EVALUATING THE FEASIBILITY OF THE *PACKER MANAGING FATIGUE PROGRAM* IN PARKINSON'S DISEASE: A MIXED METHOD PILOT RANDOMIZED CONTROLLED TRIAL

by

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Dalhousie University is located in Mi'kma'ki, the ancestral and unceded territory of the Mi'kmaq. We are all Treaty people.

DEDICATION PAGE

As I sit here, contemplating my journey, I am reminded of the words of Iran Darroudi, an exceptional artist and thinker who has inspired my way of thinking about the world. Her words have had a profound impact on shaping my perspective on passion and humility. She spoke of how true passion requires us to become nobody, to surrender ourselves to the world, and to embrace our own insignificance and become a new version of ourselves. This is a sentiment that has stayed with me throughout my journey, guiding me through the challenges, the hardships, and the triumphs.

From the first moment that I discovered my passion, I felt as though I had claimed ownership of the world. I believed that everything and everyone belonged to me, and that nothing could stand in the way of my dreams. But as I embarked on this incredible journey, I soon realized that true passion is not about ownership or possession. It is about surrendering oneself to the universe and allowing it to shape us in its own way.
Over the years, I have dedicated myself to this work, pouring all of my energy, all of my heart, and all of my soul into every project that I undertake. And while the road has been long and difficult, I have emerged from it with a renewed sense of purpose, a deeper appreciation for life, and a loving gaze towards the world. This is the essence of love - to become nobody, to let go of our own egos and to allow ourselves to be shaped by the world around us. It is a journey that required me to put down the baggage of sorrows and longings, and to move forward with a sense of purpose and determination. This work reflects all my desires, passions, and the transparency of mirrors, and begins my longer journey moving towards the future. It is a symbol of the journey that has led me to this moment and a starting point for the longer journey that lies ahead.

As I pen these words, my heart swells with gratitude and love for those who have inspired me in my life. To my beloved grandparents, whose love and kindness were a constant source of comfort and inspiration for me, I dedicate this work. Those who I missed to say goodbye to. Though they are no longer with us, their spirit lives on in me, and I hope to make them proud with my achievements.

There is one person who has been my rock, my pillar of strength, and the guiding light in my life - my father. As he battled Parkinson’s disease, his resilience inspired me to make an impact in the field. This work is a testament to my passion and dedication, and to the lessons and love my father instilled in me. His belief in me has been the wind beneath my wings and his unwavering love, the compass that guided me. Dad, I am eternally grateful for your presence in my life and for being my hero.

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ABSTRACT

Management of fatigue has been identified as an unmet need in Parkinson's disease (PD) care. To address this gap, this study evaluated the feasibility of conducting a future full-scale randomized controlled trial (RCT) of the individual version of the *Packer Managing Fatigue program* delivered via videoconference.

The study had two primary objectives: 1) to explore the perspectives of PwPD on the feasibility of the *Packer Managing Fatigue program*; and 2) to evaluate the feasibility of the proposed research protocol and outcome measures for future RCTs. This pilot, mixed-method RCT employed an assessor-blinded, two-arm design. Participants were recruited from Nova Scotia and Ontario and had to meet specific inclusion criteria: self-reported PD diagnosis, severe fatigue, English proficiency, and internet access. A total of 25 participants completed baseline measures. Standardized outcome measures and non-standardized feasibility questionnaires were used to collect quantitative data. Participants in the intervention group, were also invited to take part in a qualitative interview or focus group. Triangulation was carried out during the interpretation phase. Five themes emerged: (1) program is helpful, (2) strengths of the program, (3) areas for improvement, (4) individual online delivery feasible and (5) more support from OT would be helpful. Quantitative findings confirmed feasibility with high ratings on questionnaires and confidence in using learned strategies. Quantitative findings confirmed the feasibility of the program as well. The mixed-design ANOVA demonstrated trends toward significant improvement in occupational satisfaction and small-moderate effect sizes for occupational performance, occupational balance, and reduced motivation and physical aspects of fatigue.

In conclusion, this feasibility study demonstrated the potential application of the *Packer Managing Fatigue Program* among PWPD. The findings support the need for future full-scale RCTs to rigorously evaluate the effectiveness of the program. Additionally, this study provided valuable insights into patient perspectives and the feasibility of outcome measures for use in future RCTs. This research makes a significant contribution to the development of fatigue interventions for PD fatigue and informs future investigations.

LIST OF ABBREVIATIONS USED

PD	Parkinson's Disease
PwPD	People with Parkinson`s Disease
MS	Multiple Sclerosis
OT	Occupational Therapist
FSS	Fatigue Severity Scale
MFI	Multidimensional Fatigue Inventory
GDS	Geriatric Depression Scale
HY	Hoehn and Yahr scale
PSQI	Pittsburgh Sleep Quality Index
COPM	Canadian Occupational Performance Measure
OBQ	Occupational Balance Questionnaire
PDQ	Parkinson`s Disease Quality of Life
MMSE	Mini-Mental State Examination Conservation Strategies Assessment
SEPESA	Self- Efficacy for Performing Energy Conservation Strategies Assessment
TEDSS	Taxonomy of Everyday Self-management Strategies
PRISMS	Practical Reviews in Self-Management Support Taxonomy

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I am overwhelmed with gratitude towards the people who have played an instrumental role in my success. First and foremost, I want to express my deepest appreciation and affection for my supervisor, who has been my guiding light throughout this incredible journey. She has not only seen potential in me but also nurtured it with unwavering support, guidance, and encouragement. I feel truly blessed to have had her by my side, through thick and thin.

To my supervisory committee, I express my heartfelt gratitude for their wisdom, expertise, and encouragement. Their support has been invaluable in shaping my research and my professional growth.

I am also deeply grateful to my family, who have been my rock throughout this experience. Despite the distance between us, their endless patience, unconditional love, and generous support have helped me to overcome numerous obstacles and pursue my dreams with confidence. Their sacrifices have been immeasurable, and I am forever grateful for their unwavering faith in me.

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I cannot forget to mention all study participants who devoted their time and energy to make this research feasible. Without their contributions, this study would not have been possible.

CHAPTER 1 - INTRODUCTION

Chapter 1 of this thesis introduces the research context, providing details for the rationale for selecting the research topic and the knowledge gap that this study seeks to address. The objectives of the thesis are presented, along with a brief overview of how they were achieved through the research methodology employed. The chapter presents an overview of the structure and organization of the thesis and its chapters, offering readers a clear roadmap to navigate through the various sections. Additionally, this chapter highlights the significance of the research topic and its relevance to contemporary developments in the field. A history of the program used in this research is also described. By setting out the foundational aspects of the research, this chapter sets the tone for the thesis and offers readers a clear understanding of its scope and significance. These topics are further discussed in Chapter 2 and Chapter 4, which describe the general literature review and detailed methodology and rationale of this research respectively.

1.1 Thesis Organization and Overview

This study was planned to evaluate the feasibility of the *Packer Managing Fatigue: The Individual Self-Management Program* (1) for people with Parkinson's disease (PwPD), as well as its potential for use in a future large-scale randomized controlled trial (RCT). The program was initially introduced in 1995 as *Managing fatigue: A Six-week Course for Energy Conservation*, and was delivered in a group format (2). In 2022, a second edition titled the *Packer Managing Fatigue: A Six-Week Self-Management Group Program* was published (3). An online version of the program was also developed and evaluated in 2009 (4). Building on these earlier versions, in 2023, the author developed the individual version of the program (1) which was the main focus of this research. Additionally, other

researchers have tested adaptations of the program using various delivery formats, including teleconference (5) and individual formats (6, 7). However, these adaptations varied in length and dosage, and were not fully consistent with the original program.

The feasibility assessment in this study followed two primary pathways: 1) evaluating the program's impact, relevance, delivery, and content from the perspective of PwPD and their confidence in using the skills learned; and 2) estimating the needed sample size for future RCTs based on effect sizes, evaluating the preliminary effectiveness of the program, and assessing the efficacy of recruitment strategies.

This thesis is composed of three main sections. The first section presents a scoping review that investigated and characterized the range of information relevant to fatigue self-management programs for individuals living with chronic conditions. The second section reports a mixed-methods study that explored the feasibility of the *Packer Managing Fatigue: A Six-Week Individual Self-Management Program* from the perspective of PwPD. The third section reports a pilot RCT that assigned participants in a 1:1 ratio to either usual care or the intervention (usual care + the *Packer Managing Fatigue: A Six-Week Individual Self-Management Program*) arm. This pilot RCT assessed the feasibility of the proposed research protocol for future studies. In this research, usual care was defined as the standard treatment or care that is provided to patients by their care team. This means that participants continued receiving the same care that they would have received had they not been enrolled in the trial.

The first three chapters provide literature reviews relevant to the aims of the thesis. Chapter 1 introduces the gaps and foundations for conducting this study and explains its structure.

Chapter 2 primarily reviews fatigue in PD. However, it begins with a general overview of fatigue across various conditions due to the similarities in how fatigue presents among different conditions. Chapter 3 presents a scoping review of fatigue interventions for individuals living with chronic conditions. Utilizing this methodology, the chapter examines fatigue interventions that followed the principles of a self-management approach. This review aimed to understand and compare the content and components of available fatigue programs. Chapter 4 describes the detailed methodology and rationale for this study. Chapters 5 and 6 report the findings of the research, including the feasibility of the study program from the perspective of PwPD and the feasibility of the research protocol for conducting future full-scale RCTs. Chapter 7 is a synthesis of the thesis that discusses the overall findings of the study, including reflections on the overall work, implications, and future research directions. Chapters 1, 2, and 7 are presented in non-manuscript format and provide an overview of the research topic, a comprehensive literature review, and a discussion of the thesis findings, respectively. In compliance with the university's guidelines, the references for the chapters were compiled and provided at the end of the thesis. Chapter 3 has been published in the *Patient Education and Counseling*. Chapter 4 has been published in the *Canadian Journal of Occupational Therapy.* Chapters 5 and 6 are in preparation for submission.

As the primary investigator, I played a pivotal role in the development, conduct, and documentation of my doctoral research. This involved formulating research questions, designing, and implementing appropriate methodologies, collecting, and analyzing data and presenting findings in a clear and concise manner. Throughout this process, I collaborated closely with my research committee and supervisor to ensure that my work

met rigorous academic standards. My committee and supervisor provided invaluable feedback and suggestions to keep me on track and ensure that my research progressed in the right direction. They also reviewed all written materials to ensure their quality. In addition to my research committee and PhD supervisor, two other co-authors assisted with data analysis and finalizing the scoping review presented in Chapter 3. One co-author also contributed to qualitative interviews and managing data collection for the feasibility questionnaires used for the program. This was for the manuscript presented in Chapter 5 which aimed to evaluate the feasibility of the program from the perspective of patients. Their guidance was instrumental in helping me achieve my research objectives. A detailed list of the contributions of all authors is also provided below:

Manuscript 1: What We Know About Fatigue Self-Management Programs for People Living with Chronic Conditions: A Scoping Review

Authors and Contributions:

- Neda Alizadeh: Conceptualization; Methodology; Data curation; Analysis; Investigation; Project administration; Resources; Validation; Visualization; Roles/Writing - original draft; Writing - review & editing.
- Tanya Packer: Conceptualization; Methodology; Analysis; Investigation; Supervision; Resources; Validation; Writing - review & editing.
- Yu-Ting Chen: Analysis; Writing - review & editing.
- Yaser Alnasery: Analysis; Writing - review & editing.

Manuscript 2: Managing Fatigue in Parkinson's Disease: Protocol for A Pilot Randomized Controlled Trial

Authors and Contributions:

- Neda Alizadeh: Conceptualization; Methodology; Validation; Visualization; Roles/Writing - original draft; Writing - review & editing.
- Tanya Packer: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Ingrid Sturkenboom: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Gail Eskes: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Grace Warner: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.

Manuscript 3: Mixed-method Evaluation of the Individual Packer Managing Fatigue Program: Perspectives of People with Parkinson`s Disease.

Authors and Contributions:

- Neda Alizadeh: Conceptualization; Methodology; Data curation; Analysis; Investigation; Project administration; Resources; Validation; Visualization; Roles/Writing - original draft; Writing - review & editing.
- Tanya Packer: Conceptualization; Methodology; Investigation; Supervision; Resources, Validation; Writing - review & editing.
- Sabrena Jaswal: Data curation; Project administration; Writing - review & editing.
- Ingrid Sturkenboom: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.

- Grace Warner: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.

Manuscript 4: Managing Fatigue in Parkinson's Disease: Preparing for A Pilot Randomized Controlled Trial

Authors and Contributions:

- Neda Alizadeh: Conceptualization; Methodology; Data curation; Analysis; Investigation; Project administration; Resources; Validation; Visualization; Roles/Writing - original draft; Writing - review & editing.
- Tanya Packer: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Ingrid Sturkenboom: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Grace Warner: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.
- Gail Eskes: Conceptualization; Methodology; Supervision; Resources; Validation; Writing - review & editing.

1.2 General Introduction

PD is a neurodegenerative disorder characterized by the presence of motor symptoms such as muscle rigidity, tremor, and bradykinesia. In addition to these motor symptoms, PwPD may also experience non-motor symptoms. Non-motor symptoms have been identified as a leading cause of deterioration in the quality of life as the disease progresses (8). Research has shown that non-motor symptoms can precede the onset of motor symptoms,

highlighting the importance of early recognition and intervention (9). It is important to recognize and address both types of symptoms early to improve overall patient well-being.

Fatigue is a prevalent but under-recognized non-motor symptom (10-12). The overall prevalence estimate for fatigue in PD was found to be 50% according to a systematic review and meta-analysis (11). The sensation of fatigue is characterized by a lack of energy than can pose challenges for the performance of routine daily activities (10). Fatigue is a significant contributor to the reduction in quality of life and is a leading cause of disability for PwPD (13). It has been shown to have a profound impact on various aspects of daily living and can result in financial distress for those affected by PD (13, 14). For example, it has been reported as a barrier to routine daily tasks including work, household chores, and physical abilities such as climbing stairs moving around and exercise (13, 15). Fatigue has been identified as the most bothersome non-motor symptom and one of the most common symptoms among all symptoms in PD, even more so than common motor difficulties such as bradykinesia, rigidity, and impaired postural stability (16).

Despite its significant negative impact on life of PwPD, the management of fatigue has been identified as an unmet need in PD care and is a top priority from both research and patient perspectives (16, 17). There is a lack of consensus on how to define and measure fatigue, and few studies have investigated its management. Most studies have focused on understanding its etiology. Currently, there is a paucity of RCTs available for fatigue management in PwPD. Even when RCTs are available they often report fatigue as a secondary outcome with small sample sizes, inconsistent definitions of fatigue, and without considering the impact of confounding variables (18-20). This highlights the need for further research into effective management strategies.

One program that has shown promise in other neurological conditions is the *Managing Fatigue: A Six-Week Course for Energy Conservation* program. This is an occupational therapy intervention designed to improve the occupational performance of individuals with severe fatigue. It aims to enable individuals to participate in daily life activities that they identify as important, meaningful, or necessary for self-care, productivity, leisure, and rest. The program is client-centered and evidence-based, and it recognizes that while the disease can contribute to fatigue, its impact on occupational performance may be similar regardless of the specific diagnosis (2).

During its development, the program utilized the Predisposing Reinforcing and Enabling Constructs in Educational Diagnosis and Evaluation (PRECEDE) model (21) to prioritize and select program content. The PRECEDE model employs a structured process to select content based on the known needs of a specific group and potential solutions. At the time of development of the original program, content assessed included exercise, medication, and energy conservation. Given the suggested dosage and time frame of the program, content focused on energy conservation strategies were selected. However, in recent adaptations, content for sleep hygiene and cognitive fatigue have been incorporated into the program. In addition to its content, the program is designed to build self-efficacy using a variety of strategies. The social cognitive theory proposes four ways to change behavior: competence mastery, peer modeling, social persuasion, and cognitive restructuring (21). All of these methods are included in the original program, and in later versions, quotes, and stories of experiences of peers were incorporated into the content of the program, as well as motivational interviewing by therapists. The facilitator's role in the program is to help participants plan weekly activities and support them in testing and trialing strategies

to be used at home or in the workplace. This helps build self-efficacy and encourages choice and personalization of options. The program is designed to promote active decision-making and effective energy management, utilizing available energy in an optimal way. Participants actively choose where they can decrease the amount of unnecessary energy used, and instead use their energy for things they need and want to do. Whether the program is delivered in a group or individually, online or in-person, strategies to build self-efficacy are incorporated into the program. The *Packer Managing Fatigue* program aligns with self-management theory, which involves the active participation of individuals in managing their health and well-being (22). Based on the scoping review (23) conducted in this thesis on fatigue programs for people with chronic conditions, it appears that the *Managing Fatigue* program places a significant emphasis on key self-management components. For details see Chapter 3. This review also identified that the most commonly cited self-management strategies were activities, process strategies, and social interaction strategies. This is consistent with the aim and focus of this program in providing tools and strategies to enable those with chronic fatigue to actively take control of their energy management to optimize participation in their meaningful daily activities despite the negative impact of fatigue. Given that there are several interacting components in this program, and since the outcome of the program may depend on the behavior of the individuals, the *Packer Managing Fatigue* program is a complex intervention as defined by the Medical Research Council (24). Therefore, mechanisms for change may be related to factors that may not be distinguishable, but the above provides the foundations.

Topics covered in the program include activity simplification, task analysis, environmental modification, fatigue communication, planning and prioritization, rest and relaxation

techniques, proper body mechanics to conserve energy, utilization of tools and technology to simplify tasks, breaking down activities into manageable components, balancing, planning and scheduling to optimize energy, goal setting to promote motivation and engagement in meaningful activities, sleep hygiene to improve sleep quality, and strategies for managing cognitive fatigue. The rationale for including these topics in the program is that they directly address patients' areas of concern related to managing their fatigue effectively. While individual therapists may have their own approaches to managing fatigue, the *Packer Managing Fatigue* program provides a comprehensive and structured approach that covers a wide range of topics relevant to patients' concerns. To optimize the delivery of the program, detailed therapist training was developed in which therapists learned more about the program and general considerations for PD-specific fatigue. More details on this training can be found in Chapters 4-6.

Originally, the program included one therapist manual with patient handouts. However, in its most recent versions, it has been updated to include two separate manuals: one for patients and one for occupational therapists. In addition to changes in its format, the *Packer Managing Fatigue* program has also been adapted for delivery through various methods.

The original version of the program was first developed to be delivered in a group setting but has since been adapted for other methods of delivery including online (4), and one-on-one (1). The 2020 individual *Packer Managing Fatigue: A Six-Week Individual Self-Management Program* (1) was evaluated in our study as well as in another three-arm randomized controlled trial (25). The program was originally developed in Canada but has been evaluated in several other regions including Austria, the Netherlands, Sweden, Australia, and the United States. The effectiveness of the program has been demonstrated

through several RCTs in neurological conditions such as MS (5, 26-30) and neuromuscular diseases (31). These studies have shown that the program can decrease the impact of fatigue on daily life activities (28, 32-34) while increasing self-efficacy, and participation in daily life activities (29, 31, 35) and enhancing quality of life (7, 26, 29, 31). Despite its proven effectiveness in MS, the program has rarely been studied in patients with PD. More details on these studies can be found in Chapter 2.

This individual version was created to provide a standardized and consistent approach to one-on-one delivery. The general content, duration and frequency is consistent with the original program but includes additional content on sleep hygiene and cognitive fatigue to align with current knowledge on the importance of cognitive fatigue in neurological conditions and the impact of sleep on fatigue. The six sessions focus on trialling, evaluating, and adopting energy conservation strategies including: session 1) the importance of rest and sleep; session 2) communication and body mechanics; session 3) activity stations; session 4) priorities and standards; and session 5) balancing your schedule. Session six includes a course review and a discussion of future recommendations. Each weekly session is scheduled for 90 minutes but could be adjusted according to individual needs. The entire program is scheduled to last 6-8 weeks (1).

In summary, the original *Managing Fatigue* program is an evidence-based non-pharmacological intervention that could help address this unmet need for PwPD by providing individuals with tools to manage their fatigue more effectively. Fatigue is a common symptom among individuals with neurological conditions (36). Despite potential differences in how fatigue manifests among various neurological disorders (37, 38), its severity and impact are comparable across conditions. As such, it is anticipated that the

new individual *Packer Managing Fatigue* program will yield similar results for individuals with PD as have been observed for those with MS and neuromuscular conditions. Therefore, the program was implemented without adapting it to be specific to PwPD, but OTs received specific PD training and homework activities were tailored to each individual. This aimed to enhance the delivery and tailoring. This decision was made based on the assumption that the program would be effective for individuals with PD given the similarities in fatigue severity and effects across neurological conditions. For ease of use and consistency in this thesis, we will refer to the program as the *Packer Managing Fatigue* program.

Shortly after receiving ethics permission to begin recruitment, this research was impacted by the COVID-19 pandemic in Nova Scotia (March 2020), which was the most severe lockdown period. The COVID-19 pandemic affected research study designs in several ways (37). It necessitated a shift from in-person to remote data collection methods and impacted recruitment and retention of trial participants (39). Additionally, the pandemic introduced additional stressors or challenges for research participants which impacted their ability to engage in interventions and affect health outcomes (40, 41). As many jurisdictions adopted virtual care and telehealth interventions as usual care during that time, evidence of effectiveness for video conference-based interventions was needed more than ever (42). Telehealth has proven to be an innovative solution during pandemics like COVID-19, providing a means of limiting patient exposure to potentially infected individuals (43). It also offers a long-lasting solution for patients to receive appropriate treatment in their home environment (44). In response to these challenges, the current research also transitioned to videoconference delivery and online data collection. The

program was delivered using videoconferencing and data were collected using online surveys while participants were in videoconference meetings with the assessor. These modifications enabled the study to continue during the pandemic while increasing accessibility for participants in remote areas and reducing geographic and transportation barriers while allowing participants to follow public health restrictions imposed by the COVID-19 pandemic.

CHAPTER 2 - FATIGUE IN THE BIG PICTURE: A FOCUS ON PARKINSON'S DISEASE

2.1 Fatigue in Neurological Conditions

2.1.1 History and Epidemiology

Fatigue is considered one of the most common and disabling symptoms among people living with chronic neurologic conditions. For example, its prevalence is estimated at 54–88% in people with MS (45, 46); > 50% in those with PD (47), 30–70% for those who have had a stroke (48), and up to 73% for people with traumatic brain injury (TBI) (49).

Despite its high prevalence, fatigue, as a clinical symptom, was not studied until the late 1980s (50, 51). Previously, fatigue was generally interpreted as physiological fatigue, defined as tiredness experienced after vigorous physical or mental activity or a mismatch between an individual's perceived ability to initiate and continue tasks and what they could actually do. This type of tiredness does not interrupt daily life activities as it is often short-term and alleviated by rest (51, 52). The differences between "physiological fatigue" and "pathological fatigue," which occurs in people with chronic conditions, emerged later. Fatigue as a pathological condition is more complex and long-lasting than general tiredness. It is a chronic symptom that can persist for months or even years and can occur at any time of the day. Unlike general tiredness, this type of fatigue does not disappear with rest and can significantly interfere with daily activities (53, 54).

2.1.2 Definition and Classification

Fatigue and fatigue-related symptoms have now been described and classified in a variety of ways (55) and yet there is no universal concept across conditions and disciplines (54, 56, 57). Based on current knowledge, fatigue appears to be a common phenomenon across

neurological conditions with similar impact and severity (34). For example, in MS, fatigue is considered a state of exhaustion distinct from depressed mood or physical weakness. It can interfere with a daily functioning and is a common symptom of the condition (58). Similarly, in PD, fatigue has been described as a consistent sense of exhaustion that manifests in a multidimensional manner, making the performance of daily life activities, physical or mental, a strain for individuals (59). In stroke, fatigue is characterized by a feeling of early exhaustion during mental activity, with weariness, lack of energy and aversion to effort that interferes with daily activities (60). A recent scoping review conducted by our research team (23), investigated fatigue management in chronic conditions. The review included 15 studies representing nine conditions and found that there were no distinct differences in the definitions of fatigue across different conditions. Fatigue was generally characterized as a multifaceted and complex symptom, with excessive tiredness not proportional to activity and interference with daily activities. These findings suggest that while the specific manifestations of fatigue may vary among different conditions, the overall impact and severity of the symptom may be similar.

Fatigue can be classified in many different ways, including primary/secondary, central/peripheral, subjective/objective, and physical/cognitive fatigue (55). Fatigue can emerge as primary symptom of a condition is due to the inflammatory and/or degenerative disease process. Fatigue can also emerge secondary to other symptoms of a disease or comorbidities such as sleep, depression, or anemia (61). It has been classified into central vs peripheral fatigue. Central fatigue is due to malfunction of central neural structures such as the motor cortex, which impairs the signal transmission to the motor unit and muscle fibers. It occurs more in individuals with central nervous system and upper motor neuron

diseases including MS, PD, and stroke (62). In contrast, peripheral fatigue is related to dysfunction outside of the central nervous system and is often associated with a loss of muscle strength or atrophy (62).

Fatigue can also be described as subjective perceptions of fatigue and fatigability. Perception of fatigue usually refers to the general sensation of exhaustion that is perceived by a person (54, 63, 64). Fatigability is defined as "the magnitude or rate of change in a performance criterion relative to a reference value over a given time of task performance or measure of mechanical output" [p.411] (54). Fatigability is also described in two forms: perceived fatigability and performance fatigability. Perceived fatigability refers to capability of a person and is influenced by their psychological state and their body's ability to maintain homeostasis. Performance fatigability, on the other hand, refers to a decline in performance due to fatigue and is influenced by the nervous system and involved muscle (54). Fatigability is measured by observing changes in an outcome variable during the actual performance of a task. Fatigue, on the other hand, is estimated through self-report questionnaires that ask respondents to estimate their capacity to perform various cognitive, physical, and psychosocial tasks (65).

Fatigue can manifest in two distinct ways: physical fatigue and cognitive fatigue. Some individuals experience both physical and cognitive fatigue while some only experience one form (66). Based on available definitions, physical fatigue refers to the reduced energy to complete a task requiring physical effort and is more commonly addressed in the clinic and research (63). Cognitive fatigue, on the other hand, is a relatively newer concept and has been characterized by a lack of mental energy that can interfere with an individual's ability to initiate and sustain mental tasks (63, 67). This can manifest as difficulty concentrating,

reduced mental capacity, and decreased performance during and after prolonged cognitive effort (63). While fatigue is often associated with prolonged physical or mental effort, it can also manifest as a chronic condition that is not necessarily triggered by a difficult task. In some cases, individuals may experience persistent fatigue that limits their ability to initiate even simple tasks.

2.1.3 Etiology of Fatigue

Current imaging studies in MS, PD, and stroke have found that an impairment in a wide range of central nervous system structures relates to fatigue. There is no general consensus on the specific cortical area playing a key role in fatigue (66). However, the frontal regions (68), parietal white matter (68, 69), corpus callosum (69, 70), basal ganglia (71), internal capsule, the periventricular trigone of the lateral ventricle (66), and thalamus (71, 72) are believed to be involved. Overall, in PwPD, dysfunctions in the basal ganglia have been observed (73). In individuals who have had a stroke, dysfunctions in the reticular activating system (RAS) and subcortical grey and white matter have been found whereas individuals with MS, whereas in individuals with MS, abnormalities in the cingulate gyri and left primary sensory cortex have been reported (61, 74).

Environmental triggers such as stress, temperature, infection, and immunization may also induce fatigue (61, 75, 76). Stress may change the neuroendocrine system mainly through the hypothalamic-pituitary-adrenal axis and the sympathetic nervous system. Changes in the neurotransmitters serotonin, dopamine, and adrenaline determine the nature and severity of fatigue-associated symptoms such as muscle pain, sleep disorder, and anxiety (61).

Fatigue is often misdiagnosed or not diagnosed at all. The subjective and unique-to-each-person nature of fatigue raises the risk of failure to recognize fatigue (77). It is often difficult for patients and healthcare providers to differentiate primary fatigue from fatigue secondary to other conditions including sleep problems, apathy, and/or depression that may manifest similar experiences (66). Therefore, fatigue may be undiagnosed or excluded from treatment plans if healthcare providers do not ask specific questions about the occurrence of fatigue and its impact on daily lives using self-report subjective measurements (78, 79).

2.1.4 Impact of Fatigue in Neurological Conditions

In addition to being prevalent, fatigue is also one of the most disabling symptoms for people with chronic neurologic conditions. Up to 40% of individuals with MS (80) and about 50% of those with PD reported fatigue as their most disabling symptom (81). Fatigue limits the ability to engage in daily activities, employment, leisure, social participation (activities with family, friends and/or within community), sleep, driving, and community mobility for people with neurological conditions including MS, PD, and stroke (13, 82, 83).

Fatigue is one of the main reasons for the negative sense of productivity and reduced work capacity leading to early retirement (14, 84, 85). Individuals with MS, especially those with a minor disability, list fatigue as a major reason for their unemployment (86). Similarly, fatigue in PD is associated with early retirement and reduced work hours, sometimes resulting in financial distress (14, 59). Fatigue is also associated with restricted participation in rehabilitation and medical care and poor neurological recovery (87, 88). This may be the consequence of impaired cognitive capacity that is associated with fatigue (89), the lack of energy, or dependence on help to seek and adhere to necessary medical

treatments. This extensive negative impact of fatigue on lives of individuals with neurological conditions is therefore associated with decreased quality of life (88-90).

2.2 Fatigue in Parkinson's Disease

2.2.1 Fatigue: A Common Non-Motor Symptom of Parkinson's Disease

PD is the most common movement disorder and the second most prevalent neurodegenerative disorder after Alzheimer's disease. The exact cause of PD is not known, but several risk factors have been suggested. These include genetic factors, pre-existing depression, sleep problems, intestinal impairments, and exposure to toxins (91). PD is characterized by the gradual loss of neuronal subtypes, specifically in the nigrostriatal dopaminergic pathway, and basal ganglia motor loops interfering with normal movement onset and execution (92, 93). PD may present with a broad range of motor and non-motor symptoms and high level of disability. However, the manifestation and intensity of symptoms vary by person, as PD has a different prognosis and is also related to personal and environmental factors (77, 91, 94).

Well-recognized motor symptoms associated with PD are resting tremor, rigidity, akinesia, bradykinesia, hyperkinesia, and postural instability. The presence and impact of a variety of non-motor symptoms is a significant part of the clinical spectrum of PD that contributes to a high level of disability. The most common non-motor features of PD are fatigue, anxiety, depression, apathy, sleep problems, sensory dysregulation and/or impairment in cognitive function (12, 91, 95-97).

Fatigue in PD has been reported as one of the most bothersome non-motor symptoms of PD but is less recognized in research and underestimated in PD care (11, 96). Research into fatigue in PD is relatively recent compared to other similar conditions and has not been studied until the last two decades (47, 98, 99).

Fatigue in PD, similar to other chronic neurological conditions discussed above, impacts many aspects of everyday life. In PD, the need to measure and address different aspects of fatigue, including physical and cognitive, has been the specific focus of previous studies (100, 101). Based on the available qualitative findings that have explored the impact of fatigue from the perspective of patients with PD, fatigue was reported as a barrier to routine daily tasks including household chores, self-care, engaging with children and grandchildren, activities that need physical abilities (e.g., climbing stairs, moving around, and exercise) (15). Fatigue in PD can also cause financial distress for people living with PD as they often retire early and reduce their work hours (13, 14). In one study, up to half of the individuals with PD identified fatigue as the primary reason for their inability to work (14). Inability to accomplish life activities and employment can decrease the quality of life in patients with PD (81, 102).

Since there is minimal research on fatigue patterns in both early and advanced PD and because PD can start and progress differently in individuals, it is difficult to explicate one fatigue pattern in PD over time. However, fatigue seems to be present in both advanced and early stages of PD. It can arise before motor symptoms (103-106). Based on the finding of a longitudinal study, fatigue prevalence in PD increased over time (from 35.7% experiencing fatigue at baseline to 42.9% four years later, 55.7% at eight years, and 73.1% at nine years follow-up). This study also found a significant increase in fatigue severity

during the nine years of follow-up (103). Therefore, based on the available evidence, fatigue in PD tends to start early and worsen with disease progression over time (11, 107).

Etiology of fatigue in PD is not well understood. It may have a combination of primary and secondary causes, and it can be difficult to distinguish between the two (66). While motor symptoms such as tremor and rigidity may contribute to the development of fatigue in PwPD, the presence of cognitive fatigue and its occurrence even in those with mild motor symptoms suggest that other factors may also play a role (73). Dysfunctions in the basal ganglia have been observed in those with PD fatigue. In PD, the brain changes that occur are mostly related to the impairment and/or death of nerve cells in the basal ganglia, which normally produce dopamine. When these neurons die or become impaired, they produce less dopamine, leading to movement problems associated with PD. Additionally, people with PD lose the nerve endings that produce norepinephrine, which might help explain some non-movement features of PD such as fatigue (63, 73).

Overall, based on available evidence, fatigue in PD is related to interactions between multiple systems including upper motor neuron, lower motor neuron, and neuromuscular junction (108). As Kostić noted, the upper motor neuron involvement is connected to observed "abnormality in basal ganglia (BG)-cortical mechanisms, particularly frontal loops, and an imbalance between neurotransmitters (e.g., dopamine and serotonin), along with an altered hypothalamus-pituitary-adrenal axis, neuroinflammation, cardiac sympathetic denervation, etc." (109) (P.323.).

2.2.2 Factors Contributing to Fatigue in PD

Fatigue, although an independent symptom in PD (54), can also be secondary to other PD-related symptoms such as sleep disturbances, pain, or depression, which result in similar manifestations and impact (107, 110). The presence of comorbid medical conditions such as major depressive disorder (MDD), cerebrovascular disease, heart failure, or diabetes may contribute to the development of fatigue in PwPD. These comorbidities can exacerbate the underlying fatigue associated with PD and may require additional evaluation and treatment to manage effectively (111).

