations between demographic variables and primary analysis variables

| | Mean | SD | Range | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|---|---|---|---|
| **Primary Analysis Variables** | | | | | | | | | | |
| 1. 4-6 weeks NRS | 1.42 | 1.40 | 0-5 | | .36** | -.08 | -.31** | -.13 | .40** | .12 |
| 2. 12 months NRS | .85 | 1.35 | 0-5 | | | -.04 | -.21** | -.27** | .20* | .34** |
| 3. Optimism | 5.38 | 4.92 | -7-15 | | | | .23** | .29** | -.07 | -.12 |
| 4. 4-6 weeks PedsQL | 62.4 | 14.8 | 23.3-100 | | | | | .40** | -.46** | -.16* |
| 5. 12 months PedsQL | 82.2 | 13.2 | 41-100 | | | | | | -.18* | -.37** |
| 6. 4-6 weeks FDI | 17.0 | 9.8 | 0-39 | | | | | | | .15* |
| 7. 12 months FDI | 3.4 | 4.1 | 0-15 | | | | | | | |
| **Baseline Variables** | | | | | | | | | | |
| 8. Baseline NRS | 2.8 | 2.4 | 0-9 | .23** | .30** | -.10 | -.21** | -.12 | .13* | .15* |
| 9. Age (years) | 15.0 | 1.8 | 11-20 | .11 | -.02 | -.04 | -.07 | .06 | -.01 | -.13 |
| 10. Sex (%F) | 87 | | | .01 | 0.02 | .01 | -.01 | -.06 | .13 | .12 |
| 11. Previous Surgery (%) | 30.3 | | | -.02 | -.14 | .-06 | .08 | .11 | -.01 | -.12 |

| | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 12. | History of Chronic Illness (%) | 10.5 | | | -.03 | .04 | -.09 | -.03 | -.10 | .08 | .03 |
| 13. | Previous Hospitalization (%) | 40.5 | | | .12 | .07 | -.14* | -.12 | -.01 | .13 | -.04 |
| 14. | Prematurity (%) | 7.3 | | | .05 | -.01 | -.05 | -.05 | .07 | .06 | -.02 |
| 15. | Parent Years of Education | 14.3 | 3.0 | 8-20 | .01 | -.01 | .01 | -.06 | -.01 | .03 | -.08 |
| **Potential Psychological Covariates** | | | | | | | | | | | |
| 16. | Trait Anxiety | 34.9 | 7.4 | 13-57 | .16** | .15* | -.29** | -.41** | -.31** | .19** | .17** |
| 17. | 4-6 weeks PCS | 16.5 | 8.5 | 0-39 | .18** | .08 | -.11* | -.27** | -.16** | .25** | .02 |
| 18. | 12 months PCS | 13.6 | 9.4 | 0-37 | -.01 | .24** | -.18** | -.24** | -.33** | .14* | .20** |

Table 4.2 Predictors of Quality of Life in Acute and Chronic Recovery Phases

| Model | *B* | *SE B* | *t* | *p* | $R^2$ | *n* |
|---|---|---|---|---|---|---|
| **Acute Phase** | | | | | .42 | 146 |
| NRS 4-6 weeks | -2.15 [-3.60, -.78] | .74 | -2.90 | <.01 | | |
| Optimism | .26 [-.15, .69] | .22 | 1.16 | .25 | | |
| NRS 4-6 X Optimism | .01 [-.31, .32] | .17 | .04 | .97 | | |
| *Covariates* | | | | | | |
| Trait Anxiety | -.85 [-1.11, -.56] | .14 | -5.98 | <.001 | | |
| PCS 4-6 weeks | -.33 [ -.54, -.09] | .12 | -2.80 | <.01 | | |
| NRS Baseline | -.42 [-1.36, .43] | .46 | -.90 | .37 | | |
| **Chronic Phase** | | | | | .40 | 106 |
| NRS 12 months | -3.15 [-5.67, -1.08] | 1.38 | -2.29 | <.05 | | |
| Optimism | .65 [.16, 1.07] | .26 | 2.54 | <.05 | | |
| NRS 12 X Optimism | -.04 [-.68, .41] | .40 | -.11 | .91 | | |
| *Covariates* | | | | | | |
| Trait Anxiety | -.39 [-.77, -.06] | .20 | -1.98 | .05 | | |
| PCS 12 months | -.43 [-.70, -.17] | .14 | -3.03 | <.01 | | |
| NRS Baseline | .42 [-.57, 1.42] | .55 | .76 | .45 | | |

Table 3. Predictors of Functional Disability in Acute and Chronic Recovery Phases

| Model | *B* | *SE B* | *t* | *p* | $R^2$ | *n* |
|---|---|---|---|---|---|---|
| **Acute Phase** | | | | | .30 | 151 |
| NRS 4-6 weeks | 2.70 [1.72, 3.66] | .50 | 5.36 | <.001 | | |
| Optimism | .06 [-.21, .34] | .14 | .43 | .66 | | |
| NRS 4-6 X Optimism | -.14 [-.34, .06] | .11 | -1.29 | .20 | | |
| *Covariates* | | | | | | |
| Trait Anxiety | .17 [-.06, .38] | .12 | 1.43 | .16 | | |
| PCS 4-6 weeks | .22 [.03, .40] | .10 | 2.22 | <.05 | | |
| NRS Baseline | .15 [-.44, .76] | .31 | .49 | .63 | | |
| **Chronic Phase** | | | | | .31 | 110 |
| NRS 12 months | 1.26 [.57 1.97] | .35 | 3.57 | <.001 | | |
| Optimism | -.07 [-.22, .11] | .09 | -.79 | .43 | | |
| NRS 12 X Optimism | -.13 [-.24, -.01] | .05 | -2.62 | <.05 | | |
| *Covariates* | | | | | | |
| Trait Anxiety | .04 [-.05, .14] | .05 | .79 | .43 | | |
| PCS 12 months | .04 [-.02, .11] | .03 | 1.18 | .24 | | |
| NRS Baseline | .02 [-.31, .37] | .18 | .11 | .91 | | |

# CHAPTER 5. DISCUSSION

This overall aim of this dissertation was to contribute to our understanding of postsurgical pain and functioning following major pediatric surgery and examine predictors of recovery from a resilience perspective. Specific objectives of the dissertation were to: 1) Examine postsurgical pain trajectories and functioning following major pediatric surgery, 2) Review protective factors relevant to pediatric postsurgical recovery, 3) Examine one protective factor for pediatric postsurgical recovery in greater depth. To accomplish these objectives, three separate manuscript-style papers were presented as part of this dissertation. Paper one (Chapter two) addresses objective one using data derived from a multi-center longitudinal study to examine postsurgical pain trajectories and functional outcomes in the year after major pediatric surgery. Paper two (Chapter three) addresses objective two in the form of a topical review. Paper three (Chapter four) addresses objective three by examining optimism as a moderator of the relation between pain and functioning at two timepoints in the postsurgical period. The following sections provide a summary of the main findings of each paper and integrates the findings together within existing literature. This is followed by a discussion of the theoretical and clinical implications of the dissertation research. Next there is a discussion of strengths and limitations of the dissertation research which is followed by directions for future research.

