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Injury-Related Fear in Individuals with Chronic Ankle Instability

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ABSTRACT

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About 40% of individuals who suffer a lateral ankle sprain develop chronic ankle instability (CAI). The mechanisms for developing CAI is believed to be multi-factorial, however, most literature has focused on the physical manifestations of the condition, leaving our understanding of psychological manifestations of the condition fairly unclear. Injury-related fear has been identified as a psychological factor that may be relevant to the condition, but our understanding is limited. Therefore, the purpose of this dissertation was to better understand injury-related fear in patients with CAI through three studies.

The first study systematically reviewed the literature to understand differences in injury-related fear between individuals with and without CAI. We found those who develop CAI report higher levels of injury-related fear compared to those who fully recover after their ankle sprain and to those without a history of one. Therefore, injury-related fear is a psychological factor that likely contributes to chronicity after ankle sprain.

The Fear-Avoidance Model (FAM) is a theoretical framework hypothesizing a relationship between injury-related fear and chronicity and disability, and so the second study used the FAM framework to understand the relationships between FAM components and function and disability in individuals with CAI. We found that beyond symptoms of instability and pain, greater pain catastrophizing and injury-related fear significantly predicted lower

function and greater disability. This relationship necessitates a better understanding of these fears so that reduction strategies can be used to enhance patient outcomes.

Therefore, the third study explored perceptions and experiences of injury-related fear in individuals with CAI. Our participants described injury experiences along susceptibility and severity contributed to the magnitude and generalizability of injury-related fears and subsequent activity behaviors. Patient goals, values, and attitudes toward their condition and physical activity likely contribute to the impact of these fears and the condition on quality of life. Therefore, beyond identifying injury-related fear after ankle injury, clinicians should engage in patient discussions to understand patients' injury-related fears, values, and goals to create individualized care plans that can best enhance their quality of life.

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NOMENCLATURE

AFAQ	Athlete Fear Avoidance Questionnaire
AII	Ankle Instability Instrument
AJFAT	Ankle Joint Functional Assessment Tool
ATF	Anterior Talofibular
CAI	Chronic Ankle Instability
CAIT	Cumberland Ankle Instability Tool
CF	Calcaneofibular
COP	Ankle Sprain Coper
CON	Healthy Control
CNS	Central Nervous System
CQR	Consensual Qualitative Research
FABQ	Fear-Avoidance Beliefs Questionnaire
FAM	Fear-Avoidance Model
FAAM-ADL	Foot and Ankle Ability Measure Activities of Daily Living Subscale
FAAM-Sport	Foot and Ankle Ability Measure Sport Subscale
FI	Functional Instability
HRQOL	Health-Related Quality of Life
IAC	International Ankle Consortium
IdFAI	Identification of Functional Ankle Instability
LAS	Lateral Ankle Sprain
mDPA	Modified Disablement in the Physically Active Scale

NASA	National Aeronautics and Space Administration
NFI	No Functional Instability
PCS	Pain Catastrophizing Scale
PRO	Patient-Reported Outcome
PTOA	Post-Traumatic Osteoarthritis
Quick-FAAM	Quick Foot and Ankle Ability Measure
RAS	Recurrent Ankle Sprains
SAS	Single Ankle Sprain
TSK	Tampa Scale of Kinesiophobia

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CHAPTER 1

INTRODUCTION

1.1 Background

Lateral ankle sprains are one of the most prevalent musculoskeletal injuries affecting both physically active and general populations.[1-3] Traditionally these injuries were thought to be minor, but evidence suggests that 40% of individuals continue to suffer from ongoing symptoms that persist greater than a year after their initial ankle sprain.[4] These symptoms include perceived ankle instability, episodes of ankle "giving way," and recurrent ankle sprains, and together these characterize a condition known as chronic ankle instability (CAI).[5] Beyond the symptoms that characterize the condition, individuals with CAI also present with unique combinations of associated impairments that span patho-mechanical, sensory-perceptual, and motor-behavioral domains.[6] Collectively, these impairments are thought to lead to long-term health-related consequences such as post-traumatic ankle osteoarthritis, function and activity limitations, and health-related quality of life (HRQOL) deficits. Therefore, developing evidence-based intervention strategies for preventing and treating CAI are important to mitigate these long-term sequalae. However, specific impairments important to CAI must first be identified in order to determine the best intervention strategies for targeting and improving these impairments.

Decades of research has been dedicated to identifying impairments associated with CAI.[6] This has typically been done by comparing individuals with CAI to individuals with no ankle sprain history, or by comparing to individuals deemed ankle sprain "copers". Copers are people who have sustained an ankle sprain but recover full function and do not suffer from ongoing symptoms.[7] Throughout this time a host of impairments have been identified to exist in the CAI population through the use of both clinician- and patient-based outcomes.[6] Studies using

clinician-based outcomes to identify physical impairments have dominated the literature and provide meaningful information for clinicians. However, patient-based outcomes provide information on how the injury is affecting the person across a wide variety of health factors and are equally important to identify and track.[8] Only recently have patient-based outcomes been used to identify impairments in those with CAI and provide understanding of patients' perceived impairments, function, and HRQOL.[9]

The first systematic review to summarize patient-based outcomes in CAI was published only six years ago.[9] This review revealed that three types of patient-reported outcomes (PROs) had been examined in those with CAI, including region-specific, generic, and dimension-specific outcomes. Collectively, they confirmed HRQOL deficits were present in those with CAI. Strong and moderate evidence was found supporting these deficits in region-specific and generic outcomes, respectively, but limited evidence was found regarding dimension-specific outcomes.[9] Dimension-specific outcomes are scales that assess one specific aspect of heath, and most commonly assess psychological constructs.[8] At the time, only two studies had used psychological questionnaires in the ankle sprain populations and both chose to examine injury-related fear.[10,11] Although this construct was studied in both investigations, one study compared injury-related fear outcomes between individuals with CAI and healthy controls and found those with CAI reported significantly higher levels of injury-related fear,[11] while the other study compared individuals with CAI and ankle sprain copers, and did not find differences.[10] These combined results suggest that injury-related fear is an impairment that may arise after an ankle sprain, but also that more research is needed to understand how ankle injuries affect this dimension of health in this population.[9]

Injury-related fear is thought to develop from memories of the pain and discomfort suffered after injury,[12] encompasses fear of movements that the individual feels may make them vulnerable to pain and (re)injury, and can also promote avoidance behaviors.[13] The fear-avoidance model (FAM) is a theoretical framework that has been used to examine how injury-related fear can develop after injury and postulates that exaggerated negative beliefs about pain, known as pain catastrophizing, can lead patients into a cycle of fear, activity avoidance, and chronic pain and disability.[13,14] CAI has been associated with functional deficits and disability for decades; however, recent reports also indicate a high prevalence of mild, persistent pain.[15] The role of persistent pain and its associated cognitive-affective outcomes has yet to be examined within CAI, and the FAM may serve as an appropriate framework to begin examining them and their relationship to reported disability.

The FAM (Figure 1) and its components have been applied and empirically supported across various musculoskeletal conditions.[16-18] This evidence suggests that injury-related fear could contribute to the development of chronicity following ankle sprain injuries, and if unaddressed, may be leading these individuals towards the long-term consequences noted to exist within this population. Encouragingly, there is some evidence that rehabilitation and multimodal interventions have the ability to improve reported fear beliefs in individuals with CAI.[19-21] However, the presence of these fears may also require more specific treatment approaches that focus on modification of fear beliefs and their subsequent behavioral consequences. Although dimension-specific PROs are helpful tools for identifying fear beliefs, a better understanding of the beliefs and/or factors that underlie the PRO scores would ultimately lead to the ability to address them within the intervention if necessary. Therefore, it is critical to

explore this construct beyond the numeric representation on a PRO to further our understanding within the CAI population.

Figure 1 Fear-avoidance model

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From: Vlaeyen J, Kole-Snijders AM, Boeren RG, Van Eek H. Fear of movement/(re) injury in chronic low back pain and its relation to behavioral performance. *Pain.* 1995;62(3):363-372.

1.2 Problem

Ankle sprains are highly prevalent injuries which often result in persisting symptoms and the development of CAI. These individuals present with short-term and long-term health-related consequences that affect their quality of life. Interventions to prevent and mitigate CAI and its consequences have mainly focused on the physical impairments that are identified in these individuals, negating the potential for other underlying mechanisms such as a heightened level of injury-related fear. Anecdotally, injury-related fear has been mentioned in CAI literature for years; however, the extent to which injury-related fear is present in individuals with CAI compared to those without the condition is still unclear, as is the best tools to assess differences. These fears have shown to be barriers to recovery and rehabilitation in other musculoskeletal conditions, and are theorized to lead to cycles of chronicity and disability similar to that which is

demonstrated in the CAI population, via the FAM. It is unknown if the components of the FAM would apply to the condition of CAI and its associated impairments. Lastly, little is known about injury-related fear in the CAI population beyond its reported existence. Without true exploration of the fear beliefs in these individuals, it remains difficult for clinicians to address them within rehabilitation interventions.

1.3 Purpose

There were multiple purposes to this dissertation in order to further understand injury-related fear in patients with CAI. The first purpose was to systematically review the literature investigating differences in injury-related fears between individuals with and without CAI. The second purpose was to determine if the FAM and its components can be applied to CAI. The third purpose was to explore the perceptions and experiences that underlie elevated levels of injury-related fear in individuals with CAI.

1.4 Aims and Hypotheses

Aim 1: To examine the extent to which injury-related fear is present in individuals with CAI

Hypothesis 1.1: Individuals with CAI will report greater levels of injury-related fear compared to those without CAI

Hypothesis 1.2: Ankle sprain copers and controls will share similar levels of injury-related fear

Aim 2.1: To examine relationships between injury-related fear and pain catastrophizing beliefs in those with CAI

Hypothesis 2.1: Greater pain catastrophizing beliefs will be related to greater levels of injury-related fear

Aim 2.2: To assess the influence of pain on ankle function and global disability in individuals with CAI

Hypothesis 2.2: Pain presence will explain additional variance beyond reported instability in both ankle function and global disability outcomes

Aim 2.3: To determine the unique role of the cognitive-affective model components in predicting function and disability

Hypothesis 2.3: When controlling for instability and pain, both pain catastrophizing and injury-related fear will uniquely explain additional variance in both function and disability

Aim 3: To explore the perceptions and experiences that influence injury-related fear measured with the Tampa Scale of Kinesiophobia (TSK-11), within the CAI population

1.5 Operational Definitions

1. Chronic Ankle Instability: A condition that classifies individuals at least a year after an initial ankle sprain injury who continue to report ongoing issues of recurrent sprains, and/or perceived instability, and/or episodes of "giving way."
2. Injury-Related Fear: An emotional response to the presence of danger or the threat of harm, in this case, specific to an injury. It will be used when referring to any sub-constructs that describe it including but not limited to kinesiophobia and fear-avoidance beliefs.
3. Kinesiophobia: A sub-construct of injury-related fear which describes fears regarding movements in which an individual feels vulnerable to pain and (re)injury.

1.6 Assumptions

The primary assumptions are as follows:

Chapter 3:

1. Information in published articles was accurate and free from error
2. Data extracted from published articles was accurate and free from error
3. PRO scores included in the review are an accurate and standardized assessment of injury-related fear in participants

Chapter 4:

1. Participants read and understood the questions on the survey
2. Participants accurately and honestly answered survey questions
3. PRO scores are an accurate and standardized assessment of injury-related fear, pain, and regional and global disability in participants

Chapter 5:

1. Participants provided honest answers to the questions during interviews

1.7 Delimitations

Chapter 3:

1. Only studies using the TSK, Fear-Avoidance Beliefs Questionnaire, and Athlete Fear Avoidance Questionnaire were included narrowing the scope of injury-related fear to two sub-constructs (kinesiophobia and fear avoidance)

Chapter 4:

1. Participants were adults between 18-40 years of age
2. Participants must have met the International Ankle Consortium guidelines for CAI
3. Outcomes of interest were delimited to only the PROs used in the study

Chapter 5:

1. Participants were adults between 18-40 years of age

2. Participants must have met the International Ankle Consortium guidelines for CAI
3. Participants must have completed the survey study
4. The semi-structured interview guide was based on the TSK-11, narrowing the scope of the study to the construct of kinesiophobia

1.8 Limitations

Chapter 3:

1. Our review is limited to available peer-reviewed literature and did not include gray literature
2. Confounding variables that could affect injury-related fear outcomes are not established in the CAI population and could thus not be accounted for
3. Criteria used for defining ankle sprain populations varies in the literature

Chapter 4:

1. Survey questions were not randomized
2. Some of the PROs used in this study have not been validated in their electronic format
3. Some of the PROs used in this study have not been validated in the CAI population

Chapter 5:

1. Although a semi-structured interview guide was used, different participants may have been asked different questions based on their responses to the TSK-11, and therefore, different follow-up questions as well

CHAPTER 2

REVIEW OF THE LITERATURE

2.1 Overview of Chronic Ankle Instability

Prevalence, Impact, and Long-Term Consequences of a Lateral Ankle Sprain

Out of 11.8 million physician office visits annually, 23% involve a sprain or strain injury to the ankle or foot.[22] Specifically, lateral ankle sprains (LASs) have the highest incidence when compared to other types of ankle sprains,[4] but may even be underestimated as approximately 55% of individuals do not seek treatment from a healthcare professional after sustaining a LAS.[23] Traditionally, LASs are often regarded as benign injuries that will resolve quickly without treatment; however, evidence suggests that this is not the case. It is estimated that 40% of individuals continue to suffer from ongoing symptoms that persist greater than a year after their initial ankle sprain.[4] These symptoms include perceived ankle instability, episodes of ankle "giving way," and recurrent ankle sprains, and characterize a condition known as chronic ankle instability (CAI).[5] Beyond these characterizing impairments, patients with CAI also suffer from a host of other impairments that can lead to long-term health-related consequences such as function and activity limitations, and health-related quality of life (HRQOL) deficits.[6] Additionally, acute LASs and CAI have also been associated with the development of ankle joint degeneration and are estimated to account for up to 80% of all cases of post-traumatic ankle osteoarthritis (PTOA).[24] Further troubling, is that these individuals are also thought to develop PTOA at an earlier age than those with idiopathic osteoarthritis, which would increase the years that these individuals would be affected by the condition and associated disability.[24] CAI is thought to mediate the progression of PTOA, however, what determines whether someone will

develop CAI after a LAS is still not fully understood. The most updated theoretical model is presented below.

Updated Model for the Development of CAI after LAS

A LAS injury occurs to the lateral ligament complex after excessive episodes of inversion, and internal rotation of the rearfoot in relation to the tibia.[25-27] Disruption or stretch of the lateral ligaments, most often the anterior talofibular (ATF) and in more severe cases the calcaneofibular (CF) ligaments,[1] occurs and clinical signs of injury such as pain, swelling, and inflammation develop quickly after. Patho-mechanical impairments including ankle joint laxity, arthrokinematic and osteokinematic restrictions, secondary tissue injury, and tissue adaptations are believed to be a direct result of the biological manifestations following the tissue injury.[6] It is theorized that, although hard to discern, sensorimotor function is also disrupted immediately following the injury due to inflammatory and pain mediators which results in sensory-perceptual and motor-behavioral impairments. Common sensory-perceptual impairments include decreased somatosensation, pain, perceived instability, injury-related fears, decreased self-reported function, and global HRQOL deficits. Motor-behavioral impairments include altered reflexive action of the fibularis longus, neuromuscular inhibition, muscle weakness, balance deficits, altered movement patterns, and decreased physical activity.[6] The clinical outcome after an ankle sprain is thought to range from full recovery (ankle sprain coper) to the development of CAI, and is decided by a combination of influencing factors.

A combination of emotional and psychological responses to the injury based on personal and environmental factors are thought to be influential to perceptions and behaviors after injury.[28,29] Physiological responses to injury can create local changes about the ankle joint that may lead to changes within the central nervous system (CNS), such as neuromuscular inhibition.

Hormonal responses can also increase stress hormones and potentially have influence over perception and movement. Changes in these areas after injury show evidence of alterations in afferent feedback systems, as well as efferent motor output, resulting in theories of reorganization within the CNS as an underlying mechanism to the development of CAI.[6,30]

Neural underpinnings have been associated with CAI for over six decades, but in the latest proposed model, CAI is viewed through three theoretical perspectives.[6] At the crux of these theories is that of the neuromatrix and neurosignature.[31,32] The neuromatrix is described as the neural networks found within the brain that process the afferent information and then create an output (deemed the neurosignature) that is related to movement output as well as perceptions and emotions.[31,32] Before injury, one's neurosignature is at a homeostasis. After injury, inflammation, stress, and tissue damage are thought to then disrupt the individual's neurosignature. The neurosignature relies on the cyclical relationship between the sensory-perceptual and motor-behavioral functions.[6,33] For example, after the ankle injury there is evidence of disruption of somatosensation due to damage to the proprioceptors about the ankle. The disruption of somatosensation creates a loss of afferent feedback, or aberrant feedback being sent to the CNS.[6,34] Additionally, there may be arthrokinematic restrictions or subsequent mechanical instabilities that may also provide changes to afferent activity.[6,35] These injury consequences create organismic constraints within the system. Based on the dynamic systems theory, because the body is inherently focused on movement, the system will re-organize to account for these organismic constraints and find new ways to accomplish the tasks at hand.[36-38] These reorganization strategies affect efferent activity at both spinal and supraspinal levels of motor control, changing reflexive activity and voluntary movements that then continue to send aberrant activity back into the system.[6,35] If the impairments that are creating change to the

afferent and efferent systems are not addressed, then these movement patterns and neural functioning become the new embedded neurosignature.[6] This neurosignature adaptation, or neuroplasticity component, is thought to explain the alterations in balance ability and movement alterations in those with CAI which may predispose the individual to recurrent episodes of the ankle giving way and subsequent ankle sprains.[6]

The example used above highlights a traditional set of impairments that have been widely accepted within the CAI literature (deafferentation and mechanical insufficiency about the ankle) as a means of changing the neurosignature of patients which may lead to CAI. These clinician-based outcomes, along with others, have dominated the literature and provide meaningful information for clinicians regarding the physical manifestations of CAI. However, the new model also posits that sensory and perceptual factors can change the neurosignature as well. The role of persistent pain and accompanying stress are specifically theorized to negatively impact the neurosignature and if left unaddressed, these factors could lead to further disability. Despite pain persistence being the trait for most other chronic musculoskeletal conditions, this particular impairment has received minimal focus within the CAI literature.[6] An overview of the literature pertaining to pain follows.

2.2 Pain and CAI

Following a LAS, the typical physiological response occurs which inherently includes the presence of ankle pain. This pain is generally localized to the lateral or anterolateral area of the ankle, consistent with the anatomical location of the ATF and CF ligaments. This acute pain, along with the immediate swelling post-injury, can often disrupt the patient's ability to weight-bear which affects function and activity levels, and can often be the reason why patients seek care after ankle injury. Early intervention strategies are typically focused on alleviating these

inflammatory symptoms[39] and evidence suggests that with conventional treatment, pain diminishes quickly in the first two weeks following the sprain.[40] However, not all individuals seek medical care following ankle injuries.[23,41] Additionally, majority of those who do see a healthcare provider may not be receiving the evidenced-based standard of care.[42] In sporting populations there is often a rapid return to functional activities,[39] with reports of patients returning to normal activity only 3 days after an initial ankle sprain, and one day following a recurrent sprain.[43] Although acute pain generally guides return to functional activities, there are individuals who still report pain and demonstrate other impairments after being cleared for return to activity.[40,44,45] The evidence suggests that patients may be returning to activity too soon after their injury, which may contribute to the residual and recurrent issues associated with CAI, including the persistence of pain.

Although pain persists beyond the acute stage in ankle sprain populations,[40,44] and it is typically a major symptom in other chronic conditions, it has not received a lot of focus within the CAI literature. Recognizing this, in 2019 Adal et al[15] conducted a systematic review of the literature to begin investigating the prevalence of pain among individuals with CAI. The results revealed that there was, in fact, a high prevalence of intermittent and mild pain in patients who were classified as having CAI.[15] The included studies used various methods of assessing pain which mostly consisted of subjective methods rather than validated questionnaires. So in 2020, Adal et al[46] conducted a retrospective analysis to investigate the prevalence of pain in individuals with CAI using information from a validated self-report questionnaire. The results revealed that 60.1% of the participants with CAI in previous studies reported ankle pain. Most participants were younger adults and reported pain during moderate or vigorous physical activity, although age was a predictor of pain during daily activities. Pain presence was also found to be associated

with increased levels of perceived ankle instability.[46] A recent cross-sectional study quantified pain using the SF-36 Pain subscale and found elevated levels of bodily pain in their CAI sample when compared to the healthy group, and found relationships to exist between reported pain and function.[47] Most studies of pain are retrospective in nature, making it hard to determine whether pain in these individuals was developed secondarily to CAI, or is a contributing factor to the condition. Nevertheless, the literature indicates that persistent or chronic pain may be an important and overlooked impairment that needs to be explored more within this population. The neuromatrix theory regarding chronic pain is described below, as well as how this new finding of persistent pain may help account for other impairments identified in the CAI population through application of this theory.