A large number of factors associated with fatigue in PD have been reported in literature. However, there is heterogeneity among studies in terms of the measures used and the reported statistical significance (112). Factors include PD-related factors such as disease severity (101, 113); mood disorders such as depression, anxiety and apathy (101, 107, 113, 114); cognitive impairments (115); sleep difficulty (101, 107, 116); autonomic and/or sensory dysregulation (101), and side effects of medications (20, 111). A systematic review of 44 studies and 7,427 patients with fatigue in PD found that age, disease duration, daily medication dose, disease severity, depression, anxiety, and apathy are the most frequently reported significant correlators with fatigue in PD (11).

2.2.3 Fatigue Measurement in PD

To date, there are no sufficiently validated biomarkers to diagnose fatigue. Assessment is mostly based on self-report using a variety of questionnaires (86). In PD, fatigue has been found to be multidimensional and may appear as different mental and/or physical, which can be independent of each other and impact quality of life differently. These dimensions

are recommended to be measured specifically (63, 117). The majority of the fatigue measures evaluate the subjective experiences of patients (37). Objective fatigue/fatigability is usually examined by testing individual's performance before and after doing a demanding task (54). None of the fatigue measures can screen for confounding factors that need to be assessed and controlled for optimal fatigue management intervention. Therefore, considering the presence of these factors is important when measuring or treating fatigue (118).

In 2010, the Movement Disorders Society of PD, organized a task force to evaluate current fatigue measurements (generic and/or disease-specific) in PD (37). Fatigue measurements were included if they (1) were used in PD, (2) assessed only fatigue, and (3) has been used by groups other than the developers of the measures. In total, seven fatigue measurements were identified, comprising the Fatigue Severity Scale (FSS), the Fatigue Assessment Inventory (FAI), the Functional Assessment of Chronic Illness Therapy-Fatigue Scale (FACIT-F), the Multidimensional Fatigue Inventory (MFI), the Parkinson Fatigue Scale (PFS), the Fatigue Severity Inventory (FSI), and the Fatigue Impact Scale for Daily Use (D-FIS) (37). Each of these fatigue measurements was reviewed based on its: (1) application in PD populations; (2) application in clinical studies beyond the developers; and (3) validity, reliability and sensitivity to changes in the PD population. If a measurement fulfilled all three conditions, it was marked as a "recommended measure"; if the measurement was applied in PD studies but only in one of the two other conditions, it was marked as a "suggested measure", and finally, if a measurement was applied in PD studies but neither of the other two conditions were met, it was marked as a "listed measure". Among all included fatigue measurements in the review by Friedman and

colleagues (2010), only two scales, the FSS and the MFI were marked as “recommended measures” for measuring fatigue severity (37).

Neither recommended fatigue measure of PD was developed specifically for PD-related fatigue. The MFI is the only measure able to measure multiple dimensions of fatigue. The available information on the MFI is mostly based on other conditions and not PD. There was also no data on the reliability and validity of the MFI in PD at the time of review by Friedman and colleagues. In 2012, Elbers and colleagues evaluated the validity and reliability of the MFI in 153 patients with PD (119). They demonstrated that the four domains of the MFI (physical fatigue, mental fatigue, reduced motivation, and reduced activity) were reliable and valid to assess the multidimensional aspects of fatigue in patients with PD (38).

The only measure that was developed specifically for PD-related fatigue was PFS, developed by Brown, Dittner, Findley, and Wessely (120), which was not recommended by the Movement Disorders Society of PD. It was developed based on the experiences of fatigue in individuals with PD. A drawback of this measure is that it only measures the impact of physical fatigue and not the multidimensional impact of fatigue in PD. PFS has been tested in more than 600 individuals with PD (121). The internal consistency among 16 items from the PFS scale is high (117). Yet, its sensitivity to evaluate changes in fatigue requires more evidence (37).

The FSS was marked as a “recommended” fatigue scale in PD because of acceptable psychometric properties including its ability to discriminate fatigue in PwPD and it has been used by groups other than the developers. FSS is a self-reported, general 9-item scale

that assesses the functional impact of fatigue (122). The FSS is unidimensional and does not measure multiple aspects of fatigue (123, 124). It has been frequently used as a measure to assess general fatigue or as a screening tool in many fatigue studies and its psychometric properties have been validated in many chronic conditions including PD (125, 126).

2.2.4 Potential Fatigue Interventions in PD

Although fatigue is a common symptom in many neurological conditions, its management is an unmet need in PD care (127) and is of top priority both from research (128) and patient perspectives (16). A recent systematic review of fatigue self-management interventions, delivered by occupational therapists and physiotherapists, for multiple groups of people living with chronic conditions found that only eight PwPD were included among a total of 3109 participants from 36 primary studies (127). In another recent systematic review that evaluated effectiveness of non-pharmacological fatigue interventions, across chronic conditions, it was found that among the 28 included RCTs, 10 were in MS, one was in post-polio syndrome, and none were found for PD, TBI, or stroke (129). Similarly, in our recent scoping review, which evaluated the content of fatigue self-management programs in individuals with chronic conditions, out of the nine included disease groups, PD was missing (23). These findings confirm that there is limited evidence-based research on fatigue management approaches for PwPD and emphasize the importance of evaluating fatigue interventions specifically for this population. Therefore, in this literature review chapter, although the scope was initially focused on PD fatigue, given the limited availability of research on fatigue management in PD, it was necessary to borrow evidence from other conditions, especially MS, in some sections. This approach allowed a more

comprehensive understanding of fatigue management strategies that may be applicable to PwPD.

Most available studies have focused on understanding the etiology, evaluation, and experiences of fatigue, leaving a paucity of effectiveness studies in the literature. Even when RCTs are available, fatigue is often reported as a secondary outcome, there are small sample sizes, inconsistent definitions of fatigue and limited consideration of confounding variables (20, 130). To our knowledge, a systematic review of fatigue interventions, including pharmacological and non-pharmacological fatigue programs, specifically in PwPD was last conducted in 2015 (18). In 2019, the International Parkinson and Movement Disorder Society Evidence-Based Medicine Committee conducted a review of the literature to identify new evidence-based recommendations for treating nonmotor symptoms, including fatigue, published after 2011 (131). Four new RCTs were identified, of which only two focused on fatigue intervention. One study evaluated pharmacological approaches and the other examined acupuncture. The evidence for the efficacy of these programs in PD was deemed "insufficient" (131).

Evidence on the effectiveness of medications for PD-related fatigue is inconclusive and limited. Although some evidence suggests that rasagiline and modafinil may have a modest effect in improving physical fatigue, the side effects of other PD medications can worsen fatigue or related symptoms including depression (132, 133). Although still developing, several non-pharmacological interventions have been evaluated in PD or other conditions with similar experiences of fatigue. Among all, the three types of non-pharmacological fatigue interventions commonly used in neurological conditions are Cognitive Behavioural Therapy (CBT), exercise therapy, and energy-conservation interventions (18, 20).

The CBT-based programs focus mostly on the experiences and consequences of behaviours. Individuals are encouraged to learn from their past experiences to modify their behaviours in future to increase benefits and minimize the negative consequences of their behaviours (131). The available evidence regarding the effectiveness of CBT in managing PD fatigue is limited (18). However, evidence is available for other conditions such as chronic fatigue syndrome (CFS) and MS. In a systematic review of 15 RCTs evaluating the effectiveness of CBT interventions in people with CFS (N=1043), CBT was found to be more effective than usual care in reducing symptoms of fatigue post-treatment. However, when compared to other psychological therapies, such as relaxation, counselling, and education/support, the difference in mean fatigue scores was not statistically significant (134). Similarly, a systematic review and meta-analysis of four RCTs with a total of 193 CBT-treated patients and 210 control-treated patients with MS showed that CBT had a positive short-term and long-term effect on fatigue (135). Nonetheless, evidence of the effectiveness and feasibility of CBT-based programs for PD-related fatigue is limited (18, 136). A systematic review evaluating the effectiveness of CBT on different PD symptoms, including mood disorders, sleep, and fatigue, found that among all included studies only two have assessed the impact of CBT on fatigue management in PwPD, but they did not report an evidence of impact of CBT on fatigue (137).

Another common non-pharmacological approach to managing fatigue is exercise. The effect of exercise on fatigue has received considerable research attention in other chronic neurological conditions including MS. For example, according a metanalysis, multiple exercise regimens have been evaluated in patients with MS, including resistance training, aerobic training, yoga, aquatic exercise, and combined training (138). The effect sizes for

these interventions varied from -0.24 to 2.05 with a pooled effect size of 0.57 (P = 0.02). In total, 30% of exercise-based interventions included in this metanalysis showed a significant intervention impact (138). In stroke, there is preliminary evidence that exercise promotes sleep and reduces fatigue. However, the extent to which exercise impacts these health parameters is still unclear (139).

In PD, although exercise is widely recognized as an important tool for managing motor symptoms, its effects on fatigue are still being explored. A systematic review (140), evaluating the impact of physical activity on non-motor symptoms of PD, including depression, cognition, fatigue, apathy, anxiety, and sleep, found that exercise therapy can decrease fatigue impact. Included interventions varied greatly in terms of frequency and length of program. Overall, significant improvements in non-motor symptoms were found with different durations (4 months to three years) and frequencies (2–4 sessions/week) of intervention. Exercise type also varied across included interventions (e.g., aerobic training, treadmill training and walking; resistance training; balance training, Tai Chi; as well as customized programs such as physiotherapy or occupational therapy). Although depression was the most widely studied outcome, showing significant improvements, significant improvements were seen in fatigue in only one of the included studies (140).

However, other evidence does not support the use of exercise as a potential fatigue intervention, with some PwPD even finding that exercise triggers their fatigue (111, 141, 142). Therefore, several gaps remain unaddressed. There is limited information on the types of exercise that are most effective, and existing programs have not been sufficiently compared. The exact mechanisms by which exercise improves fatigue in individuals with PD are not well understood, and studies have used small sample sizes with participants that

have variable PD symptoms, age, and disease stage (140). It is important for future research to address these gaps and carefully consider the heterogeneity of PD participants when evaluating the impact of exercise on fatigue. Using exercise as a potential intervention to control fatigue requires precise planning and prescription. Future large-scale, high-quality randomized clinical trials are needed for validation (143).

Energy conservation is the third non-pharmacological intervention for fatigue. This therapeutic approach involves balancing activity and rest, outsourcing tasks, and making physical/environmental adaptations (144). In 2022, a scoping review (144) was conducted to identify and organize energy conservation practices. The review found that energy conservation strategies were the most common way to manage pain and fatigue. The *Managing Fatigue Program: A Six-Week Energy Conservation Co*urse (2) is one of the most recognized programs in the field, which has been evaluated and proven effective in multiple RCTs, showing evidence for medium to large effect sizes for various outcomes, including fatigue impact (5, 26, 28, 32), quality of life (26, 32), stress and anxiety (29) and self-efficacy (26, 29). In a recent systematic review (145), seven out of 10 energy conservation studies were developed based on the *Managing Fatigue* program. In our recent scoping review evaluating the effectiveness of individual fatigue programs in people with chronic conditions (23), we also found that one of the main groups of programs that incorporated the main components of self-management principles were developed based on the *Managing Fatigue* program. Out of a total of 15 interventions included in the review, five were based on the *Managing Fatigue* program. Therefore, the *Managing Fatigue* program appears to be one of the most established and effective fatigue management programs available for individuals with chronic conditions.

In summary, fatigue is a significant symptom in PD that requires attention and management, yet evidence-based research on fatigue management approaches for PwPD is limited. Most available studies have focused on understanding the etiology and experiences of fatigue, leaving a paucity of effectiveness studies in the literature. The efficacy of medications for PD-related fatigue is inconclusive and limited, and non-pharmacological interventions, such as cognitive-behavioral therapy, exercise therapy, and energy-conservation interventions, are being explored mostly in other conditions with similar fatigue impact and experiences. However, evidence of the effectiveness and feasibility of these interventions for PD-related fatigue is limited, and further research is needed to develop effective interventions for this population.

2.3 Managing Fatigue: A Six-Week Energy Conservation Course

The *Managing Fatigue* program (2), is a well-known program widely used to manage fatigue in neurological conditions (146). The original program was a group-based in-person program. Similar to other occupational therapy programs, it is focused on optimizing occupational performance, defined as the ability to perform and engage in valued activities and roles in the home or community context (147). The program includes energy management strategies such as activity simplification, task analysis, environmental modification, communicating about fatigue, planning, and prioritizing (6). The original program included six sessions: (1) the importance of rest; (2) communication and body mechanics; (3) activity stations; (4) priorities and standards; (5) balancing your schedule; and (6) course review and future plans.

The effectiveness of the *Managing Fatigue* program has been tested in several randomized controlled trials. The program is effective in reducing fatigue impact (28, 32-34). Patients who have received the program showed significant improvement in the cognitive scale (mean difference = -2.91; 95% CI: -4.32, -1.50), physical scale (mean difference =-2.99; 95% CI: -4.47, -1.52) and the psychosocial scale (mean difference = -6.05; 95% CI: -8.72, -3.37) compared to patients in control groups (146). Furthermore, the program has a positive impact on quality of life (7, 26, 29, 31) , participation (29, 31, 35), self-efficacy (5, 7, 26), depression (27), and sleep quality (27). Thus, based on the large body of evidence on the effectiveness of the Packer Managing Fatigue program on multiple outcomes, the program is an evidence-based, non-pharmacological program.

Despite the proven effectiveness of the *Packer Managing Fatigue* program in MS, PwPD have rarely been studied. The only study that recruited people living with PD included only eight patients with PD in an RCT design that evaluated the effectiveness of an internet format of the *Managing Fatigue* program in a sample of patients with MS, post-polio syndrome, or PD (29). The study findings demonstrated only marginal improvement in self-efficacy and significant reduction in stress compared to the no-intervention group. No PD-specific analysis was conducted in this study (29).

Since its development, multiple forms of the *Managing Fatigue* program have been developed and tested, mainly in MS. These include: face-to-face group-based delivery (27, 28, 34, 148); a teleconference version (32, 33); an internet version (29); and one-to-one delivery (6, 7). With a few exceptions, one-to-one delivery of the *Managing Fatigue* program has been via adaptations by researchers other than original authors. Blikman et al., (6), evaluated a 12-session version of the program delivered one-to-one for people

living with MS in a randomized controlled trial design. The primary outcomes were fatigue severity (fatigue subscale of Checklist Individual Strength-CIS20r), fatigue impact (Modified Fatigue Impact Scale-MFIS) and participation (Impact on Participation and Autonomy scale - IPA). The intervention used in this study was not more effective at reducing fatigue and participation restrictions than an information-only control condition. In another study, Van Heest et al (7), reproduced only five modules of the original program and evaluated patients with chronic conditions (MS, fibromyalgia, cancer, and stroke) in a one-group pre-test, post-test design. The last session of the original program was eliminated in this study, and the 5-session version of the program was delivered over a 4–6-week period. The findings of this study demonstrated significant improvements in post-test fatigue measured by the Functional Assessment of Chronic Illness Therapy–Fatigue Scale; quality of life, measured with the Functional Assessment of Cancer Therapy–General; and self-efficacy, measured with the Self-efficacy for Performing Energy Conservation Strategies Assessment (SEPECSA). Based on these studies, findings on effectiveness of these one-to-one versions of the program are inconclusive. Further research is needed to standardize and evaluate the one-to-one delivery of the *Packer Managing Fatigue* program. The current study addressed this gap by evaluating the feasibility and preliminary effectiveness of the newly standardized one-to-one version of the program: *Packer Managing Fatigue: A Six-week Individual Self-management Program* for PwPD. The detailed methodology can be found in Chapter 4.

CHAPTER 3 - WHAT WE KNOW ABOUT FATIGUE SELF-MANAGEMENT PROGRAMS FOR PEOPLE LIVING WITH CHRONIC CONDITIONS: A SCOPING REVIEW

This chapter is a manuscript that was published in the Patient Education and Counseling in June 2023. The manuscript provides the background and identifies the gaps in the literature on fatigue self-management programs that inform this PhD research.

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3.1 Abstract

Objective: The significant impact of fatigue on the lives of patients with chronic conditions has demanded healthcare research to respond. One response has been the development and testing of self-management programs. Little is known about what these programs have in common or how they differ. This scoping review compared the key components of fatigue self-management programs.

Methods: Scoping review methodology was employed. Databases of CINAHL, Academic Search Premier, PsycINFO, Cochrane and Medline were searched to identify relevant sources.

Results: Included fatigue programs were compared using a three-component framework: 1) self-management strategies; 2) active patient participation; and 3) self-management support. Although all programs included some aspects of these components, the extent varied with only a few domains of these components found across all programs.

Conclusion: The three self-management components employed in this study showed potential benefits in identifying similarities and differences across fatigue programs with comparable and distinct underlying theories. This three-component framework could facilitate identification of domains associated with positive outcomes.

Practice Implications: It is essential that authors of programs provide detailed descriptions to enable inter-program comparison. The three-component framework chosen for this review was capable of describing and comparing fatigue self-management programs, paving the way for more effective interventions.

3.2 Highlights

- Comparative analysis of fatigue programs is challenging due to the lack of defined self-management components.
- A three-component framework: self-management strategies, active patient participation, and self-management support, enables comparing programs.
- Active patient participation is most common; self-management support is least common in fatigue programs.
- To select best programs, therapists assess content, patient participation and support, together with their clinical judgment.

3.3 Introduction

Fatigue is considered one of the most common and disabling symptoms experienced by people with chronic conditions including multiple sclerosis (MS) (149), Parkinson`s disease (PD) (150), rheumatoid arthritis (RA) (151), and cancer (152) and results in a comparable magnitude of impact and severity across conditions (36). Multiple frameworks and conceptualizations have been proposed to define this type of chronic, ongoing fatigue (55), yet universal consensus across conditions and disciplines remains elusive (54, 56, 57). However, regardless of conditions, fatigue is complex and can persist for months or even years. Unlike transient tiredness, this type of fatigue does not disappear with rest and can significantly interfere with daily activities (53, 54).

This type of fatigue impairs performance in household activities, leisure, employment, and social participation and is frequently found to be associated with decreased quality of life (13, 15, 82, 83, 88, 153, 154). It is a complex experience that can manifest as cognitive and/or physical symptoms (155). While sleep and depression are known to be related to fatigue, the direction and nuances of this relationship are still unknown (54). Despite its huge impact, fatigue is relatively overlooked in the research (156).

Currently, there is no cure and no definitive cause has been identified for fatigue within or across conditions that emphasize the importance of non-pharmacological interventions. Among non-pharmacological treatments, interventions focusing on self-management have shown improved health outcomes in people with chronic conditions (157, 158). Self-management programs often aim to build self-efficacy and acquire helpful behaviors and strategies that enable patients to manage their health and care (159). These behaviours and

strategies are commonly referred to as medical management (e.g., monitoring and adhering to medication or diet), role management (e.g., building and maintaining daily roles) and emotional management (e.g., coping with depression due to disease (18). The goal of self-management programs is to empower patients to collaboratively and actively determine goals for their health and care that are derived from their personal choices and life requirements (160-162). The role of the "active, engaged patient" was first introduced in the Chronic Care Model developed by Wagner et al (160) Supporting individuals to learn and use their knowledge and skills to manage their condition and its impact on daily life is integral to improve disease symptoms and functional outcomes (18).

There is an extensive variety of theories underpinning and conceptualizing self-management programs in general (157-159). This points to the need to find ways to compare programs and identify the active components of successful programs (160). While many fatigue interventions are described as self-management programs, the extent to which self-management components are included, the skills presented, and the support provided vary across programs. Yet, a few comparisons have been undertaken to analyze the level of similarity and diversity of these programs (129).

The impact of fatigue on the lives of people with chronic conditions and the importance of integrating self-management programs into their care is significant. The objective of this scoping review was to provide a comprehensive understanding of fatigue self-management programs and their key components. The study investigated and characterized the breadth of information relevant to fatigue self-management programs for individuals living with chronic fatigue. Additionally, the review examined the theoretical frameworks, setting and delivery formats, and logistics of these programs, as well as the definitions of self-

management and fatigue within the context of these programs. This work sets the stage for future investigations to determine which program components/characteristics are associate with positive health outcomes.

3.4 Methods

3.4.1 *Design*

This study employed the five-step scoping review methodology suggested by Arksey and O'Malley (161). Reporting details were also guided by the PRISMA Extension for Scoping Reviews (PRISMA-ScR) (162). The protocol was registered with the Open Science Framework (https://osf.io/z9u3s).

Step 1- Identify the Research Question: The main purpose of this study was to review and compare fatigue self-management programs for people living with fatigue secondary to a chronic condition. The questions this study aimed to answer were 1) What is known about the theoretical frameworks, setting and delivery formats, and logistics of fatigue self-management programs? 2) How is self-management defined? 3) How is fatigue defined? (4) What are the self-management components in the programs and how are they implemented?

Step 2- Identify Relevant Studies: The search strategy was developed in collaboration with a university librarian with experience in scoping and systematic review methodology. CINAHL (EBSCO Publishing, Glendale, CA), Academic Search Premier, PsycINFO, Cochrane and Medline databases were searched in February 2021 to identify relevant sources. The search focused on the concepts of self-management and fatigue, with the team

identifying and selecting articles reporting on patients with chronic conditions through rigorous screening procedures. Due to the absence of a feasible approach or predetermined classification system encompassing all specific chronic conditions related to fatigue, our review refrained from imposing restrictions based on particular chronic conditions. However, the authors utilized the description of a chronic condition as defined by Bernell and Howard (163). This definition encompasses a long duration of the disease, which requires lifelong medical intervention and has a substantial impact on daily functioning. Detailed search strategies are presented in Appendix A.

Step 3 - Study Selection and Operationalizing the Definitions: To consider a program self-management, the inclusion of a very broad starting definition as proposed by Van de Velde et al. (159) was used: “Self-management is the intrinsically controlled ability of an active, responsible, informed and autonomous individual to live with the medical, role and emotional consequences of his chronic condition(s) in partnership with his social network and the healthcare provider(s).” (p.10).

According to this definition, which is also consistent with the Chronic Care Model, patients living with chronic conditions are considered experts in their own lives (164, 165). Therefore, programs that included any indication of decision making and taking actions by patients and with an active partnership between patients and interventionist were considered self-management programs. As stated by Bodenheimer et. al., (166), self-management is the shift from traditional care to collaborative care in which patients are experts in their lives and healthcare providers are experts in the disease.

Studies were included if they were published in English, peer-reviewed journals between 2001 and 2021, and focused on fatigue as the main purpose of the intervention. Participants had to be adults aged 18 years or older with fatigue secondary to one or more chronic conditions. Programs that focused strictly on medical adherence, acquisition of information, or were composed solely of symptom monitoring, dietary changes, or exercise were excluded. Programs that were not delivered by healthcare providers (e.g., delivered only by lay leaders) or programs with an absence of an active partnership between patients and interventionists were also excluded. Finally, interventions directed at clinicians or caregivers were also excluded. Studies prior to 2001 were considered to have limited currency and were excluded since self-management science has developed primarily in the last two decades (167).

All results from searches were uploaded to the Covidence Software (168) where duplicates were removed. Before beginning the abstract/title review, inter-rater reliability of the selection criteria was tested. First, reviewers (NA, YTC, TLP, YA) screened the same five articles using the preliminary selection criteria. After considering discrepancies and building a common understanding between reviewers, definitions were refined and the screening and data extraction manuals were amended. Next, reviewers individually screened the same 50 articles, chosen randomly, then discussed differences until consensus was reached. The process of reviewing articles in blocks of 50 was repeated three times until the kappa level of agreement reached ≥ 0.8, which represents a high level of agreement (169).

All citations deemed relevant were procured for subsequent full-text review. Those articles that could not be obtained through institutional holdings available to the authors

were requested by document delivery or from the source author or journal when available. The reviewers repeated the reliability process, first with five articles to gain preliminary consensus. They then reviewed 10 randomly selected articles. After three trials of 10 articles, the team's level of agreement reached ≥ 0.8 kappa score. In the full-text review, articles were included if there was evidence that the intervention program used in the study focused on fatigue and met the operational definition of a self-management program.

Step 4 - Charting the data: A systematic and purposeful approach was applied to chart the findings. A data extraction form was created by the first author to capture the characteristics of studies based on the research questions. The form was tested by reviewers who each extracted data from the same two articles. After a round of discussion, revisions were made, and the final agreed-upon form was reproduced in the web-based software platform, COVIDENCE (Table 1).

Data from each article were extracted by two reviewers. Once all data were extracted, two reviewers (NA and YTC) were assigned to clean, collapse, or consolidate the extracted text into a single entry. When there was a query, the original extractors were consulted. Finalized extracted data was then exported to an excel spreadsheet to be coded.

To categorize the self-management components, two taxonomies and thematic analysis were used. The patient active participation component was categorized using thematic coding. The Taxonomy of Everyday Self-management Strategies (TEDSS) was used to analyze self-management program content. It describes five goal-oriented and two support-oriented domains. The goal-oriented domains are the "Activities", "Internal", "Social

Interaction", "Disease Control", and "Healthy Behaviour" domains. The support-oriented includes the "Process" and "Resource" domains (170). The TEDSS Framework was designed as a patient-centred framework identifying self-management strategies used to manage a chronic condition (170).

The Practical Reviews in Self-Management Support (PRISMS) Taxonomy was used to categorize self-management support. The PRISMS proposes 14 domains that can be used by healthcare providers to support self-management for people with long-term conditions (171). Pearce et al., (2016) synthesized over 100 systematic reviews for self-management support which resulted in the PRISMS taxonomy (171). The domains of the PRISMS taxonomy were developed specifically based on self-management support studies rather than behavioral change theories (171). Therefore, it is more inclusive and has a broader lens when compared with other existing frameworks such as the taxonomy of Behavioral Change Techniques (BCT) developed by Michie et al., (172) which focus only on client interactions and excludes services required.

At least two reviewers independently coded/categorized the extracted data and each pair of reviewers then met to discuss and resolve conflicts. If consensus was not reached, the conflicts were discussed within the larger research team. Once consensus occurred, the data was coded again by the same reviewers, using the final agreed-upon codes, and the final codes with detailed examples were discussed with the whole research team.

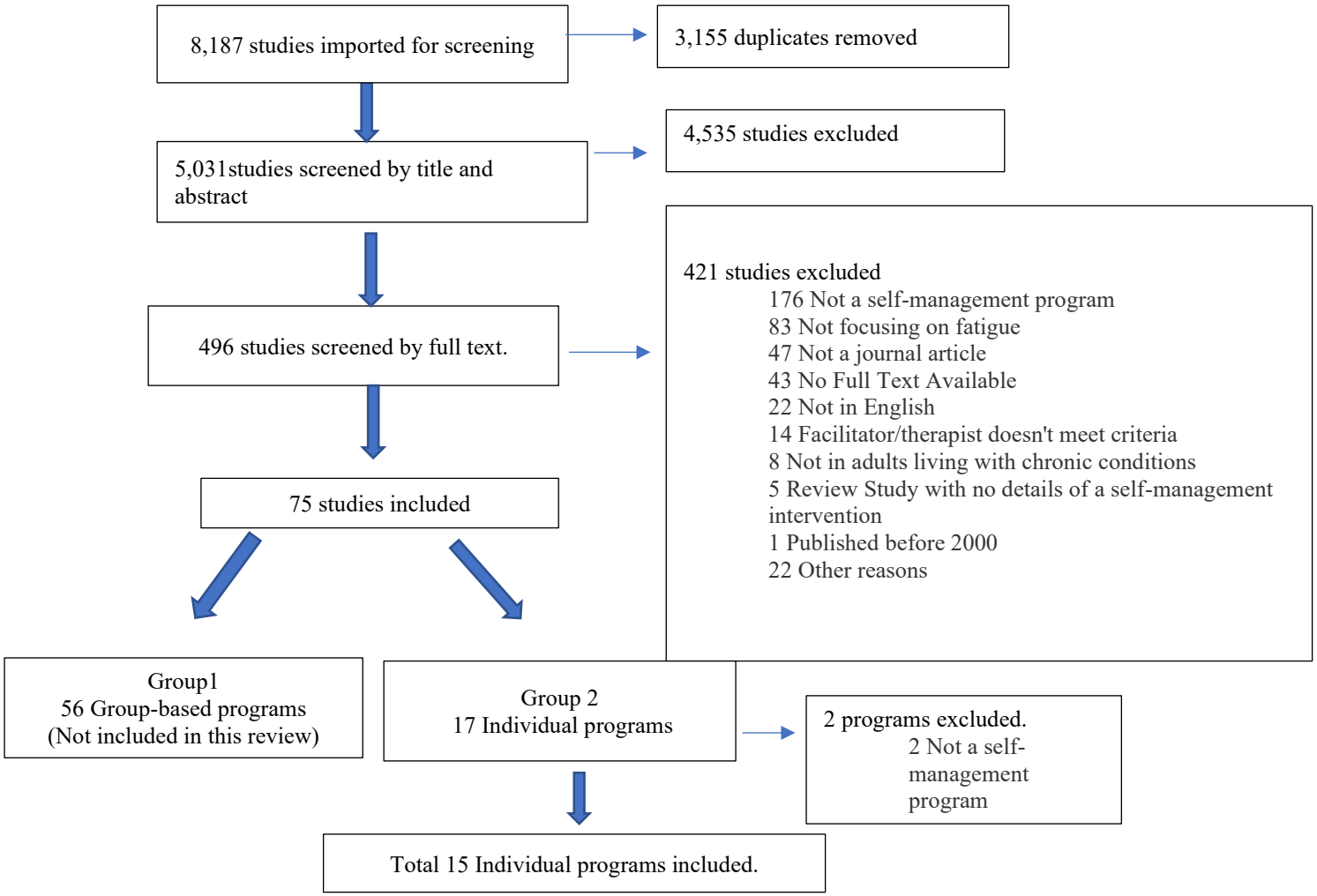
Table 1 *Extraction Form with Definitions and Examples*

Type of Data	Data Extracted, Definitions and Operationalization of Terms
Description of Studies	The following data were extracted: title, author (s), publication year, country, research objectives, study design, main diagnosis, and fatigue outcome measures (primary vs secondary).
Participants characteristics	The following data were extracted: age, gender, disease severity, disease severity (e.g., stage 4 Parkinson`s disease
Description of programs	The following data were extracted: program name given by author; underpinning theories; program goal as defined by authors, logistics, delivery mode and setting; skill and qualifications of program providers. Logistics were operationalized as the time, duration, and number of sessions. Mode of delivery was categorized as in-person, online, telephone, or videoconference. Setting was defined as the patient location when the program was delivered and categorized as if not home, hospital, clinic. research lab. Underpinning theory was defined as any theoretical framework/theory/ supporting evidence that authors used as the bases of their fatigue intervention. This could refer to a previously developed theory such as the Social Cognitive Theory or other resources such as a literature review.
Description of fatigue	Any information authors used to describe fatigue, its types and/or measures in their programs was extracted. *" Typology" refers to the focus of the program regarding type of fatigue if described such as cognitive, physical, general fatigue or not separated. *" Measures" refers to any fatigue measure used in the study to screen or measure fatigue
Description of self-management and self-management components	Any information authors used to define/describe "self-management" anywhere in the text was extracted. Any information authors used to describe the content and delivery of the program was extracted. These included details of any skills, practice, education, activities designed for patients to learn/practice in the program, and/or any kind of support that providers gave to patients with fatigue (Lecture, role playing, any type of encouragement, etc.). In coding stages, following components were specifically used to categorize the extracted data: 1- Self-management strategies as defined in the Taxonomy of Everyday Self-management Strategies (TEDSS) 2- Self-management support as defined in the Practical Reviews in PRISMS taxonomy. 3- Patient active participation which refers to any evidence for patient active decision making in the program (e.g., whether patients can choose the content, or activities in the session, or if there are any homework pieces)

Step 5 - Collating, summarizing, and reporting the results. To decipher the concept and components of fatigue self-management program a qualitative content analyzing approach was utilized (173). The programs in the included studies to the domains of three main self-management components were then mapped. A combination of a descriptive numerical summary and a thematic analysis were used to summarize and report data based on research questions. Inclusion average percentages was calculated using the average numbers of included domains for each self-management components across programs.

As presented in the PRISMA flow diagram (Figure 1), 75 full-text studies met the study criteria. There was great diversity in setting and delivery formats of the programs. Closer review revealed that the included articles were comprised of two distinct groups of programs: group vs one-to-one delivery. Some studies used a mix of formats. To reduce the variability among programs and allow better comparison, only programs that fully or partially included one-to-one delivery are reported here. Future papers will report results of the other group. During data extraction at the full text level, two additional articles were excluded as detailed reading of the articles revealed that they were not consistent with the inclusion criteria.

Figure 1 *PRISMA Diagram of Study Selection for Scoping Review of Fatigue Self-Management Intervention*



3.5 Results

In total, 15 interventions, described in 14 studies, were included in this review. The study designs varied and included nine full-scale randomized controlled trial designs (RCTs), one quasi-experimental design, and two pilot and/or feasibility studies. Two protocol papers for RCTs were also included. Overall, the findings from completed effectiveness studies demonstrated some level of positive impacts on multiple outcomes, including fatigue impact (7, 174-180) , participation (30, 174, 179), quality of life (7, 174, 180), mental health -including depression, anxiety and emotional distress (174, 176, 180), and self-efficacy (7).

3.5.1 *What Is Known about the Theoretical Frameworks, Setting and Delivery Formats, and Logistics of Fatigue Self-Management Programs?*

Included programs reported a range of underpinning theoretical frameworks, goals, delivery settings, and fatigue measures (Table 2). Fully in-person delivery was the most common approach (n =10), followed by a mixed format of telehealth and in-person (n =3) and telehealth-only (n =2). Telehealth formats included videoconferencing and teleconference calls. The mixed delivery formats were a combination of phone calls, online modules, and in-person sessions.

Programs were divided into three main types according to their theoretical foundation: 1) developed based on cognitive behavioural therapy (CBT) (n=4); 2) a version of the Managing Fatigue program developed by Packer et al. (1995) (n=5); and 3) “Other” programs that were based on one or a combination of theories (n=6). These included the energy envelop theory, energy management education, psychobiological entropy model, and the chronic care model. There was one program, "Fatigue and Activity Management

Education (FAME)", which was developed based on the results of a qualitative study [38]. Another program described a cognitive therapy treatment that focused on developing cognitive strategies to better tolerate and reduce stress and self- criticism [40].