## 5.1 Summary and Integration of Key Findings

### 5.1.1 Summary of Paper One

Paper one (Chapter two) used latent growth curve modelling to identify three distinct postsurgical pain trajectories based upon seven measurements of pain over one year following major spinal surgery for scoliosis. As hypothesized, a subset of the sample (12%) followed a trajectory characterized by persistent pain. This, 'moderate-severe pain with incomplete

resolution' trajectory was characterized by moderate to severe immediate postsurgical pain that declined over the next several weeks but then remained stable in the mild to moderate pain range for the remainder of the postsurgical year. Nearly half of the sample followed a, 'moderate-severe pain with good resolution' trajectory which was characterized by moderate to severe immediate postsurgical pain that steadily declined over the next several weeks and then declined further but at a slower rate to near resolution by 12 months. More than a third of the sample followed a 'mild-moderate pain with good resolution' trajectory which was characterized by mild to moderate immediate postsurgical pain that steadily declined over the next several weeks to near resolution that remained stable over the remainder of the postsurgical year. Also as hypothesized, individuals in the 'moderate-severe pain with incomplete resolution' trajectory reported worse functional outcomes at 12 months as compared to individuals in the other two trajectories. Partial support was found for the hypothesis about predictors of trajectory membership as baseline pain and child anxiety were predictive of membership in the, 'moderate-severe pain with incomplete resolution' trajectory, but no other parent or child factors were found to be significant predictors.

### 5.1.2 Summary of Paper Two

Paper two (Chapter three) is a topical review that asserts the importance of identifying and examining protective factors in pediatric postsurgical recovery, reviews the most recent literature on protective factors that may be relevant for pediatric postsurgical recovery and discusses future research directions. Identifying and examining protective factors is important to further our understanding pediatric postsurgical recovery so that more comprehensive and targeted interventions can be developed to help bolster the resilience of individuals identified to be at risk of poor postsurgical outcomes. There is very little research on protective factors in

pediatric postsurgical recovery but findings from the growing interest in protective factors in the pediatric chronic pain and adult postsurgical pain literature provide a starting point. At the individual level self-efficacy, pain acceptance and optimism are likely important for predicting recovery, but the timing of their measurement may be important in predicting outcomes. At the parent level parent psychological flexibility and parent pain acceptance are likely important but may be secondary to individual predictors. Future research needs to include measures of both risk and protective factors when studying pediatric postsurgical outcomes and begin to examine the mediating and moderating effects of protective factors.

### 5.1.3 Summary of Paper Three

Paper three (Chapter four) examines optimism, one of the most investigated protective factors in pain, as a moderator of the relation between pain and functioning during two time points in the postsurgical recovery period. Partial support was found for the hypothesis that optimism would moderate the relation between pain and functional outcomes during the chronic postsurgical recovery period. Optimism was found to moderate the relation between pain and functional disability during the chronic postsurgical period such that the relation between pain and functional disability was weaker amongst individuals with higher levels of optimism and was stronger amongst individuals with lower levels of optimism. Optimism was not found to moderate the relation between pain and quality of life during the chronic postsurgical period nor was optimism found to moderate the relation between pain, or either of the functional outcomes during the acute postsurgical period.

### 5.1.4 Integration of Findings with Existing Research

**5.1.4.1 Pediatric Postsurgical Pain and Functioning.** One of the major goals of this dissertation was to provide more information about postsurgical pain and functioning following

major pediatric surgery. This was important as most of what is known about postsurgical pain and functioning after major surgery has come from research conducted in adults (Glare et al., 2019; Katz & Seltzer, 2009b; Kehlet et al., 2006). At the time that data collection began for data collection in the longitudinal cohort, no studies reporting frequent measures of pain and functioning in the first postsurgical year following major pediatric surgery had been published. Results presented in paper one of this dissertation contribute to and extend what is now a growing literature on pediatric postsurgical pain and functioning (Chidambaran et al., 2017; Connelly et al., 2014; Rabbitts et al., 2015; Rosenbloom et al., 2019; Sieberg et al., 2013). First, several findings in paper one are consistent with what has recently been reported in other studies. Paper one demonstrated that the majority of adolescents who undergo major surgery tend to recover very well by 12 months with limited pain and good functioning. This is consistent with Rabbitts and colleagues (2015) who found that 49 of their 60 participants followed a pain trajectory characterized by early recovery with limited pain and activity restrictions and good quality of life by one year. This is also consistent with Rosenbloom and colleagues (2019) who found that more than half of their participants were classified into a trajectory characterized by mild baseline pain which remained stable throughout the first postsurgical year and that the majority of their participants reported little to no functional disability by one year. Also consistent with what has been reported in previous studies, paper one demonstrated that a subset of individuals follow a trajectory characterized by moderate to severe pain in the acute postsurgical period that shows some improvement over the ensuing weeks but then stabilizes in the mild to moderate pain range for the remainder of the postsurgical year. This is also consistent with Rabbitts and colleagues (2015) who found that 11 of their 60 patients followed a trajectory characterized by higher immediate postsurgical pain that was slow to resolve. This is

also somewhat consistent with Rosenbloom and colleagues (2019) who found less than half their sample followed a trajectory characterized by moderate to severe baseline pain that remained throughout the postsurgical year. A final consistency between the findings of paper one and previous studies is evidence that individuals in the persistent pain trajectory reported worse functioning at 12 months as compared to participants in other trajectories. This is consistent with Rabbitts and colleagues (2015) who reported greater activity limitations and worse quality of life for the patients in their, 'late recovery' trajectory. These findings contribute to building a consistent evidence base which helps clinicians, adolescents and their families know what to expect following major surgery so that they can make informed decisions and plan appropriately for their recovery period.