Neuromatrix Theory of Persistent Pain and Implications for CAI

The neuromatix theory posits that pain is not only a sensory experience occurring after injury, but is a multidimensional experience influenced by many factors.[31] Despite the complexity of pain, the function of pain is simplistic in that pain is an alert system triggered when the brain perceives that there is danger to the body and subsequent action is required. The subsequent action, or motor output, results in behaviors and movements that promote avoidance and lessen provocation of the injured area.[48] If pain is not addressed and persists, there is an increased sensitivity of the neuromatrix for pain-related input required to activate this neural network.[48] Therefore, this lessens the amount of input needed for the individual to perceive pain, which creates a self-sustaining cycle. This process also affects the output, which results in maladaptive motor responses.[31,48]

This theory has already been applied to CAI regarding other impairments but could certainly extend to the newly discovered impairment of persistent pain. The influence of pain on

other impairments associated with CAI is still generally unknown. Pain reported on validated questionnaires has shown to be related to greater degrees of perceived instability as well as functional limitation in activities of daily living and sport,[46,47]and has been found to explain some of the variance in spinal reflexive excitability and inhibition in patients with CAI.[49] Continued use of pain-related outcomes in CAI studies is warranted to further understand the characteristics and implications of this impairment.

It should also be noted that in addition to the persisting noxious signal, non-noxious mechanisms, such as cognitive-affective factors, are hypothesized to be contributing factors to the changes in the neuromatrix as well.[31,48] For example, an individual who appraises pain as highly threatening increases the overall sensitivity of the system by lowering the threshold for what is considered a threat to the body.[48] Additionally, these beliefs may alter the attention to pain and painful stimuli, which has shown to influence pain perception.[50] Emotional factors can also interact with pain, as negative emotions can lead to increased pain levels.[50] These cognitive and affective factors have shared neural pathways which is believed to explain their modulatory role in pain perception.[32,50] The reverse is also true in that pain seems to have an effect on cognitive and emotional function as well. In chronic pain patients, for example, cognitive and emotional changes and impairments have been noted to exist alongside pain presence.[50] Therefore, persisting or recurring pain may be detrimental by diminishing the body's ability to inhibit noxious signals through changes in modulatory pathways which can lead to maladaptive output and sustained pain, while also contributing to dysfunction within these cognitive and emotional domains. Recent studies have identified cognitive and emotion-related changes in the CAI population which will be discussed in the following section.

2.3 Cognitive-Affective Outcomes and CAI

Despite rehabilitation efforts, ankle sprain injuries continue to have high recurrence rates as patients return to their activities of daily life and sport. Often these environments can be more complex than that of a controlled clinical setting. Complex environments provide an abundance of changing and potentially conflicting information to the central nervous system and warrant attentional and cognitive processing demands. The theory of modularity suggests that there are shared neural processes that are available for both cognitive and motor systems allowing for both attentional demands and desired motor actions to be executed.[51,52] However, if two tasks that rely on the same neural processing areas are both summoned simultaneously, this is thought to create interference and performance can become degraded.[52] Individuals with CAI often display motor performance deficits which has yielded investigations into the role of cognitive function within these individuals.

A recent systematic review looked into studies of cognitive loading on motor performance in those with musculoskeletal conditions including CAI.[53] The evidence from dual-task design studies suggest those who experience ankle instability may have less automaticity and greater attentional requirements in the maintenance of balance during more challenging tasks,[54] and in more dynamic tasks, such as gait.[55,56] Therefore, it is believed that in normal conditions and movements such as walking, those with CAI have to afford a greater proportion of available neural resources to the activity. As a cognitive task is added this may create competition for resources, and therefore explains changes to biomechanical and gait patterns, which may predispose these individuals to continuous instability and subsequent injury and disablement.[53]

This greater attentional demand may be attributed to the neurocognitive functioning in these individuals.[57,58] A recent study indicates patients with CAI demonstrated lower composite memory, visual memory, and simple attention on computer-based neurocognitive testing compared to controls.[57] Poor attentional regulation and control on computer-based testing has also shown to be associated with poorer postural control in individuals with CAI, which was not present in healthy controls.[58] Another recent study investigated cognitive function with a dual-task design and found that those with CAI demonstrated slower reaction times during the cognitive task in static balance conditions with eyes open and eyes closed, despite only seeing apparent balance deficits in the eyes closed condition.[59] Together, the results suggest that CAI may affect cognitive processes and control of attention, and that these changes may be contributing to functional deficits within this population. It is unknown as to the mechanism for these changes; however, it is possible that pain and pain-related factors may play a modulatory role in some individuals.

As previously mentioned, pain is an alert system to the body. Its function is to interrupt and demand attention.[60] Chronic or persisting pain is believed to consume a portion of the available attentional resources of the brain, therefore, potentially contributing to impaired task execution.[60,61] Individuals with CAI have a high prevalence of pain, but the intensity of that pain seems to be lower,[15] which may not alone contribute to changes in attentional demand systems as it is believed that prioritizing pain over the goals of task execution is influenced by high pain intensity.[60] However, pain-related beliefs may influence pain processes and the interruptive effect of painful stimuli.[60,61] Individuals who have negative cognitive appraisals or coping toward pain, such as pain catastrophizing, often assign a higher threat value to pain. When pain is given greater value of threat, fears may emerge regarding pain and (re)injury and together,

these pain-related beliefs are thought to impact the disturbing effect of pain on cognitive tasks, which could interfere with attentional demands and result in impaired cognitive function.[61,62] These mechanisms are merely speculative as these relationships have yet to be investigated in ankle sprain populations. Additionally, pain catastrophizing has not been the focus of any known study in CAI, although has been shown to exist in college athletes who had experienced previous injury, which warrants further investigation.[63] Injury-related fear, however, has been noted in this population, and may serve as another potential modulating influence to pain, neurocognitive function, functional deficits, and disability.

Injury-Related Fear and CAI

It is well-known that after injury, athletes report increases in depression, anger, tension, and anxiety along with decreases in self-esteem and vigor.[64] Over the course of rehabilitation and recovery, cognitions and affect are shown to change to be more positive over time.[65] However, returning to sport or full activity can induce fears or anxieties regarding re-injury.[66] As previously mentioned, patients who sustain a LAS may return to full activity before deficits are resolved.[44,45] One study found these patients who had returned to high activity levels still reported high levels of perceived instability similar to that of what is used to determine CAI.[45] Lack of confidence in the injured part, incomplete recovery, and returning to activity too soon are all thought to promote the development of fear and anxieties related to re-injury.[66]

Injury-related fear is thought to be brought about by memories of the pain and discomfort suffered after an injury, and may also be associated with multiple movements or activities that could cause re-injury.[12] Injury-related fear is a psychological construct that includes but is not limited to kinesiophobia, fear-avoidance beliefs, and reinjury anxiety.[11] Kinesiophobia describes fear of movements that the individual feels may make them vulnerable to pain and (re)injury.[67]

These fear beliefs can promote avoidant behaviors which is described as fear-avoidance beliefs.[14] Although often used interchangeably, fear and anxiety constructs differ, and some believe that re-injury anxiety is a more appropriate term as it describes anticipation and uncertainty relating to (re)injury.[68] Fears regarding re-injury have been anecdotally noted in the CAI literature for years and are attributed to physical activity restrictions[69] and other avoidance behaviors, as well as stiffening strategies related to balance tasks.[70] Injury-related fear was not measured in individuals with CAI until more recently.[10,11] To date there have been few studies that have directly investigated injury-related fear in individuals with CAI, and these studies are limited to investigations of kinesiophobia and fear-avoidance beliefs using the Tampa Scale of Kinesiophobia (TSK),[67] Fear-Avoidance Beliefs Questionnaire (FABQ)[14] and Athlete Fear Avoidance Questionnaire (AFAQ)[71] self-report instruments.

Wikstrom[10] was the first to investigate kinesiophobia in individuals with CAI and did not find any significant differences between individuals with CAI and those who were considered ankle sprain copers. Because both of these populations had experienced an ankle sprain, Houston et al[11] expanded upon these investigations by examining differences in both kinesiophobia and fear-avoidance in those with CAI compared to those without a history of ankle sprain. It was found that those with CAI demonstrated elevated levels of injury-related fear with large magnitudes of difference between groups measured by TSK and FABQ instruments. More recent studies have expanded upon these by investigating these constructs in collegiate athletes. These investigations found that college athletes with recurrent ankle sprains[72] and those with perceived instability (determined by Identification of Functional Ankle Instability)[73] report higher levels of injury-related fear when compared to athletes with a single ankle sprain or no perceived instability, respectively, and those without an ankle sprain history. Recurrent sprains

and perceived instability are two of the classifying characteristics in those with CAI which supports the notion that this impairment may be important to the development or continuance of the condition.

Ankle joint laxity may also play a role in influencing injury-related fear. Hadadi et al[74] recently found that individuals with both mechanical and functional instability reported the greatest levels of injury-related fears compared to those only reporting functional instability. Additionally, Houston et al[75] found that ankle laxity, along with force plate balance performance, was predictive of injury-related fear in those with CAI. Although both of these studies included participants of both sexes, the majority were female participants which may explain why one study found associations in injury-related fear and ankle joint laxity in only the female participants with a history of ankle sprain.[73] Although limited, these studies suggest a relationship may exist between injury-related fear and other common impairments in those with CAI. More work is needed to fully understand what factors contribute to greater fear levels in ankle sprain populations and whether differences exist between those who go on to develop CAI and those who do not.

In other musculoskeletal conditions, injury-related fear has shown associations to negative outcomes after injury regarding physical impairments, recovery, and function.[17,76] These associations have not been fully investigated in ankle sprain populations despite strong evidence of functional deficit and disability in those with CAI.

2.4 Function and Disability in CAI

The key characteristics describing CAI are related to functional impairments about the ankle.[5] In order to determine whether an individual has CAI, it is required to obtain this information directly from the patient. Actual episodes of instability, such as number of sprains

and “giving way,” are easily collected from patients; however, it is recommended that discriminative patient-reported outcomes (PROs) are also used to capture a patient’s perceived level of instability to identify this key impairment..[5] The International Ankle Consortium (IAC) has endorsed three validated PROs to confirm ankle instability: Ankle Instability Instrument, Cumberland Ankle Instability Tool, and Identification of Functional Ankle Instability.[5] These PROs include questions regarding ankle sprain history as well as the characteristics associated with the perceived instability episodes and have been used widely in ankle sprain research. Cut-off values have been determined for these instruments to identify this impairment, and thus, the condition of CAI.

Beyond the classifying functional impairment of instability, these individuals also report decreased ability to perform functional tasks due to their ankle. Evaluative, region-specific PROs, are commonly reported to assess participants’ level of disability and have overwhelmingly been shown to demonstrate deficits in those with CAI compared to those without.[9] One of the most used PROs assessing regional disability is the Foot and Ankle Ability Measure (FAAM), which is also endorsed by the IAC for use in the CAI population. It is comprised of two subscales with questions pertaining to activities of daily living (FAAM-ADL) and sport (FAAM-Sport) related function. In 2016, Hoch et al[77] developed a shortened version of the FAAM (Quick-FAAM) for specific use in patients with CAI to decrease administration and scoring time. The Quick-FAAM retained five items from the FAAM-ADL and seven items from the FAAM-Sport. It has demonstrated strong internal consistency ($\alpha = 0.94$),[77] and acceptable test-retest reliability,[78] and recently was found to be able to distinguish between individuals with CAI and copers.[79]

Region-specific PROs like the FAAM and Quick-FAAM are designed to assess patients' perceptions of how their injury or condition affects function directly related to the region of interest, the foot and ankle in this case. This makes these PROs ideal for assessing effectiveness of interventions to the ankle, or ankle disability overtime, since they tend to be more responsive to ankle-related changes. However, these PROs are unlikely to capture or detect changes in other domains of HRQOL and disability. Therefore, the combined use of specific and generic PROs is recommended to detect a broader picture of disability. Generic PROs, as the name implies, are designed to capture an individual's perception of how injury influences their overall health and well-being. These PROs have not been used as commonly in the CAI literature; however, evidence for the existence of global physical impairments continues to grow, indicating that a global measure of disability is warranted for this population as well. In a 2015 systematic review, there was moderate evidence to suggest global health deficits in individuals with CAI compared to those without.[9] This was supported in two other recent studies using the SF-36, PROMIS, and the Modified Disablement in the Physically Active Scales (mDPA).[47,80] One notable finding is that the mental health subscales of the SF-36 and PROMIS have not detected differences between young or middle-aged individuals with and without CAI.[9,47,75,81] However, the mental health subscale of the mDPA did detect significant differences in young CAI participants,[80] which suggests the mDPA has an advantage over the other generic measures. Despite significant findings, the effect sizes for the mental health subscale were small, which may suggest that generic mental health concerns do not present in those with CAI differently than in healthy control subjects.

As mentioned previously, however, one psychological dimension that has shown differences in ankle sprain populations is fears regarding re-injury. The fear-avoidance model

(FAM), is a cognitive-behavioral model that has been used to examine how injury-related fear can develop after injury, and the influence of cognitions and emotions on chronic pain and disability.[13,82] As individuals with CAI likely demonstrate elevated levels of injury-related fear, and some also report persistent pain and disability, the FAM may also serve as a theoretical framework for ankle sprain populations, and thus, will be described in the following section.

2.5 The Fear-Avoidance Model

The FAM is a biopsychosocial approach to understanding how after injury or a painful experience, some individuals get stuck in a cycle of chronic pain and disability, while others are able to recover without functional limitation. It is based on the idea that people who develop and maintain chronic pain do so because of cognitive-affective factors that lead to avoidant behaviors, disuse, and disability. Since its formation, the FAM and components of the FAM have been supported across a variety of chronic or musculoskeletal conditions including low back pain,[14,82] neck and shoulder pain,[83] chronic pelvic pain,[84] whiplash disorders,[85] patellofemoral pain,[86] and anterior cruciate ligament reconstruction.[87]

Waddell's theoretical model of Fear-Avoidance[14] was created to postulate causality between low back pain and disability with the addition of cognitive, affective, and behavioral factors linking these concepts together. This was formulated based on his research which found weak relationships between pain and disability in individuals with low back pain and that fear-avoidance beliefs had stronger relationships with reported disability than did pain.[14] Further, he found little direct relationships between pain and pathological severity, and fear-avoidance beliefs, which together suggests that these fear beliefs are another dimension of impairment and that these beliefs may affect behavior more than the physical impairment itself.[14]

It was well accepted that after an acute low back injury, the physical impairment from the injury can directly cause disablement. However, when low back pain becomes a chronic issue, often times there is no evidence of structural damage, but pain and impairment can still exist. The changes from the initial injury may spur subsequent changes such as muscle guarding, disuse behavior, and aberrant and compensatory movements that create new pathological pain and physiological impairments for these individuals. This "new" pain creates a sensory experience that will lead to cognitive, affective, and behavioral consequences, which then feedback into the system and allow it to be self-sustaining. Waddell's work[14] supports the notion that one of the major cognitive pathways between pain and disability in chronic low back pain is the presence of fear-avoidance beliefs. These beliefs are fears relating to painful experiences which can subsequently lead to avoidant behaviors that are maladaptive and may lead to chronicity. Waddell created the FABQ[14] to capture these beliefs and found that they were able to explain a large proportion of disability in those with chronic low back pain, and thus, are important for identification and consideration in rehabilitation for low back pain.

This framework has been modeled and modified to identify other key features that may lead to chronicity. Vlaeyen et al,[13] created a cognitive-behavioral model that encompassed fear of movement/(re)injury as a separate component and precursor to avoidance. This model postulates that after an injury there is a painful experience that occurs. Cognitive-perceptual response to the injury can affect how pain is tolerated and the subsequent sympathetic reactions from the pain and injury. One identified appraisal often cited in chronic pain patients is "catastrophizing" which is characterized by exaggerations of their situation and attentional focus on the negative aspects. When patients catastrophize, this can create greater stress, exacerbate painful perceptions, and provoke kinesiophobia, which is fear related to movements that may

make the person vulnerable to pain or reinjury. According to Vlaeyen's model, these fears can result in avoidance and subsequent behavioral strategies including disuse, disability, and depression. These subsequently create new painful experiences or rumination of past painful experience that become self-sustaining, leading to chronicity. On the other side of the model are individuals who do not catastrophize after injury. Because they do not increase the threat value of pain and injury, they tend to not have fears regarding pain and movement and are able to confront the injury and perform movements and activities that allow for proper healing and full recovery.[13]

Relationships between components of the FAM have been found to exist across multiple conditions, but using structural equation modeling, Cook et al[18] found that catastrophizing and pain predict levels of fear, while both catastrophizing and fear predict increased pain and disability. This begins to provide predictive utility of the FAM components in chronic pain patients. These components will be discussed further below.

2.6 Components of the FAM

Pain Catastrophizing

Pain catastrophizing is described as a negative or maladaptive cognitive-affective response to actual or anticipated pain. Individuals who adopt this belief system tend to magnify the threat value of pain, feel helpless regarding pain, and ruminate about their pain.[88] There are multiple tools that have been used to assess pain catastrophizing, but the most common among these is the Coping Strategies Questionnaire (CSQ)[89] and the Pan Catastrophizing Scale (PCS).[88] The CSQ only measures the helplessness domain, whereas, the PCS measures three domains (rumination, magnification, and helplessness) and thus is believed to be a more robust measure of the construct.[90]

There are multiple theories related to the construct of pain catastrophizing which include appraisal, attention bias, communal coping, as well as CNS/neural mechanisms.[90] Collectively, it is thought that when patients catastrophize, this can create greater stress to the system and can in turn exacerbate painful perceptions.[90] As described in the neuromatrix theory, pain catastrophizing may increase the sensitivity of the neural networks related to pain processing and therefore, activate with lower levels of pain.[48] This is one of the believed mechanisms to support its association with the development and continuance of persistent pain,[90] and has support via neuroimaging studies.[91]

Catastrophizing has consistently demonstrated associations to negative pain-related outcomes across both healthy and diverse patient populations.[92] There is some variance to the degree of associations to pain severity, however it has accounted for up to 31% in some samples.[92] It has been uniquely predictive of pain intensity, chronic pain, and disability in prospective analyses,[93,94] as well as to global disability ratings in individuals with knee pain.[95] It is often found to be related to fear of pain and (re)injury, negative affect, chronic pain, and disability which support its connection to other FAM components.[18,90,92,93,95] Despite its demonstrated inter-relatedness with fear outcomes, it is believed to be an important construct to the pain experience.[90,94]

Fear

Fear is an emotional response to danger, and is believed to be a learned response from pain.[96] Although direct learning is often the focus of fear in pathological participants, fear regarding pain and/or (re)injury can also be learned via observation and verbal instruction.[96,97] It is further hypothesized that catastrophic cognitions regarding pain can promote fear as the threat value of pain is believed to be increased. Fear, in turn, is believed to promote avoidant behavior

in order to mitigate the threat, which is the basis of the FAM.[13,96-98] Different pain-related fear constructs exist in the literature, giving rise to multiple assessment tools. The most common fear construct studied in ankle sprain literature is kinesiophobia. Kinesiophobia describes fearing movements in which an individual feels may make them vulnerable to injury or reinjury.[67]

In sporting populations, injury-related fear has emerged as an important factor in patients who have had an anterior cruciate ligament reconstruction. It has shown to be related to reinjury risk,[99] functional and movement-related outcomes,[99,100] and was highlighted as a main factor of whether a patient returned to their previous sporting activity.[101] Additionally, a recent report found that kinesiophobia, as measured by the TSK, was a more significant predictor than functional outcomes regarding whether a patient returned to their previous sporting activity.[102] Across numerous other musculoskeletal conditions there is supporting evidence that kinesiophobia is related to greater levels of pain intensity and severity, disability, and HRQOL deficits.[16] There is also moderate evidence to suggest that greater levels of kinesiophobia are predictive of the progression of disability overtime.[16]

Overall, the cognitive-affective components of the FAM have emerged as important factors related to pain, function, disability, and chronicity. As such, the framework and components of the FAM may also serve as a theoretical model useful for the condition of CAI. Exploration of these factors may provide further insight into the development and continuance of chronicity in ankle sprain populations.

2.7 Application of the FAM to CAI

Although there are multiple theories already being applied to the condition of CAI, they have mainly been centered around the physical manifestations of the condition. As more psychological and perceptual constructs surface in the literature, the use of theoretical models

specific to these constructs may be helpful to understand their relationship to chronicity after ankle sprain.