Overall, five main program goals were identified in the data (Table 2). The most common were "*To improve participation in daily life activities"* (n=8) and "*To build and improve self-management behaviour/skills"* (n=5). Participants in the programs were from nine disease groups: MS (n=4), cancer (n=2), chronic fatigue syndrome (n=3), end-stage renal disease (n=2), systemic lupus erythematosus (SLE) (n=1) and acquired brain injury (n=1). One study included participants with multiple chronic conditions (MS, Guillain-Barr´e syndrome, SLE, Myasthenia gravis, and Muscular dystrophy).

Fatigue was measured for two main reasons: as a screening tool for participant inclusion and/or as an outcome measure. The only measure used to screen participants in more than one study was the Fatigue Severity Scale (FSS) (n =3). Fatigue as an outcome measure was most commonly assessed with the Fatigue Severity Scale (FSS) (n =5), Modified Fatigue Impact Scale (MFIS) (n =4) and/or Checklist Individual Strength (CIS20r) (n =3).

Table 2 *Overview of Characteristics of Included Programs*

Authors/ Year	Name of Program	Theoretical Foundation	Goal(s) of the Program	Participant Condition(s)	Fatigue Measures (screening and/or outcome measure)	Setting/Delivery Format
Picariello/2018(181)	Cognitive-behavioural therapy (CBT) for renal fatigue (BReF)	CBT	Positive believes, attitudes and behaviour to cope with disease	ESRD	Screening: CFQ Outcome measure: CFQ	Mix- In person and telephone calls-Consists of 3-5 sessions over 4–6 weeks. Delivered by the primary researcher, or a registered health psychologist
Jason/2007(182)	Cognitive Behavior Therapy (CBT)	CBT "Cognitive Therapy (Approach) Chronic Care Model"	To improve participation in daily life activities Positive believes, attitudes and behaviour to cope with disease	CFS	Screening: None Outcome measure: Fatigue Severity Scale (FSS).	In person-Consists of 13 sessions (45 minutes) every 2 weeks Delivered by registered nurses
Friedberg/2013(177)	Fatigue Self-Management (FSM)	CBT Clinical model of CFS	To build and improve self-management behaviour/skills	UCF and CFS	Screening: None Outcome measure: FSS.	In-person-Consists of two Session over three weeks. Delivered by registered nurses
Ehde/2015 (183)	Telephone-delivered self-management intervention (T-SM)	CBT	To build and improve self-management behaviour/skills	MS	Screening: MFIS Outcome measure: MFIS	Telehealth-Telephone calls_ Consists of 8 weekly sessions (45- to 60-minute) plus15-minute follow-up calls at 4 and 8 weeks post-treatment Delivered by social workers and psychologists

Authors/ Year	Name of Program	Theoretical Foundation	Goal(s) of the Program	Participant Condition(s)	Fatigue Measures (screening and/or outcome measure)	Setting/Delivery Format
Blikman/2017 (6)	Individual energy conservation management (IECM)	Managing Fatigue program	To build and improve Self-Management Behaviour/Skills To Improve Self-Efficacy To Improve energy Conservation Skills To reduce the severity of fatigue	MS	Screening: Checklist Individual Strength (CIS20r) subscale fatigue Outcome measure: Checklist Individual Strength (CIS20r) subscale fatigue and FSS	In-person- consists of 12 sessions (45 min) over 4 months. Delivered by OTs.
VanHeest/2017 (7)	one-to-one format of the 6-wk Managing Fatigue course developed by Fox (2010)	Managing Fatigue program	To improve participation in daily life activities	MS, Fibromyalgia, Cancer Poststroke, Guillain-Barr´e syndrome, SLE. Myastheniagravis, Muscular dystrophy.	Screening: FSS Outcome measure: FACIT FS	In-person-consisted of 5 modules over four to six sessions of 1–2 hours. Delivered by OTs and/or OT students
Kos/2016 (30)	Individual self-management occupational therapy intervention program (SMOoTh)	Managing Fatigue program Energy Envelope Theory Behavioural Change Theories	To improve participation in daily life activities To Improve Self-Efficacy	MS	Screening: Visual Analog Scale developed by Kos et al [58]. Outcome measure: MFIS	In-person-consisted of three sessions of 60–90 min for three consecutive weeks. Delivered by OTs.

Authors/ Year	Name of Program	Theoretical Foundation	Goal(s) of the Program	Participant Condition(s)	Fatigue Measures (screening and/or outcome measure)	Setting/Delivery Format
Plow/2020 (25)	Managing Fatigue Program	Managing Fatigue program Social Cognitive Theory	To build and improve self-management behaviour/skills To improve self-efficacy To Improve energy conservation skills	MS	Screening: Fatigue Severity Scale (FSS), Outcome measure: FIS	In-person-the number and length of sessions is tailored to participants' needs and preferences. over the 6 weeks Delivered by OTs.
Raina/2016 (175)	Maximizing Energy (MAX) intervention	Managing Fatigue program Behavior Activation Theory	To improve participation in daily life activities	TBI / ABI	Screening: FSS Outcome measure: PROMIS	Telehealth-Internet using Web-camera technology -consisted of two sessions of 30-minutes per week over an 8-week period. Delivered by OTs.
Ream/2006 (176)	Beating Fatigue intervention	Winningham 's Psychobiological Entropy model	To improve participation in daily life activities To improve energy conservation skills	SLE	Screening: None Outcome measure: Four visual analogue scales (VASs)*	In-person- The intervention program was provided over the first three treatment cycles. Diary entries were reviewed by support nurses who visited patients at home once during each treatment cycle. - Delivered by registered nurses

Authors/ Year	Name of Program	Theoretical Foundation	Goal(s) of the Program	Participant Condition(s)	Fatigue Measures (screening and/or outcome measure)	Setting/Delivery Format
ORiordan/2017(174)	Fatigue and Activity Management Education (FAME)	Based on a qualitative study for people with SLR	To improve participation in daily life activities	SLE	Screening: None Outcome measure: FSS	In-person-Mix of groups and one-to-one delivery-Consists of 6 weekly sessions (2.5 hours). OTs with multidisciplinary input (PT and Dietitian) will deliver the program.
Kos/2015 (179)	Activity Pacing Self-management (APSM)	Energy Envelope Theory	No report	CFS	Screening: None Outcome measure: CIS	In-person-Three sessions over three weeks (60–90 min) OTs and/or PTs delivered the program
Yates/2005 (178)	The psychoeducational intervention for Managing Fatigue in Women Receiving Adjuvant Chemotherapy for Early-Stage Breast Cancer	Green's PRECEDE (Predisposing, Reinforcing, and Enabling Causes in Educational Diagnosis and Evaluation) model of health behavior.	To build and improve self-management behaviour/skills	Early-Stage Breast Cancer: (Stage I or II)	Screening: None Outcome measure: Four 11-point numeric rating scales developed from the literature	Mix- In person and telephone calls-Consists of 2 sessions conducted by phone (10 minutes) and 3 in-person sessions-Two additional booster sessions were employed. Delivered by registered nurses

Authors/ Year	Name of Program	Theoretical Foundation	Goal(s) of the Program	Participant Condition(s)	Fatigue Measures (screening and/or outcome measure)	Setting/Delivery Format
Farragher/2019 (184)		Energy Management Education (EME) Clinical model of Cognitive Orientation to Occupational Performance (CO-OP)	To improve participation in daily life activities	ESRD	Screening: Fatigue Severity Scale (FSS) Outcome measure: Fatigue Severity Scale (FSS), Fatigue Management Questionnaire and Modified Fatigue Impact Scale (MFIS)	
Jason/2007 (182)	Cognitive Therapy Treatment (COG)	Cognitive Therapy Approach Chronic Care Model	To improve participation in daily life activities Positive believes, attitudes and behaviour to cope with disease	CFS	Screening: None Outcome measure: Fatigue Severity Scale (FSS).	In person-Consists of 13 sessions (45 minutes) every 2 weeks Delivered by registered nurses

SRD: End stage renal disease, FSS: Fatigue Severity Scale, CFQ: Chalder Fatigue Questionnaire, CFS: Chronic fatigue syndrome also known as Myalgic Encephalomyelitis or Myalgic Encephalopathy (ME), UCF: Medically unexplained chronic fatigue, MFIS: Modified Fatigue Impact Scale, FACIT FS: Functional Assessment of Chronic Illness Therapy–Fatigue Scale, FIS: Fatigue Impact Scale, PROMIS: Patient-Reported Outcomes Measurement Information System Fatigue Scale, SLE: Systemic Lupus Erythematosusm, CIS: the Checklist Individual Strength.

* Four visual analogue scales (VASs): subjective quantification of fatigue, subjective distress because of fatigue, and subjective assessment of effects of fatigue on chores/work and on pastimes/hobbies

3.5.2 *How Are Fatigue and Self-Management Defined?*

Nine programs included definitions of fatigue that ranged from "a sense of exhaustion or lack of physical and/or mental energy" (6, 181, 184) to "a decreased capacity to fulfill daily life activities" (7, 180, 185). Three key themes emerged after coding and categorizing the data: *"a multifaceted/complex symptom*"; "*excessive tiredness not proportional to activity"; and* "*a chronic symptom interfering with activities".* The identification and categorization of fatigue dimensions (ex. physical, mental, etc.) was not possible as it was not discussed in any of included studies.

Despite using inclusion criteria that comprised components of self-management programs (e.g., evidence for goal setting, problem-solving, active decision-making, and active partnership between interventionists and patients), only four of the included studies explicitly defined self-management. The lack of data meant coding/identifying themes was not possible.

3.5.3 *What Are the Self-Management Components in the Programs and How Are They Implemented?*

All programs reported evidence of all three self-management components: 1) self-management strategies, 2) active patient participation, and 3) self-management support. However, the combination of these components varied. Since there were multiple programs based on either CBT principles or the *Packer Managing Fatigue* program*,* data were also grouped and compared by subgroups.

3.5.4 *Self-management strategies*

All programs included content in at least one TEDSS domains with a range of 1–6 out of seven possible domains across programs (Median=4). Among program types, the inclusion range varied least in CBT-based program types (n =4-5) (Appendix B). CBT-based programs also had the highest mean number of TEDSS domains (64.28%) compared to two other program types (Table 5).

In terms of frequency of cited domains, “Activities" domain (n =13/15) was the most frequently reported, while "Resource" domain (n =1/15) was the least reported across all programs. Analysing program types showed that the "Resource" domain was least reported in both CBT-based (n=0) and *Packer Managing Fatigue*-based programs (n=1). Domains of "Activities", "Internal", and "Healthy Behaviour" were included in all CBT-based programs. All *Packer Managing Fatigue*-based programs included content in domains of "Process" and "Activities” whereas the “Healthy behaviours” domain was rarely incorporated into these programs (n =1) (Table 4).

Table 3 *Frequency of Cited Domains of Self-Management Components by Total and by Program Types*

Self-management component	Domains	*Managing Fatigue*-based programs (/5)	CBT-based (/4)	Others (/6)	Total (/15)
TEDSS	Activities	5	4	4	13
	Internal strategies	2	4	3	9
	Social interaction strategies	4	1	2	7
	Healthy behaviours	1	4	3	8
	Disease controlling strategies	3	2	5	10
	Process strategies	5	3	4	12
	Resource strategies	1	0	0	1
Active Patient Participation	Goal setting	4	2	4	10
	Problem solving	3	1	4	7
	Practice activities, experiment, discovery	4	2	3	9
	Homework	4	4	3	11
	Tracking, monitoring, self-evaluation	1	4	4	9
	Active discussion	3	1	5	9
PRISMS	Information about condition and/or its management	5	3	5	13
	Information about available resources	0	0	1	1
	Provision of/agreement on specific clinical action plans and/or rescue medication	5	4	6	15
	Regular clinical review	5	4	6	15
	Monitoring of condition with feedback	4	1	3	8
	Practical Support with adherence (Medication or Behavioral)	1	2	1	4
	Provision of equipment	0	0	0	0
	Provision of easy access to advice or support when needed	0	0	1	1
	Training/rehearsal to communicate with healthcare professionals [and others]	0	0	0	0
	Training/rehearsal for everyday activities	4	0	2	6
	Training/rehearsal for practical self-management activities	4	4	3	11
	Training/rehearsal for psychological strategies	1	4	1	6
	Social support	1	0	1	2
	Lifestyle advice and support	0	0	0	0

3.5.5 *Active patient participation*

Six main themes emerged for active patient participation (Table 3). The frequency of included themes for active patient participation ranged from 1 to 6 (median=4) across programs (Appendix B). "Homework" was the most common cited domain (11/15), followed by "Goal setting" (10/15). "Problem-solving" (7/15) was the least commonly reported domain.

Table 4 *Themes and Definitions for Active Patient Participation in Fatigue Management Programs*

Themes	Definition/Examples
Goal setting	Process of collaborative prioritizing, identifying needs and preferences, and setting goals and planning a required course of actions.
Problem-solving	The cognitive process of identifying problems and analyzing the factors, facilitators, and barriers to solve or overcome them.
Practice activities, experiment, discovery	Generating strategies, trial and error of strategies, rehearsals, and practice of an active behavior-this usually happens in sessions and is one step before agreement for final practice activities.
Tracking, monitoring, self-evaluating	Using tracking sheets, logs, and diaries to actively document and/or record behaviors and/or feelings.
Homework	Agreed upon home-based activities/tasks/assignments to practice at home and/or between sessions.
Active discussion	Active communication between patients and therapists.

Comparison among program types found that “Homework” was most commonly reported in 4 of the *Packer Managing Fatigue*-based programs and all CBT-based programs. However, these two program types differently addressed the domain of “Tracking, monitoring, self-evaluation”. While this domain was present in all CBT programs, it was only reported in one *Packer Managing Fatigue*-based program. “Goal setting", and "Practice activities, experiments, and discovery" were also frequently reported in the *Packer Managing Fatigue*-based programs (4/5 intervention) as well as “Homework”. In

CBT-based programs, domains of "Active Discussion" and "Problem Solving" were rarely cited. Overall, the mean inclusion of active patient participation was mostly seen in the *Packer Managing Fatigue*-based program type (63.3%) (Table 5).

3.5.6 *Self-Management Support*

All programs included a range between 3–8 of the total 14 PRISMS domains (median=6). This inclusion range was 5-8 for *Packer Managing Fatigue*-based programs and 4-6 across CBT-based programs (Appendix B).

Two PRISMS domains, "Provision of/agreement on specific clinical action plans and/or rescue medication" and "Regular clinical review", were included in all programs. "Information about the condition and/or its management" (n= 13) was the next most prominent, followed by "Training/rehearsal for practical self-management activities" (n= 11). Conversely, "Training/rehearsal to communicate with healthcare providers", "Provision of equipment", and "Lifestyle advice and support" were not included in any of programs (Table 4).

Although "Training/rehearsal for everyday activities" was implemented in four support domains was the highest in the *Packer Managing Fatigue*-based programs (42.8%) compared to other program types (Table 5).

Table 5 *Average Inclusion of Self-Management Components Across Programs Reported by Program Types*

Programs	Managing Fatigue-based programs						CBT-based					Others						
	Blikman 2017	Kos 2016	Plow2020	Van Heest	Raina 2016	**Total Average of Domains Included (n, %)**	Picariello 2018	Friedberg 2013	Ehde 2015	Jason 2007	**Total Average (%)**	O' Riordan 2017	Ream 2016	Kos 2015	Farragher 2019	Yates 2005	Jason 2007	**Total Average (%)**
Inclusion of TEDSS domains (/7)	5	3	6	4	3	**4.2**	5	4	5	4	**4.5**	4	4	3	1	4	4	**3.3**
Inclusion of TEDSS domains (%) *	71.4	42.8	85.7	57.1	42.8	**57.8**	71.4	57.1	71.4	57.1	**64.2**	42.8	57.1	42.8	14.2	57.1	57.1	**47.6**
Inclusion of Active Patient Participation domains (/6)	5	6	3	1	4	**3.8**	2	2	5	5	**3.5**	2	3	3	5	5	4	**3.7**
Inclusions of Active Patient Participation domains (%)	83.3	100	50	16.6	66.6	**63.3**	33.3	33.3	83.3	83.3	**58.3**	33.3	50	50	83.3	83.3	66.6	**61.1**
Inclusion of PRISMS domains (/14)	6	8	6	5	5	**6**	4	6	6	6	**5.5**	3	5	5	4	7	7	**5.16**
Inclusion of PRISMS domains (%)	42.8	57.1	42.8	35.7	35.7	**42.8**	28.5	42.8	42.8	42.8	**39.2**	21.4	35.7	35.7	28.5	50	50	**36.9**

* The average inclusion percentage of each component in programs was calculated using the average of the number of included domains for each component divided by the total number of available domains in each component multiplied by 100; TEDSS presents Taxonomy of Everyday Self-management Strategies; PRISMS presents the Practical Reviews in Self-Management Support.

3.6 Discussion and Conclusion

3.6.1 *Discussion*

In total, 15 one-to-one fatigue self-management programs were examined in this scoping review. There is a lack of an agreed-upon model to describe and compare multicomponent self-management programs. This review is the first to delineate three important self-management components, namely, self-management strategies, self-management support and active patient participation. Two established frameworks, the TEDSS and the PRISMS were used to describe and quantify the first two components. Thematic analysis was used to define and then quantify the third. The analysis led to three main findings: 1) the three self-management components selected for this review appear to have the capacity to compare programs within and between program types; 2) this framework was also helpful in identifying the most and least frequently applied domains of self-management among programs; and 3) present programs lack description for self-management and its components.

3.6.1.1 The Suggested Three-Component Framework Has the Potential to Compare Self-Management Programs. All fatigue programs in this review included all three components either fully or partially. However, the range and focus of included components and their domains varied. The application of the three-component framework in this study allowed us to compare program types with different underpinning bases in terms of their consistency and extent of incorporating self-management components and their domains.

The findings showed that each program type had a different constellation of components. TEDSS domains were consistently included or not-included in *Packer*

Managing Fatigue-based programs. This is expected in programs based on a standardized protocol. The one exception was "Internal" domain strategies, which were reported in only 40% of *Packer Managing Fatigue*-based programs. In CBT-based programs, which have a consistent theory base but not the same standardized protocol, "Healthy behaviours" strategies were consistently reported in all CBT-based programs and strategies in the "Disease Controlling" domain were reported in half the programs.

As noted, all CBT-based programs reported content from the "Healthy behaviour" domain but only one of the *Packer Managing Fatigue*-based programs did so. While "Social interaction" domain strategies were commonly reported in *Packer Managing Fatigue*-based programs, it was only present in one CBT-based program. This likely reflects the standardized protocol of the *Packer Managing Fatigue* program which includes communication with others about fatigue (2). Interestingly, *Packer Managing Fatigue*-based programs were delivered by occupational therapists while CBT-based programs were delivered by psychologists. Professional differences in approaches and theories may partially explain differences in content.

In terms of the inclusion of the active patient participation between program types, it is noted that even though the "Tracking, Monitoring, Self-Evaluation" domain was found in only 20% of *Packer Managing Fatigue*-based programs, it was implemented in all CBT-based programs. Conversely, "Active Discussion" and "Problem Solving" were less present in CBT group (25%) compared to *Packer Managing Fatigue*-based programs (60%).

Finally, self-management support for "Training/rehearsal for everyday activities" was implemented in 80% of *Packer Managing Fatigue-based* programs while it was not found in any of CBT-based programs. On the contrary, this comparison demonstrated that although “Training/rehearsal for psychological strategies” was cited in all CBT-based programs (100%), it was only present in 20% of *Packer Managing Fatigue*-based programs. This could be expected because the original *Managing Fatigue* program is primarily aimed to increase patient participation in everyday activities (2, 186), while the CBT approach is focused on understanding the relationship between thoughts, feelings and behaviours and intends to enable behaviour change by understanding internal thoughts and beliefs (187).

Overall, despite the lack of explicit identification of self-management components and their domains supplied by authors, and the small number of programs per group type, we found that the three self-management components selected for this review appear to have capacity to compare and contrast program types. However, we recommend additional research to confirm these findings.

3.6.1.2 Delineating the Inclusion of Self-Management Strategies, Supports, and Patient Active Participation Is Possible. The average number of included domains of active patient participation was the highest across all programs compared to self-management support and strategies. Although, there were domains of self-management support that were applied in all programs, the mean number of self-management support domains found in programs was the least among all three components. For example, among the PRISMS domains, the regular clinical process activities such as "Provision

of/agreement" and "Regular clinical review" were found in all programs, while there were ten domains that were implemented only in less than half of the programs.

Among self-management strategies listed in TEDSS, the three domains of “Activities”, “Process” and “Disease controlling” were the three most commonly reported content across fatigue programs. This is mostly consistent with results of a recent systematic review which found that “Process”, “Healthy behaviours”, and “Disease controlling” were the most frequent domains reported in all self-management programs for patients with long-term conditions (188). However, strategies in the “Activities” domain were found to be the most frequent (13/15) in this study. The significant impact that fatigue has on everyday activities (13, 83, 189), is a likely explanation why “Activities” predominates in fatigue programs.

Among the PRISMS domains, "Provision of/agreement on specific clinical action plans and/or rescue medication" and "Regular clinical review" were reported in all programs, consistent with the findings of a recent scoping review of e-health self-management support interventions in musculoskeletal disorders (190). However, contrary to the findings of this scoping review, which indicated that "Lifestyle guidance and support" was the most prevalent component of the PRISMS taxonomy (n =59; 94%) [50], which suggested that "Lifestyle advice and support" was the most common component of the PRISMS taxonomy (n =59; 94%), the current study's findings revealed that this domain was absent in all programs. This could be due to either authors' lack of reporting content in programs or the complexity of involved health conditions in the current study. The majority of diseases in our review were neuromuscular conditions, whereas Kelly et al. (190), solely examined musculoskeletal conditions in their scoping review.

According to our findings, two PRISMS domains, "Social support" and "Information about resources", as well as content in the TEDSS's "Resource" domain were missing or hardly seen in programs. This contradicts the results of a recent systematic review, which found these domains to be frequently identified in effective interventions for chronic conditions (191).

Finally, as expected and in accordance with the results of prior systematic reviews, active patient participation was a key component of fatigue programs. We found that all but one program implemented two or more of the six domains of active patient participation. The most prevalent were "Homework" and "Goal setting." However, we were unable to compare our results to previous evidence since this review proposed these domains for the first time. Further research needs to be done to test the different domains of active patient participation in self-management programs.

Although it is unclear how many components/domains are needed or associated with positive patient outcomes, it has been suggested that inclusion of a greater number of components may benefit people to self-manage their long-term conditions (192). Comparing all programs, this study found that the inclusion of proposed components and their domains was most frequently reported for the active patient participation (60.46%) component followed by TEDSS strategies (56.46%). PRISMS domains were the least reported (39.64%) which was slightly less than the findings of a recently published systematic review (43%) (193).

In this scoping review, the primary objective was to identify the self-management components within fatigue self-management programs. As a result, the inclusion criteria

encompassed not only experimental designs but also various study designs that described fatigue self-management interventions. The findings of this research create a way to compare and contrast self-management fatigue interventions and examine mechanisms for change, which is a prerequisite to the comparison of different programs and their outcome. Future research using meta-analysis designs is warranted to further investigate the relationship between the highlighted self-management components and their impact on changes in outcomes.

3.6.1.3 Existing Programs Lack a Description of Self-Management Programs and Their Components. Determining the active ingredient(s) in self-management programs is a well-known gap in self-management research. A contributing factor is the lack of consistent reporting of program components and the extent to which they have been implemented (191). Related to this, as found in another review by Packer et. al., (2018), is the diverse ways self-management is conceptualized, and the underlying theories used to form self-management programs. These differences lead to heterogeneity in describing and comparing self-management programs. This also contributes to the inability of systematic reviews and metanalysis to compare and identify specific strategies and active ingredients of self-management programs that may result in better health outcomes (160). As a result, it is becoming increasingly important to synthesize and compare evidence on complex interventions such as self-management programs (194). To reduce the risk of incorrect conclusions and enable more accurate comparison among programs, comprehensive descriptions of programs and their active components is essential (158).

The intent of this review was to gain an in-depth understanding of a specific type of self-management program. Therefore, this specific focus led to the consequent small

sample size as a limitation, suggesting that future research should evaluate the value of the three components across more and different self-management interventions. This three-component framework may be a preliminary step toward developing a more systematic reporting framework for self-management programs or a more consistent definition and implementation of self-management programs.

3.7 Conclusion.

Overall, there was found to be a lack of information reported by authors about the included components of existing fatigue self-management programs which makes it difficult to compare them. Moreover, there is no commonly agreed upon framework to describe, report, or compare self-management programs. To overcome the challenges, this review selected three common self-management components to compare fatigue programs. It was found that the three-component framework is able to compare fatigue self-management programs developed based on similar or different underpinning theories and has the potential to be used as a tool for comparing programs in a more consistent and reproducible way. Consistent reporting and measurement of these three components holds potential to help understand the illusive mechanisms for change in self-management interventions.

3.8 Practical Implications

The most commonly included component and its domains found in fatigue self-management programs is active patient participation. All programs included 60.46% of the domains of this component. Among all programs, the most frequently reported domains of the three self-management components chosen for this study were strategies

in the “Activities” domain in the content component and three domains of the self-management support component: “Provision of/agreement “, “Regular clinical review “, and “Information about condition and/or its management”. Although evidence is yet growing to link all these to positive outcomes, frequency of use does provide insight into best practice fatigue self-management programs.

When selecting or designing self-management programs for implementation, therapists should look for and assess three areas: the content, active patient participation and self-management support strategies. This information, together with clinical judgement is needed to select the best programs for their client groups.

The TEDSS and the PRISMS are useful tools for therapists to assess, not just structured programs, but their own practice. For active patient participation, more evidence is required to assess the domains suggested in this review.

Acknowledgements

We would like to express our gratitude to Shelley McKibbon, our exceptional Information Services Librarian, for her substantial assistance and consultation. We would also want to extend our appreciation to Sabrena Jaswal for proofreading and editing this work.

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3.9 Appendices

Appendix A *Detailed Search Strategies*

Database	Terms	Records Retrieved
CINAHL [EBSCO]	#1: (fatig* OR tired* OR (energy N2 conserv*) OR exhausted OR exhaustion) #2: (MH "Fatigue") OR (MH "Cancer Fatigue") OR (MH "Fatigue Syndrome, Chronic") OR (MH "Mental Fatigue") #3: #1 OR #2 #4: (self W1 (care OR manag*)) OR “patient activation” OR “patient education” #5: (MH "Self-Management") OR (MH "Self Care") OR (MH "Patient Education") #6: #4 OR #5 #7: #3 AND #6	2,642
APA PsycInfo [EBSCO]	#1: (fatig* OR tired* OR (energy N2 conserv*) OR exhausted OR exhaustion) #2: ((self W1 (care OR manag*)) OR “patient activation” OR “patient education #3: (DE "Fatigue" OR DE "Chronic Fatigue Syndrome") OR (DE "Neurasthenia") #4: (DE "Self-Management" OR DE "Self-Care") OR (DE "Client Education") #5: #1 OR #3 #6: #2 OR #4 #7: #5 AND #6	1,119
Academic Search Premier [EBSCO]	#1: (fatig* OR tired* OR (energy N2 conserv*) OR exhausted OR exhaustion) #2: (self W1 (care OR manag*)) OR “patient activation” OR “patient education” #3: DE "FATIGUE" OR DE "CANCER fatigue" OR DE "CHRONIC fatigue syndrome" OR DE "MENTAL fatigue" OR DE "MUSCLE fatigue" #4: (DE "HEALTH self-care") OR (DE "PATIENT education") #5: #1 OR #3 #6: #2 OR #4 #7: #5 AND #6	1,434

Database	Terms	Records Retrieved
Medline [Ovid]	#1: (fatig* or tired* or (energy adj2 conserv*) or exhausted or exhaustion).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] #2: ((self adj1 (care or manag*)) or "patient activation" or "patient education").mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] #3: Self Care/ [MESH HEADINGS] #4: Self-Management/. [MESH HEADINGS] #5: Patient Education as Topic/ [MESH HEADINGS] #6: #2 or #3 or #4 or #5 #7: fatigue/ or mental fatigue/ [MESH HEADINGS] #8: Fatigue Syndrome, Chronic/ [MESH HEADINGS] #9: #1 or 7# or #8 #10: #6 and #9	2,455
Cochrane Library, Title/Abstract/keyword Search	(fatig* OR tired* OR (energy NEAR/2 conserv*) OR exhausted OR exhaustion) AND ((self NEAR/1 (care OR manag*)) OR “patient activation” OR “patient education”)	Trials: 1069 Cochrane Reviews:19

Appendix B Average *Inclusion of Self-Management Components and their Domains Across Programs*

Self-Management Components		Packer Managing Fatigue						CBT					Other							All Programs
Fatigue Programs		Blikman 2017	Kos 2016	Plow2020	Van Heest	Raina 2016	Total (/5)	Picariello 2018	Friedberg 2013	Ehde 201	Jason 2007	Total (/4)	O' Riordan 2017	Ream 2016	Kos 2015	Farragher 2019	Yates 2005	Jason 2007	Total (/6)	Total- all programs (/15)
TEDSS	Activities	1	1	1	1	1	**5**	1	1	1	1	**4**		1	1	1		1	**4**	**13**
	Internal strategies	1		1			**2**	1	1	1	1	**4**		1			1	1	**3**	**9**
	Social interaction strategies	1	1	1	1		**4**	1				**1**	1					1	**2**	**7**
	Healthy behaviours			1			**1**	1	1	1	1	**4**	1	1			1		**3**	**8**
	Disease controlling strategies	1		1	1		**3**		1	1		**2**	1	1	1		1	1	**5**	**10**
	Process strategies	1	1	1	1	1	**5**	1		1	1	**3**	1		1		1	1	**4**	**12**
	Resource strategies					1	**1**					**0**							**0**	**1**
Total (/7)			**3**	**6**	**4**	**3**	**57.85**	**5**	**4**	**5**	**4**	**64.28**	**4**	**4**	**3**	**1**	**4**	**4**	**47.6**	
Inclusion Average (%)			42.8	85.7	57.1	42.8		71.4	57.1	71.4	57.1		42.8	57.1	42.8	14.2	57.1	57.1		
Active Participation	Goal setting	1	1	1		1	**4**			1	1	**2**	1		1	1	1		**4**	**10**
	Problem solving	1	1			1	**3**			1		**1**		1		1	1		**3**	**7**
	Practice activities, experiment, discovery	1	1	1		1	**4**			1	1	**2**				1	1	1	**3**	**9**
	Homework	1	1	1	1		**4**	1	1	1	1	**4**	1			1		1	**3**	**11**

Self-Management Components		Packer Managing Fatigue						CBT					Other							All Programs
	Tracking, monitoring, self-evaluation		1				1	1	1	1	1	4		1	1		1	1	4	9
	Active discussion	1	1			1	3				1	1		1	1	1	1	1	5	9
Total/Program (6)			6	3	1	4		2	2	5	5		2	3	3	5	5	4		
Inclusion Average (%)			100	50	16.6	66.6	63.3	33.3	33.3	83.3	83.3	58.3	33.3	50	50	83.3	83.3	66.6	61.1	
PRISMS	Information about condition and/or its management	1	1	1	1	1	5		1	1	1	3	1	1	1	1	1	1	5	13
	Information about available resources						0					0					1		1	1
	Provision of/agreement	1	1	1	1	1	5	1	1	1	1	4	1	1	1	1	1	1	6	15
	Regular clinical review	1	1	1	1	1	5	1	1	1	1	4	1	1	1	1	1	1	6	15
	Monitoring of condition with feedback	1	1		1	1	4		1			1		1			1	1	3	8
	Practical support with adherence		1				1			1	1	2						1	1	4
	Provision of equipment						0					0							0	0
	Provision of easy access to advice or support when needed						0					0					1		1	1
	Training/rehearsal to communicate with healthcare professionals						0					0							0	0

Self-Management Components		Packer Managing Fatigue						CBT					Other							All Programs
	Training/rehearsal for everyday activities	1	1	1	1		4					0			1	1			2	6
	Training/rehearsal for practical self-management activities	1	1	1		1	4	1	1	1	1	4		1	1		1		3	11
	Training/rehearsal for psychological strategies			1			1	1	1	1	1	4						1	1/6	6
	Social support		1				1					0						1	1/6	2
	Lifestyle advise and support						0					0							0	0
Total (/14)			8	6	5	5	42.8	4	6	6	6	39.2	3	5	5	4	7	7	36.9	
Inclusion Average (%)			57.1	42.8	35.7	35.7		28.5	42.8	42.8	42.8		21.4	35.7	35.7	28.5	50	50		

TEDSS presents Taxonomy of Everyday Self-management Strategies; PRISMS presents the Practical Reviews in Self-Management Support

Appendix C Preferred *Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist*

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	Page 1
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	Page 3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	Page 4
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	Page5
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	Page5
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	Pages 5,6
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	Page 5
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	Page 31
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	Pages 6-7
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	Pages 7-8
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	Page 6, and page 22 (table 1)

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
Critical appraisal of individual sources of evidence	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	N/A
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	Pages 8-9
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	Pages 9 and 23 (Figure1)
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	Pages 9-10 and 24-27 (Table 2)
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	N/A
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	Pages 9-10 and 24-27 (Table 2)
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	Pages 9-12, tables 4, 5 and Appendix B
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	Pages12-16
Limitations	20	Discuss the limitations of the scoping review process.	Page 16
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	Page 16
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	Page 17

CHAPTER 4 - MANAGING FATIGUE IN PARKINSON'S DISEASE: PROTOCOL FOR A PILOT RANDOMIZED CONTROLLED TRIAL

This chapter is a manuscript that was published in the Canadian Journal of Occupational Therapy in March 2022. The article outlines the proposed detailed methodology of this PhD research.

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4.1 Abstract

Background. Fatigue is a disabling symptom of Parkinson`s disease (PD). *Managing Fatigue: A Six-Week Energy Conservation Intervention* was developed to improve the occupational performance of people with fatigue. Efficacy of this program has not been established in PD.

Purpose. This study will assess feasibility of the *Managing Fatigue: Individual Program* (*MFIP*) delivered via videoconference, the Randomized Controlled Trial (RCT) protocol, and the preliminary effectiveness of the MFIP.