There are also some unique findings of paper one that extend what is known in the growing literature on pediatric postsurgical pain and functioning that warrant further discussion. Paper one identified three distinct pain trajectories using frequent measurements of pain over the first postsurgical year whereas all previous pain trajectory studies employing such frequent measurements during the first postsurgical year only identified two trajectories (Connelly et al., 2014; Rabbitts et al., 2015; Rosenbloom et al., 2019). The third trajectory identified in paper one labelled, 'moderate-severe pain with complete resolution' contains nearly half of the sample and was characterized by moderate to severe immediate postsurgical pain that was similar to that observed in the 'moderate-severe pain with incomplete resolution' trajectory. However, unlike the, 'moderate-severe pain with incomplete resolution' trajectory, the 'moderate-severe pain with complete resolution' trajectory then demonstrated a steeper decline in pain following discharge from the hospital that then continued to decline more gradually over the next several months. The course of pain experienced by individuals in the 'moderate-severe pain with complete

resolution' trajectory is important as it challenges some of the assumptions we have about the relation between acute and chronic postsurgical pain. Unlike what has been reported in previous studies, the findings from paper one suggest that more work needs to be done to differentiate the individuals who experience high immediate postsurgical pain but then go on to experience different rates of pain resolution in the following weeks.

**5.1.4.2 Predicting Trajectories of Postsurgical Pain and Functioning.** Findings from the adult postsurgical pain literature have identified high levels of immediate postsurgical pain as a risk factor for the development of chronic postsurgical pain (see Glare et al., 2019; Kehlet et al., 2006). Paper one of this dissertation identified two pain trajectories with high levels of immediate postsurgical pain but observed that only one of those trajectories demonstrated continued pain that persisted until one year after surgery. This finding suggests that the rate of pain resolution during the acute postsurgical period may be more important in determining the subsequent development of chronic postsurgical pain than the presence of high immediate pain alone. One recent study conducted in adults undergoing various major surgical procedures examined acute pain trajectories and their relation to postsurgical pain at six months (Althaus et al., 2018). The authors observed considerable variability in patients' acute pain over the first five days after surgery and identified three distinct trajectories, two of which were characterized by severe immediate postsurgical pain. When they examined the relation between the acute postsurgical pain trajectories and subsequent pain at six months, the authors found that the rate of acute pain resolution was a better predictor of pain at six months than immediate postsurgical pain intensity, with slower resolution predicting the presence of chronic postsurgical pain. To date, no studies have examined acute postsurgical pain trajectories in pediatric populations, but findings from paper one of this dissertation as well as recent findings in the adult literature

suggest that more attention needs to be given to the rate of pain resolution in the acute postsurgical period in order to better understand the development of chronic postsurgical pain.

Findings from paper one of this dissertation also contribute to the growing body of literature about predictors of pediatric postsurgical pain and functioning other than acute postsurgical pain. Paper one found that baseline pain was predictive of membership in the 'moderate-severe pain with incomplete resolution' trajectory and that adding baseline pain as a covariate to the model enhanced the model's fit to the data. Baseline pain has consistently been found to be a risk factor for the development of chronic postsurgical pain in adult studies (see Glare et al., 2019) but the findings thus far in pediatric studies are mixed. Connelly and colleagues (2014) modelled high and average pain trajectories in a small sample ($N = 50$) of adolescents who underwent spinal surgery for AIS and found that baseline pain was associated with slower improvement in pain over the six months after surgery. However, in a similarly sized sample of children undergoing mixed major procedures, Rabbitts and colleagues (2015) did not find presurgical pain to be predictive of membership in their 'late recovery' trajectory which was characterized by moderate pain at four months and mild to moderate pain one year after major surgery. Using a more sizeable sample of children who underwent spinal surgery ($N = 144$), Chidambaran and colleagues (2017) found that presurgical pain was predictive of the presence of postsurgical pain at three months but not one year. These findings suggest that baseline pain may be important for predicting pain resolution in the first few months after major pediatric surgery but may not be as important a predictor for postsurgical pain later in the postsurgical period. More research is needed to determine the role of baseline pain in predicting pediatric postsurgical pain outcomes across the postsurgical recovery period.

In addition to baseline pain, the adult literature has identified several psychosocial risk factors for postsurgical pain and functioning such as trait anxiety, depression and pain catastrophizing (see Glare et al., 2019). Paper one of this dissertation examined baseline child anxiety and pain catastrophizing as predictors of trajectory membership as well as parent factors known to be important risk factors for poor chronic pain outcomes (i.e., parent anxiety, parent pain catastrophizing) and found that only child trait anxiety was predictive of membership in the persistent pain trajectory. The research regarding baseline psychosocial risk factors for pediatric postsurgical pain is growing, and the findings that have been reported suggest that psychosocial factors are important but may be different than what has been reported in the adult literature. Connelly and colleagues (2014) found that greater baseline anxiety, but not negative mood was predictive of slower pain resolution. Rabbitts and colleagues (2015) found that parental pain catastrophizing, but not child pain catastrophizing was predictive of membership in the 'late recovery' pain trajectory. Chidambaran and colleagues (2017) measured several baseline psychosocial factors including child and parent anxiety, child and parent pain catastrophizing, and child anxiety sensitivity but only found child anxiety sensitivity to be predictive of postsurgical pain at one year. Similarly, in their univariate analyses, Rosenbloom and colleagues (2019) found child anxiety, pain-related anxiety, depression and post-traumatic stress to be predictive of membership in their persistent pain trajectory as well as predictive of functional disability at 12 months. Given the variability in findings, it is difficult to draw strong conclusions without systematically reviewing the findings reported in these studies. Nevertheless, it is clear that psychosocial factors of children and their parents likely play a role and that future studies need to assess these factors using standardized measures so that findings can be compiled and compared across studies.

One of the most novel contributions that this dissertation makes to the pediatric postsurgical literature is an impetus for researchers to view pediatric postsurgical populations as resilient and begin examining protective factors that lead to resilient outcomes rather than focusing solely on risk factors that lead to pain and dysfunction. Knowledge of both risk and protective factors will lead not only to a more comprehensive understanding of pediatric postsurgical recovery but can also be used to design more effective and targeted interventions for individuals who may be at risk of poor postsurgical outcomes. Paper two describes many protective psychosocial factors that may be relevant to pediatric postsurgical pain and functioning. Most of the factors reviewed come from the pediatric chronic pain literature, which is demonstrating a rapidly growing interest in resilience and protective factors. Protective psychosocial factors have only been examined in three pediatric postsurgical studies to date (Connelly et al., 2014; Richard et al., 2018; Rosenbloom et al., 2019) and in a few adult postsurgical studies (Mahler & Kulik, 2000a; Pinto et al., 2015, 2013; Ronaldson et al., 2014). Findings from these studies generally suggest that psychosocial protective factors are important predictors for postsurgical pain and functioning and that much more research is needed to determine what protective factors are important and how they function in the postsurgical period.

