

University of Alberta

**A PLURALIST CHARACTERIZATION OF PAIN MEANING AFTER
WHIPLASH**

by

GEOFFREY PAUL BOSTICK

A thesis submitted to the Faculty of Graduate Studies and Research
in partial fulfillment of the requirements for the degree of

DOCTOR OF PHILOSOPHY

in

REHABILITATION SCIENCE

FACULTY OF REHABILITATION MEDICINE

©Geoffrey Paul Bostick
Spring 2012
Edmonton, Alberta

Permission is hereby granted to the University of Alberta Libraries to reproduce single copies of this thesis and to lend or sell such copies for private, scholarly or scientific research purposes only. Where the thesis is converted to, or otherwise made available in digital form, the University of Alberta will advise potential users of the thesis of these terms.

The author reserves all other publication and other rights in association with the copyright in the thesis and, except as herein before provided, neither the thesis nor any substantial portion thereof may be printed or otherwise reproduced in any material form whatsoever without the author's prior written permission

DEDICATION

This thesis is dedicated to my family:
My parents and brother who've supported me in every one of my endeavours and
given me every opportunity to succeed,
my loving wife Danielle who has been a constant source of encouragement,
and my beautiful daughter Quin who reminds me what is important in life.

ABSTRACT

The experience of pain is multidimensional. Biological, psychological and social factors combine to inform the experience of pain. This bio-psychosocial view has become popular given shortcomings of the pervasive medical model in accounting for pain persistence in conditions such as whiplash associated disorder (WAD). However, the majority of research has examined components of this model separately. Furthermore, important pain beliefs have not been explored in the context of WAD. In order to better understand the experience of WAD, the objective of this thesis was to examine the experience of WAD-related pain beliefs with a pluralist and integrated approach. Biological and social factors are investigated in relation to WAD-related pain beliefs using multiple methods reflecting the multidimensional experience of pain.

Three chapters principally addressed the objective of the thesis. In chapter two, a narrative review of placebo hyperalgesia is presented illustrating the biological effects of belief. Neurochemical and neuroanatomical changes were shown to coincide with altered or manipulated expectations of pain. In chapter three, WAD-related pain beliefs were examined in a longitudinal mixed-method study. Initially, beliefs are explored quantitatively then more deeply with a qualitative methodology. This qualitative piece illuminated a shared inter-subjective meaning of WAD-related pain beliefs reflecting a desire to be cured or fixed. This overarching theme underscores an adaptive perspective of pain early after WAD, but becomes maladaptive as pain persists. In chapter four, the prognostic value of, and

inter-relationships among, beliefs was examined. WAD-related pain beliefs were found to be related to catastrophizing, which is consistent with theoretical assertions relating pain and catastrophizing. In addition, early WAD-related beliefs pertaining to negative expectations, catastrophizing and mystery were associated with future pain and disability. Finally, considering data collection challenges in longitudinal observation studies, chapter five was devoted to exploring these issues and proposing recommendations for future studies.

This thesis builds on growing literature highlighting the importance of cognitive factors shaping the experience of WAD. More importantly, by examining WAD-related pain beliefs from divergent perspectives the multi-dimensional nature of pain is highlighted. This pluralist approach supports the use of multiple approaches to examining pain within a study or research program.

ACKNOWLEDGEMENTS

I would like to sincerely thank my doctoral supervisors, Drs. Douglas Gross and Linda Carroll. Both have provided a depth of wisdom and tutelage that has been an important source of success throughout my doctoral work. I would particularly like to acknowledge Dr. Gross' flexibility and availability as I navigated through the doctoral program. I am also grateful for the support of Drs. Cary Brown and Dwight Harley whom have donated their valuable time and knowledge. I am also thankful for Dr. Peter Rothe's early influence in facilitating exploration of subject matter far beyond my level of comfort. I would also like to thank Dr. Lauren Beaupre who provided me opportunities to gain valuable research experience early in my training.

I would like to thank Angela Libutti whose professionalism has made traversing through the various doctoral administrative obstacles a breeze.

I am also thankful for Drs. Paul Halger and Joanne Volden's leadership and support as Associate Dean of Graduate Studies and Research.

I would also like to acknowledge the other doctoral and masters students in the Rehabilitation Medicine graduate program for their support.

I am also grateful for the support and understanding of Dr. Bob Haennel who has provided me with shelter as a new faculty member in order to facilitate successful completion of my thesis.

Finally, I am appreciative for the financial support from the Department of Physical Therapy, the Faculty of Rehabilitation Medicine, the University of Alberta, and the Canadian Institutes of Health Research.

TABLE OF CONTENTS

Chapter 1	Introduction	Page
1.1.	Overview	1
1.2.	WAD as a model for the study of pain beliefs.....	3
1.3.	A biological conceptual framework of pain	5
1.4.	A cultural conceptual framework of pain	7
1.5.	A biocultural conceptual framework of pain	11
1.6.	Cognitive-behavioural approach to pain	14
1.7.	Epistemological/Ontological stance for the thesis: Pragmatism	17
1.8.	Study purposes	21
1.9.	References	25
Chapter 2	Meaning modulation through nocebo hyperalgesia: the biological plausibility of belief	
2.1.	Introduction	30
2.2.	Descending pain modulation	32
2.3.	The Nocebo Hyperalgesic Meaning Response: an example of modulation via expectations	34
2.4.	Relevance of the Nocebo Hyperalgesic Meaning Response for Physical Therapists	41
2.5.	Conclusions	44
2.6.	References	45
Chapter 3	<i>If they can put a man on the moon they should be able to fix a neck injury: A mixed-methods study characterizing and explaining pain beliefs about WAD</i>	
3.1.	Introduction	51
3.2.	Methodology	
3.2.1.	Theoretical position	53

3.2.2.	Research question	55
3.2.3.	Design	55
3.2.4.	Participants	56
3.2.5.	Survey measures	57
3.2.6.	Interviews	58
3.2.7.	Data analysis	59
3.3.	Results	
3.3.1.	Sample characteristics	60
3.3.2.	Characterization of WAD-related pain beliefs	61
3.3.3.	Thematic analysis of experiences explaining endorsed beliefs	64
3.4.	Discussion	75
3.5.	References	81

Chapter 4 Predictive capacity of WAD-related pain beliefs and catastrophizing

4.1.	Introduction	89
4.2.	Methodology	
4.2.1.	Design	90
4.2.2.	Participants	90
4.2.3.	Measures	91
4.2.4.	Analysis	93
4.3.	Results	
4.3.1.	Sample characteristics	94
4.3.2.	Internal consistency	95
4.3.3.	Associations between beliefs and catastrophizing	97
4.3.4.	Baseline prediction of pain intensity at three and six months post-MVC	97
4.3.5.	Baseline prediction of disability at three and six months post-MVC	98
4.4.	Discussion	101

4.5.	References	105
------	------------------	-----

Chapter 5 Lessons learned: maximizing response and minimizing attrition

5.1.	Introduction	110
5.2.	Potential consequences related to response and attrition bias	110
5.3.	Mitigating response and attrition bias	
5.3.1.	Methodological strategies mitigating response bias	112
5.3.2.	Methodological strategies mitigating attrition bias	117
5.3.3.	Analytical strategies mitigating response bias	119
5.3.4.	Analytical strategies mitigating attrition bias	120
5.4.	Perceived barriers to maximizing response	123
5.5.	Lessons learned	125
5.6.	Conclusions	127
5.7.	References	129

Chapter 6 General discussion and conclusions

6.1.	Overview	132
6.2.	The nocebo hyperalgesic meaning response	133
6.3.	An integrated mixed-method characterization of WAD-related beliefs	134
6.4.	WAD-related pain beliefs associations with catastrophizing outcomes and six-month post-MVC	134
6.5.	A pluralistic conceptual model of WAD-related pain beliefs ...	135
6.5.1.	Restitution	138
6.5.2.	Baseline pain and disability predict future pain and disability	140
6.5.3.	Expectations in the meaning of WAD-related pain	140
6.5.4.	Catastrophizing in the meaning of WAD-related pain	144
6.6.	Conclusions	146
6.7.	References	148

Appendix A: Researcher reflexivity	151
Appendix B: Sample interview guide	155
Appendix C: Details of trustworthiness analysis	159

LIST OF TABLES

	Page
Table 1-1	Assumptions of the Cognitive-behavioural approach to pain management17
Table 1-2	General characteristics of pragmatism20
Table 3-1	Sample description and characterization of pain intensity, disability and beliefs over time62
Table 3-2	Characteristics of the interview sub-sample63
Table 3-3	Sub-themes generated from the qualitative analysis along with supportive verbatim quotes72
Table 4-1	Sample characteristics at baseline96
Table 4-2	Correlations between specific beliefs and catastrophizing97
Table 4-3	Univariate and multivariate associations between baseline beliefs and pain intensity 3 and 6 months post-MVC99
Table 4-4	Univariate and multivariate associations between beliefs and self-reported disability 3 and 6 months post-MVC100
Table 5-1	Methodological strategies advocated in literature and those employed to mitigate selection bias114
Table 5-2	Comparison of the study sample with a large comparable patient sample120
Table 5-3	Comparison of demographic and beliefs variables between participants with incomplete and complete data121

LIST OF FIGURES

	Page
Figure 1-1 Akparian et al's ¹⁸ working model of chronic pain	7
Figure 1-2 Kirmayer's ²⁰ illustration of cultural influences on physiology, sensation and symptom report	9
Figure 2-1 Illustration of the anatomical structures involved in the descending pain modulation pathway	33
Figure 2-2 Illustration of chemical basis of placebo and nocebo effects on pain due to placebo or nocebo suggestions	36
Figure 2-3 Illustration of the nocebo hyperalgesia mechanism	40
Figure 3-1 Diagram of data collection and analysis sequence for this mixed-methods study	57
Figure 3-2 Visual representation of the findings from the thematic analysis	65
Figure 5-1 Visual representation of participant recruitment	116
Figure 6-1 A conceptual model representing key findings from this thesis characterizing the meaning of WAD-related pain	137

LIST OF SYMBOLS AND ABBREVIATIONS

Whiplash	Neck pain attributed to a motor vehicle collision
WAD	Whiplash Associated Disorder
MVC	Motor Vehicle Collision
SOPA	Survey of Pain Attitudes
PBPI	Pain Beliefs and Perceptions Inventory
GI Pal	Basal Ganglia
Amyg	Amygdala
mPFC	Medial Prefrontal Cortex
s1/s2	Primary/Secondary Somatosensory Cortex
DLPFC	Dorsolateral Prefrontal Cortex
PAG	Periaqueductal Gray
ACC	Anterior Cingulate Cortex
fMRI	Functional Magnetic Resonance Imaging
CCK	Cholecystokinin
NCF	Nucleus Cuneiformis
DLPT	Pontine Tegmentum
Mu-opioid	A type of opioid receptor
ACTH	Adrenocorticotropic hormone
PCS	Pain Catastrophizing Scale
ANOVA	Analysis of Variance
Quan	Quantitative
Qual	Qualitative
WDQ	Whiplash Disability Questionnaire
AB	Province of Alberta
SK	Province of Saskatchewan
NRS	Numeric Rating Scale
CI	Confidence Interval
Tort	A form of insurance law

CHAPTER 1

Introduction

1.1. Overview

Meaning is intrinsic to human pain.¹ Although a banal statement, the implications are vast. It implies context is inseparable from pain. Cartesian dualism suggests the opposite: pain is of direct consequence of nociception. Despite this notion being consistently refuted,^{2,3} it remains a pervasive belief.⁴ Nociception means nothing beyond the transmission of noxious stimuli. It is not until nociception reaches the complex web of neurons within the brain containing everything we know to comprise context (e.g. the self, culture, past experience, belief, etc. both situational and global) does nociception become something perceptible. Thus, nociception is neither necessary nor sufficient to produce pain.⁵ A variety of factors including biology, psychology and context determine whether pain is experienced. As will be demonstrated throughout this thesis, meaning is intertwined with biology, psychology and context.

Though meaning is difficult to characterize, it could be defined as something that connects things; it is a mental representation of possible associations among things, events and relationships.^{6(p.15)} Thus, we might typify the meaning of pain as the mental representation of possible relationships among nociception and all things that comprise context. Pain is therefore individual, yet socially determined. This is supported by literature examining pain across race, gender and culture. For example, African-Americans have demonstrated greater sensitivity to experimental painful stimuli compared to non-Hispanic Caucasians.^{7,8} Females tend to report more frequent and intense pain compared to men.⁹ In Whiplash associated disorders (WAD), the experience of pain is thought to be influenced by varied personal and societal factors.¹⁰

As much as meaning is argued as indispensable, it is also difficult to contemplate its relevance in the context of the pervasive and dogmatic¹¹ medical model. Daniel Moerman¹², in the context of placebo, sheds light on this in his description of ‘the meaning response’: it is “the physiologic or psychological effects of meaning in the origins or treatment of illness; meaning responses elicited after the use of inert or sham treatment can be called the “placebo effect” when they are desirable and the “nocebo effect” when they are undesirable.”^{13(p.472)} The term meaning response was derived from the apparent oxymoron of the placebo effect. Placebos, by definition, are inert. How can something that does nothing have an effect? The important point is that nothing occurs in a vacuum. Blue pills mean something different from red pills, advice from someone in a white coat means something different from someone wearing shorts, and t-shirt.¹² The interpretation of an interaction differs between a practitioner who displays active listening skills from one working in a sterile research context.¹⁴ In all of these examples, the active ingredient is meaning. These data become more compelling due to recent advances in technology. Different meaning contexts lead to measurable responses within the brain¹⁵ and pharmaceuticals have been shown to attenuate or intensify responses related to altered meaning.¹⁶ Clearly, meaning matters.

Expression of belief is one way to elucidate meaning. Beliefs about pain are defined as personally formed and culturally shared understandings of pain.¹⁷ This thesis aims to better understand the meaning of pain after a motor vehicle collision (also known as whiplash associated disorder, WAD) through examination of the biology, psychology and context of WAD-specific beliefs. The rationale for using WAD as a vehicle to study pain beliefs is discussed in section 1.2. In keeping with the ‘bricolage’ theme of this thesis, this chapter provides foundation by discussing biological and cultural conceptual frameworks of pain and then finally an integrative biocultural¹ approach. These conceptual frameworks, in particular the biocultural framework, also serve to integrate the three papers in the thesis. Next, I consider the clinical application of the

biocultural concept by introducing the cognitive-behavioural approach to pain management. The tenants of these frameworks implicate the ontological and epistemological stance of the writer. However, for purposes of clarity my ontological and epistemological stance is made explicit. From this, the chapter concludes with the purpose and research question for each paper.

1.2. WAD as a model for the study of pain beliefs

WAD is an ideal model for studying pain beliefs since it is a common and burdensome condition as well as one that appears to be strongly influenced by beliefs. Data on the incidence of WAD is varied depending on when and where the study was conducted. A systematic review by Holm et al¹⁸ reported cumulative incidence rates of WAD emergency room visits varied among North American and European studies that ranged from 28 to 328 visits per 100,000 inhabitants. More recent studies tended to report higher incidence rates. Holm et al¹⁸ also summarized incidence rates based on insurance claims. They reported cumulative incidence rates of WAD claims from the Canadian provinces of Quebec and Saskatchewan to be 70 and 600 per 100,000 inhabitants respectively in the early to mid-1990s then dipping to 417 per 100,000 in Saskatchewan in the mid-1990s. Somewhat controversially the Saskatchewan WAD cumulative incidence rate, based on claim closure, was reduced further when insurance policy changed from a tort to no-fault system.¹⁹ It is not entirely clear why reported incidence rates are so varied.

The course of WAD has also been reported as varied and not well understood. One reason for this confusion is the prevalence of neck pain in the general population has been found to range between 20 and 40%.²⁰ Thus, it is difficult to know the extent persistent symptoms are due to WAD or neck pain due to other factors. In addition, it is difficult to pool data from studies examining recovery as it is defined and measured in different ways. These factors must be considered when evaluating information on the course of WAD. It is generally accepted that

the majority of recovery occurs within the first three months after the MVC.²¹ After three months, in those with persistent symptoms, little change in self-reported pain and disability occur.²¹ The extent people with WAD report persistent symptoms is not clear, however, Carroll et al²² suggest from their systematic review summarizing the course of WAD that approximately 50% of individuals with WAD report symptoms 1 year after the motor vehicle collision (MVC). In the systematic review by Kamper et al²¹ when defining the course of WAD by recovery, it would appear that their summary of persistent pain 1 year after the MVC may be closer to 40%, though none of these data could be pooled.

Although estimates of the incidence and course of WAD are varied, the literature suggests WAD is a common diagnosis and is often subject to a persistent problem. The burden of WAD is significant. When one considers the direct and indirect costs of WAD, the societal burden has been estimated to be £10 billion in the United Kingdom.²³ The individual burden is also considerable as individuals with non-resolving WAD have been shown to have a reduced capacity for long-term earning.²⁴ This coupled with persistent disability and suffering creates a problem for individuals extending beyond the unpleasantness of persistent pain.

The evidence above suggests WAD is an important topic for research given its personal and societal influence. Given this impact, an abundance of research has been directed at elucidating the cause, persistence and optimal treatment of WAD. Correctly or incorrectly, WAD has become infamous in its ability to create heated debates regarding its origins and merits. The controversy surrounding WAD could be dated back to the late 1800s with the diagnosis of Erichsen's disease (railroad spine) where spinal cord concussions were reported to occur after railroad accidents.²⁵ These injuries were thought to have a poor prognosis leading to litigation against railroad corporations.²⁵ Heated debates ensued questioning the legitimacy of Erichsen's disease. In a similar vein, the legitimacy of WAD has been argued. Some evidence suggests that persistent WAD is culturally

determined¹⁰, other evidence has suggested it is influenced by compensation status¹⁹, various anatomical injuries have been posited²⁶, as well as psychological factors²⁷ and changes in the sensitivity of the nervous system.²⁸ While many of these debates are hidden from public scrutiny, there appears to be a unique belief-set for WAD.²⁹ In addition, various common images of collars and insurance fraud is likely to create a unique meaning of WAD-related pain. This mystery surrounding WAD as well as the history and polarized views on WAD creates an ideal model to explore pain beliefs. Moreover, WAD is an exemplar condition representing the complexities of pain and illness that well suits the biocultural approach to pain presented below.

1.3. A biological conceptual framework of pain

Akparian et al³⁰ offer a perspective of pain that is primarily biological. Since a significant dearth in the understanding of chronic pain remains, this article aimed to propose a working theory of chronic pain based on current understanding. In this important and useful article, the authors began by highlighting the challenges in defining chronic pain. The authors present various inadequate definitions ranging from: pain persisting beyond expected healing timeframes³¹ to pain persisting beyond three or six months. Since signs of peripheral and central sensitization have been shown to occur in animals very early after injury, pain should be defined in terms of these biological changes to the nervous system circuitry rather than expected healing time.³⁰

Akparian et al³⁰ review other frameworks to explain the genesis of chronic pain. They argue that there is little evidence to support anatomical, biomechanical, genetic, psychological, or social explanations for the origin of chronic pain. Instead, they argue for compelling evidence related to brain chemistry, cognition (specifically emotional decision making), and brain morphology and brain activity. They outline ascending and descending neuroanatomical structures that influence pain processing and highlight the importance of the cortex. This

information is primarily based on animal studies and brain imaging using various experimental manipulations. This evidence culminated in a proposed working theory for chronic pain with two fundamental hypotheses:

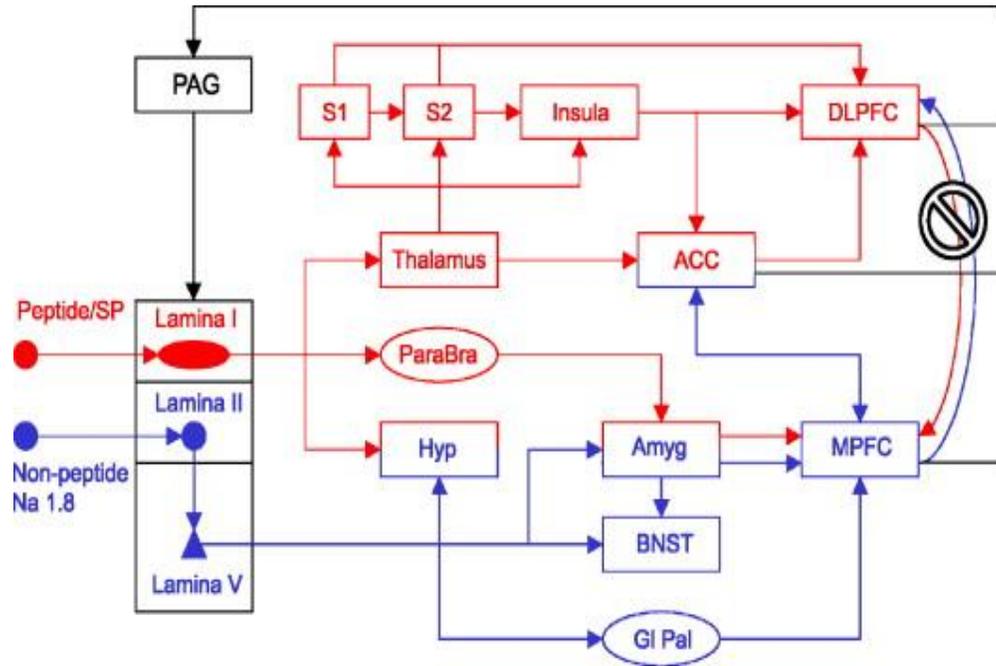
- “1) chronic pain involves distinct spinal cord nociceptive neurons with distinct supraspinal projections, resulting in distinct supraspinal modifications of pain and hedonic circuitry
- 2) chronic pain is an integrated sensory, emotional and hedonic construct, where threat value assessment and memory traces of pain directly modulate the extent to which a pain condition is rendered affective or sensory”^{30(p.94)}

Figure 1-1 is a modified diagram of detailed circuitry involved in chronic pain. On the one hand, circuitry exists that is primarily related to sensory aspects of pain. There also exists circuitry related to the emotional or affective dimension of pain. While these circuits are shown to interact, the genesis of chronic pain is due to a unique engagement of particular circuitry preferentially related to the affective dimension of pain. The perception of pain moves from an external threat of potential tissue damage to one of an internalized disease state.

The Akparian et al³⁰ working theory of chronic pain represents a reductionist view of chronic pain where pain is reduced and explained by a series of neural circuits and brain images. In addition, intentional or not, despite arguing for an integrated emotional and sensory experience, the figure below symbolizes a dualistic separation between mind and body: sensation in one circuit and affect in another. This theoretical view is representative of the medical model and is the pervasive view on pain. This view, while somewhat narrow in focus, is useful in further development of therapies aimed at reducing pain. Moreover, this conceptualization of pain has provided some vindication for chronic pain and other disorders previously characterized as ‘functional’ by illuminating biomedical markers of chronic pain; essentially turning them from unreal to real.

FIGURE 1-1

Akparian et al's³⁰ working model of chronic pain



Reprinted from Progress in Neurobiology, vol 87(2), A. Vania Akparian, Marwan N. Bailiki, Paul Y. Geha, pps 81-97, 2009, with permission from Elsevier.

1.4. A cultural conceptual framework of pain

In contrast to the biological model above, Kirmayer³² presents a cultural view of pain. He argues that basic sensorimotor events such as posture provide insight into the meaning of pain through metaphor that is rooted in culture. Whereas the biological model above primarily conceptualizes pain within the inner workings of the body, this concept describes meaning of pain through metaphoric expression. The assumption that verbal reports of symptoms represent a linear

path to the corresponding underlying physiological process is challenged. Instead, Kirmayer³² states:

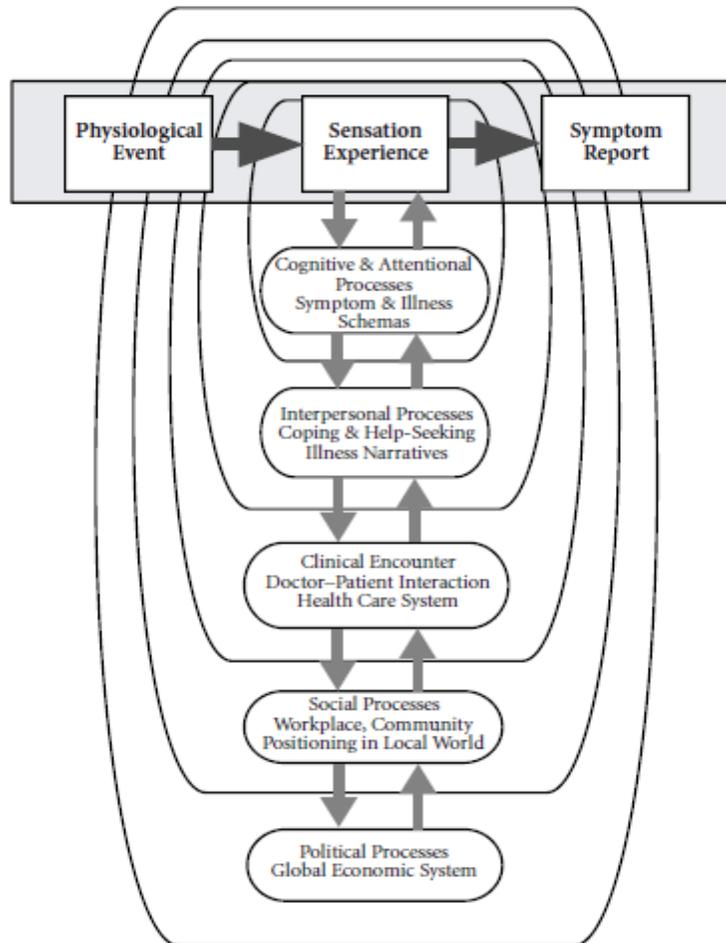
“the translation of physiological processes into sensory experience and their subsequent verbal report is deeply embedded in cultural systems of meaning. Physiological perturbations are organized and expressed in terms of a nested series of cognitive schemas involving knowledge about sensations, symptoms, illnesses or other models of affliction, and broader sociomoral [sic] notions of self and personhood.”^{32(p.319)}

This conceptualization of pain implies physiology is tempered by idiosyncratic perceptions of one’s social self. That is, physiology is relative. The inner workings of the body are interpreted through the lens of our external perceptions and values. Figure 1-2 reproduced from Kirmayer³² demonstrates that physiology, sensations and symptoms are embedded within multiple layers of culture including cognition, coping, personal interactions, community and politics. Each of these social forces colour the experience and expression of pain. To illustrate this, Kirmayer³² provides examples of how culture manifests in postural, facial and temporal expressions of pain.

Posture provides cultural meaning insofar as an upright stance tends to portray control, health, happiness and well-being while lying down or slouching gives the messages of abjection and lamentation.³³ Kirmayer³² argues that pain initiates, organizes and maintains withdrawal and avoidance behaviour. These withdrawal and avoidance behaviours clearly manifest in posture and portray messages of immobilization and calls for help. While pain dictates behaviour, behaviour may also influence pain. For example, standing tall may signify revolting against pain, while recoiling might mean acceptance. Depending on the context, revolting against or accepting pain may or may not lead to perceived reductions in the sensory experience of pain.

FIGURE 1-2

Kirmayer's³² illustration of cultural influences on physiology, sensation and symptom report.



“The social embedding of symptom and sensation experience. Medical semiotics assumes that verbal reports of symptoms are directly based on sensations, and that sensations in turn simply reflect physiological events. In fact, the transduction of physiological events to sensations, and the translation of sensations to symptom experience are shaped by a hierarchy of processes involving cognitive and attentional processes regulated by sensation and symptom schemas, interpersonal interactions involving narrative conventions and social positioning, and the exigencies of the health care system and wider social institutions. As the bi-directional arrows indicate, these processes reach up from global social and political processes to the very earliest inception of sensations to shape bodily experience.”^{20(p.322)}

Reprinted from *Transcultural Psychiatry*, vol 45(2), Laurence J. Kirmayer, pps 318-338, 2008, with permission from SAGE Publications.

The meanings associated with facial expressions move beyond those of posture since the face provides the most powerful means of expression. Kirmayer³² suggests the face not only expresses suffering, but also elicits mirrored responses from those receiving the message of suffering. He states: “the face reveals our feelings and responses to others; it is thus the primordial site of our own social presence and identity.”^{32(p.328)} Thus, if the expression of pain has deep social roots, so must the experience of pain. For example, the observation that many people turn away when faced with a face of suffering is telling about society’s view of pain. How one chooses to display their pain is in part dependent on society’s view of pain. An instance of this is stigma, a common experience for people with chronic pain. Stigma refers to an experience where a ‘marked’ person is viewed inferior or in a negative light by those who are ‘unmarked’.³⁴ If one feels stigmatized, one may choose to become recluse. This isolation has obvious implications for both physical and mental health and signifies shame.

Kirmayer³² posits that pain organizes temporal responses to injury. An early response is to withdraw. Through memory and learning, early and subsequent responses become socially formed and as memories help us remember what caused suffering.

“Culture works on and through memory at every step of this process: at the moment of registration, by framing experiences as memorable and assigning them specific meanings... To appreciate how ritual can transform painful memory, therefore, we must understand memory not only as representation in an individual, but as collective representation, and not only as representation, but as individual and collective enactment.”^{32(p.331)}

The manner in which pain is expressed provides insight into the temporality of pain. Experiences that endure are narrated as suffering, while others are just forgotten. Kirmayer³² contends that this continual construction of the relationship

between self and pain provides the texture for how the social world reconciles the immediate experience.

These metaphors, Kirmayer³² argues, provide insight into the meaning of pain. Metaphors such as posture and facial expression have deep social roots. Not only do personal and social values and norms influence the expression of pain, but also the expression itself is suggested to induce the sensory experience of pain. This conceptualization of pain embedded within culture is helpful in hypothesizing rationale for apparently different experiences of pain across cultures. However, in a society dominated by the medical model, such conceptualizations may be difficult to grasp by doubters who require biological explanations for observed phenomena.

1.5. A biocultural conceptual framework of pain

Presented above are divergent concepts of pain: a reductionist biological view and a cultural constructivist stance. Both approaches have their merits and limitations. Many have written about the shortfalls of the medical model grounded in positivist ideology.^{1,11} The proposed alternatives range from outright rejection such as a purist constructivist³⁵ to a more integrative stance.¹ A popular alternative to the medical model is the biopsychosocial model; a model famously and strenuously advocated by Engel.¹¹ Despite widespread acceptance of this model, detractors remain. Weiner⁴ published a paper criticizing the biopsychosocial model for hampering medical progress, being not falsifiable, and ubiquitous to a fault, among other things. This criticism has since been convincingly refuted.³⁶ It has also been reported that some believe the biopsychosocial model is simply viewed as a flaccid slogan, taught to medical students in lectures and forgotten by residents on the hospital floor.³⁷ Despite some dissention regarding the biopsychosocial model, this thesis supports this approach to pain insofar as biological factors are influenced by, and influence,

psychosocial factors. The tenants of this model are discussed in more detail below.

A biocultural conceptual framework¹ is presented as an alternative to a biopsychosocial framework. Biocultural and biopsychosocial frameworks are essentially the same conceptually, differing primarily semantically. Morris³⁸ argument for the term cultural is because it implies a reaching farther into context (e.g. advertising and religion) than what is implied by the term psychosocial. He argues that the term psychosocial has taken on a negative meaning for some. In addition, some simply equate psychosocial to a narrow view of work and family. Despite this constitutive difference, Morris¹ biocultural framework is conceptually very similar to the biopsychosocial model and other prominent models³⁹ insofar as matter is not rejected in favour of mind, rather the two are integrated:

“A biocultural [framework] must include whatever we can learn about the neurobiology of pain. Yet neurobiology will never encompass the entire event. An adequate model must acknowledge the ways in which the human nervous system is set into motion by the impact of mind and culture”^{40(p200)}.

A common misconception that occurs when multiple terms are fused is the assumption that each concept is valued, but separate. For example, it was not Engel's¹¹ intention that the term biopsychosocial is meant to be interpreted as biology *or* psychology *or* social factors in understanding pain. The fusion of these concepts is meant to be taken more literally. Despite this, many researchers and clinicians work within silos. Morris¹ biocultural conceptualization aims to explicitly integrate biology and culture:

“human bodies are not biological mechanisms independent of mind and culture... the person is not a self-contained unit independent of culture or body. Rather, [the biocultural model] regards human beings and complex

human events like health and illness as constructed at the intersection of culture and biology.”^{40(p200)}

Endorsement of this model requires a degree of comfort with complexity. For the bench scientist, rather than viewing pain in a sterile controlled environment, one must acknowledge, if not embrace, the idiosyncrasy of belief and the noise of culture. For the medical anthropologist, rather than viewing pain as entirely constructed, one must acknowledge, if not embrace, the reality of neurochemical and neuroanatomical representations of pain. This integration of divergent views on pain is exemplified in Morris’³⁸ application of ‘double coding’ first put forward by Jencks⁴¹. Double coding involves two different styles coexisting in a single composition. Morris goes on to say that double coding “consolidates the strengths of disparate conditions and creates innovative hybrids expressing the synergies of a pluralistic age.”^{38(p.9)}

While Morris¹ is convincing in his arguments for a biocultural framework he acknowledges that it is no panacea. Instead, it is meant to highlight that no single discipline is capable of unraveling the complexity of pain. Those who view pain holistically likely share many of the beliefs of the biocultural framework. Thus, Morris⁴⁰ directs his plea toward reductionists, challenging them to wonder whether the study of pain ...

“demands just another simple mechanistic explanation but a new, dialogical vision that sees culture as indelibly influenced by human biology and that sees human biology deeply engaged and even modified by the shaping influence of culture.”^{40(p.201)}

A biocultural framework does not make curative claims but is best summarized by Morris³⁸ as the reciprocal relation between culture and illness, it does not imply that crystals will cure cancer or that consciousness will restore a missing limb. A biocultural model means:

“beliefs shape perceptions and alter bodily processes. It means that cultures shape beliefs. It means that illnesses occur in people who live in cultures that affect illness.”^{38(p.10)}

The biocultural framework falls short of a model or theory that provides specific testable rules or hypotheses. However, it remains a useful conceptual tool highlighting the contribution of biology and culture. Perhaps more importantly, it specifically advocates an explicit integration of divergent views of pain.

1.6. Cognitive-behavioural approach to pain

Any conceptual tool within the literature is only useful to the extent that it informs/contributes/builds upon clinical practice and research. Thus far, I have argued that meaning is central to pain and one way of examining meaning is through belief. The relevance of belief is apparent in conceptual frameworks such as the biocultural approach. The clinical relevance of belief and the biocultural framework can manifest in the cognitive-behavioural approach to the management of pain. In this section, the cognitive-behavioural approach to pain is described to further justify the importance of studying the meaning of pain.