One year following an acute ankle sprain, individuals are thought to fall along a clinical spectrum of outcomes with CAI on the negative end, and full recovery or ankle sprain "coper" on the positive end.[6] After the acute ankle sprain, all individuals have the typical inflammatory response which includes a painful stimuli and experience. Individuals known as ankle sprain copers are thought to fully recover after the injury, and this is demonstrated by no or low levels of perceived instability, high self-reported functioning, no reported pain at rest or during activity.[6,7] Overall, these individuals present similar to individuals who have never had an ankle sprain.

Individuals who go on to develop CAI are known to present with movement pattern alterations, decreased perceived levels of ankle function, increased levels of global disability, and physical activity restrictions.[69] In addition, there is recent evidence of a high prevalence mild, but continued levels of pain.[15] Although pain catastrophizing has yet to be studied in ankle sprain populations, catastrophizing has recently been shown to be higher in athletes with current or previous injury,[63] and has shown, with pain to be predictive of fear.[18] As the FAM suggests it is possible that changes in movement and activity behaviors help to create new pathological pain pathways beyond the healing of the originally injured tissue, that continues these individuals down the path of chronicity. Additionally, Wikstrom et al,[103] found that increased injury-related fear and lower physical activity levels were associated with ankle joint degeneration in those with CAI, suggesting these continued behaviors could lead to early onset post-traumatic ankle osteoarthritis. Beginning to examine relationships between injury-related fear and other components of the FAM may be useful in continuing to develop an understanding of the

development of CAI and provide a theoretical framework for its development in some individuals.

CHAPTER 3

INJURY-RELATED FEAR IN INDIVIDUALS WITH AND WITHOUT CHRONIC ANKLE INSTABILITY: A SYSTEMATIC REVIEW

3.1 Introduction

Ankle sprains are highly prevalent musculoskeletal conditions in both athletic and general populations.[1-3] Although some individuals, known as ankle sprain copers, do not experience residual issues, approximately 40% of individuals who suffer an ankle sprain go on to develop chronic ankle instability (CAI)[4] which is characterized by recurrent sprains, perceived instability, and "giving way" episodes.[5] Beyond these characterizing symptoms, CAI is associated with long-term health-related consequences such as post-traumatic ankle osteoarthritis[24] and deficits in health-related quality of life.[9] Therefore, developing evidence-based intervention strategies for preventing and treating CAI is important to mitigate these long-term sequalae. Successful evidence-based interventions, however, are informed by identifying the specific factors that will need to be targeted and measured within intervention protocols. Although years of research has been dedicated to identifying impairments associated with CAI,[6] physical impairments via clinician-reported outcomes has been the major focus in the literature. It was not until more recently that patient-reported outcomes (PROs) have been used to provide information on how the injury is affecting the person across a wide variety of health factors.[9]

These investigations have added to the clinician-reported evidence supporting that CAI is associated with deficits in physical function and increased disability,[9,11] and have also identified another area regarding the potential psychological changes that may be associated with the condition. Specifically identified is that injury-related fear may arise after an ankle sprain injury.[9] Injury-related fear has been identified in patients across other musculoskeletal

conditions,[86,104,105] and has also demonstrated importance to rehabilitation and recovery.[106] It has also shown associations with individuals who suffer re-injury[99] and/or chronicity[14,16] and, therefore, may be an important area to target within rehabilitation protocols after ankle sprain.

Since the identification of injury-related fear in ankle sprain populations,[11] studies have included various PROs to attempt to capture this construct within their participants. The most commonly used PROs for assessing injury-related fear are the Tampa Scale of Kinesiophobia (TSK) and the Fear-Avoidance Beliefs Questionnaire (FABQ) which both capture elements of injury-related fear but differ in the sub-constructs in which they measure. The TSK measures kinesiophobia, which encompasses fears regarding movements in which the individual feels may make them vulnerable to pain and (re)injury,[67] while the FABQ measures fear beliefs and consequent avoidance regarding physical activity and work.[14] As these PROs were created for more general populations, the Athlete Fear Avoidance Questionnaire (AFAQ) was recently created to measure sport-specific fear avoidance in athletic populations.[71]

Despite the increased use of these PROs, the use of various instruments makes it difficult for clinicians to know which tool may be best to identify injury-related fear in their ankle sprain patients. Additionally, there remains limited understanding about the effect of CAI and injury history on this dimension of health.[9] Identifying differences in injury-related fear between those who physically recover after their ankle sprain injury and those who continue to suffer from ongoing symptoms and disability may provide insight as to the PRO and associated sub-construct most related to chronicity after ankle sprain. This knowledge could then be used to inform the most appropriate rehabilitation strategies to target this factor within rehabilitation protocols. Therefore, the purpose of this study was to conduct a comprehensive systematic review investigating differences in injury-related fear between individuals with CAI, ankle sprain

copers, and healthy controls. Based on literature supporting the associations between injury-related fear and chronicity, we hypothesize that individuals with CAI will report greater levels of injury-related fear when compared to both copers and controls. Further, we believe that copers and controls will share similar levels of injury-related fear.

3.2 Methods

Search Strategy

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement was used to guide this review. We conducted a literature search through CINAHL Plus with full text, PubMed, and SPORTDiscus with full text in November 2020 using the following terms: (ankle instability OR ankle sprain) AND (fear avoidance OR kinesiophobia). The latter terms were searched using the *all text* option as some studies used them as secondary or demographic measures only.

Selection Criteria

The search results were initially reduced by removing duplicate articles. Two independent reviewers (AS, RM) familiar with the ankle instability literature further eliminated studies by screening titles and abstracts using the inclusion and exclusion criteria. All remaining studies underwent full text review by both investigators to determine eligibility.

We included peer-reviewed articles that reported injury-related fear using the TSK, AFAQ, or FABQ as either a primary outcome or a patient descriptor in individuals with CAI, copers (COP), or controls (CON). Participants included in the CAI groups met criteria of the International Ankle Consortium guidelines (IAC),[5] or demonstrated key characteristic of CAI such as recurrent ankle sprains, perceived instability, or decreased levels of function determined by a region-specific PRO. Participants in the COP group included individuals who reported a

history of ankle sprain with no known residual symptoms. Participants were included in the CON group if they reported no history of an ankle sprain. We excluded studies that did not meet inclusionary criteria, did not compare TSK, AFAQ, or FABQ outcomes between populations, or contained duplicate data from a previously published article.

Quality Assessment

To assess methodological quality of the included studies, we used a 14-item version of the Downs and Black scale described by Moisan et al,[107] which has shown high internal consistency and interrater reliability.[108] Based on the percentage of items met, the studies were classified by quality: low (<60%), moderate (60-74.9%) or high (≥75%).[109] We independently assessed the articles (AS, RM) and any disagreement was discussed until consensus was reached.

Data Extraction

Once articles were deemed eligible, they were grouped by the between-group comparisons reported in each study. Comparisons included injury-related fear outcomes between CAI and CON, CAI and COP, and COP and CON. Study design, group criteria, and patient demographics were extracted to provide characteristics of the included studies. Sample sizes, total mean scores, and standard deviations of injury-related fear outcomes were extracted from all studies.

Statistical Analysis

Hedges *g* effect sizes (ES) and 95% confidence intervals (CIs) were calculated to examine differences between the groups. For both CAI comparisons, a positive effect was indicative of higher levels of injury-related fear in the CAI population. For the COP and CON analysis, positive effect was indicative of higher levels of injury-related fear in the COP group. ES were interpreted as weak (≤0.40), moderate (0.41–0.69), or strong (≥0.70).[110] Interpretations

of numerical findings were based on the strength of the ES and the position of the 95% CIs in relation to zero. Kappa coefficients measured inter-rater reliability of the quality assessment [none (≤0.20), minimal (0.21–0.39),weak (0.40–0.59), moderate (0.60–0.79), strong (0.80–0.90), and almost perfect (≥0.90)].[111]

Level of Evidence

We determined level of evidence for each group comparison by using the 5-level rating guidelines previously adapted from the Cochrane Collaboration Back Review Group.[9,112] Analyses in which findings were consistent in multiple high-quality studies were rated as *strong evidence*. Consistent findings in multiple moderate- or low-quality studies were rated as *moderate evidence*. Evidence from one moderate- or low-quality study was rated as *limited evidence*. *Conflicting evidence* was the rating if inconsistent findings were demonstrated from multiple studies, and *no evidence* if no studies had been done.

3.3 Results

Search Strategy

The initial search yielded 69 results, and ultimately resulted in 11 included studies reporting comparison data for injury-related fear outcomes (Figure 3.1). Injury-related fear instruments reported included the AFAQ, FABQ, and the 11-item and 17-item version of the TSK. Two included studies reported medians and interquartile ranges,[11,72] one study only reported the subscale data of the FABQ,[47] another reported outcomes separated by sex,[73] and two studies had broken the CAI group into subgroups,[74,113] so the authors were contacted to extract total mean scores and standard deviations needed for appropriate group comparisons. One author who presented their data in CAI subgroups could not be reached,[74] so only the group denoted as

those with functional instability (FI) was used for the CAI group as they fulfilled the guidelines set forth by the IAC guidelines.[5] Study characteristics are presented in Table 3.1.

Figure 3.1 Literature search flow chart

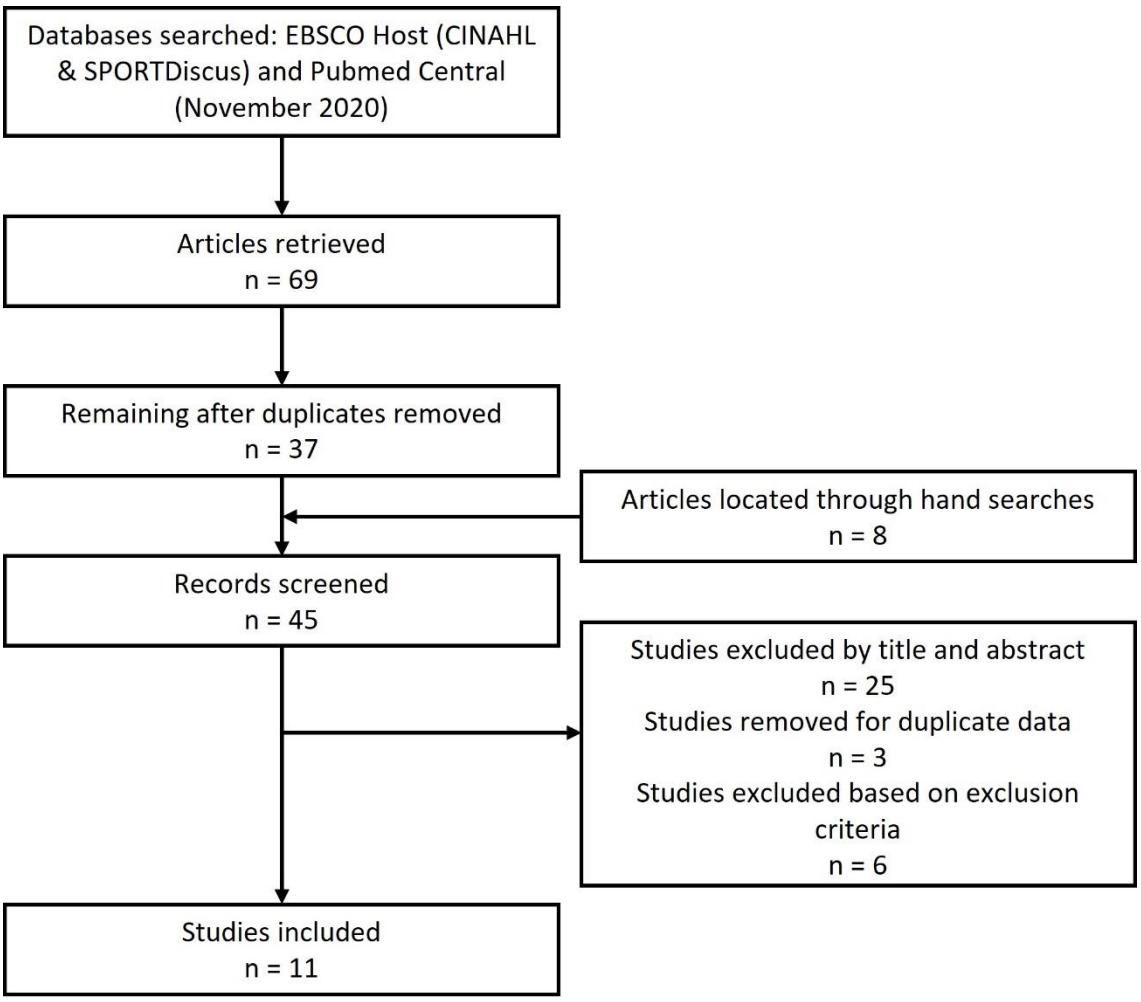


Table 3.1 Methodological summary of the included studies

Study	Study Design	Group Criteria	Population	Subject Characteristics	PRO
DeJong et al,[114] 2019	Case Control	*CAI*: at least one significant LAS ≥ 12 months prior to enrollment, no current ankle injuries ≤3 months, <90% on the FAAM-ADL and <85% on the FAAM-Sport, ≥11 on the IdFAI, and <24 on the CAIT *CON*: no history of ankle sprain	Recreationally Active (≥30 min of moderate activity 5x/week)	20 CAI (F:10, M:10; 21.7 ± 2.32 yrs; 172.74 ± 11.28 cm; 74.26 ± 15.24 kg) 20 CON (F:10, M:10; 21.2 ± 2.79 yrs; 173.18 ± 15.16 cm; 70.89 ± 12.18 kg)	TSK-17 & FABQ
DeJong et al,[115] 2020	Case Control	*CAI*: at least one significant LAS ≥ 12 months prior to enrollment with residual disability classified as ≤90 on the FAAM-ADL, and ≤85 on the FAAM-Sport *COP*: at least one significant LAS ≥ 12 months prior to enrollment with no residual disability classified as ≥99 on the FAAM-ADL, and ≥97 on the FAAM-Sport	Recreationally Active (≥1.5 hr activity/week)	14 CAI (F:14; 21 ± 3 yrs; 168.4 ± 8.4 cm; 68.9 ± 14.4 kg) 14 COP (F:14; 21 ± 2 yrs; 166.7 ± 4.4 cm; 64.3 ± 7.0 kg)	TSK-17
Fraser et al,[116] 2019	Cross-Sectional	*CAI*: LAS ≥ 12 months prior to the study who had perceived or episodic giving way and scored >10 on the IdFAI, <90% on the FAAM-ADL and <85% on the FAAM-Sport *COP*: LAS ≥ 12 months prior and no perceived or episodic giving way, and ≤10 on the IdFAI, ≥99% on the FAAM-ADL, and ≥97% on the FAAM-Sport *CON*: No history of ankle or foot sprain	Recreationally Active	20 CAI (F:15, M:5; 19.8 ± 1.3 yrs; 167.4 ± 9.3 cm; 70.4 ± 14.3 kg) 21 COP (F:13, M:8; 20.8 ± 2.9 yrs; 171.0 ± 8.9 cm; 69.3 ± 8.7 kg) 22 CON (F:13, M:9; 19.6 ± 0.9 yrs; 171.1 ± 10.1 cm; 66.5 ± 14.5 kg)	TSK-11
Fukano et al,[73] 2020	Cross-Sectional	*FI*: a history of an ankle sprain and ≥11 on the IdFAI *NFI*: a history of an ankle sprain and ≤10 on the IdFAI *CON*: no history of an ankle sprain and ≤ 10 on the IdFAI	College Athletes (football & lacrosse)	95 FI (F:51, 19.0 ± 4.9 yrs; 159.1 ± 4.4 cm; 54.3 ± 4.4 kg; M:44, 19.9 ± 1.3 yrs; 174.2 ± 6.4 cm; 67.9 ± 6.9 kg) 50 NFI (F:25, 19.8 ± 1.3 yrs; 160.0 ± 5.0 cm; 55.0 ± 5.4 kg; M:25, 19.4 ± 1.0 yrs; 171.2 ± 5.8 cm; 67.1 ± 5.0 kg) 23 CON (F:13, 19.6 ± 1.2 yrs; 159.1 ± 4.2 cm; 54.1 ± 5.1 kg; M:10, 19.6 ± 1.0 yrs; 175.3 ± 7.1 cm; 69.6 ± 7.5 kg)	TSK-17 & AFAQ
Hadadi et al,[74] 2020	Cross-Sectional	*CAI(FI subgroup)*: a history of at least 1 significant LAS ≥ 12 months prior to study participation, ≥2 episodes giving way in the past 6 months and/or RAS and CAIT score < 24, <90 on the FAAM-ADL and <80 on the FAAM-Sport *CON*: no history of ankle sprain	NA	30 CAI (F:22, M:8; 27.00 ± 6.01 yrs; 170.31 ± 6.98 cm; 70.50 ± 7.90 kg) 30 CON (F:22, M:8; 22.83 ± 2.61 yrs; 168.20 ± 7.66 cm; 67.13 ± 9.93 kg)	TSK-17 & FABQ

Table 3.1 Continued

Houston et al,[11] 2014	Case Control	*CAI*: at least 1 lateral ankle sprain and 2 episodes of ‘‘giving way’’ in the past 3 months and answered *yes* to 4 or more questions on the AII *CON*: no history of ankle sprain	Recreationally Active (≥4 on NASA Physical Activity Status Scale)	25 CAI (F:18, M:7; 21.9 ± 2.5 yrs; 170.2 ± 9.1 cm; 70.0 ± 11.4 kg) 25 CON (F:18, M:7; 22.0 ± 2.1 yrs; 167.4 ± 9.1 cm; 64.8 ± 11.2 kg)	TSK-11 & FABQ
Houston et al,[72] 2018	Cross-sectional	*RAS*: a history of ≥ 2 ankle sprains *SAS*: a history of 1 ankle sprain *CON*: no history of ankle sprain	College Athletes (varied sports)	44 RAS (F:33, M:11; 19.3 ± 1.0 yrs; 170.9 ± 8.7 cm; 68.3 ± 11.4 kg) 75 SAS (F:38, M:37; 19.5 ± 1.4 yrs; 174.8 ± 10.8 cm; 71.5 ± 12.8 kg) 28 CON (F:12, M:16; 19.4 ± 1.3 yrs; 173.1 ± 11.1 cm; 72.7 ± 14.9 kg)	FABQ
Koldenhoven et al,[117] 2019	Cross-sectional	*CAI*: a history of at least 1 significant LAS ≥ 12 months prior to study participation, ≤85% on the FAAM-Sport, and ≥11 on the IdFAI *COP*: a history of at least 1 significant LAS ≥ 12 months prior to study participation, ≥97% on the FAAM-Sport, and < 10 on the IdFAI OR they a) answered “no” to the question “Do you frequently roll your ankle or feel like it gives way?” AND b) answered “never” or “once a year” for the following questions: 1) “During activities of daily life how often does your ankle feel unstable?” 2) “During sport or recreational activity how often does your ankle feel unstable?	Recreationally Active (≥1.5 hr activity/week)	18 CAI (F:16, M:2; 21.5 ± 3.4 yrs; 167.5 ± 9.1 cm; 66.9 ± 14.4 kg) 18 COP (F:16, M:2; 20.5 ± 1.9 yrs; 168.2 ± 6.0 cm; 66.2 ± 11.3 kg)	TSK-17
Terada et al,[113] 2017	Cross-sectional	*CAI*: a previous history of an acute LAS which caused swelling, pain, and temporary loss of function at least 1 day; and repeated episodes of “giving-way” in the 6 months prior to study enrollment; and/or recurrent ankle sprains; and/or perceived ankle instability assessed by the AII and IdFAI. *COP*: had returned to full activity for at least 12 months following an initial ankle sprain without recurrent injury, episodes of giving-way, and perceived ankle instability *CON*: no ankle sprain history	NA	48 CAI: (F:25, M:23; 22.0 ± 3.8 yrs, 169.6 ± 8.6 cm, 73.1 ± 16.5 kg) 18 COP: (F:11, M:8; 21.6 ± 4.0 yrs, 169.6 ± 11.3 cm, 72.4 ± 17.3 kg) 26 CON: (F:17, M:9; 21.6 ± 3.2 yrs, 166.1 ± 8.1 cm, 66.2 ± 13.1 kg)	TSK-17

Table 3.1 Continued

Wikstrom,[10] 2011	Cross-sectional	*CAI*: have had an initial LAS that required immobilization and/or non-weight bearing for at least three days, have multiple episodes of giving way within the past year, and at least 1 recurrent sprain between 3 and 6 months prior to study participation, and required to score <22 on the AJFAT *COP*: have suffered an initial ankle sprain that required immobilization and/or non-weight bearing for at least three days but have resumed all pre-injury physical activity without limitation and without further complication for at least 12 months prior to participation, and required to score >22 on the AJFAT	NA	29 CAI: (21.9 ± 2.8 yrs, 1.77 ± 1.27 m, 72.4 ± 12.5 kg) 29 COP: (20.9 ± 1.5 yrs, 1.74 ± 1.06 m, 75.4 ± 1 6.4 kg)	TSK-17
Wikstrom,[47] 2019	Cross-sectional	*CAI*: had at least one lateral ankle sprain and at least two episodes of giving way within the past 6 months; and >11 on IdFAI *CON*: No history of ankle sprain and <10 on IdFAI	NA	45 CAI: (20.07 ± 2.07 yrs, 169.85 ± 7.43 cm, 72.65 ± 14.64 kg) 45 CON: (20.77 ± 2.35 yrs, 169.37 ± 8.34 cm, 67.22 ± 13.12 kg)	FABQ

Abbreviations: AII: Ankle Instability Instrument, AJFAT: Ankle Joint Functional Assessment Tool, CAI: Chronic Ankle Instability, CAIT: Cumberland Ankle Instability Tool, CON: Healthy Controls, COP: Ankle-Sprain Copers, FAAM-ADL: Foot and Ankle Ability Measure Activities of Daily Living, FAAM-Sport: Foot and Ankle Ability Measure Sport Subscale, FABQ: Fear-Avoidance Beliefs Questionnaire, FI: Functional instability, IdFAI: Identification of Functional Ankle Instability, LAS: Lateral ankle sprain, NFI: No functional instability, NASA: National Aeronautics and Space Administration, RAS: Recurrent ankle sprains, SAS: Single ankle sprain, TSK: Tampa Scale of Kinesiophobia

Quality Assessment

The quality index scores for all studies are presented in Table 2. Two studies were determined to be high-quality (80.0% and 86.7%); whereas, the other nine were moderate-quality (60.0-73.3%). None of the studies were low quality. The inter-reliability of the raters was strong ($\kappa = 0.87$).