**5.1.4.3 Optimism and Postsurgical Outcomes.** The findings from study three of this dissertation add to the large body of literature that supports optimism as a construct related to positive health outcomes (Rasmussen et al., 2009; Scheier & Carver, 2018; Wise & Rosqvist, 2006). This dissertation also adds to the growing body of literature that has begun to examine optimism as a protective factor in pediatric pain (Cousins et al., 2015; Pence et al., 2007) and pediatric postsurgical functioning (Richard et al., 2018). Findings from paper three

demonstrated that optimism predicts quality of life one year after major surgery even after accounting for pain, anxiety, and pain catastrophizing. This is consistent with findings reported by Cousins and colleagues (2015) who found that optimism predicted quality of life even after accounting for pain intensity and duration, pain-related fear and pain-catastrophizing. Together these findings suggest that optimism may act as a protective factor regardless of the level of pain an adolescent is experiencing. Findings from paper three are also the first to demonstrate optimism as a moderator of the relation between pain and functional disability one year after major pediatric surgery. Findings regarding the relation between optimism and physical functioning in the postsurgical period have been mixed in adults (Singh et al., 2010; Jasvinder A. Singh et al., 2016). Findings from this dissertation suggest that optimism may only have a protective effect on physical functioning when pain is still present in the chronic postsurgical period. The moderation analysis used in paper three included pain catastrophizing as a covariate but a study of adults with chronic shoulder pain suggests that optimism may also moderate the negative effect of pain catastrophizing on physical functioning (Coronado et al., 2017).

### 5.2 Theoretical Implications

The findings from this dissertation have implications for theories regarding interpersonal factors, resilience and optimism as they relate to pain and functioning. First, the findings of this dissertation have implications for the application of the interpersonal fear-avoidance model (IFAM; Goubert & Simons, 2014; Simons, Smith, Kaczynski, & Basch, 2015) of pain in a pediatric surgical context. The IFAM extends the fear-avoidance model of chronic pain (Vlaeyen & Linton, 2000) through the inclusion of parent cognitive-affective and behavioral factors that have either been shown to impact child pain-related outcomes or are theorized to impact such outcomes. Parent pain catastrophizing is one parent cognition proposed by the

IFAM to be related to children's pain experience based upon literature from both acute (T. Vervoort, Goubert, & Crombez, 2009; Vervoort et al., 2011) and chronic pain (Birnie et al., 2016; Feinstein et al., 2018; Lynch-Jordan, Kashikar-Zuck, Szabova, & Goldschneider, 2013). Parent pain catastrophizing has also been found to be predictive of pediatric postsurgical pain in some studies (Esteve, Marquina-Aponte, & Ramírez-Maestre, 2014; Rabbitts et al., 2015). Thus, it was expected that parent pain catastrophizing would emerge as a predictor of postsurgical pain trajectories. Findings of study one did not support this hypothesis, which suggests that the role of parent pain catastrophizing may be somewhat different in a postsurgical setting. Results of another study published using the same dataset as was used in this dissertation suggest that parent pain catastrophizing is a dynamic construct subject to change over time (Birnie, Chorney, & El-Hawary, 2017). The IFAM states that child and parent cognitions reciprocally interact, in part, through child pain experience and pain expression and parent interpretation. Thus it is possible that as a child's postsurgical pain fails to resolve and persists over time, parent pain catastrophizing may reciprocally be influenced and thus a stronger relation between parent pain catastrophizing and child pain may be observable at a later point in the postsurgical recovery period.

Whereas part of this dissertation examines pediatric postsurgical pain and functioning from a risk-based perspective, postsurgical recovery is also considered and examined from a resilience perspective. Resilience can be defined as effectively negotiating, adapting to, or managing significant sources of stress or trauma by utilizing protective factors within the individual or their environment (Masten, 2001; Masten, 2018; Windle, 2011). Variable-focused models of resilience propose that protective factors can impact a resilience outcome either directly (as a main effect) or indirectly (as a mediator or moderator). Results of paper three

provide support for these approaches to studying resilience and demonstrate that a single protective factor may impact resilience outcomes in pediatric postsurgical recovery through both main effects and as a moderator. In the chronic postsurgical period, a main effect of optimism was found for quality of life while a moderator effect was found for optimism on the relation between pain and functional disability. These findings also suggest that a protective factor can be associated with more than one resilient outcome but that the relations between the protective factor and each outcome may be different. While not tested in the current dissertation, it is possible that optimism and other protective factors may function as mediated moderators. For example, optimism was found to moderate the relation between pain and functioning in this dissertation but other studies suggest that optimism may have this effect via lowered pain catastrophizing (Coronado et al., 2017; Cousins et al., 2015).

Findings from this dissertation also speak to the concept of adaptability inherent in the definition of resilience. Paper three found that optimism, a proposed protective factor, was related to resilient outcomes in the chronic postsurgical period but that it was not related to the same resilient outcomes in the acute postsurgical period. These findings suggest that there may be different and more salient protective factors that help an adolescent to adapt while experiencing a more acute stressor like acute postsurgical pain, but that different protective factors may become useful once the stressor has become chronic.

Findings from paper two also have implications for a debate within the resilience theory literature. The concept of resilience and the way that it is studied has been criticized for placing too much emphasis on the individual and individual interventions and not enough emphasis on the social structure and wider social forces that impact an individual's ability to be resilient and to demonstrate resilient outcomes (Collins, 2017; Garrett, 2016; Singh & Cowden, 2015; Ungar,

2020). These critics argue that such a focus makes the concept of resilience vulnerable to being co-opted by a neo-liberal agenda aimed at responsibilizing individuals for coping, competence and success. Instead of individual interventions, they argue that radical structural change should be the target of study and intervention in resilience research. Paper two attempts to address this criticism by reviewing factors beyond the individual (i.e., family and peer factors) that may be protective and lead to resilient outcomes, but the lack of research on more distal factors (i.e., community, cultural) to include in the review supports the criticism that resilience research tends to focus on the individual. Responders to these critics, many of whom conduct their research within the discipline of social work, also point out that a pure focus on structural factors that impact resilience is at odds with the foundational social work notion of, 'person-in-environment'(Harrison, 2013; Van Breda, 2018; 2019). The person in environment principle states that both people and structures must be engaged to address life challenges and enhance wellbeing (IFSW, 2014). In her critical response, Van Breda (2019) cites several papers that approach the study of resilience with the person-in-environment principle in mind and that conclude the mere presence of social structures is insufficient to promote well-being but rather the interaction between individual agency characteristics and social structure is the key to understanding resilient outcomes. While paper two does not propose a resilience model for postsurgical outcomes, the inclusion of both individual and social factors as well as some discussion about the interaction between them lays the groundwork for an ecological postsurgical resilience model consistent with the person-in-environment principle.