A primary tenant of the cognitive-behavioural approach is that reactions to pain are viewed as self-activated based on learned experiences as opposed to automatically evoked.⁴² Thus, we have potential to control how we respond to pain. Over time, we develop a repertoire of cognitive schemas to facilitate responses to incoming stimuli. Schemas are defined as general patterns of concepts that reflect experience and influences, as well as expectations for the present and future.³⁹ Thus, how one reacts to a current situation occurs within the context of their idiosyncratic schemas. Comparison of existing schema and present context engages a meaning analysis that is the basis for how the situation is interpreted, labeled and acted on.⁴² An illustration of this process is a story from a pain education book called *Painful Yarns* in which the author tells stories to

illustrate the processing of pain.⁴³ In this story, the author describes walking barefoot in the bush when he experiences mild pain in the bottom of the foot attributed to a scratch. This attribution was based on experience. It turns out what actually occurred was a life threatening snakebite. Years later, again walking barefoot in the bush, he experienced excruciating pain after stepping on a twig that was attributed to a snakebite. In the first instance, a significant threat was presented that was minimized based on previous benign experiences of walking barefoot through the bush. In the second case, a benign stimulus to the foot was perceived as severe based on a previous improper response to a potentially life threatening situation. In both cases, the responses were inappropriate because of the available schemata to compare the current situation. Thus, the specific thoughts and feelings that patients experience prior to exacerbation of pain, during an exacerbation, as well as following a pain episode, can greatly influence the experience of pain and subsequent episodes.^{44,45}

The self-activation of responses to stimuli such as pain that are based on previous learning is just one of numerous assumptions of the cognitive-behavioural approach to pain. In Table 1-1, Turk and Okifugi⁴² summarize five assumptions particularly salient to pain management. The example above reinforces the notion emphasized in the first two assumptions: a standard passive response to stimuli does not exist. It is generally well accepted that the Cartesian hypothesis of linear responses to stimuli is not tenable. There are numerous examples where contextual factors modify even a sterile and standardized experimental painful stimulus.¹² Thus, thoughts influence biology, which influences behaviour. Conversely, biology and behaviour affect thoughts.

The third assumption states behaviour is reciprocally determined by personal and social environments. In the cultural conceptual framework described above,³² it was argued that social perceptions of pain influence personal responses to pain. For example, an adversarial compensation system may lead sufferers of pain to

respond to pain negatively due to a belief that they must prove their disability (as opposed to focusing on their abilities). On the other hand, the way individuals respond to pain might influence the larger social context. For example, responses to pain that are in part grounded in motives to receive help has implications for the healthcare system compared to those who respond to pain more internally and choose not to seek help. These illustrations highlight the importance of understanding the personal and social context in forming beliefs and behaviours about pain.

The fourth and fifth assumptions relate to the recognition that if learning contributes to responses or behaviours that are not helpful in managing pain, then addressing the maladaptive ways of thinking and feeling are both possible and encouraged. If pain is indeed complex and subjective, then knowledge about idiosyncratic beliefs, appraisals, and coping repertoires become critical for optimal treatment planning and for accurately evaluating treatment outcome.⁴⁶ More specifically, the focus is to help sufferers gain control over the effects of pain on their lives through active modification of the affective, behavioural, cognitive, and sensory facets of the experience.⁴² The behavioural piece helps to provide a sense of self-confidence in performing activities one assumed to be too difficult. The cognitive component helps to place affective, behavioural, cognitive, and sensory responses under the patient's control.⁴² Consistent with learning theory, long term maintenance of behavioural change occurs only if the patient has learned to attribute success to his or her own efforts.⁴²

TABLE 1-1

Assumptions of the Cognitive-behavioural approach to pain management adapted from Turk DC and Okifuji A⁴²

1. All people are active processors of information rather than passive reactors to environmental contingencies. Environmental stimuli are filtered through organized schemata derived from prior learning and general heuristics guiding information processing. Anticipated consequences are as important as actual consequences.
2. Thoughts can elicit or modulate affect and physiological arousal, both of which might serve as impetuses for behaviour. Conversely, affect, physiology and behaviour can instigate or influence thinking.
3. Behaviour is reciprocally determined by both the environment and the individual.
4. If people have learned maladaptive ways of thinking, feeling and responding, then successful interventions designed to alter behaviour should focus on maladaptive thoughts, feelings, physiology, and behaviours and not one to the exclusion of others.
5. In the same way that people are instrumental in developing and maintaining maladaptive thoughts, feelings and behaviours, they can, are, and should be considered active agents of change of their maladaptive modes of responding. People with chronic pain are not helpless pawns of fate. They should become instrumental in learning and carry more effective modes of responding to their environment.

1.7. Epistemological/Ontological stance for the thesis: Pragmatism

To this point, I have advocated a biocultural conceptual framework of pain and discussed how this manifests clinically by outlining the cognitive-behavioural

approach to pain management. This information implies an epistemological and ontological stance underlying this thesis. However, further clarification is required to situate the epistemology and ontology informing this thesis. Various worldviews such as post-positivism and constructivism coloured the discussion above. This is intentional as the epistemological and ontological perspective governing this thesis is pragmatism. If one accepts that the nature of reality is understood in terms of degrees of relativism or realism, then pragmatism represents a viable perspective. Furthermore, if one acknowledges the possibility that objectivism and subjectivism could be context dependent, then pragmatism may be an appropriate lens to examine phenomena. Pragmatism embodies a moderate view of purist philosophical stances. Pragmatism seeks to reconcile these perspectives insofar as the resulting resolution provides an ideal outlook of a complex phenomenon.⁴⁷

Pragmatism is based on a contingency approach to research methods and concept selection.⁴⁷ There are instances where qualitative methods based in naturalistic inquiry are useful, others where quantitative methods based on the scientific method are preferred, and yet others where mixing methods is appropriate. A moderate and consequentialist worldview permits a pluralist approach to research. This is not to say that pragmatism is an approach void of assumptions. Nor is it a method that conveniently subverts restrictions of purist philosophical foundations. Table 1-2 summarizes the general characteristics of pragmatism that are particularly relevant.

A comprehensive reflection of epistemology and ontology requires examination of the weaknesses of this approach. Johnson and Onwuegbuzie⁴⁷ provide a thorough summary of these weaknesses. Those most relevant to this thesis are presented. Since basic research is not viewed as immediately applicable, it might receive less attention than more applied research. This is not necessarily a rule as a pragmatist evaluates the potential consequences of research. As will be seen in

Chapter 2, basic research can be linked to applied research and theory and vice versa, strengthening understanding. Thus, while basic science does not immediately address real world problems it is nonetheless important in achieving a broad understanding of pain.

Pragmatism is criticized for vague conceptualizations of what constitutes useful or consequential research.⁴⁷ Thus, researchers must be clear in how research rooted in pragmatism is useful. This was addressed through discussion of the biocultural conceptual framework of pain. In order to gain a broad understanding of pain, particularly the cognitive dimension, multiple philosophical positions are required. Since little advocacy is needed for the pervasive medical model, an approach that unites relativism and subjectivity with realism and objectivity is likely to be viewed more saliently.

Given the focus of practicality in pragmatism, many philosophers argue that pragmatism fails in its logical philosophical arguments.⁴⁷ Thus, pragmatism is not effective in resolving competing philosophical disputes. While many purists may not be convinced by a pragmatist's argument, in health care, efforts must be made to ensure research has practical consequences. To disregard a valid approach to a problem due to incongruence with a philosophical stance may not lead to balanced understanding of complex, non-linear problems such as pain. Nevertheless, the criticism of a logically lacking sound philosophical position is not taken lightly. It is acknowledged that contradictions with theoretical perspectives, particularly in Chapter 3 of this thesis, exist. Consequently, transparency is provided to demonstrate how philosophy guided decision-making and how these decisions were tempered in lieu of a non-purist pluralist approach.

TABLE 1-2

General characteristics of pragmatism adapted from Johnson and Onwuegbuzie⁴⁷

- Pragmatism aims to find a middle ground between philosophical dogmatisms to find a workable solution to many longstanding philosophical dualisms.
- Rejects traditional dualisms and generally prefers more moderate and commonsense versions of philosophical dualisms based on how well they work in solving problems.
- Recognizes the importance of the natural or physical world *and* the emergent social and psychological world that includes language, culture, human institutions, and subjective thoughts.
- Knowledge is both constructed *and* based on the reality of the world we experience and live in.
- Endorses fallibilism
- Endorses eclecticism and pluralism (e.g., different, even conflicting, theories and perspectives can be useful).
- Endorses a strong and practical empiricism as the path to determine what works.
- Views current truth, meaning, and knowledge as provisional.
- Prefers action to philosophizing (pragmatism is, in a sense, an anti-philosophy).
- Takes an explicitly value-oriented approach to research that is derived from cultural values; specifically endorses shared values such as democracy, freedom, equality, and progress.
- Pragmatism is a dynamic homeostatic process of belief, doubt, inquiry, modified belief, new doubt, new inquiry . . . The present is always a new starting point.
- Generally rejects reductionism (e.g., reducing culture, thoughts, and beliefs to nothing more than neurobiological processes).

Despite criticisms of integrating divergent philosophies, Johnson and Onwuegbuzie⁴⁷ cite deJong⁴⁸ in making a compelling argument for the pragmatic approach:

“In the social and behavioural sciences, this goal of understanding leads to the examination of many different phenomena, including holistic phenomena such as intentions, experiences, attitudes, and culture, as well as more reductive phenomena such as macromolecules, nerve cells, micro-level homunculi, and biochemical computational systems. Thus, there is room in ontology for mental and social reality as well as the more micro and more clearly material reality.”^{47(p.15)}

Mixing methods does not necessitate implementation of a pragmatic perspective. Others have rationalized the mixing of methods differently.⁴⁹ What is consistent is the belief that mixed methods research offers an important contribution. Greene³⁷ exemplifies this belief:

“a mixed methods approach to social inquiry distinctively offers deep and potentially inspirational and catalytic opportunities to meaningfully engage with the differences that matter in today’s troubled world, seeking not so much convergence and consensus as opportunities for respectful listening and understanding.”^{49(p.22)}

1.8. Study purposes

Two important themes have emerged from this introductory chapter: (1) meaning (as measured by examining belief) is central to the experience of WAD-related pain, and (2) biological and cultural forces collide in the conceptualization of WAD-related pain requiring a pluralist approach to research and clinical management of WAD-related pain. This thesis provides a pluralist examination of WAD related to these themes. In chapter two, a narrative review of the nocebo hyperalgesic meaning response provides a discussion about physiological and

anatomical processes underlying the specific belief expectations. In chapter three, quantitative and qualitative results are mixed to provide a broad characterization of WAD-related pain beliefs. The quantitative data served to characterize WAD-related beliefs and to formulate a qualitative interview guide that explored the experiences informing these beliefs. Finally, in chapter four, the potential consequences of WAD-related pain beliefs are assessed for their relationship with catastrophizing and their capacity to predict pain and disability six months post-MVC. Each study purpose is detailed below.

Chapter 2 (Study 1): *Meaning modulation through placebo hyperalgesia: the biological plausibility of belief*

Pain modulation by cognitive and emotional factors appears to be tacitly understood. However, this tacit acceptance does not necessarily lead to changed clinical behaviour in line with models reflective of contemporary views of pain such as the biopsychosocial and biocultural models. Advances in technology have permitted an improved understanding of how cognitions such as expectations influence the experience of pain. This paper attempts to summarize evidence highlighting the specific anatomical and neurochemical mechanisms leading to pain modulation via expectations. Placebo hyperalgesia provides an illustration of a meaning response mediated by the context created by expectations to influence the experience of pain. Potential clinical manifestations of the placebo hyperalgesic meaning response are also discussed.

Chapter 3 (Study 2): *“If they can put a man on the moon they should be able to fix a neck injury”:* A mixed-methods study characterizing and explaining pain beliefs about WAD

Meaning is intrinsic to human pain and has broad social and institutional determinants, but it is also individual.¹ Expression of one’s beliefs is one way of understanding meaning. Traditionally, beliefs are captured through quantitative surveys and summarized statistically. However, important WAD-related pain

beliefs and how they behave over time have yet to be explored. In addition, given the idiosyncratic nature of belief, a dearth exists in the representation of the detailed voices of the sufferer. Both quantitative and qualitative characterizations of beliefs are thus needed to achieve a broad perspective of WAD-related pain beliefs. Quantitative findings permit a group mean characterization of beliefs that can be compared to other conditions or cohorts and be subjected to statistical analysis. Qualitative findings provide detailed lived experiences leading to a shared meaning of what informs belief endorsement. The purpose of this study was to use quantitative data that characterizes WAD-related pain beliefs over time to develop a qualitative method that provides additional inter-subjective meaning. Specifically, this qualitative component explores experiences that inform belief endorsement as recorded on a quantitative survey measuring WAD-related beliefs.

Chapter 4 (Study 3): *Predictive capacity of WAD-related pain beliefs and catastrophizing*

The prevalence of persistent WAD-related pain is high and constitutes a significant burden to healthcare.^{22,50} Prognostic research suggests WAD outcomes are largely determined by psychological factors such as catastrophizing.^{22,51} Despite numerous studies examining psychological factors such as pain beliefs, these data remain inconsistent necessitating further clarification. In addition, pain beliefs is a broad construct that requires a broad representation of pain beliefs to examine their association with constructs known to be important in WAD such as catastrophizing and for predicting outcomes such as pain and disability. Pain beliefs measured by the Survey of Pain Attitudes (SOPA) and Pain Beliefs and Perception Inventory (PBPI) have been useful in chronic pain settings. However, these tools have not been used to examine beliefs in early to late stages of WAD. We examined the capacity of SOPA and PBPI beliefs and catastrophizing to predict pain intensity and disability six months post-MVC.

Chapter 5: *Lessons learned: maximizing response and minimizing attrition*

These three studies provide a pluralist perspective of WAD-related pain beliefs and contribute significant novel findings to the literature. However, additional lessons were learned throughout the research process. In particular, there was a significant challenge presented throughout the data collection process that adversely affected the studies in chapters three and four. Thus, this chapter provides a discussion of the consequences of response and attrition bias, strategies advocated and used to mitigate these consequences, and recommendations for similar future endeavours.

Chapter 6: *General discussion and conclusions*

In this chapter, key findings are summarized. In addition, the principal focus is on integrating the major findings therefrom chapters two through four.

1.9. References

1. Morris DB. Sociocultural and Religious Meanings of Pain. In: Gatchel RJ and Turk DC editors. *Psychosocial Factors in Pain: Critical Perspectives*. New York (NY): The Guilford Press; 1999.
2. Beecher HK. Pain in Men Wounded in Battle. *Ann Surg*. 1946;123(1): 96-105.
3. Moseley GL. A pain neuromatrix approach to patients with chronic pain. *Man Ther*. 2003;8(3):130-140.
4. Weiner BK. The biopsychosocial model and spine care. *Spine*. 2008;33(2):219–23.
5. IASP Taxonomy [internet]. 2011 [cited 2011 Jun 17]. Available from: <http://www.iasp-pain.org/Content/NavigationMenu/GeneralResourceLinks/PainDefinitions/default.htm>
6. Baumeister RF. *Meanings in life*. New York (NY): The Guilford Press; 1991.
7. Edwards CL, Fillingim RB, Keefe F. Race, ethnicity and pain. *Pain*. 2001; 94(2):133-137.
8. Sheffield D, Biles PL, Orom H, Maixner W, Sheps DS. Race and sex differences in cutaneous pain perception. *Psychosom Med*. 2000;62(4):517–523
9. Unruh AM. Gender variations in clinical pain experience. *Pain*. 1996;65(2-3):123–67.
10. Obelieniene D, Schrader H, Bovim G, Miseviciene I, Sand T. Pain after whiplash: a prospective controlled inception cohort study. *J Neurol Neurosurg Psychiatry*. 1999;66(3):279-83.
11. Engel GL. The need for a new medical model: A challenge for biomedicine. *Science*. 1977; 196(4286):129-136.
12. Moerman DE. *Meaning, medicine and the “placebo effect”*. West Nyack (NY): Cambridge University Press; 2002.

13. Moerman DE, Jonas WB. Deconstructing the placebo effect and finding the meaning response. *Ann Intern Med.* 2002;136:471-476
14. Kaptchuk TJ, Kelley JM, Conboy LA, et al. Components of placebo effect: randomised controlled trial in patients with irritable bowel syndrome. *BMJ.* 2008;336(7651):999-1003.
15. Sawamoto N, Honda M, Okada T, et al. Expectation of pain enhances responses to nonpainful somatosensory stimulation in the anterior cingulate cortex and parietal operculum/posterior insula: an event-related functional magnetic resonance imaging study. *J Neurosci.* 2000;20(19):7438-7445.
16. Benedetti F, Amanzio M, Vighetti S, Asteggiano G. The biochemical and neuroendocrine bases of the hyperalgesic nocebo effect. *J Neurosci.* 2006;26(46):12014-12022.
17. Wrubel J, Benner P, Lazarus RS. Social competence from the perspective of stress and coping. In: Wine J, Syme M editors. *Social competence.* New York (NY): Guilford Press, 1981.
18. Holm LW, Carroll LJ, Cassidy JD, Hogg-Johnson S, Côté P, Guzman J, Peloso P, Nordin M, Hurwitz E, van der Velde G, Carragee E, Haldeman S. The burden and determinants of neck pain in whiplash-associated disorders after traffic collisions. Results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders. *Spine*;33(4):S52-S59.
19. Cassidy JD, Carroll LJ, Cote P, Lemstra M, Berglund A, Nygren, A. Effect of eliminating compensation for pain and suffering on the outcome of insurance claims for whiplash injury. *New Eng. J. Med.* 2000; 342:1179-1186.
20. Hogg-Johnson S, van der Velde G, Carroll LJ, Cassidy JD, Côté P, Guzman J, Peloso P, Nordin M, Hurwitz E, Carragee E, Haldeman S. The burden and determinants of neck pain in the general population. Results of

- the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders. *Spine*;33(4):S39-S51.
21. Kamper SJ, Rebbeck TJ, Maher CG, McAuley JH, Sterling M. Course and prognostic factors of whiplash: A systematic review and meta-analysis. *Pain*. 2008;138(3):617-629.
 22. Carroll LJ, Holm LW, Hogg-Johnson S, Cote P, Cassidy DJ, Haldeman S, Nordin M, Hurwitz EL, Carragee EJ, van der Velde G, Peloso PM, Guzman J. Course and prognostic factors for neck pain in whiplash-associated-disorders (WAD): Results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and its Associated Disorders. *Spine*. 2008;33(4):S83-S92.
 23. Radanov BP, Dvorak J. Spine update. Impaired cognitive functioning after whiplash injury of the cervical spine. *Spine* 1996;21(3):392-397.
 24. Leth-Petersen S, Rotger GP. Long-term labour-market performance of whiplash claimants. *J Health Econ*. 2009;28(5):996-1011).
 25. Keller T, Chappell T. The rise and fall of Erichsen's disease (railroad spine). *Spine*. 1996;21(13):1597-1601.
 26. Taylor J, Taylor M. Cervical spinal injuries: an autopsy study of 109 blunt injuries. *J Musculoskel Pain*. 1996;4(4):61-79.
 27. Williamson E, Williams M, Gates S, Lamb SE. A systematic literature review of psychological factors and the development of late whiplash syndrome. *Pain* 2008;135(1-2):20-30.
 28. Sterling M. Differential development of sensory hypersensitivity and a measure of spinal cord hyperexcitability following whiplash injury. *Pain*. 2010;150(3):501-506.
 29. Bostick GP, Ferrari R, Carroll L, Russell AS, Buchbinder R, Krawciw D, Gross DP. A comparison of population beliefs across whiplash work related neck and upper extremity injury in Canada. *Eur J Pain*. 2009; 13(3):300-4.

30. Akparian AV, Baliki MN, Geha PY. Towards a theory of chronic pain. *Prog Neurobiol.* 2009;87(2):81-97.
31. Merskey H, Bogduk N. Pain Terms: a current list with definitions and notes on usage. In: Merskey H, Bogduk, editors. *Classification of Chronic Pain, IASP Task Force on Taxonomy.* 2nd ed. Seattle (WA): IASP Press; 1994.
32. Kirmayer LJ. Culture and metaphoric mediation of pain. *Transcult Psychiatry.* 2008;45(2):318-338.
33. Lakoff G, Johnson M. *Metaphors we live by.* Chicago (IL): University of Chicago Press; 1980.
34. Jones EE, Farina A, Hastorf AH, Hazel M, Miller DT, Scott RA. *Social Stigma.* New York (NY): W. H. Freeman; 1984.
35. Lincoln YS, Guba EG. Paradigmatic controversies, contradictions, and emerging confluences. In: Denzin NK, Lincoln YS, editors. *The landscape of qualitative research,* 2nd ed. Thousand Oaks (CA): Sage Publications; 2003.
36. Gatchel RJ, Turk DC. Criticisms of the biopsychosocial model in spine care: creating and then attacking a straw person. *Spine.* 2008;33(25):2831-2836.
37. Sullivan MD. The Quality of our lives. *Advances in mind-body medicine.* 1998;14:253-255
38. Morris DB. How to speak postmodern: medicine, illness and cultural change. *Hastings Center Report.* 2000;Nov-Dec:7-16.
39. Chapman CR, Nakamura Y, Flores LY. Chronic pain and consciousness: A constructivist perspective. In: Gatchel RJ, Turk DC. *Psychosocial factors in pain: critical perspectives.* New York (NY): The Guilford Press, 1999.
40. Morris DB. Placebo, pain and belief: a biocultural model. In Harrington A, editor. *The Placebo Effect.* Cambridge: Harvard University Press; 1997.

41. Jencks C. What is post-modernism? 2nd ed. New York (NY): St. Martins' Press; 1987.
42. Turk DC, Okifuji A. A cognitive-behavioural approach to pain management. In: Wall PD and Melzack R, editors. Textbook of Pain, 4th ed. London: Churchill Livingstone; 1999.
43. Moseley GL. Painful yarns: metaphors and stories to help understand the biology of pain. Canberra: Dancing Giraffe Press; 2007.
44. Jensen MP, Turner JA, Romano RM, Lawler BK. Relationship of pain-specific beliefs to chronic pain adjustment. *Pain*. 1994;57(3):301-9.
45. Turk DC and Okifuji A. Psychological factors in chronic pain: Evolution and revolution. *J Consult and Clin Psychol*. 2002;70(3):678-90.
46. Turk DC, Meichenbaum D, Genest M. Pain and behavioural medicine: a cognitive-behavioural perspective. New York (NY): Guilford Press; 1983.
47. Johnson RB, Onwuegbuzie AJ. Mixed methods research: a research paradigm whose time has come. *Educational Researcher*. 2004;33(7):14-26.
48. deJong HL. Causal and functional explanations. *Theory and Psychology*. 2003;13(3):291-317.
49. Greene JC. Is mixed methods social inquiry a distinctive methodology? *J Mix Methods Res*. 2008;2:7-22.
50. Chappuis G, Soltermann B, CEA; AREDOC; CEREDOC. Number and cost of claims linked to minor cervical trauma in Europe: results from the comparative study by CEA, AREDOC and CEREDOC. *Eur Spine J* 2008;17(10):1350-1357.
51. Walton DM, Pretty J, MacDermd JC, Teasell RW. Risk factors for persistent problems following whiplash injury: results of a systematic review and meta-analysis. *J Orthop Sports Phys Ther* 2009;39(5):334-350.

CHAPTER 2

Meaning modulation through nocebo hyperalgesia: the biological plausibility of belief

2.1. Introduction

Advances in imaging technology have expanded the view of pain from primarily a ‘bottom-up’ process to also include a ‘top down’ process. Whereas the state of the tissues is the primary variable in a bottom-up process, the dynamic relationship between cognitive, emotional, and contextual factors as well as nervous system structure and function comprise top-down processing of pain.¹ Many physical therapists have now come to recognize this tacitly. However, acceptance of this dynamic relationship may be difficult to conceptualize without knowledge of possible underlying mechanisms. In this paper, an illustrative example of how pain is influenced by cognitions is presented. Converging lines of evidence including anatomical, neurochemical and clinical data are summarized and potential applications to clinical practice are hypothesized.

Beecher² published a study that is widely cited as an example of the complex interplay between bottom-up and top-down pain processing. He states:

“There is a common belief that wounds are inevitably associated with pain, and, further, that the more extensive the wound the worse the pain. Observation of freshly wounded men in the Combat Zone showed this generalization to be misleading.”^{2(p.96)}

Observations such as Beecher’s suggest that injured tissue is not the only variable determining the experience of pain, rather transmission of noxious stimuli undergo some form of integration and evaluation prior to pain becoming conscious. This is consistent with the widely advocated biopsychosocial model.³ While this and other helpful conceptual models exist to explain the processes described above,⁴⁻⁶ the specific mechanisms in which cognitive factors modulate

pain at biological levels may be helpful for healthcare professionals involved in the management of patients with pain. Moreover, considering the predominance of the medical model, explanation of cognitive modulation of pain may require mechanistic justification to gain more widespread acceptance.

How do cognitions influence pain? Three cognitive mechanisms have been proposed to influence the perceptions of pain: attention, re-appraisal and expectation.⁷ These mechanisms are not the only factors involved in cognitive modulation (i.e. genetics and nervous system injury/neuroplasticity are also important), nor are they fully understood. However, an emerging body of evidence has facilitated an improved understanding of cognitive modulation of pain. A cognitive mechanism of particular interest for physical therapists is expectation (for a further review of pain modulation mechanisms underlying attention and re-appraisal see^{7,8}). Expectation has been defined as patient predictions about the future outcome and consequences of their health condition.⁹ Based on past knowledge and the immediate context, expectations may prepare or attune the nociceptive system for a decreased or increased pain response. Recent reviews, particularly in low back pain, highlight the importance of expectations in influencing pain.^{10,11}

The study of placebo and nocebo responses has provided a window to examine the physiological and anatomical responses to expectations. Traditionally, these responses were believed to occur with inert treatments. However, if the treatment were truly inert, then no response would be expected. Thus, something must be responsible for these types of responses. Moerman and Jonas¹² suggest placebo and nocebo responses are better characterized as meaning responses. That is, a hyperalgesic response to negative expectations is attributed to the meaning of that stimulus. The rich context characterizing the experience of pain is intimately tied to the chemical inner-workings of the body.^{4,5} Context is more than the setting of a research experiment, or a clinic. It comprises previous experiences, beliefs,

cultural values and norms, gender roles, and more.⁵ These influences form the lens in which pain is viewed and constitutes what is referred to as the meaning of pain. Thus, when faced with a stimulus, the response is not standard, it means something. It is coloured by personal, social, and situational context.

This paper attempts to summarize evidence highlighting specific anatomical and neurochemical mechanisms leading to cognitive pain modulation via expectations. The nocebo hyperalgesic meaning response provides a biological illustration of how expectations could influence the pain experience. Potential clinical manifestations of are also hypothesized.

2.2. Descending Pain Modulation

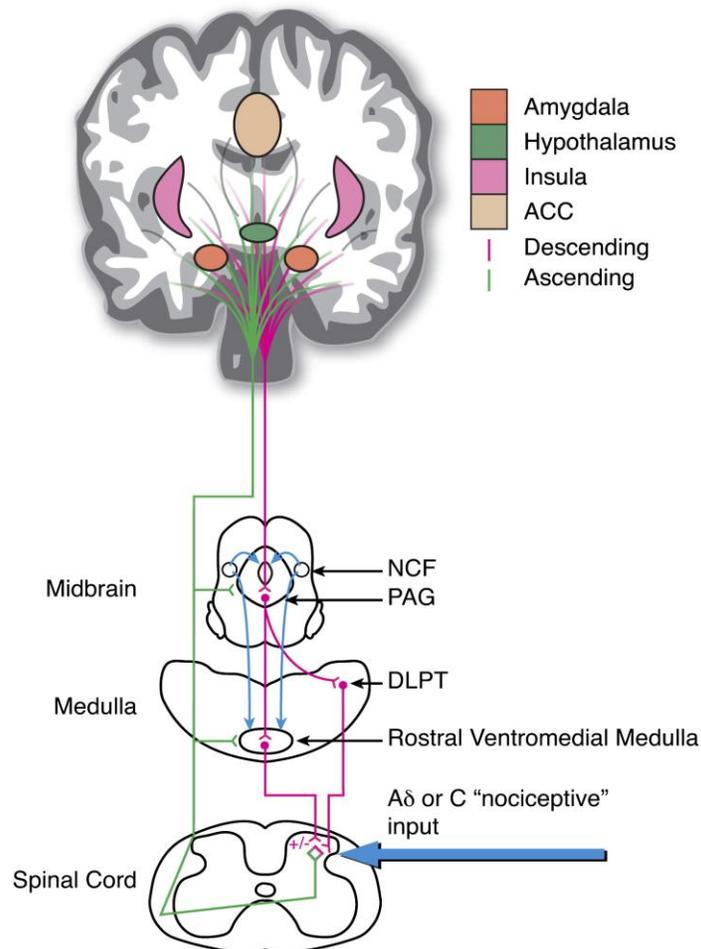
Prior to discussing pain modulation via expectations, a well-known and common pathway involved in ‘top-down’ pain modulation is reviewed: the opioid descending pain modulation pathway (Figure 2-1). Early animal experiments uncovered that injection of morphine (an opioid analgesic) into the periaqueductal gray (PAG) facilitated hypoalgesia.¹³ Subsequent research more clearly established the anatomical make-up of this descending pathway.¹⁴ The components comprising this pathway principally include cortical structures such as the prefrontal cortex, anterior cingulate cortex (ACC), somatosensory cortices, and insula; brainstem structures such as PAG; while the rostral ventral medulla represents the last supraspinal stop before spinal cord nociception transmission is modulated.¹ (Figure 2-1) The coupling of the ACC and PAG in this pathway predicts activity in the somatosensory cortex suggesting influences on the sensory aspect of pain.¹⁵ Though initially conceptualized as a pain inhibitory system, this pathway is now known to be bi-directional. That is, modulation can be either pro- or anti-nociceptive.¹⁴

This descending pain modulation pathway is believed to provide a mechanism whereby cognitive and contextual factors interact with biology to modulate pain.

Negative expectations may alter incoming nociception through the presupposition that pain will be experienced. If an individual is about to experience a potentially noxious stimulus, they may either expect it to be of high or low pain intensity.

FIGURE 2-1

Illustration of the anatomical structures involved in the descending pain modulation pathway.



NCF (nucleus cuneiformis); PAG (periaqueductal gray); DLPT (dorsolateral pontine tegmentum); ACC (anterior cingulate cortex); +/- indicates both pro- and anti- nociceptive influences, respectively

Reprinted from Neuron, vol 55(3), Irene Tracey, Patrick W. Mantyh, pps 377-91, 2007, with permission from Elsevier.

This expectation would be formulated by factors such as background knowledge that the stimulus will be painful, previous similar experience(s), or another person's suggestions of impending pain. In cases where the expectation of the painful stimulus is high but the stimulus is low, one of two things may happen: the perception of pain may be reported as being of high intensity, in which case a nocebo response would have occurred: pronociceptive modulation over-rides information from the periphery and/or changes the pain experience. Alternatively, the perception of pain may be more congruent with the stimulus intensity suggesting the incoming nociception was not modulated. For the former to happen, reasonable hypotheses would include: (1) a neural mechanism involving some type of pre-activation of brain areas capable of engaging descending pain modulation pathway(s) and/or (2) changing the pain experience. Some research has investigated how expectancies modulate normally noxious or non-noxious stimuli, and illuminated the specific anatomical regions and neurochemical mechanisms. We will discuss this in the context of the nocebo hyperalgesic meaning response.

2.3. The Nocebo Hyperalgesic Meaning Response: an example of modulation via expectations

Understanding nocebos via placebos

The nocebo hyperalgesic meaning response is opposite to that of the more familiar placebo hypoalgesic meaning response yet shares many conceptual similarities. Placebo has been defined as a phenomenon whereby expectation or anticipation of a positive outcome induces improvement of a symptom.¹⁶ As understanding of the anatomy and neurochemistry of the placebo phenomenon has improved, it has become of great interest to clinicians due to its numerous implications. However, this raises potential ethical concerns.¹⁷ Although placebo hypoalgesia is most commonly discussed in regard to pharmaceuticals, it has been reported in other non-pharmacological treatments.¹⁸⁻²⁰ Placebo responses have

also been linked to a wider patient-provider treatment context and expectations are thought to be a key influence.²⁰

Evidence from neuroimaging studies using functional magnetic resonance imaging (fMRI) have shown decreased activation of brain structures involved in pain during a noxious stimulus treated with inert cream (placebo treatment) and suggestions of symptom improvement.^{15,21} This suggests an alteration of the pain experience. In addition, hypoalgesia is thought to be mediated via the opioid descending pain modulation pathway discussed above. This is generally accepted in part due to evidence from neuroimaging studies showing activation of the rostral ACC and PAG.^{15,22} Furthermore, the ACC and PAG have been associated with prefrontal cortex activity (an area activated with anticipation of pain) suggesting an ability to recruit the opioid descending modulation pathway prior to stimulation.²¹

Opioids and their associated receptors have been identified as important in this descending modulation of pain.²³ Congruent with anatomical findings, neurochemical evidence indicates that hypoalgesia achieved by recruitment of the opioid descending modulation pathway can be activated by mu-opioid receptors.¹⁶ (Figure 2-2) For example, a placebo hypoalgesia response can be blocked by administration of an opioid antagonist.^{24,25} Mechanistic data such as this are important as it contradicts the allegation that placebo hypoalgesia is simply a result of methodological limitations in controlled trials such as response or publication bias.²¹ Further details of the anatomical and neurochemical basis of placebo hypoalgesia have been discussed elsewhere.^{15,16,22,26}

Nocebo hyperalgesia meaning response

Research in the area of placebo responses has created a foundation for learning more about nocebo. Nocebo is defined as a phenomenon whereby expectation or anticipation of a negative outcome may induce the worsening of a symptom.¹⁶ It

entails a negative response to administration of an ‘inert’ substance or treatment coupled with negative verbal suggestions. Particularly relevant for physical therapists, the phenomenon has been expanded to include broader nocebo-related

FIGURE 2-2

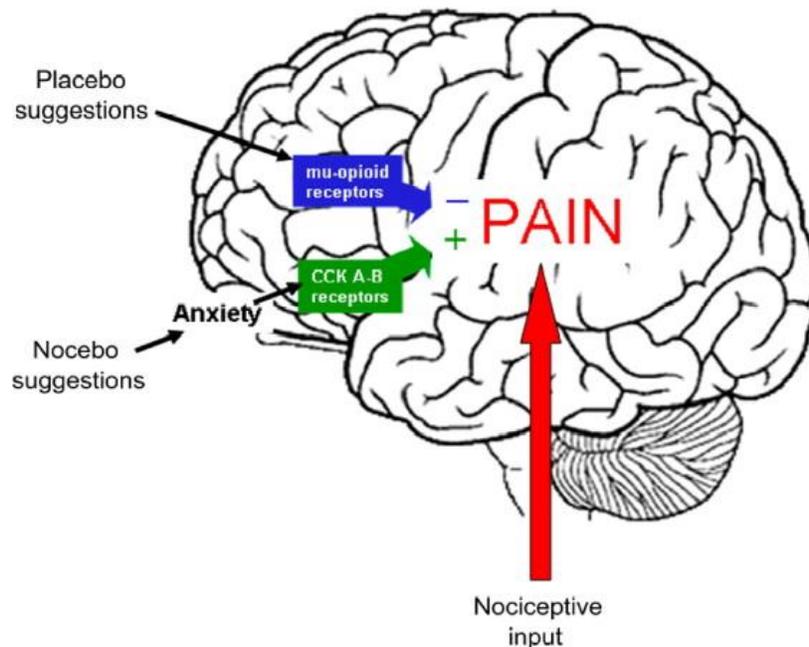


Illustration of the pain attenuating effects on incoming nociception by the stimulation of opioid receptors via placebo suggestions (positive expectations) and the pain exacerbating effects of incoming nociception by the stimulation of cholecystokinin receptors via nocebo suggestions (negative expectations).