Methods. A mixed-methods approach nested in a pilot RCT, randomizing 54 participants 1:1 to usual care or MFIP arms will be employed to evaluate the feasibility and preliminary effectiveness of MFIP. Qualitative and quantitative data will be collected simultaneously.

Implications. Results will identify evidence for establishing protocol requirements for a full-scale RCT. Knowledge of the effectiveness of the one-to-one videoconference delivery format of the program has the potential to enhance the accessibility and the quality of care of the PD population. ClinicalTrials.gov identifier: NCT04267107

Keywords. Occupational Therapy, Self-Management**,** Occupational Performance, Energy Conservation

4.2 Introduction

Parkinson`s disease (PD) is a progressive neurodegenerative condition caused by the loss of dopamine cells located within the basal ganglia (195). PD is associated with a variety of motor and non-motor symptoms (77) with fatigue being one of the most common non-motor symptoms. Fatigue impacts occupational performance, participation, and quality of life (13). The exact etiology of fatigue is unknown but it can arise at early or later stages of the disease (196).

Fatigue can impact everyday function and occupational performance (15). For example, fatigue is associated with reduced social participation, increased risk of social isolation and psychological distress (13). For people with PD, fatigue is also associated with early retirement and reduced working hours, sometimes resulting in financial distress and a diminished sense of productivity (14, 59).

Despite the impact of fatigue on health and daily life, it is under-recognized in many health care settings (79). Even when recognized, currently available meta-analyses and systematic reviews in PD report no definitive pharmacological solution (20, 133, 197), emphasizing the importance of developing and testing non-pharmacological interventions. Among non-pharmacological treatments, cognitive-behavioral therapy (CBT), exercise, and energy conservation interventions have been reported in the literature (136). However, the scarcity of high level RCTs hinders adoption of non-pharmacological interventions, especially in people living with PD (20, 133, 197). One of the most frequently studied fatigue interventions is the '*Managing Fatigue: A Six-Week Energy Conservation Course'. Managing Fatigue* is a client-centered occupational

therapy program which is aligned with the Person-Environment-Occupation (PEO) model (147). The program teaches and supports energy management strategies such as activity simplification, task analysis, environmental modification, communicating about fatigue, planning, and prioritizing (198).

The *Managing Fatigue* intervention was originally developed and evaluated in a face-to-face group format (26-28). Since then, it has been adapted for multiple delivery formats including teleconference (5), internet (29), and one-to-one formats (6, 7).

The one-to-one format has only been tested in two studies. Blikman et al., (6). using an RCT design, evaluated a twelve-session version of the program for people living with MS, while Van Heest (7) in a one-group pre-test, post-test design, evaluated a version with five out of the six program modules of the program (the last module was excluded) in people with chronic conditions (MS, fibromyalgia, cancer, and stroke).

Blikman et al., (2017), found no significant difference in fatigue between the experimental and information-only control groups as measured with the Checklist Individual Strength (CIS20r), the Modified Fatigue Impact Scale (MFIS), and the Fatigue Severity Scale (FSS). Moreover, no significant changes were found for social participation, measured with the Impact on Participation and Autonomy questionnaire (IPA), the Medical Outcomes Study Short Form 36 (SF-36), or the Rehabilitation Activities Profile (RAP). However, Van Heest (2017) found significant improvements in post-test fatigue measured by the Functional Assessment of Chronic Illness Therapy–Fatigue Scale; quality of life, measured with the Functional Assessment of Cancer Therapy–General; and self-efficacy, measured with the Self-efficacy for Performing Energy Conservation Strategies Assessment (SEPECSA).

The heterogeneity in the delivery of the program, as well as the differing outcome measures and populations in each of these studies makes the results difficult to interpret, and more studies are required to assess the one-to-one delivery approach of the *Managing Fatigue* intervention.

While the *Managing Fatigue* intervention has been tested extensively in people with MS, only one study has included people living with PD to date. Ghahari et al., (29), evaluated fatigue due to MS (n=74), post-polio (n=13), and PD (n=8) in an RCT design testing the internet format of the program. Results demonstrated marginal improvement in self-efficacy and significant reductions in stress compared to the non-intervention group. However, results were not reported separately for each condition.

In summary, prior studies have shown promising results for the *Managing Fatigue* intervention, however, there is limited evidence of effectiveness for people living with PD, and evidence for the one-to-one format is contradictory. The planned study will address these gaps by evaluating the feasibility of a one-to-one videoconference delivery format of the *Managing Fatigue* intervention, named "*The Managing Fatigue: Individual Program (MFIP)",* for people living with PD.

The MFIP consists of six weekly sessions adapted for one-to-one delivery via videoconferencing by our research team, which includes the original author of the *Managing Fatigue* intervention. Videoconference reduces geographic and transportation barriers and allows participants to benefit from the program while following public health restrictions imposed by the COVID-19 pandemic. Regulations enforced by the government in response to the pandemic have changed the landscape of healthcare practices. With many

jurisdictions now adopting virtual care and telehealth interventions as usual care, evidence of effectiveness of video conference-based interventions is needed more than ever (42).

4.3 Methods

4.3.1 Study Design and Ethics

A mixed-methods approach (199) nested in a pilot RCT, assigning participants in a 1:1 ratio to either usual care or MFIP arms will be employed. Qualitative and quantitative data will be collected simultaneously to evaluate the feasibility and preliminary effectiveness of the MFIP. Consistent with the feasibility/RCT pilot study design (200), we will assess two main aims. Aim 1 is to evaluate the feasibility of the MFIP and the pilot protocol in people living with PD. For this aim, the following research questions will be addressed:

- To what extent is the MFIP relevant, acceptable, and impactful from the perspective of people living with PD?
- How effective are recruitment strategies to include participants from a range of sociodemographic backgrounds?
- To what extent are the selected outcome measures acceptable to people living with PD?

Aim 2 is to assess the preliminary effectiveness of the MFIP, more specifically to answer:

- Are there any statistical differences among study outcome measures between the MFIP and usual care arms?
- What is the required minimum sample size for a future full-scale RCT based on differences between study arms?

As this is a feasibility study, there will be no primary versus secondary outcomes. The outcomes for evaluation of preliminary effectiveness will be occupational performance,

occupational balance, fatigue impact, quality of life, sleep quality, and self-efficacy. Results will be used to inform the primary outcome of any future RCT. This protocol has been approved by the Nova Scotia Health Research Ethics Board (ref: 1027048).

4.3.2 Participants

Estimating an attrition rate of 20%, a convenience sample of 54 participants (27 in each arm) will be recruited in Canada. We conducted both a sample size calculation and consulted previous literature to inform our estimation. To calculate the sample size, the type-1 error was set at 5% and the type-2 error at 20% for a power of 80%. Consulting previous relevant studies on fatigue in PD, effect sizes were only available for two of the outcome measures to be used in this study: The Multiple Fatigue Inventory (MFI) with an effect size of 0.664 (201) and the Canadian Occupational Performance Measure (COPM) with an effect size of 0.37 (202). Thus, using the minimum effect size reported for the COPM, a total sample size of 42 will be required. Considering a 20% attrition rate, 54 participants will be recruited. The calculated sample size also aligns with methodological reviews that recommend a sample size of 10 to 50 participants for pilot/feasibility studies (203-205).

The inclusion criteria for this study will be: ability to provide informed consent; age 18 or older; having been diagnosed with PD (self-report); a score of ≥4 on the FSS; ability to read and communicate in English; access to the internet and an electronic device (i.e., Smart cellphone, tablet, computer); and a private place for videoconferences. Exclusion criteria will be: previous completion of the *Managing Fatigue* intervention, or a co-morbidity that causes severe fatigue. Since participants are required to take an active role

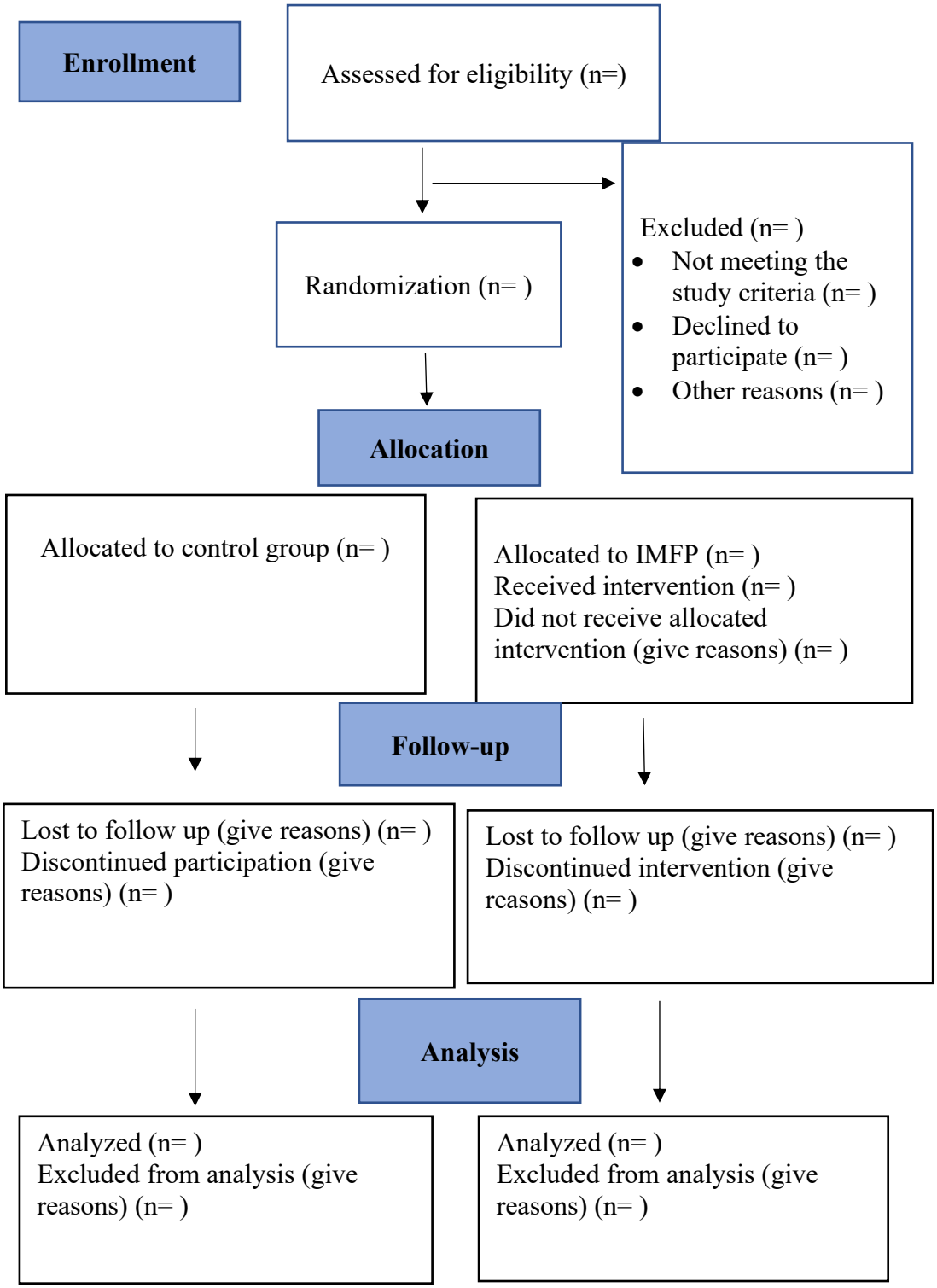
in the program, having severe cognitive impairment, demonstrated by a score of <13 on the Mini-Mental Status Exam (MMSE), will also be an exclusion criterion.

This study will recruit participants in several different ways: (a) web-based advertisements, (b) posters, (c) social media, (d) community advertising, and (e) word of mouth. Individuals who are interested in participating will contact the research team by email. Potential participants will first complete an email screening procedure to confirm they meet the preliminary eligibility criteria (are 18 years or older, live with PD, experience fatigue, and have access to an electronic device). Once confirmed, they will receive the study information and the consent form by email. Final eligibility will be confirmed after completion of the FSS and the MMSE during a secure videoconference. Consent will be confirmed electronically before any data collection. Participants will be informed that they may withdraw from the study at any time and for any reason. Whenever possible, reasons for withdrawals will be recorded.

After the screening process, a research staff member, not involved in the screening or data collection, will randomly assign eligible participants to either the usual care or the MFIP arms using sealed envelopes. After randomization, the research staff will assign participants in the MFIP arm to a therapist and will respond to any queries from participants. The assessor who is responsible for screening and data collection will be masked to group assignment. Study participants will be instructed not to share any information on group allocation with the assessor. The masking of the assessor will be monitored by documenting any information received during pre, post, and follow-up testing. A full CONSORT flow diagram is presented in Figure (2).

In this study, participants cannot be masked to their assigned group (MFIP versus usual care) as they will be actively involved in the sessions. Participants in the MFIP arm will receive the six-week MFIP in addition to their current healthcare services. Those in the usual care arm will not receive the MFIP and will continue receiving usual care. After completion of the study, participants in the usual care arm will have the option to receive the program's manual and attend an online, three-hour workshop on fatigue management held by occupational therapists at no cost. In this workshop, they will receive an abbreviated program that includes the program content and an introduction to the pre-session activities, in-session activities, and homework.

Figure 2 *CONSORT Flow Diagram*



4.3.3 Intervention

The MFIP consists of weekly sessions of structured discussion between an individual participant and a qualified occupational therapist. The MFIP is described in two manuals, one for participants and one for therapists. The content is similar to that in the original program; however, two additional topics have been added: sleep hygiene and cognitive fatigue. The content is arranged in six sessions: (a) the importance of rest and sleep; (b) communication and body mechanics; (c) activity stations; (d) priorities and standards; (e) balancing your schedule; and (f) course review and future plans. Content related to cognitive fatigue is threaded through all sessions. Each session includes three main parts: 1) pre-session where participants are asked to complete activities prior to the session so they are prepared for discussion with their therapists; 2) in-session activities/information where participants discuss and learn about topics, based on their priorities; 3) homework where participants practice at home to ensure they are building the skills discussed in sessions. Participants will be supported to evaluate and tailor the strategies suggested in the program according to their prioritized occupational performance issues. Each weekly session will be approximately 90 minutes, although therapists may adjust the pace depending on participants' needs and preferences. The goal is to support participants to complete the program within six to eight weeks.

Licensed occupational therapists, who complete an online training course designed for this study, will deliver the program. Therapists will be required to complete all modules and successfully answer all corresponding quiz questions. Therapists will have the opportunity to review content and re-take quizzes until all questions are correctly answered.

The therapist training covers: (a) history and development of the MFIP; (b) evidence of the effectiveness of the program; (c) fatigue; (d) an introduction to fatigue measurement; (e) energy management; (f) self-management and chronic disease management; (g) building self-efficacy; (h) motivational interviewing; (i) the transtheoretical model of behavioral change; (j) put it all together; (k) using secured videoconferencing; (l) Parkinson's disease; and (m) fatigue in Parkinson's disease.

4.3.4 Data Collection

Data for study outcome measures will be collected using Opinio Survey Software (206) a secure, university supported platform. Participants will complete questionnaires online during a videoconference call with the assessor.

Demographic information will be collected after participants are enrolled and have consented. Participants will answer questions regarding age, gender, years since diagnosis, living status, employment status, and any treatments they are currently receiving to manage their fatigue. Data collected for age, gender, years since diagnosis and living status will be used to assess the effectiveness of recruitment strategies to include participants from a range of sociodemographic backgrounds.

Data collection for aim 1 will consist of feasibility questionnaires and focus groups to understand the perspectives of participants in the MFIP arm. Two feasibility questionnaires were developed by our research team to assess, from the perspective of participants, the relevance, acceptability, and perceived impact of the program. Individualized links to the feasibility questionnaires will be sent by a research assistant to participants via email. Feasibility Questionnaire #1 will be completed weekly to evaluate

the relevance, acceptability, and perceived impact of the content of each session. Feasibility Questionnaire #2 will be completed at the end of the program and will assess relevance, acceptability, and perceived impact of the entire program. The response scale for both is a five-point Likert scale (207). Collected data will be exported to Stata: Software for Statistics and Data Science (208).

After completion of the program, 15 participants will be recruited to take part in one of three focus groups (n=5 per group). Selection of participants will be based on maximum variation sampling accounting for disease duration, fatigue severity, and gender. Participants will be provided additional information about the focus groups and will be asked to provide a separate informed consent before participation. An experienced research assistant will lead the focus groups via a secure videoconference using a prepared interview guide. Participants will be encouraged to discuss the feasibility of the program including relevance, acceptability, barriers to completion of the program, acceptability of study measures, and perceived impact/changes.

To evaluate the effectiveness of recruitment strategies for future RCT designs, the following data will be collected and analyzed: how participants learned about the program; proportion of participants who withdraw or are lost to follow-up, and, where possible reasons for withdrawal; and sociodemographic characteristics associated with each of the above.

To assess the preliminary effectiveness of the MFIP (Aim 2), data will be collected using standardized outcome measures administered at baseline, following completion of the program, and three months after completion. The COPM will be conducted using the

standard interview-based protocol during a synchronous videoconference call. All other study measures are self-report surveys and will be completed online. To our knowledge, only the Occupational Balance Questionnaire (OBQ) and the Pittsburgh Sleep Quality Index (PSQI) have been used online. Formatting of the paper versions of all measures (COPM excluded) selected for this study, will be faithfully reproduced on the online platform. To maximize similarity to the original method of administration, the assessor will be available via videoconference during completion to respond to participants' questions.

4.3.5 Study Outcome Measures

The outcome measures were selected based on the content and expected impact of the program. In addition, studies that employed the *Managing Fatigue* intervention or focused on fatigue in people living with PD were reviewed to identify potential outcome measures.

Occupational Performance. The *Managing Fatigue* intervention was originally developed to increase the occupational performance of people who experience severe fatigue. Occupational performance will be measured with the Canadian Occupational Performance Measure (COPM). The COPM is a standardized, client-centered, occupation-focused measure administered using a semi-structured interview. It measures perceived occupational performance and occupational satisfaction (209).

Respondents will identify three to five self-selected occupational performance issues and then rate each one on a 10-point Likert scale. The average performance and satisfaction scores will be calculated by summing individual occupational issue scores then dividing by the number of issues (209). The psychometric properties of the COPM have been confirmed in chronic conditions (210, 211). This measure is responsive to change in

individuals with chronic conditions (N=150) (212) with high sensitivity (209), and has acceptable test-retest reliability for both performance and satisfaction scores (ICC=0.63 and 0.84 respectively). The COPM was developed based on the Canadian Model of Occupational Performance (209) and has previously been used as a primary outcome in an RCT evaluating an intervention for people with PD (213). In that study, the COPM was shown to be sensitive to change and able to detect significant differences between groups (213).

Fatigue Impact. Fatigue impact will be measured with the Multidimensional Fatigue Inventory (MFI)) (214). The MFI is a self-report fatigue tool with 20 items measuring five dimensions: general fatigue, physical fatigue, mental fatigue, reduced motivation, and reduced activity. Elbers et al., (119) evaluated the reliability and validity of the MFI in the PD population (N=153). After combining general fatigue and physical fatigue dimensions they reported that the reliability and validity of the four-domain scale (physical fatigue, mental fatigue, reduced motivation, and reduced activity) were higher than the original five-domain scale. The MFI shows reliable internal consistency (Cronbach's alpha >0.80) and has construct validity compared to a Visual Analogue Scale measuring fatigue ($0.22<r<0.78$) (214). The MFI is the only multi-dimensional measure recommended by the Movement Disorders Society for PD that also assesses the multidimensional aspects of fatigue in this population (150).

Occupational Balance. Occupational balance will be measured with the Occupational Balance Questionnaire (OBQ) () (215). The OBQ is an 11-item measure, developed to assesses individuals' satisfaction and perception with the amount and variation of meaningful occupations. The OBQ aligns well with the expected outcomes of the MFIP

related to scheduling, planning, and prioritizing activities.

The OBQ measures satisfaction with the amount of time that a person takes to accomplish tasks. It uses a 4-level ordinal response scale for each item ranging from 0 “completely disagree” to 3 “completely agree”. The OBQ total score ranges from 0 (no occupational balance) to 33 (maximum occupational balance). The psychometric properties of the OBQ have not been explored in PD. However, in the general population, it has shown high internal consistency (Cronbach’s alpha= 0.936) and test, re-test reliability (Spearman’s Rho= 0.926) for its total score (N=67). Neither ceiling nor floor effects were reported with this measure (215).

Quality of Life. Quality of life will be measured with the Parkinson`s Disease Quality of Life-8 (PDQ-8) (216). The PDQ-8 is the short-form of the Parkinson Disease Questionaire-39 which assesses the impact of PD on HRQoL over the past month. The PDQ-8 is a summary index with eight items, each representing one dimension of the PDQ-39. These items are mobility, activities of daily living, emotional well-being, stigma, social support, cognition, communication, and bodily discomfort. It uses a 0-4 response scale and scores are summed, then converted into a percentage. Lower scores indicate better quality of life (217). Psychometric properties have been confirmed in several studies (63, 216, 218). Studies by Franchignon et al., (218) and Tan et al., (219) demonstrated internal consistency (Cronbach's alpha 0.72, 0.81) and construct validity between PDQ-8 and the measure of autonomy and participation (IPA-I) (rs>0.50) and other clinical PD-specific measures (UPDRS-ADL, UPDRS-ME, HY, and SE), (rs = 0.30–0.50).

Sleep Quality. Sleep quality will be measured with the Pittsburgh Sleep Quality Index

(PSQI) (220), the most common assessment tool used to evaluate sleep quality (221). This 19-item self-report scale measures: subjective sleep quality, sleep latency, sleep duration, sleep efficiency, sleep disturbance, use of sleep medication, and daytime dysfunction (220). Component scores range from 0 (no difficulty) to 3 (severe difficulty) and are summed to produce a global score (range 0 to 21). Higher scores indicate worse sleep quality. A meta-analysis by Mollayeva et al. (221) demonstrated that the PSQI has good internal consistency based on Cronbach's alpha, strong reliability and validity, and moderate structural validity in a variety of samples. This review found that the reported Cronbach's alpha coefficient met the cut-point for a positive rating for within- and between-group comparisons (ranging from 0.70 to 0.83). The PSQI has been used as an outcome measure to test the effectiveness of the Managing Fatigue intervention for MS population which was able to detect a significant change in sleep quality (27).

Self-Efficacy. Self-efficacy will be measured by the Self-efficacy for Performing Energy Conservation Strategies Assessment (SEPECSA). The SEPECSA was developed based on the *Managing Fatigue* intervention content for a prior study and measures the individual's self-confidence to perform the strategies they learned in the program (34). The item response scale ranges from 1 (not at all confident) to 10 (completely confident). The final score is the mean of item scores. Liepold & Mathiowetz (34) demonstrated in a study with people with MS that the SEPECSA has high test and retest reliability (r = 0.776, ICC =0.771), good validity, and very high internal consistency (Cronbach's alpha =0.953). The SPECSA has not yet been used with the PD population, however, it has been used in previous similar studies (29, 222).

Two additional measures will be used to assess disease severity (measured by Estimated

Hoehn and Yahr scale) and depression (measured by Geriatric Depression Scale). The GDS-15 is a short, yes/no self-report measure used to screen for depression in the elderly. Although not extensively tested in PD, it appears to have adequate discriminant validity for a diagnosis of major and minor depressive disorder in PD at a cut-off of 4/5 (223). The Estimated Hoehn and Yahr scale (HY) are a widely used clinical rating scale that identifies the broad categories of motor function in PD (224).

4.3.6 Data Analysis

Focus groups will be audiotaped and data transcribed verbatim. Any potentially identifying information will be removed prior to analysis. Data will be analyzed using the six-stage content analysis framework by Braun and Clarke (225). The text will be coded verbatim without changing the meaning. Coded material will be categorized semantically until themes emerge. Codes and themes will be reviewed and refined until the final distinctive themes can be created.

Quantitative data will be downloaded from Opinio to Stata. After cleaning, data will be examined for skewness, outliers, and systematic missing data. Extreme outliers, defined as greater than ±2SD from the mean, will be removed if they are less than 5% of all data (226).

Data from the Feasibility Questionnaires will be analyzed using descriptive analysis, including frequencies and proportions for categorical data, and means and standard deviations for continuous data. Baseline data on age, depression, and disease severity will be examined for equality of groups. In the case of significant differences between groups in any of these variables, a series of sensitivity analysis will be used to test the effect of the

variables.

Because all measurements will be completed during a videoconference call and under the supervision of the assessor, we do not expect significant missing data at the item level. Any missing data, not at random, will be managed by following the protocol of each measurement tool or imputed using mean substitution. If the maximum number of missing items has not been included in the measurements' protocol, it will be set at 20%.

A general linear mixed model will be used to assess the preliminary effectiveness of the outcome variables: fatigue impact, occupational balance, occupational performance, quality of life, sleep quality, and self-efficacy. The models will include the group assignment variables, time and interaction of time and group. We will conduct analyses using both intention-to-treat and per-protocol principles. Similar to previous studies studies (227), for per protocol analysis, participants having complete data or received at least four intervention sessions will be included in the analysis.

Data from outcome measures will be used to calculate effect sizes to estimate the preliminary effectiveness and the required sample size for future RCTs and evaluate the sensitivity and responsiveness of the study outcome measures. Measurement tools with the highest effect size and the smallest significance level will be defined as the most sensitive measures (228). Measurement tools with higher levels of mean variability at baseline will have a smaller effect and, therefore, will be defined as less responsive measures (229). The sensitivity and responsiveness of measurement tools alongside their acceptability, obtained from the focus group data, will be used to identify the most suitable outcome measures for the future RCT.

4.4 Discussion

The effectiveness of fatigue interventions in people with PD has been investigated in only a few RCTs and fatigue has rarely been a primary outcome measure (20, 73, 133). Moreover, the use of small sample sizes, variable definitions for fatigue, and a lack of consistency in accounting for confounding variables, such as depression and sleep problems (130), makes it difficult to draw reliable conclusions from these studies. Therefore, there is a need for more robust studies that explore how best to manage fatigue, especially in people living with PD. In order to conduct a rigorous RCT, robust preliminary data on outcome measures, design, and feasibility of delivering a program are required. This preliminary pilot study is specifically designed to address these issues and provide answers needed for future larger and fully powered studies.

The *Managing Fatigue* intervention has been proven to be effective in reducing the impact of fatigue associated with neurological conditions, thus there is potential for a similar result for people with PD. Recommendation for clinical use, however, requires evidence of effectiveness from RCTs. Further, our planned study will put emphasis on occupation related outcomes which have not been adequately measured and evaluated in previous studies that tested the *Managing Fatigue* program. To best of our knowledge, only Veenhuizen et al., (230) evaluated occupational performance as an outcome for this program. Our study will use occupational performance and occupational balance as study outcomes to better allow a more accurate evaluation of the impact of the program on everyday activities and occupational functioning of participants.

In our pilot RCT design, we are using a one-to-one delivery approach via videoconference.

Although one-to-one delivery allows therapists to focus on tailoring the program to participants' distinct priorities and situations, and improves access for people in remote areas, or where lower population density makes forming groups more difficult, there is still insufficient evidence to support its benefits in the PD population. This study will therefore contribute to the body of evidence examining the feasibility of using a one-to-one delivery approach.

This study will also contribute to the growing body of knowledge related to virtual care. Since the COVID-19 pandemic in 2020, the feasibility of using telehealth has been amplified. Therefore, developing and evaluating studies to assess the delivery of health programs using videoconference is becoming more relevant. However, using online communications and delivery approaches, when conducting studies, introduce additional challenges. For example, in the current study, there are no validated online versions for many of the study's self-report outcome measures. Thus, we had to adapt these tools for online administration. This is a potential limitation of our planned study, but we will implement measures to mitigate this limitation including consulting with the the authors of the outcome measures and ensuring that assessors will be available via videoconferencing to help address any issues or questions that might arise for participants. Future studies to validate online versions of outcome measures will be needed to fully understand the effectiveness of virtual care.

Another possible limitation of using the videoconference delivery is participants' unfamiliarity with the technology, potentially causing frustration and/or fatigue, especially in older individuals. Although these challenges exist, in this pilot study we will learn about the perspectives of people with PD regarding the acceptability of the online versions of the

intervention and the study outcome measures. This will contribute to our understanding of the face validity of the online measures.

4.5 Conclusion

The proposed study will evaluate the feasibility of the MFIP in people living with PD. If the known beneficial effects of this program for other conditions extend to the PD population, this research will provide the preliminary evidence needed to support further studies that will help guide integration into the process of care for people living with PD.

Key Messages

- Effective fatigue management interventions are not currently available for people with PD.
- Testing the feasibility and preliminary effectiveness of the *Managing Fatigue: Individual Program* will provide occupational therapists with evidence to support their practice in addressing fatigue with individuals with PD.

Acknowledgments

We would like to thank our research team at the International Chronic and Complex Condition Research Group (IC3RG) at Dalhousie University for their assistance in running this trial. We would also like to thank the occupational therapists who will deliver the program and the participants who will give of their time to help improve the lives of others.

CHAPTER 5 - MIXED-METHOD EVALUATION OF THE INDIVIDUAL PACKER MANAGING FATIGUE PROGRAM: PERSPECTIVES OF PEOPLE WITH PARKINSON`S DISEASE

5.1 Abstract

Background: Fatigue is a common symptom in Parkinson's Disease (PD). Limited treatment options are available to address it.

Objectives: The aim was to explore the feasibility of the individual version of the *Packer Managing Fatigue* program for people with PD (PwPD).

Methodology: A concurrent mixed-method design collected data from 12 adults with PD through videoconferencing using Zoom for Healthcare.

Findings: Five themes emerged: program is helpful; strengths of the program; areas for improvement; individual online delivery is feasible; and more support from OT would be helpful. Quantitative findings confirmed feasibility with high ratings on questionnaires and confidence in using learned strategies.

Conclusion: The findings support the use of the *Packer Managing Fatigue* program for PwPD and provide insight into their unique needs. Future studies may investigate tailoring the program to address PD-related fatigue and its effectiveness.

5.2 Introduction

Fatigue is the most common non-motor symptom of Parkinson's disease (PD), a neurodegenerative disorder that presents with a wide range of motor and non-motor symptoms (16, 150). More than 50% of people with PD (PwPD) experience fatigue (16, 150), affecting occupational performance and quality of life (13, 107). Fatigue is a major impediment to completing a broad range of daily activities, including self-care and leisure (231), as well as employment and productivity (232). Reduced participation can diminish social engagement and increase the risk of social isolation and psychological distress for PwPD (13).

The high prevalence and significant impact of fatigue have driven the development and evaluation of fatigue interventions for PwPD (128). Based on currently available meta-analyses and systematic reviews, pharmacological management of fatigue is limited (20, 133, 197). Among non-pharmacological approaches, *Managing Fatigue: A Six-Week Course for Energy Conservation* (2) was one of the first programs developed to help people with fatigue due to a neurological condition. The original program was a six-week, group-based intervention developed in 1995 for delivery by occupational therapists (OTs); an updated second edition, called *Packer Managing Fatigue: A Six-Week Group Self-Management Program* is now available (3). An individual version is also available and developed by original author and their research team (1).

The program supports patients to learn and practice energy management strategies to reduce the impact of fatigue on daily activities and to optimise occupational performance. Strategies include balancing energy, budgeting and expenditure such as: how to rest; task

analysis and activity simplification; environmental modification; communicating about fatigue; planning, prioritizing and completing activities (2).

Since 1995, various adaptations and delivery formats of the *Managing Fatigue* program have been developed, including group (4), teleconferencing (5), and in-person individual versions (6, 7, 30). Effectiveness has been demonstrated in conditions such as multiple sclerosis (MS) and neuromuscular diseases (28, 31-34, 233). However, little is known about its feasibility and effectiveness for PwPD (29). The individual delivery method was the main focus here as it is more feasible in clinical settings and allows for maximum patient tailoring. Inconsistent findings have been reported in previous studies evaluating individual formats of the program. This may be attributed to variations in the implementation of the program, including changes to session content, number and duration that deviate from the original protocol (32). For instance, one RCT evaluated a 12-session program for effectiveness in people with MS. No significant difference in fatigue or participation was found between the experimental and information-only control groups (6). In contrast, another study used only five modules of the original group program, delivered in 4-6 individual sessions, and found a significant improvement in fatigue impact and quality of life (7).