Lastly, findings from paper three also have implications for the way optimism is conceptualized. The construct of optimism encompasses two separate, but theoretically linked schools of thought. One defines optimism as generalized favorable expectancies for the future

(Scheier & Carver, 1985), while the other defines optimism as individual differences in how people habitually explain good and bad events they encounter in life (Gillham et al., 2002). The first definition is commonly referred to as, 'dispositional optimism' while the other is referred to as, 'explanatory style or attributional style.' The vast majority of research that has examined optimism's relation to health outcomes has utilized a definition of optimism reflective of dispositional optimism (Rasmussen et al., 2009). In contrast, paper three of this dissertation utilized a measure of optimism that reflects the explanatory style of optimism.

Explanatory Style Optimism emerged from research on Reformulated Learned Helplessness Theory and the Hopelessness Theory of Depression (Reivich & Gillham, 2003). The Reformulated Learned Helplessness Theory states that an individual's causal attributions about uncontrollable events directly impact cognitive, emotional and motivational processes and can lead to feelings of helplessness (Abramson et al., 1978). The Hopelessness Theory described explanatory style as a risk factor within a diathesis-stress model of depression emphasizing how stable and global explanations for negative events yield hopelessness (Abramson et al., 1989). Unsurprisingly, much of the literature on explanatory style has examined it as a risk factor rather than a protective factor and much of this research has been conducted looking at mental health outcomes (Alloy, Lipman, & Abramson, 1992; Calvete, Villardón, & Estévez, 2008; Carter & Garber, 2011; Park, Bang, Kim, Lee, & An, 2018). Findings from this dissertation provide some evidence to support the application of the explanatory style definition of optimism in a physical health context as well as for it to be conceptualized as a protective factor.

### 5.3 Clinical Implications

Findings from this dissertation have several important clinical implications. First, findings from paper one underscore the importance of measuring and managing both baseline

pain as well as acute postsurgical pain for the prevention of chronic postsurgical pain. It is still not clear whether high levels of postsurgical pain observed in individuals who go on to develop chronic postsurgical pain is new onset pain due to the surgery or if it is instead an exacerbation of existing pain that was present before surgery. More research is needed to disentangle the complex relation between baseline pain and immediate postsurgical pain. Nevertheless, it seems important to assess pain at both of these timepoints so that it can be properly treated.

Findings from this dissertation also highlight the importance of understanding pain as more than just a nocioceptive experience. There are several psychosocial aspects to the experience of pain that should be examined when considering targets for intervention. Findings from paper one, which are consistent with adult postsurgical literature, demonstrate that baseline anxiety is likely an important target for intervention in order to prevent the development of chronic postsurgical pain. A pre-surgical intervention that used cognitive-behavioral therapy principles to treat anxiety (e.g., identifying and challenging maladadaptive thoughts) could be used to reduce presurgical anxiety and provide individuals identified to have anxiety with the opportunity to build skills that could be used to manage their postsurgical anxiety. Contrary to findings in the adult postsurgical literature, paper one of this dissertation did not observe baseline pain catastrophizing to be a predictor of pain trajectory membership. While this dissertation did not examine the ability of *postsurgical* pain-catastrophizing to predict pain trajectory membership, findings from other studies suggest that pain catastrophizing may not be as stable a construct in youth as in adults (Birnie, Chorney, & El-Hawary, 2017). Consequently, it may be more effective to target pain catastrophizing beliefs in the postsurgical period rather than the baseline or presurgical period.

Along with psychosocial risk factors, findings from this dissertation also point to the importance of considering factors that may be protective for pediatric postsurgical recovery. Paper two reviewed several factors at the individual, family and peer levels that warrant further examination in a pediatric postsurgical context and findings from paper three provided support for optimism as a protective factor for functional outcomes one year after surgery. While optimism is generally considered to be trait-like, interventions aimed at improving optimism do exist and have proven effective in increasing levels of optimism (Meevissen, Peters, & Alberts, 2011; Peters, Flink, Boersma, & Linton, 2010; Seligman, Schulman, & Tryon, 2007; Traxler, Hanssen, Lautenbacher, Ottawa, & Peters, 2019). Thus, interventions aimed at preventing poor postsurgical outcomes should include ways of improving protective factors such as optimism as well as mitigating risk factors such as anxiety.

In terms of applying the above-mentioned findings to intervention, they could be used to inform the development and testing of a pediatric transitional pain service. Transitional pain services have been described in the adult postsurgical literature (Jensen, Stubhaug, & Breivik, 2016; Katz et al., 2015, 2019; Tiippana et al., 2016). These services aim to offer timely and effective treatment to patients deemed to be at high risk for developing chronic postsurgical pain. This is accomplished by providing both pre and postsurgical pain management for high risk patients and providing services to improve coping and functioning in both the immediate and long-term postsurgical period.

## 5.4 Strengths and Limitations

### 5.4.1 Prospective Longitudinal Research Design.

The prospective longitudinal design utilized to generate data for papers one and three of this dissertation is a strength as it conveys several advantages over other designs. First, unlike

retrospective studies, the data is not subject to participant recall bias nor is it limited by what might have been gathered in the patient charts at several different healthcare centres. This design also allowed for the measurement of several different a priori baseline variables that could be controlled for as well as examined as predictors of postsurgical outcomes across the postsurgical recovery period. Perhaps most importantly, this design allowed us to see how pain changed over the postsurgical period for different groups of individuals and how pain and psychosocial variables related to functioning at different points in the postsurgical period.