Table 3.2 Modified Downs and Black Quality Index scores for the included articles

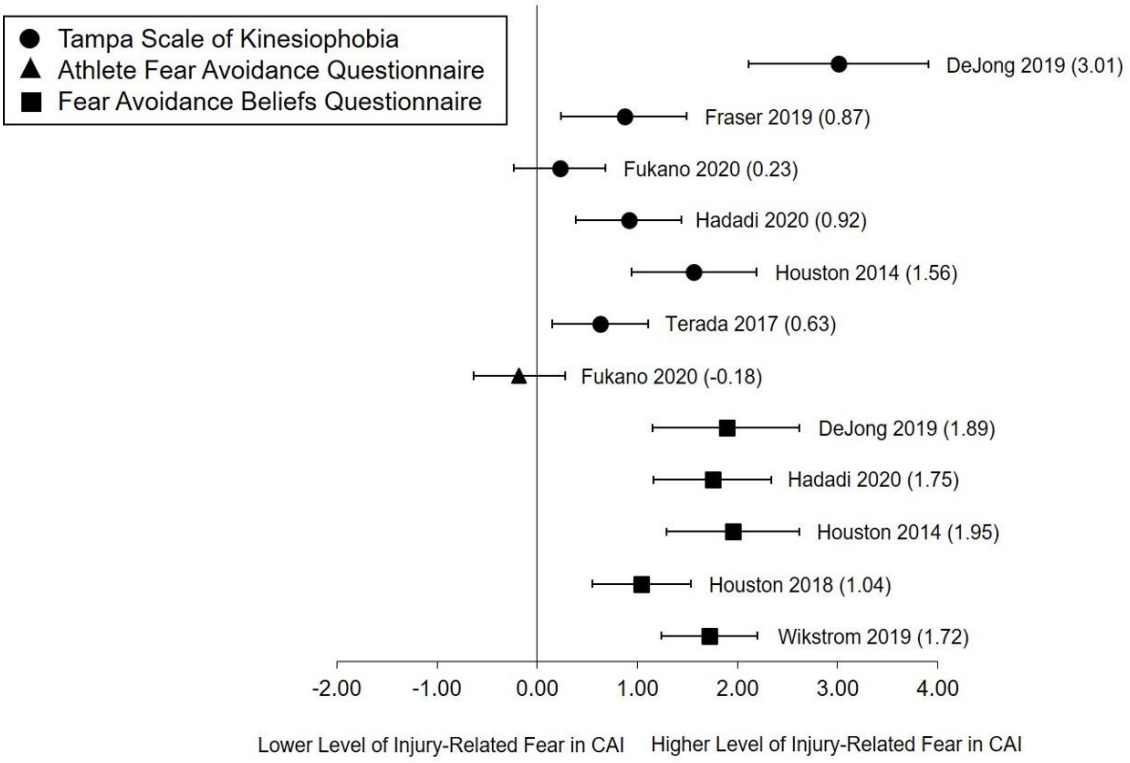
Study	Quality Index Score (%)
DeJong et al,[114] 2019	66.7%
DeJong et al,[115] 2020	73.3%
Fraser et al,[116] 2019	73.3%
Fukano et al,[73] 2020	73.3%
Hadadi et al,[74] 2020	73.3%
Houston et al,[11] 2014	73.3%
Houston et al,[72] 2018	80.0%
Koldenhoven et al,[117] 2019	73.3%
Terada et al,[113] 2017	86.7%
Wikstrom,[10] 2011	60.0%
Wikstrom,[103] 2019	60.0%

Data Synthesis

Chronic Ankle Instability and Healthy Control Groups. Eight studies provided data comparing CAI to CON.[11,47,72-74,113,114,116] Two of the studies used a version of the TSK (TSK-11 or TSK-17),[113,116] two of them used the FABQ,[47,72] three articles reported both TSK and FABQ outcomes,[11,74,114] and one reported TSK-17 and AFAQ outcomes.[73] The ES and 95% CIs are presented in Figure 3.2. All but one estimate indicates that individuals with CAI report higher

levels of injury-related fear when compared to CON. ES for the TSK instruments ranged from weak to strong (0.23-3.01), with one of the comparison's CIs crossing zero.[73] ES for the lone AFAQ comparison was the only negative effect (-0.18), but similarly, the CIs crossed zero.[73] Comparisons of the FABQ instrument were strong (1.04-1.95) and none of the CIs crossed zero.

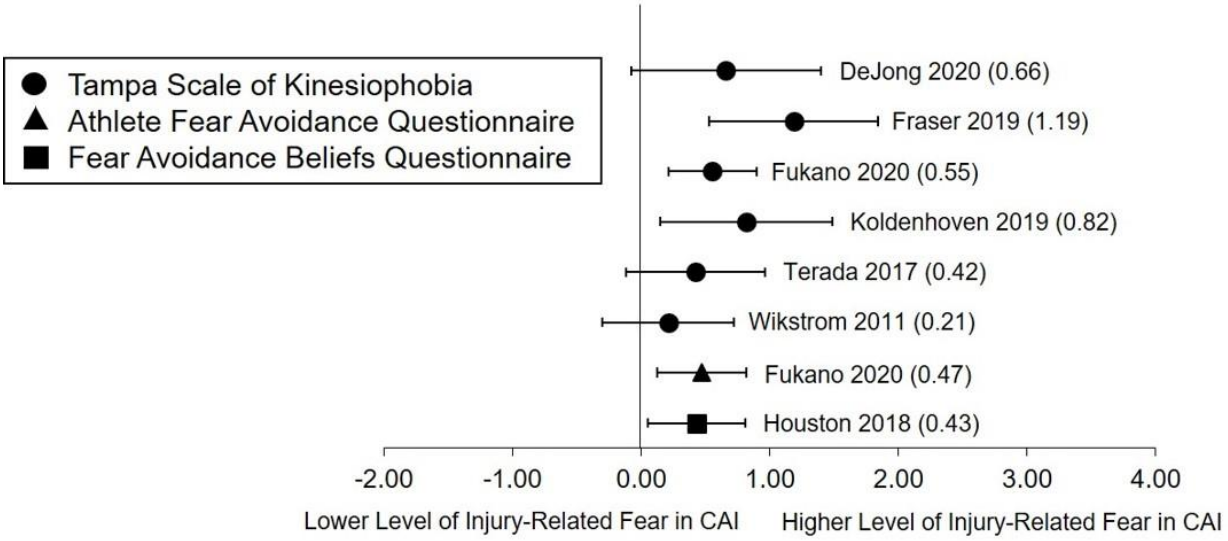
Figure 3.2 Summary of Hedges g effect sizes and 95% confidence intervals for the comparison between the chronic ankle instability and healthy control groups



Chronic Ankle Instability and Ankle Sprain Coper Groups. Seven included studies compared CAI to COP.[10,72,73,113,115-117] Six studies used the TSK,[10,113,115-117] one used the FABQ,[72] and one reported both TSK-17 and AFAQ outcomes.[73] All point estimates indicate that individuals with CAI reported higher levels of injury-related fear when compared to COP (Figure 3.3). ES for the TSK instruments ranged from weak to strong (0.21-1.19), with three of

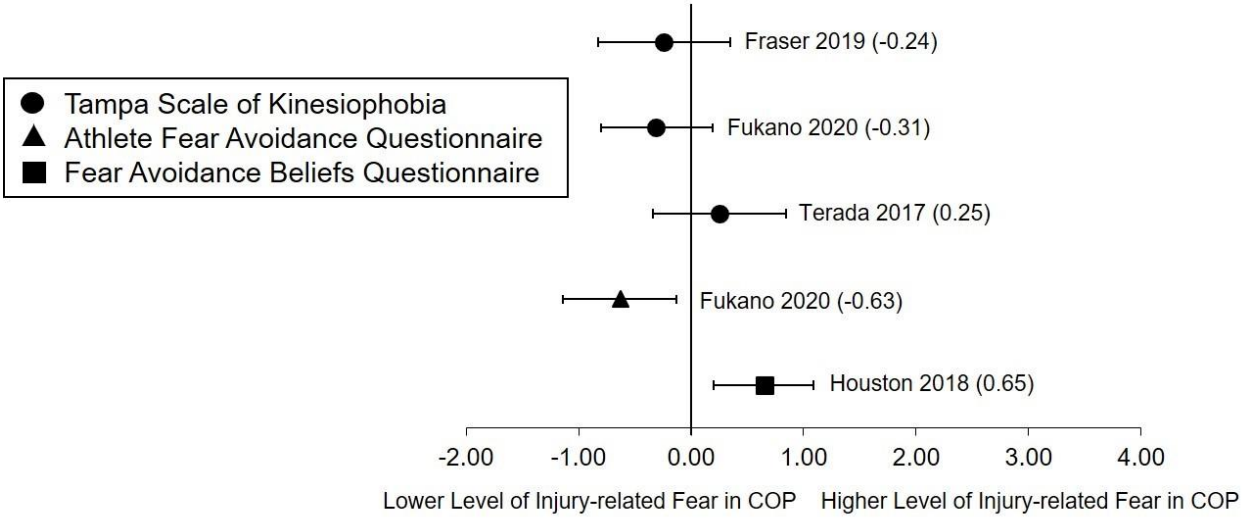
the comparisons crossing zero. The lone comparisons for the AFAQ and FABQ both had a moderate effect (0.47 and 0.43, respectively) with CIs that did not cross zero.

Figure 3.3 Summary of Hedges g effect sizes and 95% confidence intervals for the comparison between the chronic ankle instability and ankle sprain coper groups



Ankle Sprain Coper and Healthy Control Groups. Four studies compared COP to CON.[72,73,113,116] Three studies used the TSK,[73,113,116] one study used the FABQ,[72] and one reported both TSK-17 and AFAQ outcomes.[73] The ES and 95% CIs are presented in Figure 3.4. Inconsistent findings were found for this comparison between instruments. Comparisons of the TSK indicate there were no differences in between COP and CON as they demonstrated inconsistent ES (-0.31-0.25) and all CIs crossed zero. The one AFAQ comparison demonstrated lower reported fear avoidance in the COP compared to CON with a moderate effect (-0.65), while the one FABQ comparison had a positive moderate effect (0.65), suggesting higher levels of fear-avoidance beliefs in this COP group compared to CON.

Figure 3.4 Summary of Hedges g effect sizes and 95% confidence intervals for the comparison between the ankle sprain coper and healthy control groups



3.4 Discussion

The purpose of this study was to conduct a comprehensive systematic review investigating differences in injury-related fear between individuals with and without CAI. We identified eleven studies reporting injury-related fear data allowing for comparisons between individuals with characteristics of CAI, COP, and CON. Our review found strong evidence that individuals with CAI report higher levels of injury-related fear compared to CON and moderate evidence that individuals with characteristics of CAI have greater levels of injury-related fear when compared to COP. Comparisons between COP and CON groups yielded some inconsistencies between instruments, but our overall findings suggest that those who have fully recovered after one ankle sprain, do not seem to differ from those who have no history of ankle sprain. Together these results begin to identify the potential importance of this psychological factor in the development of chronicity after ankle sprain injury. Injury-related fear is an umbrella term which can include multiple sub-constructs. As mentioned, the various PROs that have been used throughout the literature slightly differ in the injury-related fear sub-construct in

which they measure. Therefore, a closer look at the results of the various instruments is warranted to determine which PROs may be the most appropriate to identify injury-related fear within these populations.

Both the AFAQ and the FABQ were created to measure fear-avoidance beliefs. In our review, Fukano et al[73] provided the data for the lone AFAQ comparison between all groups. In this study, the CON group surprisingly reported a higher mean score than both groups with a history of ankle sprain. One potential explanation for this result is that the AFAQ asks the patient to answer the fear belief statements as if they were in pain from a sport injury,[71] as opposed to speaking directly to their ankle/ankle injury. It is possible that this instrument captured a more generalized fear regarding sports injury and was less specific to their fears regarding their ankle. It should also be noted that all participants enrolled in the study were high-contact college athletes,[73] which could also have influenced these findings; however, the actual reasoning for the higher than average scores in the CON participants is largely unknown. Due to this and the limited available studies using the AFAQ, it is difficult to make conclusions on its usefulness in identifying injury-related fear in the ankle sprain populations.

Conversely, the FABQ consistently exhibited moderate to strong effect sizes indicating higher levels of fear-avoidance beliefs in those with CAI when compared to COP and CON, and in COP when compared to CON. Additionally, none of the confidence intervals from the FABQ comparisons crossed zero. This could indicate that the FABQ is more sensitive than the AFAQ at detecting fear-avoidance belief differences between the three groups; however, CAI-COP and COP-CON comparison data were limited to a single study sample. In the CAI and CON comparisons, the FABQ demonstrated strong ES across all 5 studies highlighting the usefulness of detecting fear-avoidance beliefs between these groups.

The Houston et al[72] comparisons demonstrated the slightest dispersion from the rest, yet still had a very strong ES at 1.04 and was the same study that provided the sole effect sizes for the FABQ when comparing CAI to COP, and COP to CON. The criteria used to create the ankle sprain groups was strictly based on number of ankle sprains – so those reporting recurrent sprains were used in our CAI group and those reporting a single ankle sprain were used in our COP group. Although these characteristics are representative of both groups, it is unknown if the participants in these groups meet the full criteria for being an individual with CAI or a true COP, as there was no reported data of physical function or perceived instability for these individuals. It should also be noted that participants in this study, like participants in Fukano et al's[73] study, were college athletes. Therefore, college athletes with an ankle sprain history report higher levels of fear-avoidance than those without one, and athletes who have experienced multiple ankle sprains report the highest levels of fear-avoidance. Interestingly, the CON group in this study also provided the greatest mean FABQ score of all available CON groups which provides further evidence of greater levels of fear-avoidance in college athlete participants regardless of group when compared to non-athlete studies. It is unknown if higher injury-related fear in these athletes is related to unaddressed fears from past injury, greater physical demands, or emotional consequences of injury being enhanced in these individuals, but further explorations may help to elucidate this.

The TSK was the most commonly used PRO amongst our included studies, and measures level of kinesiophobia. Unlike the FABQ, the TSK demonstrated inconsistent ES across group comparisons. Differences in the survey designs likely contribute to the dispersion of effects. The TSK uses a 4-point scale ranging from 1 (*strongly disagree*) to 4 (*strongly agree*) which yields possible scores between 11-44 (TSK-11) or 17-68 (TSK-17); whereas the FABQ uses a 6-point

scale ranging from 0 (*completely disagree*) to 6 (*completely agree*) resulting in possible scores from 0-96. Therefore, the difference between someone *completely/strongly disagreeing* to all belief statements and *completely/strongly agreeing* to all belief statements is 96 points for the FABQ and only 33 (TSK-11) or 51 (TSK-17) points for the TSK, which allows for greater possible mean differences when using the FABQ. This in turn, would affect the magnitude of the calculated effects. There were also discrepancies in which TSK instrument was used between studies. The TSK-17 has been subjected to multiple psychometric analyses, and the four reversely scored items have consistently been problematic.[118] The shortened, 11-item version was created to address these concerns and others in order to make the measure more sound,[67] and may therefore be a better option for clinicians when interested in identifying kinesiophobia in their ankle sprain patients.

Beyond instrument design, there were differences noted between the studies which may also help to explain differences in effects across group comparisons. For the CAI and CON comparison, all point estimates were positive, and only one study showed no effect. As mentioned earlier, the Fukano et al[73] study enlisted college athletes and their CON group showed abnormally high levels of injury-related fear, which resulted in only a small difference in scores between groups. If that study is removed, the point estimates for this group comparison range from moderate to strong (0.63-3.01), suggesting that the TSK is useful for identifying kinesiophobia in those with CAI compared to CON. The CAI and COP comparisons all demonstrated positive effects ranging from weak to strong. The non-significant effects and dispersion in point estimates in this group comparison are likely related to a small sample sizes,[115] challenges defining and determining coper groups, and variations in study quality. Two moderate-quality studies found COP groups to have lower reported TSK scores compared to

CON, and the highest quality study found their COP group had greater scores. Despite the inconsistent findings, all TSK comparisons demonstrated weak effect sizes with CIs that crossed zero, suggesting that there is no difference in kinesiophobia between COP and CON groups. Overall, these findings suggest that the TSK instruments are useful in identifying kinesiophobia related to a history of ankle sprain, and exploration of this construct may produce more clarity between factors that can influence kinesiophobia levels between individuals after ankle sprain. Limited evidence suggests that ankle laxity measures as well as force plate balance outcomes are associated with injury-related fear in some CAI samples, which may suggest that some pathomechanical and motor-behavioral impairments that result from CAI may influence injury-related fear, or vice versa.

Clinical Implications

Our systematic review has demonstrated that the FABQ and TSK-11 may be the best available instruments in detecting heightened levels of injury-related fear in patients with previous ankle sprains and CAI. Injury-related fear, via the Fear-Avoidance Model, is theorized to contribute to the promotion of avoidance behaviors and overtime, lead to cycles of chronic pain and disability.[13,14] Therefore, high levels of injury-related fear could partially explain neural adaptations that further promote avoidance and lead to other movement-behavior impairments such as poor balance, movement pattern alterations, reduced physical activity, and higher levels of reported disablement.[6,82] Encouragingly, there is some evidence of reduced fear after various physical rehabilitation protocols in individuals with CAI.[19-21,119-121] Areas targeted in these interventions include combinations of calf stretching, joint mobilizations, intrinsic foot, ankle, and lumbopelvic complex strengthening, static and dynamic balance, and functional and gait training.[19-21,119-121] Most of these studies targeted multiple aspects of function which also led to

multidimensional improvements in other outcomes including perceived functional ability. So, although no one physical rehabilitation protocol can be recommended at this time, using a multimodal approach that targets the individual needs of the patient is likely the preferred approach to improving both clinical and patient-based outcomes, including reductions in injury-related fear. Beyond typical physical rehabilitation strategies, psychologically informed practice aims to bridge the gap between the management of physical and psychosocial factors following injury and may specifically be of interest to these patients.[122,123] Common psychological frameworks incorporated into rehabilitation protocols include education, imagery, self-talk or reframing, graded exposure, social support strategies, goal-setting, and relaxation.[17] More work is needed to investigate the application of psychologically informed practice in sport injury and specifically ankle sprain populations; however, the literature is promising for the benefits that it can have in individuals following injury.[124-126] More research is also needed to further understand the depth and breadth of fear beliefs held in ankle sprain populations, and the best intervention strategies for addressing these concerns.

Limitations

This review is not without limitations. We conducted our search in electronic databases we believed would capture the literature regarding the population of interest, and also performed a hand search to locate as many studies as possible, but there is still a chance that pertinent literature was missed. We did not include any gray literature and only included peer-reviewed studies, however, since the outcomes of interest for this study were also found in studies that only used these tools as patient descriptors, we believe that this helps against potential publication bias as injury-related fear was not the research question of interest in these studies. Although the IAC has issued guidelines for what should classify an individual as having CAI,

there were some differences in the group criteria amongst the studies, and one study pre-dated the issuance of these guidelines. There were also differences in the group representing COP, and not all groups met criteria suggested to distinguish this group. Although the studies that veered from these criteria were included, we were careful to interpret their contribution to the results appropriately and believe it contributes to the overall look of injury-related fear in the ankle sprain populations.