Reprinted from *Neuroscience*, vol 147(2), F. Benedetti, M. Lanotte, L. Lopiano, L. Colloca, pps 260-71, 2007, with permission from Elsevier.

effects, such as symptom worsening following negative expectations without the administration of an inert substance or treatment.²⁷ Recall symptom experience (noxious or otherwise) does not occur independent of context or meaning. Thus,

nocebo hyperalgesia is best characterized as an undesirable physiologic or psychological effect of meaning.¹² Similar to placebo, the nocebo hyperalgesic meaning response is thought to be a conscious response and therefore mediated by conscious processes such as expectations, however, other mechanisms such as conditioned responses cannot be ruled out.²⁷

Anatomical and neurochemical mechanisms of the nocebo hyperalgesic meaning response

If nocebo hyperalgesia and placebo hypoalgesia are essentially opposite sides of the same coin and share expectations as a mechanism for pain modulation, it is reasonable to expect the opioid descending modulation system would also be involved in generating nocebo effects. This is especially likely considering the evidence that this pathway has both pro- and anti-nociceptive effects. Indeed, expected pain has been shown to activate similar brain areas involved in modulation of noxious stimuli as those discovered with placebo.²⁸

Similar to placebo studies, the ACC has been implicated in the modulation of pain via expectation.²⁹ Sawamoto and colleagues³⁰ found that expectation of an experimentally induced painful stimulus increased perceived unpleasantness even when the stimulus was innocuous. This subjective finding was confirmed objectively insofar as the expectation of pain enhanced activity in the ACC. Furthermore, unpleasantness was greater when there was uncertainty regarding the nature of the stimulus. This finding is important as it has been shown that uncertainty provokes anxiety,³¹ that as discussed below, is believed to have an important role in nocebo meaning responses. A related study involving manipulated expectations and responses to experimental pain included both subjective (reported pain) and objective (neuroanatomical activation) outcomes, and supports Sawamoto's findings.³² Increased expectations for pain were accompanied by higher reported pain intensity and activation of pain-related areas such as the ACC, insula and primary somatosensory cortex, even when the

stimulus was below the pain threshold.³³ That hyperalgesia occurred when the stimulus was below pain threshold suggests modulation from structures related to anticipation of pain. Moreover, brain regions consistent with expected pain have been found in close proximity to regions mediating the pain experience, suggesting a mechanism to predict impending pain.^{34,35} The association with pre-stimulus expectations and activation of anatomical structures involved in the opioid descending modulation pathway imply this pathway is also involved in modulating pain. Whether pain modulation occurs by changing the pain experience centrally or via this descending pain modulation pathway (other pathways are also likely to be involved) is not entirely clear. However, both cases provide anatomical support for observations that negative expectations alter the pain experience.

If placebo hypoalgesia and nocebo hyperalgesia potentially share a similar modulation pathway, how is hyperalgesia produced in one case and hypoalgesia in the other? Negative expectations, such as thoughts of pain exacerbation, are often anxiety provoking.²⁷ An expectation of pain may evoke feelings of stress or fear regarding the impending stimulus. Physiologically, anxiety appears to be mediated by the neurotransmitter cholecystokinin (CCK).³⁶ CCK has been shown in animal studies to block the analgesic effects of morphine and produce hyperalgesia.³⁷ Further illustrating the potential importance of CCK in nocebo and placebo phenomena, substances known to block CCK enhance placebo hypoalgesia.²⁷ Also of interest is the distribution of CCK in the brain is similar to that of opioid peptides.¹⁶ Thus, placebos and nocebos seem to anatomically and chemically consist of the opposite side of the same coin. On one side, negative expectations activate CCK, facilitate anxiety and pain perception, and interfere with opioid anti-nociception. On the other, opioid peptides facilitate a decreased pain experience.

In order to examine CCK's involvement in the nocebo hyperalgesia meaning response, neurochemical studies have been performed involving exposure to an experimentally induced painful/non-painful stimulus, administration of an inert substance, and manipulation of expectations. For example, patients with mild pain after thoracoscopy were administered a saline injection and a verbal suggestion of worsening of pain (nocebo group). The other groups in the experiment were given nocebo suggestions and various doses of proglumide to block CCK. Results demonstrated an increase in perceived pain in the nocebo group and a complete block of nocebo hyperalgesia in the groups given higher doses of proglumide.³⁸ This finding supports the suggestion above that nocebo effects are likely anxiogenic and mediated in some way by CCK.

Whether the effects of CCK act directly on pain or are mediated through other mechanisms is not entirely clear. In an attempt to differentiate this, Benedetti and colleagues³⁹ performed an experiment where healthy volunteers were randomly allocated to four groups: a natural history, nocebo, nocebo plus pre-treatment of diazepam (blocking anxiety), and nocebo plus pre-treatment of proglumide (blocking CCK). Nocebo hyperalgesia was observed when verbal suggestions of symptom worsening were provided. Administration of proglumide eliminated the nocebo effect, but did not decrease physiological measures of anxiety. Diazepam eliminated nocebo hyperalgesia and anxiety. These results more clearly delineate the relationship between nocebo suggestions, anxiety and CCK (shown graphically in Figure 2-3). Nocebo suggestions of symptom worsening appear to introduce anxiety into processing of painful stimuli, which in turn may activate the CCK-ergic system and descending pain modulation structures. Thus, CCK antagonists likely act on anxiety-induced hyperalgesia rather than nocebo-induced anxiety, and agents that act directly on anxiety are likely to minimize nocebo effects.³⁹

Similar to the placebo hypoalgesic meaning response, numerous neurochemicals are likely involved in the mechanisms underlying nocebo.^{40,41} While more modulation pathways, chemicals and receptors are likely to be discovered, the current evidence is compelling insofar as it provides tangible mechanistic explanations for background theory and clinical studies that highlight the role cognitive factors play in the pain experience.

FIGURE 2-3

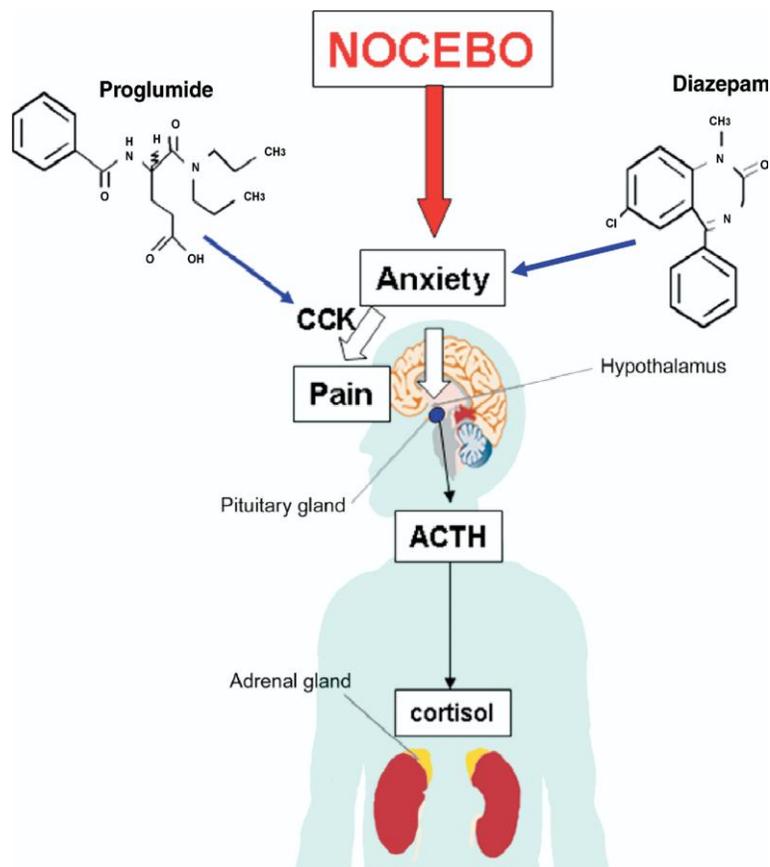


Illustration of the nocebo hyperalgesia mechanism. Verbal suggestions of symptom worsening lead to anxiety which increases the perception of pain, mediated by CCK. The nocebo effect can be blocked by CCK antagonists like proglumide, or by agents that act on anxiety directly (e.g. Diazepam)

Reprinted from Neuroscience, vol 147(2), F. Benedetti, M. Lanotte, L. Lopiano, L. Colloca, pps 260-71, 2007, with permission from Elsevier.

2.4. Relevance of the Nocebo Hyperalgesic Meaning Response for Physical Therapists

As discussed, cognitive and contextual factors can initiate various biological processes leading to perceived worsening of painful stimuli in experimental settings. Does this manifest clinically? Regarding the meaning response placebo hypoalgesia, clinical manifestations may include contextual factors such as positive relationships or therapeutic alliance between therapist and patient, patient acceptance of the theoretical rationale or conceptual scheme for a specific treatment, as well as a therapeutic clinical or healing setting.⁴² Although clinical applications are less developed for nocebo hyperalgesia meaning responses, it is useful to note that significant relationships between pain and negative expectations have been shown in clinical studies. For example, Boersma and Linton⁴³ reported associations between negative expectancies, average pain, pain frequency and pain intensity in patients with neck or back pain. In another study, expectations of recovery after an injury sustained in a MVC were examined against global and region-specific indicators of recovery.⁴⁴ Those who believed they would recover quickly recovered 80% faster than those who did not even after controlling for important confounders.

Somewhat paradoxically, Crombez and colleagues⁴⁵ found expectations for increased pain in a maximal knee flexion-extension task in patients with chronic low back pain did not cause an increase in pain experienced during the task. Rather, expectations for increased pain lead to a fear of re-injury that lead to a modification of the task in an effort to avoid potential pain. This may be partially explained by the mechanism outlined earlier where anxiety mediates the relationship between negative expectations and hyperalgesia.

There are a variety of pain beliefs that have been found to be associated with expectations of a poor outcome. For example, belief that pain will be permanent

or is mysterious has been associated with higher levels of pain intensity and anxiety.⁴⁶ Recently, it has been shown that general population beliefs about neck pain from whiplash tend to be more pessimistic compared to similar conditions including work-related neck pain and shoulder pain.⁴⁷ Moreover, patients who believe their neck pain after MVC is caused by ‘whiplash’ tend to have poorer outcomes compared to those patients who attribute their neck pain to strains or sprains.⁴⁸ Thus, it appears that some conditions such as whiplash-associated disorders are associated with more pessimism that may lead to negative recovery expectations or increased levels of anxiety.

Negative expectations clearly have important implications for pain processing and a negative therapeutic context may activate descending modulation mechanisms that alter the pain experience. In experimental settings, the researcher verbally suggesting symptom worsening typically creates these nocebo-meaning responses. In the clinic, the nocebo meaning response may be generated based on prior experiences, via interactions with healthcare providers or through response to specific interventions that unintentionally evoke anxiety and negative expectations. When health care providers evoke a negative painful response due to treatments or interaction, this is referred to as nocebo iatrogenesis.⁴⁹ Language used by healthcare providers can be extremely important in shaping expectations of impending symptoms. Moreover, patients in distress are prone to take literally what they hear from someone they consider an expert.⁵⁰

Consider the term “unstable” which is commonly used in practice when treating patients with painful spinal conditions. The overall concept of stability is poorly conceptualized and not uniformly applied among researchers and healthcare providers,⁵¹ so it is not far-fetched to think that some patients may have varied views of stability. The term may be interpreted ominously and adopting a threatening label such as ‘unstable’ influences patient’s meaning of pain.⁵² A study by Hafner⁵³ further illustrated the potential consequences of maladaptive

pain beliefs. In this study, the interpretations of patients with chronic low back pain were examined after interactions with a health care provider. Some explanations were construed by patients as frightening and led to an enhanced perception of vulnerability and pessimism about recovery. For example, explanations for chronic low back pain reported by patients included “deterioration of the spine”, “nerve damage”, or “worn out spine”. Some patients were fearful of these explanations and wondered if they might become “paralyzed” or “end up in a wheelchair”. In some cases, patients reported that their health provider actually stated that their condition might lead to paralysis or wheelchair use. Not surprisingly, many patients had pessimistic expectations for recovery stating “recovery is hopeless”, “the spine had come to the end of its life”, and “the more the spine hurts the more it wears away”. These examples highlight unintended misinformation or misinterpretation that may lead to a meaning response characterized by increased anxiety and negative expectations about recovery. While many of the interactions were between patients and surgeons, commonly used jargon by physical therapists (such as ‘spinal instability’) may also be interpreted in an unintended negative way by patients. It is unlikely that healthcare providers intend to paint a pessimistic picture; however care must be taken in attempting to rationalize or describe the source of pain.

Whether a healthcare provider is engaged in conversation with a patient about pain or a patient has formulated their own expectations about treatment or recovery, it is important to acknowledge the manner in which patients view their condition. In medicine, authors have suggested that attempts be made to ameliorate nocebo meaning responses through gentle and reassuring explanations⁵⁴ and by developing collaborative relationships with patients to assist them in understanding potential negative side effects of treatment.⁵⁵ Nocebo meaning responses are not limited to medication side effects and may include responses that are strictly the result of negative cognitions and context. In this regard, given the emerging evidence of the mechanisms underlying cognitive

modulation, physical therapists should consider effective methods of providing reassurance and education^{11,56} as well as cognitive-behavioural strategies when patients already hold negative expectations or beliefs.⁵⁷

2.5. Conclusions

Mechanisms of cognitive pain modulation appear to include a common descending neurological modulation system. Though more is yet to be known about the anatomical and neurochemical mechanisms, research illuminating known mechanisms support theoretical assertions that cognitive and affective factors play an important role in shaping pain. While causal inferences are not possible, we have discussed experimental and clinical evidence supporting the modulation process. Negative expectations about pain underlying placebo hyperalgesia meaning responses have been presented as an example of a cognitive mechanism influencing the experience of pain and recovery. Some clinical applications of these concepts were presented including the importance of providing reassurance as well as avoiding negative labeling of a patients' painful condition.

2.6. References

1. Tracey I, Mantyh PW. The cerebral signature for pain perception and its Modulation. *Neuron*. 2007;55(3):377-91.
2. Beecher HK. Pain in men wounded in battle. *Ann of Surg*. 1946; 123(1): 96-105.
3. Engel GL. The need for a new medical model: a challenge for biomedicine. *Science*. 1977;196(4286):129-136.
4. Chapman CR, Nakamura Y, Flores LY. Chronic pain and consciousness: a constructivist perspective. In: Gatchel RJ, Turk DC, editors. *Psychosocial factors in pain: critical perspectives*. New York (NY): The Guilford Press, 1999.
5. Morris DB. Sociocultural and Religious Meanings of Pain. In: Gatchel RJ and Turk DC editors. *Psychosocial Factors in Pain: Critical Perspectives*. New York (NY): The Guilford Press; 1999.
6. Moseley GL. A pain neuromatrix approach to patients with chronic pain. *Man Ther*. 2003;8(3):130-140.
7. Wiech K, Ploner M, Tracey I. Neurocognitive aspects of pain perception. *Trends Cog Sci*. 2008;12(8):306-313.
8. Villemure C, Bushnell MC. Cognitive modulation of pain: how do attention and emotion influence pain processing? *Pain*. 2002;95(3):195–199.
9. Janzen JA, Silvius J, Jacobs S, et al. What is a health expectation? Developing a pragmatic conceptual model from psychological theory. *Health Expectations*. 2006;9(1):37–48.
10. Iles RA, Davidson M, Taylor NF, O'Halloran P. Systematic review of the ability of recovery expectations to predict outcomes in non-chronic non-specific low back pain. *J Occup Rehabil*. 2009; 19(1): 25-40.
11. Main CJ, Foster N, Buchbinder R. How important are back pain beliefs and expectations for satisfactory recovery from back pain? *Best Prac Res Clin Rheumatol*. 2010;24(2):205–217.

12. Moerman DE, Jonas WB. Deconstructing the placebo effect and finding the meaning response. *Ann Intern Med* 2002;136(6):471-476.
13. Tsou K, Jang CS. Studies on the site of analgesic action of morphine by intra-cerebral micro injection. *Scienta Sinica*. 1964; 13(): 1099-109.
14. Fields HL, Basbaum AI, Heinricher MM. Central nervous system mechanisms of pain modulation. In: McMahon SB and Koltzenburg M, editors. *Textbook of Pain*, 5th ed. Philadelphia (PA): Elsevier/Churchill Livingstone, 2005.
15. Bingel U, Lorenz J, Schoell E, Weiller C, Bushel C. Mechanisms of placebo analgesia: rACC recruitment of a subcortical antinociceptive network. *Pain*. 2006;120(1-2):8-15.
16. Benedetti F, Amanzio M. The neurobiology of placebo analgesia: from endogenous opioids to cholecystokinin. *Prog Neurobiol*. 1997;52(2):109-125.
17. Colloca L, Benedetti F. Placebos and painkillers: is mind as real as matter? *Nature Rev Neurosci*. 2005;6(7):545-552.
18. Hashish I, Feinman C, Harvey W. Reduction of postoperative pain and swelling by ultrasound: a placebo effect. *Pain*. 1988;33(3):303-311.
19. Roche PA, Tan H, Stanton W. Modification of induced ischaemic pain by placebo electrotherapy. *Physiother Theory Pract*. 2002;18(3):131-139.
20. Kaptchuk TJ, Kelley JM, Conboy LA, et al. Components of placebo effect: randomised controlled trial in patients with irritable bowel syndrome. *BMJ*. 2008;336(7651):999-1003.
21. Wager TD, Rilling JK, Smith EE et al. Placebo-induced changes in fMRI in the anticipation and experience of pain. *Science*. 2004;303(5661):1162-7.
22. Eippert F, Finsterbusch J, Bingel U, Bushel C. Direct evidence for spinal cord involvement in placebo analgesia. *Science*. 2009;326(5951):404.

23. Heinricher MM, Morgan MM, Fields HL. Direct and indirect actions of morphine on medullary neurons that modulate nociception. *Neurosci.* 1992;48(3):533-43.
24. Levine JD, Gordon NC, Fields HL. The mechanism of placebo analgesia. *Lancet.* 1978;312(8091):654-7.
25. Benedetti F, Arduino C, Amanzio M. Somatotopic activation of opioid systems by target-directed expectations of analgesia. *J Neurosci.* 1999;19(9):3639-48.
26. Scott DJ, Stohler CS, Egnatuk BS, Wang H, Koeppe RA, Zubieta JK. Placebo and nocebo effects are defined by opposite opioid and dopaminergic responses. *Arch Gen Psychiatry.* 2008;65(2):220-231.
27. Benedetti F, Lanotte M, Lopiano L, Colloca L. When words are painful: unraveling the mechanisms of the nocebo effect. *Neurosci.* 2007; 147(2):260-271.
28. Porro CA, Baraldi P, Pagnoni G, Serafina M, Facchin P, Maieron M, Nichelli P.. Does anticipation of pain affect cortical nociceptive systems? *J Neurosci.* 2002;22(8):3206-3214.
29. Rainville P. Brain mechanisms of pain affect and pain modulation. *Curr Opin Neurobiol.* 2002;12(2):194-204.
30. Sawamoto N, Honda M, Okada T, Hanakawa T, Kanda M, Fukuyama H, Konishi J, Shibasaki H. Expectation of pain enhances responses to nonpainful somatosensory stimulation in the anterior cingulate cortex and parietal operculum/posterior insula: an event-related functional magnetic resonance imaging study. *J Neurosci.* 2000; 20(19): 7438-7445.
31. Carleton NR, Sharpe D, Asmundson, GJG. Anxiety sensitivity and intolerance of uncertainty: requisites of the fundamental fears? *Behav Res Ther.* 2007;45(10):2307-16.
32. Hsieh JC, Stone-Elander S, Ingvar M. Anticipatory coping of pain expressed in the human anterior cingulate cortex: a positron emission tomography study. *Neurosci Letters.* 1999;262(2):61-64.

33. Koyama T, McHaffie JG, Laurienti PJ, Coghill RC. The subjective experience of pain: where expectations become reality. *Proc Natl Acad Sci USA*. 2005;102(36):12950-12955.
34. Ploghaus A, Tracey I, Gati JS, Clare S, Menon RS, Matthews PM, Rawlins JNP. Dissociating pain from its anticipation in the human brain. *Science*. 1999;284(5422):1979-1981.
35. Keltner JR, Furst A, Fan C, Redfern R, Inglis B, Fields HL. Isolating the modulatory effect of expectation on pain transmission: a functional magnetic resonance imaging study. *J Neurosci*. 2006;26(16):4437– 4443.
36. Lydiard RB. Neuropeptides and anxiety: focus on cholecystokinin. *Clin Chem*. 1994;40(2):315-318.
37. Xu XJ, Hokfelt T, Hughes J, eld-Hallin ZW. The CCK-B antagonist CI988 enhances the reflex-depressive effect of morphine in axotomized rats. *NeuroReport*. 1994;5(6):718-720.
38. Benedetti F, Amanzio M, Casadio C, Oliaro A, Maggi G. Blockade of placebo hyperalgesia by the cholecystokinin antagonist proglumide. *Pain*. 1997; 71(2):135-140.
39. Benedetti F, Amanzio M, Vighetti S, Asteggiano G. The biochemical and neuroendocrine bases of the hyperalgesic placebo effect. *J Neurosci*. 2006; 26(46): 12014-12022.
40. Hebb ALO, Poulin JF, Roach SP, Zacharko RM, Drolet G. Cholecystokinin and endogenous opioid peptides: interactive influences on pain, cognition, and emotion. *Prog Neuropsychopharmacol Biol Psychiatry*. 2005;29(8):1225-1238.
41. Enck P, Benedetti F, Schedlowski M. New insights into the placebo and placebo responses. *Neuron*. 2008;59(2):195-206.
42. Frank JD, Frank JB. *Persuasion and healing: a comparative study of psychotherapy*. 3rd ed. Baltimore, Maryland: John Hopkins University Press; 1993.

43. Boersma K, Linton SJ. Expectancy, fear and pain in the prediction of chronic pain and disability: a prospective analysis. *Eur J Pain*. 2006;10(6):551-557.
44. Carroll LJ, Holm LW, Ferrari R, Ozegovic D, Cassidy DJ. Recovery after whiplash associated disorders: do you get what you expect? *J Rheumatol*. 2009;36(5):1063-1070.
45. Crombez G, Vervaeke L, Baeyens F, Lysens R, Eelen P. Do pain expectancies cause pain in chronic low back patients? A clinical investigation. *Behav Res Ther*. 1996;34(11):919-925.
46. Williams DA, Robinson ME, Geisser ME. Pain beliefs: assessment and utility. *Pain*. 1994;59(1):71-78.
47. Bostick GP, Ferrari R, Carroll L, Russell AS, Buchbinder R, Krawciw D, Gross DP. A comparison of population beliefs across whiplash work related neck and upper extremity injury in Canada. *Eur J Pain*. 2009;13(3):300-4.
48. Buitenhuis J, de Jong PJ, Jaspers JP, Groothoff JW. Catastrophizing and causal beliefs in whiplash. *Spine*. 2008;33(22):2427-33.
49. Roche PA. Pain and placebo analgesia: two sides of the same coin. *Phys Ther Rev*. 2007;12(3):189-198.
50. Schenk PW. 'Just breathe normally': word choices that trigger nocebo responses in patients: six language traps and how to avoid them. *American J Nurs*. 2008;108(3):52-57.
51. Reeves NP, Narendra KS, Cholewicki J. Spine stability: the six blind men and the elephant. *Clin Biomech*. 2007;22(3):266-74.
52. Bogduk N. What's in a name? The labelling of back pain. *Med J Aust*. 2000;173(8):400-1.
53. Hafner C. The information we give may be detrimental. In: Gifford L, editor. *Topical issues in pain 4: Placebo and nocebo, pain management, muscles and pain*. Falmouth: CNS Press; 2002.

54. Varelmann D, Pancaro C, Cappiello EC, Camann WR. Nocebo-induced hyperalgesia during local anesthetic injection. *Anesth and Analg.* 2010; 110(3):868–70.
55. Barsky AJ, Saintfort R, Rogers MP, Borus JF. Nonspecific medication side effects and the nocebo phenomenon. *JAMA.* 2002;287(5):622–627.
56. Moseley GL, Nicholas MK, Hodges PW. A randomized controlled trial of intensive neurophysiology education in chronic low back pain. *Clinical Journal of Pain.* 2004;20(5):324-30.
57. Lamb SE, Hansen Z, Lall R, et al. Group cognitive behavioural treatment for low-back pain in primary care: a randomised controlled trial and cost-effectiveness analysis. *Lancet.* 2010;375(9718):869-70.

CHAPTER 3

If they can put a man on the moon, they should be able to fix a neck injury: A mixed-methods study characterizing and explaining pain beliefs about WAD

3.1. Introduction

Meaning is intrinsic to human pain and has broad social and institutional determinants, but it is also individual.¹ Expression of one's beliefs is one way of understanding meaning. Pain beliefs are defined as personally formed or culturally shared cognitive understandings of pain.^{2,3} Beliefs about cause, control, duration, outcome and blame have been shown to be important in determining pain and function.⁴⁻⁶ In experimentally evoked pain, beliefs related to the expected intensity of a painful or non-painful stimulus can modify reported pain,⁷ the neuroanatomical representation of pain,⁸ as well as influence the neurochemistry of pain modulation.⁹

Neck pain after a motor vehicle collision (MVC) (also known as whiplash associated disorder or WAD) can carry with it unique contextual factors such as litigation and strong cultural and institutional influences.¹⁰⁻¹² Thus, beliefs about pain in WAD are particularly salient. In spite of this, there is a shortage of data about WAD-specific beliefs; especially in non-chronic populations. Of the available data, much of it is quantitative and suggests beliefs about WAD are negative when compared to other similar conditions.¹³ Pain catastrophizing, fear-avoidance, self-efficacy, causation and disability beliefs are thought to be important predictors of WAD-related pain and disability.^{6,14-17} However, the research examining the predictive capacity of many of these beliefs is inconsistent and more understanding is required.¹⁸ There are other measures of beliefs found to be useful in chronic pain contexts that have yet to be used in WAD such as the Survey of Pain Attitudes (SOPA) and Pain Beliefs and Perception Inventory (PBPI). Thus, the scope of WAD-specific beliefs also requires clarification.

Considering the idiosyncratic nature of pain and belief, a further dearth in understanding exists: a lack of representation of a voice for the sufferer. Morris¹⁹ suggests that this subjectivity embodies the complexity of illness necessitating a biocultural model. He describes the characteristics of the model as: “beliefs shape perception and alter bodily process, cultures shape beliefs, and illnesses occur in people who live in cultures that affect illnesses.”^{19(p.10)} Thus, while representing beliefs about illness biologically with numbers and images have a place in understanding illness, so does the shared inter-subjectivity captured by qualitative research. Put another way, understanding meaning cannot simply be discovered through surveys, there is no finally correct interpretation, rather it is a matter of coming to terms.²⁰ This requires a depth of exploration that is beyond the scope of a quantitative survey.

Little is known about the subjectivity of WAD-related pain beliefs. Some literature regarding beliefs in the context of low back pain illustrates the utility of qualitative data in understanding pain beliefs. Verbeek et al.²¹ performed a systematic review of qualitative and quantitative studies evaluating a specific belief about low back pain (expectations of treatment) and found patients expect a patho-anatomical diagnosis. This tangible diagnosis led to a feeling that their pain was legitimized. On the other hand, a lack of a diagnosis was associated with feelings of embarrassment and perceptions of being treated cruelly. Qualitative data such as this are helpful in understanding the patient’s perspective and may inform motives for particular beliefs or behaviour. Much of qualitative evidence relates to the context of chronic pain.²²⁻²⁵ There is little known from a qualitative perspective throughout early stages of WAD.

Given the idiosyncrasy of belief and subjectivity of pain, a need exists in the literature to not only obtain quantitative results but to explain such results in more detail, especially in terms of detailed voices and participants’ shared perspectives.

This perspective requires a theoretical framework that can capture this idiosyncrasy and subjectivity. Pragmatism is a useful philosophy to the study of pain beliefs that requires consideration of both quantitative and qualitative viewpoints. While purist approaches (positivism and constructivism) in and of themselves may be inadequate to gain a broad conceptualization of WAD-related pain beliefs, pragmatism represents a contingency approach to addressing problems.²⁶ That is, pragmatism advocates a pluralist approach to inquiry that permits determining how mixing research methods is used most fruitfully to address complex problems.²⁷ In Morris'¹⁹ biocultural model of pain, a prominent attribute is double coding. Double coding represents two differing styles within the same composition.²⁸ The advantage is that “differing styles may unite strengths of disparate traditions and create innovative hybrids expressing the synergies of a pluralistic age”.^{19(p.9)} An example application of double coding in research is integrating quantitative and qualitative methods. These two different methods with disparate assumptions combined create a hybrid approach known as mixed-methods research. In this case, a mixed-methods approach permits examining pain from a group mean/statistical lens as well as from a personal, lived-experience view. Accordingly, the purpose of this study was to use quantitative data that characterizes WAD-related pain beliefs over time to develop a qualitative method that provides additional inter-subjective meaning. Specifically, this qualitative component explores experiences that inform belief endorsement as manifested on a quantitative survey measuring WAD-related beliefs.

3.2. Methodology

3.2.1. Theoretical position

The theoretical position in quantitative approaches tends to be hidden and is usually implied (e.g. positivism or post-positivism). However, qualitative methods

can have foundation in a variety of theoretical positions.²⁰ Identifying ones theoretical stance is important, especially in qualitative and mixed-methods research, as it informs the research process. As mentioned, a pragmatic philosophy congruent with a biocultural model of pain provided the underlying impetus for the research question below and thus the decision to use mixed-methods. Pragmatism permits researchers to reconcile the challenges in incorporating purist perspectives into a mixed-methods study.²⁶ For example, a constructivist supports relativism, while positivists support realism. Pragmatism is based on a belief that knowledge is both constructed and based on the reality of the world we experience.²⁶ Whereas a pragmatic approach holds that there is no *correct* way to understand pain, it is also action-oriented and supports that some beliefs about pain are maladaptive/adaptive for some people under various circumstances. This represents a moderate view for both relativism and realism. In this study, questionnaire subscale scores situate individual and WAD-specific pain beliefs within a broader view of what may constitute helpful or unhelpful thinking. The qualitative piece aims to gain an additional, deeper understanding of these beliefs by adding a differing worldview that assumes beliefs are based on idiosyncratic lived experience. In addition, the qualitative findings arose through an interaction between researcher and participant with explicit efforts made to restore the label of “knower” to the participant. Thus, both post-positivist and constructivist perspectives influence the quantitative and qualitative pieces of this mixed-methods study. While purists would reject such an investigation within one study, the eclecticism and pluralism of pragmatism reconciles differences by focusing on the consequences of integrating divergent approaches. In this case, benefits of providing a broad characterization of the meaning of WAD-specific pain that is congruent with contemporary views of pain such as Morris,¹⁹ biocultural approach outweigh drawbacks of integrating divergent methods. Refer to Appendix A for a detailed account of the researcher’s reflexivity.

3.2.2. Research question

What beliefs do patients have about WAD-related pain throughout the recovery period (as measured via belief endorsement on a series of surveys) and what experiences inform these reported beliefs (as determined through face-to-face interviews)?

3.2.3. Design

Mixed-methods involve using qualitative and quantitative approaches to collect and analyze data in a single study.²⁹ The purposes of this study, based on Greene's³⁰ categorization, were both developmental and complimentary. That is, quantitative data was used to inform a qualitative approach. In addition, the qualitative data served to ascertain the experiences that inform belief endorsement to compliment the quantitative characterization of WAD-related pain beliefs. In other words, the qualitative data explored experiences informing endorsement of WAD-related pain beliefs thus providing additional meaning to the quantitative portrayal of beliefs. The timing of the data collection was concurrent and occurred longitudinally. The paradigm emphasis was qualitative. None of the design typologies outlined by Creswell²⁹ or Johnson and Onwuebguzie²⁶ neatly fit the design characteristics of this study. The design of the study is portrayed visually in (Figure 3-1). The integration of paradigms occurs in the data collection phase, while analysis of quantitative and qualitative data occurred independently. WAD-related pain beliefs were quantitatively measured longitudinally at baseline and at three and six months post-MVC. A sub-group of these participants also agreed to participate in an interview after completing each survey. Interview participants' scores from the surveys guided the interview. This qualitative component explored the experiences that informed endorsement of beliefs reported on the surveys.

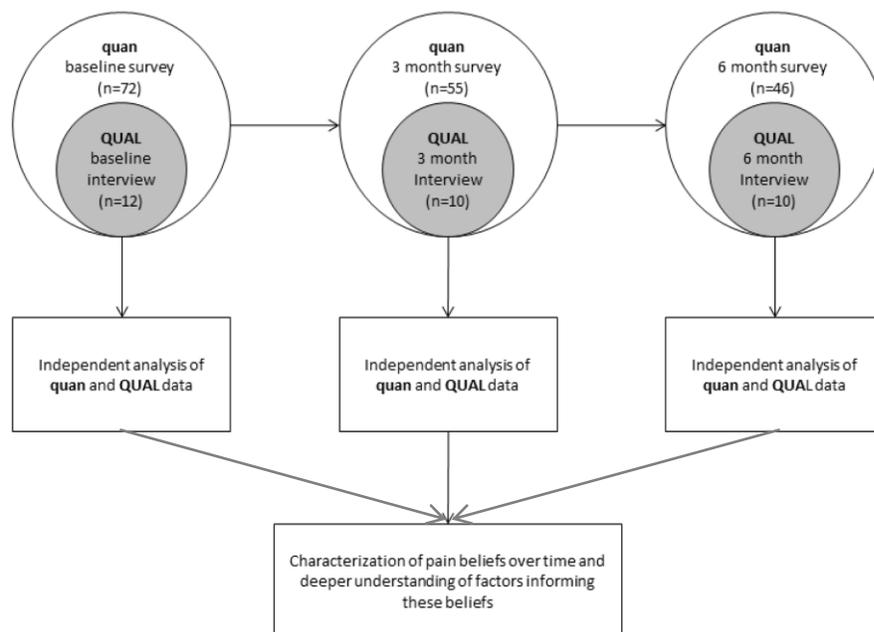
3.2.4 Participants

A convenience sampling strategy was used to recruit study participants between December 2008 and May 2010. Adults with acute (< six weeks) WAD attending physical therapy and chiropractic clinics in Alberta and Saskatchewan were invited to complete a paper-based survey at baseline and three and six months post-MVC. Potential participants under the age of eighteen, unable to speak or read English, not reporting neck pain, or the time since the MVC was longer than six weeks were excluded. At the end of the survey, participants were invited to complete a one-hour face-to-face interview at each measurement occasion.

Interview participant recruitment continued until varied ages and genders were represented. Variation in pain experience (i.e. recovered, not recovered/chronic) was also desired. Although up to 50% of people experience WAD symptoms one-year after the injury³¹ suggesting varied pain experiences would be likely in this sample, defining recovery in terms of pain or timelines alone is problematic.^{32,33} Thus, we aimed to ensure a chronic pain perspective by purposively selecting four additional subjects with long-standing WAD-related chronic pain, separate from the nested qualitative interview sample. These participants ensured maximum variation in sampling. Seeking information from discordant contexts aids in obtaining varied data from multiple perspectives, increasing the likelihood that findings will be of sufficient detail and transferrable to others.³⁴ Interviews continued until no new information was gained (thematic saturation).³⁵

FIGURE 3-1

Diagram of data collection and analysis sequence for this mixed-methods study.



3.2.5. Survey measures

The survey included items related to demographics (age, sex, previous history of WAD, type of insurance, number of painful body parts, and whether or not the injury resulted in an overnight hospital stay), pain intensity, disability and beliefs. An 11-point numerical rating scale and the Whiplash Disability Questionnaire³⁶ measured pain intensity and self-reported disability respectively. Both have supportive validity evidence.^{36,37} Three questionnaires measured beliefs and are discussed in detail.