Along with these strengths, there were also some limitations associated with the design that warrant discussion. First, as with all longitudinal designs, this design was subject to data loss mainly through participant attrition over the course of the postsurgical year which can lead to bias regarding the representativeness of the sample. Attempts were made to retain participants during the postsurgical year, however, retention efforts were somewhat limited by ethical considerations aimed at respecting participants rights to drop out of the study. Data for the present dissertation was mainly collected by phone, but may have been more successful if multiple methods that are now available could have been used such as email, text, social media, or a smartphone application specifically designed for the study. In order to appropriately account for the missing data and minimize bias, statistical analysis techniques based on expectation-maximization algorithms (i.e., multiple imputation, full-information maximum likelihood methods) were used in papers one and three. These methods are strongly recommended for the analyses of longitudinal data in order to minimize bias due to participant attrition and other causes of data loss (Graham, 2009). However, given the novelty of these techniques, particularly in some academic disciplines, the strength and utility of these techniques may be underestimated.

Another limitation associated with the prospective longitudinal design concerns the postsurgical period and the amount of time needed to collect the data. Conclusions drawn from this dissertation about pediatric postsurgical recovery are limited to the first postsurgical year; Based on studies that have examined postsurgical outcomes beyond the first year (Chidambaran et al., 2017; Sieberg et al., 2013), it is likely that postsurgical pain and functioning continued to evolve in this cohort and thus conclusions made from this dissertation may not apply in a later postsurgical period.

### 5.4.2 Multi-Centre Data Collection

The data utilized for papers one and three of this dissertation were collected at eight different children's health centres across Canada as part of a multidisciplinary research endeavor to better understand the postsurgical recovery experience of children after major spinal surgery. This recruitment strategy is unique in the literature as other comparable studies recruited participants from a single institution (Connelly et al., 2014; Rabbitts et al., 2015; Rosenbloom et al., 2019; Sieberg et al., 2013). A multi-center recruitment strategy conveys several benefits including the ability to collect a more diverse sample than what would have likely been obtained if data were collected at a single institution. A multi-center recruitment strategy also allowed for the collection of one of the largest Canadian datasets of scoliosis surgery patients. While a multi-centre study conveys these benefits it also introduces a challenge for a cohort study in that differences in institutional practices could confound the findings. However, any potentially confounding differences can be examined in preliminary analyses and accounted for by inclusion of study site as a covariate in subsequent analyses.

### 5.4.3 Measurement of Postsurgical Outcomes

The importance of measuring patient-oriented outcomes such as pain and quality of life only gained recognition within the last twenty years and has since become more common in medical discipline research (Dworkin et al., 2005; Wright, 1999). The findings from this dissertation contribute to the growing body of postsurgical literature that measures patient-oriented outcomes including pain, functional disability and quality of life. These outcomes were measured at several time points during the postsurgical period which, as mentioned above, provided information about how they change over time. While the inclusion of these measures is certainly a strength and a contribution to the postsurgical literature, there have been several advances in the field of pain research that point to some limitations of the outcome measures used in this dissertation. The primary measure of pain was self-reported average pain in the last 24 hours using a numerical rating scale. While self-report of pain, when it is possible, is regarded as a primary aspect of children's pain assessment upon which to base clinical care (Twycross, Voepel-Lewis, Vincent, Franck, & Von Baeyer, 2015), this particular measure does not capture other aspects of pain such as unpleasantness, activation (i.e., with movement), or quality (i.e., burning, aching) which may be important for predicting chronicity when measured in an acute pain context (Jaaniste et al., 2019; Pagé, Campbell, et al., 2013).

### 5.4.4 Protective Factors and Postsurgical Outcomes

A strength of this dissertation is its attention to protective factors in postsurgical recovery; thus, contributing to a more comprehensive understanding of the recovery process and aiding in development of intervention programs for those at risk of poor postsurgical outcomes. The topical review in paper two provides an up-to-date overview of an area that is rapidly developing and provide some future directions to guide further development in the field (Palermo, 2013). A topical review was an appropriate starting point, as protective factors and

resilience has only just begun to garner attention in pediatric pain and there is little to no research in pediatric postsurgical recovery. Although paper two provides a novel and important contribution to the literature, it is limited in its ability to make strong conclusions founded on exhaustive and rigorous methodology as is typical in other types of reviews (i.e., systematic reviews, scoping reviews). Additionally, while paper two reviews some protective factors outside the individual (i.e., parent factors, peer factors), there is a distinct lack of literature to review on broader factors at the community, cultural and societal level that could also be protective for adolescent postsurgical outcomes. The World Health Organization defines social determinants of health as the conditions in which individuals are born, grow, live, work and age that are shaped by the distribution of money, power and resources. The research in this dissertation was conducted with a sample of predominantly Caucasian adolescents with well-educated parents accessing publicly funded healthcare in a high-income country. Similar research conducted in a different setting with a more diverse sample may yield different results and future research should consider examining protective factors at broader levels that reflect social determinants of health in addition to individual, parent and peer factors.

### 5.5 Future Directions

Findings of this dissertation suggest several future directions. First, future studies should attend to measuring predictor variables early in the acute postsurgical period and how they relate to chronic postsurgical outcomes. Pain in the acute postsurgical period is an important factor to assess, particularly over the course of several days. Studies in the adult postsurgical literature have begun to examine acute pain trajectories as predictors of chronic postsurgical pain (Althaus, Arránz Becker, & Neugebauer, 2014; Althaus et al., 2018), but this has not yet been examined in pediatric postsurgical patients. Future studies should examine both the levels of pain in the

immediate postsurgical period as well as the rates at which they resolve in the acute postsurgical period as predictors of chronic postsurgical pain. Furthermore, assessment tools that capture other important aspects of pain such as unpleasantness and quality of pain should be used in postsurgical pain research as these other aspects of pain may be important for predicting chronicity (Jaaniste et al., 2019; Pagé, Campbell et al., 2013).

In addition to pain, functional outcomes should also continue to be assessed in the acute postsurgical period. Models of chronic pain development and maintenance describe a cycle of pain, cognitions, behavior and functioning as inter-related and self-perpetuating (Vlaeyen & Linton, 2000; Asmundson et al., 2012; Simons & Kaczynski, 2012). Understanding how pain and functioning in the acute postsurgical period relates to pain and functioning in the chronic postsurgical period is important for both patients and clinicians. Information about acute pain and functioning can be used to better prepare adolescents for what to expect as they begin to re-engage in important activities (e.g., school, social events). Information about acute pain and functioning as they relate to chronic postsurgical outcomes can also be used to inform interventions for individuals identified to be at risk of poor postsurgical outcomes.