This study evaluated a research version of the new individual protocol that the original author and her research team developed in 2023 (1). It was delivered using synchronous videoconferencing due to the COVID-19 pandemic. This version increases the opportunity to tailor the content to each individual, while the videoconference format improves accessibility in remote areas. The research objectives were to explore, from the perspective

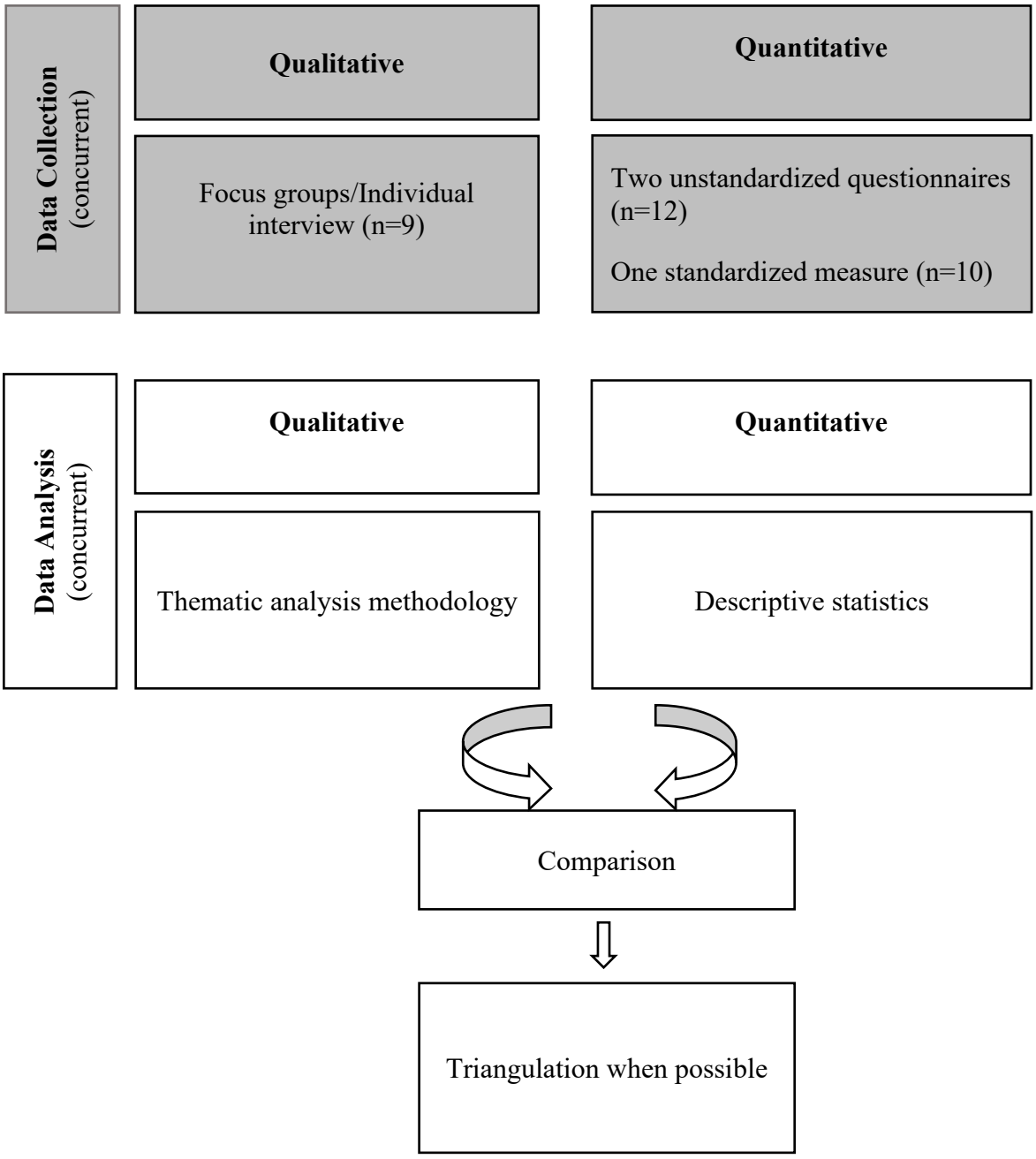
of PwPD, the perceived impact on daily life activities, relevance, feasibility of delivery and content, and perceived confidence to use the skills and strategies learned in the program.

5.3 Materials and Methods

5.3.1 Study Design

The current study is part of a larger pilot study (186) with 25 PwPD testing the *Packer Managing Fatigue: The Individual Self-Management Program* (1). Using a concurrent mixed-methods approach (234), both qualitative and quantitative data were collected through surveys, interviews, and focus groups (see Figure 3). The mixed methods design was employed to ensure comprehensive data collection and to draw more robust conclusions. Qualitative data provided insight into individuals' perceptions, while quantitative data added validity to these findings. By combining both types of data, a more nuanced interpretation can be achieved. The two strands of data collection were implemented separately. Triangulation was carried out during the interpretation phase, where the results from each strand were compared, synthesized, and used to draw conclusions (235). The research protocol was approved by the the Nova Scotia Health Research Ethics Board (ref: 1027048). To report the findings, this study used the checklist for mixed method research manuscript preparation and review proposed by Lee et al. and is attached as an appendix (Appendix A) (236).

Figure 3 *Design of Mixed-Method Research Design*



5.3.2 Participants and Recruitment-

As part of the larger study, participants in the intervention group completed the questionnaires used in this study. They were also invited to take part in a qualitative interview or focus group. To participate in the main study, all participants had to reside in one of two Canadian provinces, Ontario, and Nova Scotia, due to OT licensure regulations. All were adults with a self-reported diagnosis of PD who scored 4 or higher on the Fatigue Severity Scale (FSS), could read and speak English, and had access to the internet, an electronic device, and a private place for videoconferences. Participants with scores of 13 or lower on the Mini-Mental Status Exam (MMSE), indicating severe cognitive difficulties, were excluded. To participate in this sub-study, participants had to be a participant in the intervention group of the main study.

To recruit participants for the focus groups and individual interviews, an information sheet for this sub-study and a consent form were sent to those who completed the program in the intervention group of the main study. Those who provided consent were contacted to schedule an interview based on their preference. Participants were informed that they could withdraw from the study at any time without reason.

5.3.3 Study Intervention

The *Packer Managing Fatigue: The Individual Self-Management Program* consisted of six semi-structured individual sessions. This individual version of the program is manualized for occupational therapy interventionists and enhanced with a dedicated participant manual. The content remains consistent with the original program and incorporates two additional topics, sleep hygiene and cognitive fatigue management. Sleep hygiene

education is integrated into the first session while cognitive fatigue management strategies are incorporated throughout all sessions. Content in sessions one to five focuses on: (1) the importance of rest and sleep, (2) communication and body mechanics, (3) activity stations, (4) priorities and standards, and (5) balancing your schedule. Session six included a program review and a discussion of future plans.

During each session, delivered using the Zoom for Healthcare platform (237), a licensed OT assisted participants in trialling, evaluating, and adopting fatigue management strategies relevant to their daily life. Each session had three main components: 1) pre-session activities to prepare participants for in-session discussion; 2) in-session activities and discussions focused on potential skills and strategies to save energy and spend it wisely, problem-solving, and action-planning; and 3) homework activities designed to test and trial strategies introduced during the session. Homework activities were initiated during the session and completed independently following the session and were tailored to the individuals. Successes and challenges were discussed in the next session. Each weekly session was expected to take about 90 minutes. However, OTs were instructed to adjust times depending on patient needs and to complete the entire program in 6–8 weeks.

Prior to working with clients, OTs completed a 15-module, asynchronous online training course of approximately 3-4 hours (Table 6) that included embedded quizzes. A fidelity checklist, designed by the research team, was completed by therapists after each session.

Table 6 *Overview of Online Training for Occupational Therapists Delivering the Packer Managing Fatigue Program*

Course	Modules	Content
Facilitating the Managing Fatigue Program	Self-Management	The Role of Self-Management in Chronic Disease Management; Taxonomy of Everyday Self-Management Strategies; Self-Management in Managing Fatigue; The Role of The Practitioner in Supporting Self-Management
	Building Self-Efficacy	Define Self-Efficacy; Factors Affecting Self-Efficacy; Strategies to Build Self-Efficacy; Building Self-Efficacy Using the *Packer Managing Fatigue* Program.
	Behavioural Change	Transtheoretical Model of Behavioural Change; Stages of Behavioural Change; Stages Statistics; Context of Fatigue
	Motivational Interviewing	Key Features of Motivational Interviewing; Facilitate Changes and Types of Questions Used in Motivational Interviewing; Power Balance in Motivational Interviewing
	History and Development	The Purpose of the Program; The Target Population(s) For the Program; The Topics Covered in the Program.; The Rationale for Including Topics in The Program; The Similarities and Differences Between What Is Covered in The Program and What the Practitioner Typically Cover in Their Clinical Practice
	Evidence of Effectiveness	The Different Formats of The *Managing Fatigue* Program; Where Has the Program Been Delivered? Who Does the Program Help? What Are the Program Outcomes?
	Fatigue	Experiences Of Fatigue; Definitions and Types of Fatigue; Factors Influencing Fatigue; Measuring Fatigue; Occupational Therapy in Fatigue
	Energy Management	Introduction To Energy Management; Explaining Energy Management; Energy Management Strategies
	Concepts and Practice	Program Layout; Program Content; How Psychoeducational Groups Develop Over Time; Facilitator's Role
	Apply Your Skills	Scenarios And Questions/Answers
Managing Fatigue Online	Using Videoconferencing for Delivery	Videoconferencing: The Basics; Technology; Set Up for Success
	Zoom - What, When, and How	Schedule Visits with Participants; The Basic Functions of Zoom; Record Sessions on Zoom
	When Things Go Wrong	Troubleshoot and Deal with Challenges in A Virtual Environment; Handling A Missed Visit; Handling Adverse Events;

Course	Modules	Content
Managing Fatigue in Parkinson`s disease	Parkinson`s Disease	Parkinson's Disease: Overview; Motor Symptoms; Non-Motor Symptoms; Impact of Parkinson's Disease; Severity and Progressions of Symptoms; Response Fluctuations in Parkinson's Disease; Occupational Therapy and Parkinson's Disease
	Fatigue in Parkinson's Disease	Overview; Causes of Fatigue; Contributing Symptoms to Fatigue in Parkinson's Disease: Cognitive Difficulties; Mental and Physical Fatigue in Parkinson's Disease; Impact of Fatigue in Parkinson's Disease

5.3.4 Data Collection

As part of the original study (188), participants in the intervention group (n = 12) completed the questionnaires for this sub-study using Opinio Survey Software (206). Virtual interviews and focus groups (depending on participant preference) were conducted using Zoom for Healthcare which enables compliance with Canadian data protection regulations, such as the Personal Information Protection and Electronic Documents Act (PIPEDA) and the Personal Health Information Protection Act (PHIPA) (238).

Qualitative Data Collection: An experienced researcher (SJ), not involved in delivering the interventions, scheduled, and conducted individual interviews and focus groups at mutually agreed-upon times. An interview guide was developed to explore the perspectives and personal experiences of PwPD who participated in the program. Semi-structured, open-ended, non-directional interviews were conducted, during which participants were encouraged to discuss the feasibility of the program's content and delivery, barriers that prevented them from fully applying the skills learned in the program to their daily lives, the perceived impact of the program, and modifications they would make to the program.

Quantitative Data Collection: As part of the main study, participants completed demographic questionnaires at baseline. The Fatigue Severity Scale (FSS) (122), the Mini-

Mental State Examination (MMSE) (238), the estimated Hoehn and Yahr scale (HY) (224), and the Geriatric Depression Scale: Short version (GDS-15) (239), were used to assess fatigue severity, cognitive status, disease stage, and depression, respectively.

To assess the feasibility of the program, the research team developed two feasibility questionnaires (see Appendix C), one administered weekly and one at the end of the program. Participants' perceived confidence in using the energy conservation strategies taught in the program was measured using the Self Efficacy for Performing Energy Conservation Strategies Assessment (SEPECSA) (34). Table 7 summarizes the characteristics and timing of all questionnaires. The main study protocol (186) includes further details on the properties of these measures.

Table 7 *Measurements in the Study and their Properties.*

Questionnaires & description	Measurement timing
Feasibility Questionnaires: Two brief questionnaires assessed the program's relevance, substance, design, delivery, and perceived impact. A 12-item questionnaire, completed at the end of the six-week intervention, assessed overall program feasibility, while a 10-item questionnaire was completed weekly to assess feasibility of each session. Both utilized a five-point Likert scale (1= "strongly disagree" to 5= "strongly agree") **(207)**. Scores of three and above were considered feasible.	At completion of each session and completion of program
The SEPECSA (34): A 14 items questionnaire, based on a worksheet in the original *Managing Fatigu*e program, and further developed by Mathiowetz and colleagues (34) , the SEPECSA measures self-confidence to perform strategies learned in the program. The item response scale ranges from 1 (not at all confident) to 10 (completely confident). The final score is the mean of the item scores.	At the completion of program,
The GDS-15 (239): A short, yes/no self-report measure the GDS-15 is used to screen for depression in the elderly. Adequate discriminant validity for a diagnosis of major and minor depressive disorder in PD was found**(223)**. Scores of 0-4 are considered normal; 5-8 indicate mild depression; 9-11 indicate moderate depression; and 12-15 indicate severe depression.	At baseline
The self-report HY (224): This popular clinical rating scale classifies motor function in PD into five stages : (1) one-sided symptoms only, minimal disability; (2) both sides affected, balance is stable; (3) mild to moderate disability, balance affected; (4) severe disability, able to walk and stand without help; and (5) confinement to bed or wheelchair unless aided. We used an adapted version in which participants self-report the stage of the disease.	At baseline
The FSS (122): A self-report scale that contains nine items on physical, mental and social aspects of fatigue. These are rated on a seven-point Likert scale from completely disagree (1) to 'completely agree (7). A score of 4 or more indicate severe fatigue.	At baseline
The MMSE (238): A screening tool (30 items) for cognitive function that evaluates attention and orientation, memory, registration, recall, calculation, language, and ability to draw a complex polygon. Scores of 24 and above indicate normal cognitive functioning.	At baseline

SEPECSA: Self Efficacy for Performing Energy Conservation Strategies Assessment; GDS-15: Geriatric Depression Scale: Short version; HY: Hoehn and Yahr scale; FSS: Fatigue Severity Scale; MMSE: Mini-Mental State Examination (MMSE)

5.3.5 Data Analysis

The study used descriptive statistics to examine demographic and clinical features and data from questionnaires. These statistics included mean, SD, quartiles (median and 25th and 75th percentiles), and outlier analysis.

All interviews were recorded and transcribed verbatim using NVivo transcription software (240). A research assistant then reviewed all transcriptions and removed any potentially identifying information. Thematic analysis, combining both inductive and deductive coding, was used to analyze interview and focus group data (241). The coding process was primarily guided by Creswell & Creswell's work in 2017 (235). The first author (NA) created a list of a priori codes to organize the findings. A priori codes were based on the research questions, specifically the feasibility of the program content and manual, facilitators, and barriers to completing the program, perceived effectiveness of the program, PD-specific needs, and future considerations to improve the program for PwPD. The applicability of the codebook was tested by two coders, including the first author. No modifications were required, and the codebook was reproduced in the NVivo coding software. To ensure inter-coder reliability, the two coders independently coded two transcripts and discussed conflicts until consensus was reached. The coded data were exported from NVivo, further clustered manually, and treated as conceptual categories, allowing connections to be made and relationships to be explored using constant comparative methods by the two coders. This promoted the emergence of distinctive themes and enhanced qualitative trustworthiness (242). Any disagreements between coders were discussed and resolved based on consensus. Once the data analysis was completed, the final list of themes with a summary of de-identified sample quotes was sent by email

to participants and the interviewer for validity review. No additional changes were suggested. The names used to report findings from the qualitative analysis are pseudonyms and were selected based on the most popular names in 2022.

5.4 Results

5.4.1 Demographics

Twelve participants in the intervention group of the main study completed the feasibility questionnaires; 10 completed the SEPECSA, and nine also participated in an interview (n=7) or a focus group (n=2). Quantitative results are reported as mean ± standard deviation (SD) unless stated otherwise. The majority of participants identified themselves as male (n =9), with an average age of 68.6 ± 9 years and a disease duration of 5.8 ± 3.4 years. Consistent with inclusion criteria participants reported severe fatigue (FSS = 5.4 ± 0.9) and normal global cognition (MMSE = 28.5 ± 1.3). The mean depression score was 6.2 ± 3.4, and disease stage ranged from 1-3 on the HY scale (median=2).

5.4.2 Findings From the Qualitative Data

A total of five themes clustered into three main domains emerged and are described below. The first domain was the Perceived Impact of the Program, which included one major theme: Theme 1- Program Was Helpful. The second domain was Program Content and Structure, which included two themes: Theme 1- Strengths of the Program, Theme 2- Areas for Improvement. The third domain was Support and Delivery, which included two themes: Theme 1 - Individual, Online Delivery is Feasible and Theme 2 - More Support from OTs Would Be Helpful. Detailed themes and example quotes are provided in Appendix B.

Domain 1: Perceived Impact of the Program

Theme 1: Program Was Helpful.

Participants reported the program to be beneficial. They perceived access to the program as a positive opportunity, given the lack of fatigue management resources in healthcare. They described it as a learning opportunity to adopt behaviours and strategies to manage their energy to complete their daily life activities.

> *I found the lessons on dividing tasks into smaller chunks rather than tackling a whole project at once particularly helpful. Before the program, I would often spend a whole or half day in the garden without taking breaks, and the next day I would feel completely exhausted. It didn't make sense to me, but the program taught me that it's much more sensible to take frequent breaks and rest, so that you have energy for the next day. When I started the program, I didn't realize the importance of breaking tasks into smaller pieces, but now I see how much it can help conserve my energy. (Sophia)*

Examples of these behaviours, reported by participants, were: task simplification; using assistive/adaptive devices; proactively planning and organizing tasks to manage their energy; realizing the importance of proper body posture and understanding that rest is a permitted and useful activity. Participants also gained more self-acceptance, optimism, and self-care motivation.

> *I used to put off things that should have come first, such as taking care of myself, so that you can actually have the energy to do your activities and to help you slow the progression of your disease, your stress, your sleep, your fatigue, and everything else. So yeah, that's very inspiring that it really helped do that. (Noah)*

Some participants reported experiencing decreased fatigue after learning how to say "no" and express their needs. Participants realized that communicating their concerns is important but found it challenging because of the belief that others may not fully understand or accept their situation.

> *I know it's important to communicate my needs, and while I try to share as much as I can, I don't usually go into detail about everything because it can become overwhelming. I usually stick to simpler things like saying, I take medication and eat food at these times. People can easily understand those things. Explaining things like fatigue can be more difficult to convey to others. (Jack)*

Even though the program was helpful, participants reported that their fatigue levels did not decrease significantly. Some attributed this to the progression of their disease and the timing of the study, which coincided with the COVID-19 pandemic. Sophia acknowledged this when she said, *"I think I still get fatigued and I know it is due to the progression of the disease, not the program."* Freddie added, "*The COVID-19 pandemic halted many of our activities while also adding new ones. Coping with the pandemic was tiring in itself.*"

Domain 2: Program Content, and Structure

Theme 1: Strengths of the Program.

Participants greatly appreciated the printed manual and described the inclusion of pre-session and homework activities as strengths of the program. These elements supported them to prepare for and practice program content and activities. Mohammad said, "*Having the physical manual was really helpful. I could go back and review it whenever I needed to. It made me feel confident that I had all-time access to the content*."

Most participants found the program relevant to their needs as they were experiencing fatigue. Therefore, since the program was specifically focused on fatigue management, it was important and relevant for them. Sophia said: *"Well, the word itself, fatigue, was like bingo for me to say this is relevant to me. I didn't know how really to deal with fatigue. That's what attracted me."*

Theme 2: Areas for Improvement.

Participants identified two potential improvements. They recommended a more user-friendly page structure and the incorporation of content pertaining to other factors that impact their fatigue. A few participants regarded the program as intensive, and found its structure and procedures complex, which made it difficult for them to locate particular topics in the manual. Ava found the program complex and said, *some of the manual material was a little complex. The table of contents, session priorities, and standards were confusing at first. But once I got the hang of it, I understood what was happening.*

Participants also wanted information and strategies to deal with additional factors that influence PD fatigue. Medication dose and timing are critical considerations for PwPD, as they impact fatigue and general functioning. Other factors identified were anxiety, mood swings, and poor physical fitness which participants felt were not adequately addressed in the program. For example, Ava mentioned, "*I find my energy level low, both physical and mental. There were no exercise components in the program".* Jack suggested, "*I have bad and good times, mostly based on my medications. I think some information on that would be helpful for patients."*

Although participants found the program relevant, they hoped to see a stronger connection to PD. This included the program's language, as well as the examples and scenarios presented in the manuals. Megan expressed this as, "*It was definitely relevant, but I think the program wasn't specifically designed for fatigue in PD, it was for fatigue in general*." Similarly, Ava noted: " *I kept looking for the word Parkinson's and it wasn't there. It was all MS*."

Domain 3: Support and Delivery

Theme 1: Individual, Online Delivery Is Feasible.

All participants believed that both individual and videoconference delivery were feasible, especially during the pandemic. Sophia stated, "*I think it's great that the sessions were just for me. Having a therapist in an individual session helped me learn more as she could pace things based on my needs*." A few participants mentioned that they would have preferred videoconference delivery even if the study had not been conducted during the COVID-19 pandemic as they found it more convenient and less stressful. Lucas described this as: *"In a virtual way, you're not overawed by the presence of another person, you can concentrate on the questions and topics. So, I am in favour of the virtual approach."* On the other hand, there were a few participants who said they preferred videoconference delivery because of the pandemic, as they found it safer. For example, Freddie, indicated a preference for in-person meetings not restricted by the pandemic. *"I think meeting over Zoom worked well. Obviously, it's much nicer to be in person. I like being in a room with my therapist together, so I could actually feel more involved."*

Theme 2: More Support from OTs Would Be Helpful.

Participants expressed a desire for additional support from their OTs and more follow-up after the program. Murphy said, "*Maybe a reminder or a phone call to review the materials I had to do would have helped me to keep up with the program*." Additionally, some participants expressed a desire for more feedback from their therapists. Mohammad said, "*I work better with more feedback than free flow."*

Participants also voiced a preference for a more tailored pace and duration of the sessions. Many recommended that sessions be prioritized or intensified based on individual participant priorities. Ava suggested shortening the sessions, "*The sessions were too long considering that it was meant to help people with fatigue. I think they need to be shortened".* Jack also added, "*It was all useful, however, I do not know if a different order would have been better. Perhaps it would be more efficient to begin with sessions that are more relevant to each individual."*

5.4.3 Findings From the Quantitative Data

All items on the questionnaires evaluating overall program feasibility (rather than session-specific feasibility) had a mean item score of three or higher (out of a possible five) [mean range: 3.5-4.3] (see Appendix B). Consistent with the thematic data "tailoring" and "pace and duration" received the lowest scores (Figure 4, Appendix C).

In addition to overall program feasibility, participants rated the feasibility of each session individually. Overall, all sessions were considered feasible, with mean scores of 3.7 and above out of five. The first session was judged the least feasible, while the last session was considered the most feasible. The lowest feasibility score was for "appropriate pace" in session one, while the highest feasibility score was for the "encouraging aspects" of session six (Table 8).

Figure 4 *Tukey Box Plot for the Overall Feasibility of the Packer Managing Fatigue Program.*

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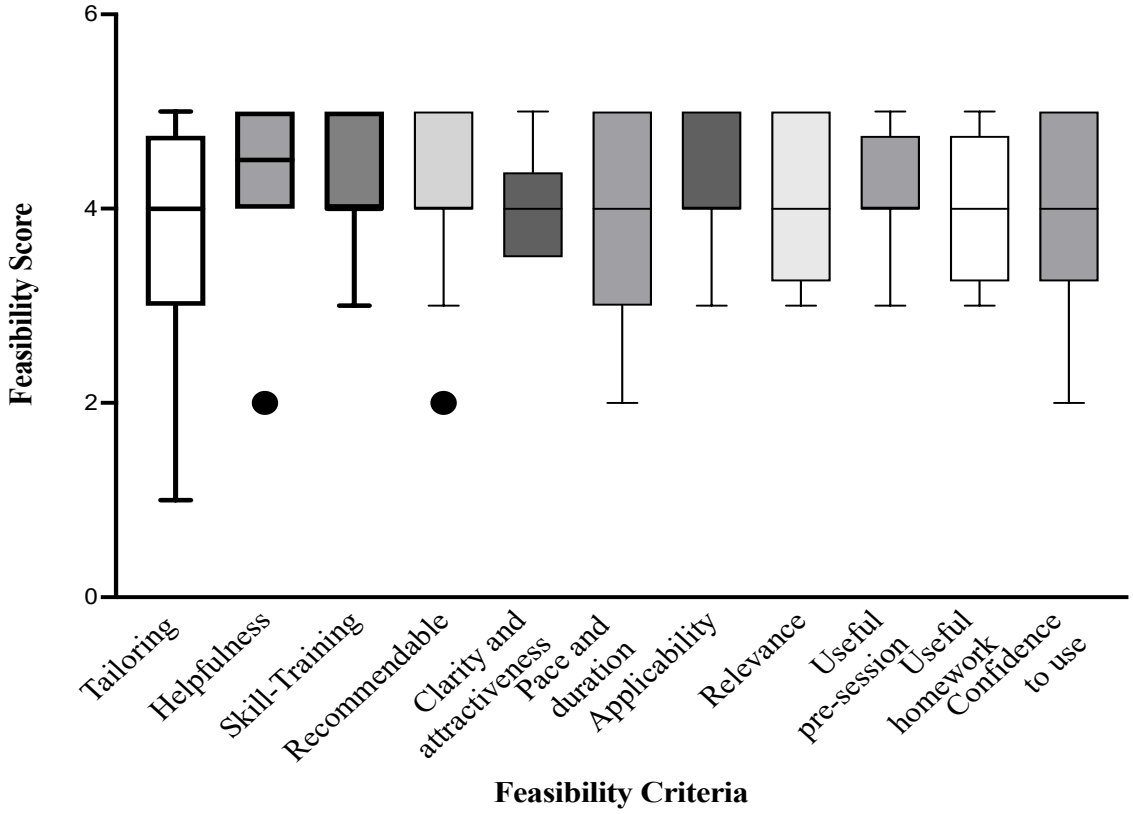


Table 8 *Feasibility by Session of the Packer Managing Fatigue: A Six-week Individual Self-Management Program*

	session 1	session 2	session 3	session 4	session 5	session 6
Criteria	*Results are represented as mean (SD)*					
Clear and organized	3.7(0.9)	4.0(0.7)	4.0(0.6)	4.3(0.5)	4.2(0.5)	4.1(0.5)
Tailoring	3.8(0.8)	3.9(0.7)	4.0(0.7)	4.1(0.6)	3.7(0.8)	4.0(0.6)
Appropriate pace	3.2(1.2)	3.7(0.8)	4.3(0.6)	3.9(0.6)	4.0(0.6)	4.0(0.6)
Encouraging	3.8(0.8)	4.5(0.5)	4.0(0.9)	4.3(0.6)	4.4(0.5)	4.6(0.5)
Relevant	3.7(1.5)	4.1(0.8)	4.0(0.8)	4.0(0.7)	4.0(0.5)	4.2(0.7)
Applicability	4.2(0.7)	4.2(0.7)	4.3(0.4)	4.1(0.8)	3.9(0.5)	4.0(0.3)
Confidence	3.0 (0.8)	4.0(0.9)	3.9(0.6)	3.9(0.7)	4.1(0.3)	4.3(0.5)
Average feasibility rating (Mean of means)	3.7(1)	4.1(0.7)	4.1(0.7)	4.1(0.7)	4.0(0.5)	4.2(0.5)

Perceived Confidence to Use Energy Conservation Strategies Learned in The Program:

Ten participants also completed the SEPECSA questionnaire following completion of the program. Participants were generally confident to apply the learned strategies. The mean scores for every strategy were above six out of possible 10 score [Mean range:6.4-8.5] (Table 9). Upon further investigation of the wide range of responses across all strategies (1-10), outlier analysis (Tukey's fences) showed that two participants consistently had the lowest scores (Table 9). They were older and had longer disease duration than the other participants. A sensitivity analysis, removing these two participants, resulted in a narrower range (4-10 vs. 1-10) and 95% CI (8.0 –8.5 vs. 7.2–7.9) across all strategies.

Table 9 *Confidence to Use Energy Conservation Strategies Based on the SEPECSA*

Variables	***All participants (N=10)***		***Excluding outliers(N=8)***	
	Mean (SD)	*Median (IQR)*	*Mean (SD)*	*Median (IQR)*
Identify Work Height	7.8 (2.6)	8.5(7-10)	8.3(2.4)	9.0(7.7-10.0)
Change Location of Equipment	8.3(1.5)	8.5(7.2-9.7)	8.6(1.1)	8.5(7.7-10.0)
Use Adaptive Equipment	8.2(2.1)	8.0(8.0-9.7)	8.8(0.9)	8.5(8.0-10.0)
Change Body Position	8.3(1.4)	8.0(8.0-9.0)	8.6(0.9)	8.5(8-9.2)
Eliminate Activity	6.8(2.6)	7.5(5.2-8.7)	7.7(1.7)	8.0(6.0-9.2)
Delegate Activity	6.9(2.2)	7.0(6.0-8.7)	7.4(1.7)	7.0(6.0-9.2)
Communicate Needs	7.8(2.5)	8.0(7.0-9.7)	8.5(1.3)	8.5(7.7-10.0)
Modify Standards	7.5(2.3)	8.0(6.2-9.0)	8.3(1.2)	8.0(7.7-9.2)
Adjust Priorities	8.1(2.0)	8.5(7.2-9.7)	8.7(1.2)	9.0(8.0-10.0)
Simplify Activities	7.5(2.5)	8.0(6.2-9.7)	8.1(1.9)	8.5(6.8-10)
Plan the Day	6.8(2.6)	7.0(5.2-8.7)	7.7(1.6)	7.5(6.7-9.2)
Change Time of Doing Activities	7.0(2.7)	7.0(5.2-9.7)	7.9(1.7)	7.5(6.7-10)
Include Rest in a Day	8.5(1.6)	9.0(7.2-10)	8.5(1.7)	9.0(7.0-10)
Rest During Longer Activities	6.4(1.4)	8.1(5.2-9.0)	7.9(1.6)	8.0(6.7-9.2)

5.5 Discussion

To our knowledge, this is the first study to offer the *Packer Managing Fatigue* program, one of the most widely recognized programs for managing fatigue secondary to neurological conditions, to PwPD and to explore their perspectives on the program`s feasibility. Analysis found unique themes, and when possible, the quantitative results were

used to further elaborate and explain the qualitative findings. The combined analysis reveals three main findings: 1) PwPD perceived the *Packer Managing Fatigue: The Individual Self-Management Program* to be helpful and feasible; 2) the individual delivery via videoconferencing was a feasible alternative when in-person sessions were not possible; and 3) to maximize the feasibility of fatigue programs in PD, it may be beneficial to further tailor the program and delivery to the unique characteristics of PD-related fatigue taking into consideration the personal needs of each individual.

Both qualitative and quantitative findings demonstrated that the program supported participants to learn and implement new behaviours and strategies to manage their energy to complete their life activities, reframe their attitudes toward life, and appreciate the significance of communication and rest. The published body of evidence is strong for effectiveness of the original group version of the *Packer Managing Fatigue* program (5, 6, 27, 34). However, less is known about how patients perceive the impacts of the program (222, 243). A previous study evaluated the perceived effectiveness of the strategies included in the *Packer Managing Fa*tigue program for the MS population. It found that the two strategies *"including rest in the day*" and "*planning the day to balance rest and work"* were regarded as the most effective (222). In the present study, quantitative analysis similarly found participants were most confident in their application of "*including rest in the day."* The qualitative findings also emphasized the significance of including rest breaks and planning daily activities as effective approaches to managing the impact of fatigue on daily life. Based on the theoretical foundation of occupational therapy, rest is considered an essential aspect of an individual's daily activities, and an optimal balance between work, self-care, and rest is crucial to achieve maximum function and overall health (244). This

evidence supports the theory and suggests that occupational therapy interventions aimed at understanding and practising rest are beneficial for both PD and MS populations. Strategic resting appears to be a critical strategy of the *Packer Managing Fatigue* program, with participants reporting that a change in rest schedule can lead to an instant reward and a sense of control over their fatigue.

Participants in this study, though, found that, although they had learned a lot about managing their fatigue, they still experienced fatigue. Previous effectiveness studies of the individual and teleconference versions of the *Packer Managing Fatigue* program in the MS population found similar results. While the adverse impact of fatigue was decreased for individuals who received the program, the severity of their fatigue, mainly evaluated by the FSS, did not differ over time (5, 6). This is not surprising, given that the program's goal is not to reduce the intensity of fatigue but to manage the negative impact of fatigue (2). It is also possible that, as fatigue is better managed, people add or increase activities with no reduction in fatigue.

Although earlier research and systematic reviews have highlighted the importance of group settings and the benefits of peer support and interaction in fatigue programs (129, 245), participants in the present study did not mention the lack of peer support in the individual format. In contrast, the majority of individuals felt the individual format allowed for devoted attention of the OT. Similarly, in a previous review study which reviewed qualitative papers (n=15 articles) focused on telehealth interventions supporting patients with long-term conditions (LTCs), authors did not find compelling evidence to add peer-support in group-based delivery as an active ingredient of successful interventions (246).

Participants in this study were satisfied with the videoconferencing sessions and found them convenient. Videoconferencing may have increased access regardless of geographical obstacles and during times of physical distancing. telehealth delivery has proven valuable for individuals with chronic conditions. It has enabled them to build trust-based relationships with practitioners and to participate more actively in distance programs (246). It is notable that while virtual delivery was found to be feasible, a few participants expressed a personal preference for live interaction with their OT if a safe opportunity was available. The importance of human interaction and personal contact for individuals with LTCs has been highlighted in previous research (247). The use of videoconferencing was especially valuable during the COVID-19 pandemic.

Similar to explanations found in a previous qualitative study (231), participants in the current study reported that their fatigue was linked to various factors. In another qualitative study (243) that explored the perspective of people with MS on the teleconference version of the *Packer Managing Fatigue* program, it was also reported that the experience of fatigue is dependent on numerous factors, varies by time, and is not limited to involvement in an intense activity. This variation may influence the implementation of the program's offered strategies (243).

In PwPD, a large number of factors contribute to fatigue, which makes its management extremely difficult (112). This complexity reinforces the need for a patient-centered, multi-aspect approach with regular access to specialists and timely follow-up (248, 249). Participants in the current study suggested that considering symptoms associated with fatigue, such as medication regimens, mood, anxiety, physical activity, and disease progression, may have potential benefits for managing fatigue. One way to do this is to

embed the fatigue program in larger multi-disciplinary settings to manage other factors contributing to fatigue in PwPD. A recent systematic review found that incorporating physical exercises inspired by self-management principles into fatigue management programs may reduce the impact of fatigue (145). In another recent study, the group-based *Packer Managing Fatigue* program was combined with aerobic training and relapse prevention for people with neuromuscular disease, resulting in improved social participation and functional endurance (31). From the qualitative interviews, participants in that study were satisfied with the combination of physical training and the *Packer Managing Fatigue* program (245). Yet, there is a lack of evidence available for the use of the *Packer Managing Fatigue* program in PwPD solely or in combination with other programs, and future trials are needed to examine this hypothesis.

To enhance the support provided to patients, it is also important to ensure that the content, pace, and duration of the program meet each individual's unique needs. Although participants considered all assessed aspects of the program to be feasible, tailoring, and appropriate pace of delivery received the lowest scores among all the aspects.

Individualized care and developing therapeutic plans based on individual priorities and needs are critical in a complex and multidimensional condition like PD. This is because PD can manifest and progress differently (77, 94). To design and promote tailored health programs, identification of critical personal and disease related characteristics that require additional support is needed (250). Currently, little is known about specific characteristics that can be used to tailor interventions. A previous study in the MS population found those with a younger age and less disease impairment were more confident in using the learned strategies in the program and more likely to benefit from a teleconference group delivery

(251). Larger studies should be conducted to identify important characteristics that can be used to tailor fatigue management programs more specifically to PwPD.