Survey of Pain Attitudes (SOPA-35):³⁸ the SOPA-35 includes seven subscales with items scored on a five-point Likert agreement scale. The seven factors include Control, Disability, Harm, Emotion, Medication, Solicitude, and Medical Cure. There is considerable validity evidence for the SOPA, though it is primarily located in chronic pain populations.³⁹⁻⁴⁴

Pain Beliefs and Perceptions Inventory (PBPI):⁴⁵ The PBPI is a 16-item questionnaire designed to evaluate patient beliefs about pain. It contains four subscales labeled Mystery, Self-Blame, Pain Permanence, and Pain Constancy. Respondents indicate their agreement with the items on a four-point Likert scale. The validity evidence for the PBPI is primarily in chronic pain settings.⁴⁶⁻⁴⁹

Pain Catastrophizing Scale (PCS):⁵⁰ The PCS is a 13-item instrument that measures the extent of exaggerated negative mental thoughts during actual or anticipated painful experience.⁵¹ It is reported to have three correlated factors: rumination, magnification, and helplessness. The PCS has a considerable amount of validity evidence in both acute and chronic pain populations^{14,52} and has been examined as a prognostic factor in WAD.⁵³ For all three measures, higher scores indicate more endorsement of the belief(s).

3.2.6. Interviews

Qualitative data were collected via semi-structured interviews. Prior to the interview, belief subscales from the measures above were scored. Those with relatively high or low scores (indicating a relatively strong or weak endorsement of a specific belief) formed the content of the interview guide. (Appendix B) Thus, participants were directed to discuss particular beliefs from their survey. Congruent with the research aims of this study, questions focused on eliciting descriptions of experience (e.g. what experiences led you to believe ...?) and opinion/value questions that inform one's goals, intentions, desires and values

(e.g. Why do you believe ...?).⁵⁴ The interview was digitally recorded and professionally transcribed.

3.2.7. *Data analysis*

Quantitative analysis

Descriptive statistics summarized demographic information and characterized beliefs of the sample at each measurement occasion. One-way repeated measures ANOVA determined if beliefs statistically changed over time. Post-hoc tests were conducted with Bonferroni correction. The sample size needed to detect moderate effects ($f^2 = 0.0625$), $\alpha = 0.05$, $\beta = 0.80$, a conservative estimate of correlation among measures, and considering corrections for violations of sphericity, was $n=35$. We also compared age, gender and baseline pain intensity in the study sample with a large database of physical therapy clinics in Alberta and Saskatchewan provided by two physical therapy companies ($n=2759$) to ascertain the representativeness of the study sample.

Qualitative analysis

Upon receipt of the transcribed interview, the researcher performed multiple readings to become thoroughly familiar with the data. Consistent with the research aims, the data was analyzed using a meaning-focused approach, which is typical of analytic procedures employed in research with a constructivist philosophy.³⁴ This approach consisted of content analysis according to the procedures outlined by Bowling.⁵⁵ Coding occurred within the same interview in short segments or paragraphs. Once the analysis revealed mutually exclusive codes, further linkages between these units of data led to the generation of themes. As understanding increased, themes were refined. Thematic analysis underwent three iterations over approximately a six month period. During this period, readings that were influential in framing the final stage of analysis included Kleinmann's Illness Narratives⁵⁶ and Frank's Wounded Storyteller.⁵⁷ In the final

stage of the analysis, a framework was constructed that best represented the data and linked themes into a meaningful interpretation.

In order to evaluate study trustworthiness⁵⁸ (qualitative validity), the following procedures were performed. Member checking ensured that the researcher's interpretations adequately represented the interviewee's voice. Within each interview, the interviewer summarized notes and preliminary interpretations to the participant. The interviewer asked for verification/clarification of these interpretations. In addition, participants received the results of the interview portion of the study and were given the opportunity to anonymously comment on the findings and provide clarifications. The interviewee could document clarifications and return them to the investigator by mail. Finally, an independent researcher performed an audit of the data analysis process and product. This auditor was provided three samples of transcripts with associated documents demonstrating coding and thematic analysis, summary of methodology and the analyst's reflexivity. The auditor evaluated the acceptability of whether the interpretations were grounded in the data, logical, clear and of sufficient explanatory power. In addition, the auditor commented on the likelihood of inquirer bias based on the researcher's reflexivity.

3.3. Results

3.3.1. Sample characteristics

Seventy-two participants comprised the study sample. The characteristics of the participants are found in Table 3-1. Regarding representativeness, the study sample was similar to a large physiotherapy database (n=2759) for age and initial pain intensity [mean age (std dev) = 39.1 (13.9) and 40.4 (14.2); mean baseline pain intensity (std dev) = 5.2 (2.0) and 5.9 (2.1) respectively]. However, the study sample had a higher proportion of females (79.2%), compared to the larger clinic sample (60.1%). The characteristics of the interview sample are also found in

Table 3-1. Based on these descriptive data, the interview sub-sample consisted of a higher proportion of females, those with a history of whiplash, and insurance under a tort (litigious) system. Thirteen people comprised the interviewee sub-sample (Table 3-2). Of these participants, eight completed all interviews. Two participants were lost to follow-up after the baseline measurement and three participants could not attend one interview due to conflicts. As mentioned, four additional participants with WAD-related chronic pain were included to provide increased heterogeneity. Thus, 36 interviews (12, 10, and 14 at baseline, 3 and 6 months respectively) were conducted with 17 participants. Aliases were used to protect interviewees' anonymity.

3.3.2. Characterization of WAD-related pain beliefs

At the three- and six-month follow-up, 55 (76%) and 48 (67%) participants returned a survey respectively. Comparison of participants with complete and missing data showed both groups were similar concerning age, number of painful body parts, previous history of whiplash, insurance system, baseline pain and disability, and beliefs. Male participants were more likely to be lost to follow-up. Pain intensity, disability and beliefs of participants over time are portrayed in Table 3-1. Pain intensity and disability decreased over time from the baseline to three-month follow-up. Little change occurred after three-months. When the interview and non-interview groups were combined in analysis, beliefs appeared to be relatively stable with the exception of beliefs about disability, medical cure and pain permanence and constancy. Post-hoc tests revealed no changes between the three- and six-month measurements for these beliefs. Beliefs were generally similar between the no interview and interview groups with the exception of some minor differences at the six-month follow-up. Beliefs related to medical cure, medication and control were generally endorsed, while disability, solicitude, permanence, mystery and self-blame beliefs were endorsed less.

TABLE 3-1

Sample description and characterization of pain intensity, disability and beliefs over time

Variable	Baseline*		3-months*		6-months*	
	n=59	n=13	n=44	n=11	n=35	n=11
Age	39.1 (14.1)	38.8 (14.1)				
Gender (female)	46 (78.0%)	11 (84.6%)				
Previous WAD history	15 (25.4%)	5 (38.5)				
Insurance (tort)	42 (72.4%)	13 (100%)				
Painful body sites	2.9 (1.7)	2.8 (1.6)				
Hospitalization	0 (0.0%)	0 (0.0%)				
Pain intensity	5.2 (2.0)	5.2 (1.9)	2.9 (2.5)	2.5 (1.8)	3.1 (2.6)	2.6 (1.9)
Disability (WDQ)	59.8 (32.6)	55.9 (29.8)	34.5 (33.7)	32.5 (29.8)	34.7 (35.9)	30.7 (31.7)
SOPA-Control	11.2 (3.8)	11.8 (5.0)	12.0 (4.0)	13.7 (4.3)	11.3 (5.4)	12.1 (5.6)
SOPA-Disability	7.5 (3.5)	7.6 (4.0)	5.9 (4.7)	5.5 (4.9)	5.8 (5.1)	5.7 (4.9)
SOPA-Harm	8.8 (3.3)	8.0 (4.5)	7.5 (3.9)	7.6 (4.0)	6.6 (4.2)	7.3 (5.8)
SOPA-Emotion	9.7 (5.5)	9.6 (5.2)	9.0 (4.7)	10.4 (5.4)	8.7 (4.6)	9.9 (6.6)
SOPA-Medication	11.8 (3.8)	11.6 (4.5)	10.7 (4.4)	9.9 (5.3)	11.5 (4.5)	10.0 (4.8)
SOPA-Solicitude	7.3 (4.8)	6.2 (4.2)	5.1 (4.0)	7.5 (4.4)	5.8 (5.2)	5.1 (4.4)
SOPA-Medical cure	11.9 (3.2)	11.6 (4.5)	10.0 (3.4)	11.7 (4.8)	9.2 (4.4)	8.9 (4.9)
PBPI-Mystery	-2.6 (3.8)	-3.4 (3.3)	-2.3 (4.0)	-1.7 (4.4)	-2.2 (4.4)	-3.3 (4.1)
PBPI-Permanence	-4.4 (4.0)	-4.5 (3.8)	-3.0 (4.7)	-2.3 (6.9)	-1.5 (5.4)	-1.1 (5.6)
PBPI-Constancy	1.1 (4.0)	1.5 (4.0)	-1.2 (5.0)	-0.4 (5.9)	-1.4 (5.4)	-0.2 (4.9)
PBPI-Self-blame	-4.3 (2.1)	-4.2 (1.7)	-3.9 (2.1)	-4.9 (1.6)	-3.2 (3.3)	-5.1 (1.3)
Catastrophizing	24.9 (9.2)	24.0 (10.4)	22.6 (9.9)	21.1 (8.5)	24.2 (11.5)	23.8 (14.9)

*values reported as mean (standard deviation)

Bold indicates significant at the $\alpha < 0.02$ (Bonferroni correction)

WAD=whiplash associated disorder, WDQ=whiplash disability questionnaire, SOPA=survey of pain attitudes, PBPI=pain beliefs and perceptions inventory

TABLE 3-2

Characteristics of the interview sub-sample.

Participant alias	Characteristics				
	Age (years)	Gender	Baseline pain	History of WAD?	Recovery status*
Cheryl	22	female	8/10	No	recovered
Jennifer	39	female	6/10	Yes	not recovered
Randall	40	male	6/10	No	not recovered
Claire	37	female	6/10	Yes	recovered
Stephanie	66	female	1/10	No	recovered
Stacey	30	female	4/10	Yes	unknown
Sheila	45	female	6/10	Yes	recovered
Danielle	34	female	6/10	Yes	not recovered
Dana	62	female	5/10	Yes	not recovered
Kim	27	female	7/10	No	unknown
Jason	29	male	3/10	No	recovered
Charlene	24	female	6/10	No	not recovered
Vanessa	52	female	missing	Yes	recovered
Leanne	52 year-old female with a 17-year history of chronic pain attributed to MVC				
Dawn	47 year-old female with a 8-year history of chronic pain attributed to MVC				
Candice	44 year-old female with a 8-year history of chronic pain attributed to MVC				
Linda	22 year-old female with a 3-year history of chronic pain attributed to MVC				

*recovered' based on interview discussions; MVC=motor vehicle collision

3.3.3. *Thematic analysis of experiences explaining endorsed beliefs*

Based on the interviewee's survey scores, four categories of beliefs from the SOPA, PBPI and PCS were most commonly identified as important within the interviewee sub-sample: SOPA-Medical Cure, SOPA-Control, SOPA-Emotion, and PBPI-Mystery. Thus, the thematic analysis presented below explained the experiences informing these four specific beliefs. In addition, the proceeding thematic analysis provides adjunctive inter-subjective meaning to that provided by the quantitative representation of meaning above.

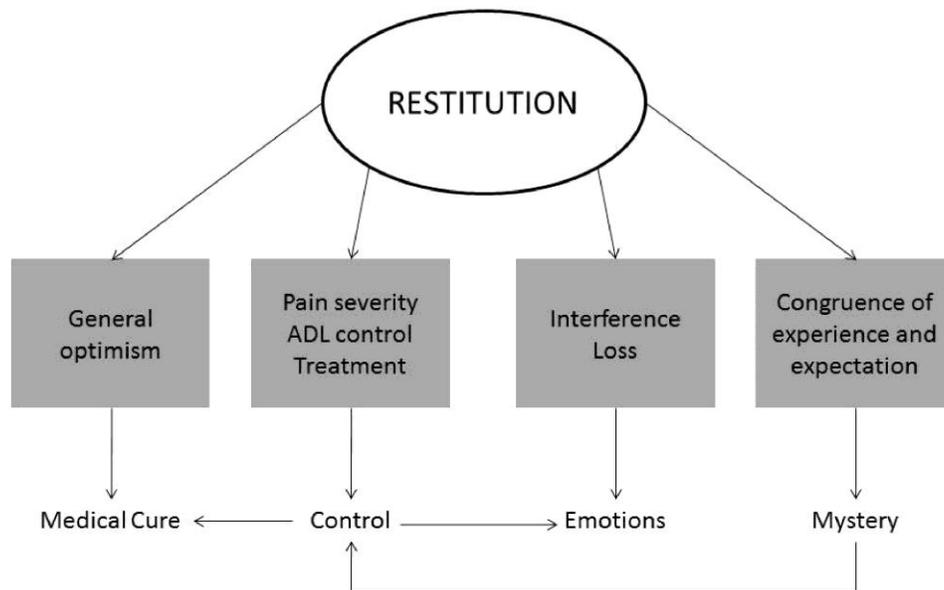
Figure 3-2 gives a visual representation of the outcome of the thematic analysis. The specific beliefs medical cure, control, emotions and mystery identified from the survey were informed by sub-themes. The sub-themes informing a belief in medical cure included and 'general optimism'. Control beliefs were determined by 'pain severity', 'control of activities of daily living' and 'treatment success'. Whether participants believed in a relationship between emotions and pain was governed by the sub-themes 'interference' and 'loss'. The belief in the mysteriousness of pain was based on the degree of 'congruence' between expectations and experience. In addition, the specific beliefs control and mystery appeared as sub-themes informing medical cure and emotions and control respectively. Finally, these sub-themes were found to be informed by the meta-theme restitution.

Presentation of the qualitative findings begins with an overview of the origin and description of the meta-theme. Following this, sub-themes informing each specific belief are briefly discussed with supportive verbatim quotes presented in Table 3-3. When possible, two quotations for each theme are provided in Table 3-3: the first from participants whose survey and interview suggested recovery and the second from participants who had not yet recovered. The participant's alias,

measurement occasion, and associated beliefs sub-scale score are also provided with each quote in Table 3-3. Finally, since the quantitative and qualitative beliefs suggested stability over time, the findings are not separated according to measurement occasion.

FIGURE 3-2

Visual representation of the findings from the thematic analysis.



The meta-theme of restitution was found throughout the sub-themes informing the specific beliefs medical cure, control, emotions, and mystery. Thematic analysis revealed relationships between control and emotions and mystery and control. ADL = activities of daily living

Meta-theme: restitution

The meta-theme restitution appeared to be a unifying construct across all sub-themes informing the four beliefs: SOPA-Medical Cure, SOPA-Control, SOPA-Emotion, and PBPI-Mystery. This term was derived from Arthur Frank's⁵⁷ writings on illness narratives. The restitution illness narrative is one commonly told by those who are recently injured and is congruent with modernist views of health (i.e. the medical model).¹⁹ That is, anyone who is recently injured wants to return to his or her pre-injury level of function. Frank⁵⁷ cites Talcott Parson's^{59,60} to capture the essence of the restitution narrative: "the journey into the kingdom of illness is a limited one, from which return is both expected and possible".^{57(p.82)} This meta-theme initially emerged as a theme within the specific belief medical cure wherein participants used language such as "fix" to describe the desired outcome of treatment.

"I don't know if it's my belief in science or something like that... They can put a man on the moon they should be able to fix a (neck) injury." – Randall

Although some participants had difficulty interpreting the term 'cure' when applied to pain and physical therapy or chiropractic treatment, interviewees expressed a desire to be returned to a pre-injury level of pain and function.

"Pain is really difficult to cure... With cure always mean not just taking the symptoms away but actually removing the reasons for and in that perspective I don't think there is a cure for whiplash. I think there is in a certain way there is a treatment, there is a treatment to help the body cure itself." – Claire

This quote also highlights potential difficulty when viewing pain through a restitution lens. While a restitution narrative provides impetus to seek remedy, matters are complicated when pain persists. For example, the patho-anatomical cause of pain is often elusive when pain persists. If a cause is not ascertained, then

fixing the problem is difficult. In a restitution narrative, a once potentially adaptive therapeutic goal of fixing or curing early after injury becomes a maladaptive belief as pain persists. This conflict was apparent for this participant:

“I haven't accepted it. I still think that hopefully somewhere down the line that they will find something that will help... I think I just think that because I don't want to deal with the reality of it. Because I think that after eight years and everybody telling me, there's nothing, there's nothing, there's nothing, that really I should accept the fact that there is nothing out there for me.” – Candice (participant with chronic pain)

Sub-themes informing beliefs

The SOPA beliefs pertaining to a belief that medicine can cure pain, pain is controllable, emotions have an impact on pain, and the PBPI belief that pain is mysterious were informed by the sub-themes described below with specific examples using verbatim quotes provided in Table 3-3. Participants had varying degrees of endorsement of each belief. The sub-themes identified for each belief represent potential reasons for endorsing or rejecting them. As mentioned, restitution was an over-arching meta-theme unifying these sub-themes. Evidence of restitution within these sub-themes is provided.

SOPA-Medical cure:

As described above, participants often used terms such as ‘fix’ when discussing recovery predictions. This language implies the body is a machine. Machines are indeed fixable, and if not they can be rebuilt. Viewing the body mechanistically is characteristic of restitution. Discussions of cure inevitably led to the topic of optimism in most interviews. Optimism implied a belief that pain experiences are likely remediated. Optimism could also be construed as positive expectancies, which are defined as “patient predictions about (positive) future outcomes and consequences of their health condition”.⁶¹ The two sub-themes explaining optimism included: (1) the controllability of pain and (2) general belief in

remaining optimistic (Table 3-3). Control of pain was an important factor in experiencing optimism. Exerting control over pain meant experiencing predictability, leading to a sense that pain will be overcome. Conversely, difficulties with pain control led to challenges in retaining a positive outlook about recovery.

Optimism related to WAD was also a consequence of a general belief in optimism. This theme may reflect a natural desire to get well and stay well. Many participants spoke of the importance of being an optimistic person in all aspects of life. This is characteristic of restitution insofar as society prefers stories of perseverance and recovery.⁵⁷ The notion that we are socialized to achieve restitution was well captured in one participant's account:

“I feel like no one cares, just because in our society, when we have a person who falls down ... they're expected to recover, ... it's just the way our society wants us to deal with it.” – Danielle

SOPA-Control:

The desire for cure is likely to lead to actions to control the situation. The injured person wants the body's former predictability back again: not simply regaining mechanical functioning but also staving off the contingency represented by illness itself: disability.⁵⁷ In these data, the controllability of one's pain was determined by four sub-themes: (1) perceived severity of the injury/pain, (2) understanding (3) control over daily demands, and (4) treatment success. (Table 3-3) The first sub-theme, perceived pain intensity or injury severity, was tacitly understood as a factor influencing pain controllability. That is, low levels tended to equate to less injury leading to endorsement of the belief that pain is controllable. Conversely, higher levels of pain were suggestive of injury that is more significant and tended to lead to a belief that pain is not well controlled. Another factor that led to the perception of pain controllability, was the experience that one's pain was understandable. The experience of pain could be viewed as a riddle needing

answers, especially in the context of unexpected unresolved pain. For many participants, an ability to understand one's pain leads to predictable strategies to control it.

The need to reconnect with the pre-injury self may lead to the desire to remain productive. For example, interviewees often indicated it was not necessarily pain that was difficult to control, but rather the inability to avoid various activities of daily living. Thus, the need (or desire) to complete certain activities could not be avoided leading to pain exacerbation and thus the belief that pain was not well controlled. Finally, Frank's⁵⁷ contention that restitution is filled with talk about tests and their interpretation, treatments and their possible outcomes, the competence of physicians, and alternative treatments was also seen in these narratives.⁵⁷ Indeed, participants equated their perception of the controllability of pain, especially in the latter stages of recovery, with the degree of success experienced with treatments.

SOPA-Emotion:

The relationship between pain and emotions appeared to be tacitly understood as one participant said:

“I think that's common knowledge ... I think that's just common sense, isn't it?” – Stacey

Two sub-themes primarily explained the endorsement that pain and emotions were related: (1) the ability to control one's pain and (2) interference and loss (Table 3-3). In addition, a potential third sub-theme surfaced: stigma. Although only superficially explored, it is worth mentioning as it provides additional context to a belief in the association between emotions and pain after WAD.

A perceived lack of control over pain impaired the peace of mind sought when attempting to recover from injury and could lead to emotional strain. For example, uncontrolled pain was disruptive and often led to frustration and feelings of fear,

distress, sadness and worry. Conversely, gaining control or predictability meant becoming closer to restoring pre-injury daily function. Approximating normalcy led to positive feelings such as happiness, self-satisfaction and optimism.

“In the face of illness or injury, the body may turn in upon itself and separate from the self ... this temporarily broken-down body becomes “it” to be cured.”^{57(p.85)} Until “it” is cured and reintegrated with the self, role fulfillment is difficult. The emotional strain associated with pain had much to do with this forced separation of body and self due to interference and loss. Pain interfered with accomplishing usual activities. Interference led to lost opportunities for social interaction and role fulfillment. Thus, the degree of departure from normalcy appeared to create emotional strain. As pain persisted and puzzles remained unsolved, it was not surprising that frustration followed.

While most participants believed in a relationship between pain and emotions, ascertaining the specific relationship between the two was challenging. The tacit assumption that emotional reactions occur only as result of pain (and not the other way around) was initially prevalent. However, deeper probing into this relationship revealed a possible mediation by a lack of control, interference and loss. The difficulty in participants ascertaining how pain and emotions were related was prevalent and mirrors the challenges interpreting the SOPA Emotion scale: is a belief that emotions affect pain adaptive, or a consequence of psychological distress?

As mentioned, stigma surfaced as a potentially important factor in explaining beliefs about the relationship between pain and emotions. Stigma refers to an experience where a ‘marked’ person is viewed inferior or in a negative light by those who are ‘unmarked’.⁶² A number of participants either experienced or perceived a stigma associated with WAD. The invisibility of pain is a well-established characteristic in the chronic pain literature, especially in the absence

of a clear explanation.⁶³ Specific to WAD, this participant provided an illuminating metaphor:

“The hood actually was just wrinkled up a bit and the bumper was pushed in, but everything under the hood was moved over six inches, making the vehicle completely unstable. The guy that did the assessment just said to me, “I’m not even gonna look at it anymore... It’s a write-off.” And that’s kind of, almost [laugh] what I feel like sometimes, like, um, you can’t see that I’m injured, because I go to work and I do stuff with my daughter’s school, and I carry on my life as normal as I can, but everything is harder... So, I kind of feel like that car, like fine on the outside, but inside all broken” – Jennifer (baseline)

While pain is not often associated with an outwardly observable mark leading to isolation from those unmarked, stigmatization is nonetheless experienced and perhaps is exaggerated in the case of WAD. Not only is pain invisible, but sufferers may also experience explicit or implicit insinuations of malingering and secondary gain.

PBPI-Mystery:

Many participants desired understanding of their injury/pain as it provided a means of eliminating the pain or fixing the problem. For example, one participant stated

“... if you think you understand it at least then you take what you think are appropriate actions to help fix it.” – Stephanie

In general, the belief that pain was mysterious appeared to be most prevalent during follow-up interviews. In other words, pain early on after the MVC was not surprising. However, for some participants, as the pain persisted beyond expected timelines, the discussion about the mysteriousness of pain was more relevant. Whether pain was viewed as mysterious was primarily due to the sub-theme of incongruence. (Table 3-3) That is, when expectations for recovery were not congruent with the experience, confusion resulted.

TABLE 3-3

Sub-themes generated from the qualitative analysis along with supportive verbatim quotes.

Belief	Sub-theme(s)	Verbatim quote
Medical cure	<p>Optimism</p> <p>1. Control over pain</p> <p>2. Generally an optimist</p>	<p><i>... its nothing he cannot fix it, so I believe in him, he did a good job last time ... – Sheila (baseline; 20/20)</i> <i>The more time goes on and you don't see a difference, it's harder to remain optimistic... – Jennifer (baseline, 9/20)</i></p> <p><i>Because you have to believe something... I don't know, I read something years ago about if you go to a doctor he has to believe he can help you and you have to believe he can help you otherwise you are not going to experience much... - Stephanie (baseline; 11/20)</i> <i>I think if you leave hope all together you're done. You would never (get) past the depression. – Dawn (chronic pain participant; 2/20)</i></p>
Control	<p>1. Pain/injury severity</p> <p>2. Understanding</p> <p>3. Control over daily demands</p> <p>4. Treatment success</p>	<p><i>I'm still walking [laughter] and talking, so, I might have a, something that's being fixed, but it's not all broken – Vanessa (baseline; 18/20)</i> <i>But I constantly feel something, so I don't really have much control... – Charlene (3-months; 9/20)</i></p> <p><i>I knew what make it worse and what would make it better, so that helped a lot. I think that's what made me feel like I could do something to change it, like take control... – Jason (6-months; 18/20)</i> <i>Sometimes I can do something that I don't think is gonna be a problem and it is... So it feels like, how much less can I do and still get the things done that I need to do ... Gosh, I don't have a clue. I have no clue – Jennifer (6-months; 6/20)</i></p> <p><i>... say, I have a full day of events, I definitely will be taking that Advil or ibuprofen just to, just to ease the pain a little bit more – Cheryl (baseline; 9/20)</i> <i>I mean I can't avoid doing the dishes and I can't avoid getting a little stressed out when some one in my life is upsetting me... I'm no where near the mind master to be able to not let it bother me – Linda (participant with chronic pain; 2/20)</i></p> <p><i>It's important to be able to talk it out with somebody, too, to have the support ... they've been good at helping set some limits or giving you some direction – Vanessa (baseline; 18/20)</i> <i>Well I tried chiropractic and physio and massage and IMS and nothing is really, I mean they are all temporary – Jennifer (6-months; 6/20)</i></p>

Emotions	1. Control	<p><i>You feel, um, like you can carry on, whereas before, um, day-to-day it was difficult to know how things would be and it was really distressing because one day looked exactly like the next day – Vanessa (baseline; 18/20)</i></p> <p><i>Oh, it makes me feel like hell, because there's nothing you can do about it... Um, everything hurts, everything aches, and I don't have to go over it, there's nothing I can do... I kind of feel helpless – Danielle (baseline; 7/20)</i></p>
	2. Interference and Loss	<p><i>a little bit frustrated because now ... a lot of my own personal time I have to devote to working that I've missed, time working. I have to make up that time that I've missed... – Stacey (baseline; 12/20)</i></p> <p><i>Work wise, I can't go. My marriage isn't very stable at this point at all. I'd be surprised if it's even there in six months. I lost out on all kinds of family functions... I don't see friends anymore. I have really no life. It revolves around doctors and medicine. – Dawn (participant with chronic pain; 10/20)</i></p>
	3. Stigma*	<p><i>I think in my case even when I felt my neck was a little sore I was reluctant to say maybe that's whiplash, because whiplash has a stigma of fraud about it. – Stephanie (6-months; 3/20)</i></p> <p><i>I think it's just because they can't see it, they can't prove it on a scientifically basis. It's frustrating because I know I get it. I know how bad it gets ... It's very stressful, it's very depressing. And you feel like nobody's on your side. – Charlene (6-months; 1/10)</i></p>
Mystery	1. Incongruence	<p><i>... so if I'm not getting the right results and I'm doing what they tell me to do, then maybe their diagnosis is wrong... I should've seen some sort of benefit and I didn't. – Jennifer (3-months; 3)</i></p> <p><i>Generally, I ask her if it's normal for this to be going on this long. And she goes, yes it is... things will just fix themselves – Randall (6-months; 0)</i></p>

*not stigma was superficially explored and is highlighted for its potential salience for further study

When recovery from pain does not progress in a manner that is expected, it highlights one of the most important limitations of living in a restitution narrative: when restitution no longer ‘fits’. If there is no longer a sensible way to make meaning of one’s injury, it is both difficult to resume one’s pre-injury self or move toward creating a new self. When one believes in the power of modern medicine’s reductionist assumption that all pathology can be remediated, one is inevitably confused when this expectation is unmet. However, it is important to highlight that simply understanding one’s pain might not lead to reducing it. In analyzing negative cases and member check materials, one anonymous participant stated:

“My recovery did not match the expected recovery however, I understand my pain ... understanding pain helps you to learn your limitations ... It does not change how much pain you have. It does not help you deal with the effects on your life or relationships/work.” - Anonymous

This comment highlights the complexity of pain – simply understanding one’s pain does not necessitate an understanding of suffering.

Trustworthiness of the Qualitative Analysis

Appendix C contains findings from the trustworthiness analysis. Included are the findings from the member check process and suggests that participants agreed with the study’s interpretations. Participants were contacted three times and encouraged to send comments especially if they did not agree with the study’s interpretations. They were also told that no response to the member check forms would be considered as meaning there were no major concerns with the interpretations. Of the 17 interview participants, 5 participants returned the member check form. The confirmability audit findings are also included and support the researchers’ interpretations and analysis process. Finally, an illustration of generated sub-themes dissimilar to, or in contrast with, the

presented thematic analysis is provided (i.e. negative case analysis). Anonymous verbatim transcripts are available upon request to the corresponding author (GB).

3.4. Discussion

This study represents a novel examination of WAD-related pain beliefs and the experiences that inform them in a longitudinal mixed-methods study. Of particular salience is the identification of the meta-theme restitution, or the desire to be ‘fixed or ‘cured’ from pain, and numerous sub-themes that explain the endorsement or rejection of WAD-related pain beliefs. While much of the knowledge regarding the meaning of pain in musculoskeletal conditions such as WAD is situated in chronic pain literature, we found that even in early stages of the condition, WAD pain meaning shares similarities with themes from the chronic pain literature. We also found that beliefs were relatively stable over time. Finally, stigma arose as a potentially important characterization of the experience of WAD.

Grounded in the qualitative data was the emergence of the meta-theme restitution. For example, the desire to be returned to pre-injury level of health was manifest in the data by participants seeking control, normalcy and cure. This emphasizes the adaptive role this narrative assumes in the early stages of injury. However, it also stresses the difficulty in viewing pain through this lens when it does not respond in an anticipated manner. This raises important questions about how pain is best understood by the sufferer: at what point is the restitution narrative no longer helpful? What role do health professionals have in facilitating an awareness of alternative narratives? Since health providers have been encouraged to shift from a curative approach to pain toward a management approach,⁶⁴ explicit identification of the restitution narrative is important as it potentially represents an incompatibility in the conceptualization of pain by patient and provider.

Alternate narratives to restitution include chaos and quest.⁵⁷ In the chaos narrative, the individual living within this story is unable to find meaning in their pain or illness. In the quest narrative, the individual is defined as a new person and finds enjoyment in life despite pain. These narratives clearly differ from restitution and it is thus reasonable to expect that WAD-specific beliefs would also differ if the narrative changed. Analysis of negative cases demonstrated some evidence of these other narratives, but they did not dominate any interview. Measuring and probing beliefs within a six-month period may not be enough time to move in and out of various narratives. However, with the addition of four interviews with people with chronic WAD pain, it was hypothesized that more variety in pain meaning would occur. Interestingly, these participants also demonstrated evidence of experiencing their pain within a restitution narrative. This may be supportive of the assertion that society prefers to hear stories of restitution.⁵⁷

In addition to the meta-theme restitution, deeper understanding of factors informing beliefs was achieved. This provided adjunct inter-subjective meaning to the quantitative meaning characterized in the survey measures. While some sub-themes informing beliefs appeared tacitly understood, qualitative data reinforced the importance of constructs in the quantitative literature. For example, the primary sub-theme informing the belief that pain is mysterious was incongruence. Incongruence occurred when expectations did not match experience, highlighting the importance of appropriate expectations. Negative expectancy is an important factor in influencing negative outcomes in a variety of conditions,⁶⁵ including WAD.⁶⁶ Considering that the majority of patients with WAD have a favourable prognosis,^{67,68} expectations more in line with this research may be helpful. The thematic analysis also illuminated inter-relationships among beliefs. For example, medical cure, a similar construct to expectations, highlighted the importance of control. Data such as these may contribute to strategies to empirically examine a

priori relationships to help refine theoretical models that have been recently scrutinized.⁶⁹

In general, quantitative data suggested most beliefs were stable over time. This finding supports other evidence that suggest, in the absence of targeted interventions, beliefs remain stable over time.¹⁴The qualitative data also suggested relative stability insofar as the unifying meta-theme (restitution) appeared to be pervasive throughout the thematic analysis. It is thus possible that restitution also influences the degree of endorsement of other beliefs. Moreover, if pain beliefs are partly culturally determined then it is likely that the public would share similar beliefs characterized by restitution narratives. Indeed, Frank⁵⁷ contends that the restitution narrative is society's preferred story. This has implications for targeted education initiatives. If observed beliefs are the target of change, perhaps the underlying narrative restitution should be the factor addressed. Considering the frequency of restitution throughout the data and the apparent stability of this belief across early and late stages of injury, a paradigm shift away from viewing pain as something to be 'fixed' may be required to modify maladaptive beliefs.

No known literature exists examining the meaning of pain as characterized by qualitative analysis of belief in a longitudinal cohort of patients with WAD. Despite the unique context of WAD, some shared meaning exists with studies examining chronic pain. For example, the observed themes of loss and interference are also common in the chronic pain literature.^{70,71} In addition, while emotional responses to pain are often presumed to occur primarily in people with chronic pain, similar emotional responses were reported in this study, particularly as it related to poor control of pain. Finally, much is written about the invisibility of chronic pain and its associated stigma.^{22,72} In addition to the sub-themes informing beliefs, the emergence of stigma within the qualitative data, even in the acute/sub-acute stages, provided an important context for WAD. As Elaine Scarry⁷³ states: "to have pain is to have certainty, to hear of another's pain is to

have doubt.”^{73(p.4)} This stigma of doubt and perceived malingering likely adversely affects one’s meaning of pain and could be characterized by a state of liminality.⁷⁴ Liminality can be thought of a state where one lacks social status. Liminality creates stigma since dogmatic views of pain characterized by dualism are incompatible with the experience of living with WAD; a condition not readily characterized by observable patho-anatomy, even in early stages of injury. Thus, communicating pain becomes difficult as current societal views have no way to rationalize chronic pain other than being a fault of the sufferer.⁷⁴ A reduced comfort in communicating pain due to stigma could lead to isolation and therefore an impairment in social status. Although the sub-theme of stigma surfaced and represents a potentially important factor in elucidating the meaning of WAD pain, it was only superficially explored in this study and thus warrants further investigation.

There are limitations of this study that require consideration. This cohort does not entirely represent the wider WAD population. While some important demographics are similar (age and pain intensity), this cohort contained a higher proportion of females compared to the sampling frame. However, given differences cited in perceived pain and belief between sexes,^{14,22,75} this participant sample provides an important female perspective. In addition, the rate of attrition in this study was relatively high. This occurred despite efforts to maximize response, such as follow-up letters and phone calls. Comparison of demographic data and measures of pain, disability and beliefs between participants with and without complete data suggest a systematic difference for sex only.

A limitation of the qualitative data is different interpretations may have been made by other researchers. It is impossible to view and interpret data from a completely blank slate. Thus, other analysts with differing reflexivity are likely to interpret the data differently. In addition, the fact that quantitative data determined the interview guide means that it constrained the qualitative results. Thus,

participants were not able to construct their own meaning. To mitigate this, interviewees were asked an open-ended question about pain meaning at the start of the interview to determine if meaning was not adequately captured within the prescribed survey categories (data not presented here). No novel beliefs were volunteered with the exception of stigma. To maximize trustworthiness, measures were taken to ensure the interpretations were transparent, credible, dependable and confirmable.