Finally, a meta-analysis was not included as a part of our analysis due to the inability to reliably quantify or explain the dispersion between studies. The use of random-effects modelling takes into account the variability between studies; however, best practices include reporting prediction intervals as a way to quantify the variance of the effects and a minimum of 10 comparisons are needed to begin to accurately provide useful prediction intervals.[127,128] Additionally, statistical measures used to explain any dispersion include sub-group analysis or meta-regression. These statistical procedures are not useful without a substantial amount of studies (at least 10) per group, or per characteristic.[129] Further, there is a lack of knowledge on what characteristics may be important to use for these investigations in relation to injury-related fear in the ankle sprain populations. As such, we believe that we could not provide reliable quantitative data that would further inform our systematic review. We do recommend that future investigations aim to identify factors that may help to explain the differences in injury-related fear levels in ankle sprain populations to further our understanding of this construct and to inform best practices.

3.5 Conclusions

Our systematic review suggests that there are heightened levels of injury-related fear in individuals with CAI when compared to both COP and CON. Individuals who have fully

recovered after one ankle sprain do not seem to differ from those who have no history of ankle sprain, which further emphasizes the importance of this impairment to individuals with CAI. Therefore, appropriate PROs should be considered in the treatment of ankle sprains to identify and monitor individuals who report injury-related fear as it may contribute to the development of future instabilities and associated impairments. Some associations have been made between injury-related fear and other impairments in this population, but more research is needed to understand influential factors contributing to these fears and how this impairment may affect movement behavior and disability. Although some typical physical rehabilitation protocols have shown to reduce injury-related in those with CAI, more research investigating psychologically informed practice strategies in this population may be needed to determine the best interventions to address these concerns.

CHAPTER 4

THE FEAR-AVOIDANCE MODEL AND CHRONIC ANKLE INSTABILITY

4.1 Introduction

Soft tissue sprain and strain injuries are the most common musculoskeletal injuries seen across healthcare facilities, with estimates of 4.2 million of these injuries involving the ankle and foot annually.[22] In physically active populations, sprains to the ankle ligaments are substantially more common,[22] with lateral ankle sprains demonstrating the highest incidence.[4] Lateral ankle sprains are often regarded as benign injuries that will resolve quickly with minimal treatment. While there are patients who seem to fully recover after their ankle sprain injury, known as ankle sprain copers, evidence suggests that 40% of individuals continue to suffer from recurrent sprains, episodes of instability, and perceived ankle instability for over one year after their initial sprain.[4] These persistent symptoms characterize a condition known as chronic ankle instability (CAI).[5] Many other impairments have been identified within the CAI population including stability and movement pattern alterations, decreased perceived levels of ankle function, increased levels of global disability, and physical activity restrictions.[69] Acute and chronic ankle sprains are also estimated to account for up to 80% of all cases of post-traumatic ankle osteoarthritis.[24] Despite years of research, it is still not fully understood which specific factors, or combination of factors, lead some patients down this continuum of disability.

The characterizing symptoms of CAI are specifically focused on symptoms of ankle instability, whereas other chronic musculoskeletal conditions are typically characterized by pain. Up until more recently, pain had not been a real focus in the CAI literature despite evidence of persisting pain after ankle sprains beyond the typical acute stage.[40,44] A recent retrospective analysis revealed 60% of CAI participants in previous research studies reported pain during

different levels of activity.[46] The role of pain in CAI is still unknown but it has shown associations with perceived instability[46] and function[47] in recent reports. Despite this, the intensity of recurrent pain in this population was reported to be a mild intensity[15] which may not alone contribute to changes in function. It is well-documented that pain is inextricably linked to emotional and cognitive functions.[50] Specifically of interest is the potential role of injury-related fear as this has been identified in individuals with CAI and has shown associations to negative outcomes after injury regarding physical impairments, recovery, and function in other conditions.[17,76] The fear-avoidance model (FAM) is a cognitive-behavioral model that has been widely applied to explain this phenomenon in those populations.[13]

The FAM (Figure 1.1) postulates that exaggerated negative beliefs about pain, known as pain catastrophizing, can lead patients into a cycle of fear and activity avoidance.[13] These changes can lead to disuse which can often create new pathological pain pathways beyond the healing of the originally injured tissue, that continues these individuals down the path toward chronic pain and disability. On the other side of the model, individuals who do not prioritize pain-related thoughts after injury are thought to be able to then confront their pain and injury, which leads them towards full recovery and function.[13] Considering that after an ankle sprain injury, we know that there is a clinical spectrum of outcomes with one end of the spectrum being copers, or full recovery and function, and the other end being CAI, or those who suffer from residual ankle instability that leads to dysfunction and disability, the FAM and its components may also serve as a theoretical model for understanding the development of CAI in some individuals post-ankle sprain.

Therefore, the purpose of this study was to determine whether the FAM and its components may be applicable to patients with CAI by examining relationships between pain

catastrophizing, injury-related fear, pain, ankle function and global disability. This was tested through three specific aims. Our first aim was to examine the relationship between the two cognitive-affective model components – pain catastrophizing and injury-related fear. Pain catastrophizing is thought to contribute to the development of injury-related fear, but it is also possible that those who are fearful of re-injury may adopt pain catastrophizing cognitions that increase focus on the feared stimuli of pain. Thus, our first hypothesis was that greater pain catastrophizing beliefs would be related to greater levels of reported injury-related fear. Our second aim was to determine the influence of pain presence on reported function and disability. We hypothesized that the presence of pain would explain additional variance beyond reported instability in both ankle function and global disability outcomes. Our third aim was to determine the unique role of the cognitive-affective model components in predicting function and disability. We hypothesized that when controlling for instability and pain, both pain catastrophizing and injury-related fear would uniquely explain additional variance in both function and disability.

4.2 Methods

This study used a cross-sectional survey design and was approved as exempt research by the Old Dominion University Health Sciences Human Subjects Review Committee in December 2020. Recruitment for potential participants occurred over a 4-week period and was done via email in a university setting, and through shareable social media posts (Facebook and Twitter) to broaden our geographical and demographic reach. Participants could be of any gender or ethnicity but were required to be between the ages of 18 and 40 years old. Inclusion and exclusion criteria for potential CAI participants followed the guidelines set forth by the

International Ankle Consortium[5] and questions pertaining to this criteria were included in the survey to determine eligibility.

Participants were classified as having CAI if they reported at least one significant ankle sprain which was sustained at least 12 months prior to the survey and also reported residual symptoms including recurrent ankle sprains, and/or 2 or more giving away episodes in the previous 6 months, and/or perceived instability classified as a score ≥ 11 on the Identification of Functional Ankle Instability (IdFAI).[5] Individuals were excluded if they had sustained an acute lower extremity injury within the past three months, or had a history of lower extremity fracture or surgery.

We used Qualtrics (Provo, Utah) to create the anonymous survey which consisted of 37 total questions. This included the informed consent, a demographic section, general inclusion and exclusion criteria, specific questions and tools to determine the classification of CAI,[5] and the patient-related outcome assessments for collecting pain catastrophizing, injury-related fear, pain, ankle function, and global disability outcomes. As each of the patient-related outcome assessments have established validity and reliability levels, no additional validation was completed for our survey. Additionally, the patient-related outcome assessments were organized into matrix-type questions to lower the overall total number of question in the survey.

Pain Catastrophizing

The Pain Catastrophizing Scale (PCS) was used to assess pain catastrophizing beliefs[88] as it has demonstrated strong internal consistency (α=0.93), good test-retest reliability (ICC=0.75), and validity.[88,130,131] The PCS is a 13-item scale assessing the frequency of negative pain-related beliefs and ranges from 0 (*not at all*) to 4 (*always*). Total scores are calculated (ranging from 0-

52), along with three subscale scores assessing magnification, rumination, and helplessness, with higher scores indicating higher levels of pain catastrophizing.

Injury-Related Fear

The Tampa Scale of Kinesiophobia-11 (TSK-11) was used to assess fear of movement and re-injury.[67] It has demonstrated good internal consistency (α=0.79), test-retest reliability (ICC=0.81), and validity when compared to the original 17 item scale,[67] and has demonstrated differences between individuals with and without CAI.[11] It is an 11-item scale ranging from 1 (*strongly disagree*) to 4 (*strongly agree*) yielding total scores ranging from 11-44, with higher scores indicating higher levels of fear related to movement and re-injury.

Pain

Pain was used as a binary outcome (present or not present) for the purpose of this study, and was determined using the answer on two survey questions. The first question is from the Cumberland Ankle Instability Instrument (CAIT) and states, "I have ankle pain" and has six potential answers (walking on level surfaces, walking on uneven surfaces, running on level surfaces, running on uneven surfaces, during sport, or never). Participants who reported pain during any level of physical activity were considered to have pain. Because this question describes conditional pain activities, the use of a numerical rating scale for pain was also used secondarily to determine pain presence. Participants were asked to rate their highest level of ankle pain they have experienced within the past week on a scale from 0 (*none*) to 10 (*worst pain imaginable*). Any participant who responded with reported pain > 0 was considered to have pain.

Ankle Function

The Quick-FAAM is a regional scale designed to determine functional limitations in those with foot and ankle conditions.[77] It is a shortened version of the Foot and Ankle Ability

Measure (FAAM)[132] and retained five items from the FAAM-Activities of Daily Living and seven items from the FAAM-Sport subscales. It is a 12 item scale ranging from 4 (*no difficulty at* all) to 0 (*unable to do*). Scores are totaled and transformed into percentages, with 100% being representative of no functional loss. It has demonstrated strong internal consistency ($\alpha = 0.94$),[77] and acceptable test-retest reliability,[78] and recently was found to be able to distinguish between individuals with CAI and copers, with CAI patients demonstrating lower scores.[79]

Global Disability

The modified Disablement in the Physically Active Scale (mDPA) is a global scale designed for individuals who are physically active.[133] The mDPA has demonstrated high test-retest reliability (ICC=0.943) and internal consistency (α=0.890–0.908).[133] The mDPA is 16 items ranging from 0 (*no problem*) to 4 (*severe*) and addresses both physical and mental factors. Total scores range from 0-64, with higher scores being indicative of increased disablement. The mDPA has shown to detect differences in those with and without CAI, with individuals with CAI reporting higher disablement.[11]

Statistical Analyses

Statistical analyses were performed using IBM SPSS Statistics, version 27 (IBM Corporation, Armonk, NY) on all participants who were classified as CAI. Individuals were excluded if the survey was not completed in its entirety or if they did not meet the full inclusion and exclusion criteria. Demographic variables are summarized as either mean ± standard deviation or as n (%) overall. To test the first hypothesis, Pearson-product moment correlations were used to evaluate the relationships between pain catastrophizing (PCS) and injury-related fear (TSK-11) and correlation coefficients (r) were interpreted as (negligible < 0.3, low = 0.3-0.49, moderate = 0.5-0.69, high = 0.7-0.89, very high = 0.9-1.0).[134]

To test our second hypotheses, two hierarchical linear regression models were used to determine the influence of pain presence on function and disability. The Quick-FAAM and mDPA served as the outcome variable in their respective models. For both models, the IdFAI score was used as a control variable and therefore entered in the first block. Pain was then entered as a two-level predictor (0=no pain; 1=pain) into the second block to determine its additional utility in predicting function and disability.

To test our final hypotheses, two hierarchical linear regression models were used to determine the influence of the cognitive-affective outcomes on function and disability. Again, the Quick-FAAM and mDPA served as the outcome variable in their respective models. For these analyses, both IdFAI and pain were used as control variables and entered in block one. PCS and TSK-11 were then simultaneously entered into the second block to determine their additional utility in predicting function and disability.

The data were assessed for bias by identifying any cases that may be outliers or influential, and although in all models, a few cases were found to have residuals >±2 standard deviations and one case in the mDPA model was found to have residuals >±3 standard deviations, all cases proved not be influential (Cooks distance <1) to their models. Linearity and additivity were assessed by plotting the predictors and outcome to ensure this assumption was satisfied. Effects due to multicollinearity were limited by ensuring the Pearson's correlation coefficients between predictor variables in the final model were less than 0.9, inspecting variance inflation factors and tolerances, and examining the variance distribution on the eigenvalues in the collinearity diagnostics table. The assumption of homoscedasticity was verified by inspection of the regression of standardized residual versus regression of standardized predicted value plot. Durbin-Watson testing yielded no problem with the assumption of independent errors, and

although normality of errors testing indicated a slight skew in the data, we assumed normality based on the central limit theorem (>30 participants), and used bootstrapping to re-estimate the robustness of the significance testing of the model parameters, and to obtain 95% bias corrected (BCa) confidence intervals using 1,000 iterations. All assumptions were tested with strategies presented by Field.[135] Overall performance of the final model was evaluated using R^2 and significance was set to *a priori* at $p < 0.05$.

4.3 Results

Due to the nature of our recruitment strategy, we were unable to determine the number of potential participants that our survey could have reached, however, of those that accessed the survey (n = 314), 259 completed and submitted their answers, for a completion rate of 82.5%. Of those who completed the survey, 114 did not meet the basic inclusion and exclusion criteria (8 due to age, 56 due to history of surgery, 36 due to history of fracture, 13 due to recent acute injury, and 1 reporting no history of a significant ankle sprain). An additional 19 did not meet our CAI criteria, which left a total of 126 CAI participant responses that were included in our analysis. Demographic data and mean outcome measure scores for participants are presented in Table 4.1.

We found a significant, low, positive relationship between PCS and TSK-11 scores (r =0.493, 95% BCa CI [0.357, 0.606], $P < .001$), indicating that as reported levels of pain catastrophizing increased so did reported levels of injury-related fear.

The model with IdFAI entered as a single predictor significantly explained 23.4% of the variance in Quick-FAAM scores ($R^2 = .234$, $P < .001$), and the addition of pain significantly improved the Quick-FAAM model by accounting for an additional 8.9% of the variance ($F\Delta$ = 16.099 (1, 123) $P < .001$). For the final model, both IdFAI and pain were found to be

significantly negatively related to Quick-FAAM (R^2 = .322, P < .001) and each predictor demonstrated unique predictive utility (Table 4.2).

Table 4.1 Participant demographics and patient-reported outcome data

Demographic or Outcome	n (%) or mean (SD)
Gender Identity	n = 126
Male	17 (13.49%)
Female	107 (84.92%)
Other*	1 (0.79%)
Prefer not to specify	1 (0.79%)
Age	32.69 (4.38)
Physical Activity Score**	n = 126
1	5 (3.97%)
2	11 (8.73%)
3	17 (13.49%)
4	45 (35.71%)
5	48 (38.10%)
IdFAI	17.31 (4.90)
Pain Presence	n = 126
No Pain	44 (34.92%)
Pain	82 (65.08%)
Pain Catastrophizing Scale	7.32 (7.46)
Helplessness	2.30 (2.94)
Magnification	2.16 (2.25)
Rumination	2.87 (3.12)
TSK-11	21.36 (5.53)
Quick-FAAM	83.22 (14.95)
mDPA	10.50 (10.67)
Physical	8.68 (8.87)
Mental	1.82 (2.85)

*Participant identified as non-binary **As described by Jurca et al[136] 2: Regular (≥5 days/week) low level exertion >10 minutes at a time; 3: Aerobic exercise, vigorous sport, or similar exertion for 20-60 minutes/week; 4: Aerobic exercise, vigorous sport, or similar exertion for 1-3 hours/week; 5: Aerobic exercise, vigorous sport, or similar exertion for over 3 hours/week

Table 4.2 Perceived instability and pain as predictors of function

Model		*b* (95% BCa CI)	SE *B*	β	*P* value
1	(Constant)	108.778 (101.081, 116.909)	3.764		.001*
	IdFAI	-1.477 (-1.904, -1.044)	.223	-.484	.001*

2	(Constant)	107.066 (100.162, 114.319)	3.405		.001*
	IdFAI	-.979 (-1.450, -.527)	.233	-.321	.001*
	Pain Presence	-10.604 (-14.536, -6.257)	2.191	-.339	.001*

Confidence intervals, standard error, and significance are based on 1000 bootstrap samples *<0.001

The model with IdFAI entered as a single predictor significantly explained 21.4% of the variance in mDPA scores (R^2 = .214, P < .001), and again, the addition of pain significantly improved the mDPA model by accounting for an additional 6.6% of the variance (FΔ = 11.198 (1, 123) P = .001). For the final model, both IdFAI and pain were found to be significantly positively related to mDPA (R^2 = .280, P < .001), and each predictor demonstrated unique predictive utility (Table 4.3).

Table 4.3 Perceived instability and pain as predictors of disability

Model		*b* (95% BCa CI)	SE *B*	β	*P* value
1	(Constant)	-6.876 (-12.152, -1.099)	2.932		.022
	IdFAI	1.004 (.644, 1.353)	.183	.463	.001*
2	(Constant)	-5.830 (-11.175, -2.56)	2.920		.046
	IdFAI	.700 (.316, 1.103)	.213	.322	.003
	Pain Presence	6.482 (2.929, 10.242)	1.883	.292	.002

Confidence intervals, standard error, and significance are based on 1000 bootstrap samples *<0.001

As noted in the previous Quick-FAAM analysis, both IdFAI and pain presence were found to be significant predictors of Quick-FAAM scores, accounting for 32.2% of the variance. The addition of the cognitive-affective outcomes (PCS and TSK) to the model significantly

improved the Quick-FAAM model by accounting for an additional 16.5% of the variance (FΔ = 19.434 (2, 121) $P < .001$). For the final model, all predictors were significantly negatively related to Quick-FAAM ($R^2 = .487$, $P < .001$), and each predictor demonstrated unique predictive utility (Table 4.4).

4.4 Perceived instability, pain, and cognitive-affective variables as predictors of function

Model		*b* (95% BCa CI)	SE *B*	β	*P* value
2	(Constant)	120.620 (112.037, 129.231)	4.515		.001*
	IdFAI	-.650 (-1.104, -.216)	.230	-.213	.006
	Pain Presence	-10.045 (-13.664, -6.072)	2.023	-.322	.001*
	PCS	-.393 (-.714, -.095)	.163	-.196	.016
	TSK	-.783 (-1.182, -.375)	.210	-.290	.001*

Confidence intervals, standard error, and significance are based on 1000 bootstrap samples *<0.001

Similarly, in the previous mDPA analysis, both IdFAI and pain presence were found to be significant predictors of mDPA scores, accounting for 28.0% of the variance. The addition of the cognitive-affective outcomes (PCS and TSK-11) to the model significantly improved the mDPA model by accounting for an additional 16.2% of the variance (FΔ = 17.578 (2, 121) $P < .001$). For the final model, all entered predictors significantly positively related to mDPA ($R^2 = .442$, $P < .001$), and each predictor demonstrated unique predictive utility (Table 4.5).

Table 4.5 Perceived instability, pain, and cognitive-affective variables as predictors of function

Model	*b* (95% BCa CI)	SE *B*	β	*P* value

2	Constant	-14.152 (-20.570, -7.159)	3.355		.001*
	IdFAI	.475 (.083, .890)	.206	.219	.026
	Pain Presence	6.169 (2.660, 9.247)	1.644	.278	.001*
	PCS	.346 (.098, .585)	.120	.243	.003
	TSK	.463 (.167, .743)	.147	.241	.002

Confidence intervals, standard error, and significance are based on 1000 bootstrap samples *<0.001

4.4 Discussion

The purpose of our study was to apply the FAM to the CAI population by investigating specific relationships between some of the model components. We were first interested in investigating whether a relationship existed between pain catastrophizing and injury-related fear variables as no literature has investigated the use of pain catastrophizing in the CAI population thus far. Our hypothesis was supported in that higher levels of pain catastrophizing were significantly related to higher levels of injury-related fear. This relationship is hypothesized to exist because individuals who catastrophize pain and injury, appraise pain as highly threatening. This increase in the value given to the threat of pain is therefore believed to lead someone to develop fear regarding movements that are associated with pain and injury.[13] Although our study cannot infer the direction of this relationship, our results demonstrate that they are significantly related constructs. There is some debate in the literature on the uniqueness of these inter-related variables,[90] however, the strength of this relationship was just under moderate, so although the constructs were found to be related, our results indicate they are unique and independent constructs and could both be used in further analyses. Others studying these variables have produced similar findings to ours.[76,90,137] Further, as injury-related fear is an established factor

related to CAI, this relationship does suggest that pain catastrophizing may be another cognitive-affective variable warranting further investigation in the ankle sprain population.