To deliver integrated client-centered care for fatigue management may also depend on the expertise of the healthcare provider including OTs working with PwPD. The OTs' expertise also affects the quality of the therapeutic relationship with the client. This can result in better communication, trust, patient engagement, and a personalized approach for every individual (252). This proficiency may require specific training for OTs who work with PwPD. A recent observational study (n =51,464) examined the association between the level of expertise in PD care of healthcare providers, including OTs, and patient-reported PD-related complications (253). The results showed that participants who had access to specialized OTs experienced fewer PD-related complications (253). In the current study, none of the OTs were specialized in PD care. Drawing from this evidence, implementing even more extensive PD-specific training modules is recommended in future studies. It would also be interesting to compare the outcomes of the program when delivered by OTs with or without PD expertise in future studies.

As common in small sample size studies, the generalizability of findings is limited. A larger sample size is more likely to include individuals with a wider range of disease stages, cognitive function, and gender, allowing a deeper understanding of the potential benefits of the program and its relationship to the clinical and sociodemographic characteristics of PwPD.

5.6 Conclusion

In conclusion, this study provides preliminary evidence for the potential use of the *Packer Managing Fatigue* program by occupational therapists. The program was feasible from the patient's perspective. However, its effectiveness in clinical settings needs to be tested in future studies before it can be implemented successfully. These findings contribute significantly to the field of occupational therapy and PD research by offering new insights into the development and implementation of effective fatigue interventions from the perspective of PwPD. Future research may explore the program's effectiveness in larger scale trials and compare it to other approaches for managing fatigue in this population.

5.7 Appendices

Appendix A *Checklist for Mixed-Methods Research (MMR) Manuscript Preparation and Review.*

Rational and description of MMR design	**Provide a clear statement of the study purpose**
	☒ Explicitly describe the MMR design in accordance with Creswell's (2015) typology and use a diagram to illustrate the relationship and sequence of qualitative and quantitative research components
	☒ Justify why the MMR design is appropriate for meeting the study purpose
Transparency in describing method details	☒ Describe the study population(s) and sample (s, e.g., who, what, how many)
	☒Describe the sampling procedures (including inclusion and exclusion criteria, recruitment)
	☒ Describe qualitative data collection processes (how often data were collected, who collected the data, what kind of data collection instruments were used, how data were recorded—e.g., notes, transcripts)
	☒ Describe quantitative data collection processes (how often data were collected, who collected the data, what kind of data collection instruments were used measurements, validity/reliability)
	☒ Describe qualitative data analysis processes (coding, single or multiple coders, replication logic, credibility)
	☒ Describe quantitative data analysis procedures (missing data and how they are handled, statistical tests used)
Integration of qualitative and quantitative research components	☒ Interpret qualitative analysis results with appropriate quotes if necessary
	☒Interpret quantitative analysis results in consideration of statistical significance, selection bias, and threats to validity
	☒ Compare qualitative and quantitative results
	☒Address divergencies and inconsistencies between qualitative and quantitative results

Appendix B *Emerging Themes, Codes and Examples*

Themes	Codes	Sample Phrases
Program was helpful	I have learnt/modified behaviours and strategies to manage my energy and complete my daily activities	*"I found the lessons on dividing tasks into smaller chunks, rather than tackling a whole project at once, particularly helpful. Before the program, I would often spend a whole or half day in the garden without taking breaks, and the next day I would feel completely exhausted. It didn't make sense to me, but the program taught me that it's much more sensible to take frequent breaks and rest, so that you have energy for the next day. When I started the program, I didn't realize the importance of breaking tasks into smaller pieces, but now I see how much it can help conserve my energy".* *"Instead of piling everything in my arms, I'm going to put the books in the shelves. Now I use the trolley that I have, and I put all the books there and pushed the trolley. It's a little bit longer, but it's not as tiring as doing it the old way."* *"I was suggested to use a mixer, an automatic mixer, as I found it tiring to mix up cookies and so forth just in a bowl with a spoon. So, the result is that we went out and bought a mixer. It's much easier to do now".* *"What I found from the program that turned my fatigue problem completely around was that we plan a day ahead of time. I built into my week three days at the gym, two days of Thai chi, and other ordinary life stuff. So, in order to control my fatigue, we now plan our days around my energy levels. It was a nightmare prior to the program."* *"What stands out to me about posture and arranging things in your home so that, you know, your posture was good, like one of the things I did is put my monitor up high. I, you know, so I had to look up to it, and that stood out for me. You know, arranging tools in your kitchen and all. You know, making things easier."* *"It's an overall lesson that naps and daily rests are allowed. It was just the identification of this as a permitted activity, which I did not realize before, and it helps me a lot"*
	Gain a new outlook on life and improved motivation	*"This program was good for me to express how I felt about my condition and this made me more accepting of the way I am."* *"I used to put off things that should have come first, such as taking care of myself, so that you can actually have the energy to do your activities and to help you slow the progression of your disease, your stress, your sleep, your fatigue, and everything else. So yeah, that's very inspiring that it really helped do that."* *"I come to see myself as an older person with Parkinson's and not be so ashamed of it. I think that was a result of the study"*
	Communicating with others about my needs is important but challenging	*"Yes, I'm more conscious of things now, whereas before I was just a people, people pleaser for a long, long time and still am to a certain extent. So, it was good for me to rethink what is good for me, not just to please everybody else. So that kind of came through during this session. So, I was more aware of that."* *"But also, when I go to one of those social evenings, I'm usually on cash and sometimes I get a brain fog if I'm over tired and when people are putting things through and I'm putting them through the square system, I get confused and then that's very*

Themes	Codes	Sample Phrases
		embarrassing. And the couple of times when I was too tired, I said I was just exhausted. I wasn't able to go to those evenings, whereas before I would have pushed myself to go and then put myself in the bad place because I wasn't functioning well with the brain fog. So that definitely helped." *"I know it's important to communicate my needs, and while I try to share as much as I can, I don't usually go into detail about everything because it can become overwhelming. I usually stick to simpler things like saying, 'I take medication and eat food at these times.' People can easily understand those things. Explaining things like fatigue can be more difficult to convey to others."* *"Actually, because it's I think the session focuses on how you're talking about really, it depends on the person who's interpreting the information, how they process and understand it."*
	One of the only places that talked about our fatigue	*"Because I had nothing else to help me except my neurologist and my family doctor. This is like a whole new aspect that I think was really important. And I found it quite well put together, actually."* *"I don't get any of that from my family doctor or my neurologist. It's just a totally, you know, this is the way it is."* *"I find this this program is so focused on fatigue, OK, here are the actual physical steps I have to take and the mental, how it's affecting me mentally in order to change this. And I don't get any of that from my family doctor or my neurologist"*
	Fatigue is still there; no blame on program but disease progression	"I *think it's the progression of disease that do not allow improvement. I do not see anything in program is lacking but the progression of disease."* *"I am not certain of major changes for my fatigue. I still get fatigued, but I am doing new things, which makes it more manageable for me."* *"My disease is progressive, and I expected the fatigue to get worse all the time. But since I started the program, it didn't always get worse. There were times that I felt I was getting better, that I could survive a whole day without a rest. Like just yesterday, I felt like, boy, am I getting tired again. So, you're dealing with a progressively worse disease. You don't know what it would be if you didn't have this session, if you didn't go through these things, if you didn't go through this study, it might even get worse."*
	COVID-19 pandemic played a role	*"The COVID-19 pandemic halted many of our activities while also adding new ones. Coping with the pandemic was tiring in its."* *"Because of the COVID-19 restriction, I had to pause my exercise sessions, which helped with my energy. I believe that's why I've been feeling overwhelmed with less energy recently."*
Strengths of the program	Homework and Pre-session helped me learn and be prepared for next sessions	*"There was homework that I had to report on. I wrote my plans for the week and then I was to report back on how they were implemented."* *"Homework was helpful. It covered the areas that you have studied the previously, the things that were important and needed practice were there so we could practice at home."* *"The pre-session was sort of telling you what the session was going to be about and how you could prepare."*

Themes	Codes	Sample Phrases
	Providing a patient manual was helpful	*"The manual was excellent resource so we could go back when we wanted."* *"It's very helpful that I have the book here so I can refer to it when I need."* *"I really liked that I got the manual which had lots of examples of what to do which I did not have similar experience before."* *"Having the physical manual was really helpful. I could go back and review it whenever I needed to. It made me feel confident that I had all-time access to the content"*
	Program Is Relevant	*"Well, the word itself, fatigue, I mean, that was like bingo for me to say this is relevant to me. I didn't know how really to deal with fatigue. That's what attracted me."* *"I was satisfied with the link of the topics to me. Well, nothing was unknown to me. So. I'm happy with the outcome of the study and the amount of time I had to put into it."*
Areas for improvement	Program was intensive and layout was complex	*"There was too much going on. It was a little bit overwhelming."* *"I just seemed to go through page after page until I could actually find which one the homework was and what I was supposed to do. And but that was if I'd left it three or four days and I didn't remember what I'd been told before, it just took me a little while to find out where it was written down."* *"Sometimes when I went back a few days later to do the tasks it, I had difficulty to find where my homework was and I went to different sections and it took a little while for me to find out where it was, I was supposed to do my homework from."* *"Some of the manual material was a little complex. The table of contents, session priorities, and standards were confusing at first. But once I got the hang of it, I understood what was happening"?* *"I had some difficulty finding the material. There's not a clear break then saying, you know, this is session one done and now it's session two."* *"The one comment I was going to make is that some of these materials such as the importance of rest and sleep in session one, there was a homework and then the signs of fatigue. So, session one goes up to Page 17 up to page 18. And it's not entirely clear all the time. I'm always still in session one, right? OK? I realize when I was moving into to session two, but it made sense, but I had almost finished the session one before I realized where I was."* *"Some of the manual material was a little complex. Let's just say the table of contents, then the session priorities and standards. Until I got the hang of it, I was a little lost, but once I got the hang of it, I understood what was happening"* *"One of the things I could have said, and I propose a bullet point to make learning easier and more step-by-step, when you're doing something new, like when you're making a dish for the first time with a new recipe of something of this sort. It comes out better the second time."*
	Additional factors to improve fatigue management.	*"I find my energy level low, both physical and mental. There were no exercise components in the program."* *"I wish there was more emphasis on the exercise, and its types."*

Themes	Codes	Sample Phrases
		"It's needed to understand the medication's on/off impact. I plan based on my medication schedule. Plan your day from the start and emphasize what you can and cannot do." *"I have bad and good times and its mostly based on my medications. I think some information on that would be helpful for patients."* *"Mood swing is important a because your energy level is relatable to my mood."* *"I find that when I get anxiety throughout the day, I won't be able to do anything else. It drains my energy. So, some help with that would improve fatigue management. "* *"I am having more problems lately with my memory. For example, during one of the sessions, I learned how to use assistive technology to plan my day. But I forgot about it too soon, and I could not figure it out when I came home. I'm hoping to get some handouts or a step-by-step guide that I can use later."*
	It's important in this manual to put in the language of PD	*"It was relevant for sure but I think this program wasn't specifically for fatigue in Parkinson's, it was for fatigue."* *"All examples were for patients with MS not PD. I liked to see some more specific things about Parkinson's."* *"" I kept looking for the word Parkinson's and it wasn't there. It was all MS."* *"I kept wondering how focused it was on Parkinson's, I thought it should have language in there that brought that into the communication"*
Individual, online delivery is feasible	Although in-person sessions are more preferred, Zoom is comparable to in-person sessions	*"To me both are fine but having it virtual will allow people from different cities or areas to get benefit of these programs too."* *"Zoom was alright; I am okay with technology. I had no issues with Zoom and I was able to communicate with the OT and ask my questions. I did not find it problematic at all."* *"I like that this study was done over the Zoom. I'm not very good at all of the technology, but if I were in a setting this direct Individual with someone I didn't know, I'd be quiet and shy. I don't want to express myself out loud. I liked it this way. I mean, it's cozier."* *"In a virtual way, you're not overawed by the presence of another person, you can concentrate on the questions and topics. So, I am in favour of the virtual approach."* *"Using the Zoom was pretty effective. Videoconferencing was almost as useful as the in-person."* *"I think meeting over Zoom worked well. Obviously, it's much nicer to be in person. I like being in a room with therapist together, so I could actually feel more involved."* *"I think it was fine doing it on Zoom. I think I would have preferred it to doing it live."* *"Zoom is quite compatible with me; I have no trouble with it"*
	Individual session with therapist is helpful	*"I think it's great that the sessions were just for me. Having a therapist in an individual session was helping me to learn more as she could pace things based on my needs".* *"* *"I think it's important that you have a therapist to focus only on you, deliver the program, and walk you through."*

Themes	Codes	Sample Phrases
		"The Individual session was beneficial to me. So, if I was having trouble, I asked my therapist right away and they explained it"
More support from OTs in helpful	More access/support is needed to improve adherence	*"I am getting more problems lately with my memory. For example, I have learnt in one of the sessions to use Siri assistive technology for planning my day. But I forgot that soon and I could not figure it out when I came home. I hope I had some handouts or step by step guide so I could use it later."* *"How the session will run if you're having a good day or a bad day is important. Sometimes, I could take more some days it was overwhelming."'* *"I think most of the issues were really with me not getting out of it and not just kind of understanding where what applies to me and what doesn't. I think I could have discussed it more with my therapists to make that clear for myself."* *"I did the homework before the next session. I did not do the homework immediately following a session. Maybe a reminder or a phone call to review the materials I had to do was helping me to keep up with the program."* *"I needed to contact the therapist between sessions but there was no phone contact."* *"I wish I could have access to my therapist even after completing the program."* *"I mean what's the next stage of learning. I work better with more feedback rather than free flow, right."* *"One of the things that I could have said and I suggest a bullet point to learn easier and step by step. I know it's, maybe something because of the way I think. When you're doing something, you're making a dish for the first time with a new recipe of something of this sort. It comes out better the second time."* *"It was all useful, however I do not know if a different order would have been better. Perhaps it would be more efficient to begin with sessions that are more relevant to each individual. It took me a couple of weeks to learn and practice body positions, which were important to me"*
	The session duration and pace can be more tailored	*"The timing and content should fit into my energy consumption picture quite well. I sometimes was losing my concentration in middle of the session."* *"The sessions were too long considering that it was meant to help people with fatigue. I think they need to be shortened."* *"It was usually mostly an hour and a half, and one time I was very tired. I think I had brain fog at the time and we decided to rebook. I did find the hour and a half was a bit long. I think I would have been more comfortable with an hour."* *"It was all useful but I don't know if the order could have worked better in a different order. Maybe start the order with sessions that was more relevant to each person is more attracting. I had to wait for couple of weeks to for example, learn and practice about body postures".*

Appendix C Descriptive *of Feasibility Criteria for Packer Managing Fatigue: A Six-week Individual Self-Management Program*

Feasibility Criteria	***All participants (N=12)***			***Excluding outliers(N=10)***		
	Mean (SD)	*Median (IQR)*	*Range*	*Mean (SD)*	*Median (IQR)*	*Range*
Tailoring	3.5(1.2)	4.0(3-4.7)	1-5	3.8(1)	4.0(3-5)	2-5
Helpfulness	4.3(0.8)	4.5(4-5)	2-5	4.5(0.5)	4.5(4-5)	4-5
Skill-Training	4.3(0.6)	4.0(4-5)	3-5	4.4(0.6)	4.5(4-5)	3-5
Recommendable	4.0(0.9)	4.0(4-5)	2-5	4.3(0.6)	4.0(4-5)	3-5
Clarity and Attractiveness	4.0(0.4)	4.0(3.5-4.3)	3.5-5	4.0(0.4)	4.0(3.5-4.5)	3.5-5
Pace and Duration	3.8(1.1)	4.0(3-5)	2-5	4.0(1)	4.0(3-5)	2-5
Applicability	4.2(0.6)	4.0(4-5)	3-5	4.4(0.5)	4.0(4-5)	4-5
Relevance	4.0(0.7)	4.0(3.2-5)	3-5	4.3(0.6)	4.0(4-5)	3-5
Useful Pre-session	4.0(0.6)	4.0(4-4.7)	3-5	4.2(0.6)	4.0(4-5)	3-5
Useful Homework	4.0(0.7)	4.0(3.2-4.7)	3-5	4.1(0.7)	4.0(3.7-5)	3-5
Confidence to Use skills	4.0(0.9)	4.0	2-5	4.4(0.6)	4.5(4-5)	3-5

Appendix D *Feasibility Questionnaires*

Feasibility Questionnaire 1: Weekly Feasibility Questionnaire

Week# [To be populated by research assistant]

Participant ID code number: [To be populated by research assistant]

Now that you have completed this session, we would like to ask you for your opinions.

Instructions

Please read each of the following statements and indicate the extent to which you agree or disagree with each statement by putting a check mark below the appropriate response.

1. This week's session was organized and easy to follow.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

2. Content in this week's pre-session part for this week was presented in a clear, understandable way.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

3. Content in this week's pre-session part was tailored to my personal situation.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

4. Content in this week's homework section was presented in a clear, understandable way.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

5. The homework component of this session was tailored to my personal situation.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

6. The pace of the week`s session fit my needs.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

7. This week`s session encouraged my active participation.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

8. The content of this week's session was relevant to me.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

9. I can use what I learned this week.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

10. I am confident that I can use the skills I learned this week.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

Feasibility Questionnaire 2: Whole Program Feasibility Questionnaire

Participant ID code number: [To be populated by research assistant]

Now that you have completed the whole program, we would like to ask you for your opinions on it.

Instructions

Please read each of the following statements and indicate the extent to which you agree or disagree with each statement by putting a check mark below the appropriate response.

1. Six weeks was a reasonable timeline to complete the program.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

2. The content and activities in the program were paced to my energy.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

3. The information provided in the manual was relevant to me.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

4. The program was clear and attractive**.**

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

5. The pre-session activities were useful and helped me to prepare for discussion with my therapist.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

6. The homework component was useful to learn skills.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

7. This program was tailored to my needs.

1	2	3	4	5

Strongly Disagree	Disagree	Somewhat Agree	Agree	Strongly Agree

8. Participating in this program was helpful for me.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

9. This program has allowed me to learn new skills to manage my fatigue.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

10. I am confident using the skills I learned in this program.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

11. I will use what I learned in the program in my daily life.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

12. I would recommend this program to a friend.

1 Strongly Disagree	2 Disagree	3 Somewhat Agree	4 Agree	5 Strongly Agree

Appendix E *Participant Interview Guide*

Welcome and Introduction

You have been asked to join this session because of your participation in the Managing Fatigue: Individual Program (MFIP). Our research team is interested in your opinions on the feasibility of this research and the program we used in this study, for example the relevance of the program to you, if you will use the skills you learnt, if the manual was understandable and interesting, the logistics, and if you see any impact the program might have had on you. We are interested in hearing any suggestions you may have on how to improve the program.

This focus group/individual interview will be an open discussion. Your opinions and views are valuable to us. There are no right or wrong answers, and any positive or negative comments are appreciated so that we can improve the program for other people in the future.

You all have received the information/consent form for this part of the study. At the beginning, we want to check with you one more time and make sure you are still interested in proceeding. We want to remind you to only share information that you are comfortable talking about. You may decline to answer any questions or stop participating at any time during the discussion. This discussion should take about 60 minutes. We will audiotape this session, and then transcribe it. When we transcribe the sessions, identifying information will be removed. Your identity will always be kept confidential and will never be revealed or connected to your comments in any way. While we may report quotes collected during this session, these quotes will never be connected with

your identity. Only the research team members will have access to the tape and transcripts. The OTs who administered the program will not have access to the tapes and will never be aware of who said what during this discussion.

The objective of the focus group/individual interview is to understand people's perspectives. So, we would like to hear from everyone as each opinion is valuable to us.

FOR GROUP DISCUSSIONS ONLY: "In order to keep the meeting running smoothly, I would like to ask for your help following some ground rules.

1- Please allow one person to speak at a time without interruption.

2- Make sure that everyone has a chance to speak.

3- Feel free to ask each other for more details or clarification of your ideas.

We are taking steps to protect the anonymity and confidentiality of the participants in this study. Therefore, we ask that everyone respect the confidentially of others here today by not repeating any of the conversations outside of this group. Please also refrain from discussing these conversations with any participants outside of the group meetings".

Before we begin, we would like to ask if you have any questions.

[Answer any questions]

So, let's begin.

Please tell us about your experience with this program. We are interested in your views about the program, particularly things like the organization of the manual, the way

the sessions were run, the content in the manual, if each session matched with your priorities, the logistical aspects of the program, and what you found worked for you and what did not.

Prompts:

How did you find the manual of the program?

- The comprehensiveness of the content.
- The amount of content in each session.
- The understandable content.
- The barriers to completion that you experienced.
- The homework in the manual.
- The pre-session part of the manual.
- The pace and time duration of the program
- The convenience of the location

Thank you for all of that information. Now I would like to ask you to talk about whether the program led you to make any changes in your everyday life. Please share your perspective about any positive or negative experiences.

Prompts:

- Things you took away from the program.
- Any positive/negative impact
- Any noticeable changes in:
  - Your fatigue levels?
  - The things you're able to do every day.

- Your relationship with your family, friends, or colleagues?
- Your outlook on life?

Now, as we approach the end of this session, we would like to ask for your opinion on what how we can improve this program in future.

Prompts:

- Characteristics that make a program more favorable for people living with Parkinson's disease
- Main needs of people living with Parkinson's diseases that needs more research on
- Factors that can encourage you to participate in a program/research study.
- The impact of current pandemic on participation of people living with Parkinson's disease.

Is there anything else that you would like to share with us that we did not cover in our discussion?

Thank you very much for your time and participation!

CHAPTER 6 - MANAGING FATIGUE IN PARKINSON'S DISEASE: PREPARING FOR A PILOT RANDOMIZED CONTROLLED TRIAL

6.1 Abstract

Objective: The aim was to evaluate the feasibility of the research protocol and prepare for a future Randomized Controlled Trial (RCT) to test the *Packer Managing Fatigue: The Individual Self-Management Program* for people with Parkinson's disease (PD)

Methods: This two-arm, assessor-blinded pilot study recruited participants with a self-reported diagnosis of PD, severe fatigue, who were fluent in English, and had access to the internet. Recruitment strategies included social media, healthcare centers, community outreach, patient organizations, and local web-based advertisements.

Results: In total, 25 participants were included. The sample was diverse in clinical and demographic characteristics. Analysis, using Mixed-design ANOVA, found a significant difference in occupational satisfaction between groups over time ($p=0.09$). Additionally, a paired t-test showed a significant difference within the intervention group over time for occupational satisfaction ($p=0.04$). No significant difference for the control groups over time was detected. The level of statistical significance was set at $p < 0.1$ in this pilot study. Moderate effect sizes were observed for outcomes of occupational performance and satisfaction. Small-moderate effect sizes were found for occupational balance, as well as for the Reduced Motivation and Physical Fatigue subscales of the Multidimensional Fatigue Inventory. The effect of the program using other measures was very small.

Conclusion: The results provide preliminary evidence supporting the potential benefits of the program for people with PD. Significant improvement in occupational satisfaction and moderate effect sizes observed for several outcomes suggest that a future full-scale RCT is warranted. This feasibility study provides important insights into recruitment strategies and effect sizes that can inform the design of future RCTs.

6.2 Introduction

Fatigue in Parkinson's disease (PD) is the most commonly reported non-motor symptom (16) and is defined as a consistent sense of exhaustion that manifests in a multidimensional manner and impairs performance of routine physical and mental activities (59, 100). PD fatigue impacts occupational performance and participation (13) and is associated with early retirement and reduced work hours, which can cause financial distress (14). This extensive negative impact can lead to increased social isolation and poor quality of life (90). As a result, fatigue management has been recognized as one of the top research priorities in PD research (17).

Fatigue is difficult to measure and differentiate from other clinical symptoms, such as depression, anxiety, and apathy, making its assessment and management difficult (11, 109) There are very few pharmacological options to treat fatigue in PD. According to meta-analyses and systematic reviews, evidence for the use of doxepin and rasagiline to reduce PD-related fatigue is evolving, but additional research is required, particularly due to the their side effects (19, 20, 133). Another line of research to manage fatigue is implementation of non-pharmacological approaches, although based on current evidence, there is not much known about effective fatigue interventions in PwPD (18, 20, 73). Based on evidence found for people with other conditions such as multiple sclerosis (MS) that experience similar fatigue (38), non-pharmacological approaches such as cognitive behavioural therapy (254), physical exercise (255), and energy conservation (138) may help decrease the negative impact of fatigue on the life of PwPD. Nonetheless, additional randomized controlled trials are required to assess the efficacy of these approaches (20). In this study, we were specifically interested in evaluating a self-management program

developed based on energy conservation strategies for PwPD. This program was originally developed by Packer et al in 1995 (2) as a six-week, in-person, group-based program for managing fatigue in post-polio syndrome. It focuses on enabling individuals to learn and practice energy management strategies to manage their fatigue and its impact on daily activities. Energy-management strategies included in this program are strategic resting, activity simplification, task analysis, environmental modification, communicating about fatigue, planning, and prioritizing (2).

Since first developed, multiple delivery formats of the program have been evaluated including the original group-based, in-person version (26, 27, 31), internet (29), teleconferencing (5) and one-on-one formats (6, 7). These studies were mostly conducted in individuals with fatigue due to MS (5, 28, 34, 233); while other conditions such as neuromuscular diseases, fibromyalgia, cancer and stroke have also been studied (7, 31). Overall, the program has been shown to significantly reduce the negative impact of fatigue on daily life activities (28, 32-34), and improve quality of life (5, 7, 26, 29), participation (29, 31, 35), self-efficacy (5, 148), depression (27), and sleep quality (27). When effect sizes were reported, analysis showed medium to large effect sizes for program outcome measures (5, 26, 28, 29, 32). Additionally, the program has been combined with other interventions, such as aerobic exercise training, goal-setting physical activity, and the Envelope Theory of energy conservation (31, 32, 35), which have also shown positive outcomes. Results of these versions show improvement in perceived occupational performance and satisfaction measured by the Canadian Occupational Performance Measure (COPM) in people with MS (35) and neuromuscular conditions including (31), as well as decreased fatigue impact and improved physical activity in MS population (32).

The findings on the effectiveness of the program are more consistent in studies that used the original group program (28, 32-34). However, in studies that evaluated adapted one-to-one versions of the program, mixed conclusions were reached possibly due to variations in length and content. For example, Blikman et al (2017) evaluated a 12-session one-to-one version of the program in an RCT design in people living with MS and found no significant difference in fatigue between the intervention and information-only control groups. While Van Heest (2017) evaluated only five modules of the original program in a pre-test design for multiple groups of people with chronic conditions including MS, fibromyalgia, cancer, and stroke (7). Contrary to Blikman et al., (256) this study found significant improvements in post-test fatigue, quality of life, and self-efficacy.

In 2020, the original author and a research team developed and began testing a one-to-one format, now called the *Packer Managing Fatigue: The Individual Self-Management Program* (1). While this version is being tested in other studies, no findings have been yet published (25). In the present study, we evaluated the feasibility of the first research version of this new protocol to inform and prepare for a large-scale RCT for PwPD.

Although there is substantial evidence that the *Packer Managing Fatigue* program is effective in multiple outcomes for other populations, it has not been studied for PwPD. Only one study included a small number of PwPD (n =8 of 87 participants) (5, 29). Even though this study found that the program was effective, drawing any conclusions about its outcomes for the PD group remains debatable because results by diagnosis was not reported. Thus, this study is the first to evaluate the feasibility and preliminary effectiveness of the *Packer Managing Fatigue* program in PwPD. Since the outbreak of COVID-19, the majority of interventional research has shifted toward telemedicine.

Similarly, our study was designed to be a one-to-one videoconferencing format of the *Packer Managing Fatigue: The Individual Self-Management Program*. This mode of delivery has the potential to increase accessibility, particularly for those living in remote areas or when in-person sessions are impractical.

This study was part of a larger research project (186) that aimed to determine the feasibility of conducting a future definitive RCT for the *Packer Managing Fatigue: The Individual Self-Management Program*. Two aims were addressed to achieve this goal: (1) to evaluate the feasibility of the program from the perspective of PwPD; and (2) to explore the feasibility of this suggested pilot protocol for a full-scale RCT. The results for aim 1 have been reported in Chapter five. For aim 2, this study specifically aimed to: (1) explore the preliminary effectiveness of *Packer Managing Fatigue: The Individual Self-Management Program*; (2) identify effect sizes to inform a power calculation for sample size of a definitive RCT; and (3) assess recruitment efficacy and sociodemographic variability of participants recruited in terms of age, gender, years since diagnosis and living status.

6.3 Methods

6.3.1 Study Design

This pilot RCT was an assessor-blinded, two-armed randomized controlled trial. Eligible participants were randomized in a 1:1 ratio to either an intervention group, receiving the *Packer Managing Fatigue: The Individual Self-Management Program* + usual care, or a control group, receiving no specific intervention other than usual care. After completing the study, participants in the control group were offered the program manual and an online training workshop to learn about the program.

Due to the nature of the intervention, participants were not blinded to group allocation. The control group did not receive any additional intervention while individuals in the intervention group were required to actively participate in the program. The protocol of the study was registered with the ClinicalTrials.gov PRS (ID: NCT04267107) and received approval from the Nova Scotia Health Sciences Research Ethics Board (ref: 1027048). The Consort guideline recommended for pilot and feasibility trials (200) was used as a reporting guideline and is attached as an appendix. Minor deviations from the original protocol occurred during implementation (186). The recruitment process posed a challenge during the COVID-19 pandemic, resulting in a smaller than expected sample size. Over the two-year period of recruitment, only 25 participants were enrolled, compared to the anticipated \ 54. Due to the small sample size and therefore low power, only two time points (baseline and post-test) were included in the analysis and reported here. Additionally, one of the proposed outcome measures, the SEPECSA, was not used in the analysis. Instead, it was used to assess the confidence of participants in the intervention group after completion of the program and was included when assessing feasibility from the perspective of PwPD. The reason for this change was based on participant feedback during data collection indicating that some items on the measure were not easily understood before attending the program sessions.

As this study was a feasibility and exploratory study, no primary versus secondary outcomes were pre-determined. The outcomes were occupational performance and satisfaction, occupational balance, fatigue impact, quality of life, and sleep quality.

6.3.2 Sample Size

Effect sizes for fatigue interventions on the selected outcome measures have rarely been reported in the PD population. Therefore, given the exploratory nature of our study and its aim to assess the feasibility of a full-scale RCT, we relied primarily on methodological reviews, which recommend a minimum total sample size of 10 to 50 for pilot studies (203-205, 257).

6.3.3 Inclusion and Exclusion Criteria

All participants were adults, residing in the Canadian provinces of Nova Scotia or Ontario and provided informed consent prior to any data collection. Participants were included if they self-reported having PD, scored at least four on the Fatigue Severity Scale (FSS), could read and converse in English, had access to the internet, an electronic device, and a private location for videoconferences. Participants were excluded if they had previously completed any version of the *Packer Managing Fatigue* program, had a co-morbidity that causes severe fatigue such as heart failure or diabetes, or had significant cognitive difficulty as measured by the Mini-Mental Status Exam (MMSE): score < 13.

6.3.4 Recruitment

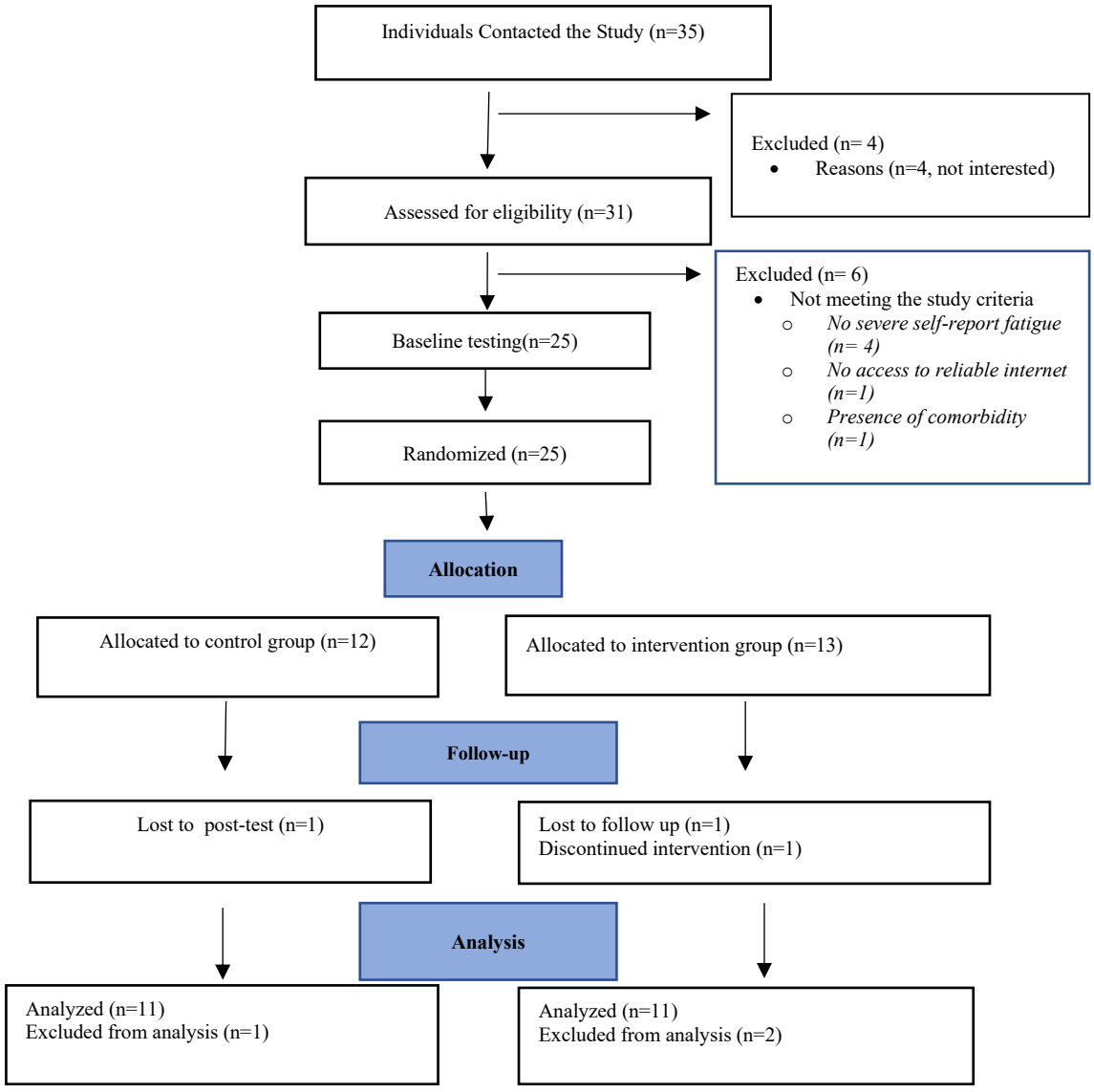
As recommended in a previous methodology study (45) and to assess the feasibility of finding an adequate sample size, multiple recruitment strategies were used. These included recruiting from patient organizations such as Parkinson Canada, through movement disorder clinics and patient-focused conferences, local web-based advertisements (e.g., KIJIJI websites), support group email listservs and websites of Parkinson Canada, social media platforms (Twitter and Facebook), and word of mouth.