To our knowledge, this is the first study using mixed-methods to explore the meaning of pain in WAD. This approach appears useful for characterizing beliefs and explaining factors underlying the formation of WAD-related pain beliefs. In addition to allowing us to characterize meaning through a quantitative portrayal of beliefs, an additional deeper understanding of meaning was achieved through exploring detailed personal experiences. Providing this adjunct inter-subjectivity to the quantitative portrayal of beliefs enhances the credibility of the study.⁷⁶

It is acknowledged that some authors argue quantitative and qualitative methods are too divergent in epistemology to permit integration.²⁰ Despite apparent incompatibilities in philosophy and epistemology cited by these authors, others are able to reconcile the two.⁷⁷ For example, Greene³⁰ provides a number of potential grounds to justify mixing methods ranging from alternative paradigms such as pragmatism, to the complementary strengths stance that posits the two paradigms are not overly divergent,⁷⁸ or a dialectical stance that view these paradigms as social constructions and not sacrosanct.⁷⁹ This study followed a pragmatic stance where purists perspectives are valued and a moderate position employed.

Congruent with contemporary views on pain, illness and injury,¹ this study characterized beliefs from a quantitative statistical perspective and attained an additional deeper understanding of inter-subjectivity by integrating a qualitative

perspective. The restitution narrative was the dominant story unifying the experienced underlying patterns of belief endorsement. The desire to be 'fixed' or 'cured' was manifest in sub-themes related to achieving control and normalcy and expectations of health care providers to provide a remedy. In addition, the data demonstrated a general pattern of stability in beliefs over time. Finally, the meaning of pain throughout early and late stages shared commonalities with meaning in chronic pain. In particular, stigma appeared to be an emerging factor that may be important in further illuminating what it means to experience WAD.

3.5. References

1. Morris, DB. Sociocultural and Religious Meanings of Pain. In: Gatchel RJ and Turk DC eds. Psychosocial Factors in Pain: Critical Perspectives. 1999. The Guilford Press: New York.
2. Fishbein M, Ajzen I. Belief, attitude, intention and behavior: An introduction to theory and research. Reading, MA: Addison-Wesley, 1975.
3. Wrubel J, Benner P, Lazarus RS. Social competence from the perspective of stress and coping. In: Wine J, Syme M editors. Social competence. New York (NY): Guilford Press, 1981.
4. Williams DA & Thorn BE. An empirical assessment of pain beliefs. Pain 1989; 36(3):351-358.
5. Jensen MP, Turner JA, Romano JP, Karoly P. Coping with chronic pain: A critical review of the literature. Pain 1991; 47(3): 249-283.
6. Buitenhuis J, de Jong PJ, Jaspers JP, Groothoff JW. Catastrophizing and causal beliefs in whiplash. Spine 2008; 33(22):2427-2433.
7. Colloca L, Benedetti F. Placebos and painkillers: is mind as real as matter? Nature Rev Neurosci. 2005; 6(7): 545-552.
8. Bingel U, Lorenz J, Schoell E, Weiller C, Buchel C. Mechanisms of placebo analgesia: rACC recruitment of a subcortical antinociceptive network. Pain. 2006; 120(1-2): 8-15.
9. Benedetti F, Lanotte M, Lopiano L, Colloca L. When words are painful: unraveling the mechanisms of the nocebo effect. Neuroscience. 2007; 147(2): 260-271.
10. Schrader H, Obelieniene D, Bovim G, Surkiene D, Mickeviciene D, Miseviciene I, et al. Natural evolution of late whiplash syndrome outside the medicolegal context. *Lancet* 1996;347:1207-11.
11. Obelieniene D, Schrader H, Bovim G, Miseviciene I, Sand T. Pain after whiplash: a prospective controlled inception cohort study. *J Neurol Neurosurg Psychiatry* 1999;66:279-83.

12. Cassidy JD, Carroll LJ, Cote P, Lemstra M, Berglund A, Nygren, A. Effect of eliminating compensation for pain and suffering on the outcome of insurance claims for whiplash injury. *New Eng. J. Med.* 2000; 342:1179-1186.
13. Bostick GP, Ferrari R, Carroll LJ, Russell AS, Buchbinder R, Krawciw D, Gross DP. A population-based survey of beliefs about neck pain from whiplash injury, work-related neck pain, and work-related upper extremity pain. *Eur J Pain* 2009; 13(3):300-4.
14. Sullivan MJL, Thorn B, Haythornwaite JA, Keefe F, Martin M, Bradley LA, Lefebvre JC. Theoretical perspectives on the relation between catastrophizing and pain. *Clin J Pain* 2001; 17:52-64.
15. Bunketorp,L.; Lindh,M.; Carlsson,J.; Stener-Victorin,E. The perception of pain and pain-related cognitions in subacute whiplash-associated disorders: its influence on prolonged disability. *Disabil Rehabil* 2006;28(5):271-279.
16. Adams H, Ellis T, Stanish WD, Sullivan MJL. Psychosocial factors related to return to work following rehabilitation of whiplash injuries. *J Occup Rehabil* 2007;17:305-315.
17. Vangronsveld K, Peters M, Goossens M, Linton S, Vlaeyen J. Applying the fear-avoidance model to the chronic whiplash syndrome. *Pain* 2007;130:258-61.
18. Williamson E, Williams M, Gates S, Lamb SE. A systematic literature review of psychological factors and the development of late whiplash syndrome. *Pain* 2008;135(1-2):20-30.
19. Morris, DB. How to speak postmodern: medicine, illness, and. cultural Change. *Hastings Center Report* 2000; 30(6):7-16.
20. Lincoln YS, Guba EG. Paradigmatic controversies, contradictions, and emerging confluences. In: Denzin NK, Lincoln YS, editors. *The landscape of qualitative research*, 2nd ed. Thousands Oaks (CA): Sage Publications; 2003.

21. Verbeek J, Sengers MJ, Riemans L, Haafkens J. Patient expectation for treatment of back pain: a systematic review of qualitative and quantitative studies. *Spine* 2004;29:2309-2318.
22. Johansson EE, Hamberg K, Westman G, Lindgren G. The meanings of pain: an exploration of women's description of symptoms. *Soc. Sci. Med.* 1999; 48:1791-1802.
23. Osborn M, Smith JA. Living with a body separate from the self. The experience of the body in chronic benign low back pain: an interpretative phenomenological analysis. *Scand J Caring Sci* 2006; 20:216-222.
24. Allcock N, Elkan R, & Williams J. Patients referred to a pain management clinic: beliefs, expectations and priorities. *J Adv Nurs* 2007;60:248–256.
25. Krohne K, Ihlebaek C. Maintaining a balance: a focus group study on living and coping with chronic whiplash-associated disorder. *BMC Musculoskeletal Disorders* 2010; 11:158-164.
26. Johnson RB, Onwuegbuzie AJ. Mixed methods research: a research paradigm whose time has come. *Educational Researcher.* 2004;33(7):14-26.
27. Hoshmand LT. Can lessons of history and logical analysis ensure progress in psychological science? *Theor Psychol.* 2003;13(1):39-44.
28. Jencks C. *What is Post-Modernism?* 2nd ed. New York (NY): St. Martins' Press; 1987.
29. Creswell, JW. *Research design: qualitative, quantitative, and mixed-methods approaches.* 3rd ed. 2009 Sage Publications Ltd.: Thousand Oaks, CA.
30. Greene JC. Is mixed methods social inquiry a distinctive methodology? *J Mix Methods Res.* 2008;2:7-22.
31. Carroll LJ, Holm LW, Hogg-Johnson S, Cote P, Cassidy DJ, Haldeman S, Nordin M, Hurwitz EL, Carragee EJ, van der Velde G, Peloso PM, Guzman J. Course and prognostic factors for neck pain in whiplash-associated-disorders (WAD): Results of the Bone and Joint Decade 2000-

- 2010 Task Force on Neck Pain and its Associated Disorders. *Spine* 2008;33(4):S83-S92.
32. Beaton DE, Tarasuk V, Katz JN, Wright JG, Bombardier C. "Are you better?" A qualitative study on the meaning of recovery. *Arthritis Care Res* 2001;45:270-279.
 33. Akparian AV, Baliki MN, Geha PY. Towards a theory of chronic pain. *Prog Neurobiol.* 2009;87(2):81-97.
 34. Fossey E, Harvey C, McDermott F, Davidson L. Understanding and evaluating qualitative research. *Aust and New Zeal J Psychiatry* 2002;36:717-732.
 35. Carter S, Henderson L. Approaches to qualitative data collection in social science. In: Bowling A, Ebrahim S. *Handbook of health research methods.* New York (NY): Open University Press, 2005.
 36. Pinfold M, Niere KR, O'Leary EF, Hoving JL, Green S, Buchbinder R. Validity and internal consistency of a whiplash-specific disability measure. *Spine* 2004;29:263-268.
 37. Dworkin RH, Turk DC, Farrar JT, Haythornthwaite JA, Jensen MP, Katz NP, Kerns RD, Stucki G, Allen RR, Bellamy N, Carr DB, Chandler J, Cowan P, Dionne R, Galer BS, Hertz S, Jadad AR, Kramer LD, Manning DC, Martin S, McCormick CG, McDermott MP, McGrath P, Quessy S, Rappaport BA, Robbins W, Robinson JP, Rothman M, Royal MA, Simon L, Stauffer JW, Stein W, Tollett J, Wernicke J, Witter J, Impact. Core outcome measures for chronic pain clinical trials: IMMPACT recommendations. *Pain* 2005;113:9-19.
 38. Jensen MP, Turner JA, Romano JM. Pain belief assessment: A comparison of the short and long versions of the Survey of Pain Attitudes. *The Journal of Pain* 2000;1:138-150.
 39. Jensen MP, Karoly P, Huger R. The development and preliminary validation of an instrument to assess patients' attitudes toward pain. *J Psychosom Res* 1987;31:393-400.

40. Jensen MP, Turner JA, Romano JM. Relationship of pain-specific beliefs to chronic pain adjustment. *Pain* 1994;57:301-309.
41. Jensen MP, Turner JA, Romano JM. Correlates of improvement in multidisciplinary treatment of chronic pain. *J Consult Clin Psychol* 1994;62:172-179.
42. Jensen MP, Romano JM, Turner JA, Good AB, Wald LH. Patient beliefs predict patient functioning: Further support for a cognitive-behavioural model of chronic pain. *Pain* 1999;81:95-104.
43. Jensen MP, Turner JA, Romano JM. Changes in beliefs, catastrophizing, and coping are associated with improvement in multidisciplinary pain treatment. *J Consult Clin Psychol* 2001;69:655-662.
44. Jensen MP, Turner JA, Romano JM. Changes after multidisciplinary pain treatment in patient pain beliefs and coping are associated with concurrent changes in patient functioning. *Pain* 2007;131:38-47.
45. Williams DA, Thorn BE. An empirical assessment of pain beliefs. *Pain* 1989;36:351-358.
46. Williams DA, Keefe FJ. Pain beliefs and the use of cognitive-behavioural coping strategies. *Pain* 1991;46:185-190.
47. Herda CA, Siergeris K, Basler H. The Pain Beliefs and Perceptions Inventory: further evidence for a 4-factor structure. *Pain* 1994;57:85-90.
48. Williams DA, Robinson ME, Geisser ME. Pain beliefs: assessment and utility. *Pain* 1994;59:71-78.
49. Dysvik E, Lindstrom TC, Eikeland OJ, Natvig GK. Health-related quality of life and pain beliefs among people suffering from chronic pain. *Pain Manag Nurs* 2004;5:66-74.
50. Sullivan MJL, Bishop S, Pivik J. The Pain Catastrophizing Scale: Development and validation. *Psychol Assess* 1995;7:524-532.
51. Sullivan MJL, Bishop SR, Pivik J. The Pain Catastrophizing Scale: development and validation. *Psychol Assess* 1995;7:524-32.

52. Sullivan MJ, Adams H, Rhodenizer T, Stanish WD. A psychosocial risk factor-targeted intervention for the prevention of chronic pain and disability following whiplash injury, *Phys Ther* 2006; 86:8–18.
53. Sullivan MJL, Stanish W, Sullivan ME, Tripp D. Differential predictors of pain and disability in patients with whiplash injuries. *Pain Res Manag* 2002; 7: 68 – 74.
54. Erlandson DA, Harris EL, Skipper BL, Allen SD. *Doing naturalistic inquiry: A guide to methods*. Newbury Park: Sage Publications; 1993.
55. Bowling A. *Research methods in health: Investigating health and health services*. Philadelphia: Open University Press, 2002.
56. Kleinman A. *The illness narratives: suffering, healing and the human condition*. New York (NY): Basic books; 1988.
57. Frank AW. *The wounded storyteller: Body, illness, and ethics*. 1995 The University of Chicago Press: Chicago IL.
58. Lincoln YS, Guba EG. *Naturalistic inquiry*. Newbury Park: Sage Publication, 1985.
59. Parsons, T. (1978). Health and disease: A sociological and action perspective. In: Parsons T, *Action theory and the human condition*. New York (NY): Free Press; 1978.
60. Williams SJ. Parsons revisited: from the sick role to ...? *Health (London)* 2005; 5:123-145.
61. Janzen JA, Silvus J, Slaughter S, Dalziel W, Drummond N. What is a health expectation? Developing a pragmatic conceptual model from psychological theory. *Health Expectations* 2006;9(1):37-48.
62. Jones EE, Farina A, Hastorf AH, Hazel M, Miller DT, Scott RA. (1984). *Social Stigma*, W. H. Freeman, New York.
63. Clarke KA, Iphofen R. A phenomenological hermeneutic study into unseen chronic pain. *Brit J Nurs*. 2008;17(10):658-663.

64. Nicholas MK, George SZ. Psychologically informed interventions for low back pain: An update for physical therapists. *Physical Therapy* 2011;91(5):765-776.
65. Mondloch MV, Cole D, Frank J. Does how you do depend on how you think you'll do? A systematic review of the evidence for a relation between patients' recovery expectations and health outcomes. *CMAJ* 2001;165:174-9.
66. Carroll LJ; Holm LW, Ferrari R, Ozegovic D, Cassidy DJ. Recovery after whiplash associated disorders: do you get what you expect? *J Rheumatol* 2009;36:1063-1070.
67. Kamper SJ, Maher CG, Hush JM, Pedler A, Sterling M. Relationship between pressure pain thresholds and pain ratings in patients with whiplash-associated disorders. *Clin J Pain*. 2011;27(6):495-501.
68. Sterling M, Hendrikz, Kenardy J. Similar factors predict disability and posttraumatic stress disorder trajectories after whiplash injury. *Pain*; 2011;152(6):1272-1278.
69. Wideman TH, Adams H, Sullivan MJL. A prospective sequential analysis of the fear-avoidance model of pain. *Pain* 2010;145:45-51.
70. Bowman JM. The meaning of chronic low back pain. *AAOHN J*. 1991;39(8):381-384.
71. Seers K, Friedli K. The patients' experiences of their chronic non-malignant pain. *J Adv Nurs*. 1996;24(6):1160-1168.
72. Nettleton S. "I just want permission to be ill": towards a sociology of medically unexplained symptoms. *Soc Sci Med*. 2006;62(5):1167-1178.
73. Scarry E. *The body in pain*. Oxford: Oxford University Press; 1985.
74. Jackson JE. Stigma, liminality, and chronic pain: mind-body borderlands. *American Ethnologist*. 2005;32(3):332-353.
75. Riley, JL, Robinson ME, Wise EA, Myers CD, Fillingim RB. Sex differences in the perception of noxious experimental stimuli: a meta-analysis. *Pain*, 1998; 74:181-187.

76. Caron-Flinterman JF, Broerse JEW, Bunders JFG. The Experiential Knowledge of Patients: A New Resource for Biomedical Research? *Soc Sci Med* 2005; 60:2575-2584.
77. Howe KR. Against the quantitative-qualitative incompatibility thesis or dogmas die hard. *Educational Researcher*. 1988;17(8):10-16.
78. Morse JM. Principles of mixed methods and multimethod research design. In: Tashakkori A, Teddlie C, editors. *Handbook of mixed methods in social and behavioural research*. Thousand Oaks (CA): Sage; 2003.
79. Maxwell JA, Loomis DM. Mixed methods design: an alternative approach. In: Tashakkori A, Teddlie C, editors. *Handbook of mixed methods in social and behavioural research*. Thousand Oaks (CA): Sage; 2003.

CHAPTER 4

Predictive capacity of WAD-related pain beliefs and catastrophizing

4.1. Introduction

The prevalence of persistent neck pain after a motor vehicle collision (MVC), also known as whiplash associated disorder (WAD), is high and a significant burden to healthcare.^{1,2} Prognostic research suggests WAD outcomes are largely determined by initial pain intensity. However, psychological factors such as catastrophizing have also been shown to be important.^{1,3} The important role of psychological factors has increased attention toward cognitive-behavioural approaches to understanding pain.⁴ Understanding idiosyncratic beliefs underlying the experience of pain represents a central component of the cognitive-behavioural approach and has been deemed critical for optimal treatment.⁵

Useful models such as the fear-avoidance model⁶ have developed in low back pain, and synthesized literature has identified specific beliefs that influence low back pain outcomes.⁷ The study of WAD-related pain beliefs are less understood, especially outside the context of chronic pain. Evidence suggests that beliefs related to causation, catastrophizing, fear-avoidance, and self-efficacy are associated with WAD pain and disability.⁸⁻¹² Interventions falling under the umbrella of cognitive-behavioural theory propose that beliefs influence pain perception and adjustment to pain.¹³ Thus, beliefs deemed maladaptive are expected to relate with outcomes such as increased pain and disability.¹⁴⁻¹⁹ However, prognostic evidence regarding specific beliefs is inconsistent in WAD and requires further clarification.²⁰

Belief is a broad construct necessitating a broad range of measurement tools to capture the diversity of the construct. Measures such as the Survey of Pain Attitudes (SOPA) and Pain Beliefs and Perceptions Inventory (PBPI) have proven useful in chronic pain settings,^{15,16,19,21} but their prognostic utility is unknown

outside this context. Some debate remains whether catastrophizing is best conceptualized as a cognition or coping strategy.^{17,22} Despite this lack of clarity, catastrophizing has emerged as an important construct influencing pain and disability. However, the literature examining the relationship between catastrophizing, beliefs and pain and disability is less developed in WAD. Based on the incomplete knowledge of the capacity of beliefs to predict future WAD-related pain and disability, we examined the extent beliefs and catastrophizing predict future pain and disability. Secondly, we examined the relationships between specific beliefs and catastrophizing.

4.2. Methodology

4.2.1. Design

A prospective cohort survey design was used. We measured WAD-related beliefs in a clinical setting from an acute stage (< six weeks post-MVC) to six months post-MVC. Baseline measurements were recorded within six weeks of the MVC and follow-up measures occurred at three and six months post-MVC. The University of Alberta Health Research Ethics Board approved this study.

4.2.2. Participants

From December 2008 to May 2010, we recruited patients with acute WAD using a convenience sampling strategy. Eighteen physical therapy and chiropractic clinics in Alberta and Saskatchewan, Canada, were approached to distribute study invitations. The 18 clinics were distributed in urban (15 clinics) and rural (3 clinics) areas of Alberta (15 clinics) and in Saskatchewan (3 clinics). Fifteen were physical therapy clinics and three were chiropractic. Study packages that included the survey were distributed by clinic administrative staff to patients attending the clinics with complaints of neck pain of less than six weeks duration as a result of a MVC. Exclusion factors included age <18 years, inability to speak

or read English, not experiencing neck pain from a MVC, or duration of greater than 6 weeks.

Sample size was based on suggestions for multiple linear regression. A commonly used rule of thumb suggests ten subjects per variable entered in the model.²³ Therefore, 50 participants were required to examine 5 baseline variables in predicting self-reported pain and disability.

4.2.3. *Measures*

Pain beliefs:

Three tools were used to measure a wide range of WAD-related pain beliefs. These included the Survey of Pain Attitudes (SOPA-35), Pain Beliefs and Perceptions Inventory (PBPI) and the Pain Catastrophizing Scale (PCS). Each will be described in detail.

SOPA-35:^{15,16,19,24} The SOPA-35 includes seven subscales with items scored on a 5-point Likert agreement scale. The subscales include control, disability, harm, emotion, medication, solicitude, and medical cure. The higher the score the more the belief is endorsed. The subscales control and emotion are considered adaptive beliefs, and the remainder maladaptive. Internal consistency coefficients for the seven subscales have been reported to range from 0.66 to 0.84 with only the harm scale having a marginal coefficient value.¹⁵ Test-retest reliability for the SOPA-35 factors was also acceptable (0.71 to 0.82).¹⁵ In chronic pain settings, there is evidence supportive of construct validity for the SOPA demonstrating expected associations with pain, disability, depression and coping.^{19,24}

PBPI:¹⁴ The PBPI is a 16-item questionnaire designed to evaluate patient beliefs about pain, with items rated on a 4-point Likert agreement scale. It was originally found to be comprised of three factors¹⁴ with subsequent studies demonstrating a four-factor structure.^{21,25-27} The subscales are labeled mystery, self-blame, pain

permanence, and pain constancy. Negative scores on the subscales indicate disagreement with the belief, while positive scores indicate endorsement. The PBPI has been shown to have adequate internal consistency (Cronbach's $\alpha = 0.65-0.80$).¹⁴ The construct validity evidence for the PBPI is primarily in chronic pain settings. Subscales on the PBPI have demonstrated expected relationships with measures of pain quality and intensity, depression, anxiety, physical functioning, and coping strategies.^{15,19,21,25-28} The constancy scale was not considered in this analysis since it measures a description of pain rather than a time-related pain belief as originally described.¹⁴ Qualitative interviews with a sub-group of our participants were conducted and will be discussed elsewhere, but confirmed that the constancy scale was interpreted as a description of pain experienced. (Bostick, unpublished data).

PCS:²⁹ - The PCS is a 13-item instrument that measures the extent of exaggerated negative mental thoughts during actual or anticipated painful experiences, with each item scored on a 5-point Likert scale.^{17,29} It is reported to have three correlated subscales: rumination, magnification, and helplessness. This three-factor structure was not reproduced in an exploratory factor analysis performed on this data set. Thus, only the overall PCS score is used in this study. Higher scores indicate higher levels of catastrophizing. Each factor has shown acceptable levels of internal consistency (Cronbach's $\alpha = 0.66-0.87$).²⁹ The PCS has been found to correlate with outcomes such as pain intensity in a variety of conditions including WAD.¹⁷ In addition, catastrophizing is associated with pain and illness behaviours⁹ and disability after WAD at, one- and three-years post-MVC.^{17,30} The PCS has a considerable amount of validity evidence in both acute and chronic pain populations¹⁷ and is an important prognostic factor in the WAD injury context.^{9,10}

Outcome variables:

Pain intensity and disability were our key outcomes of interest and were measured at each measurement occasion. Pain and self-reported disability at three and six months post-MVC were the dependent variables used in the examination of predictive validity. Pain intensity was measured on an 11-point numerical rating scale. A 30% reduction in pain intensity is considered clinically important.³¹ Disability was measured using the Whiplash Disability Questionnaire (WDQ).³² The WDQ contains 13 items, each scored on an 11-point scale. Scores range from 0 (indicating no disability) to 130 (indicating complete disability). Psychometric properties have demonstrated high internal consistency (Cronbach's $\alpha = 0.96$),³² excellent short-term test-retest reliability, reproducibility, and responsiveness in a physiotherapy setting with a range of acute to chronic WAD patients.³³

Potential moderating variables:

The survey included the following demographic variables: age, sex, previous history of WAD, vehicle insurance (tort or no fault) and whether or not the injury resulted in an overnight hospital stay. In addition, the number of painful body parts was measured as another indicator of injury severity. Baseline pain intensity, sex, history of WAD and age have been suggested as important potential moderating variables in prognostic studies examining psychological factors in WAD.²⁰ Baseline disability was considered a potential moderator variable for disability at three and six months post-MVC.

4.2.4. Analysis

Initially, descriptive statistics were calculated including means and standard deviation (std dev) for continuous variables and percentages for categorical variables.

Next, Cronbach's alpha (α) was calculated for each sub-scale at each measurement occasion in order to examine the scales' reliability (internal

consistency). Then, two analyses were used to examine our study aims described above:

(1) Simple correlational analysis was used to examine relationships between specific beliefs and catastrophizing.

(2) Multiple linear regression was used to examine the capacity of baseline beliefs to predict three and six-month pain intensity and self-reported disability. An exploratory modeling strategy identified baseline beliefs predictive of future pain and disability. Baseline beliefs from each measure (SOPA, PBPI and PCS) were examined for the presence of a significant relationship with pain intensity and disability after adjusting for potential moderating variables. Unique variance explained beyond the moderating variables was assessed for each belief.

We also compared age, gender and baseline pain intensity in the study sample with a large database of physical therapy clinics in Alberta and Saskatchewan provided by two physical therapy companies (n=2759) to ascertain the representativeness of the study sample. PASW for Windows, version 18 (© IBM SPSS Inc., 2009, Chicago, IL) was used to conduct the analysis.

4.3. Results

4.3.1. Sample characteristics

The study sample was comprised of 72 patients. The mean age of the study sample was 39.0 years (std dev = 14.0). Seventy-nine percent of the study sample was female, 27.8% had a previous history of WAD and 76.4% were insured in a tort system. The average pain intensity in the cohort was 5.2 (std dev = 2.0). The average number of reported painful body sites (including the neck) was 2.9 (std dev = 1.7). The mean baseline disability was 59.1 (std dev = 32.0). Additional details of the sample are found in Table 4-1. Regarding representativeness, the study sample was similar to a large physiotherapy database (n=2759) for age and initial pain intensity [mean age (std dev) = 40.4 (14.2); mean baseline pain

intensity (std dev) = 5.9 (2.1) respectively]. However, the study sample had a higher proportion of females, compared to the larger clinic sample (60.1%).

At the three and six-month follow-up, 55 (76%) and 48 (67%) participants returned a survey respectively. Comparison of the available baseline measures between patients with complete data and those who were lost to follow-up demonstrated two systematic differences: males and participants with higher levels of solicitude were more likely to be lost to follow-up compared to females. However, sex was adjusted for in the analyses to account for the differential follow-up. Adjusting for solicitude did not make meaningful changes in the multivariate models and was therefore not reported. No other statistically significant differences were discovered between these groups concerning the outcome variables or any of the beliefs subscales from the SOPA, PBPI and PCS.

4.3.2. Internal consistency

The internal consistency of the SOPA, PBPI and PCS subscales ranged from 0.44 to 0.85, 0.80 to 0.82, and 0.62 to 0.91 at baseline respectively. The medical cure scale and harm scales had the lowest internal consistency values and were scrutinized further. Examination of the individual items of the medical cure scale showed consistent response patterns for all items except one: “when I find the right doctor, he or she will know how to reduce my pain”. Since the cohort included physical therapy and chiropractic clinics as opposed to physicians, some participants may have had difficulty interpreting this item. In addition, the other medical cure items use language related to ‘curing pain’ rather than ‘reducing pain’, which may have led to inconsistent responses. Regarding the harm scale, the low internal consistency coefficient may be explained by the possibility that two separate constructs exist within the harm scale: exercise perceived as harmful and pain as a sign of damage. Removal of the troublesome items did not change findings in the subsequent analysis. Thus, the original structure was retained for the analysis. The pattern of internal consistency observed at the three-month

assessment was similar to those at baseline. All scales achieved a minimum internal consistency coefficient of at least 0.76 at the six-month measurement.

TABLE 4-1
Sample characteristics at baseline

Variable	Mean (standard deviation) or Frequency (percent)
Age	39.0 (14.0); range 18-73
Sex (female)	57 (79.2%)
History of WAD	20 (27.8%)
Insurance (tort AB + SK)	55 (53 AB, 2 SK) (76.4%)
Pain intensity (NRS)	5.2 (2.0)
Number of painful body sites	2.9 (1.7)
Hospitalization	0
WDQ (0 to 130)	59.1 (32.0)
SOPA Control (0 to 20)	11.3 (4.0)
SOPA Disability (0 to 20)	7.5 (3.6)
SOPA Harm (0 to 20)	8.6 (3.6)
SOPA Medication (0 to 20)	11.7 (3.9)
SOPA Emotion (0 to 20)	9.7 (5.4)
SOPA Solitude (0 to 20)	7.1 (4.7)
SOPA Medical Cure (0 to 20)	11.8 (3.4)
PBPI Mystery (8 to -8)	-2.7 (3.7)
PBPI Permanence (10 to -10)	-4.4 (3.9)
PBPI Constancy (8 to -8)	1.2 (4.0)
PBPI Self Blame (6 to -6)	-4.3 (2.0)
PCS Total (13 to 65)	24.7 (9.4)

*WAD = whiplash associated disorder, AB + SK = provinces of Alberta and Saskatchewan, NRS = numerical rating scale, WDQ = whiplash disability questionnaire, SOPA = survey of pain attitudes, PBPI = pain beliefs and perceptions inventory, PCS = pain catastrophizing scale

4.3.3. Associations between beliefs and catastrophizing

The following beliefs were consistently associated with catastrophizing at each measurement occasion: control, disability, harm, medication, mystery and permanence. Emotion and solicitude were associated with catastrophizing at baseline and three months. Table 4-2 summarizes the magnitude of these Pearson correlation coefficients.

TABLE 4-2

Correlations between specific beliefs and catastrophizing*.

	Catastrophizing (PCS)		
	Baseline n=72	3-months n=55	6-months n=46
SOPA-Control	-0.56	-0.59	-0.81
SOPA-Disability	0.43	0.66	0.78
SOPA-Harm	0.44	0.33	0.70
SOPA-Emotion	0.40	0.40	0.18
SOPA-Medication	0.31	0.36	0.64
SOPA-Solicitude	0.35	0.45	0.27
SOPA-Medical cure	0.03	-0.11	-0.28
PBPI-Mystery	0.45	0.46	0.61
PBPI-Permanence	0.36	0.53	0.73
PBPI-Self-blame	-0.01	0.06	0.05

Values in bold indicate statistical significance ($p < 0.05$)

PCS=Pain Catastrophizing Scale, SOPA=Survey of Pain Attitudes, PBPI=Pain Beliefs and Perceptions Inventory.

4.3.2. Baseline prediction of pain intensity at three and six months post-MVC

Survey of Pain Attitudes: After adjusting for the potential moderator variables baseline pain intensity, age, sex and history of WAD, there were no baseline SOPA beliefs predictive of pain intensity at three months post-MVC. However,

baseline control and medical cure were significant predictors of pain intensity at six months (Table 4-3). The moderating variables explained 19% of the variance in pain intensity at 6 months with control (n=46) and medical cure (n=45) beliefs explaining an additional 5% and 14% of variance respectively. More endorsement of beliefs about control and a medical cure for pain were associated with lower levels of pain intensity six months post-MVC.

Pain Beliefs and Perception Inventory: After adjusting for the potential moderator variables, the baseline belief in the permanence of pain was predictive of pain intensity at three (n=53) and six months (n=43) post-MVC (Table 4-3). The moderating variables explained 20% and 19% of the variance in pain intensity at 3 and 6 months with the belief in pain permanence explaining an additional 6% at 3 months and 16% at 6 months respectively. More endorsement of the belief in pain permanence was associated with higher levels of pain intensity.

Pain Catastrophizing Scale: After adjusting for the potential moderator variables, the baseline PCS was predictive of pain intensity at three (n=53) and six months (n=45) post-MVC (Table 4-3). The moderating variables explained 20% and 19% of the variance in 3 and 6-month pain intensity with catastrophizing contributing additional 9% and 8% unique variance respectively. Higher levels of catastrophizing were associated with higher levels of pain intensity.

4.3.3. Baseline prediction of disability at three and six months post-MVC

Survey of Pain Attitudes: After adjusting for the potential moderator variables baseline self-reported disability, age, sex and history of WAD, there were no baseline SOPA beliefs predictive of self-reported disability three or six months post-MVC.(Table 4-4)

TABLE 4-3

Univariate and multivariate associations between baseline beliefs and pain intensity 3 and 6 months post-MVC

	3 months		6 months	
	Univariate (95% CI)	Adjusted β -coefficient (95% CI)*	Univariate (95% CI)	Adjusted β -coefficient (95% CI)*
SOPA Control	-0.46 (-0.65, -0.22)	-0.12 (-0.26, 0.02)	-0.42 (-0.63, -0.15)	-0.19 (-0.37, -0.01)
SOPA Disability	0.39 (0.14, 0.59)	0.06 (-0.11, 0.23)	0.25 (-0.04, 0.50)	0.02 (-0.22, 0.26)
SOPA Harm	0.24 (-0.03, 0.48)	0.01 (-0.14, 0.17)	0.18 (-0.12, 0.45)	0.08 (-0.13, 0.29)
SOPA Medication	0.20 (-0.07, 0.44)	-0.07 (-0.21, 0.07)	0.28 (-0.01, 0.53)	0.05 (-0.15, 0.26)
SOPA Emotion	0.17 (-0.10, 0.42)	-0.01 (-0.09, 0.09)	-0.04 (-0.33, 0.25)	0.01 (-0.12, 0.14)
SOPA Solicitude	0.17 (-0.10, 0.42)	0.02 (-0.11, 0.14)	-0.09 (-0.37, 0.21)	-0.07 (-0.24, 0.10)
SOPA Medical cure	-0.22 (-0.46, 0.05)	-0.13 (-0.27, 0.02)	-0.40 (-0.62, -0.12)	-0.28 (-0.47, -0.10)
PBPI Mystery	0.22 (-0.05, 0.46)	0.04 (-0.11, 0.19)	0.26 (-0.04, 0.52)	0.13 (-0.05, 0.31)
PBPI Permanence	0.48 (0.24, 0.66)	0.17 (0.03, 0.30)	0.53 (0.27, 0.72)	0.25 (0.09, 0.41)
PBPI Self-blame	0.13 (-0.14, 0.38)	0.10 (-0.14, 0.35)	0.11 (-0.19, 0.39)	0.13 (-0.26, 0.53)
PCS	0.58 (0.37, 0.74)	0.09 (0.03, 0.16)	0.39 (0.11, 0.61)	0.10 (0.0, 0.18)

*adjusted for baseline pain intensity, age, gender and history of WAD

Values in bold indicates statistical significance ($p < 0.05$)

CI=confidence interval

TABLE 4-4

Univariate and multivariate associations between beliefs and self-reported disability 3 and 6 months post-MVC

	3 months		6 months	
	Univariate (95% CI)	Adjusted β -coefficient (95% CI)*	Univariate (95% CI)	Adjusted β -coefficient (95% CI)*
SOPA Control	-0.46 (-0.65, -0.22)	-1.28 (-2.83, 0.26)	-0.49 (-0.69, -0.22)	-1.49 (-3.32, 0.34)
SOPA Disability	0.33 (0.07, 0.55)	-0.85 (-2.72, 1.03)	0.34 (0.04, 0.58)	-0.31 (-2.62, 2.00)
SOPA Harm	0.34 (0.08, 0.56)	1.09 (-0.51, 2.68)	0.23 (-0.08, 0.50)	0.89 (-0.94, 2.73)
SOPA Medication	0.28 (0.01, 0.51)	0.13 (-1.41, 1.67)	0.49 (0.22, 0.69)	1.29 (-0.70, 3.28)
SOPA Emotion	0.29 (0.02, 0.52)	-0.33 (-1.45, 0.79)	0.07 (-0.24, 0.36)	0.45 (-1.80, 0.91)
SOPA Solicitude	0.18 (-0.09, 0.43)	0.11 (-1.28, 1.51)	0.04 (-0.26, 0.34)	-0.19 (-1.91, 1.53)
SOPA Medical cure	-0.16 (-0.41, 0.12)	-0.69 (-2.34, 0.97)	-0.31 (-0.56, -0.01)	-1.81 (-3.72, 0.11)
PBPI Mystery	0.34 (0.07, 0.56)	1.69 (0.15, 3.23)	0.45 (0.17, 0.66)	2.29 (0.82, 3.77)
PBPI Permanence	0.45 (0.20, 0.65)	2.15 (0.76, 3.55)	0.36 (0.06, 0.60)	1.33 (-0.17, 2.83)
PBPI Self-blame	0.06 (-0.22, 0.33)	1.51 (-1.23, 4.25)	0.13 (-0.18, 0.42)	3.37 (-0.07, 6.81)
PCS	0.63 (0.43, 0.77)	1.08 (0.34, 1.82)	0.45 (0.17, 0.66)	0.79 (-0.05, 1.63)

*adjusted for baseline disability, age, gender and history of WAD

Values in bold indicates statistical significance (p < 0.05)

CI=confidence interval

Pain Beliefs and Perception Inventory: After adjusting for the potential moderator variables, the baseline belief that pain is mysterious was predictive of self-reported disability at three (n=50) and six months (n=40) post-MVC. In addition, the baseline belief in pain permanence was associated with three month disability (n=49).(Table 4-4) The moderator variables explained 63% and 65% in the variance in 3 and 6 month disability respectively. Additional unique variance attributed to mystery was 3% at 3 months and 7% at 6 months. Pain permanence explained an additional 6% at 3 months. Higher levels of mystery and permanence were associated with higher levels of self-reported disability at three and six months post-MVC.