It is well-established that CAI can result in individuals reporting deficits in ankle function and greater levels of global disability. The FAM postulates that pain, pain catastrophizing, and injury-related fear lead an individual to avoidant behavior which then sends them down the road of disability. Therefore, our remaining hypotheses had specific interest in how pain, pain catastrophizing, and injury-related fear related to reported ankle function and disability. Our second aim was to determine the predictive utility of symptom-related factors that have been established in the CAI population on function and disability with a special interest in determining the additional utility of pain presence on these outcomes as the role of persistent pain in the CAI population has been somewhat overlooked. Our results indicate that greater levels of perceived instability were associated with lesser reported ankle function and greater reported disability within our CAI participants. Perceived instability significantly predicted 23.4% of variance in reported ankle function and 21.4% of variance in reported disability. Perceived instability is one of the characterizing symptoms of CAI[5] so it is not surprising that this variable would serve as an important predictor. Our hypothesis was further supported in that the models significantly improved when adding pain presence as an additional predictor which accounted for an increased 8.9% and 6.6% of the variance in reported ankle function and disability, respectively. This finding is consistent with a recent cross-sectional study that found relationships exist between reported pain and function in their CAI sample[47] and suggests that beyond perceived instability, individuals who reported pain during activities specified by the CAIT or reported pain within the past week, reported lower levels of ankle function and greater disability. Perceived instability and pain have recently demonstrated to have a relationship in a previous investigation,[46] but

despite this, we found both variables to be unique predictors of function and disability, and contribute similar weight to the model.

Our final models, including all four variables, explained 48.7% of the total variance in reported ankle function and 44.2% of the total variance in reported disability. Each predictor was found to significantly add to the model and reveals that greater perceived instability, pain presence, greater pain catastrophizing, and greater injury-related fear were related to lesser reported ankle function and greater reported disability. Our hypothesis was supported in that the models significantly improved when pain catastrophizing and injury-related fear were added as predictors, when controlling for both instability and pain. Together, they accounted for an additional 16.5% and 16.2% of the variance in reported ankle function and disability, respectively, which highlights their importance to the models. The use of the FAM framework has garnered support across multiple musculoskeletal conditions,[18,97,138,139] including those with foot and/or ankle pain,[140] and overall, our results demonstrate relationships that are similar to the theoretical framework presented in the FAM, suggesting it may prove useful for continued study of these variables within ankle sprain populations. Although there is an innate protective mechanism of fear which may be beneficial to the patient during certain periods of recovery, it is not well-understood when this fear may become debilitating to a patient's function or ability. Therefore, our results further support the continued pursuit of understanding the role of persistent pain and cognitive-affective factors, such as pain catastrophizing and injury-related fear, on the development and continuance of CAI and its associated impairments. Additionally, investigating intervention strategies that mitigate persistent pain and lowering injury-related fear would likely assist in improving function and disability.

Pain is often lumped in as a solely physical symptom, however, it is well-established that pain – specifically persisting or recurring pain - is a multidimensional experience influenced by many factors.[31] So although interventions specific to pain in the ankle sprain populations are warranted, our results suggest that psychologically informed intervention strategies may assist in the efficacy of reducing pain by targeting the interrelated cognitive-affective factors. Common psychological frameworks incorporated into rehabilitation protocols include education, imagery, self-talk or reframing, graded exposure, social support strategies, goal-setting, and relaxation.[17] More work is needed to investigate the application of psychologically informed practice in sport injury and specifically ankle sprain populations; however, the literature is promising for the benefits that it can have in individuals following injury.[124-126]

Limitations

This study is not without limitations which should be considered when interpretating our results. The biggest limitation is that due to the cross-sectional design, we cannot infer causality. Further, all of our participants were individuals with CAI, which limits our ability to determine the predictive utility of these variables in the development of the condition. Future research could perform prospective analyses, measuring these variables overtime and determine their use in predicting CAI, and its associated impairments.

Another limitation of our study is that there was still approximately 50% of the variance that was not explained by our variables. Due to institutional COVID-19 research restrictions that prohibited in-person data collection, only patient-reported outcomes were used and limited the availability of clinician-rated measures. For example, balance performance is established in the CAI literature as an important variable related to reported function and disability, and likely

another variable that could help to inform our models. This and other established clinician-rated variables may be considered in future investigations.

Lastly, we recognize there are inherent limitations when using self-report outcomes measures that can include memory and recall bias and can play a role in skewing the data collected and used within our models. Despite the limitations, we do believe that our study lends support for the FAM model being an important consideration to the CAI population.

4.5 Conclusions

Our study examined the influence of perceived instability, pain, pain catastrophizing, and injury-related fear on reported ankle function and disability in individuals with CAI. All of these variables were found to serve as predictors of function and disability, which continues to support the notion that the condition is multifactorial and that these variables are important for clinicians to consider when examining or treating an individual after ankle sprain(s). Our design limitations further warrant investigations focused on the role these variables play in the transition from an acute ankle sprain to CAI, and how these variables may relate to other known impairments within these populations.

CHAPTER 5

UNDERSTANDING INJURY-RELATED FEAR IN INDIVIDUALS WITH CHRONIC ANKLE INSTABILITY: A QUALITATIVE APPROACH

5.1 Introduction

Chronic ankle instability (CAI) is estimated to affect 40% of patients who suffer a lateral ankle sprain[4] and is characterized by recurrent instability episodes which may result in recurrent sprains, as well as feelings of instability in their ankle.[5] These continuous bouts or feelings of ankle instability often result in challenges to physical function, movement, and activity which have been well-established in the literature.[24] More recently, it has been revealed that many individuals with CAI also report having heightened levels of injury-related fear. Injury-related fear is an umbrella term that includes but is not limited to kinesiophobia, fear-avoidance beliefs, and reinjury anxiety,[11] and is thought to develop from memories of the pain and discomfort suffered after injury.[12] The most common element of injury-related fear studied in the CAI population is kinesiophobia, which is described as fear beliefs regarding movements, which the individual feels may make them vulnerable to pain and (re)injury.[67] Considering that those with CAI often experience multiple ankle injuries, or continually experience bouts of ankle instability that could cause re-injury during movement and activity, it is not surprising that these patients report experiencing fears regarding re-injury.

In other musculoskeletal conditions, kinesiophobia has been shown to be a barrier to rehabilitation, and has also been associated with pain, disability, and low quality of life outcomes.[16] In Chapter 4, it was revealed that kinesiophobia was also predictive of function and disability ratings in those with CAI suggesting it is an important variable to the condition. There is some evidence that rehabilitation and multimodal interventions have the ability to improve

reported fear beliefs in individuals with CAI,[19-21] but the presence of these fears may also require more specific treatment approaches that are focused on the modification of fear beliefs and their subsequent behavioral consequences. However, in order to create appropriate intervention strategies, it is important that we have a deeper understanding of injury-related fear within the CAI population as our current understanding of this construct is limited to quantitative scores.

Patient-reported outcomes (PROs) offer a standardized approach to measure psychological constructs such as injury-related fear,[8] and the Tampa Scale of Kinesiophobia (TSK)[67] is most commonly used to assess levels of kinesiophobia in musculoskeletal conditions, including patients with CAI. PROs like the TSK serve an important role in identifying this dimension of health and are also important for tracking improvements or changes to the dimension throughout treatment and rehabilitation. However, PROs should be used by clinicians and researchers beyond just the quantitative data they can provide as the overall score doesn't necessarily give the clinician a clear picture of specific fear beliefs a patient may hold. For example, a patient could disagree with all 11 items on the TSK, which would yield a total score of 22; whereas, another patient may indicate agreement with 5 out of 11 items and strongly disagree with the remaining 6 and produce an overall score of 21. If comparing scores only, these patients would look quite similar, and one would even consider patient 1 (with a score of 22) to have higher levels of fear compared to patient 2 (with a score of 21). In reality, patient 2 had an overall greater display of specific fear beliefs, despite a slightly lower overall score. Therefore, when creating individualized interventions, it is important for the clinician to look beyond the overall score and look at each item individually. Discussing the elements of the PRO with patients allows for an understanding of the experiences and/or factors that underlie the scores and would ultimately lead to the ability to address them within the intervention if necessary.

At this point in the CAI literature, it is unknown if those who present with greater levels of fear demonstrated by higher TSK scores, have similar experiences regarding fears of movement and reinjury, or if there is vast variation in experiences. Further, although CAI is known to be a heterogenous condition, it is also unknown if there are commonalities in the fear beliefs held in those with CAI that are specifically related to the condition that may not be gleaned from PRO scores alone. Therefore, the purpose of this study was to explore the perceptions and experiences that influence injury-related fear, measured with the TSK-11, within the CAI population**.**

5.2 Methods

This study was approved as exempt research by the Old Dominion University Health Sciences Human Subjects Review Committee in December 2020 as a secondary analysis to a larger study presented in Chapter 4. The larger study used a cross-sectional survey design to collect varying patient-reported outcomes to determine the use of the Fear-Avoidance Model as a framework for CAI. The survey was used as a recruitment platform for this study and was also used to confirm consent to participate, determine CAI eligibility,[5] and to collect the TSK-11 data. The exploration of injury-related fear via the TSK-11 instrument was the priority for this portion of the study and we used a consensual qualitative research (CQR) approach to explore individuals' experiences and perceptions underlying their agreement with TSK-11 items.[141] CQR includes aspects of grounded theory,[142] phenomenology,[143] and comprehensive process analysis,[144] and focuses on consistent data collection strategies and diverse teams to reduce bias.[141]

Sixty-one individuals with CAI who had completed the survey portion of our study and indicated interest in participating in qualitative interviews were considered potential participants

for this study. We then contacted participants using purposive sampling based on reported level of kinesiophobia determined by the percentage of agreement with the statements on the TSK-11, starting with the participants reporting the highest value. We used this instead of the total score to capture perceptions for as many fear belief statements as possible, and due to the issues with total score interpretations noted in the introduction. The primary researcher (AS) contacted potential participants via email over a one-month period and data saturation was used to guide this process. Due to the exploratory nature of CQR, it is suggested that 8-15 participants are needed to achieve data saturation,[141] and for our study was determined to be reached after nine participant interviews. Participant characteristics and TSK-11 agreement data can be found in Table 5.1.

Table 5.1 Participant characteristics and patient-reported outcome data

Participant Pseudonym	Gender Identity	Age (yr)	Physical Activity Score*	Percentage of Agreement on TSK-11	Statements of Agreeance	TSK-11 Score
Jessica	Female	30	2	81.82%	1,3,4,6-11	31
Melissa	Female	35	4	63.64%	1,4-7,9,11	31
Luke	Male	23	4	54.55%	1,2,6,7,9,11	29
Elizabeth	Female	30	5	45.45%	1,2,5,7,11	26
Megan	Female	29	3	45.45%	2,7,8,9,11	24
Sarah	Female	35	4	45.45%	1,3,4,7,11	27
Amber	Female	33	4	45.45%	1-3,7,10	26
Tiffany	Female	31	4	36.36%	5-7,9	22
Phil	Male	33	3	36.36%	6,7,9,11	22

*****As described by Jurca et al[136] 2: Regular (≥5 days/week) low level exertion >10 minutes at a time; 3: Aerobic exercise, vigorous sport, or similar exertion for 20-60 minutes/week; 4: Aerobic exercise, vigorous sport, or similar exertion for 1-3 hours/week; 5: Aerobic exercise, vigorous sport, or similar exertion for over 3 hours/week

Instrumentation

A semi-structured interview guide containing open-ended questions was created to explore individuals' reasoning behind their agreement level to the fear belief statements on the TSK-11. This instrument has 11 fear belief statements with a scale ranging from 1 (*strongly*

disagree) to 4 (*strongly agree*), with higher scores indicating higher levels of fear related to movement and re-injury.[67] Each participant's interview guide, specifically the direction of the questions they received, was pre-determined based on their reported level of agreement to each of the TSK-11 statements collected during the survey portion of the study. The specific statements each participant indicated agreement with are provided in Table 5.1 for reference. Before data collection, the interview guide was reviewed for clarity by the research team and the final interview guide (Table 5.2) was successfully piloted, without any further changes. The individual who completed the pilot interview was identified as a potential participant from the survey portion of the study, however, due to his agreement percentage on the TSK-11 being very low, and our purposive sampling strategy, he was not included as one of our nine participants as many others would have been contacted before him.

All interviews were conducted virtually (Zoom, San Jose, CA) by the primary researcher (AS) in order to provide consistency and credibility for data collection. Each participant consented verbally to have their interview recorded and automatically transcribed by Zoom services, and all interviews lasted between 35 and 60 minutes. At the completion of each interview, the primary researcher reviewed the transcripts along with the audio recordings in order to correct transcription errors. Reviewed transcripts were then de-identified and sent back to participants who were allowed one week to perform member checking to enhance trustworthiness of the data. If the participant did not respond within a week's time, the transcript was used as-is, and of those who did respond, all felt the transcripts accurately described their perceptions and experiences.

Data Analysis

The CQR approach suggests the formation of a diverse research team in order to mitigate bias within individual team members.[141] Three of the authors with varying levels of expertise and backgrounds composed the primary research team for this analysis, and an individual (not an author) with extensive CQR experience served as the external auditor to ensure the data were represented appropriately and accurately by the primary research team.

CQR involves a multi-step consensual data analysis to provide credibility throughout this process. The first three transcripts to return from member checking were reviewed independently by all members of the research team to begin identifying core ideas. After each member had completed this step, we came together to discuss our independent analysis and began constructing an initial codebook. This initial codebook was then used by each researcher to code one of the originally reviewed transcripts along with two new transcripts to continue to refine the codebook by specifically identifying themes and subthemes. Full consensus could not be reached at this point, so another round of coding using the refined codebook was performed on one of the original transcripts and two new transcripts which resulted in consensus of the final codebook. It was at this point that data saturation was also confirmed. Each researcher then coded all transcripts using the final codebook and met over the span of a month to discuss and reach consensus on all coded transcripts and to ensure no new data needed to be collected.[141] At the completion of this step, the external auditor was contacted and examined two fully coded transcripts and the final consensus codebook with all coded quotes. The auditor included minor feedback regarding naming of categories and themes, and suggested we collapse our third theme into another to better represent participant voice. Lastly, we established the frequency of data presented in each theme. Frequency data is provided in Table 5.3

Table 5.2 Semi-Structured Interview Guide

TSK Belief Statement	Strongly Agree/Agree Questions and Probes	Disagree/Strongly Disagree Questions and Probes
You indicated that you (*level of agreement*) with the statement…		
1. "I am afraid that I might injure myself if I exercise…"	*Can you describe why you are fearful of re-injury to your ankle during exercise.* • *Probe: Can you identify any specific activities that are particularly concerning and why?* • *Probe: How has this fear affected your ability to exercise?*	*Why do you think you are able to exercise without fear of re-injury despite your ankle instabilities?*
2. "If I were to try to overcome it, my pain would increase…"	*Why do you believe your pain would increase if you try to overcome your ankle injury/instability?* • *Probe: How does this belief impact your life and activity participation?*	*Can you please expand on why you disagree with this?*
3. "My body is telling me I have something dangerously wrong…"	*Can you explain what your body is telling you and why you think it is dangerous?*	*Can you describe how you know you don't have something dangerously wrong?*
4. "People aren't taking my medical condition seriously enough…"	*Can you explain why you feel people aren't taking your ankle condition seriously enough?*	*What have people done to relay that they take your ankle condition seriously?*
5. "My injury has put my body at risk for the rest of my life…"	*Please explain how you feel your ankle injury has increased your risk and how it affects your life.*	*Can you please expand on why you disagree with this statement.*
6. "Pain always means I have injured my body…"	*Describe how you know that pain always means you have injured your body.*	*If you are not using pain as an indicator of an injury – tell us how you know that you have injured your body.*
7. "Simply being careful that I do not make any unnecessary movements is the safest thing I can do to prevent my injury from worsening…"	*Why do you think that being careful to avoid making unnecessary movements is the safest thing to prevent your injury from worsening?* • *Probe: What movements would you consider "unnecessary" and therefore, try to avoid?*	*What do you feel is the safest thing you can do to prevent your injury from worsening?*
8. "I wouldn't have this much pain if there wasn't something potentially dangerous going on in my body…"	*Can you explain your pain level/intensity and why you think it is dangerous.*	*Can you describe any pain that you are having, and how you know that it is not dangerous.* • *Probe: If you don't have pain – does the absence of pain indicate that nothing is wrong in your body? Why or why not.*

Table 5.2 Continued

9.	"Pain lets me know when to stop exercising so that I don't injure myself…"	*Describe the specific characteristics of pain (type, level, location) that would alert you to stop exercising.* • *Probe: Why do you feel that stopping the activity would prevent you from injuring yourself?*	*Tell me why pain would not alert you to stop exercising.* • *Probe: What would be an indicator to you that you should stop exercising to prevent injury, if anything?*
10.	"I can't do all the things normal people do because it's easy for me to get injured…"	*Why do you feel you are more susceptible to injury than others? What types of things are you limiting based on this belief? How does that make you feel?*	*Can you please expand on why you disagree with this?*
11.	"No one should have to exercise when he/she is in pain…"	*Why do you believe pain should be a limiting factor to participating in exercise?* • *Probe: Do you avoid any exercise or activity because of actual or anticipated pain?*	*Why do you believe pain should not be a limiting factor to participating in exercise?*

Table 5.3 Frequencies of themes, categories, and sub-themes

Theme	Category and Sub-Theme	Frequency of Code Use	Number of Participant Cases
Pain and Injury-Related Fear	Cause of Pain and Fear	124	9
	Severity of Pain and Injury	91	9
	Pain Statements	*54*	*9*
	Consequences of Pain and Injury	*37*	*9*
	Behavioral Response	166	9
	Activity Alteration	*136*	*9*
	Planning Behavior	*30*	*8*
Assessment and Impact of CAI	Self-Assessment	160	9
	Response from Others	32	9
	Positive Outlook	118	9

5.3 Results

Data from 9 participants with CAI were available for data analysis. Seven of our 9 participants identified as female, whereas, the remaining two identified as male. Participant ages ranged from 23-35, with an average age of 31 years old. All of our participants reported regular participation in physical activity ranging from low to high exertion, with level 4 (aerobic exercise for 1-3 hours/week) as the most commonly reported activity level. Our participants' TSK-11 scores ranged from 22-31, with an average score of 26.4. These scores yielded a range of agreement percentages from 36.36%-81.82%, yielding an average of 50.5% agreeance. Participant characteristics and PRO data are presented in Table 5.1.

Two major themes were identified from the interview data including perceptions and influence of pain and injury-related fear, and assessment of their condition (Table 5.3). These themes and related sub-themes are represented by supporting quotes from our participants.

Perceptions and Influence of Pain and Injury-Related Fear

Within the theme of perceptions and influence of pain and injury-related fear, three categories emerged from the interviews including cause of pain and fear, severity of pain and injury, and behavioral response. All participants were represented within each of the three categories.

Cause of Pain and Fear. Six out of nine participants had indicated on the TSK that they were fearful of injuring their ankle during exercise. However, during the interviews, all nine participants identified that they did have concerns about reinjuring themselves, or experiencing instability or pain in their ankle during certain activities. When participants were asked why they feared injuring themselves when exercising four individuals described single past experiences of pain and injury that were perceived as severe and served as an anchor for their fears. For example, Melissa said:

> *I injured my ankle during exercise to begin with. In 2016 I injured my left ankle - so I've had ankle sprains throughout my life, but nothing to the magnitude I had in 2016…I stepped off a 20 inch box onto the ground in the middle of a workout and…I double sprained my ankle with a grade three sprain on both sides tearing all ligaments, tendons, etc…I was on crutches for seven weeks and a boot and during that time, I became pregnant. And because of…how your blood increases in volume when you're pregnant and being immobilized I developed blood clots and now I have lots of forever elements after this one simple injury.*

The other two individuals who specifically agreed that they had fear of re-injury while exercising on the TSK-11 described experiencing more regular instances of pain, instability, and/or injury. Jessica stated “I just know how frequently and how easy it happens.” Elizabeth similarly described:

> *Often during exercise I have instances where my ankles will roll unexpectedly, just in basic movements like a lot of side to side movements or any kind of uneven ground…as soon as there's like hills or trails or uneven pavement kind of in the mix then I, I think it's part of its worry and part of it just is I have like chronic weakness in my ankles.*

Although not in direct response to our initial question regarding why individuals feared injury during activity, similar sentiment was echoed by others in that their ongoing instabilities were reasoning for their increased concerns. Phil stated:

> *There is pain associated with the instability moments, but it's because I roll my ankles when I'm doing a specific activity, so like general exercise activity, there's not [pain], but there's that risk of rolling.*

Participants also described feeling that because of their instability and ankle sprain history, they were more vulnerable or susceptible to injury as Jessica and Megan describe:

> *Just because it's so easy for me to sprain my ankle or just even hurt it that it's constantly there. I don't know how else, or what else to kind of say about it but it's always [um] there ready, I feel like that I could potentially just hurt it again… And so it's always just in the back of my mind that, oh, you might get injured.* -Jessica

> *I know that my ankle is not as stable as other people's because, like, I can bend it in really weird ways that other people are like you shouldn't be able to do that so I'm afraid that, like because of that, even though it doesn't hurt that it would go to a point where would get hurt. Because it doesn't have those like things that are supposed to keep it in place.* -Megan

Our participants injury-related fear and pain experiences were described as being task- or activity-specific and not necessarily generalized to all exercise or movements. All participants described a variety of activities that were associated with actual or anticipated pain and/or injury. Eight participants described functional activities such as jumping, landing, direction change, or single-leg activities as concerning. Sarah stated her concerns with jumping and landing, " I guess again going back to like a jumping movement. I feel very unstable and like it would be really easy for me to roll my ankle doing such a thing." Interestingly, both of our male participants specifically described that the actual jumping and landing task was not worrisome, but specifically performing those tasks near others was their main cause of concern, for example, Luke described his experience relative to playing basketball, "Rebounds with contenders. Okay,

that's the main thing that I worry about just like jumping up with others and then landing in the same vicinity as them."