6.3.5 Randomization

Interested individuals contacted the research team via email. Participants consented electronically before any data collection. Those who met the inclusion criteria completed baseline measurements. They were then assigned a number and randomly allocated to one of two groups by a research assistant. This was done using sequentially numbered, sealed opaque envelopes that were only opened after each participant completed baseline measurements. No pre-determined confounding factors were used in the randomization process.

The assessor was blinded to group allocation. To ensure blinding was maintained, participants were instructed not to disclose their group allocation during post-test assessments. Blinding integrity was monitored by documenting any instances where information about a participant's allocation was received during assessments. Figure 5 presents a CONSORT flow diagram of participant progress through the study.

Figure 5 *CONSORT Flow Diagram of Participants*



6.3.6 Intervention

Participants in the intervention group received the 6-week fatigue program called *Packer Managing Fatigue: A Six-week Individual Self-management Program* in addition to their usual healthcare services. The *Packer Managing Fatigue: A Six-week Individual Self-management Program* was modeled on the original program and retained its core content.

However, it also incorporated the additional content of sleep hygiene and cognitive fatigue in accordance with evidence-based recommendations for managing fatigue (63, 258). The six sessions of the program focused on trialling, evaluating, and adopting energy conservation strategies: Session 1) the importance of rest and sleep; Session 2) communication and body mechanics; Session 3) activity stations; Session 4) priorities and standards; and Session 5) balancing your schedule. Session six included a course review and a discussion to determine future plans. The program was delivered via the Zoom for Healthcare platform (237). Zoom for Healthcare is committed to protecting the security and privacy of its customers' data and is compliant with Canadian Data Protection regulations such as the Personal Information Protection and Electronic Documents Act (PIPEDA) and the Personal Health Information Protection Act (PHIPA) (259).

Each session comprised pre-session activities completed at home to prepare participants for in-session discussions, in-session activities focused on developing skills and strategies for energy conservation and expenditure, problem-solving, and action-planning, and post-session homework assignments designed to reinforce the application of strategies introduced during the session. Sessions were expected to last approximately 90 minutes but could be adjusted based on individual patient needs with the entire program completed within 6-8 weeks. The COVID-19 pandemic introduced several challenges for research studies. In response to these challenges and the need for physical distancing and to limit patient exposure to the virus, in this study the program was delivered via videoconferencing session.

The program was delivered by licensed OTs who were eligible to practice in Nova Scotia and recognized by the College of Occupational Therapists of Ontario (COTO) to provide

virtual services to clients in Ontario. The OTs who delivered the program completed an online training course developed by the research team, consisting of two sections: general training to learn and deliver the program, and PD-specific modules. More information about the program's content and therapist training is available in Chapter five. A fidelity checklist, designed by researchers, was completed by OTs after each session.

6.3.7 Outcome Measures to Assess Preliminary Effectiveness

All data were collected online using the Opinio Online Survey Software (206) while participants were videoconferencing with the blinded assessor using Zoom for Healthcare. Self-reported outcome measures as well as a demographic questionnaire were completed by participants. Measurements were completed at two time points: at baseline, before randomization, and at post-test, approximately 8-10 weeks after baseline measurements. Included outcome measures were fatigue impact (the Multidimensional Fatigue Inventory: MFI) (214), occupational performance (COPM-P) and satisfaction (COPM-S) (209), occupational balance (Occupational Balance Questionnaire:OBQ-11) (260), quality of life (Parkinson`s Disease Quality of Life: PDQ-8) (261), and sleep quality (Pittsburgh Sleep Quality Index: PSQI) (220). These outcomes were selected based on the content and expected impact of the program as well as a review of studies that tested the *Packer Managing Fatigue* program or focused on fatigue in PwPD. Detailed explanations of these selections and the rationale behind them are reported elsewhere (188). To assess for potential covariates of fatigue in PwPD, disease severity and depression were measured at baseline by the self-reported Hoehn and Yahr scale (HY) (224), and the short version of the Geriatric Depression Scale (GDS-15) (239) respectively.

Table 10 *Measurements Used in the Study and their Properties.*

Outcome measures & description	Measurement timing
Multidimensional Fatigue Inventory (MFI) (221): 20 items self-report scale measuring five dimensions: General Fatigue, Physical Fatigue, Mental Fatigue, Reduced Motivation, and Reduced Activity. Higher scores indicate a higher level of fatigue	Baseline (Post-consent) and post program completion
Occupational Balance Questionnaire (OBQ) (260): 11-item self-report scale measuring satisfaction and perception with the amount and variation of meaningful occupations. Higher scores indicate higher occupational balance.	Baseline (Post-consent) and post program completion
Canadian Occupational Performance Measure (COPM) (209): a standardized client-centred, occupation-focused measure using a semi-structured interview. It measures perceived occupational performance and occupational satisfaction. Higher scores indicate better occupational performance and satisfaction.	Baseline (Post-consent) and post program completion
Parkinson`s Disease Quality of Life-8 (PDQ-8) (261): an 8-items self-report scale, each representing one dimension of the PDQ-39 (Mobility, Activities of Daily Living, Emotional Well-being, Stigma, Social Support, Cognition, Communication, and Bodily Discomfort). PDQ is a short-form version of the Parkinson Disease Questionaire-39 which assesses the impact of PD on HRQoL over the past month. Higher scores indicate worse health.	Baseline (Post-consent) and post program completion
Pittsburgh Sleep Quality Index (PSQI) (220): a 19-item self-report scale that measures seven components: subjective sleep quality, sleep latency, sleep duration, sleep efficiency, sleep disturbance, use of sleep medication, and daytime dysfunction. Higher scores indicate worse sleep quality.	Baseline (Post-consent) and post program completion
Geriatric Depression Scale: Short version (GDS-15) (239): a 15-item self-report scale with yes/no questions to screen for depression in the elderly. Higher scores indicate more depressive symptoms.	Baseline (Post-consent)
Estimated Hoehn and Yahr scale (HY) (224): a self-report clinical rating scale that identifies the broad categories of motor function in PD. It includes five stages: (1) one-sided symptoms only, minimal disability, (2) both sides affected, balance is stable, (3) mild to moderate disability, balance affected, (4) severe disability, able to walk and stand without help, (5) confinement to bed or wheelchair unless aided.	Baseline (Post-consent)

To assess the efficacy of recruitment strategies, a tracking form was utilized to systematically document the number of individuals who contacted the research team, met, or did not meet study criteria. The tracking form also captured information on the date of screening and how participants learned about the study. Sociodemographic characteristics of participants enrolled were recorded using a demographic questionnaire.

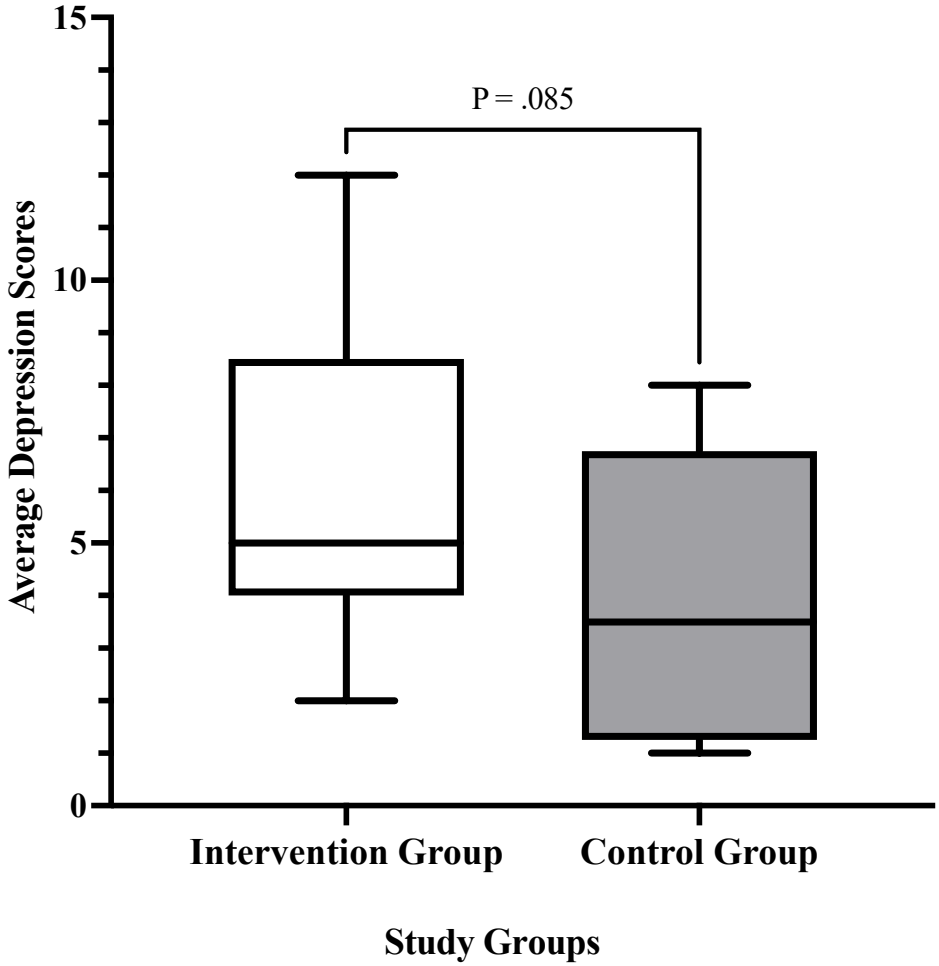
6.3.8 Data Analysis

All data were analyzed following consultation with a statistician using the Statistical Package for the Social Sciences (SPSS) version 27.0 for Windows (262). Data were first examined for skewness, outliers, and systematic missing data. If there were no per-protocol instructions for handling missing data provided with the measure, and there was less than 20% missing data at the item level, mean substitution was used. In total, only nine values (0.003%) out of 2596 possible values across all tests and time points were missing. The analysis of missing values did not demonstrate any significant pattern. Total scores for each measure were calculated based on measure-specific guidelines. Missing data at the outcome level were handled by listwise deletion, the default method in SPSS. This method excluded participants who did not complete the post-test from the analysis.

Descriptive statistics, frequency tests, and Tukey fences were used to test for outliers. Extreme outliers were defined as greater than ±2 standard deviation (SD) from the mean (226). No outliers were detected. Normality and homogeneity of variance were assessed using the Shapiro-Wilk and Levene tests, respectively. All data were normally distributed, and all measures had homogeneous variances except for one measure (COPM-P), which was therefore analyzed using non-parametric tests (Kruskal-Wallis and Wilcoxon signed-rank test). All other analyses were carried out using parametric statistics. The level of statistical significance was set at $p < 0.1$ due to the pilot nature of the study, and small sample size (263). The less stringent significance level ($p < 0.1$) was used to identify trends or effects for further investigation.

Potential covariates (age, depression, fatigue severity, disease duration, disease stage, and gender) were compared between groups at baseline using independent-sample t-tests (for continuous outcomes) and chi-square tests (for binary variables). With a mean difference between groups (MD = 2.16, 95% CI: -0.34 to 4.66, p = 0.08), depression was treated as a covariate in further analyses (Figure 6). Analyses were conducted both with and without depression as a covariate.

Figure 6 *Mean Differences Between Groups for Depression at Baseline*



The effect of the intervention on study outcomes (dependent variable) was evaluated using a mixed repeated-measures ANOVA. The time (baseline and post-test) and group

(intervention or control) were treated as independent variables, with time as a within-subject factor and group as a between-subjects factor. The results for the time*group interaction effect are reported. Effectiveness of the program on the COPM-P was evaluated using the non-parametric Kruskal-Wallis and Wilcoxon signed-rank tests. Additionally, paired-t tests were conducted separately for each group.

Effect sizes for each potential outcome measure were calculated using partial eta squared (η^2) as the measure of effect size in SPSS. To interpret the strength of effects according to the partial eta squared effect sizes, the following rules of thumb was used: a value of 0.01 represents a small effect size, a value of 0.06 represents a medium effect size, and a value of 0.14 or higher represents a large effect size (264). In this study, the interpretation of the effect of the program was based on η^2 effect sizes that were calculated for each potential outcome measure.

However, to inform the sample size calculation for a future definitive RCT using the G*Power 3.0.10 software (265), the η^2 values were converted to Cohen's f effect sizes as it is commonly used in ANOVA tests and is needed for G*Power to calculate sample sizes for mixed repeated-measures ANOVA statistic. To convert the partial eta squared (η^2) effect sizes to Cohen's f values, the following formula was used: $f = \text{sqrt}(\eta^2 / (1 - \eta^2))$ (266). The Kruskal-Wallis test value (H) was converted to partial eta squared effect size using the formula: eta2[H] = (H - k + 1)/ (n - k), where k represents the number of groups and represents the total number of observations (267).

To estimate the sample size needed for future studies, a power analysis was conducted using G*Power 3.0.10 software, based on the effect sizes obtained from the data analysis

for each measure (265). The test family and type were specified as F tests and ANOVA: repeated measures, within-between interaction. F-tests were chosen because they are commonly used to compare the means of multiple groups in ANOVA analysis. The Cohen effect sizes obtained for each measure were used, and the type I error rate was set at 0.05 with a power of 0.8. To align with common RCT designs, the number of measurements was set at three time points in two groups.

The effectiveness of each recruitment strategy, as well as all strategies collectively, was assessed by analyzing data on the number of individuals who contacted the research team, the source through which they learned about the study, and their eligibility based on study criteria. Variations in sociodemographic characteristics among participants enrolled were also examined.

6.4 Results

In total, 25 participants agreed to take part in the study and completed baseline questionnaires (13: intervention group, 12: control). There were no significant differences between study groups for any baseline characteristics except for a trend toward higher average depression scores for those in the intervention group. Three individuals (12%) withdrew from the study prior to completion, with reasons for discontinuing being illness or feeling over-committed. One participant from the intervention group withdrew after the first session of the program, and one participant from each group withdrew at the post-intervention measurement time point. Characteristics of the participants are provided in Table 11. The fidelity of the program was measured and reported as 100% following the program protocol.

Table 11 *Participant Characteristics at Baseline: Mean (SD) or N (%)*

Characteristics		*Total (N=25)*	Intervention (*n* =13)	Control (*n* =12)
Age (years)		66.8(8.6)	68.0(8.7)	65.4(8.7)
Diagnosis Duration		6.1(3.5)	6.0(3.3)	6.3(3.8)
Male gender		18.0(72%)	10.0(76%)	8.0(66%)
Living with family		22.0(88%)	11.0(84%)	11.0(91%)
Education	Graduate degree	6.0(24%)	3.0(23%)	3.0(25%)
	Post-secondary education or less	19.0(76%)	11.0(84%)	9.0(75%)
Receiving health services other than neurologists		5.0(20%)	1.0(7%)	4.0(33%)
Using an assistive device		8.0(32%)	4.0(30%)	4.0(33%)
Self-reported HY	Stages 1, 2	9.0(69%)	7.0(58%)	
	Stages 3, 4	4.0(30%)	5.0(41%)	
FSS		5.4(0.9)	5.4 (0.8)	5.5 (0.8)
MMSE		28.5(1.3)	28.8	28.2
GDS*		5.1(3.1)	6.1(3.3)	4.0(2.6)

GDS-15: Geriatric Depression Scale: Short version; HY: Hoehn and Yahr scale; FSS: Fatigue Severity Scale; MMSE: Mini-Mental State Examination (MMSE).

*Scores of 0-4 are considered normal, depending on age, education, and complaints; 5-8 indicate mild depression; 9-11 indicate moderate depression; and 12-15 indicate severe depression

6.4.1 Preliminary Effectiveness of the Program.

Outcomes were assessed at two time points: at baseline after completion of screening and prior to randomization, and post-intervention, approximately 8-10 weeks after baseline measurements, approximately two weeks following completion of the program for those in the intervention group. COPM-S scores changed significantly over time in the intervention group (mean change: 1.22, p=0.04), with a trend toward a significant time by group interaction effect [F (1,1) = 3.07, p = 0.09)], suggesting that the mean differences between groups changed over time in favour of the intervention group (Table 12).

Non-significant mean differences were observed between groups on the COPM-P, OBQ, and two subscales of the MFI: Reduced Motivation and Physical. Estimated effect sizes were medium for the COPM-P, and small-moderate for both OBQ and both subscales of the MFI: Reduced Motivation and Physical Fatigue. Details for both the mixed ANOVA and paired t-test analyses can be found in Table 12.

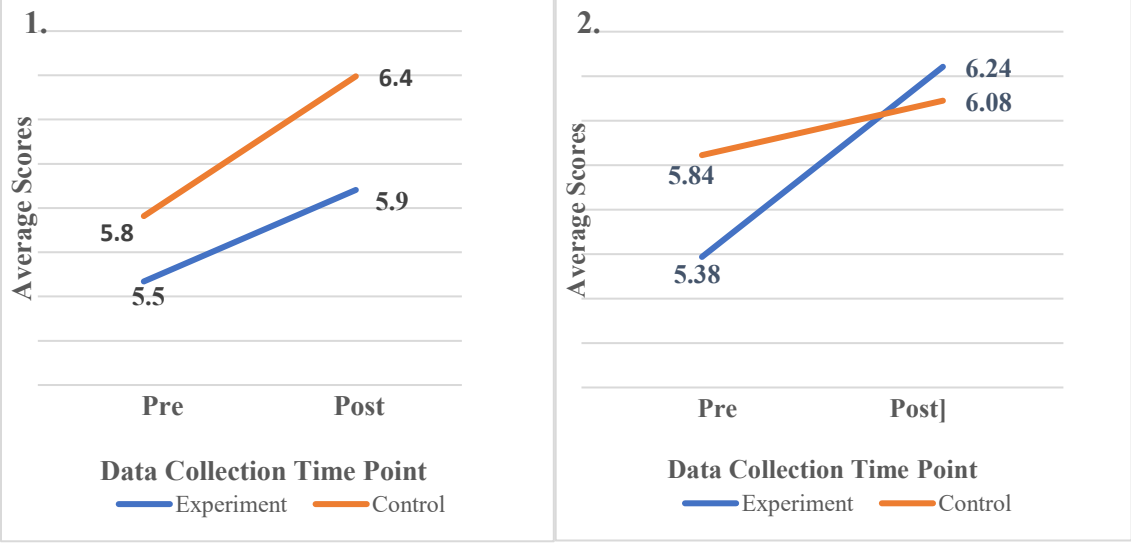
On the COPM-P, non-parametric tests were used to analyze the data as the assumptions for a mixed-design ANOVA were not met. As a result, it was not possible to test for a time by group interaction effect for this outcome. However, the Kruskal-Wallis test was used to evaluate the effect of the intervention on the change in COPM-P scores (time 2 -time 1) between groups. Wilcoxon signed-rank test was used to compare the changes of COPM-P scores within each group over time. No significant differences in the COPM-P outcome between the intervention and control groups following program completion were found (Kruskal-Wallis $H=0.24$, $p=0.62$). Within-group analyses using the Wilcoxon signed-rank test also revealed no significant differences in COPM-P scores between baseline and after intervention for either the intervention group ($Z = -.53$, $p = 0.59$) or the control group ($Z = -1.55$, $p = 0.12$).

Due to possible baseline differences in depression between the control and the intervention groups, the analysis was rerun with depression as a covariate. Both adjusted and unadjusted analyses were conducted. Although the significance levels remained unchanged (Appendix 1), there were interesting variations in the patterns of changes over time for the COPM-P across groups. Without adjusting for the depression effect, both intervention and control groups indicated non-significant improvement over time, with the control group showing slightly more improvements. However, after accounting for depression, the intervention

group demonstrated more pronounced improvements compared to the control group (Figure 7).

The effect sizes for the outcome measures, which were calculated using Partial eta squared effect size, ranged from 0.13 (COPM-S) to 0.0003 (PDQ), as shown in Table 12. Sample size calculations using G*Power 3.0.10 software and calculated Cohen f effect sizes indicated that the COPM-S required the smallest total sample size (N=46) while the PDQ-8 required the largest (N=48,177) (see Table 12). The COPM-P, MFI-Physical, OBQ-11, and MFI-Reduced Motivation had effect sizes of 0.07, 0.05, 0.05, and 0.03, respectively, and required minimum total sample sizes of 70, 96, 96, and 154 participants.

Figure 7 *Average Observed Scores for Canadian Occupational Performance Measure: Performance Subscale (COPM-P) Across Time*



1.=COPM-P without controlling for depression
2.=COPM-P after controlling for depression

6.4.2 Recruitment Efficacy and Sociodemographic Variability.

The start date of our study coincided with the onset of the COVID-19 pandemic in February 2020. As a result, recruitment was suspended in late February and did not resume until approximately eight months later in October 2020.

From February 2020 to February 2022, 35 individuals contacted the researcher. Of these, 31 completed initial screening and 26 (74%) met the eligibility criteria. A total of 25 (71%) completed baseline measurements. Participants learned of the study through patient organizations and support groups (85%), social media advertisements (11%), and clinics (2%). A total of 22 participants completed the post-intervention testing.

Although the sample size was small, it was diverse in terms of clinical and personal characteristics. Participants ranged in age from 55 to 81(median=67) and disease duration ranged from 1 to12 years (median= 5). However, diversity in disease stage, living status, and gender was less evident. The majority of participants were males (18/25) and mostly in disease stages one and two, measured with the estimated HY scale (n =16/25), followed by eight people in stage three and only one in stage four. Additionally, the majority of participants reported living with family members, including partners or children.

Table 12 *Parametric and Non-parametric Analysis: Comparison of Intervention and Control Group, Including Time effects and Time×Group Effects*

Outcomes	Group	T1	T2	Time Effect‡	Time ×group effect*					
		Mean (SD)	Mean (SD)	P	F	P	η^2‡	Cohen's f	Observed Power	Required sample size (Total)
COPM-S	Intervention	4.50 (1.96)	5.81 (2.08)	0.04	3.07	0.09	0.13	0.34	0.40	46
	Control	5.18(1.72)	5.00 (2.19)	0.69						
COPM-P*†	Intervention	5.55 (1.80)	5.69(2.00)	0.59	0.24(1)*	0.62	0.07	0.27	0.05	70
	Control	5.75(1.05)	6.45(1.29)	0.12						
MFI-General	Intervention	13.54(1.86)	13.10(1.758)	0.55	0.36	0.85	≤ 0.01	0.04	0.05	3016
	Control	13.45(1.80)	13.18(1.60)	0.46						
MFI-Physical	Intervention	12.55(4.29)	13.36(4.92)	0.44	1.09	0.30	0.05	0.23	0.17	96
	Control	13.81(4.66)	13.27(3.58)	0.63						
MFI-Mental	Intervention	12.36(3.80)	12.80 (3.83)	0.56	0.18	0.67	≤ 0.01	0.09	0.06	600
	Control	9.91(2.80)	9.82(3.12)	0.93						
MFI-Reduced Activity	Intervention	12.54(3.80)	13.81(4.89)	0.54	0.12	0.72	≤ 0.01	0.07	0.06	988
	Control	12.55(4.27)	13.00(3.09)	0.63						
MFI-Reduced Motivation	Intervention	9.73(3.60)	9.27(3.19)	0.57	0.69	0.41	0.03	0.18	0.12	154
	Control	11.18(3.12)	11.54(3.83)	0.58						
OBQ-11	Intervention	15.24(4.69)	18.64(3.95)	0.53	0.93	0.34	0.05	0.21	0.15	96
	Control	18.63(3.95)	17.9 (5.59)	0.46						
PDQ-8	Intervention	9.64(3.74)	10.36(3.9)	0.19	≤ 0.01	0.93	≤ 0.01	0.01	≤ 0.01	48177
	Control	9.27(3.46)	9.91(4.01)	0.51						
PSQI	Intervention	8.64(4.34)	8.91(3.7)	0.7	0.04	0.83	≤ 0.01	0.04	0.05	3016
	Control	6.18(2.67)	6.90(1.64)	0.43						

*Kruskal-Wallis H(df)- For the COPM-P measure, a Kruskal-Wallis test was used to analyze the differecnes between groups instead of the time by group interaction, due to unmet assumptions for a parametric mixed-design ANOVA; †Wilcoxon signed-rank test; ‡Time effect represents the paired-t test analysis and η^2 represents the partial eta squared.

COPM-S, Canadian Occupational Performance Measure-Satisfaction subscale; COPM-P, Canadian Occupational Performance Measure-Performance subscale; MFI, Multinational Fatigue Inventory; OBQ, Occupational Balance Questionnaire; PDQ, Parkinson`s Disease Quality of Life; PSQI, Pittsburgh Sleep Quality Index

6.5 Discussions

To our knowledge, this is the first pilot RCT to evaluate the feasibility and preliminary effectiveness of the *Packer Managing Fatigue: The Individual Self-Management Program* in PwPD. Additionally, this is the first report of the results obtained from the newly developed individual version of the program. Findings shed light on understanding the requirements and planning for future RCTs. The key findings of this study suggest that conducting a larger RCT to evaluate the effectiveness of the *Packer Managing Fatigue* program for PwPD is feasible considering the effect sizes found for multiple outcome measures. Despite challenges posed by the COVID-19 pandemic, the study successfully recruited a diverse group of participants, primarily through patient organizations and support groups. These results suggest that direct patient recruitment is the most effective strategy.

The main findings showed trends toward significance for satisfaction with performance of self-identified priorities in daily activities, as measured by the COPM-S, in favor of the intervention group. Significant changes over time within the intervention group were also found for COPM-S. Additionally, a medium-large effect size was found for outcome measures of COPM-S, as well as a medium effect size for the impact of the program on the performance of self-identified priorities in daily activities, measured by the COPM-P. Previous studies that evaluated the program in people with MS and neuromuscular conditions have also reported significant improvements in COPM-S (268, 269) and COPM-P (230) for the intervention group compared to a usual care control.

Regarding the direction of the effect in our study, the COPM-S showed improvement in the intervention group compared to the control group over time. For the COPM-P, the

performance of those in the intervention group appeared to improve more than those in the control group when the effect of depression was adjusted in the analysis. Notably, the control group had better average scores at baseline. Further investigation showed that those with the largest decrease in COPM-P subscale at post-test measurement were in the intervention group (n=3), and interestingly, they also had the highest depressive scores among all participants. Conversely, in the control group, four participants mentioned starting other health interventions such as physiotherapy or using assistive devices for community mobility, which was not seen in the intervention group. When the analysis was adjusted for the effect of depression, those in the intervention group showed more improvement over time compared to the control group. In PwPD, previous research has also indicated that depression is a strong predictor of fatigue (107, 109). Therefore, it is suggested that future studies also consider evaluating the impact of depression on study outcomes and/or include it as an outcome measure.

Participants also mentioned that their performance in their daily occupation had been interrupted because of the Covid-19 pandemic which may explain non-statistically significant findings in addition to the low sample size. A cross-sectional study (270) that explored the effect of the pandemic on PwPD found that the pandemic had a negative impact on essential daily life activities. Specifically, 57% of PwPD reported a negative impact on their social activities, 15% on their house chores, and 21% on their exercise activities. In our study, many participants also reported that the COVID-19 pandemic affected their daily occupations, particularly their social life and outdoor leisure activities like exercise. Given that our measurement was during the pandemic time, it was challenging to rate their occupational performance and satisfaction during pandemic

restrictions as many restrictions were still in place at post-test measurements for most of participants. While this may suggest a potential negative impact of COVID-19 on the outcome of our study, further research is needed to compare these findings during the pandemic with those during a normal time for this population.

Using the estimated effect sizes for the COPM-P and COMP-S, a minimum sample size of 28 and 20 is necessary to achieve 80% power with 5% type 1 error. Considering the preliminary findings of this study for PwPD and previous evidence for effectiveness of the program on occupational performance and satisfaction measured by the COPM (268, 269), it is recommended to conduct further research with a larger sample size to obtain more precise and reliable results in the PD population. This will facilitate more accurate conclusions about the impact of the program on this specific population.

Small-moderate effect sizes were also found for OBQ, MFI-Reduced Motivation, and MFI-Physical. Given the small sample size of this study and the independence of effect sizes from sample size (271, 272), discussion is mainly focused on the effect sizes of the study outcome measures which are necessary for understanding the feasibility of a program.

Occupational balance, an important concept used in occupational therapy refers to having the perceived right number of occupations and the right variation between the different activities that one does in daily life, including self-care, leisure, and productivity activities (273). The current study has revealed a small-moderate effect size for the program's impact on occupational balance, as measured by OBQ-11. This underscores the need for a future RCTs with a minimum sample size of 38 if this outcome is needed. To our knowledge, prior studies have overlooked the potential impact of the *Packer Managing Fatigue*

program on occupational balance. Given that the program explicitly emphasizes prioritizing and balancing activities throughout the day, changes in occupational balance can be expected in future studies. In light of these findings, it is essential to incorporate measures of occupational balance as an outcome in future research on the program's effectiveness. The preliminary results of this study highlight the potential of using OBQ-11 to assess the program's effectiveness in improving occupational balance. Including this outcome measure in future investigations can lead to valuable insights into how the *Packer Managing Fatigue* program can enhance occupational balance and point towards promising avenues for future research.

Small-moderate effect sizes were observed for the Reduced Motivation and Physical subscales of the MFI. However, the effect sizes for the General, Mental, and Reduced Activity subscales were negligible. The average effect size for the MFI was small (η^2= 0.02), with a standard deviation of 0.02 among different subscales, necessitating a large total sample size (n=970) for adequate statistical power. Previous research has demonstrated that the program significantly decreased the negative impact of fatigue in multiple dimensions in other conditions, including physical, cognitive, and psychosocial fatigue, as measured by the Fatigue Impact Scale (FIS) and Modified Fatigue Impact Scale (MFIS) (5, 28, 32, 146). Given the strong evidence supporting the ability of the MFIS to detect changes following participation in the program (27, 274), and its emerging psychometric properties in PD (275), it is cautiously recommended against using the five-subscale MFI measure in future RCTs and suggest that future studies evaluating the program in PwPD consider incorporating the MFIS.

Although there was a trend toward significant differences in depression scores measured by the GDS-15 between study groups at baseline, controlling the analysis for depression did not result in any significant differences in study outcomes compared to not controlling for depression; however, we have observed variations in direction of changes for COPM-P. Previous studies have shown that the program can lead to improved scores in depression, anxiety, and stress (27, 29). In PwPD, previous research has also indicated that depression is a strong predictor of fatigue in PD (11, 109, 113). A recent study focusing on PwPD also suggested that fatigue programs may be most beneficial for younger individuals who have higher depression scores (276). Although the study did not provide evidence for the potential impact of the depression on the impact of the program, future research can explore the relationship between depression and the program in PwPD.

This study utilized a range of recruitment strategies to optimise recruitment. The most effective approach was sending information direct to patients through patient organizations and support groups. In health research studies, each recruitment strategy has its own advantages and disadvantages. For instance, while social media recruitment can be time-efficient and may require minimal interaction with or knowledge about the local population, it can still present challenges in reaching specific patient populations (277). In this study, recruitment was limited to the provinces of Ontario and Nova Scotia due to occupational therapists' license requirements. Research ethics board restrictions for clinics outside of the province meant that we could only advertise the study in local clinics in Nova Scotia, while recruitment in Ontario was limited to patient organizations and social media. Overall, the recruitment rate through healthcare systems was low. This might be due to the limited number of PD-specific clinics in the province, ethics requirements for recruiting

from health clinics in multiple regions, and the need for well-established relationships with clinics and health centers (277). This was further compounded by pandemic-imposed restrictions and limited in-person visits to clinics.

Despite our best efforts, the recruitment process proved to be challenging and time-consuming. Based on current reports from Parkinson Canada, it was estimated that more than 100,000 Canadians have been diagnosed with Parkinson's disease. From these, 46,000 individuals live in Ontario, and more than 1,000 live in Nova scotia (278). According to Parkinson Canada's Manager of Strategic Projects and Knowledge Mobilization Research, the study information was shared with at least 1,900 people in the two provinces of Ontario and Nova Scotia. There is no precise estimate of the reach of recruitment via social media and other methods in the study.

The recruitment for this study was conducted during the most severe lockdown and restrictions imposed by the COVID-19 pandemic, a time of numerous changes and uncertainties. PwPD were not exempt from these influences. Recent case-control studies have shown that COVID-19 had a detrimental impact on the physical symptoms (279, 280), (279, 280), their daily life activities (279), and mental health (279-281) of PwPD. Furthermore, PwPD were found to be more susceptible to COVID-19 infection and to have a worse disease course (282). Finally, using digital technologies may discourage participation for some individuals depending on their age, education, income level, language barriers, and cognitive and motor impairments (283, 284).

Low recruitment may have been related to several other factors, including barriers encountered by PwPD and their caregivers in participating in research studies (285), the

presence of apathy, a common symptom of PD characterized by reduced motivation and interest in activities (286), and limited proficiency in utilizing technology and telehealth sessions, which may have also acted as a barrier to participation (283).