Pain Catastrophizing Scale: After adjusting for the potential moderator variables, the baseline PCS was predictive of self-reported disability at 3 months post-MVC (n=50) (Table 4-4). The moderator variables accounted for 63% in 3-month disability with catastrophizing explaining an additional 6% of variance. Higher levels of catastrophizing were associated with higher levels of pain intensity.

Regression diagnostics revealed no evidence of multicollinearity or significant violation of homoscedasticity, normality or linearity assumptions

4.4. Discussion

Beliefs about pain measured in the early stages following MVC appear to provide important information for predicting future recovery in patients with WAD. Cognitive-behavioral approaches suggest that cognitions should predict behaviour. For example, the fear-avoidance model predicts negative thoughts such as catastrophizing lead to fear-avoidance beliefs and disability.⁶ These assertions have been recently examined to determine if these relationships are indeed causal.^{34,35} While these studies suggest the assumptions of these

relationships require refinement, there is ample evidence implicating the importance of cognitive factors as determinants of behavior.^{1,3,7,20} In this study, WAD-related beliefs were important variables in predicting outcome. In addition to catastrophizing, beliefs pertaining to permanence were consistently predictive of pain intensity. Beliefs related to the mysterious nature of pain were consistently predictive of self-reported disability. The importance of catastrophizing in predicting both pain and disability has been well supported elsewhere³⁶ and these data build on existing evidence specific to WAD.^{9,18}

Similar to previous research, baseline pain intensity and disability account for the majority of variance in future pain and disability.³ However, beliefs about future outcome (permanence and medical cure) were one of the strongest predictors of pain intensity and disability in this study. Previous research has demonstrated constructs similar to permanence and medical cure, such as expectations, are related to pain intensity. For example, in a large longitudinal cohort study, Carroll and colleagues³⁷ found that those who expected to recover quickly experienced a cessation of pain 80% faster compared to those who expected that they would never recover, after adjusting for confounding. Recovery or outcome expectancies have also been shown to be related to actual outcomes in a variety of health conditions such as low back pain and myocardial infarction.³⁸

Little research is available to situate raw beliefs scores observed in this study with other similar study populations. In a study by Adams et al.,¹⁸ pre-treatment PCS scores for work-disabled patients with WAD II entering a cognitive-behavioral treatment program at 4-12 weeks, 3-6 months and 6-18 months post-MVC were reported as 19.0 (9.4), 20.1 (8.4) and 30.4 (11.5) respectively. The PCS scores for the sub-acute and early-chronic participants were lower than we observed in this study, while the scores for in the chronic stage were higher compared to our 6-month data. Since previous literature suggests females tend to report higher levels of catastrophizing compared to men,¹⁷ the higher proportion of females in this

study may account for the differences in catastrophizing observed compared to the Adams et al study. No known literature exists to compare pain beliefs as measured by the SOPA and PBPI in a similar cohort of patients.

Caution must be exercised in the interpretation of these findings. The baseline sample is relatively small (n=72). In comparison with a database (patients in Alberta and Saskatchewan during the study recruitment period) from a large physical therapy company with clinics throughout the Canada (n=2759), the sample is generalizable concerning age and baseline pain intensity. However, this study over-represents females. Literature suggests that women report higher levels of pain intensity, display more pain behaviours, seek more healthcare and score higher on measures of catastrophizing.^{39,40} Thus, findings from this study may be more generalizable to female patients with WAD. Follow-up data at the three and six month measurements were subject to significant attrition. Twenty-four percent of the sample was lost to follow-up at the three-month measurement occasion and an additional twelve percent were lost to follow-up at six months. Attrition occurred despite two reminder letters and one phone call. While the consequences of this attrition bias cannot be completely ascertained, sensitivity analysis of baseline characteristics (demographics, pain, disability and beliefs) between those with complete and incomplete data demonstrated systematic differences only in the variable sex (males were more likely to be lost to follow-up). Since sex was not related to the outcome, this difference alone does not adversely impact the internal validity of the study. In addition, the effects of attrition limited statistical power. In particular, the small sample size limited more detailed regression analyses. Finally, while efforts were made to control potential confounding, there are likely unmeasured moderating and/or mediating variables that could influence the findings. For example, although this exploratory study was focused on beliefs, variables such as emotional distress may influence the relationships between beliefs, catastrophizing and outcome^{21,41-43} and warrant consideration in explanatory studies.

This study contributes important information to the growing body of knowledge regarding cognitive factors in determining WAD outcome. In addition to the more established catastrophizing literature, pain beliefs measured by the SOPA and PBPI represent potentially important predictors of outcome after WAD. In particular, future beliefs about outcome and cure appear to have promise as predictors of poor prognosis after WAD. Furthermore, these beliefs have the possibility to contribute to future research aimed at clarifying theoretical perspectives on pain-related catastrophizing. Both the conceptualization of pain-related catastrophizing as an appraisal and communal coping strategy was supported in this study. Future studies examining beliefs and catastrophizing pertaining to WAD with larger and more representative cohorts will further clarify the role of cognitions in shaping the experience of WAD.

4.5. References:

1. Carroll LJ, Holm LW, Hogg-Johnson S, Cote P, Cassidy DJ, Haldeman S, Nordin M, Hurwitz EL, Carragee EJ, van der Velde G, Peloso PM, Guzman J. Course and prognostic factors for neck pain in whiplash-associated-disorders (WAD): Results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and its Associated Disorders. *Spine* 2008;33:S83S92.
2. Chappuis,G.; Soltermann,B.; Cea; Aredoc; Ceredoc. Number and cost of claims linked to minor cervical trauma in Europe: results from the comparative study by CEA, AREDOC and CEREDOC. *Eur Spine J* 2008;17:1350-1357.
3. Walton DM, Pretty J, MacDermd JC, Teasell RW. Risk factors for persistent problems following whiplash injury: results of a systematic review and meta-analysis. *J Orthop Sports Phys Ther* 2009;39:334-350.
4. Eccleston C, Williams AC, Morley S. Psychological therapies for the management of chronic pain (excluding headache) in adults. *Cochrane Database Syst Rev* 2009;15:CD007407.
5. Turk DC, Meichenbaum D, Genest M. *Pain and behavioural medicine: A cognitive-behavioural perspective*. New York: Guilford Press, 1993.
6. Vlaeyen JWS, Linton SJ. Fear-avoidance and its consequences in chronic musculoskeletal pain: a state of the art. *Pain*, 2000;85:317-332.
7. Main CJ, Foster N, Buchbinder R. How important are back pain beliefs and expectations for satisfactory recovery from back pain? *Best Prac Res Clin Rheumatol* 2010; 24:205-217.
8. Kyllback M, Theirfelder T, Soderlund A. Prognostic factors in whiplash-associated disorders. *Int J Rehabil Res* 2002;25:181-187.

9. Sullivan MJL, Stanish W, Sullivan ME, Tripp D. Differential predictors of pain and disability in patients with whiplash injuries. *Pain Res Manag* 2002;7:68–74.
10. Nederhand MJ, Ijzerman MJ, Hermens HJ, Turk DC, Zilvold G. Predictive value of fear avoidance in developing chronic neck pain disability: consequences for clinical decision making. *Arch Phys Med Rehabil* 2004;85:496501.
11. Buitenhuis J, de Jong PJ, Jaspers JP, Groothoff JW. Catastrophizing and causal beliefs in whiplash. *Spine* 2008;33:2427-2433.
12. Thompson DP, Oldham JA, Urmston M, Woby SR. Cognitive determinants of pain and disability in patients with chronic whiplash-associated disorder: A cross-sectional observational study. *Physiother* 2010;96:151-159.
13. Turk DC and Okifuji. A cognitive-behavioural approach to pain management. In: Wall PD and Melzack R, editors. *Textbook of Pain*, 4th ed. London: Churchill Livingstone, 1999. pp 1431-44.
14. Williams DA, Thorn BE. An empirical assessment of pain beliefs. *Pain* 1989;36:351-358.
15. Jensen MP, Turner JA, Romano JM. Pain belief assessment: A comparison of the short and long versions of the Survey of Pain Attitudes. *J Pain* 2000;1:138-150.
16. Jensen MP, Turner JA, Romano JM. Changes in beliefs, catastrophizing, and coping are associated with improvement in multidisciplinary pain treatment. *J Consult Clin Psychol* 2001;69:655-662.
17. Sullivan MJL, Thorn B, Haythornthwaite JA, Keefe F, Martin M, Bradley LA, Lefebvre. Theoretical perspectives on the relation between catastrophizing and pain. *Clin J Pain* 2001;17:52-64.
18. Adams H, Ellis T, Stanish WD, Sullivan MJL. Psychosocial factors related to return to work following rehabilitation of whiplash injuries. *J Occup Rehabil* 2007;17:305-315.

19. Jensen MP, Turner JA, Romano JM. Changes after multidisciplinary pain treatment in patient pain beliefs and coping are associated with concurrent changes in patient functioning. *Pain* 2007;131:38-47.
20. Williamson E, Williams M, Gates S, Lamb SE. A systematic literature review of psychological factors and the development of late whiplash syndrome. *Pain* 2008;135(1-2):20-30.
21. Williams DA, Robinson ME, Geisser ME. Pain beliefs: assessment and utility. *Pain* 1994;59:71-78.
22. Thorn BE, Keefe FJ, Anderson T. The communal coping model and interpersonal context: problems or process? *Pain* 2004;20:275-82.
23. Norman GR, Streiner DL. *Biostatistics: the bare essentials*. 2nd ed. Hamilton: BC Decker, 2000.
24. Jensen MP, Karoly P, Huger R. The development and preliminary validation of an instrument to assess patients' attitudes toward pain. *J Psychosom Res* 1987;31:393-400.
25. Herda CA, Siergeris K, Basler H. The Pain beliefs and perceptions inventory: further evidence for a 4-factor structure. *Pain* 1994;57:85-90.
26. Morley S, Wilkinson L. The Pain Beliefs and Perceptions Inventory: a British replication. *Pain* 1995;61:427-433.
27. Strong J, Ashton R, Chant D. The measurement of attitudes towards and beliefs about pain. *Pain* 1992;48:227-236.
28. Dysvik E, Lindstrom TC, Eikeland OJ, Natvig GK. Health-related quality of life and pain beliefs among people suffering from chronic pain. *Pain Manag Nurs* 2004;5:66-74.
29. Sullivan MJL, Bishop SR, Pivik J. The Pain Catastrophizing Scale: development and validation. *Psychol Assess* 1995;7:524-32.
30. Sullivan MJL, Stanish W, Waite H, Sullivan ME, Tripp D. Catastrophizing, pain, and disability following soft tissue injuries. *Pain* 1998;77:253-260.

31. Farrar JT, Young Jr JP, LaMoreaux L, Werth JL, Poole RM. Clinical importance of changes in chronic pain intensity measured on an 11-point numerical pain rating scale. *Pain* 2001;94:149–58.
32. Pinfold M, Niere KR, O'Leary EF, Hoving JL, Green S, Buchbinder R. Validity and internal consistency of a whiplash-specific disability measure. *Spine* 2004;29:263268.
33. Willis C, Niere KR, Hoving JL, Green S, O'Leary EF, Buchbinder R. Reproducibility and responsiveness of the Whiplash Disability Questionnaire. *Pain* 2004;110:681-688.
34. Vangronsveld KLH, Peters M, Goosens M, Vlaeyen JW. The influence of fear of movement and pain catastrophizing on daily pain and disability in individuals with acute whiplash injury: A daily diary study. *Pain* 2009;139:449-457.
35. Wideman TH, Adams H, Sullivan MJL. A prospective sequential analysis of the fear-avoidance model of pain. *Pain* 2010;145:45-51.
36. Haythornthwaite JA. It's a belief. It's an appraisal. It's Copin ... No, it's catastrophizing. In: Castro-Lopes J, editor. *Current topics in pain*. Proc 12th World Congress on Pain, Seattle: IASP Press, 2009. pp. 271-287.
37. Carroll LJ; Holm LW, Ferrari R, Ozegetic D, Cassidy DJ. Recovery after whiplash associated disorders: do you get what you expect? *J Rheumatol* 2009;36:10631070.
38. Mondloch MV, Cole D, Frank J. Does how you do depend on how you think you'll do? A systematic review of the evidence for a relation between patients' recovery expectations and health outcomes. *CMAJ* 2001;165:174–9.
39. Sullivan MJL, Tripp DA, Santor D. Gender differences in pain and pain behavior: the role of catastrophizing. *Cog Ther Res* 2000;24:121–34.
40. Unruh AM. Gender variations in clinical pain experience. *Pain* 1996;65:123–67.

41. Sterling M, Jull G, Vicenzino B, Kenardy J, Darnell R. Physical and psychological factors predict outcome following whiplash injury. *Pain* 2005;114:141–148.
42. Sterling M, Pettiford C, Hodkinson E, Souvlis T, Curatolo M. Psychological factors are related to some sensory pain thresholds but not nociceptive flexion reflex threshold in chronic whiplash. *Clin J Pain* 2008;24:124–30.
43. Sullivan MJ, Thorn B, Rogers W, Ward LC. Path model of psychological antecedents to pain experience: experimental and clinical findings. *Clin J Pain* 2004;20:164-73.

CHAPTER 5

Lessons learned: maximizing response and minimizing attrition

5.1. Introduction

We recruited patients from physical therapy and chiropractic clinics to ascertain their beliefs about pain, how they influence outcome, the stability of these beliefs, as well as factors that inform endorsement of beliefs. Study participants were sampled in a clinical environment. This presented significant challenges related to participant recruitment and retention. These issues were primary limitations for studies two and three.

Much has been written about consequences of poor response and attrition as well as strategies to mitigate them.¹ Despite this, there are no clear guidelines about what constitutes adequate response and follow-up.² Criteria are provided, but meeting them does not necessarily mean generalizability is achieved or bias avoided.³⁻⁵ Thus, it is paramount to understand the potential for response and attrition bias within a study. In this chapter, a detailed reflection of the potential for bias within studies two and three is discussed. Potential consequences of these biases and strategies used to mitigate bias are reviewed. Finally, I hypothesize potential reasons for poor response and make suggestions for planning similar studies.

5.2. Potential consequences related to response and attrition bias

Response and attrition bias fall under the umbrella of selection bias. Selection bias refers to systematic error resulting from the manner subjects are selected or retained in a study.⁶ It occurs when characteristics differ between people that do and do not participate, and/or those with complete and incomplete follow-up data.⁷ Selection bias affecting the internal validity of a study results when systematically different variables are related to an outcome. In this case, selection

bias can either dilute or exaggerate a true relationship. For example, volunteers for research tend to be better educated, more active in community affairs, less likely to smoke and more concerned about health matters.⁶ Whether variables such as these are associated with the study outcome determines whether the internal validity of the study has been compromised. Selection bias affecting the external validity of the study occurs when the sample under study differs from the larger sampling frame such that study findings are not generalizable outside of the study. When responders systematically differ from non-responders, results of the study should be generalized only to people with characteristics similar to the study sample. A secondary consequence to non-response is insufficient power to carry out statistical procedures. This is a prominent concern in cohort studies involving complex disorders such as whiplash where many variables are required to disentangle complex relationships. As an example, multiple linear regression requires approximately 10 to 20 participants per independent variable.^{8,9}

Unless the entire population of interest can be sampled, selection bias is likely to occur to some extent. The sampling method is an important determinant of selection bias. For example, a convenience sample that recruits participants from an emergency room may be systematically different from participants recruited in other settings. Côté et al¹⁰ demonstrated this to be an important consideration in a systematic review of the prognosis of acute whiplash injury. The reviewers separated studies based on recruitment setting and suggested that sample characteristics could depend on the recruitment context. The possibility that recruitment setting influenced prognosis was evaluated in a subsequent review¹¹ and found only modest differences; however power may have been insufficient to make firm conclusions.¹¹ Thus, convenience sampling can lead to selection bias that impacts both the internal and external validity of a study.

Bias due to attrition occurs when follow-up samples systematically differ from the original sample.⁶ Attrition is inevitable in human prospective studies as people

move away, choose to withdraw, become deceased, etc. When research participants retained in the study are systematically different from those who are lost to follow-up and when these variables are related to the outcome, internal validity is compromised. In addition, as research participants are lost, the generalizability of the sample may also be adversely impacted. Attrition also has consequences to study power. An additional related issue is missing data. Non-random missing data due to loss to follow-up can become a possible predictor of outcome thus threatening internal validity.

5.3. Mitigating response and attrition bias

5.3.1. Methodological strategies mitigating response bias

Two concerns of selection bias pertained to this study: (1) systematic differences between responders and non-responders (at baseline or over follow-up measurements) possibly leading to a threat to internal validity and (2) misrepresentation of the target population by the sample population threatening external validity. A sample that is not representative of the target population is most likely to arise from sample selection procedures and non-response. The ideal method of minimizing selection bias is to prevent it prior to data collection. Selection bias is highly unlikely to occur when probability sampling is used. Given an equal opportunity for selection, there is also an equal opportunity for having all characteristics of the target population represented.¹² If probability sampling is not feasible then efforts should be made to ascertain, to the extent possible, the differences between the sample and population (see analytical strategies below).

Convenience sampling strategies are prone to response bias if patients decline to participate in the study. Response bias is best controlled at the planning stage of a study by implementing strategies to maximize response rate. McColl et al¹³

provide a thorough review of strategies to enhance response rate. The strategies identified as having the most evidence to support their use include multiple contacts, saliency, enclosed monetary incentives, and a highly personalized letter with assurances of anonymity. A popular approach is Dillman's tailored design method¹⁴ that consists of: (1) a respondent-friendly questionnaire; (2) use of four contacts by first-class mail, with an additional 'special' contact (e.g. certified mail, telephone call); (3) use of return envelopes with real first-class stamps; (4) personalized correspondence and (5) a token financial incentive sent with the survey. In addition, a recent systematic review examined 481 studies using 110 methods to improve response rate.¹⁵ Table 5-1 summarizes these methods and others found to be effective in improving response.

Strategies used to mitigate response bias in this thesis are also summarized in Table 5-1. In addition, to illustrate these strategies and to provide context for the recruitment and retention challenges faced in this thesis, details of the participant recruitment strategy are provided visually. (Figure 5-1) The recruitment strategy included: (1) construction of a respondent friendly and salient questionnaire; (2) recruitment from a wide range of physical therapy clinics in Alberta that were committed to assist with data collection; and (3) distribution of surveys in clinics with minimum burden on administrative staff. Details for each of these strategies are discussed below.

Respondent Friendly Questionnaire

McColl et al¹³ advocate a number of steps to make the survey respondent friendly. For example, limiting the length of the questionnaire, providing clear instructions, using the 'circle the item' response format, and making sure the font is at least 10-point. This study's questionnaire was piloted on lay people without neck pain and found to be readable and was completed within 12 minutes. Saliency of a survey is difficult to determine, however, it was believed to be salient insofar as the research project aimed to increase understanding of a condition that potential

participants were currently seeking care for. Moreover, the information letter inviting participation stated that involvement might lead to improved education strategies to better manage the condition.

TABLE 5-1

Methodological strategies advocated in literature and those employed to mitigate selection bias.

Strategies to mitigate selection bias from literature¹³⁻¹⁶	Studies 2 and 3 of this thesis
Response bias	
• Probability sampling	
• Salient	√
• Respondent friendly	√
• Multiple contacts (including a special contact)	
• Personalized correspondence	
• Monetary incentives	
• Recorded delivery	
• ‘Teaser’ on the envelope	
• Pre-notification	
• Obligation to respond	
Attrition bias	
• Multiple contacts (including a special contact)	√
• Monetary incentives	
• Obligation to respond	
• Postage-paid envelopes	√
• Community involvement (engagement)	
• Study identity (consistent use of logo and colours with contacts)	√
• Study personnel (respectful, accommodating)	?
• Detailed study description	√
• Systematic contacts and scheduling	√
• Provide benefits to participation (e.g. free educational booklet)	
• Nonmonetary incentives (e.g. small tokens of appreciation)	√
• Special tracking for hard to reach participants	

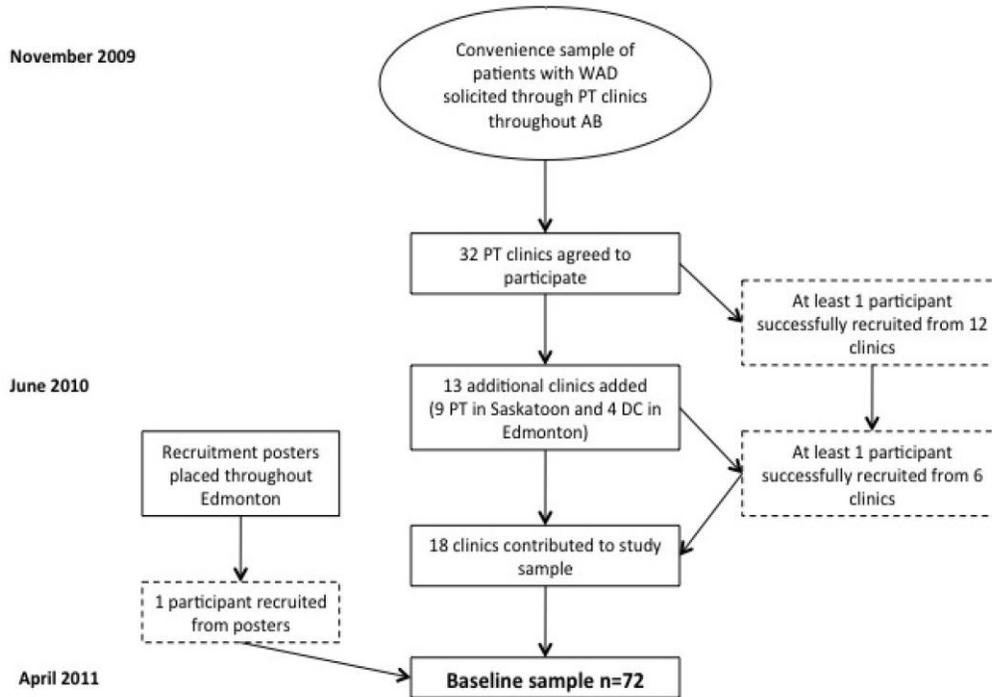
Recruitment From a Wide Range of Physical Therapy Clinics

Recruitment was intended to be wide in scope and included multiple strategies. The aim was to enrol as many patients as possible from varied contexts to maximize response and idealize generalizability. Initially, a large medical general practice organization with medical clinics in Edmonton and Calgary was approached to participate in data collection. This company was targeted as it had research support infrastructure and a large number of clinics. Unfortunately, this medical group declined to participate. Hospital emergency rooms were also approached. However, these departments did not permit data collection within the emergency room without at least one of their physicians' being part of the study team. We were permitted to place a recruitment poster in the emergency waiting room. We also aimed to send study packages through an insurance company but were not successful in securing participation. Therefore, the primary data collection source was private physical therapy clinics (Figure 5-1). Two large Canadian physical therapy companies were initially approached. These companies own 49 clinics throughout the province of Alberta. We solicited support from regional managers and the study was pitched to clinic managers. Twenty-two of these clinics agreed to participate. In addition, private physical therapy clinics in Alberta listed on the University of Alberta Department of Physical Therapy student clinical placement database were contacted via an intermediary. Finally, physical therapy clinics listed in the Calgary and Edmonton yellow pages were directly contacted. Thus, we contacted the majority of private practices in Edmonton and Calgary. An additional 10 clinics agreed to participate. A relatively smaller proportion of rural practices were invited. As a token of our appreciation for participation, we offered to share study findings with clinic participants.

After 6 months of data collection, only 12 of the 32 clinics had successfully enrolled patients into the study. We contacted regional and clinic managers to

address barriers and requested reminders sent to the staff about the study. To help increase response, additional physical therapy clinics in Saskatoon, Saskatchewan

FIGURE 5-1
Participant recruitment flow



were approached to participate (nine additional clinics agreed to distribute surveys). Of these, three clinics successfully enrolled participants. We also invited chiropractic clinics to participate to further increase the sampling frame. To do this, the first three characters of the postal codes of physical therapy clinics participating were matched against a list of registered chiropractic clinics in Edmonton and Calgary. We consulted the yellow pages to identify matching chiropractic clinics in Saskatoon. Personalized letters were sent to each clinic that matched the first three characters of the postal code of participating physical

therapy clinics. Follow-up phone calls were made within 2 weeks to clinic owners who did not respond to the initial contact. Four chiropractic clinics agreed to participate, but just four participants were enrolled. In addition, we contacted every listed physical therapy clinic in the Edmonton yellow pages and requested they place a recruitment poster within their clinic. This strategy was not effective as just one participant entered the study after seeing a recruitment poster. Finally, we provided a thank you card and a \$10 gift certificate to all study contacts within the clinics along with written encouragement to continue to distribute surveys. This expense was covered outside of the allocated operating budget.

Minimal Burden on Clinic Administrative Staff

In total, 45 clinics agreed to serve as data collection sites. Of these, 18 enrolled at least one research participant. In order to maximize clinic involvement, managers requested minimum staff burden. Thus, clinic staff were simply asked to offer a study package to every patient attending the clinic for assessment of pain because of a motor vehicle collision (MVC). The potential participant answered four questions to determine their inclusion into the study. If the patient did not meet the requirement, they were asked to return the survey to the researchers in a provided postage-paid envelope.

5.3.2. Methodological strategies mitigating attrition bias

The majority of evidence about minimizing attrition is in the context of ‘lessons learned’. A systematic review summarized this evidence by identifying themes capturing strategies to idealize follow-up.¹⁶ These themes and other strategies were outlined in Table 5-1. The authors of this review claimed that attrition is generally lower when more strategies are used to mitigate attrition. Though much of these data are experiential, a recent study² examined a number of strategies including different forms of incentives to decrease attrition. In this study, various forms of incentives were compared (gift cards, £2.00, £5.00). They found that incentives decreased attrition, but the type of incentive did not seem to matter.²

The strategies outlined in maximizing response¹³⁻¹⁵ could also apply to minimize attrition.

The retention strategies used in this study were found in Table 5-1. For example, we used multiple follow-up contacts including one special contact, and provided postage-paid envelopes. In addition, the principles of a respondent friendly questionnaires and saliency were applied. We implemented a systematic approach to secure follow-ups. Once a baseline survey was submitted, the date of the MVC was recorded as well as dates for the three- and six-month follow-up mailings. One week prior to the follow-up date, the survey was mailed, included a short letter thanking the participant for their participation and a request to complete the follow-up questionnaire. The package also included an information letter and a postage-paid envelope. If the survey was not received within two weeks, two reminder letters were mailed one week apart. Finally, if we continued to receive no response after three weeks, the participant was given a reminder phone call.

We were unable to implement many of the strategies to increase response from Table 5-1. The most common reason for this was insufficient resources. Monetary incentives appear to be the most effective strategy to maximize response and minimize attrition. This strategy was not used because there were insufficient funds within the operating grant (\$10,000.00). Approximately 650 surveys were sent to clinics. Thus, even a small incentive represented a significant cost. Probability sampling was also not used. Given the wide range of possible entry points into the healthcare system after a MVC-related injury, it was not feasible to sample in a manner that ensured each participant had an equal probability of being in the study. We could not use multiple contacts, personal correspondence, and pre-notification as we relied on an intermediary (clinic staff) to make initial contact.

We were able to use many of the advocated strategies to minimize attrition. The primary exception was the use of incentives to secure returned surveys. As mentioned, budgetary constraints did not permit offering incentives. Since the study was anonymous, we did not know the extent of loss to follow-up due to participants moving addresses (packages were addressed to “Neck Pain Study Participant”).

5.3.3. Analytical strategies mitigating response bias

The primary concern in attempting to understand the consequences of response bias is to ascertain potential differences between responders and non-responders and whether these differences are associated with the outcome(s). In addition, we are interested in the degree to which the study sample represents the sampling frame. It is often difficult to obtain data to examine differences between responders and non-responders. However, two large physical therapy companies with 57 physical therapy clinics in Alberta and Saskatchewan were able to provide some demographic data for all patients who attended their clinics in the three major centres involved in this study during the study period. Thus, this served as a proxy measure for determining response bias. We were able to compare the study sample and the larger patient sample on age, sex and initial pain intensity. (Table 5-2) We found the study sample was comparable to the larger sample with regard to age and initial pain intensity, but differed on sex. Initial pain intensity is a consistent predictor of future pain and disability and was related to the study outcomes in this study. While age is an inconsistent predictor of future outcome in the wider whiplash literature, it was found to be associated with future pain and disability in the multivariate models in this study. These potential confounding factors were similar in the study population and the larger comparison database.

TABLE 5-2

Comparison of the study sample (n=72) with a large comparable patient sample (n=2759)

	Study sample	Proxy sampling frame
Age: mean (stand dev)	39.1 (13.9)	40.4 (14.2)
Sex: (% female)	79.2	60.1
Baseline pain intensity: mean (std dev)	5.2 (2.0)	5.9 (2.1)

While gender appeared to differ systematically between responders and non-responders, it was not related to the outcomes pain intensity and disability at any measurement occasion in either univariate or multivariate analysis. In addition, sex was adjusted for when examining relationships between beliefs and the outcome variables. Thus, it is unlikely that the observed difference in proportions of females in responders and non-responders adversely impacted the internal validity of the study.

The primary consequence of the observed difference in the larger proportion of females participating in this study compared to the proportion participating in physical therapy in general is impaired generalizability. Thus, findings from this study are principally applicable to women accessing physical therapy for WAD-related pain. Considering women tend to access physical therapy more than men, these data remain clinically relevant.

5.3.4. Analytical strategies mitigating attrition bias

In order to determine the potential bias related to attrition, we compared baseline characteristics and beliefs between participants with complete and incomplete follow-up data. Table 5-3 provides a detailed comparison. Participants with complete data were more likely to be female compared to male. In comparing

TABLE 5-3

Comparison of demographic and beliefs variables between participants with incomplete and complete data

	Baseline – 3-months attrition		Baseline – 6-months attrition		Baseline – 3-months – 6-months attrition	
	missing n=17	not missing n=55	missing n=26	not missing n=46	missing n=32	not missing n=40
Pain intensity	5.5 (2.0)	5.1 (2.0)	5.3 (2.3)	5.2 (1.8)	5.3 (2.2)	5.2 (1.8)
Disability	64.7 (31.4)	57.4 (32.2)	56.0 (32.5)	61.0 (31.9)	57.3 (34.4)	60.6 (30.2)
No. body parts	2.4 (1.4)	3.0 (1.8)	3.0 (1.9)	2.8 (1.6)	2.8 (1.8)	3.0 (1.7)
Age	34.6 (9.7)	40.4 (14.9)	37.9 (12.5)	39.7 (14.9)	36.7 (11.8)	40.9 (15.4)
Gender*						
Female	8 (14.0%)	49 (86.0%)	19 (33.3%)	38 (66.7%)	22 (38.6%)	35 (61.4%)
Male	9 (60.0%)	6 (40.0%)	7 (46.7%)	8 (53.3%)	5 (33.3%)	10 (66.7%)
History of WAD						
History	5 (25.0%)	15 (75.0%)	7 (35.0%)	13 (65.0%)	8 (40.0%)	12 (60.0%)
No history	12 (23.2%)	40 (76.9%)	19 (36.5%)	33 (63.5%)	24 (46.1%)	28 (53.9%)
Insurance						
Tort (AB/SK)	14 (25.5%)	41 (74.5%)	19 (34.5%)	36 (65.5%)	23 (44.2%)	32 (55.8%)
No fault (SK)	3 (23.7%)	13 (76.3%)	7 (43.8%)	9 (56.2%)	9 (56.3%)	7 (43.7%)
SOPA-Control	10.5 (3.7)	11.6 (4.1)	11.3 (4.0)	11.3 (4.1)	11.3 (4.1)	11.4 (4.0)
SOPA-Disability	7.9 (4.3)	7.4 (3.5)	7.4 (4.2)	7.6 (3.3)	7.2 (4.0)	7.8 (3.3)
SOPA-Harm	10.2 (2.2)	8.2 (3.8)	8.5 (3.3)	8.7 (3.7)	9.0 (3.4)	8.3 (3.7)
SOPA-Medication	12.7 (4.0)	11.4 (3.9)	12.1 (4.3)	11.5 (3.8)	12.0 (4.2)	11.6 (3.8)
SOPA-Emotion	9.8 (4.1)	9.7 (5.8)	9.1 (5.3)	10.1 (5.5)	9.1 (5.1)	10.2 (5.6)
SOPA-Solicitude	8.9 (4.9)	6.5 (4.5)	8.6 (4.5)	6.2 (4.6)	8.1 (4.5)	6.2 (4.7)
SOPA- Medical cure	11.9 (2.7)	11.8 (3.6)	12.4 (3.4)	11.5 (3.4)	12.1 (3.2)	11.6 (3.6)
PBPI-Mystery	-2.2 (4.1)	-2.9 (3.6)	-2.9 (3.3)	-2.6 (4.0)	-2.6 (3.3)	-2.8 (4.0)
PBPI-Permanence	-3.9 (4.1)	-4.5 (3.9)	-4.4 (3.3)	-4.4 (4.3)	-4.1 (3.4)	-4.6 (4.3)
PBPI-Constancy	1.1 (4.4)	1.2 (3.9)	0.6 (3.9)	1.5 (4.0)	1.0 (4.0)	1.3 (4.0)
PBPI-Self-blame	-4.7 (1.7)	-4.2 (2.1)	-4.2 (2.3)	-4.4 (1.8)	-4.2 (2.2)	-4.4 (1.9)
PCS	25.9 (9.2)	24.4 (9.5)	24.7 (9.1)	24.7 (9.6)	24.8 (9.2)	24.7 (9.6)

Bold indicates a possible systematic difference between participants with complete and incomplete data

baseline beliefs, it appeared that SOPA-solicitude scores were different between all follow-up comparisons. That is, the difference pattern was stable. Those with incomplete data had relatively higher endorsement of solicitude. There also appeared to be a difference in the SOPA-harm scale when baseline scores were compared for those with complete data and those lost to follow-up at three months. However, this difference was not stable in other comparisons raising the possibility that the difference could be due to chance. Sex and baseline solicitude was not related to future pain or disability. However, the belief that pain and exercise is harmful was related to disability in univariate analysis but not when after adjusting for a priori potential confounding factors. Thus, the systematic differences in sex and the solicitude and harm beliefs represent a potential threat to internal validity. However, sex was not related to the outcomes and was adjusted for in multivariate analysis. Moreover, considering solicitude had no relationship with the outcomes and the relationship between harm and disability was inconsistent and disappeared in multivariate analysis, the likelihood of threat to internal validity is minimized. Thus, similar to the discussion above regarding response bias, the primary consequence of attrition in this study pertained to generalizability.