All nine of our participants described either running and hiking as other physical activities in which they experience pain or anticipate pain and injury. Amber describes having concerns when hiking, "Because usually I roll my ankle at least twice, while I'm hiking. It happens and I'm gonna fall at some point." Specifically noted to be reason for pain or fear during these activities seemed to be the increased risk of injury on uneven terrain as Tiffany describes, "I know that's [running on uneven terrain] when it is most likely going to happen the most or has like the highest risk versus just running on like the road or sidewalk, or something," and the length of the run as Amber states, "[Um] typically it's only for the long run, so usually I can run one to five miles and be fine but anything after that it hurts…"

As for daily activities that were mentioned as cause of pain or fear, participants mostly described walking or standing for long periods of time, or again walking or navigating uneven or unpredictable terrains:

> *And so things like walking on uneven surfaces is something that just causes me to sprain my ankle. Here in Michigan, we have lots of uneven surfaces, especially in the winter time when snow and just the sidewalk buckles or starts to crack. I literally just sprained my ankle walking down the driveway falling on a crack… I definitely like feel like my body tensing up when like it's slippery or when there's ice or something that um, it's there.* -Jessica

> *Walking. I have increased pain in terms of extensive walking… [when traveling] It gets stiff and the stiffness causes discomfort… I would say the largest impact is when I'm doing like walking at Disney world, or walking at a fair, or walking at like hiking or anything that takes lots of…You know it's lots of impact on the leg.* -Melissa

> *Like even sometimes walking so you know when you of course I'm at home, you know I mainly just wear tennis shoes and things like that, but it seems like when I go on vacations, especially in the summer it's warm you want to wear sandals things like that and not necessarily I don't - I never wear flip flops because I can't; but like say if I wanted to wear flat sandals or…I don't know what tom's or just anything like that I am scared because usually by the end of the day, my ankle and my foot starts hurting*

> *because where I'm flat footed in that flat shoe so sometimes even walking sometimes it bothers it.* -Amber

Severity of Pain and Injury. Participants' pain and injury perceptions and behaviors went beyond just the fear that they could or do experience injury and pain, but also seemed to be related to perceived levels of severity regarding pain and injury. Sub-themes identified in this category were pain statements and consequences of pain and injury.

The threat value one gives to pain is inherently weaved into the kinesiophobia construct and thus was at the center of some of our interview questions. However, as pain is subjective to one's own experience it is hard to discern an individual's true beliefs, and so we coded statements that were related to pain as "pain statements" and included them as they likely inform participants' perceptions and behaviors regarding injury and fear of injury. Participants pain statements included pain being described as a symptom or signal of injury like Jessica states, "I just feel like in the back of your head pain is a way of telling your body that something is wrong. I think that's what I've been drilled from such an early age…" and also describe experiences or knowledge regarding varying levels of pain and that it does not always equate to an injury.

> *I mean for the most part, it does [pain signals injury]. Obviously pains can be good, too, but when it comes to like the ankle I feel like pain is not necessarily a good thing, just because that means something has been aggravated or pushed further than it should type thing.* -Tiffany

> *Pain is definitely used as an indicator [of injury] at times, but there's also just naturally, your body will feel pain sensation when you're doing something strenuous… I can have ankle pain and not have what I think is an injury …in an alignment sense I can have pain with that but it not be something I need to do anything about… I feel like working out is painful, but it's not like I'm hurt.* -Elizabeth

Pain statements made by our participants also spoke to how pain is one piece of information that they use to guide their activity and movement decisions. Luke describes, "It's [pain] a very good indicator to stop because, number one, like you could further complicate

whatever is happening." These statements, again, reflect the subjective nature of "pain" as described by Megan and Tiffany:

> *I think discomfort vs pain are two different things, and so, when you're like truly in pain [um] like it is your body like telling you that you need to stop doing whatever you're doing, I think that the way that we perceive discomfort sometimes makes us think that we're in pain and that's why we - Like some people will be like Oh, this is painful, I have to stop that kind of thing so but like true genuine pain is our body like really telling us that like something is not right…* -Megan

> *It depends on like the level of soreness I guess you could say. Like you know if it's like a one or two like mild soreness, then I would probably just push through it and run, but if it was like higher you know, like maybe - Four or five soreness I may like just limit what I'm doing or do something else cardio-wise instead…Like I said, I think it's more based on the pain. Obviously soreness versus like a sharp pain, then I would probably stop what I was doing it that happened during that activity, then I would, you know, stop just because that's not a normal pain that I typically feel in the ankle.* -Tiffany

Participants also perceived severity of pain and injury by describing consequences of ankle pain and injury. Often these statements were related to why they are fearful of injury and/or given as reasoning for why they change their activity behavior. Participants described consequences of injury in regards to physical ailments, like Jessica who stated, "But I think like when it happens, it's like a 10 like I am like in tears, ice, like all I can do is just lay down. All I do is think about it." Consequences were also described as secondary issues that have followed injury or could arise in the future:

> *I think just knowing that like a more serious injury could come out of it. [Um] I have had some…other like calf injuries related to that same ankle and I think it's always lateral calf, so I think something to do with like the muscle weakness in that ankle* -Tiffany
> *And so, whenever my - the ankle flares up, then I start having the pain, you know, on the top of my foot and it just throbs. And so I kind of get that fear that, oh no is this going, is it going to turn into a stress fracture? am I going to be the boot? am I going to not be able to move around for six weeks? So that's - I get that fear.* -Amber

Participants also described how an injury would affect their movement or activity, or as reasoning for why they changed their movement behavior, and also described how an injury would affect other aspects of their life:

> *Exercise is like a really big part of not just like my life like something that I enjoy but, like my mental health and so for me, I know, like if that was taken away from me for any part of time that it would really, really, really affect not just like Oh, I might gain weight or anything like that, just like. It would affect my whole life…if I had a major ankle injury that surgery could be a possibility being booted could be a possibility and it's just one of those things where it's like you know….even if it's a small chance that possibly happening and taking all of those things away from me, even for a limited amount of time it would like really derail those six to eight weeks or whatever it was.* -Megan

> *Since I am married with two kids, I don't because of that. I don't want to roll my ankle and be out for work and we don't get paid – you get a couple weeks of PTO and that's it, so it's a lot of those things.* -Phil

Behavioral Response. All of our participants described varying their activity behaviors based on their concerns regarding pain or their injury-related fears. Two sub-themes emerged including activity alteration and planning behavior. Participants generally used terms like cautious or careful to describe their approach to activity and movement. For example, Jessica stated "I think I'm more cautious about what it is that I'm going to be doing." Melissa shared similar sentiment "I am more cautious in how I - how I do everything with that foot." Participants also shared that being cautious or careful is one way they feel they can avoid injury. Elizabeth stated, "I do find that I can totally avoid injury if I'm really careful with my movement." Tiffany similarly stated:

> *I mean because of knowing like how it happens, I think, just being cautious and you know careful and doing what I need to, to like not have it happen again type thing is probably, like I said, why you just want to be careful.*

Participants shared how they are cautious in different ways. All but one of our participants described simply avoiding movements or environments in which they feel susceptible to injury. For example Sarah said, "I mean, again, there's things that I don't do because I want to avoid. Like, I won't jump very high and I won't run." Luke and Amber describe their avoidance strategies:

> *The giving way or rolling your ankle. It's happened a lot I guess within just playing sport…So it's something that I look out for, or try to like avoid as much as possible… So with that, meaning like not trying to do those motions that put me in that situation.* -Luke

> *Maybe if I'm walking and you need to you can either jump down, you know from where that is or you could do five other steps to move around that, I would always do the five other steps instead of like jumping down.* -Amber

Although avoidant behavior was described by our participants for certain tasks or movements, they shared that they did not avoid activity altogether, but instead described making modifications to their activity or movements to continue to participate but in a way that felt safe. Sarah stated, "Again doing modifications to not completely stop the activity, but to do it in such a way that it's safer or at least feels safer." Our participants describe that this sometimes meant changing how they play or participate. Luke stated, "I would still engage in the exercise/sport, but not necessarily the movement. Or would like try to not put myself in that situation. [For example] So I just wouldn't go for rebounds as often as I would." Others describe changing their awareness or focus of attention while performing the activity, changing their effort or intensity, or their volume of activity:

> *When I'm doing lunges, you know, I really have to think about the way that I'm placing my foot down, you know and my ankle, because sometimes especially that right ankle - like if my leg is behind my body it wants to kind of roll to the side. So I just, you know, I think about things like that you know as far as making sure that my foot placement is where it needs to be, and that - you know, I have to make sure my knee and everything is in alignment otherwise my ankle will compensate. Kind of the same way, probably like when I go hiking I do like shorter steps you know, so I like short quick steps, because that way I'm not on that one foot too long – that kind of thing.* -Amber

> *I really only pay attention when I'm like doing exercise like if I was walking I don't really pay attention as much , but I think just exercise in general I just if I'm running with no brace just I'll be like more cautious of the sidewalk like if there's raised or cracks or you know uneven surface there like I'll just be more cautious and try to pay attention, I guess, you could say to like my footing.* -Tiffany

> *…Like I still want to play and have fun , but like, I tone it back a little bit versus you know jumping as high as I can. Like I said, I love to play volleyball – I love to block, I love to spike, but there's just times, where it's just like because of that, I am not going to*

> *go all out because I would rather play next week also versus rolling my ankle and not being able to play , so I tend to have that guarded mentality just so I can continue to play, and do the things I like to do. So, I don't do it as hard as I would have if I was fearless in that and it's primarily because my ankles.* -Phil

This was also described as modifying or exchanging the risky activity for something that is similar or in which the individual felt was less risky or they were more confident with as Megan states, "I feel like I do like avoid some things like I'll choose different movements or things like that that I'm just like more comfortable with." Sarah similarly describes:

> *I'm really careful and will modify like as soon as I read that other people were not actually hopping over their dumbbells, but like behind their dumbbells with like that visual of how high they have to go – I'm like, oh, I'm going to start doing that. So I'll still do workouts, but maybe modify some certain aspects of it.*

The second sub-theme was related to planning behaviors. Participants shared that sometimes they also employed pre-emptive behaviors in anticipation of injury, or in the prevention of injury. Participants described planning their activities specifically to avoid or prevent risk, ensuring they had the appropriate means for caring for an injury if one were to happen, and/or employing the use of specific footwear or bracing to feel safe:

> *Like when I hike, I do pick trails that have either like, you know, more like round about. Or it's just like flat or I mean, or just say you and I were I intentionally wear high shoes or high socks or stability for my ankles, because it's the right thing for me.* -Elizabeth
> *I think just picking out my outfit for the day. And what shoes I'm going to wear and things like that are all constantly in the back of my head. Um, making sure that I have ibuprofen in my car in case something happens.* -Jessica

> *Or like if I'm going to try to run again a lot, I need to make sure that I have new shoes, and that I wear my ankle brace or ice right afterwards or do my like ankle stability exercises and things like that I guess.* -Amber

> *The easiest way to explain it is like running on the uneven terrain, like, I know I have to be careful, because even just walking that like - You don't know, like I said, roots or you know a little divots, so just being cautious of that and careful so that's why I just wear the brace to be like extra cautious.* -Tiffany

Assessment of their Condition

Within the assessment of their condition theme, three categories emerged from the interviews including self-assessment, response from others, and positive outlook. All participants were represented within each of the three categories.

Self-Assessment. Participants shared their assessment of their injuries and current condition, as well as their assessment of the care strategies that have been used on their ankle. Regarding their current condition, participants shared some issues that they still currently deal with regarding symptoms like instability, pain, swelling, and scar tissue, or regarding function. Melissa said, “I have very weak ankles, I have very limited calf mobility, now. I had better calf mobility before my injury…” and also how this affects her, “I can't do the same level of movements that I did pre-injury.” Most participants, even when they disclosed having bouts of discomfort or pain, discounted it as described by Melissa and Megan:

> *The level of pain that I was in when the injury happened versus the level of pain that I feel on a more regular but still intermittent basis - they're just night and day they're nowhere near the same threshold of pain. It was unbearable pain when the injury occurred and even through the first couple of weeks of recovery, to the point where I feel a little to no pain, unless I have overused it and, in that level of pain is still just pebbles compared to the mountain of pain that I felt. It’s – It’s just not comparable.* -Melissa

> *I get really bad pains like through my ankle and then like into my foot kind of thing, and I know that, like… It’s probably not like it's not like surgery required kind of thing but it's also one of those things where it's like [um] my ankle does not work, the same way that other people who maybe didn't have an injury.* -Megan

When participants were asked about their perceived level of seriousness of the condition, or whether their ankle put them at risk for life, participants typically described viewing CAI as annoying or something they have to deal with. Phil described “it's just annoying. It’s an annoying, stupid, thing that I have to deal with.” But, generally participants believed that their condition was not serious despite some of their ongoing issues:

> *I would say it's in the middle, it's, it's probably not serious. I would say closer to not serious. I mean, I do have, like, swelling, like always my ankles are swollen for life from injuries and crack - they crack constantly everywhere I walk - I like tiptoe around the baby's crib because they're like, they crack and I think that that's probably not like ideal, but it's not something that's going, doesn't.. you know, day to day, it doesn't affect me and could it be better? Yes, but it could be super worse.* -Elizabeth

> *It's more annoying to me than anything, just because I feel like it's something that I'm have to deal with forever, so I don't think there is a quick fix for it , though, but so I think it's more - It just annoys me more than anything, and, of course, when it starts hurting a lot, then I think it's serious, but then other times I just kind of ignore it and think it'll be fine. I'll just do this or that, and you know it should get better.* -Amber

Participants described the information that led them to believing that their condition was not serious. For example pain, function, and life impact were noted as ways participants gauged the level of seriousness. Luke said, "The absence of pain indicates there's nothing wrong with my ankle and then that could also be seen through like, for the most part, my participation in activities, exercises." Megan and Phil describe similar assessments:

> *I don't have like mobility issues… overall, like, I feel like my life experience like when I go about my day to day is not like diminished by the pain level that I'm in or the my mobility issues because of my ankle.* -Megan

> *So currently I have not done anything that makes me feel like I'm going to be impacted for my life, since I don't have chronic pain, or anything. As I said, it's more - has changed my lifestyle habits, but I don't think I've done it, I don't think so far, I don't think, yet it has caused anything that I'm like - Oh no that's gonna affect me when I'm 70. I think there's back pain, there is that stuff that I'm more worried about then like the general instability of my ankle as an older individual.* -Phil

Tiffany described knowing that ankle instability may be more serious for others, but in her case it was not, "So, like mine doesn't hinder me in any way really so like I don't necessarily think it's serious in my standpoint but, like in other people standpoints, it could be serious, it could hinder them." Participants also described that their ability to manage their condition was used to guide their judgement regarding the seriousness of their condition as Phil and Sarah state:

> *I think it's not very serious, as long as I choose activities… if I was doing everything without caution, it would be a lot more serious, but I think it's something that can be managed with the right amount of caution and choice of activities.* -Phil

> *I mean, I would say it's a minor injury, it's nothing necessarily to, like, you know, have anyone hub-ub about um Yeah, minor. It's, um, it's a minor, minor thing. It's true. Like, you know, rest, ice, compression, whatever is usually enough to fix it.* -Sarah

Participants also displayed some ambivalence in how they viewed their condition, its effects on their life, and whether it could be modified. For example, Amber described that because she does not have constant pain, she is able to put it out of her mind although admitted she still knows something is wrong:

> *Out of sight out of mind, I guess, I don't know I mean, I know that there's always something wrong. Because I was thinking about that I guess you always know there's something wrong, otherwise you wouldn't try to change your movements, you know or think about your movements, I feel like if you didn't have any ankle problems you, you would never think about those things, so I guess I know there's always something wrong, but if it's not hurting me then I'm like that's okay today.*

Jessica discussed how her instability has persisted despite making efforts to change it "I've done numerous things about it: PT, supports, and it's just constantly there." But later indicated that she is still hopeful that her condition can change:

> *I know that there are things that you can do to overcome it. And so I don't feel like for the rest of my life, I have to endure these things. I haven't had an ankle sprain, knock on wood, in quite a bit. And so um do I feel like it's something that it's going to inhibit me forever? Hopefully not… Yea, so, I just, I don't feel like it's gonna affect me forever.*

All participants described care strategies that they have used for their ankle and their assessment of these strategies. Some of these were self-driven and some were related to care received from others. There were varying experiences and encounters with health care providers. For example, Amber describes her positive experience, "they [my healthcare providers] made me rest, and so it got better at the time, so I felt like they listened to me." Melissa on the other hand, described how she went to multiple doctors before she felt like she received the appropriate care:

> *So I had seen two different offices that basically brushed it off as you'll be fine, this is no big deal [um] and then, finally, my own [primary care] doctor took it seriously…and then I could actually start seeing the correct type of orthopedic doctor and get the services, I needed to recover…*

Despite the variance in experiences, participants described perceived benefit from either care received from others or self-driven care, and some discrepancies were noted. For example, Jessica described rest as being helpful after injury, "I know my body and I know what usually happens after… usually getting off of it and resting it is something that helps me a lot." Whereas Phil described working through it was beneficial:

> *I feel that a light ankle sprain a lot of times for myself actually improves with pushing through the little bit of the pain and kind of getting the blood flow and everything that - I've had better success through that.*

All of the participants who described experiences with rehabilitation, perceived them as beneficial. Megan specifically describes how after doing rehabilitation, she wished she would have done it sooner:

> *I was always one of those people that was like - I'm not - like PT doesn't work, I'm not going to do PT - that's ridiculous. And like I've been through PT now and I was like oh my gosh my - my ankle probably would have been so much better if I would have just done this in high school when I when I originally like hurt it. So it's, it's funny, they've always tried to like present the plan to me it was me being a bad patient.*

Interestingly, Tiffany and Phil are both rehabilitation professionals and both shared perceived benefit of rehabilitation, but also noted that it may not completely "fix" their instability:

> *I just think, from my experience, I've done plenty of ankle stabilization exercises that do help, and I've done… yeah mostly ankle stabilization type things/exercises - of the all the angles and doing the thera band – and I think that's helped, but it doesn't prevent the unloaded, or like the.. If all those muscles aren't active at the same time, the joint's still loose.* -Phil

Response from Others. Our participants often described interactions with others throughout their experiences including healthcare providers, coaches, family, and peers and

shared their perception of the response from others regarding their injury or condition. These interactions likely inform their current perceptions regarding their injuries and condition.