Much more research is needed to identify and examine the role of protective factors in pediatric postsurgical recovery. Findings from the pediatric chronic pain literature provide a useful starting point, but this work does not speak to potential differences in the role of protective factors in acute and chronic postsurgical period. Findings from paper three of this dissertation suggest that at least one factor, optimism, is predictive of resilient outcomes in the chronic postsurgical period, but not in the acute postsurgical period. More studies are needed to examine other protective factors relevant to pediatric postsurgical recovery. Once more studies begin to examine both risk and protective factors in pediatric postsurgical recovery, a more

systematic approach to reviewing studies such as scoping review could be done to begin building a theoretical model of pediatric postsurgical recovery.

Paper three of this dissertation used a variable-focused approach to preliminarily examine the relation between optimism, a protective factor, and resilient outcomes in pediatric postsurgical recovery. However, other approaches to studying resilience could also be used to understand protective factors that are important. For example, person-centred approaches to the study of resilience can be used to identify comparative groups of individuals who experience similar levels of risk but who show different patterns of adaptation (e.g., aggregated case studies, trajectories of resilience outcomes). Researchers can then compare these groups on multiple criteria to determine protective factors that may lead resilient outcomes. Given the differences in he acute versus chronic postsurgical period observed in paper three, it may be useful to attempt to examine resilient outcomes using a person-focused approach that observes how resilient outcomes change over time.

In order to further knowledge in the field of pediatric postsurgical recovery, it is important that data are collected in a way that allow for comparison across studies. Agreement and use of a standard set of outcome measures would help to achieve this goal. The Scoliosis Research Society patient questionnaire is one example of a patient-reported outcome measure that has been widely adopted. While this measure has allowed for ease of comparison across studies investigating postsurgical outcomes for scoliosis patients, its specificity limits its generalizability to other pediatric major surgery populations. The Patient Reported Outcomes Measurement Information System (PROMIS), an initiative by the National Institutes of Health, may be a more useful set of outcome measures to adopt for postsurgical research. Developers of the PROMIS recognized there was a lack of standardization in patient-reported outcomes which

did not allow for meaningful comparison across trials or populations, often resulted in a burden to patients who would be required to fill out multiple measures, and that measures were not necessarily sensitive to change (Cella et al., 2007). In order to address these limitations, PROMIS was developed using a rigorous mixed methods approach which has resulted in several item banks for use across adult and child health conditions (Cella et al., 2007; DeWalt, Rothrock, Yount, & Stone, 2007; Reeve et al., 2007). Several PROMIS domains have been validated for use in pediatric chronic pain (Kashikar-Zuck et al., 2016) and a scoping review of 21 adult surgical studies that used PROMIS domains found that they performed efficiently, accurately and reliability across surgical types. Additionally, a recent study that looked at postsurgical outcomes across both adult and pediatric spinal deformity patients found strong to moderate correlations between SRS questionnaire outcomes with corresponding PROMIS domains as well as reduced ceiling and floor effects on the PROMIS as compared to the Scoliosis Research Society questionnaire (Bernstein et al., 2019).

## 5.6 Conclusion

In conclusion, this dissertation sought to further our understanding of postsurgical pain and functioning following major pediatric surgery and examine predictors of recovery from a resilience perspective. The first paper identified three distinct pain trajectories with the majority of the sample demonstrating good pain resolution and functioning by one year and a small subset of the sample following a trajectory characterized by high immediate postsurgical pain that persisted over the course of the first postsurgical year. This trajectory was predicted by baseline pain and anxiety and individuals classified in this trajectory had worse functional outcomes at one year as compared to individuals in the other trajectories. The second paper is a topical review that provides a rationale for identifying and examining protective factors in pediatric

postsurgical recovery, reviews the most recent literature on protective factors relevant to pediatric postsurgical recovery and recommends directions for future research. Finally, the third paper examined one protective factor, optimism, in more depth by examining its moderating effects on the relation between pain and functional outcomes at two time in the postsurgical period. Taken together the findings of this dissertation demonstrate that a small subset of adolescents are at risk of developing chronic pain and poor functioning but that the vast majority of adolescents recover well from major surgery. Future research should study not only the factors that put adolescents at risk of poor postsurgical outcomes, but also the factors that promote good postsurgical outcomes so that they can be used to inform effective interventions.

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## APPENDIX A: TOPICAL REVIEW SEARCH STRATEGY DETAILS

| OVERVIEW | |
|---|---|
| Databases: | PubMed – MEDLINE<br>PsycINFO<br>CINAHL |
| Date of Search: | April 13 & 22, 2020 |
| Limits: | Date limit: 1990 – 2020<br>Language limit: English language |

| SYNTAX GUIDE | |
|---|---|
| MeSH | Medical Subject Heading |
| * | Before a word, indicates that the marked subject heading is a primary topic; or, after a word, a truncation symbol (wildcard) to retrieve plurals or varying endings |
| TI | Title |
| AB | Abstract |

| MULTI-SEARCH STRATEGY | |
|---|---|
| Concept | Search Terms |
| Children/Adolescents/Young Adults | adolescent*[TI] OR teen*[TI] OR "young-adult"[TI] OR "young-adults"[TI] OR youth*[TI] OR girl[TI] OR girls[TI] OR boy[TI] OR boys[TI] OR preteen*[TI] OR "pre-teen"[TI] OR "pre-teens"[TI] OR "school-aged"[TI] OR "high-school"[TI] OR "secondary-school"[TI] |
| | OR |
| | "Adolescent"[MeSH] OR "Adolescent Development"[MeSH] OR "Adolescent Health Services"[MeSH] OR "Adolescent, Institutionalized"[MeSH] OR "Adolescent, Hospitalized"[MeSH] OR "Psychology, Adolescent"[MeSH] OR "Adolescent Psychiatry"[MeSH] OR "Adolescent Medicine"[MeSH] OR "Adolescent Behavior"[MeSH] OR "Adolescent Health"[MeSH]) |
| Surgery | surgery[Title] OR surgeries[Title] OR "major surgery"[Title] OR "major surgeries"[Title] OR "spine surgery"[Title] OR "spinal surgery"[Title] OR "spine surgeries"[Title] OR "spinal surgeries"[Title] OR "scoliosis surgery"[Title] OR "scoliosis surgeries"[Title] OR "spinal fusion"[Title] OR "complex surgery"[Title] OR "complex surgeries"[Title] OR "facial surgery"[Title] OR "facial surgeries"[Title] OR "orthognathic surgery"[Title] OR "orthognathic surgeries"[Title] OR "bariatric surgery"[Title] OR "bariatric surgeries"[Title] OR "obesity surgery"[Title] OR "obesity surgeries"[Title] |