An additional consequence of attrition is missing data. This may reduce the power of statistical tests leading to type II errors. A number of strategies exist to handle missing data. First, it must be determined if the missing data is extensive enough to influence power and whether the missing data is random or systematic. Random missing data that does not influence power can simply be deleted. Since the sample size of the study was small, simple list-wise deletion was avoided. Tabachnick and Fidell¹⁷ provide examples of various strategies to impute missing data. Estimation of missing data can be accomplished by substituting the variable's mean or predicting its value through regression, however this reduces the variance for that variable. The most valid method of imputation is multiple imputation.¹⁷ This procedure involves logistic regression where cases with and

without a missing value on a particular variable form the dependent variable. Once predictors are determined, several (three to five) random samples are drawn to estimate the value(s) of the missing data for that variable. The derived data sets are analyzed statistically with the average of the parameter estimates reported.¹⁷ However, while multiple imputation is more valid than other strategies, it still does not entirely mitigate selection bias. Considering observations of systematic differences in the data the benefits of imputation was carefully weighed with consequences of selection bias.

In this thesis, missing data was considered in two ways. First, if missing data occurred in individual items, the measure's score was pro-rated. This only occurred when one or two items were missing within a measure. The assumption in this case was items within a scale have been previously shown through factor analysis and measurement of internal consistency to represent the same construct. Thus, it is likely that missing values would be similar to completed items within the same scale. If data was missing due to attrition, imputation was not used. Considering systematic differences were discovered between participants with and without missing data, imputation would not solve the bias and may even perpetuate it. Moreover, particularly with regression, smaller sample sizes can over-estimate parameters. Thus imputing means or predicting missing values with regression is likely to decrease variance and further increase the risk of spurious associations being observed.

5.4. Perceived barriers to maximizing response

There are numerous reasons for non-response in research. For example, research participation may be viewed as not salient, overly burdensome, or perceived to be untrustworthy or harmful.^{16,18} Some or all of these barriers may have been present in this thesis. In particular, respondent burden was likely a significant factor. While the questionnaire itself was not overly burdensome, patients must complete

paperwork pertaining to their injury in the clinic. In addition, insurance paperwork also requires completion. Thus, a request to complete a survey may not have been viewed favourably in this context. Strategies to mitigate these potential concerns to maximize response were discussed above, as well as barriers to implementing these approaches.

While implementing more of the measures to maximize response outlined in Table 5-1 may have been successful, a number of other perceived barriers are worth mentioning. Much of this discussion has centred on maximizing response of research participants. However, many participants were not given the opportunity to respond. This statement is based on discussions with study contacts. Some staff were not aware of the study and those that were occasionally forgot to offer the survey. A related issue is primarily speculative. It was possible that many staff were not engaged in the data collection process. This lack of engagement may have contributed to reduced perceived saliency, which was identified as an important factor in increasing response.

A recent study¹⁹ highlighted the challenges of engaging clinicians in research. In this study,¹⁹ over 200 stakeholders (e.g. clinicians, scientists, professional and pharmaceutical representatives) were interviewed regarding the feasibility of clinician participation in research within their own practice setting. Their findings revealed three major categories of themes that impede clinician participation in research: (1) a need for greater attention to concerns of clinicians; (2) absence of infrastructure; and (3) (mal)alignment of financial structures. The first theme highlights a need to engage the clinician in the research process. That is, clinicians should not be simply passive partners. Active involvement increases the saliency of research for the clinician and assures a voice within the research endeavour. Active involvement could manifest as participation in the development of research questions and contributing to development of research methods. These contributions increase the likelihood that the study question is

meaningful and study procedures are not overly burdensome for clinicians. In addition, clinicians must feel that their contributions, large or small, are valued.

Absence of infrastructure pertains to clinicians' awareness of research opportunities and their lack of technical knowledge about research. In the context of this study, infrastructure was available insofar as the study's investigators provided technical knowledge and addressed ethics and other research operational requirements. However, infrastructure in the context of administrative research support within clinics would greatly decrease burden on clinic staff. Improved awareness and understanding of the basics of research may also lead to clinicians reaching out to researchers as opposed to researchers soliciting clinician participation. Related to infrastructure is alignment of financial structure to facilitate clinical research. Clinical practice in the context of private practice occurs within a business model. Thus, economics are of concern for clinicians. Participation in research must not be overly disruptive to practice and clinicians require fair compensation for their time. Whereas some physicians' motives to participation in research include altruism and prestige,²⁰ this cannot be relied on to maximize clinician participation and may lead to burn out of clinicians who repeatedly volunteer time for research.

5.5. Lessons learned

Reflection on the procedures used to recruit and retain research participants and review of recent literature have led to three recommendations for future recruitment and retention strategies: (1) increasing clinician engagement; (2) initial participant contact made by a member or employee of the research team; and (3) an incentive strategy for both clinicians/administrative staff and patients. Rather than the clinician perceiving their role to be passive, approaching clinicians as partners may improve engagement. Face-to-face meetings to propose the study question and ask for clinician input might be useful in increasing or

assessing their perceived saliency of the topic. While prospective clinical partners were consulted about the approach to data collection, a more thorough discussion might have been helpful to enable clinician awareness of the various strategies used to maximize response. This discussion could lead to negotiating a more effective recruitment strategy. These initial engagement meetings would also serve to emphasize the importance of maximizing participant response and the consequences of poor response to the validity of a study. Furthermore, the value of clinician participation and how this value is most fairly quantified with monetary and/or nonmonetary tokens of appreciation could be discussed. These strategies target many of the specific barriers outlined by Kahn et al.¹⁹

The second recommendation pertains to removing the burden of ‘selling’ the study from clinic administration staff and/or clinicians. Rather than clinic staff distributing surveys, a simple card is provided to all patients requesting their consent to be contacted by researchers. At the end of each week, a member of the research team would gather contact information. This strategy addresses a number of issues. The first is that the card is routinely given to all participants and would contain a small amount of information analogous to a ‘teaser’ statement mentioned in Table 5-1. For example: “Neck pain from a car accident? Learn how you can help researchers improve treatment; check this box if you are willing to be contacted by a researcher about a new study on whiplash”. This card could be routinely placed with each patient’s assessment form minimizing the chance that a study package is not distributed and requires no ‘sales pitch’ on the part of clinic staff. In addition, it ensures that researchers, who clearly view the study to be important, can promote the study at a time when participants are not burdened with other forms. Finally, this approach would permit multiple personalized contacts as recommended in the literature.¹³⁻¹⁵ Scheduled weekly meetings or phone calls with the clinic’s study contact might also be helpful in troubleshooting and increasing engagement.

The final recommendation is to include a monetary incentive strategy for both study contacts at clinics and research participants. In this thesis study, a relatively modest amount of the operating budget was devoted to reimbursing participants time for face-to-face interviews (\$900.00). A more substantial amount would be required to provide an incentive for the completion of each survey. In order to make sure incentives are not coercive, a modest amount would be offered (e.g. \$5.00/per survey completed). Small monetary incentives has been shown to improve response.^{2,21} Thus, to attain the sample originally desired (n=130) and factoring in an improved attrition rate (25%) than what was observed (34%), 162 participants would need to be recruited costing \$2,430.00. This cost would increase depending on response rate. While the operating budget for this study could not accommodate this cost, future studies could delay initiation until appropriate funds are raised. As was done in this thesis study, \$10.00 gift cards along with a thank you card could be given to each clinic study coordinator. However, tokens of appreciation that are more frequent (e.g. early, midway and late data collection) might help maintain recruitment momentum. If 20 clinics participated, this would cost an additional \$600.00. Finally, while dissemination was offered as a token of appreciation to clinicians participating, other non-monetary gestures could be offered such as educational pamphlets citing new literature in the assessment and treatment of whiplash injuries.

5.6. Conclusion

Maximizing response and minimizing attrition is vital to conducting valid and meaningful research. Consequences of each can include limited statistical power and threats to internal and external validity. Numerous methodological and analytical approaches are available to mitigate selection bias. Researchers must weigh the costs and benefits of implementing these strategies. While much attention in the literature is centred on maximizing response from research participants, considerable attention should be directed to maximizing saliency and

minimizing burden for clinicians as well. This is crucial if clinical physical therapy research is to be conducted. Recruiting participants from private clinical practices adds a layer of complexity due to potentially varied agendas of stakeholders. Future clinical research should consider multiple strategies to maximize response and minimize attrition. In particular, monetary incentives and efforts to improve clinician engagement ought to be prioritized.

5.7. References

1. Olson SH. Reported participation in case-control studies: changes over time. *Am J Epidemiol.* 2001;154(6):574-581.
2. Fumagalli L, Laurie H, Lynn P. Experiments with methods to reduce attrition in longitudinal surveys. Institute for Social and Economic Research. 2010;2010-04:
3. Templeton L., Deehan A, Taylor C, Strang J. Surveying general practitioners: does a low response rate matter? *Br J Gen Pract* 1997;47(424):91-94.
4. Harig R, Alte D, Volzke H, Sauer S, Wallaschofski H, Ulrich J, Schmidt CO. Extended recruitment efforts minimize attrition but not necessarily bias. *J Clin Epidemiol.* 2009;62(3):252-260.
5. Schmidt CO, Raspe H, Pflingsten M, Hasenbring M, Basler HD, Eich W, Kohlmann T. Does attrition bias in longitudinal population-based studies on back pain? *Eur J Pain.* 2011;15(1):84-91.
6. Oleckno WA. *Essential epidemiology: principles and applications.* Long Grove (IL): Waveland Press Inc., 2002.
7. Rothman KJ. *Epidemiology: an introduction.* New York: Oxford University Press, 2002.
8. Norman GR, Streiner DL. *Biostatistics: the bare essentials.* Lewiston, NY: BC Decker, 2000.
9. Katz MH. *Multivariable analysis: a practical guide for clinicians.* New York (NY): Cambridge University Press, 2006.
10. Cote P, Cassidy JD, Carroll L, Frank JW, Bombardier C. A systematic review of the prognosis of acute whiplash and a new conceptual framework to synthesize the literature. *Spine* 2001;26(19):E445-58.
11. Scholten-Peeters GG, Verhagen AP, Bekkering GE, van der Windt DAWM, Barnsley L, Oostendorp RAB, Hendriks EJM. Prognostic factors of whiplash-associated disorders: a systematic review of prospective cohort studies. *Pain* 2003;104(1-2):303-322.

12. Portney LG, Watkins MP. Foundations of Clinical Research: applications to practice. Upper Saddle River: Prentice Hall Health, 2000.
13. McColl E, Jacoby A, Thomas L, Soutter J, Bamford C, Steen N, Thomas R, Harvey E, Garratt A, Bond J. Design and use of questionnaires: a review of best practice applicable to surveys of health service staff and patients. *Health Technol Assess* 2001;5:1-256.
14. Dillman D. Mail and Internet Surveys: The Tailored Design Method, 2nd ed. New York (NY): John Wiley Co.; 2000.
15. Edwards PJ, Roberts I, Clarke MJ, DiGuseppi C, Wentz R, Kwan I, Cooper R, Felix LM, Pratap S. Methods to increase response to postal and electronic questionnaires. *Cochrane Database of Systematic Reviews*. 2009;3:Art. No.: MR000008. doi:10.1002/14651858.MR000008.pub4.
16. Robinson KA, Dennison CR, Wayman DM, Pronovost PJ, Needham DM. Systematic review identifies number of strategies important for retaining study participants. *J Clin Epidemiol*. 2007;60(8):757-765.
17. Tabachnick BG, Fidell LS. Using multivariate statistics, 5th ed. New York (NY): Harper Collins, 2007.
18. Barclay S, Todd C, Finlay I, Grande G, Wyatt P. Not another questionnaire! Maximizing the response rate, predicting non-response and assessing non-response bias in postal questionnaire studies of GPs. *J Fam Pract*. 2002;19(1):105-111.
19. Kahn K, Ryan G, Beckett M, Taylor S, Berrebi C, Cho M, Quiter E, Fremont A, Pincus H. Bridging the gap between basic science and clinical practice: a role for community clinicians. *Implement Sci*. 2011;6(1):34.
20. Mainous AG, Hueston WJ: Characteristics of community-based primary care physicians participating in research. *J Fam Pract* 1995, 40(1):51-56.

21. Deehan A, Templeton L, Taylor C, Drummond C, Strang J. The effect of cash and other financial inducements on the response rate of general practitioners in a national postal study. *Br J Gen Pract.* 1997;47(415):87-90.

CHAPTER 6

General Discussion and Conclusions

6.1. Overview

Beliefs about pain are personally formed or culturally shared cognitive understandings of pain.^{1,2} Examining pain beliefs provides insight into meaning that is central in the experience of pain.³ The International Association for the Study of Pain explicitly states that pain should not be considered synonymous with nociception, since nociception is neither necessary nor sufficient to produce pain.⁴ While nociception provides a true representation of a noxious stimulus, pain is not experienced until it is filtered through meaning. In this thesis, meaning of whiplash associated disorder (WAD)-related pain was examined. In chapter two, a novel application of the placebo hyperalgesic meaning response summarizes emerging neurochemical and neuroanatomical processes that underlie cognitive modulation of pain. Expectancy, a particular belief about pain or illness, is one proposed mechanism underlying this modulation. In chapter three, a mixed-methods study, WAD-related pain beliefs were characterized both quantitatively and qualitatively. Quantitative findings portrayed these beliefs numerically, permitting a standard comparison with future studies and with other populations. In addition, the stability of beliefs over time was examined statistically. The qualitative component addressed an absence of individual voices in representing the meaning of WAD. Shared inter-subjectivity revealed themes that formed endorsement of self-reported beliefs as measured via survey. Chapter four, provided a consequentialist examination of the potential importance of measuring beliefs early after injury in predicting outcome six months after the motor vehicle collision (MVC). In addition, WAD-related pain beliefs were examined for their association with catastrophizing. Finally, chapter five presented a reflection on data collection challenges for this research, proposing strategies to minimize the effects of selection bias on future studies.

Key findings from each of the three principal studies comprising this thesis are highlighted. Next, I present a conceptual model that explicitly integrates chapters two to four through observed commonalities. This model is situated within current-related literature.

6.2. The placebo hyperalgesic meaning response

Objective

To summarize evidence of the placebo hyperalgesic meaning response highlighting specific anatomical and neurochemical mechanisms leading to pain modulation via expectations.

Key findings

- 1) Mechanisms underlying cognitive pain modulation support theoretical assertions and clinical observations that cognitive factors are important in shaping pain experiences.
- 2) Negative expectations about pain can form a meaning response to noxious or normally non-noxious stimuli that can lead to, or increase, pain. This is known as placebo hyperalgesia or placebo allodynia.
- 3) In the context of a negative expectation, anxiety could be produced that activates CCK receptors in the brain. CCK can block descending hypoalgesia or increase descending hyperalgesia. Alternatively, negative expectations could change the experience of pain as demonstrated by brain imaging.
- 4) Physical therapists and other healthcare providers have the potential to contribute to negative expectations through well-meaning explanations that are interpreted negatively by patients.

6.3. An integrated mixed-method characterization of WAD-related beliefs

Objective

To use quantitative data that characterizes WAD-related pain beliefs over time to develop a qualitative method that provides additional inter-subjective meaning. Specifically, this qualitative component explored experiences that inform belief endorsement as manifested on a quantitative survey measuring WAD-related beliefs.

Key findings

- 1) The meta-theme restitution, representing a desire to be ‘fixed’ or ‘cured’, was foundational and integrative in the qualitative data.
- 2) Restitution may be adaptive early after WAD, but could become problematic for meaning construction later on.
- 3) In addition to the quantitative characterization of WAD-related pain beliefs, adjunctive meaning related to experiences informing endorsement of the SOPA-Medical cure, SOPA-Control, SOPA-Emotion and PBPI-Mystery scales was achieved.
- 4) Both quantitative and qualitative data suggested stability in beliefs over time.
- 5) Stigma emerged as a potentially important contextual descriptor in the formation of WAD meaning that warrants future study.

6.4. WAD-related pain beliefs associations with catastrophizing and pain and disability six-month post-MVC

Objective

To examine the relationship between beliefs, pain, disability and pain-related catastrophizing; and whether beliefs predict future pain and disability.

Key findings

- 1) Belief in the permanence of pain at baseline was predictive of pain intensity and disability six months post-MVC.
- 2) Belief in the mysteriousness of pain at baseline was predictive of disability six months post-MVC.
- 3) Consistent with previous research, baseline catastrophizing was predictive of pain and disability six months post-MVC.
- 4) WAD-related pain beliefs were associated with catastrophizing in a manner supportive of theoretical assertions related to catastrophizing.

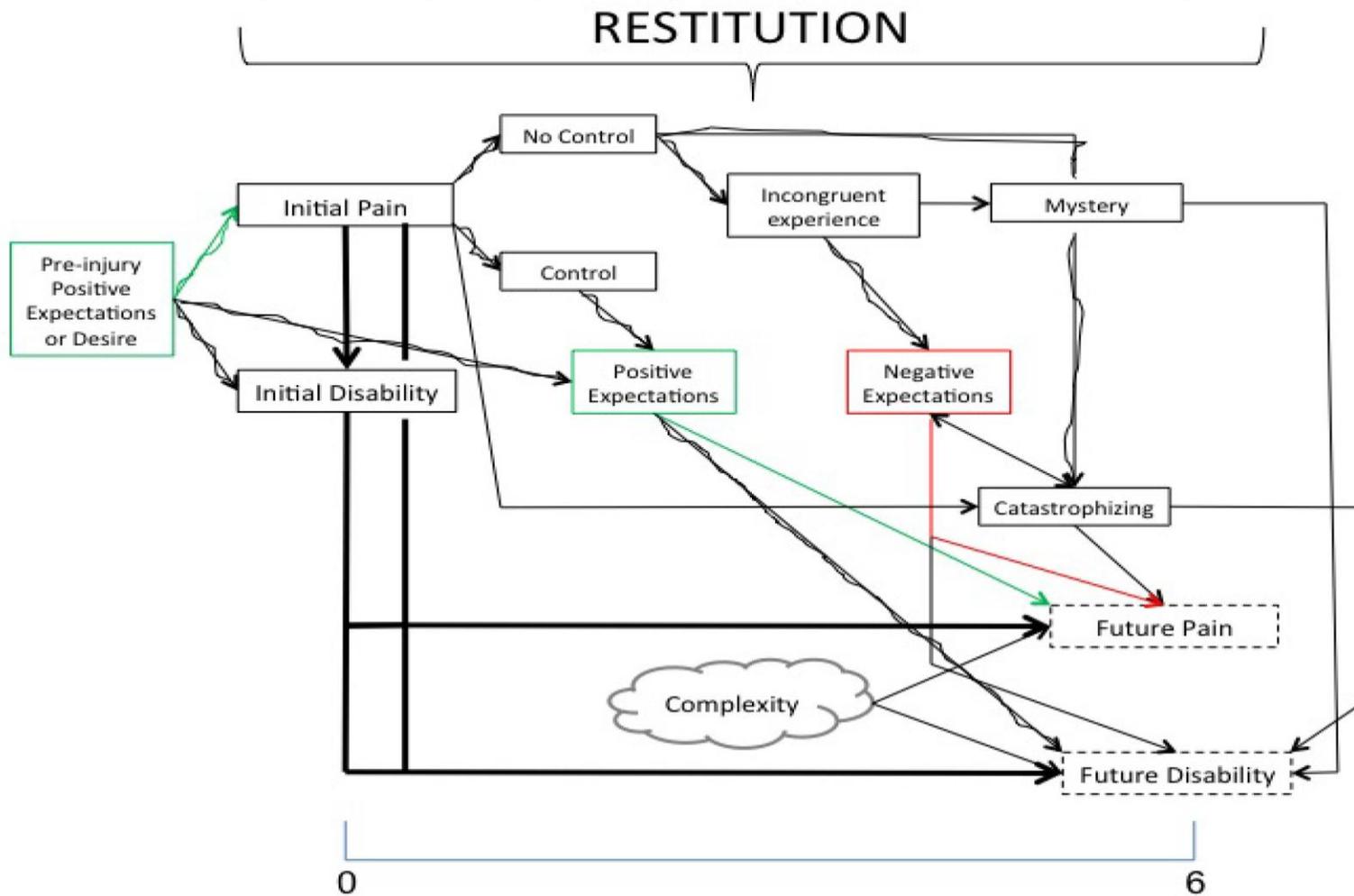
6.5. A pluralistic conceptual model of WAD-related pain beliefs

Figure 6-1 visually represents a conceptual model integrating findings from this thesis. Moving through the model from left to right, factors influencing the six-month outcomes pain and disability are shown. Green boxes illustrate a context of positive expectations or meaning response, while red boxes illustrate a negative expectancy. The red arrows represent the nocebo hyperalgesic meaning response as a biological mechanism influencing pain. The green arrows signify nocebos antithesis, placebo mediating pain response which is on the positive side of the meaning response coin. Thick arrows portray the strong and consistent relationships demonstrated in this study and others. Restitution is visualized on top of the model signifying its over-arching influence on the construction of meaning. While this model represents a parsimonious representation of the thesis findings, the intent was to maintain a degree of complexity. The experience of pain and the determinants of function are complex and multifaceted. To further illustrate this complexity, ‘squiggled’ lines were drawn on top of straight lines signifying the mixing of methods. The straight arrows imply statistically generated relationships while the ‘squiggled’ lines represent the necessarily ‘imperfect’ relations generated from idiosyncratic shared meaning. Furthermore, since arrows (squiggled or straight) imply linear relationships, a complexity/non-

linear term is introduced that represents a proportion of variance beyond measurement error that cannot predict outcomes owing to the unpredictability of inherently complex systems. The experience of pain, given its social, individual, biological and psychological determinants share characteristics of a complex adaptive system.⁵ For example, while pain and disability are considered outcomes, the boundaries pertaining to recovery are not clear. A system becomes complex when it is influenced by, and embedded within, other systems. The introductory chapter of this thesis illustrated this in discussing how the expression of pain is, in part, socially determined. Thus, the experience of pain is embedded within a complex social system (e.g. pain is expressed differently across cultures). In creating a model for the experience of pain, it is possible that no model will be accurately predictive or static. Attempting to represent such a model two-dimensionally is necessarily difficult. Thus, in lieu of the apparent complexity of the experience of pain it is prudent to include a term that attempts to account for a degree of non-linearity and unpredictability.

The placebo hyperalgesic meaning response (chapter two) highlighted the biological plausibility of meaning modulation of pain via expectations. The prognostic study (chapter four), emphasized belief related to the permanence of pain (similar to expectations) and catastrophizing as contributing unique individual variance for future pain and disability. Finally, the mixed-methods study (chapter three), the desire to become ‘cured’, ‘fixed’ or returned to a pre-MVC self (restitution) coloured motives for belief endorsement. This restitution narrative may be adaptive early after injury and in the context of congruence with expectations and experience. Thus, the model accentuates both a scenario where restitution fits, and one where it does not. When positive recovery expectancies are congruent with the recovery experience, restitution is a useful narrative. However, when the experience of pain persists, one is faced with a cognitive predicament.

FIGURE 6-1: A conceptual model representing key findings from this thesis characterizing the meaning of WAD-related pain



This conceptual model highlights key findings within the thesis. In addition, it highlights a common theme unifying the three studies. Expectations was found to be prominent in the review of the nocebo hyperalgesic meaning response, the thematic analysis of experiences that informed belief endorsement, and in predicting future outcomes. The major components of the conceptual model are discussed with specific emphasis on expectations. Supportive literature is integrated within the discussion.

6.5.1. Restitution

An overarching concept within the model summary is the meta-theme generated from chapter three, labeled restitution. The argument is that a sufferer's story is told through a restitution narrative.⁶ Thus, the meaning of one's pain (including one's belief) is filtered through restitution. The restitution narrative is a consequence of modernist views on illness and health. That is, the body can be cured by commodity.⁶ Drugs, surgery, and other services can remediate illness and injury. This was explicitly observed in chapter three as endorsement of a belief in cure and rejection of the belief in pain permanence. These beliefs represent an expectation for remediation or, as will be discussed below, a desire for remediation. The importance of restitution became even more prevalent in later interviews in chapter three when expectations for remediation were incongruent with the experience of pain. This conflict in meaning revealed an important limitation of restitution:

“The first limitation of restitution stories is the obvious but often neglected limitation of the modernist deconstruction of mortality: when it [restitution] doesn't work any longer, there is no other story to fall back on. Restitution stories no longer work when the patient is dying or when the impairment will remain chronic.”^{6(p.94)}

While expectations for cure are helpful and lead to seeking health care provider assistance in facilitating health, for a small but significant portion of people who experience WAD, pain will become chronic. Chronic pain is simply incompatible

with restitution. To live with chronic pain through the lens of restitution means an endless search for meaning through observable patho-anatomy. In addition, exotic and sometimes expensive treatments are sought to deliver cure. This meaning construction of pain is maladaptive in the context of chronic pain, as no known 'cure' exists for managing the pain. Overcoming this meaning of pain is a focus in cognitive-behavioural approaches for managing chronic pain.

The nocebo hyperalgesia meaning response (chapter two) and prognostic data (chapter four) suggests negative expectations may exacerbate the experience of pain. In study two, inter-subjective interpretations of meaning also placed value in maintaining a positive outlook. However, the danger comes when expectations do not match experience. This is not to say that maintaining positive expectations are not important. Rather, it is important that one's conceptualization of pain must leave room for a framework that can accommodate two scenarios: (1) an uneventful recovery from WAD, and (2) an incomplete or absence of recovery. As described in the introduction, conceptual frameworks such as the biocultural model⁷ are capable of rationalizing both scenarios. The so-called dogmatic medical model⁸ is only capable of explaining scenario one and is congruent with narratives of restitution. Frank⁶ argues that restitution stories are preferred stories and are strongly maintained by institutional and media forces. Thus, shifting pain meaning would likely require what Kuhn⁹ calls a paradigm shift. For this to occur, a collective must experience a crisis.⁹ For many people, the medical model works nicely. Despite fervent calls for alternate paradigms,^{7,8,10} modernist views on illness and pain remain pervasive. This thesis suggests meaning rooted in restitution has important consequences. Strategies to translate alternate ways of conceptualizing injury, illness and pain are needed.

6.5.2. Baseline pain and disability predict future pain and disability

Baseline pain intensity and disability are strong and consistent predictors of future pain and disability. This has been supported in numerous other studies.^{11,12} However, it would be useful to understand in more depth about the importance of baseline pain intensity. Sterling¹³ has shed some light on this by examining measures of nervous system hypersensitivity. For example, combinations of predictor threshold scores have been used as a way to ascertain potential interactions with baseline variables that may better explain the impact on future disability.¹⁴ An individual with a cold temperature pain threshold above 13 degrees, is older in age and with high initial pain has an 84% likelihood of reporting persistent disability.¹⁴ While hypersensitivity is believed to characterize chronic pain, evidence suggests these changes occur early on and are thus important in shaping the early experience of pain.¹⁵ These data are important in gaining understanding about factors that determine self-reported baseline pain.

The conceptual model proposed in this thesis is heavily focused on cognitive determinants of future pain and disability. Sterling's work^{13,14} could be integrated and would contribute biological and affective mediators/moderators adding to the meaning of WAD. A recent review¹⁶ on catastrophizing provides yet another heuristic integrating various theoretical mechanisms in determining outcome. Thus, while the early pain/disability late pain/disability relationship is solid, complexity is superimposed when the multitude of biological, psychological and cultural factors are considered.

6.5.3. Expectations in the meaning of WAD-related pain

The red and green arrows within the model suggest the possible underlying biology of expectations in determining the experience of pain. In the model, pre-MVC expectations are hypothesized to contribute to the initial pain response. While these were not measured pre-MVC, there is theoretical¹⁷ and empirical¹⁸ literature supportive of this. Expectations, in this study, were one of the strongest

beliefs predictive of outcome for both pain intensity and disability at six months. Expectations were operationalized within the pain beliefs and perceptions inventory as a belief in pain permanence. Though measured differently, expectations about future outcome have been shown to be predictive of similar outcomes in a variety of conditions,¹⁹ including WAD.²⁰ Interestingly, despite evidence to suggest beliefs about WAD are negative²¹ the over-arching theme of restitution was that people expected cure. This could be construed as a positive expectation. Specifically, Maddux¹⁷ considers this a behaviour-outcome expectancy. That is, an expectation that a specific behaviour will probably lead to a specific outcome. In this study, there was an assumed expectation that seeking care (behaviour) would remediate pain and disability (outcome).

Expectation consists of a wide range of sub-constructs. For example, the perceived negativity surrounding WAD might be considered a stimulus-response expectancy. This is an expectation that a stimulus (MVC) signals the probable occurrence of a nonvolitional response (e.g. prolonged pain). Maddux¹⁷ suggests a stimulus-response expectancy might be a trigger for a behaviour-outcome expectancy. Qualitative data from this study suggested a general belief in remaining optimistic in all parts of life. This could also represent a stimulus-response expectancy insofar as any negative or challenging situation (stimulus) will be overcome (response). This would be consistent with the overarching theme of restitution as well. Thus, competing stimulus-response expectancies could influence the behaviour-outcome expectancy above.

An alternate consideration is related to the construct desire. Jensen and Karoly²² considered desire to be the degree to which one wants to experience a symptom change. This could also be construed as motivation and possibly resembles the meta-theme restitution. Alternatively, expectancy is the expectation for symptom change. Clearly, these constructs are closely related. However, motivation has been shown to contribute more variance in placebo responses compared to

expectancy.²² Thus, separating these constructs may be important.¹⁸ In most cases, patients likely desire to have pain remediated. This was certainly the case in chapter three. In the context of negative expectations, is it possible to desire to have pain abolished, yet expect it not to be? The relative consistency in believing in cure could, in part, be related to desire, while considering negative future outcomes more closely approximates negative expectations. Many patients likely wrestle with this conundrum. That is, a desire to achieve restitution but a belief that this may not occur. This problem was evident in the chronic pain participants from chapter three. Future studies examining expectancy beliefs should distinguish between desire and expectancy.

Of course, expectations are simply predictions based on probabilities. A number of patients who experience WAD may have positive behaviour-outcome expectations but have an experience incongruent with their expectation. This was illustrated in Figure 6-1. Attribution, similar to expectancies, is concerned with perceived control. While expectations predict future events, attribution explains current scenarios.¹⁷ Attribution was discovered within the qualitative data when expectations were incongruent with experience leading to a belief in pain mysteriousness. Attribution was sought to reconcile this. Olsen et al²³ suggests that expectancy-disconfirming events are often attributed to external and unstable causes. This would be consistent with the restitution narrative and anecdotal observations that people experiencing chronic pain use significant resources when seeking expert opinion and/or sophisticated imaging to explain the cause of their pain. The explicit appraisal process described here also fits within theoretical assertions of catastrophizing discussed below.

Perceived control shares similarities with expectations. The SOPA-control scale examines one's belief in their ability to control pain. This scale constitutively approximates what Maddux¹⁷ defined as personal-outcome expectancy beliefs. Personal-outcome expectancies are those related to the probability that one will or

will not attain an outcome.¹⁷ These expectancies comprise self-efficacy and behaviour-outcome expectancies.¹⁷ Constitutive similarity is supported by strong correlations observed between SOPA-control and future outcome beliefs measured by the PBPI-permanence scale. The importance of self-efficacy has been emphasized elsewhere²⁴ providing further conceptual support for the relationship between SOPA-control and PBPI-permanence (expectancy) observed.

The discussion including expected outcomes and responses of pain based on behaviours and stimuli could be substituted for alternative outcomes in the affective domain. Cognition and affect are known to be intimately related.¹⁶ A possible outcome of decreased worry and helplessness could be expected from seeking care for WAD. Indeed, proxy measures of expectations such as the PBPI-permanence and SOPA-control scales were correlated with catastrophizing, suggesting a relationship. In addition, qualitative data indicated negative emotional responses such as worry and helplessness occurred when participants felt they had difficulty controlling their pain. While affective measures of pain were not assessed in this study, there is evidence to suggest that expectations preferentially influence pain unpleasantness.¹⁸ This is not to say that the sensory component of pain is not influenced, but rather there appears to be a strong support for a relationship between pain and mood. The nocebo hyperalgesic meaning response also supports this assertion as evidenced by the mediating effect of anxiety. Thus the association arrow between catastrophizing (catastrophizing is considered a cognitive and affective construct^{16,25}) and expectations is justified. Whether negative expectations lead to maladaptive affect, or if negative affect leads to negative expectations is not clear. Thus, a bi-directional association is depicted in Figure 6-1.

6.5.4. Catastrophizing in the meaning of WAD-related pain

Another prominent feature of the conceptual model is catastrophizing. In this model, similar to expectations, catastrophizing was associated with future pain and disability. This finding is consistent with previous research.^{16,25} While chapters three and four were primarily exploratory, some data support theoretical mechanisms underlying catastrophizing. Of particular salience is the view that catastrophizing influences pain and disability through an appraisal process. This theory is based on Lazarus and Folkman's transactional model of stress and coping²⁶ where primary and secondary appraisal processes interact to shape the cognitive and behavioural coping strategies initiated.¹⁶ As this theory relates to pain, a primary appraisal consists of an assessment of the threat-value of pain. A secondary appraisal comprises judgments of available coping options and whether they could be successfully executed.

The relationship between expectations and catastrophizing demonstrated in this model supports appraisal theory. Catastrophizing (specifically rumination and magnification) might be considered a primary appraisal of perceived threat²⁷ and one's expectations a secondary appraisal. A threatening appraisal of pain (primary appraisal) would likely consist of endorsements of statements such as "I wonder whether something serious might happen" and "I keep thinking about how much it hurts". As mentioned above, one's personal-outcome expectancey includes both self-efficacy expectations and behaviour-outcome expectations, which would fit the concept of a secondary appraisal. Moreover, the model suggests perceived control influences catastrophizing. This was determined through cross-sectional associations in chapter four and has been reported elsewhere as well.²⁵ Further support stems from the qualitative arm of the study that suggested factors influencing endorsement of SOPA-control beliefs included the perceived severity of the injury. This represents a primary appraisal of threat value that may inform catastrophic thinking directly or through the assessment of one's capacity to control pain.

The preceding discussion is not meant to suggest an appraisal conceptualization of catastrophizing is the only valid theoretical mechanism. Since there were no measures capable of assessing competing theories such as attention bias, the central nervous system (CNS), or neurophysiological mechanisms, it is not possible to say how these could be integrated into the conceptual model. However, emerging data suggest that biological mechanisms (e.g. CNS and neurophysiological) could be applied in a similar manner to the application of the nocebo hyperalgesic meaning response to expectancy.¹⁶ A communal coping mechanism has also been put forth as a potential mechanism. The suggestion is that catastrophizing represents a motive to garner support from others.^{16,25,28} In chapter four, solicitude was associated with catastrophizing, which may provide superficial support for this proposed mechanism.

In summary, the conceptual model integrating the three principle chapters within this thesis emphasizes expectations as an important component of forming WAD-related meaning. Expectations provide a biological explanation for altered pain experiences. In addition, beliefs rooted in modernism suggest that an expectation or desire for cure broadly informs meaning. Incongruent experiences and expectations can lead to a negative orientation about future outcomes, despite a desire for cure, leading to internal conflict. In addition to the importance of expectations in shaping meaning, they appear to be important in contributing variance in predicting future pain and disability. Finally, conceptual models such as the one presented here visually depict simple linear relationships. By combining divergent qualitative and quantitative methods, models become necessarily complex and relationships appropriately blurred due to the idiosyncratic experiences of pain and illness.