Luke was the only participant we interviewed who did not seek care from a healthcare provider after his self-described worst ankle injury and said, “I think it's like more of like, I don't want to do it [seek medical care] knowing that my parents could find out… they’ll get mad.” The other eight participants described interacting with urgent care, primary care, or orthopedic physicians, athletic trainers, and physical therapists. Participants described different responses to their injuries included imaging, immobilization, rest, and rehabilitation. Two of our participants also described other complaints that led them to pursuing rehabilitation that led to focusing on their ankle:

> *My doctors were saying, that they believe that instability in my knee was because of the instability in my ankle and then I was like compensating for things so it [pursuing physical therapy] was kind of for both ankle and knee.* -Megan

> *I actually went to see the physical therapist for my back…and he said. You know, because …everything's on the right side, I think that your - your back and your hip is bothering you because of your ankle instability, so this was before even I saw the sports medicine doctor, so he was kind of on it. And so, he kind of taught me some different exercises and so really me strengthening my, my hip and my glutes kind of helps my ankle more too.* -Amber

One of the questions on the TSK-11 relates to how serious the individual feels others are taking their injury or condition. Regarding healthcare providers, some participants felt as though they did not take their ankle injuries seriously, like Sarah:

> *Anytime that I've had a sprain or like brought it up to a doctor. They were just like rest - like there was no real like - we should look into this… I would have to say that it's not being taken seriously.*

Melissa interacted with two doctors before feeling as though her injury was taken seriously. She first describes:

> *I went to our urgent care, which is our walk in for our doctor's practice, and they evaluated and slapped on this chintzy little boot that didn't do anything, it was awful and they said okay by Monday you'll be fine.*

When she continued to have pain she went for another opinion:

> *I went to the orthopedic walk in where they said okay here's a more stabilizing boot, which is the one I wore for seven weeks and they said. 'Here, some anti-inflammatories. In a week you'll be fine.'*

Melissa described that her pain continued, and eventually connected with her primary care doctor who happened to go to the same gym where she felt her injury was finally taken seriously:

> *If it weren't for my primary doctor - forget the doctors who have actually seen my ankle, but if it weren't for her, she ordered an MRI and I got that done and then that's when we realized the extent of all the damage that had been done.*

On the other hand, Megan admitted that her doctors took her injury seriously, but that she did not, "Okay, so when I originally hurt it… they like were …I felt like I didn't take it as seriously as they did - and they were like we need to do PT, we need to do all this stuff." Regarding that specific question, participants also included parents, coaches, and peers into who they described as "others." Tiffany describes how her athletic trainer and coaches were both responsive to her injuries and in the prevention of ankle injury:

> *During high school…I was practicing, rolled one, like or you know sprained one and then like the very next day sprained other, so then the athletic trainer was like well you know you need to do something about it, because obviously there's, a problem… then like after that we had to wear braces like playing basketball when I was on another team, and we had to do like…ankle stability exercises before practice and then wear our braces, so I think it was just like part of my whole high school career was just doing rehab to strengthen because the coaches knew the severity too of ankle sprain and loss of time, I think is what they were worried about from sport.*

Phil described his coaches in high school also told him to "wear the braces and everything to make sure you're not. Same thing with like basketball, is wear the high tops and everything…" but perceived that this response was not serious, "I think they also interpreted as

it's not that big of a deal, just do this to compensate for it or to adapt with it." Elizabeth mentioned receiving care from her athletic trainer, coaches, and others and said, "I've never felt like people thought I was making it up or it wasn't a factor..." Whereas, Phil describes, "My wife thinks I am a baby, when I roll my ankles she was like you're fine - that type of thing. Like when I rolled it on the street and had like pretty solid swollen ankles she was just like yeah.." Jessica shared a similar sentiment from her family, "Like my family says, like, 'Oh, you did it again' type of thing."

Positive Outlook. Throughout our interviews, participants shared how CAI and their past ankle injuries affect their life in negative ways. Despite this, individuals generally displayed attitudes and cognitions that provided them a positive outlook regarding their condition. As mentioned, although participants did describe fears regarding certain movements and activities, it didn't stop them from remaining active. Sarah says, "I guess that kind of goes along with what I said about being stubborn and like still doing the workout anyway. Like, I guess I'm not so fearful of re injury that I'm going to stop moving." All of our participants described specific activities or tasks in which they felt confident in their abilities, or in their overall ability:

> *So I feel that I can do everything that someone with no injury can do… I would say, like people without ankle injuries… that's like who I'm comparing to – you know like someone that doesn't have injuries that they have to worry about or pain type thing.* -Tiffany

Two participants described a gradual process that led to improving their confidence during exercise that had previously been associated with pain or fear:

> *So, I just.. I use it more. I work out more regularly and that helps just keeping it moving better. I guess my confidence, the more I do the more confident I feel… you know a little bit at a time builds up to be a lot of time.* -Melissa

> *I think some of it was just like the gradual build up that I allowed myself to have so like allowing myself to go back to working with much lighter weights than maybe I knew I was capable of, and so like gradually building back up to it and, like being able to like expose myself to these things like repeatedly without pain um and then just knowing like*

> *if it did hurt, I was allowed to stop. Like I had already agreed ahead of time like if something hurts that I'm not continuing that - that helped a lot.* -Megan

Participants described having to adjust or alter their activities, and often make modifications due to their ankle in order to continue to exercise. However, most participants were open to the idea of finding new ways to move and be active as Elizabeth describes, "I learned to like other types of exercise like even we got a peloton like.. That's no risk." Participants also shared that they didn't find that the modifications they had to make were inhibiting the intended goal of the activity. For example, Sarah said:

> *Hopping over a dumbbell like taking the dumbbell out of that equation makes it safer and I'm probably not jumping as high- but still getting cardio in so I think I'm still meeting an objective, but like taking a factor of it out that would make it dangerous to me…if the stimulus is to, like, you know, intend to be jumping and like increase your heart rate, I'm doing that.* -Sarah

Some participants described feeling that they have accepted their ankle condition for what it is. Tiffany says:

> *I would say it's just been something I've dealt with for a long time, so it doesn't like – if it rolls or anything like that it doesn't bother me like type thing. I just go on with what I'm doing because I might just be walking down the sidewalk and it could roll or you know running whichever so it's just part of life, type thing.*

Amber and Phil also described that due to their priorities, goals, and values, their condition did not affect the quality of their life:

> *I don't feel like I can't play with my kids or do my job effectively and for the most part, like the type of exercise that I choose to do I don't have issues with it, and again, some of that is just because, like my goals are different.* -Amber

> *Just at this stage of my life.. if you asked me 10 years ago, I would have a whole different story…there's things that I needed to accomplish back then, in my male ego that I… Now that I'm like- I'm in a different stage of life that those activities are not as important to me…Can I get up and down with my girls and can I hold them in my arms – those are the things I got to make sure I can still do.* -Phil

5.4 Discussion

This study is the first to explore the injury-related fear construct in individuals with CAI using a qualitative approach. We chose to explore this by trying to understand the experiences and factors that patients used to assign their agreement level to the TSK-11 statements, as this is the most commonly used PRO for assessing injury-related fear in this population. Our participants' level of kinesiophobia, as measured by the TSK-11 was varied; however, all 9 participants described feeling concerned about their ankle experiencing instability or injury during certain activities. Our results reveal that although some similarities exist, each individual's experience likely contributes to their fear perceptions, behavioral responses, and overall quality of life differently, and thus an individualized care approach is necessary for both understanding and addressing injury-related fear in CAI patients. Additionally, our results support the use of the TSK-11 as a discussion tool for clinicians to begin understanding these individualized experiences in their patients.

Our results also point to some specific areas that may be useful for clinicians to discuss with their patients to understand factors influencing their injury-related fear and how it affects their activity and life, and can inform the best ways of addressing them. Firstly, our results suggest that understanding patients' injury, instability, and pain experiences may be an ideal place to begin. It is well-established that an injury can be a perceived stressor to an individual's life, and there are multiple factors that are thought to interact which result in the cognitive appraisals of the injury experience, and also affect the emotional response that occurs after injury - including a fear response.[145,146] One of these factors is related to the characteristics of the injury, and the specific injury experiences that were mentioned by our participants to cause their injury-related fears were related to the perceived severity of the event or based on an increased

frequency of instability, pain, and injury episodes to their ankle. This is similar to findings from other studies that found that athletes who have experienced major injuries requiring more time-loss, reported greater injury-related fear than those who experienced minor injuries;[147] and those who have experienced multiple ankle sprains reported greater injury-related fear than those who have experienced one ankle sprain.[72] Understanding whether patients' fears are anchored to one injury experience or if they are from ongoing instability and/or painful experiences would be helpful for identifying the major cause of their fear and how generalized it may be across tasks and activities. A lack of support from others is another factor identified to increase perceived stress after injury[145,146,148] and was noted by some of our participants during their past injury experiences as well, and thus may be another important area related to past injury experiences to discuss with patients to understand the stress they have associated with their past ankle injury experiences. Although clinicians are unable to change the past experiences that may have caused or promoted the acquisition of injury-related fear in these patients, they may still be able to reduce the stress associated with their current condition. One qualitative study identified connectedness as a specific factor that patients described as helping them manage their injury-related fear after anterior cruciate ligament reconstruction (ACLR).[149] Specifically regarding the support provided by rehabilitation providers was that they felt valued and understood and felt as though their rehabilitation was individualized to their specific needs.[149] Therefore, if given the opportunity to work with patients with CAI, clinicians should aim to provide support and assist them in reducing their injury-related fears using multiple strategies on an individualized basis. Some potential strategies, based on our results, will be discussed further.

Our participants described that their injury, instability, and pain experiences also provided them with evidence towards believing they were susceptible or vulnerable to pain and

re-injury of their ankle, as well as perceiving that future pain and injury would be associated with harm or unwanted consequences. Perceived susceptibility and severity are two specific factors identified to affect how an individual appraises a threat, with greater perceived threat of injury contributing to greater level of fear regarding re-injury.[150,151] Therefore, clinicians should also attempt to gauge patients' beliefs regarding their susceptibility to ankle re-injury as well as their perceived ramifications or harm associated with future injury as these cognitions may underlie some of the behaviors associated with their fear, and may need to be challenged during their rehabilitation. One potential strategy that may be useful for altering cognitions and may also help alleviate the fear associated with threatening situations, is imagery.[152] Of specific interest is the evidence that using imagery scripts producing a challenge-appraisal versus a threat-appraisal state during a stressful scenario resulted in more positive interpretations and more adaptive coping responses in athletes.[153] Perhaps similar strategies can be used in patients with CAI, using imagery to manipulate the threat-appraisal state during a situation in which they feel susceptible to re-injury, by introducing scripts that promote challenge-appraisal states by enhancing their self-efficacy and control within the scenario.[153] Imagery and relaxation techniques have also been shown to aid in typical rehabilitation protocols enhancing both physical and psychological outcomes in ACLR patients.[154] Together, these studies support the use of these techniques as potential strategies that could assist patients with CAI reporting injury-related fears.

Direct experiences of instability, pain, and injury also informed the tasks, activities, and situations that our participants identified as a cause of fear and pain and subsequent avoidant behavior. This finding supports the notion that fear acquisition and learning develops via classical conditioning.[97] The mechanism of a lateral ankle sprain likely serves as a proprioceptive cue that was followed by pain and/or injury, and therefore the situation in which it occurred is

then associated with injury and pain. Therefore, any activity or environment in which individuals experience subsequent feeling of their ankle giving way or pain, their body may associate these new movement experiences with pain and injury and learn to fear them as well. Direct experiences are noted to enhance fear learning,[97] which is why understanding actual experiences of pain, injury, and instability would certainly be useful for beginning to identify the specific activities and movements patients have learned to fear based on these experiences. We also found that in some cases, participants identified activities or situations based on the anticipation of pain or injury without ever having directly experienced pain or injury during the activity. This is supported in that fear learning can also generalize to situations that an individual perceives as similar to a learned fear situation,[155] and so clinicians should aim to identify all situations their patient associates with pain or fear, regardless of past injury history. This can be accomplished in various ways, but in low back patients and in patients after undergoing anterior cruciate ligament reconstruction (ACLR), Photographic Series of Daily Activities[156] and Photographic Series of Sport Situations[157] have demonstrated to be helpful tools for patients to identify fear invoking activities and sports situations. Future researchers could test the use of these tools, or develop specific photo series for the ankle sprain population, as this may be helpful to gain understanding of patients' situation-specific fears and can also be used to better inform individualized intervention strategies.

Based on our participants demonstrating situation-specific injury-related fear, another strategy that may be beneficial for reducing these fears in patients with CAI is graded exposure therapy. Graded exposure therapy is individualized to the patient and combines cognitive and behavioral approaches with progressions in activity to build up activity tolerance.[158] The goal with this therapy comes from the fear extinction literature which describes that exposure to the

activity or movement without having the unconditioned stimulus, pain and injury in this case, has shown to lead to a dampening of the excitatory association by creating inhibitory associations that reduce fear and the generalization of fear.[97] Therefore, movements and/or activities which are identified as feared activities by the patients would be included in these progressive rehabilitation programs to begin to dissociate the previous relationships between the activity and injury starting with the least feared.[158] Two of our participants specifically described gaining confidence in previously feared activities through progressive and gradual exposure to them, and reported now being able to perform those activities without problem. These two accounts, although certainly limited, does support the idea that this may be a ripe area for exploring within CAI patients regarding their situation-specific fears. Graded exposure has been investigated by others and shows promise for reducing fears and increasing function in patients,[159-163] although results seem to be maximized when they are used in combination with other intervention strategies.[161,163] Education to alter pain and injury memories may be helpful in addition to graded exposure,[164] and other psychologically informed intervention strategies mentioned previously such as social support and imagery, as well as goal setting,[165] may also be helpful in relieving fears and anxieties as well as enhancing confidence.[17]

Lastly, our results also support understanding patients' values, goals, and perspectives towards activity. These will help shape the approach for their individualized care plans, and are likely driving factors to the way patient's fears and condition impact their physical activity and quality of life. Our results support that kinesiophobia was not applicable to all physical activities,[164] as our participants were able to maintain levels of physical activity despite reporting specific activity avoidance and alterations. Despite these changes to their activity behavior, almost all of our participants shared that they did not mind having to make these changes.

Furthermore our participants reported that despite ongoing symptoms and issues associated with CAI, they perceived their condition as being a minor annoyance, and not impacting their overall quality of life. Some of our participants, as also described by Filbay et al,[166] shared that it took time to reach levels of acceptance of their condition and the adaptations to their activity, and in some cases changed as their life priorities, goals, and values changed. For example a few of our participants stated that had we asked them these same questions a few years ago, they likely would have felt differently, but due to where they were now or their current goals, they had reached a level of acceptance. It is unknown if the positive outlook of our participants would continue to be seen in patients with CAI that were unable to continue participating in activities of interest or were not open to other modalities of exercise and were not able to accommodate their fears or reach a level of acceptance. Individuals who are similarly aged to our cohort but who report lower levels of activity due to their ankle may be especially important to investigate as increased injury-related fear and lower physical activity levels have shown to be associated with ankle joint degeneration in those with CAI.[103] This also may suggest critical time points in the progression of the condition which could benefit from strategies that assist in the reduction of injury-related fear and participation of physical activity that maintains individuals' quality of life.

Limitations

Our study is not without limitations. One limitation is that we only used the TSK to begin to explore one aspect of injury-related fear construct (kinesiophobia) within the CAI population. We also relied on self-report measures and interviews to guide this study which increase the potential for recall bias from our participants which could affect their recollection of their injury experiences and memories. Additionally, using a qualitative design and small cohort does not

allow for the generalizability of our findings to all patients suffering from CAI. However, further investigations using themes identified within our study to continue to explore this construct within this population would serve useful in expanding our knowledge on potential areas to intervene in order to best enhance continued physical activity and quality of life.

5.5 Conclusions

Our study supports the use of the TSK-11 as a useful means of evaluating injury-related fear in individuals with CAI. However, it emphasizes the importance of discussing patients' injury experiences and factors that influence agreement with TSK-11 statements as this can lead to a deeper understanding of their fear and may reveal specific areas that need to be addressed within their care plan. Specific areas that could be important are related to an individual's perceived susceptibility and severity of future injury, as well as the perceived risk of injury associated with specific tasks and activities, as these may become feared activities in which the patient begins to avoid. Patient values, goals, and perspectives towards activity may also help shape the impact of both injury-related fear and the condition of CAI on their quality of life.

CHAPTER 6

SUMMARY

6.1 Purpose, Aims, and Hypotheses

There were multiple purposes included in this dissertation to further understand injury-related fear in patients with CAI. The first purpose was to systematically review the literature investigating differences in injury-related fears between individuals with and without CAI. The second purpose was to determine if the FAM and its components can be applied to CAI. The third purpose was to explore the perceptions and experiences that underlie elevated levels of injury-related fear in individuals with CAI. These studies were designed to address the following aims and hypotheses:

Aim 1: To examine the extent to which injury-related fear is present in individuals with CAI

Hypothesis 1.1: Individuals with CAI will report greater levels of injury-related fear compared to those without CAI

Hypothesis 1.2: Ankle sprain copers and controls will share similar levels of injury-related fear

Aim 2.1: To examine relationships between injury-related fear and pain catastrophizing beliefs in those with CAI

Hypothesis 2.1: Greater pain catastrophizing beliefs will be related to greater levels of injury-related fear

Aim 2.2: To assess the influence of pain on ankle function and global disability in individuals with CAI

Hypothesis 2.2: Pain presence will explain additional variance beyond reported instability in both ankle function and global disability outcomes

Aim 2.3: To determine the unique role of the cognitive-affective model components in predicting function and disability

Hypothesis 2.3: When controlling for instability and pain, both pain catastrophizing and injury-related fear will uniquely explain additional variance in both function and disability

Aim 3: To explore the perceptions and experiences that influence injury-related fear measured with the Tampa Scale of Kinesiophobia (TSK-11), within the CAI population

6.2 Summary of Findings

The findings from all studies and specific aims are described below:

Aim 1: To examine the extent to which injury-related fear is present in individuals with CAI

Findings: Our hypotheses were supported as moderate to strong evidence was demonstrated supporting heightened levels of injury-related fear assessed with the FABQ and TSK, in individuals with CAI when compared to both COP and CON. Additionally, individuals who have likely recovered after their ankle sprain do not seem to differ from those who have no history of ankle sprain further emphasizing changes to this psychological variable in those who develop chronicity after ankle sprain.

Aim 2.1: To examine relationships between injury-related fear and pain catastrophizing beliefs in those with CAI.

Findings: Our hypothesis was supported as a significant weak positive relationship was found between PCS and TSK-11 scores indicating that greater pain catastrophizing was related to greater injury-related fear.

Aim 2.2: To assess the influence of pain on ankle function and global disability in individuals with CAI

Findings: Our hypothesis was supported as the addition of pain significantly improved both the ankle function and global disability models. Pain accounted for an additional 8.9% of the variance in the Quick-FAAM model, and 6.6% of the variance in the mDPA model above and beyond IdFAI scores. Both perceived instability and pain significantly explained 32.2% and 28.0% of the variance in function and disability outcomes, respectively, and both predictors emerged as unique predictors to the model.

Aim 2.3: To determine the unique role of the cognitive-affective model components in predicting function and disability

Findings: Our hypothesis was supported as the addition of pain catastrophizing and injury-related fear significantly improved both the ankle function and global disability models. The addition of the cognitive-affective outcomes (PCS and TSK) accounted for an additional 16.5% of the variance in Quick-FAAM model, and 16.2% of the variance in mDPA model above and beyond both IdFAI and pain. The final models significantly explained 48.7% and 44.2% of the variance in function and disability outcomes, respectively, and all predictors emerged as unique predictors to the model.

Aim 3: To explore the perceptions and experiences that influence injury-related fear measured with the Tampa Scale of Kinesiophobia (TSK-11), within the CAI population

Findings: Due to the qualitative nature of this study, there was no hypothesis driving this study beyond the stated purpose. Nonetheless, our study findings do support the use of the TSK-11 as a useful means of beginning to identify injury-related fear in individuals with CAI, but supports that understanding the injury experiences and factors that patients use to identify their agreement with statements on the TSK would serve as more beneficial in formulating strategies to reduce these fears.

6.3 Conclusions

This dissertation set out to understand the presentation and impact of injury-related fear in individuals with CAI. Our results demonstrate that the FABQ and TSK are appropriate tools to begin to identify injury-related fear in individuals after ankle sprain and that those who develop chronicity after ankle sprain report higher levels of fear compared to those who recover after injury. Within the CAI population, perceived instability, pain presence, pain catastrophizing, and injury-related fear are related to lower reported ankle function and higher disability. These results continue to support the use of the FAM in understanding CAI, and specifically lend support to the notion that these cognitive-affective outcomes may play a role in the development and continuance of chronicity within these individuals.

Our qualitative findings continue to support the FAM as individuals who reported various levels of injury-related fear described avoiding activities and tasks that they believed would put them at an increased risk of re-injury. Additionally, an individual's perceived susceptibility and severity of future injury may contribute to the magnitude and generalizability of these fears and subsequent avoidant behavior and attitudes. Despite individuals demonstrating injury-related fear and avoidant behavior, patient values, goals, and perspectives towards activity showed that they may help alleviate the overall impact of both injury-related fear and the condition of CAI on patients' physical activity level and quality of life ratings.

In summary, clinicians should consider the use of appropriate PROs in their patient evaluations to begin to identify levels of perceived instability, pain catastrophizing, and injury-related fear after ankle sprain. Additionally, discussing elements of these PROs and specific activities and tasks in which the patient feels increase their risk of re-injury are likely important in identifying areas in which enhancing confidence and reducing fears will be important within

rehabilitation plans. Continued exploration of these concepts is still necessary to expand our knowledge on potential areas to intervene in order to best enhance continued physical activity and quality of life.

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PUBLICATIONS

Published Refereed Manuscripts

1. **Suttmiller, AMB** and McCann, RS. Injury-related Fear in Individuals with Chronic Ankle Instability: A Systematic Review. *Journal of Sport Rehabilitation*. In Press.

2. McCann RS, **Suttmiller AMB**, Gribble PA, Cavallario JM. Athletic Trainers' Methods for Determining Return to Activity for Patients with an Ankle Sprain Part I: Types of Measures Used. *International Journal of Athletic Therapy & Training*. In Press.

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