| | |
|---|---|
| | OR |
| | "surgery"[MeSH] OR "bariatric surgery"[MESH] OR "bariatric surgeries"[MESH] OR “surgeries, bariatric”[MESH] OR “surgery, bariatric”[MESH] OR "scoliosis"[MESH] OR "spinal fusion"[MESH] OR “spinal fusion/instrumentation”[MeSH] OR “spinal fusion/psychology”[MeSH] OR “spinal fusion/rehabilitation”[MeSH] OR “spine/surgery”[MeSH] OR "spinal fusions"[MESH] OR “orthognathic surgeries”[MESH] OR “orthognathic surgery”[MESH] OR “orthognathic surgeries, maxillofacial”[MESH] OR “orthognathic surgery, maxillofacial”[MESH] OR “maxillofacial orthognathic surgery”[MESH] OR “orthognathic surgical procedure”[MESH] OR “orthognathic surgical procedures”[MESH] OR “procedure, orthognathic surgical”[MESH] |
| | OR |
| | ("post-operative"[Title] OR postoperative[Title] OR "post operative"[Title] OR "post-surgical"[Title] OR post-surgical[Title] OR "post surgical"[Title] OR "surgical recovery"[Title] OR "post-operative recovery"[Title] OR "post operative recovery"[Title] OR "post-surgical recovery"[Title] OR "post surgical recovery"[Title] OR "after surgery"[Title] OR "after operation"[Title]) |
| Pain, Chronic Pain | pain[Title/Abstract] OR "chronic pain"[Title/Abstract] OR "persistent pain"[Title/Abstract] OR "post-surgical pain"[Title/Abstract] OR "post surgical pain"[Title/Abstract] OR "neuropathic pain"[Title/Abstract] OR "arthritis"[Title/Abstract] OR "musculoskeletal pain"[Title/Abstract] OR "widespread pain"[Title/Abstract] OR “pain psychology” OR “postoperative pain” OR “postoperative rehabilitation” |
| | OR |
| | “pain”[MeSH Terms] OR “pain/prevention and control”[MeSH Terms] OR “pain/psychology”[MeSH Terms] OR “pain/rehabilitation”[MeSH Terms] OR “pain/surgery”[MeSH Terms] OR “pain management”[MeSH Terms] OR “pain management/psychology”[MeSH Terms] OR “pain threshold”[MeSH Terms] OR “pain threshold/psychology”[MeSH Terms] OR “pain, postoperative”[MeSH Terms] “pain, postoperative/prevention and control”[MeSH Terms] OR “pain, postoperative/psychology”[MeSH Terms] OR “pain, postoperative/surgery”[MeSH Terms] OR “chronic pain”[MeSH Terms] OR “chronic pain/prevention and |

| | |
|---|---|
| | control”[MeSH Terms] OR “chronic pain/psychology”[MeSH Terms] OR “chronic pain/surgery”[MeSH Terms] |
| Postsurgical Outcomes | recovery[Title] OR "physical recovery"[Title] OR function[Title] OR "physical function"[Title] OR "physical ability"[Title] OR "functional ability"[Title] OR "return to activities"[Title] OR "return to activity"[Title] OR "daily living"[Title] OR "activities of daily living"[Title] OR "ADLs"[Title] |
| | OR |
| | “recovery of function”[MeSH] OR “activities of daily living”[MeSH] OR “activities of daily living/psychology”[MeSH] OR “return to sport”[MeSH] OR “return to sport/psychology”[MeSH] |
| | OR |
| | "psychological growth"[Title] OR "personal growth"[Title] OR "self-knowledge"[Title] OR "post-traumatic growth"[Title] OR "posttraumatic growth"[Title] OR "benefit finding"[Title] |
| | OR |
| | "psychological growth"[Title/Abstract] OR "personal growth"[Title/Abstract] OR "self-knowledge"[Title/Abstract] OR "post-traumatic growth"[Title/Abstract] OR "posttraumatic growth"[Title/Abstract] OR "benefit finding"[Title/Abstract] |
| | OR |
| | "quality of life"[Title] OR QoL[Title] OR "mental health"[Title] OR "mental wellness"[Title] OR "psychological distress"[Title] OR "self-efficacy"[Title] OR "self confidence"[Title] OR "confidence"[Title] OR "self-esteem"[Title] OR satisfaction[Title] OR social[Title] OR peers[Title] OR "peer relations"[Title] OR “peer group”[Title] OR school[Title] OR "return to school"[Title] OR academics[Title] OR “academic performance”[Title] OR “academic success”[Title] OR leisure[Title] OR "friends"[Title] |
| | OR |
| | resilience[Title] OR resilient[Title] OR resiliency[Title] OR adapt[Title] OR adapting[Title] OR adaptation[Title] OR adapted[Title] OR flexible[Title] |
| | OR |
| | “resilience, psychological”[MeSH] OR “resilience, psychological/classification”[MeSH] OR “adaptation, psychological”[MeSH] |

| | |
|---|---|
| Protective Factors | "positive psychology"[Title/Abstract] OR acceptance[Title/Abstract] OR "body image"[Title/Abstract] OR optimism[Title/Abstract]] OR "explanatory style"[Title/Abstract] OR "attribution style"[Title/Abstract] OR "psychological flexibility"[Title/Abstract] OR "values-based"[Title/Abstract] OR "values"[Title/Abstract] OR "social support"[Title/Abstract] OR "parenting style"[Title/Abstract] OR "parent support"[Title/Abstract] OR "community"[Title/Abstract] OR "peer support"[Title/Abstract] OR “peers”[Title/Abstract] OR “peer group”[Title/Abstract] OR "empathy"[Title/Abstract] OR “attachment”[Title/Abstract] OR “socioeconomic status”[Title/Abstract] OR “SES”[Title/Abstract] OR “income”[Title/Abstract] OR “income level”[Title/Abstract] OR “ethnicity”[Title/Abstract] OR “race”[Title/Abstract] OR “culture”[Title/Abstract] OR “neighbourhood”[Title/Abstract] OR “built environment”[Title/Abstract] OR “housing”[Title/Abstract] |
| | OR |
| | “psychology, positive”[MeSH] OR “acceptance and commitment therapy”[MeSH] OR “body image”[MeSH] OR “body image/psychology”[MeSH] OR “optimism”[MeSH] OR “optimism/psychology”[MeSH] OR “parenting”[MeSH] OR “parenting/psychology”[MeSH] OR “social support”[MeSH] OR “peer group”[MeSH] OR “peer influence”[MeSH] OR “empathy”[MeSH] |