6.6. Conclusions

This pluralistic bricolage provides a broad view of WAD-related pain beliefs consistent with contemporary biopsychosocial or biocultural frameworks. The review of the nocebo hyperalgesic meaning response provided an example of a biological rationale for meaning modulation of pain through expectations. While arguments were made in chapter three that cognitive modulation of pain should not be reduced to biology, the overall theme of the thesis supports pluralism. That is, biological rationales of meaning are valuable but do not preclude a social constructivist perspective.

In chapter three, quantitative and qualitative data were mixed to provide an intersubjective account of factors that lead to endorsement of beliefs as measured through survey. Thematic analysis revealed potential relationships between prominent features within a proposed conceptual model. For example, beliefs related to control impacted expectations. When expectations were incongruent with experience, mystery resulted. Mystery was related to future disability, which underscores one problem with restitution narratives. In addition, data suggested a lack of control could be associated with affective constructs such as catastrophizing. Finally, the meta-theme of restitution was argued as one that coloured the meaning of pain for participants and was informative of the generated themes. Restitution is a narrative that views pain and disability as a temporary state that is to be restored to a pre-MVC state; to be fixed. This view can be helpful early on in seeking help, shaping positive expectancies for outcome, and limiting perceived threat. However, as pain and disability persists, this concept can be problematic as expectancies are challenged, control is lost, treatment becomes unsuccessful, and the puzzle of pain remains unsolved; all of which is distressing. An alternate narrative may be required to better frame beliefs to minimize distress.

Finally, in chapter four the central role of the meaning representations of expectancy and catastrophizing in determining future outcomes such as pain and disability was emphasized. Both expectancy and catastrophizing offered unique variance in the prediction of pain intensity and disability six months post-MVC after controlling for potential confounding (baseline pain intensity and disability, age, sex, and previous history of WAD). In addition, relationships between beliefs and catastrophizing provided support for theoretical mechanisms underpinning catastrophizing, particularly appraisal theory.

This thesis represents a unique contribution to the conceptualization of the meaning of WAD-related pain (as examined via belief). A pluralistic approach was taken that viewed meaning through biological, inter-subjective and statistical lenses. The result was not only a broad, yet deep, characterization of beliefs, but also an integrated précis of the potential role of expectancy in shaping WAD meaning and contributing to WAD outcomes.

6.7. References

1. Fishbein M, Ajzen I. Belief, attitude, intention and behavior: An introduction to theory and research. Reading, MA: Addison-Wesley, 1975.
2. Wrubel J, Benner P, Lazarus RS. Social competence from the perspective of stress and coping. In: Wine J, Syme M editors. Social competence. New York (NY): Guilford Press, 1981.
3. Morris, DB. Sociocultural and Religious Meanings of Pain. In: Gatchel RJ and Turk DC eds. Psychosocial Factors in Pain: Critical Perspectives. 1999. The Guilford Press: New York.
4. IASP Taxonomy [internet]. 2011 [cited 2011 Jun 17]. Available from: <http://www.iasp-pain.org/Content/NavigationMenu/GeneralResourceLinks/PainDefinitions/default.htm>.
5. Plsek PE, Greenhalgh T. The challenge of complexity in health care. *BMJ* 2001; 323(7313):625-8.
6. Frank AW. The wounded storyteller: Body, illness, and ethics. 1995 The University of Chicago Press: Chicago IL.
7. Morris, DB. How to Speak Postmodern: Medicine, Illness, and. Cultural Change. *Hastings Center Report* 2000; 30(6):7-16.
8. Engel GL. The need for a new medical model: A challenge for biomedicine. *Science*. 1977; 196(4286): 129-136.
9. Kuhn TS. The structure of scientific revolutions, 2nd ed. Chicago (IL): The University of Chicago Press; 1970.
10. Gatchel RJ, Turk DC. Criticisms of the biopsychosocial model in spine care: creating and then attacking a straw person. *Spine*. 2008;33(25):2831-2836.
11. Carroll LJ, Holm LW, Hogg-Johnson S, Cote P, Cassidy DJ, Haldeman S, Nordin M, Hurwitz EL, Carragee EJ, van der Velde G, Peloso PM,

- Guzman J. Course and prognostic factors for neck pain in whiplash-associated-disorders (WAD): Results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and its Associated Disorders. *Spine* 2008;33:S83S92.
12. Walton DM, Pretty J, MacDermd JC, Teasell RW. Risk factors for persistent problems following whiplash injury: results of a systematic review and meta-analysis. *J Orthop Sports Phys Ther* 2009;39:334-350.
 13. Sterling M. Differential development of sensory hypersensitivity and a measure of spinal cord hyperexcitability following whiplash injury. *Pain*. 2010;150(3):501-506.
 14. Sterling M, Hendrikz J, Kenardy J. Similar factors predict disability and posttraumatic stress disorder trajectories after whiplash injury. *Pain*. 2011;152(6):1272-1278.
 15. Akparian AV, Baliki MN, Geha PY. Towards a theory of chronic pain. *Prog Neurobiol*. 2009;87(2):81-97.
 16. Quartana PJ, Campbell CM, Edwards RR. Pain catastrophizing! A critical review. *Expert Rev Neurother*. 2009;9(5):745-758.
 17. Maddux JE. Expectancies and the social-cognitive perspective: basic principles, processes, and variables. In: Kirsch I (editor). *How expectancies shape behaviour*. Washington (DC): American Psychological Association; 1994.
 18. Price DD, Barrell JJ. Expectation and desire in pain and pain reduction. In: Kirsch I (editor). *How expectancies shape behaviour*. Washington (DC): American Psychological Association; 1994.
 19. Mondloch MV, Cole D, Frank J. Does how you do depend on how you think you'll do? A systematic review of the evidence for a relation between patients' recovery expectations and health outcomes. *CMAJ* 2001;165:174-9.

20. Carroll LJ; Holm LW, Ferrari R, Ozegovic D, Cassidy DJ. Recovery after whiplash associated disorders: do you get what you expect? *J Rheumatol* 2009;36:1063-1070.
21. Bostick GP, Ferrari R, Carroll LJ, Russell AS, Buchbinder R, Krawciw D, Gross DP. A population-based survey of beliefs about neck pain from whiplash injury, work-related neck pain, and work-related upper extremity pain. *Eur J Pain* 2009; 13(3):300-4.
22. Jensen MP, Karoly P. Motivation and expectancy factors in symptom perception: a laboratory study of the placebo effect. *Psychosom Med*. 1991;53(2):144–152.
23. Olsen JM, Roese NJ, Zanna MP. Expectancies. In: Higgins ET, Kruglanski AW, editors. *Social Psychology: handbook of basic principles*. New York (NY): Guilford; 1996.
24. Kyllback M, Theirfelder T, Soderlund A. Prognostic factors in whiplash-associated disorders. *Int J Rehabil Res* 2002;25:181-187.
25. Sullivan MJL, Thorn B, Haythornthwaite JA, Keefe F, Martin M, Bradley LA, Lefebvre. Theoretical perspectives on the relation between catastrophizing and pain. *Clin J Pain* 2001;17:52-64.
26. Lazarus R, Folkman S. *Stress, appraisal and coping*. New York (NY): Springer; 1984.
27. Severeijns R, Vlaeyen JWS, van den Hout MA. Do we need a communal coping model of pain catastrophizing? An alternative explanation. *Pain*. 2004;111(3):226-229.
28. Thorn BE, Keefe FJ, Anderson T. The communal coping model and interpersonal context: problems or process? *Pain* 2004;20:275-82.

APPENDIX A

Researcher reflexivity

Demographic reflexivity:

I am a 34-year-old PhD candidate in the Faculty of Rehabilitation Medicine at the University of Alberta performing the current study as part of my PhD thesis project. I am a white Canadian male whose socio-economic status would be classified as “middle-class”. Prior to entering the PhD training program, I was trained as a physical therapist and worked clinically for 5-years. Throughout my practice years, I obtained specialized clinical training in manual therapy. I also developed an interest in chronic pain clientele and an interest in pain; in particular, the cognitive aspects of pain.

Personal reflexivity:

Throughout training and practice as a physical therapist, my view on health and illness was primarily through the lens of the medical model (positivist/post-positivist). Literature that I read was primarily quantitative and I viewed research questions in a positivist/post-positivist perspective. Thus, entering the PhD program my comfort level was viewing health and illness in quantitative and positivist/post-positivist perspectives. As my studies in pain and research progressed I became more aware of the naturalistic paradigm. Given the large shift in considering research questions from purely quantitative methods to one that includes naturalistic inquiry, I faced considerable challenges framing this positivist ‘upbringing’. This was compounded by the choice of using a mixed-methods approach. Moving from paradigm to paradigm required considerable reflection on my part to re-familiarize myself with the tenants of the naturalistic paradigm. While a qualitative researcher aims to explore inquiry with a blank-slate, this is pragmatically not possible. Thus, I needed to be explicit in reflecting on how my considerable experiences in the quantitative paradigm potentially influenced my interpretations. In other words, qualitative analysis and

interpretation does not occur tacitly for me. Throughout this process, I required peer debrief sessions to help ensure that I was facilitating subjectivity.

As a novice researcher undertaking my first qualitative analysis and interpretation as principal investigator, I was acutely aware of the immense responsibility of appropriately representing the research participants in the data analysis. I have employed measures to facilitate appropriate representation of the data (member checking), however, appropriate representation of the data remained at the forefront of my mind throughout the process. Moreover, given the positivist/post-positivist influence from my clinical training and practice and the need to frame this aspect of my experience further increased my sensitivity to adequately represent participants voice throughout the process. As a consequence, the analysis and interpretation of the data tips toward description and deduction as opposed to interpretation and induction. That is, I purposively stayed very close to the language used by the participants in the analysis process. In addition, the interviews are framed based on quantitative data that were constructed on views (cognitive-behavioural approach to pain) that I am familiar with. Therefore, existing theory either explicitly or implicitly influenced my interpretations (appropriately so given the design). This study is not theory building, so an inductive process is less appropriate.

I am strongly influenced by literature advocating a biopsychosocial approach to pain. More specifically, cognitive-behavioural based physical therapy interventions such as patient education. I am also engaged in literature in the followings areas: (1) consciousness theory applied to pain, (2) Arthur Frank's illness narratives (restitution, chaos, and quest), and (3) biological research related to placebo and nocebo effects. While much of this reading is congruent with a naturalistic paradigm, my beliefs in a biopsychosocial and subjective reality are likely to influence my interpretations. Indeed, I encouraged participants to discuss

their subjective experiences and psychosocial aspects of their injury in cases when the focus was primarily biological.

As a researcher, I have an interest in understanding a particular subject matter. This is likely not lost on the research participants and the relationship between researcher and participant may very well yield discourse that differs from other relationships. Moreover, the research participant may view the researcher differently from a social perspective that may also influence a particular discourse unique to this relationship. Therefore, the experiences elicited in these interviews likely reflect motives surrounding increasing understanding of an individual's experience for the purpose of illuminating a broader understanding that may be applied to others.

Ontological/Epistemological reflexivity:

The primary factor to consider here is that the qualitative research occurred within the context of a quantitative study (i.e. a mixed-methods study). In other words, the inquiry was not purely naturalistic. Some may argue that mixed-methods research is in contrast to principles of naturalistic inquiry. However, considering the research question, a mixed-methods design was deemed most appropriate. Thus, the interview guide was based on quantitative data so the interview was focused on particular beliefs as opposed to a purely naturalistic inquiry that may permit a more open inquiry to beliefs that may not be captured in a pre-fabricated measure. This issue is discussed above and I was aware of the need to frame the quantitative perspective when engaged in the qualitative analysis.

As a mixed-method approach integrates quantitative and qualitative paradigms that have very different views of reality, adhering to a particular framework is challenging. Thus, a purist perspective is difficult to defend. Instead, a pragmatic worldview guided the research process. That is, a moderate perspective to purist's views was used. Much of the qualitative portion was conducted through a

constructivist lens. Constructivism supports realism and subjectivism. Pragmatism values this perspective but views it more of a continuum. Whereas realism and subjectivism were influential in this study, it is acknowledged that a purist perspective is not entirely congruent with a mixed-method approach.

APPENDIX B

Sample interview guide

SURVEY RESPONSES

WDQ: 89/130

SOPA:

- Control: $5^* = 2$, $10^* = 3$, $21 = 2$, $24 = 1$, $29^* = 2$: 10/20 (A)
- Disability: $4 = 3$, $13^* = 2$, $23 = 2$, $26^* = 1$, $35 = 3$: 11/20 (M)
- Harm: $1 = 4$, $7^* = 3$, $16^* = 2$, $20 = 1$, $34^* = 2$: 12/20 (M)
- Emotion: $9 = 2$, $15 = 3$, $19 = 2$, $28 = 1$, $30^* = 2$: 10/20 (A)
- Medication: $2 = 1$, $8 = 1$, $17 = 0$, $32^* = 4$, $33^* = 3$: 9/20 (M)
- *Solicitude*: $3 = 2$, $11 = 1$, $14 = 1$, $18 = 1$, $22 = 1$: 6/20 (M)
- Medical Cure: $6^* = 1$, $12 = 3$, $25 = 1$, $27^* = 3$, $31 = 1$: 9/20 (M)

WBQ:

- *Recovery Pessimism*: $2^* = 4$, $3^* = 4$, $6^* = 3$, $8^* = 5$: 16/20
- Active coping: $5 = 4$, $10^* = 4$, $11 = 4$: 12/15
- Passive coping: $7^* = 4$, $9^* = 4$, $12^* = 2$: 10/15
- Treatment Pessimism: $1^* = 3$, $4^* = 4$: 7/10
- End up in w/c = disagree
- Alternative treatments = don't know

PBPI:

- *Mystery*: $1 = -1$, $4 = 2$, $8 = 1$, $14 = 1$: 3
- *Permanence*: $2 = -1$, $5 = -1$, $9^* = -1$, $12^* = -1$, $15^* = -1$: -5
- *Constancy*: $3^* = 2$, $6 = 1$, $10 = 2$, $16 = 2$: 7
- *Self-Blame*: $7 = -1$, $11 = -1$, $13 = -1$: -3

PCS:

- Rumination: $8 = 3$, $9 = 2$, $10 = 1$, $11 = 2$: 8/20
- Magnification: $6 = 3$, $7 = 1$, $13 = 1$: 5/15
- Helplessness: $1 = 3$, $2 = 1$, $3 = 1$, $4 = 3$, $5 = 2$, $12 = 2$: 12/30

QUALITATIVE INTERVIEW GUIDE

Research question: How do the lived experiences of patients with neck pain from whiplash inform their beliefs about their pain condition?

I. INTRODUCTION:

“The purpose of this interview is for you to tell me more about your pain and to expand on what you told me in your survey. I am very interested in your information and experience. This information will help us to better understand neck pain after whiplash injury. I would like to remind you that the interview will be recorded. All of your information is confidential and you may choose not to answer any question that you are not comfortable with. You can turn off the recorder at any time”.

II. CONTEXTUALIZE THE INTERVIEW:

a. Can you describe to me what happened in the car accident? [Follow-up: what were you feeling immediately after it happened? What were your initial thoughts about the accident?]

b. What does your neck pain mean to you. Follow-ups: what are some of the things you think about when you are in pain – what do you think those things mean; describe some of the feelings you have when you experience your pain – what does it mean when you feel that way?

III. DETAILS ABOUT SPECIFIC BELIEFS:

- Based on your questionnaire, I interpreted that you believed/felt that your pain is mysterious, is that accurate?
 - Tell me why you feel that way.
 - Give me an example why your pain confuses you?
 - What does it mean to you if you do not fully understand your pain (or finds their pain confusing)?
 - What do you think about/how does it make you feel not knowing enough about your pain?

- Mysterious items: No one has been able to tell me exactly why I am in pain, my pain is confusing, I don't know enough about my pain, I can't figure out why I am in pain

- Based on your questionnaire, I interpreted that you believed/felt that your pain will not be permanent, is that accurate?
 - Why do you feel that way?

 - Tell me about some of your experiences that have led to this belief.

 - Tell me about how being optimistic makes you feel (or optimism influence your recovery)

 - Do you believe there is a cure for your pain (how do you define cure)?

 - Permanence items: I used to think my pain was curable, but now I am not so sure, my pain is here to stay, pain is temp pblm in life, someday I'll be 100% painfree, there is a cure for my pain

- Based on your questionnaire, I interpreted that you believed/felt your pain was constant?
 - Tell me why you feel that way?

 - What experiences have you had that lead to this belief?

 - What thoughts or feelings do you have when you reflect on experiencing continuous pain?

 - Constancy items: there are times when I am pain free, I am continuously in pain, wake up and go to sleep with pain, my pain varies in intensity but is always with me

- Based on your questionnaire, I interpreted that you believe pain is an sign of damage being done, is that accurate?
 - Why do you feel that way?
 - Can you give me an example?
 - Have you ever experienced a time when you have felt pain, but not believed it to be a sign of damage being done? Tell me about this?
 - Some people think emotions influence pain, what do you think about that?

Now that we have talked a little about your pain, compared to when we starting talking, would you change anything about your meaning of pain?

Interviewer summation: “What I heard in the interview was ... (summarize key points), did I understand correctly? Is there anything that you were thinking about that we didn’t discuss that you would like to tell me about?”

Close interview: “Thank you for your time and for sharing your story”

APPENDIX C

Details of Trustworthiness Analysis

Confirmability Inquiry Audit

*Confirmability criteria	Acceptable	Not acceptable
1. Was the interpretation grounded in the data? <ul style="list-style-type: none"> • Sample A • Sample B • Sample C 	X	
Comments:		
2. Are the interpretations logical? <ul style="list-style-type: none"> • Sample A • Sample B • Sample C 	X	
Comments:		
3. Is the category structure clear and of adequate explanatory power? <ul style="list-style-type: none"> • Sample A • Sample B • Sample C 	X	
Comments:		
4. Is inquirer bias likely based on the investigators reflexivity account? <ul style="list-style-type: none"> • Sample A • Sample B • Sample C 	X	
Comments:		
5. Overall, was the degree of confirmability acceptable?	X	
Comments:		

*refer to the page following for details regarding each confirmability criteria.

Detailed description of confirmability criteria to guide inquiry audit (per Lincoln and Guba, 1985):

1. Is the interpretation grounded in the data?
 - Evaluate whether there is convergence between the thematic interpretation, coding and raw data.

2. Are the interpretations logical?
 - Evaluate whether an appropriate analytic technique was selected?
 - Evaluate whether the analytic technique was applied properly?
 - Do category labels accurately describe the concepts?

3. Is the category structure clear and of adequate explanatory power?
 - Is there an unintended mixture of levels of analysis?
 - Is there an unclear method of analysis?
 - Do the categories support an exhaustive account of the data?
 - Do the categories describe the data/phenomena at the same level?

4. Is inquirer bias likely based on the investigator's reflexivity account?
 - While bias likely exists, does this bias misrepresent the data?
 - Is there an imposition of inquirer's own terminology in the data?
 - Is there a sufficient description of the inquirer's tacit processes (i.e. reflexivity account)?

5. Overall was the degree of confirmability acceptable?
 - Are the efforts to ensure confirmability acceptable?
 - Did the inquirer account for negative evidence?
 - Did the inquirer accommodate negative examples?

Member check

The table below summarizes the findings from respondents (5) returning member check forms.

Belief	Extent of agreement with thematic analysis	Comments
Medical cure	All respondents agreed or mostly agreed	<p>“A positive attitude is important in every aspect of my life. Pain or no pain. Being optimistic is good for the immune system.”</p> <p>“Agree that optimism plays a huge role in recovery and ability to keep going even when in pain etc. For me “cure” was never really an option the chances of being pain free were limited after surgery but after 4 there is nerve damage, mobility and strength loss etc. So cure wasn’t ever a word I’d use. I think of cure as cured of a disease or illness not skeletal/nerve damage caused by an accident. And cure would, yes, be total to pre-illness state.”</p>
Control	All respondents agreed or mostly agreed	<p>“Unsure if (???) and stronger meds were helpful or if the injury itself healed on its own with the help of exercise and stretches or a combination of all the above?”</p> <p>“When not in control of pain it is very severe pain, frustrating because of debilitation and inability to work – which is also stressful – the disability and inability to work and do preaccident activities, be painfree etc is what makes/is the cause of future concern – mostly the disability more than not feeling in control though they are loosely connected.”</p>
Emotions	All respondents agreed or mostly agreed, except one who was unsure	<p>Regarding negative attitudes experienced – “Very much so for me as I also felt that whiplash was claimed fraudulently. I don’t anymore.”</p> <p>“I think society needs to be better educated about whiplash injury and the healing process. Yes of course pain interfering with my life caused stress and frustration. Due to quality of life and lost opportunities such as missed work and not being able to spend time with my son.”</p> <p>Regarding understanding/emotions – “Same (respondent understood pain) its still just as frustrating maybe in a slightly different way.”</p> <p>Regarding negative attitudes – “ agree – people don’t understand what they can’t see and/or haven’t experienced even if they understand the theory”</p> <p>“Very difficult you feel helpless, hopeless can see people’s doubt in the way they look at you and respond to you – it is totally different than pre MVA when working etc. When people ask what you do and you say on disability they don’t see</p>

		one – you feel like a total loser and so much less of a person, having no value and no contribution to home, family or society.”
Mystery	All respondents agreed or mostly agreed, except one who disagreed	<p>“When a therapist or health care professional predicts that a treatment will be successful and the anticipated time for relief is met, it promotes trust in the caregiver. I found that this relieves stress”</p> <p>“My recoveries did not match expected recovery time or extent of damage (had multiple surgeries) however, I understand my pain and did understand risks prior to surgery thought you are always optimistic hoping for the best case scenario it doesn’t always happen – dealing with it is harder than understanding it by far. Understanding pain helps you to do proactive things to minimize it as best you can and to learn limitations – what aggravates it, what may alleviate it, even for a short period of time. It does not change how much pain you have or the injury, or the disability you are left with. It does not help you deal with the affects on your life or relationships/work – those issues are psychological not just a matter of understanding your pain”</p>
General comments	<p>“Information like how many other neck injuries I’ve had or other knee/ligament injuries or broken bones etc. Overall I healed very well but I’ll always have arthritis as a <u>direct</u> result of these injuries.”</p> <p>“It is so much more than understanding your pain, it is the effects the disability has on you physically, psychologically, family, friends your ability to do things you took for granted. Then there’s how you believe yourself, how society, friends, family, etc perceive you – It can be dark and resources are limited – psychologist \$160/hr rehab 6 visits free pay rest, gym membership fees - \$250/year exercise \$600/year etc etc and CAD pension and disability is limited. Mortgage etc bills etc are still due and you have ½ what you had working to pay so losing everything you worked for is an issue also.”</p>	

*Note, member checking also performed within interviews.

Negative case analysis

The table below represents the negative case analysis. The table is inclusive of all negative cases. The negative cases either represent contradictions to the themes or themes that were not extracted, but potentially important, due to a lack of enough reoccurrence within the data.

Meta-theme or theme	Negative cases
Restitution	<p>Though some terminology below suggests acceptance rather than restitution, strict adherence to the quest narrative, goes beyond acceptance and into acceptance in the context of embracing a new way of life (Frank, 1996). These ‘negative cases’ likely represent the fluidity of narrative (Frank, 1996).</p> <p>“I think that it. See I don't know if I would use the word curable? Because I think when it comes to an injury in any way, I don't know so much about being 100% pain free. I think you adapt to your pain at whatever level it is and when it's really bad you can adapt and fit it into your life but as you get better, you can be close to 100% and still adapt and feel like you are 100% of your person and not be 100% pain free. If that made any sense. You make concessions. I still make concessions for my ankle injury from years, and years, and years ago. I don't think about it, I just know that have to be careful if I go skiing or if I go do this or that. So you make adaptations to compensate for it. I thought in the beginning that it was going to be like a fleeting thing, like you feel it and then you recover, you get hurt and then you get better. I thought it was that black and white. Because I started to see it happening from my other accident. So I didn't that it would linger and linger. Curable, hum? I don't know about that one. Manageable.” – <i>Jennifer (3-months)</i></p> <p>“I guess what I wonder about further down the line, I wonder if it's always going to be like it is now where I'm optimistic and then I'm pessimistic, and I'm optimistic and I wish that it would completely just level off, where I knew where I had some concrete answer. Someone said to me, you are always, always going to have this pain, it's always going to be nagging you in the back of your ... it would almost feel like a burden being off my shoulders because then I would know. And not, be where I don't have pain, and I think oh great, I'm getting better and then be hit with it again. So I would rather know for sure then just kinda be, I won't say whine but because it's not really whine but there are days when I feel really great, things are good, I just need to keep up doing what I'm doing and I'll keep feeling good and then I don't. So some say yes, if you do this you're never gonna have any pain, or no matter what you do you're are always gonna have pain. I think it would take off some of the burden of the ... all the time”. – <i>Jennifer (6-months)</i></p> <p>Yep. I don't think there's anything that can entirely cure it, 'cause, honestly, it's kind of like scarring a muscle, the way I look at it, and a scar's always permanent. It's just a matter of getting to learn how to work with it. So, I don't think there's any medication that'll just take it away, like a cold or, you know, anything like that.” – <i>Charlene (3-months)</i></p>

	<p>“It’s just now that it’s more permanent and I’m more at ease with it, ‘cause I know it’s gonna be there. [pause] It’s kind of like you have to grieve, that grieving process after, and then it takes awhile and then you’re okay with the thought of it. It’s similar to that”. – <i>Charlene (3-months)</i></p> <p>“Yeah, I guess it would ... yeah because I thought I wouldn't feel anything and then now I realize that it's just going to be like this. Just have to take a little more time to soak in the tub, put something under my neck.” – <i>Stephanie (6-month)</i></p> <p>“Because I'm not. I'm not overly optimistic either. So I'm kind of neutral. I just kind of learnt to accept it. The aches and pains ... moments of back pain ... there, but I'll just have to manage it on my own. So I'm neither optimistic or pessimistic in that situation, I just am. I just that's just the way that it is life ... Yes. And the injury it is what it is, it will always be there. Everything that's been done that can be done has been done, you know, except for ... and like I said, if I could afford it, I would go to massage, to chiropractor, you know, I would go get extra services, if I was rich and famous one of those people, then I would, but I'm not. I have to take the Tylenol and if I have aches and pains. Does that make sense? ... Well it doesn't mean anything to me other than I just want to move on with my life. My husband and I are trying to have a baby, mother nature will decide I have obligations. Right now I'm just I have obligations I can't let pain get in my way I can't let pain get in my way, I have to on. I have to be the mother and I have to be the wife and I have the friend, I have life has to go on, so whether I have pain or not. So that's just how I deal with it.” – <i>Danielle (6-months)</i></p> <p>“I think if it's there it's going to be there. I don't think there's any particular activity or thing or medication that can make that particular movement, go away. If there was people would say don't do this or whatever. And it's not going to hurt there. If it's going to be there, it's going to be there, but I don't see where it's pessimistic in way ... if you still have pain. I've lived a whole life besides that accident and I've had seven car accidents, who's to say where it was from or what it's from, or maybe I just slept the wrong way or did whatever. (Another example) is . I don't kneel, I sit down or I kneel on the other knee or I use a lot of padding, but I'm not going to change my whole life because I can't kneel. I go to church, I go down on one knee instead of two knees or I'm going to sit on the ground and get myself back up again. I'm not going to kneel on that knee because I know it's going to hurt. But to say that I'm pessimistic, no, it's a fact, it's never going to be better because I have an incision right across the knee and nothing's going to change that.” – <i>Dana (6-months)</i></p> <p>“Just tolerable pain. Like I can tolerate a lot, I've got a high pain threshold I was told. And so for me to be hurting and not being able to move like this, its really high” – <i>Leanne (participant with chronic pain)</i></p> <p>“Even if I just have some kind of life where, of course, I would like</p>
--	---

	<p>to be able to go back to work full time, but to go back at all, doing something even volunteer on my own, would better than what I have. Because right now I don't really have much quality of life. So you don't feel like you are very productive at all. I'm not. And I think if I could get to where I could do something that would help. It changes your outlook too which also helps your mental well being and I think it does affect what do and what you can do physically too.” – <i>Dawn (participant with chronic pain)</i></p> <p>“I really think and they told me I wouldn't and now I actually believe that I won't (be painfree). That would be OK, if you could learn to deal with it in a different way than just being angry about it or depressed about it and we can get passed it enough that you can do normal things, then I would be OK with that. Because then it's not consuming your whole life. I mean, thank God, I'm only 47 now what am I going to be like in 10 years. It sucks, you know one day you are going home from work and you have everything and then you don't.” – <i>Dawn (participant with chronic pain)</i></p> <p>“Yeah, I would like to have everything 100% back but it's not going to happen and that took me a long time. Probably only in the past couple of months that I really truly accept and that isn't easy either. But you never know. Maybe I can do something else, it doesn't have to be what I was doing before.” – <i>Dawn (participant with chronic pain)</i></p>
Optimism – pain controllability	<p>“For some, optimism was associated with motivation to pursue treatment (or no optimism means given up): They may not heal or not heal as quickly. Um, if you don't, if you don't try or if you don't, um, if you don't give it a chance, it's, you know, nothing ventured, nothing gained.” – <i>Randall (3-months)</i></p>
Optimism – general belief in optimism	<p>“Some difficulty in distinguishing between hope and optimism: I'm hoping that it won't and I don't feel a reason why it would be. Like I say, if I could see exactly what happens in that type of that injury, what the dynamics are of it and what it does to your body then I would understand it a little bit more and then maybe make an accurate prediction of how it will affect me in the future, like if it is just simply, stay in physio and do these stretches and stuff for a year and you'll may not be able to start to see the results for a while. When I first started the treatment before it actually made me feel worse before it started making me feel better. So maybe it's just a longer curve for this where I'm going to feel worse for six months and then for the remaining six months I will start to feel better. I don't know yet. I'm hopeful that it won't be permanent but the path that it's been taking it almost feel like it's getting worse makes me think that there is a possibility of it.” – <i>Jennifer (3-months)</i></p> <p>“Yes. And the injury it is what it is, it will always be there. Everything that's been done that can be done has been done, you know, except for ... and like I said, if I could afford it, I would go to massage, to chiropractor, you know, I would go get extra services, if I was rich and famous one of those people, then I would, but I'm not. I have to take the Tylenol and if I have aches and pains. Does that make sense?” – <i>Danielle (6-months)</i></p> <p>“I haven't accepted it. I still think that hopefully somewhere down the line that they will find something that will help.” – <i>Candice (participant with chronic pain)</i></p>

	Pessimism when framed positively can be a good thing: “Someone said to me, you are always, always going to have this pain, it's always going to be nagging you in the back of your ... it would almost feel like a burden being off my shoulders because then I would know” - <i>Jennifer (6-months)</i>
Control – pain/injury severity	Intensity of pain in and of itself may not lead to a lack of control – what is more telling is whether a solution is available: “I definitely feel less overwhelmed and more positive even if I have bad pain, like if I have really bad sciatica or something. There's a treatment that I can do myself at home that it's very effective on it. So even when I have that extreme pain and it's very overwhelming it isn't because I know I can do something about it and I can do it myself and it's gonna work. So it's less daunting when I suppose you know there's an end because in the grand scheme I suppose, I don't really know when I'm going to be pain free.” – <i>Linda (participant with chronic pain)</i>
Control – understanding pain	Sometimes understanding does not lead to meaningful control: “My recoveries did not match expected recovery time or extent of damage (had multiple surgeries) however, I understand my pain and did understand risks prior to surgery thought you are always optimistic hoping for the best case scenario it doesn't always happen – dealing with it is harder than understanding it by far. Understanding pain helps you to do proactive things to minimize it as best you can and to learn limitations – what aggravates it, what may alleviate it, even for a short period of time. It does not change how much pain you have or the injury, or the disability you are left with. It does not help you deal with the affects on your life or relationships/work – those issues are psychological not just a matter of understanding your pain” – <i>anonymous from member check</i>
Control – control over daily demands	
Control – treatment success	Sometimes a practical solution to gaining control is not apparent: “Yeah medication half the time it doesn't even work, so I'm taking it for nothing and then I get addicted to it and then they have to wean me off and try something different. It's not worth the hassle.” – <i>Leanne (participant with chronic pain)</i>
Emotion – interference	Perhaps not so much interference from pain, but the interference from being involved in MVA (supportive of unique context of compensation injuries): “just dealing with insurance companies – oh, my God. [laughter] That's probably just the biggest hassle, getting a hold of them, getting all your paperwork done – that's just exhausting. Um, sending things in, going and getting a new vehicle, like, getting a rental vehicle, which is, it's more than just an injury. Actually, the injury's probably the easiest part of it all. [laughter] I can deal with it. But, um, everything just starting to get processed and, and better. The longest part is gonna just get, uh, my disability, like, getting covered for work that I missed, which, that'll take three months. So, it won't be over for another three months.” – <i>Cheryl (baseline)</i> Distinguishes between pain and emotional pain: You're like crying and upset and you're in pain, you're probably gonna feel more pain, 'cause you're having emotional pain at the same time, so, you know

	<p>[pause] it does kind of make sense. – <i>Kim (baseline)</i></p> <p>Emotions were not important for all: “I’m like that (emotional) by nature. I don’t get excited, or yell or get angry very often. I’m still able to come to work, I can still do my normal day to day activities. That’s probably why.” – <i>Randall (6-months)</i></p> <p>“I told them that there’s mind over matter. And I’d tell them that’s a false, that they are just saying that to whine and get attention. Unless they are depressed because depression can, they could be in a depression that might be something else that I would tell them because depression can lock up someone’s ... it can make them feel pain and stuff. But it wouldn’t be due to an accident. I think that’s another reason why doctor’s might not be paying 100% towards people that have pain that are internally and they can’t see it because they think they might be seeking attention or something like that.” – <i>Charlene (6-months)</i></p>
Emotion – loss	<p>Another possible theme, but not observed frequently enough: lack of empathy:</p> <p>“I am feeling that way because of the car accident, because of the injuries. Everything has cause and effect. That’s why I’m feeling, feeling depressed, because I can’t do things I normally would. I’m feeling depressed because I’m in pain. And feeling depressed would be, [unclear, 37:59, people don’t understand?]. [pause] They don’t understand the severity of the injury. They don’t understand anything about the injury, especially my boss, because they have never been there. So, I just have to, I just kind of expect it [pause] and go on and just” – <i>Danielle (baseline)</i></p>
Mystery - congruence	<p>“My recoveries did not match expected recovery time or extent of damage (had multiple surgeries) however, I understand my pain and did understand risks prior to surgery thought you are always optimistic hoping for the best case scenario it doesn’t always happen – dealing with it is harder than understanding it by far. Understanding pain helps you to do proactive things to minimize it as best you can and to learn limitations – what aggravates it, what may alleviate it, even for a short period of time. It does not change how much pain you have or the injury, or the disability you are left with. It does not help you deal with the affects on your life or relationships/work – those issues are psychological not just a matter of understanding your pain” – <i>anonymous from member check</i></p> <p>Perhaps mystery is also about others’ understanding (connected to stigma?): I think if, if, you know, certainly, if I had a broken leg or something, people would say, “Oh, I understand. You can’t do that. You have a broken leg.” But, when you say it’s your back or your neck, I think, in general – and I’m guilty of it myself – is like, “Oh, get over it.” Right? – <i>Jennifer (baseline)</i></p> <p>Attempts to understand may also be maladaptive: “But I don’t wanna get so focused and fixated on it that I [laughter] uh, can’t live my life.” – <i>Vanessa (baseline)</i></p>