To enhance generalizability to the intended population, effectiveness studies should include a diverse sample that accurately reflects relevant characteristics (287). A diverse sample that includes a sample with a range of clinical and demographic characteristics, such as sex, living status, and disease severity is helpful for ensuring that the results of effectiveness studies are applicable to a wider range of individuals and minimize bias and increase the external validity of the findings, making them more relevant to real-world clinical settings (287). However, the level of this variety may depend on the research design and questions, as a diverse sample may also lead to a lack of homogeneity in the study population, which can make it difficult to draw conclusions about the effectiveness of the intervention being studied. Future studies should choose their recruitment strategies, criteria, and analysis according to their goals and focus. Although the sample size was small in our feasibility study, the recruitment plan was still able to recruit people with a variety of clinical and demographic characteristics, including age, living status, education, disease duration and severity. Fewer females and people in the late stages of PD were recruited. This is expected as PD is more common in males, with the relative risk of developing the condition being about 1.5 times greater in males than females (288). However, because the disease and fatigue experiences can present differently among males and females (251, 289), application of strategies to include both sexes in relatively equal sizes may help to better understand the impact of the program. Also, evidence suggests that although fatigue tends to start early, it can progress over time with disease severity (103).

A recent longitudinal study found that more severe motor disabilities predict fatigue in PwPD (290).

In summary, this feasibility study found that recruiting participants from patient organizations and support groups was the most feasible strategy in terms of both number and diversity of participants, which is consistent with previous research that highlights the benefits of recruiting from patient groups for ensuring the inclusion of a diverse sample in effectiveness studies (291). The findings emphasize the importance of involving patient organizations and support groups in recruitment efforts for future effectiveness studies to ensure that the results can be generalized to a wider population. Collaborating with patient organizations and support groups for recruitment efforts can lead to more meaningful and representative research findings that can benefit a broader population.

Despite its contributions, the feasibility study conducted in this research has limitations that should be acknowledged. This study was conducted during the COVID-19 pandemic lockdown, which imposed various restrictions on the participants' activities and might have influenced their willingness to participate. As a result, the generalizability of the findings may be limited, and future research conducted at different times or in different contexts may yield different results. Additionally, the COVID-19 pandemic may have affected the participants' behaviors, and responses to the study's measures. Therefore, caution should be exercised when interpreting the study's results, as they may have been impacted by the pandemic's effects. Additionally, using videoconferencing may have limited participation of those that do not have access to reliable internet. Furthermore, the small sample size made it challenging to draw definitive conclusions about the effectiveness of the program.

6.6 Conclusion

In conclusion, our study provides preliminary evidence for the feasibility and potential benefits of the individual version of the *Packer Managing Fatigue* program for PwPD. According to the effect sizes found, outcome measures of COPM and OBQ-11 seem to be feasible to measure the impact of the program in future trials. Given the small sample size and wide confidence intervals for these measures, future full-scale RCTs are needed to make definitive conclusions about the program's effectiveness on PwPD. Additionally, a longer follow-up with an adequate sample size may provide more insight into the program's long-term impact. This study was intended to form the basis of a larger trial and can serve as an important foundation for future research in this field.

6.7 Appendices

Appendix 1 *Parametric And Non-Parametric Analysis: Comparison of Intervention And Control Group With Depression as a Covariate*

	Repeated Measure-ANCOVA and Equivalent Non-Parametric Tests				
	Mean DifferencesControl-intervention (CI)	Cohen f	Observed Power	F	P
COPM-S	-0.52(-2.36-1.30)	0.47	0.08	4.19	**0.05**
MFI-General	1.01(-0.13-2.16)	0.03	0.05	0.02	0.88
MFI-Physical	2.83 (-0.80-6.04)	0.18	0.11	0.64	0.43
MFI-Mental	-1.34(-4.25-1.56)	0.13	0.08	0.36	0.55
MFI-Reduced Activity	1.56(-1.48-4.60)	0.10	0.07	0.18	0.67
MFI-Reduced Motivation	3.96(1.34-6.59)	0.11	0.07	0.27	0.60
OBQ	2.09(-1.46-5.65)	0.19	0.12	0.72	0.40
PDQ	0.30(-2.39-3.00)	0.01	0.00	0.00	0.95
PSQI	1.01(-2.98-5.01)	0.03	0.05	0.02	0.88
COPM-P*		Cohen f2	DFH	DF	P
		0.27	1	20	0.32

* For the COPM-P outcome, the non-parametric Quade ANCOVA test was used, which is an equivalent to the parametric ANCOVA in the SPSS software.

Appendix 2 *Prioritized Meaningful Occupational Performance Issues Among Participants Measures with Canadian Occupational Performance Measure (COPM)*

Prioritized meaningful occupational performance issues	**Percentage (All participants)**
Personal self-care	22%
Instrumental Activity of daily living	32%
Family roles and relationships	24%
Social participation outside family	44%
House chores and maintenance	52%
Work and productivity	56%
Leisure and play	64%
Exercise	84%

CHAPTER 7- CONCLUSION

7.1 Summary of Findings

This chapter provides a general overview and a reflection on findings of this thesis. Specific discussion points related to each manuscript have been provided in each chapter, thus, this final chapter focuses on the overall discussion, limitations, and potential implications for future studies.

The objective of this study was to explore the feasibility of implementing the *Packer Managing Fatigue: A Six-Week Individual Self-Management Program* for PwPD. This was achieved by exploring participants' experiences and perceptions regarding the program's content and delivery, as well as its perceived impact, relevance, and feasibility. Participants' confidence in utilizing the skills and strategies acquired through the program was also assessed. In addition, this study aimed to evaluate the feasibility of conducting future large-scale RCTs by estimating the required sample size for future RCTs using effect sizes found for selected outcome measures, as well as analyzing preliminary effectiveness and recruitment efficacy.

Chapter 3 reports findings of a scoping review aimed to understand individually delivered self-management programs available for those with fatigue due to chronic conditions including PD by delineating three important self-management components, namely, self-management strategies, self-management support and active patient participation. Overall, 15 fatigue interventions were included. Although a wide range of diseases were found to be included, PD was missing from the literature. Findings from this scoping review showed that generally, three main types of programs exist: 1) those developed based on cognitive

behavioural therapy (CBT); 2) the original or an adapted version of the *Packer Managing Fatigue* program; and 3) "Other" programs based on one or a combination of other theories including energy envelop theory, energy management education, psychobiological entropy model, and the chronic care model. All types of fatigue programs indicated evidence of inclusion of the three self-management components examined in this review. However, the number and range of domains within a self-management component and the combination of these components varied across program types. Based on the findings of the scoping review, it is evident that self-management components are an important aspect of fatigue programs in chronic conditions. It is interesting to note that the *Packer Managing Fatigue* program group had the greatest level of self-management support and active patient participation compared to other groups. The findings also reveal that the most commonly cited self-management strategies, as defined by the TEDSS framework, in this program were Activities, Process, and Social Interaction strategies. This is consistent with the aim and focus of this program in providing tools and strategies to enable those with chronic fatigue to actively take control of their energy management to optimize participation in their meaningful daily activities despite the negative impact of fatigue. Overall, this scoping review provides valuable insights into the potential of this program in fatigue management in chronic conditions. Although findings of this scoping review showed that there is limited utilization of fatigue self-management programs among PwPD, there are evidence-based programs including the *Packer Managing Fatigue* program, which have been shown to be effective in other conditions and may potentially offer similar benefits for managing fatigue in PwPD.

In Chapters 5 and 6, this pilot RCT evaluated the feasibility of the program and the feasibility of the proposed research protocol for future RCTs. In Chapter 5, the main question asked was "to what extent is the *Packer Managing Fatigue: The Individual Self-Management Program* feasible for PwPD". A mixed-methods evaluation was conducted with 12 PwPD with self-reported fatigue to explore the feasibility of the program from their perspective. Feasibility questionnaires and a standardized self-efficacy measure, designed specifically for the program, were used to collect quantitative data. Qualitative data was collected through individual and focus group interviews. Overall, participants found the program helpful and feasible. They found the program provided a learning opportunity to better understand and plan their energy expenditure in order to manage completing meaningful daily life activities. Participants appreciated the strategies included in the program, such as the importance of rest, breaking activities and using tools and technology, and found them beneficial tools to manage their fatigue. The mean scores for all feasibility criteria were three or higher out of a possible five. Participants also suggested some areas for improvement to fit the needs of people with PD. For example, although participants found all aspects of the program to be feasible, the tailoring and pace of delivery received the lowest scores. Some participants found the session too long while others found them appropriate. Some participants also wanted more follow-up and support between sessions. In addition, some participants suggested that incorporating additional strategies to manage factors that contribute to their fatigue, such as mood and physical activity management, may enhance fatigue management in PD. Participants were generally confident in their ability to apply the strategies learned in the program to their daily lives.

Chapter 6 evaluated the feasibility of the research protocol for a future RCT for PwPD. Recruitment strategies were assessed, and effect sizes were determined. These effect sizes were then used to calculate the sample size needed for a definitive study.

The recruitment strategies employed in this study effectively recruited participants with a variety of characteristics, despite the small sample size. The included participants varied in age, disease duration, and disease stage. However, the self-identified gender of the participants was less diverse compared to other characteristics. Out of the total sample, 72% self-identified as men. This is consistent with PD being more prevalent in males than females. Among all recruitment strategies used in this study, recruiting through patient organizations and support groups was the most effective strategy (85% of total participants).

Analysing differences between the study groups over time found a positive trend toward improvement for the COPM-S measuring individual`s satisfaction with performance on self-identified priorities in daily activities in the intervention group compared to the control group. In terms of effect sizes, an approximate medium effect was observed for this outcome, as well. A moderate effect was also seen for the COPM-P measuring the perceived performance on self-identified priorities in daily activities. These findings also suggested a feasible required minimum sample size of N= 70 for future RCTs.

In addition, small-moderate effects were observed for the OBQ-11, and two subscales of MFI: Physical Fatigue and Reduced Motivation. However, the overall effect of the program measured by the MFI was small based on criteria for interpreting the partial eta squared effect size. The program had a negligible effect when measured with PSQI and PDQ-8. No

significant differences between the study groups over time were found for any of these measures.

7.2 General Discussion

Based on the findings from Chapters 5 and 6, it is suggested that the *Packer Managing Fatigue* program has potential application in PwPD and is feasible to be tested in a large-scale RCT design. According to patients, receiving the program was a positive experience that enabled them to better understand their fatigue and learn ways to minimize its negative impact on their lives. These findings are comparable to those found from the pilot RCT which showed trends toward improvement in satisfaction on prioritized occupational performance issues and small-moderate effect sizes for four other outcomes of occupational performance on prioritized activities, occupational balance, and physical and reduced motivation aspects of fatigue. Future studies with adequate sample sizes should be conducted to rigorously evaluate the effectiveness of the program. The following sections discuss the reflection and application of findings from this feasibility study for future studies, including (1) Patient Perspective; (2) Recommended Outcome Measures for Future Randomized Controlled Trials (RCTs): Fatigue, Participation in Daily Life Activities, and Occupational Balance; and (3) Pandemic-Era Research Recruitment.

7.2.1 Patient Perspective

The program was reported to be beneficial by participants who perceived it as a positive opportunity. They found it to be a learning opportunity to adopt behaviors and strategies to manage their energy. For instance, they learned about task simplification and the importance of taking frequent breaks to conserve energy. Additionally, most participants

reported gaining self-acceptance, optimism, and motivation for self-care, which may have contributed to their ability to engage in daily activities with greater satisfaction. These findings are consistent with the observed trends toward improvement in satisfaction with completing self-identified prioritized daily life activities, which suggests that the program may have an impact on participants' ability to engage in meaningful activities and manage their fatigue.

Participants also appreciate the printed manual and the pre-session and homework activities in the program, helping them prepare for and practice program content and activities. The physical manual was particularly helpful, as it gave participants all-time access to the content. Overall program feasibility was rated high, and participants were generally confident in applying the learned strategies.

Some participants suggested areas for improvement in the program content and delivery that can be further evaluated in future studies. Participants suggested including management of other factors that impact fatigue in PwPD, such as medication side effects and physical inactivity. They also felt that incorporating PD-specific terminology and examples and tailoring the pace and content of the program to be more PD-related may improve their engagement in a program. More specifically, some participants mentioned that they appreciated shorter sessions with increased follow-up between sessions due to concentration problems and memory difficulties. Some also mentioned that they would have preferred to focus more comprehensively on specific sessions they were interested in first.

A study on bothersome symptoms and coping preferences in PwPD suggested that interventions addressing complex PD symptoms such as fatigue need to be comprehensive, including education on physical activity and emotional support (292). Understanding and education about additional factors that contribute to worsening of fatigue in PD, such as anxiety and depression, medication side effects, and the importance of physical activity were also highlighted in previous studies (100, 112, 293). For example, some participants in the current research believed exercise to be a valuable activity to help increase their energy and function in daily activities. According to a large study that explored the perspective of PwPD on triggering and alleviating factors to their fatigue, nearly half of the study participants felt that exercise alleviated their fatigue. On the other hand, also about half of the participants report that physical exertion either did not affect fatigue or worsened fatigue. Thus, the relationship between exercise and fatigue appears complex (111). Those who believed exercise is alleviating their fatigue had significantly lower scores on measures of fatigue and depression compared to other groups (111).

Based on current evidence, some research suggests that exercise may improve fatigue in (294, 295), while others did not find a significant impact (141, 142). Currently, there are several gaps in the literature on this topic (296). Available studies have small sample sizes with heterogeneous exercise programs and types, limiting their generalizability heterogeneous exercise programs, limiting their generalizability (140). Information on specific exercise regimens and types of beneficial exercise is limited and the mechanisms by which exercise improves fatigue are not well understood.

When exercise was used as an additional strategy to the *Packer Managing Fatigue* program for people with MS and neuromuscular conditions (31, 32), mixed results were found as

well. Although some evidence suggested a positive effects when physical activity interventions were used in addition to the *Packer Managing Fatigue* program, differences were seen in the types of physical activities used. One study used a multidisciplinary approach that included various approaches such as medical treatment, psychosocial support, physiotherapeutic approaches, and energy-saving methods based on the *Packer Managing Fatigue* program but did not find significant differences in study outcomes between the intervention and control groups (35). Another study found that the physical activity plus *Packer Managing Fatigue* program significantly decreased self-reported fatigue and physical activity compared to a social support intervention but did not show significant differences compared to a physical activity-only intervention (32). The third study used an exercise approach in addition to *Packer Managing Fatigue* program that involved aerobic exercise training, exercise education, energy conservation management, and implementation and relapse prevention. This study demonstrated significant improvements in performance, satisfaction, and walking test in the intervention group compared to the control group (31).

In summary, although some participants felt exercise appears to be a valuable strategy for participants, the evidence on the effectiveness of exercise for managing fatigue in PD is mixed and further research is needed to determine the most effective types, dosages, and lengths of exercise approaches for managing fatigue in PwPD and in combination with *Packer Managing Fatigue* program.

Some participants in the current study expressed the need for more support from healthcare providers to apply the strategies learnt in the program. This support was mainly referred to as tailoring the pace and the duration of the sessions in order to align it with their unique

needs with increased follow up between sessions. Some found the program sessions were long and beyond their energy, others needed more time to follow the sessions and activities. Barriers such as disease progression and memory difficulties were also mentioned as factors that may hinder the implementation of learned content and necessitate specific support from therapists. Also, as people were in different stages, the priority of sessions may be different for different people based on their individual characteristics. For example, some people appreciated more support from the therapists to learn proper body mechanics and ergonomics, while others found the scheduling and rest as the most needed strategy.

PD symptoms and experiences vary greatly between individuals in terms of symptoms, disease progression and response to treatment (77). Therefore, careful consideration of personal characteristics and confounding factors is important when delivering patient-centred programs including fatigue interventions. In addition, numerous factors have been identified as triggers or correlates of fatigue in PD, including age, disease duration, medication dose, disease severity, motor impairments, cognitive deficiency, depression, anxiety and apathy, and sleep disturbance (11, 109).

Tailoring health programs to the unique characteristics of an individual can be a challenging task as well as consideration of potential confounding variables is challenging, as it requires identifying critical personal characteristics that necessitate additional support. There is currently little known about specific participant characteristics that can inform the tailoring of fatigue interventions (250). Despite the limitation of the small sample size, descriptive analysis in this study suggests that older participants with longer disease duration were less confident in implementing energy conservation strategies in their daily

lives. This finding implies that this subgroup may require more support to meet their unique needs.

Multiple techniques included in the therapist training to maximise the outcome for patients. These included principles of self-management support, the Transtheoretical Model of Behavioural Change, also known as the stages of change model, motivational interviewing, and principles of social cognitive theory (1, 3). However, efficient delivery of programs for a complex condition such as PD may also depend on the disease-specific expertise of OTs. The expertise of an OT can play a pivotal role in optimizing patient-centeredness and tailoring fatigue interventions to meet the unique needs of each client living with PD (297). Providing optimal care for complex chronic neurological diseases such as PD requires highly specialized healthcare. Specialized healthcare professionals can enhance intervention engagement, active participation, and care delivery for PwPD (297). In addition, effective therapeutic relationships are related to the expertise of the healthcare providers. As expertise increases, communication, trust, patient engagement, and tailoring the approach for each individual can improve (252). Future studies may benefit from measuring the impact of OTs' expertise and the therapeutic relationship on program outcomes which may provide valuable insights into optimizing care delivery for PwPD.

It is important to consider a limitation of RCT design research which is the potential tension between adhering to an evidence-based structured intervention protocol and customizing it to meet each participant's unique needs. Maintaining internal validity and reducing variability requires strict adherence to the protocol. However, this may constrain therapists' ability to tailor the intervention to individual participants (298, 299)

7.2.2 Recommended Outcome Measures for Future RCTs

Previous reviews have examined tools for measuring neurological fatigue and recommended a wide range of measures, making it difficult to choose which to use (36). The evaluation of fatigue measurement tools is not equally established across chronic neurological disorders, with less research available regarding PD (37, 38). In a systematic review (38) that assessed the psychometric properties of fatigue questionnaires in neurological conditions, of all 38 studies investigating 31 different self-report fatigue scores, only five studies, including four fatigue measures, were found to be used previously for PwPD; three studies were for stroke, and the rest were for MS (n =30).

By measuring different dimensions of fatigue impact, multidimensional measures provide a more comprehensive assessment of the effectiveness of fatigue interventions (63). This can be particularly useful in conditions such as PD, where fatigue often manifests in different ways (63). At the time this project was developed in 2016, no multidimensional questionnaires had been adequately validated for use in PwPD (37, 63). Among the available multidimensional self-report fatigue measures, the MFI was the only measure suggested by the Movement Disorders Society for the multidimensional assessment of PD fatigue despite not being specifically validated in PD (38). The MFI is a generic measure that evaluates the impact of fatigue on daily life (38). Measures that evaluate the severity of fatigue were not included as an outcome measure since the aim of the program was not to change the severity of fatigue but the negative impact that it has on daily life.

Previous studies evaluating different versions of the program have shown that the perceived severity of fatigue, mostly measured with the FSS (5, 274), or Checklist Individual Strength (CIS-20R), does not change post-intervention (6, 31). However, when

the impact of fatigue on daily life activities was measured, positive effects of the program have been seen (5, 27, 28, 274). According to findings for the MS population, the *Packer Managing Fatigue* program improves multiple dimensions of fatigue impact, including physical, cognitive, and psychosocial, particularly when measured with the Fatigue Impact Scale (FIS) (28, 29, 32, 33) and its modified version, the Modified Fatigue Impact Scale (MFIS) (27, 35, 274).

Based on the findings of this study, the feasibility of using the MFI to measure the effectiveness of the *Packer Managing Fatigue* program remains uncertain. Although, the analysis of differences between groups showed small-moderate effect sizes for the Reduced Motivation and Physical subscales of the MFI after program completion, the average effect size of the program using MFI was small. While patient data indicate that the program was helpful in improving their understanding of fatigue and providing strategies to manage its negative impact on daily activities, we observed a very small effect for the Reduced Activity subscale of the MFI. Reduced Activity in the MFI was defined as a potential consequence of subjective fatigue on daily life activities, mostly in terms of decreased or incomplete performance in a day. This subscale includes items such as 'I think I do very little in a day' (36).

Sample size calculations also indicated a wide range of required sample sizes, from 38 for the Physical Fatigue subscale to 3016 for the General Fatigue subscale. These findings indicate that not all MFI subscales may be sensitive for measuring the program's impact in future RCTs. Considering the average small effect size with required large sample size (n =970), we do not recommend using the five-subscale MFI measure in future studies.

The Modified Fatigue Impact Scale (MFIS) is another common multidimensional measure used to assess the effectiveness of fatigue programs on multiple dimensions of fatigue. Previous studies, evaluating the effectiveness of the *Packer Managing Fatigue* program on fatigue impact, found improvements in multiple dimensions including physical, cognitive, and psychosocial fatigue as measured by the MFIS (27, 35, 274).

Although in previous evidence in 2004, the Modified Fatigue Impact Scale (MFIS) was recommended for use in the MS population (300), its psychometric properties were later confirmed in PwPD in 2017 (275). According to a systematic review of neurological fatigue, most available fatigue measures are adapted based on the two measures of the FSS and FIS. Considering the strong evidence for the MFIS's ability to measure changes after receiving the *Packer Managing Fatigue* program in previous studies (27, 35, 274) , and the emerging evidence for its psychometric properties in PD (275), we recommend that future studies include the MFIS when evaluating the program in PwPD.

The preliminary findings of the current study indicated trends toward significant improvement for the satisfaction with performance in daily activities, measured by COPM-S, in favor of the intervention group. In addition, a moderate effect size for occupational performance, measured by the COPM-P, with no significant differences between groups was observed. Approximately half of the previous studies that used a version of the *Packer Managing Fatigue* program used occupational performance or participation as an outcome domain. Occupational performance of an individual refers to participation in daily tasks and activities for self-care, productivity, and leisure in response to the demands of their environment (209).

Several measures were used in previous studies to assess the effectiveness of the program on participation in daily life activities. These tools included the Impact on Participation and Autonomy Scale (IPA) (6, 301); the COPM (30, 31); the Utrecht Scale for evaluation of rehabilitation participation; the Activity Card Sort (ACS) (29, 31), the Rehabilitation Activities Profile (RAP)(6), and the Community Participation Indicators (25).

In this particular study, the COPM was chosen as a client-centered outcome measure to assess perceived occupational performance and satisfaction on prioritized activities/participation issues (302). The COPM measures changes in the perceived ability to perform and satisfaction with relevant and important daily activities (209). The use of the COPM enhances client-centered practice as participants are actively involved in identifying their occupational performance priorities. This active involvement increases the client's motivation and allows for individualization of clinical programs to meet the unique needs of each person (302, 303).

The preliminary findings of this study are consistent with previous research that used COPM as an outcome measure to evaluate the impact of the *Packer Managing Fatigue* program. Previous studies have shown that completing the program resulted in improvements in COPM subscales for people with neuromuscular (230) and MS (268). The ability of the COPM to measure changes in a one`s performance and satisfaction with their important daily activities aligns well with the aims of the *Packer Managing Fatigue* program, which is to enable individuals to participate in their daily activities despite experiencing fatigue (2). Most of participants in this study mentioned that using the strategies and skills they learnt in the program, they were able to manage their energy during the day to complete their important daily tasks. Therefore, future studies should

consider using participation in daily life activities measured by the COPM as an outcome when evaluating the program's effectiveness for PwPD.

Fatigue can hinder an individual's ability to achieve occupational balance, thereby impacting their overall occupational performance. Consequently, it is crucial to assess occupational balance when evaluating the occupational therapy interventions that aim to empower individuals to actively participate in a variety of important and meaningful occupations, as this is linked to their mental and physical health and well-being being (304, 305). Achieving a sense of balance between meaningful activities can promote health and improve quality of life (306). The OBQ-11 is a tool developed to measure an individual's satisfaction with the amount and variation of their occupations and has shown promising reliability and construct validity in general populations (215). However, further research is needed to examine its psychometric properties in PD and other neurological conditions (307).

The *Packer Managing Fatigue* program is expected to enhance occupational balance due to its emphasis on balancing daily activities. The program includes an activity to help participants balance their day, which involves discussing with the OTs their current balance of self-care, productivity, and leisure. To facilitate understanding, participants are asked to reflect on the proportion of these activities in their day, whether they are satisfied with the balance, areas that may be neglected, and areas that consume more time and energy than desired. In the current study, analysis of between-group differences found a small-moderate effect size for occupational balance as measured by the OBQ-11, suggesting a potential impact on occupational balance if tested on a larger scale. Therefore, by incorporating the OBQ-11 to measure occupational balance in future RCTs of the *Packer Managing Fatigue*

program, researchers can gain valuable insights into the program's effectiveness and contribute to the available evidence.

7.2.3 Pandemic-Era Research Recruitment

In March 2020, shortly after receiving ethical approval to begin recruitment for the current study, Canada experienced the most severe lockdown due to the pandemic. Similar to many other research studies, our research design was significantly impacted by the COVID-19 pandemic.

Despite all efforts, the recruitment rate remained suboptimal, which may be attributed to the challenges imposed by the COVID-19 pandemic. Nevertheless, the study was able to recruit a diverse sample of participants with a variety of clinical and demographic characteristics, including age, living status, education, disease duration and severity.

The COVID-19 pandemic has significantly affected the design of research studies, including recruitment and retention of trial participants, a shift from in-person to remote data collection methods, ethical considerations, and the impact on trial outcomes. The pandemic has also increased the burden on participants, including additional stressors or challenges related to COVID-19, which can impact their ability to engage in the intervention and affect health outcomes (39, 41). Consequently, this study transitioned to videoconference delivery and online data collection. Although these modifications made it possible to conduct the study during the pandemic and may have increased the program's accessibility to remote areas (308), some people may not have access to or were uncomfortable with the use of internet connections and electronic devices. Higher income, higher education, and telehealth use prior to the COVID-19 pandemic were associated with

telehealth use during the pandemic (40). Previous studies also found that some factors including older age, less education, lower income level, language barriers, and increased cognitive and motor impairments are barriers to use telehealth delivery for healthcare users (283, 284).

Multiple factors may have made it challenging for PwPD to engage in research activities during the COVID-19 pandemic. For example, COVID-19 caused significant decrease in quality of life and health outcomes of people with chronic neurological conditions. In PwPD, the pandemic has also negatively impacted the physical symptoms due to decreased physical activity, as well as their daily life activities such as work, household, and leisure (279-281). The pandemic has had a detrimental effect on the mental health of PwPD, with increased anxiety and depression being the most commonly reported symptoms (270, 309). Furthermore, research has demonstrated that PwPD are more susceptible to contracting COVID-19 and are at risk of experiencing a more severe disease course. The pandemic has also made it challenging to engage in research activities, particularly for individuals with pre-existing health issues such as PD (310).

The COVID-19 pandemic may have had an impact on the outcomes of research studies conducted during this period. For instance, in our study, many participants had to limit their social participation and outdoor activities due to the pandemic, which could have potentially impacted their occupational performance, occupational balance, and quality of life. Therefore, given the unique circumstances of the pandemic, such as social distancing measures, lockdowns, and other restrictions, it is important to acknowledge and consider the potential impact of the pandemic when interpreting and generalizing the findings of

studies conducted during this period and comparing the results of research studies completed during the COVID-19 pandemic with those of studies conducted at other times.

7.3 Conclusion

This research addressed a significant gap in the current literature by preparing for and informing future RCTs to evaluate the effectiveness of an evidence-based fatigue management program for PwPD: the *Packer Managing Fatigue* program. Given the lack of RCTs evaluating fatigue management approaches in PD, this study provides valuable insights into the design of future RCTs evaluating fatigue management approaches in this population. Additionally, this study provides valuable insights into the patient perspective on the program.

The results of this feasibility study indicate that the *Packer Managing Fatigue: The Individual Self-Management Program* delivered through videoconferencing sessions was feasible and also the research protocol was feasible. Therefore, it is recommended to test effectiveness in future full scale RCTs. This conclusion is based on findings from the parts of the study. The program received positive feedback from participants indicating that it was helpful in managing their fatigue and completing their daily activities. In addition, an observed trend toward significant improvements in occupational satisfaction with a medium-large effect, as well as small-moderate effect sizes found for occupational performance, occupational balance, as well as physical and reduced motivation subscales of fatigue were found. Some areas for improvement were suggested, including considering how best to adjust for or tailor delivery based on disease-related factors and individual characteristics. Participants also suggested considering additional approaches that manage

other contributing factors to fatigue in PD, such as management of physical activity and stress. These preliminary positive findings and given that there is no other proven fatigue intervention for this population, the findings of this study suggest that the *Packer Managing Fatigue* program has potential to be used in fatigue management in PwPD if its effectiveness is established in full scale RCTs.

Future studies are also suggested to evaluate the efficacy of the program in conjunction with other possible fatigue management strategies as recommended by patients such as physical activates. Additionally, other aspects of PD fatigue management should be measured and explored, such as the impact of the therapist's expertise and allied healthcare relationships on study outcomes. The positive feedback from participants indicate that this program can be a valuable tool for both PwPD and OTs working with them to manage their fatigue and improve their daily life activities.

The study also had some limitations that should be noted. The study was conducted during the COVID-19 pandemic lockdown and under various restrictions that might have influenced the participation rate. Therefore, the generalizability of these findings may be limited, and research conducted at different times may yield different results. Moreover, the study was conducted using videoconferencing, which might have excluded many potential participants who were unable or unwilling to use electronic devices due to physical limitations or personal preferences.

7.4 Future Recommendations

1. Given the preliminary evidence for the feasibility and potential benefits of the *Packer Managing Fatigue* program for PwPD, it is recommended that full-

scale RCTs with larger sample sizes and adequate power be conducted to rigorously evaluate the program's effectiveness in managing fatigue in PwPD.

2. Consider testing the Modified version (MFIS) to measure fatigue impact in future studies with PwPD.

3. Future RCTs evaluating the effectiveness of the *Packer Managing Fatigue* program in PwPD may consider using the COPM and OBQ as outcome measures, given the preliminary evidence for their potential effect on occupational performance, satisfaction, and balance. However, their limitations such as the subjective nature of occupation selection and prioritization by participants should be taken into account.

4. Based on the findings of this study, it is recommended that future research efforts focus on nationwide recruitment to obtain a larger and more diverse sample, with greater representation across disease stages and gender. This will facilitate more comprehensive analysis of the impact of these factors on the outcomes of fatigue interventions. Given the success of patient organizations as a recruitment strategy in this study, it is suggested that future research should prioritize collaboration with these organizations to enhance recruitment efforts.

5. To better understand the effectiveness of the *Packer Managing Fatigue* program in managing fatigue in PwPD, future RCTs should evaluate it both as a standalone intervention and in combination with other approaches such as exercise and anxiety and stress management. In the meantime, PwPD who report fatigue may work with their healthcare providers to implement a comprehensive

fatigue management plan that may include the *Packer Managing Fatigue* program. It is also suggested that future studies consider partnering with clinicians and decision-makers in real-world clinical settings to explore the perspective of OTs on barriers and facilitators for tailoring the program to PwPD and resources required to ensure its implementation.

6. It is suggested that therapist training be expanded for PD-specific needs, and occupational therapists tailor the delivery of the program based on disease-related factors and individual characteristics to potentially optimize its effectiveness. In addition, it is suggested to incorporate PD-specific examples when delivering the program to PwPD

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GENERAL APPENDICES

Standardized Measures Used in This Study

The following is a list of measures used in this study. Due to copyright limitations, only their citations and sources are provided:

- Occupational Balance Questionnaire -11 (OBQ-11): Håkansson, C., Wagman, P., & Hagell, P. (2019). Construct validity of a revised version of the Occupational Balance Questionnaire. Scandinavian Journal of Occupational Therapy, 27(6), 441-449. I couldn't find any specific information on where to obtain this measure or if permission is needed to use it. You may want to contact the authors of the study for more information.
- Parkinson's Disease Quality of Life Questionnaire (PDQ-8): Jenkinson C, Fitzpatrick R, Peto V, Greenhall R, Hyman N. (1997). The Parkinson's Disease Questionnaire (PDQ-39): development and validation of a Parkinson's disease summary index score. Age Ageing. 26(5):353-7. To use the PDQ-8 the copyright holder (Oxford University Innovation) will require you to have a license. Licenses for academic users do not typically attract fees. Full details can be found here: https://innovation.ox.ac.uk/outcome-measures/Parkinsons-disease-questionnaire-pdq-39-pdq-8/.
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available for non-profit research, but for pharmaceutical studies, permission is required for use.

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Sociodemographic Questionnaire

DALHOUSIE UNIVERSITY

Evaluating the Feasibility of the Individual Managing Fatigue Program for Individuals with Parkinson`s Disease

Participant ID: **Date:**

We are interested in learning about you. Please answer each question below by choosing the option that best suits you or giving short answers to the questions.

Age: Disease Duration (years):	Gender: ◯ 1-Male ◯2- Female ◯3- Others -Please specify ◯4- Rather not to say
Marital Status ◯Married ◯Single ◯Widowed ◯Common-law relationship ◯Other (please specify):	Highest level of education completed: ◯Less than high school completion ◯High school completion ◯ Postsecondary education (Bachelor's, Trade, College ◯ Graduate Degree (Master's, PhD) ◯ Other (please specify):
Are you currently working or employed? ◯1- Yes ◯2- No	**If you are not working,** is PD the main reason for not being employed? ◯ 1-Yes ◯ 2-No **If you are working,** how are you employed? ◯1-Full time work ◯2-Part time work ◯3-Casual
What is your current living arrangement? ◯1-Living alone ◯2-Living with your partner ◯3-Living with your children ◯4-Others-Please specify	Do you use any assistive devices? ◯1-Yes ◯2-No ◯If yes, please specify:
How do you manage your fatigue? ◯1-Medication ◯2-Rehabilitation ◯3- Both ◯4- Other (please specify)	If you have been receiving rehabilitation interventions in last 6 month, please specify: ◯1-Occupational Therapy ◯ 2-Physiotherapy ◯3-Speech Pathologist ◯4-Psychologist/ Mental Health Professional ◯5-Home care professional ◯6-Others (please specify) ◯N/A

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March 29, 2023

Managing Fatigue in Parkinson's Disease: Protocol for a Pilot Randomized Controlled Trial

Published, Canadian Journal of Occupational Therapy

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